Massive digestive bleeding as a result of gastric heterotopy: a case report

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Abstract

Introduction. Gastric heterotopy is a rare entity in the pediatric population. It occurs in the gastrointestinal tract, leading to digestive bleeding.

Clinical case. This is the case of a 10-year-old boy with gastric tissue in the proximal jejunum, which caused two massive digestive bleeding episodes. Diagnostic techniques included endoscopic capsule, enteroscopy, and biopsy. The patient was scheduled for laparotomy and resection. After one year of follow-up, he remained asymptomatic.

Discussion. Gastric heterotopy approach represents a diagnostic challenge. Owing to how rare it is, there is no global consensus in terms of treatment. However, surgery is the definitive therapy. In this case, decision was made not to perform intestinal resection and anastomosis, but resection of the compromised intestinal wall. No malignity was reported in the literature reviewed.

Key Words: Gastric heterotopy; Pediatrics; Jejunum; Gastrointestinal bleeding.

Sangrado digestivo masivo por heterotopia gástrica. Reporte de un caso

Resumen

Introducción. La heterotopia gástrica es una entidad infrecuente en la población pediátrica. Se presenta en el tracto gastrointestinal llevando a cuadros clínicos de sangrado digestivo.

Caso clínico. Se reporta el caso de un escolar de 10 años, el cual presentó tejido gástrico en el yeyuno proximal, originando sangrado digestivo masivo en dos ocasiones. La secuencia de apoyos diagnósticos requirió cápsula endoscópica, enteroscopia y biopsia. Fue llevado a laparotomía y resección de la lesión. En el seguimiento al año se mantuvo asintomático.

Discusión. Su abordaje genera un reto diagnóstico. Debido a su infrecuente presentación no hay un consenso global para el tratamiento, sin embargo, la intervención quirúrgica es la terapia definitiva. En este caso no se hizo resección intestinal y anastomosis sino resección de la pared intestinal comprometida. No se reportó malignidad en la literatura revisada.

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Palabras Clave: Heterotopia gástrica; Pediatría; Yeyuno; Sangrado gastrointestinal.

INTRODUCTION

Gastric heterotopy (GH) is a rare entity characterized by the presence of ectopic gastric mucosa. It is caused by an abnormality in embryonic development at intrauterine week 4, with subsequent tissue hyperplasia over time[1,2].

It should be suspected in the presence of intestinal bleeding, intestinal obstruction, or chronic anemia, among others[3]. The most frequent locations include the esophagus, the colon, and Meckel's diverticulum (MD)[4]. We report the case of a boy diagnosed with GH in the proximal jejunum, which is rare in this pathology. Patient management and clinical outcome, as well as a literature review in the pediatric population, are presented.

CASE REPORT

A previously healthy 10-year-old boy presented at the pediatric gastroenterology unit of our institution with history of two tarry stool episodes within 10 months. Both episodes had been treated at another healthcare facility. The first required blood product transfusion and ICU stay as a result of hemodynamic instability. While in hospital, an upper GI endoscopy (UGIE) was carried out, which showed no intestinal bleeding. Diagnostic laparoscopy was subsequently performed, which revealed no abnormal findings, followed by exploratory laparotomy, which found no lesions potentially accountable for this episode.

Five months later, he had a new episode, which required red blood cell transfusion. A new UGIE was conducted, without bleeding evidence. He was referred to our institution after undergoing an endoscopic capsule study, which showed an active bleeding area in the proximal jejunum, without anatomical lesions. Edema of the adjacent intes-

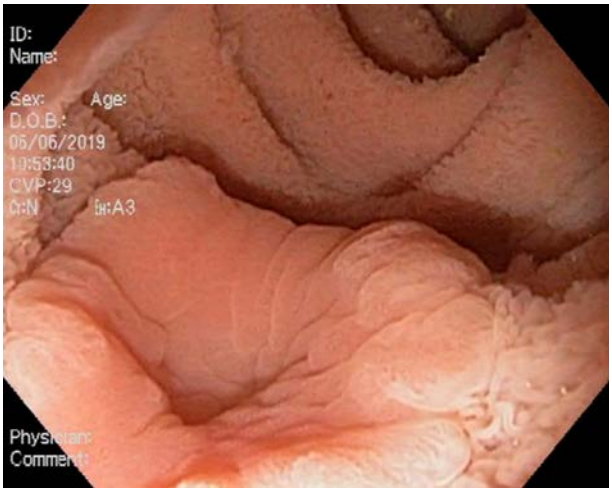

Figure 1. Enteroscopy image showing a lesion with elevated borders, a depressed center, and a smooth surface in the proximal jejunum.

