

Complicating Recurrent Gastric Heterotopia of the Small Intestine: Case Report

İnce Bağırsakta Komplikasyonlarla Birlikte Tekrarlayan Gastrik Heterotopi

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ABSTRACT Heterotopic gastric mucosa (HGM) occurrence outside of Meckel's diverticulum and intestinal duplication is a very rare pathology of the gut. Herein we report a complicating recurrence of unusual localization HGM in the small intestine. A 13-year-old boy presented with abdominal pain and melena. He had a medical history of prior surgery due to ectopic gastric mucosa, complicated with perforation and bleeding. In the differential diagnosis, Tc 99m pertechnetate scintigraphy demonstrated ectopic gastric mucosa in the midgut. In laparotomy, approximately 90 cm of the small bowel including the abnormal segment was resected and the patient did well. Gastric heterotopia of the small intestine is a rare pathology. These lesions can be confused with other disorders of the intestine (intussusception, infectious diarrhea, inflammatory bowel disease, etc). Total resection cures the condition. However, it should be kept in mind that microscopic residuals can cause relapses.

Key Words: Gastric heterotopia, small intestine, gastrointestinal bleeding

ÖZET Heterotopik gastrik mukoza (HGM)'nin, Meckel divertikülü ve bağırsak duplikasyonlarının dışında gastrointestinal sistemin başka bir yerinde bulunması nadir bir durumdur. Bu olgu sunumunda, ince bağırsakta yerleşim gösteren, komplikasyonlara neden olan ve tekrarlayan gastrik heterotopili bir hastayı sunduk. On üç yaşında erkek hasta karın ağrısı ve melena nedeni ile hastanemize getirildi. Hastanın daha önce HGM'nin neden olduğu ince bağırsak delinmesi nedeni ile opere olduğu ve operasyon sonrasında tekrarlayan gastrointestinal kanamalarının olduğu öğrenildi. "Tc-99m-pertechnetate" sintigrafisinde incebağırsakta uzun bir segmentte yaygın tutulum izlendi. Laparatomide gastrik heterotopi olan segment eksize edildi ve hasta sorunsuz olarak taburcu edildi. İnce bağırsakta gastrik heterotopi nadir bir klinik durum olmasına karşın, açıklanamayan sindirim kanalı kanamalarının, delinmelerinin ve tekrarlayan invajinasyonların ayırıcı tanılarında düşünülmelidir. "Tc-99m-pertechnetate" sintigrafisi bu hastaların tanı ve izleminde önemlidir. Kesin tanı ve tedavide cerrahinin yeri hayli önemlidir. Gastrik heterotopi olan segmentin eksizyonu tedavi için yeterli olmakla birlikte, mikroskopik kalıntıların hastalığın tekrarına neden olabileceği unutulmamalıdır.

Anahtar Kelimeler: Gastrik heterotopi, ince bağırsak, gastrointestinal kanama

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Heterotopia, from the Greek "heteros" (different) and "topos" (location), is defined as the occurrence of normal tissue in an abnormal location. Synonymously, the term "choristoma", from the Greek "choristos" (separated), has also been used.¹ HGM may be found anywhere in the gastrointestinal tract. Gastric ectopic tissue has been found in the tongue and mouth, in the esophagus, duodenum, biliary tree, liver, pancre-

as, small bowel, Meckel's diverticulum, colon, vermiform appendix, and rectum.²⁻⁵ Larynx, thyroid gland, adrenal gland, umbilical polyps, the urinary bladder, and placenta as well as intrathoracic, intra-abdominal, and spinal cysts have all harbored gastric tissue.^{2,4,6,7}

In the alimentary tract HGM is found most commonly in a Meckel's diverticulum and in intestinal duplications respectively.^{5,8} The presence of HGM in the small intestine is a very rare condition and may be life-threatening for children. It may be complicated with gastrointestinal bleeding, inflammation, bowel obstruction, intussusception and perforation.⁹

We reported a 13-year-old boy with HGM of the small intestine without congenital abnormalities, which was complicated with intestinal perforation, recurrent bleeding and intussusception.

CASE REPORT

A 13-year-old boy was admitted to our hospital with a 4-day history of abdominal pain, melena, debility and lack of appetite. His medical history was significant for prior abdominal surgery. He had been operated for intestinal perforation six years ago in a peripheral hospital. His previous medical notes revealed that he had had two perforated sites 60 centimeters distal to the ligament of Treitz in the previous operation and had undergone resection and end-to-end anastomosis. The histopathological examination at that time had revealed ectopic gastric mucosa, inflammation and perforation. After the prior operation, his severe abdominal pain and gastrointestinal bleeding had persisted. He was hospitalized two times within the last two years because of severe gastrointestinal bleeding and intussusception, but no identical etiology could be determined. He was diagnosed with type I diabetes mellitus when he was 9 years old.