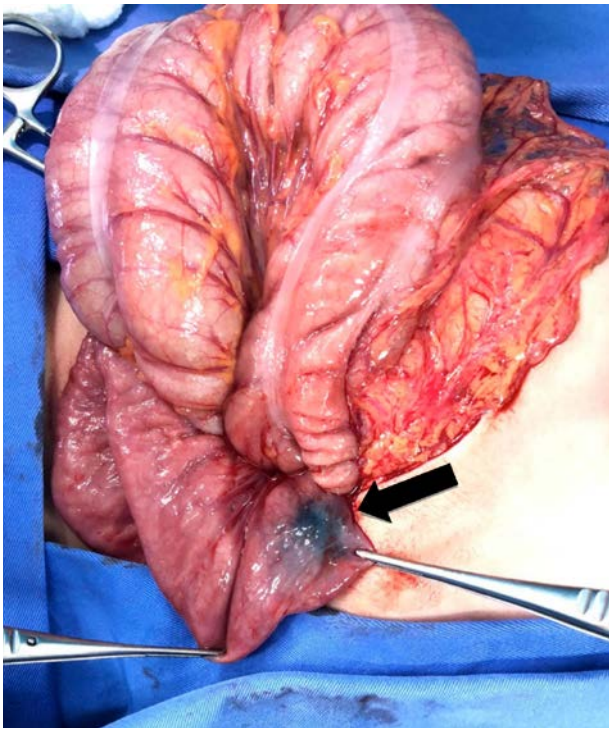

Figure 2. India-ink-marked lesion at the beginning of the proximal jejunum (*black arrow*).

tinal mucosa was also observed. Omeprazole treatment at 20 mg every 12 hours was scheduled for 1 month, followed by 20 mg once a day for 4 months.

Two months later, he was scheduled for enteroscopy, which revealed a hypervascularized lesion with elevated borders, a depressed center, and a smooth surface in the proximal jejunum (Fig. 1). Biopsy was carried out and marked with India ink. The pathological study reported heterotopic gastric mucosa without malignity. He was assessed by the pediatric surgery department and scheduled for open resection with intraoperative enteroscopy. During laparotomy, the India-ink-marked lesion was found 7 cm away from Treitz angle (Fig. 2), which was confirmed under enteroscopic vision. Wedge resection was carried out in the compromised segment, and the defect was closed in 2 layers. The remaining gastrointestinal tract was analyzed, but no further lesions were found. The pathological study confirmed diagnosis of GH, along with an area of gastric mucosa with glands covered with parietal (oxyntic) and principal cells (Fig. 3). In the immediate postoperative period (POP), he was referred to the ICU for monitoring purposes and remained there for one day. On POP day 4, oral nutrition was resumed without complications. He was discharged on POP day 5. One month later, he was hospitalized as a result of partial intestinal obstruction, which healed without surgery. He was discharged on POP day 5. In the one-year follow-up period, no further bleeding symptoms or intestinal obstruction episodes were noted.

DISCUSSION

In GH, the acid secretion of the ectopic gastric tissue causes tissue changes in the adjacent mucosa (erosion and ulceration), which are accountable for symptoms and signs, including intestinal bleeding. GH should be suspected in the presence of chronic intestinal bleeding in children in whom other causes (polyp, MD, arteriovenous fistula) have been ruled out[5]. According to Abdulrahman Al-Hussaini et al.'s review, GH is more frequent in men than in women (M:W – 19:5), presenting at a mean age of 7 years, consistent with the characteristics of this patient[6]. There are no epidemiological studies describing prevalence and incidence in the pediatric population. In our literature review, GH cases were found in various locations along the gastrointestinal tract[1,5].

Rosa et al. reported the case of an older infant with a single nodular lesion at the base of the longue that was surgically resected, without recurrence during follow-up[7].

Location at the distal esophagus represents a diagnostic challenge. Valeriu et al.[8] described the case of an adolescent with recurrent episodes of epigastric pain. Endoscopic findings revealed multiple secreting polypoid lesions in the distal esophagus, with histopathological examination demonstrating the presence of ectopic gastric mucosa.

Tanioka et al. identified a stenotic GH lesion in the pylorus of an adolescent. Conservative treatment with dilatations was initially applied. However, as a result of recurrence, distal gastrectomy was decided upon as a definitive treatment[9]. Alexander et al. found gastric mucosa in the ampulla of Vater, which caused recurrent pancreatitis episodes[10].

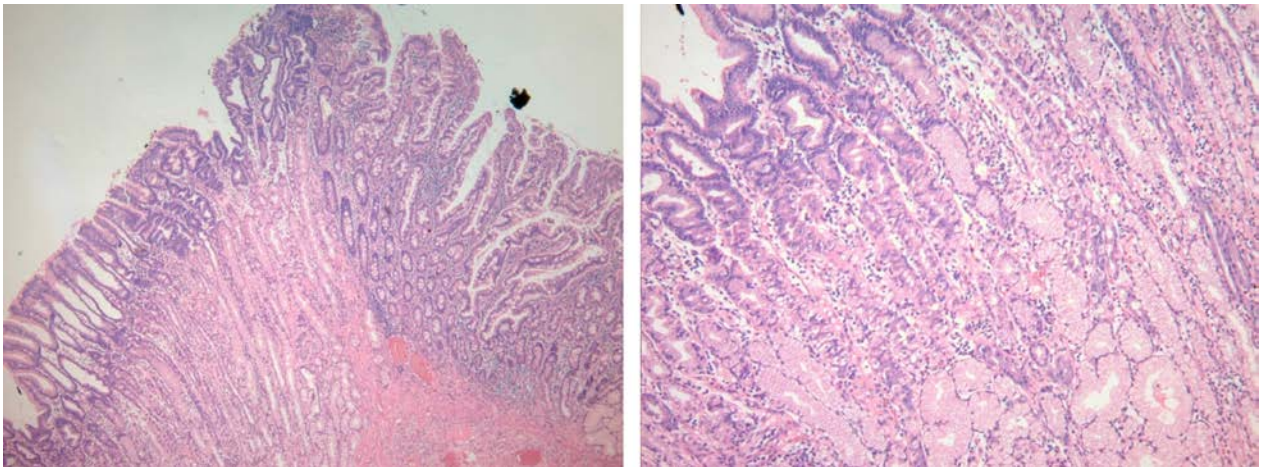

Figure 3. Histological image of the lesion. An area of gastric mucosa with glands covered with parietal and principal cells can be seen.

Another study found gastric mucosa in the proximal jejunum of a 9-year-old patient with chronic intestinal bleeding[2]. In the report by Cai et al., a large gastric tissue polypoid lesion was found in the ileum. It was surgically treated with intestinal segment resection and anastomosis[3]. In these last 2 cases, surgical resection of the compromised segment turned out to be the definitive treatment, contrarily to our case, where wedge resection instead of intestinal resection was carried out, given the proximity of the lesion to Treitz angle. The need for resection of a whole segment depends on the lesion's size, extension, and location.

Conservative management with proton pump inhibitors in order to stop acid secretion helps reduce symptoms. However, following treatment discontinuation, symptoms may reoccur, in which case surgery is required as a definitive treatment[11].

GH-related intussusception is another clinical presentation[12,13]. Ahn et al. reported the case of a child with recurrent intussusception episodes. Enteroscopic assessment revealed polypoid lesions in the proximal jejunum, with the histological study showing gastric tissue. Definitive treatment was based on enteroscopic resection, which stands as a safe strategy in the management of this pathology[14].

A retrospective study analyzed the clinical and demographic parameters allowing the presence of gastric mucosa to be predicted in MD[4]. 2 groups of MD patients were established, according to whether they had GH or not. A statistically significant difference in favor of the presence of GH when MD's base was > 1.5 cm (79.4% sensitivity and 87.7% specificity) was found.

The use of video-capsule as a diagnostic method can prove convenient and safe when it comes to characterizing intestinal bleeding[2,3]. The swallowing process of the video-capsule can be expensive in the pediatric population, but introduction through UGIE has been demonstrated to be safe[15]. This report is an example of its diagnostic use in those cases where access to more distal portions of the small bowel proves difficult with other techniques.

GH has also been associated with gastrointestinal tract malformations. The study by Schapiro et al. found a large area of GH in the small bowel of a girl with history of intestinal malrotation, annular pancreas, and gastroesophageal reflux surgery at an earlier age[16]. Seyde et al. reported the case of an adolescent with history of intestinal atresia repair undergoing laparotomy as a result of intestinal obstruction. Once the compromised segment had been resected, the pathological study showed GH in the anastomosis carried out in the neonatal period[17].

GH associated with helicobacter pylori infection has also been described. This contributes to the deterioration of the adjacent mucosa and increases malignity risks[1,18].

CONCLUSION

Ectopic gastric mucosa is rarely seen in the pediatric population. It should be suspected based on clinical manifestations suggestive of intestinal bleeding, with endoscopy being a useful diagnostic technique. Up until now, no malignity cases in this ectopic tissue have been reported. Surgical resection represents the definitive treatment, and in cases like ours, wedge resection is feasible without the need for intestinal resection.

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