In initial physical examination, he was pale and tachycardic. He had left paramedian incision scar and no peritoneal irritation symptoms. Digital rectal examination was normal. The laboratory test including complete blood count, coagulation tests and blood biochemistry were within normal

limits except for severe anemia (Hb 3.9 g/dL) and high blood glucose level (450 mg/dL). Abdominal US scan revealed intussusception. Hydrostatic enema did not demonstrate an ileocolic intussusception and the abdominal computed tomography did not reveal an abdominal mass. Gastroduodenoscopy and colonoscopy could not demonstrate any bleeding site. Tc-99m-pertechnetate scan showed extensive tracer accumulation throughout the long segment of the small intestine that appeared simultaneously with the activity of the gastric mucosa (Figure 1). Upper GI tract series with barium demonstrated multiple filling defects thought to be polypoid lesions in the small bowel (Figure 2).

Recurrent severe gastrointestinal bleeding in addition to abnormal scintigraphic and radiological findings warranted laparotomy. Laparotomy, which allowed complete exploration of the intesti-

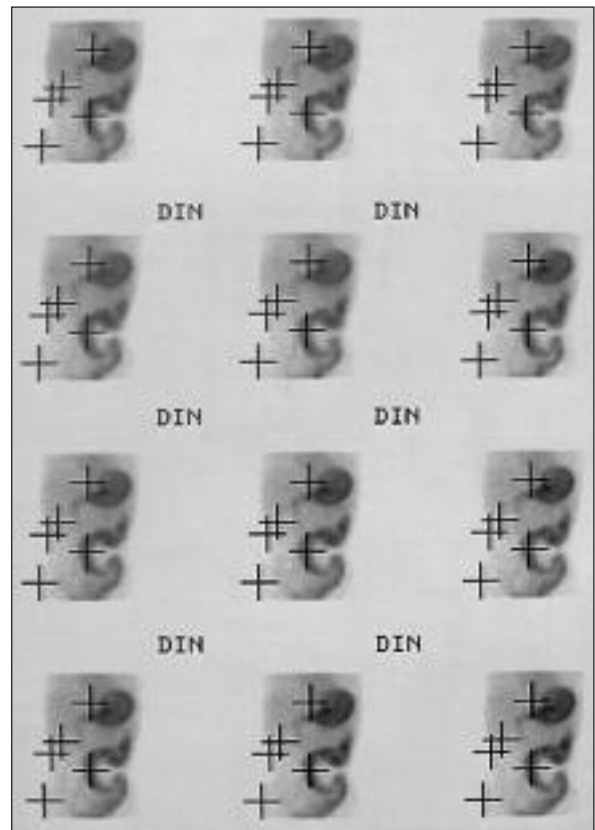


FIGURE 1: Tc-99m-pertechnetate scan revealed an extensive tracer accumulation throughout the small intestine that appeared simultaneously with the activity of the gastric mucosa.



FIGURE 2: Multiple filling defects thought to be polypoid lesions in the small bowel.

ne, revealed that small bowel loops were adhered to the left upper quadrant and adhesiolysis was performed to separate the loops. The bowel was edematous and thickened between 40 cm distal to the ligament of Treitz and 210 cm proximal to the ileo-cecal valve. Polypoid structures were palpated within this segment. We performed enterotomy and explored vegetative polypoid and sessile lesions along the whole lumen of this bowel segment. Approximately 90 cm of the small bowel including the abnormal segment was resected and an end-to-end anastomosis was performed.

Macroscopic evaluation of the specimen revealed coarse, rugose and partly polypoid mucosa involved globally (Figure 3). On microscopic examination, the grossly abnormal areas showed extensive replacement of the small bowel mucosa by full-thickness fundic type glands. (Figure 4).

The patient was discharged on postoperative day 10 uneventfully. At postoperative 3 months, repeated Tc-99m-pertechnetate scintigraphy showed no abnormal tracer accumulation. The hemoglobin level was between normal ranges and there

was no occult blood in feces. Proton pump inhibitors were initiated to prevent recurrent complications. The patient is doing well now with no signs of intestinal bleeding and abdominal pain.

DISCUSSION

Isolated heterotopic gastric mucosa without any associated morphological abnormalities, such as Meckel's diverticulum or intestinal duplication, is a very rare condition. The first case of HGM in the small intestine without congenital anomalies was reported by Poindecker in 1912 in the ileum.¹⁰ Approximately 30 cases of ectopic gastric mucosa beyond the ligament of Treitz, not associated with Meckel's diverticulum, have been reported in the literature.⁹

Gastric heterotopia of the intestine may develop in both sexes and all age groups, but symptomatic cases are more common in children.¹¹ Although the exact mechanism of HGM is not known, the lesions are suggested to arise from the epithelium of the primitive gut as a congenital anomaly, become separated from the primordial stomach and undergo hyperplasia over time.^{12,13}

The origin of ectopic gastric mucosa has been divided into either heterotopic (congenital) or metaplastic (acquired) lesions based on its histological architecture. Skandalakis et al suggested metaplasia of pluripotent endodermal cells of the embryonic foregut as an origin for heterotopic tissues.¹⁴ HGM has the characteristic glandular structure and cellular elements of normal gastric fundus.^{2,10} In contrast, metaplastic gastric mucosa, which occurs within the damaged intestinal mucosa, consists of focal gastric type surface epithelium with a relatively small number of chief and parietal cells. In contrast to "true" HGM, metaplasia does not have the appearance of fully developed fundic mucosa and does not involve the full mucosal thickness. Mucosal damage and inflammation are responsible for metaplastic type of gastric mucosa as seen in the inflammatory bowel disease, celiac disease, necrotizing enterocolitis (NEC), Barrett's esophagus and peptic damage seen in the duodenum.^{9,15-18} Therefore, the metaplastic gastric epithelium that



FIGURE 3: Macroscopic image of the specimen.

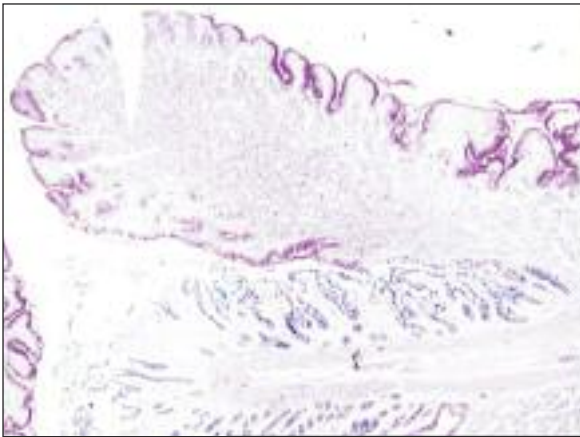


FIGURE 4: Histopathological examination revealed a prevalent heterotopic gastric mucosa (alcian blue-periodic acid-Schiff, pH 2.5, x40).

occurs in these diseases are distinct processes from the rare heterotopic lesions of the jejunum and ileum.¹⁰

HGM may become symptomatic with inflammation, bleeding, or perforation due to acid secretion of ectopic gastric mucosa and furthermore by intussusception or intestinal obstruction due to mass effect. The source of bleeding from diverse lesions of the small intestine is often difficult to determine. The outstanding symptom is commonly melena, but the need to examine the many feet of small and large intestine in order to determine the source is usually a vexing problem.¹⁹ Endoscopic methods may aid the diagnosis of HGM, if the localization of the lesion is accessible such as esophagus, stomach, duodenum or colon.

Tc-99m-pertechnetate scintigraphy is a well-established diagnostic technique to evaluate children with lower gastrointestinal bleeding. Tc-99m-pertechnetate is taken up and secreted by the tubular glands of the gastric mucosa.⁸ Although Tc-99m-scintigraphy has false positive and false negative results, it is the gold standard for the diagnosis of heterotopic gastric mucosa. The reported sensitivity ranges from 50-92%. In experimental studies, Priebe et al found that at least 1.8 cm² of gastric mucosa was necessary for imaging by gamma camera techniques.²⁰ Pharmacological intervention using H2 antagonists may increase the sensitivity.²¹ False positive results noted in the literature include intussusception, bowel inflammation, gastrointestinal bleeding unrelated to HGM, uterine blush, retention of tracer in the urinary collecting system, vascular lesions such as hemangiomas, and arteriovenous malformations.^{8,22,23} False negative Tc-99m-pertechnetate scintigraphy may result from the small size of the island of HGM. Residual contrast in bowel from previous studies may hinder the detection of HGM. Tracer in the urinary bladder may obscure a focus of HGM if it is located near the bladder.^{8,22,23}

Although HGM is a rare condition, it should be considered in the differential diagnosis of gastrointestinal tract bleeding in infants and children. Treatment involves resection of the involved intestinal segment. The main problems after surgery are recurrence and microscopic residue of the HGM. Most cases are already complicated at the time of presentation. Our patient was admitted to a peripheral hospital with intestinal perforation. An intestinal segment involving the perforation and ectopic gastric tissue was resected. It seems that the microscopic residual HGM was responsible for the complaints of the patient allowing residual remnants to grow up until they were resected. Jimenez et al reported a 4-year-old boy with recurrent gastrointestinal tract bleeding secondary to jejunal gastric heterotopia that was explored twice for the resection of the involved intestinal segment.⁹ In addition, Bueno et al reported a case of a child who was operated for HGM in the small intestine for four times; at the final operation they used intraoperative Tc-99m-pertechnetate scintigraphy for the

localization of the HGM.²³ HGM is difficult to determine during the operation; microscopic residual tissues or disseminated lesions may be overlooked. Intraoperative endoscopic examination or enterostomy during laparotomy may be useful for unidentified lesions.²² Tc-99m-pertechnetate with a handheld gamma probe has also been used for unidentified lesions.²³

Long-term treatment with proton pump inhibitors may prevent recurrent ulceration. Periodic screening with hemoglobin level, stool tests for oc-

cult blood, and Tc-99m-pertechnetate scanning may detect lesions before they result in ulceration of the adjacent mucosa.⁹

Summing up, although HGM in the small bowel is a rare clinical condition it should be considered in the differential diagnosis of unexplained gastrointestinal bleeding, perforation and recurrent intussusception. Tc-99m-pertechnetate scintigraphy is important for the diagnosis and the follow-up of such patients and surgical resection is the treatment of choice.

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