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CASE REPORT | COLON

An Unusual Case of Gastric Heterotopia Presenting as Rectal Prolapse

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ABSTRACT

Heterotopic gastric mucosa (HGM) involving the rectum is an uncommon finding. It is especially rare in young children. Rectal prolapse is an uncommon presentation of HGM. We report a case of HGM in the rectum of a 2-year-old previously healthy girl, who presented with rectal prolapse and painless bleeding. Endoscopic mucosal resection was performed to completely resect the lesion after the patient failed to respond to proton pump inhibitors. This case underscores the importance of considering HGM involving the rectum as a cause of rectal prolapse in young pediatric patients.

KEYWORDS: rectum; prolapse; hematochezia

INTRODUCTION

Heterotopic gastric mucosa (HGM) is characterized by the presence of normal gastric mucosa distinctly demarcated from the surrounding mucosa and completely separated from the anatomical location of the stomach.[1] HGM is the most commonly reported epithelial heterotopia and is believed to be due to congenital heteroplasia occurring during organogenesis.[2,3] HGM has been reported throughout the gastrointestinal tract, and most cases are observed in the esophagus, duodenum, and Meckel diverticulum.[4]

HGM in the rectum is rare. Studies reviewing its occurrence have reported less than 80 cases with much lesser incidence in the pediatric population.[5,6] In children, clinical features vary from incidental finding to bowel habit changes, epigastric pain, anal pain, and recurrent pancreatitis, with most commonly reported symptom being hematochezia.[7,8] Rectal prolapse has not been reported as a presenting symptom of HGM in young children.

We report a 2-year-old girl, who presented initially with rectal prolapse and hematochezia with evaluation revealing the cause to be HGM.

CASE REPORT

A 2-year-old girl was referred to gastroenterology service for evaluation of a 6-month history of painless rectal bleeding. Parents had also noticed rectal prolapse after every bowel movement that spontaneously reduced minutes after defecation. The blood was noted to be bright red in color, mixed with the stool, with occasional mucus. The parents denied painful defecation, constipation, diarrhea, or any other gastrointestinal symptoms.

Her medical history was unremarkable. Her newborn screen was negative. Her growth and development were normal. There was no family history of cystic fibrosis, any bleeding disorders, or any gastrointestinal pathology.

On physical examination, no abnormalities were noted. Specifically, her abdomen was soft and nontender without organomegaly. Perianal examination revealed no anal fissures or skin tags. Her rectal examination was normal.

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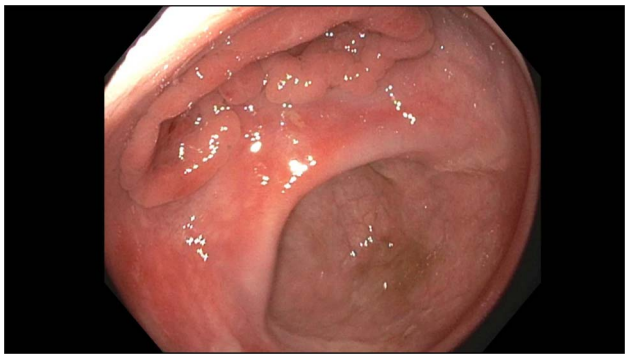

Figure 1. Flexible sigmoidoscopy #1 polypoid, nonobstructing mass (15 × 5 mm) in distal rectum.

Flexible sigmoidoscopy was performed, which revealed a sessile polypoid lesion with raised edges and a central depression with erythema in the surrounding tissue. The lesion was about 5 cm in diameter and was located in the posterior wall about 5 cm from the anal verge (Figure 1). Biopsies of the lesion revealed heterotopic gastric fundic-type mucosa with adjacent rectal mucosa. No dysplasia, cryptitis, or granulomas were noted. The tissue was negative for *Helicobacter pylori* on immunohistochemical staining. The patient was started on high doses of omeprazole with only a transient improvement in symptoms. Endoscopic mucosal resection (EMR) using Orise gel to create a submucosal cushion was performed without complications (Figure 2). The lesion was resected completely in piecemeal fashion (Figure 3). The patient has since noted complete resolution of the rectal prolapse and hematochezia on follow-up clinic visits 12 months after the EMR.

DISCUSSION

HGM in the rectum was first reported by Ewell and Jackson in 1939.[9] Since then, only a few cases have been reported in young children. Rectal prolapse has not been reported as a presenting symptom for HGM. HGM-related complications include ulcers, fistulas, or bowel perforation, and they are more common in the pediatric population.[10] The risk of malignant transformation of HGM is poorly understood, although cases of

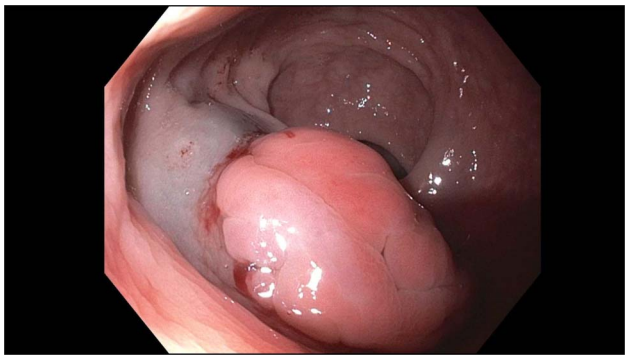

Figure 2. Flexible sigmoidoscopy #3 Orise gel used for elevation of heterotopic mucosa with submucosal-lifting gel.

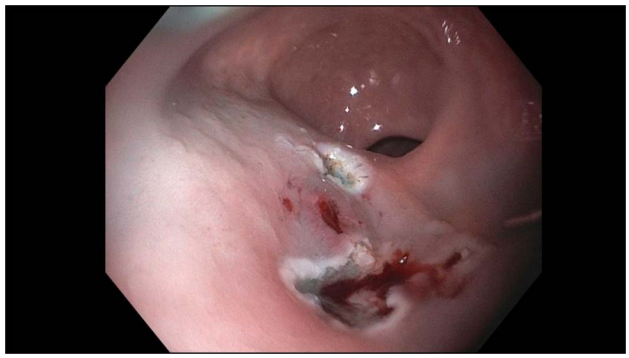

Figure 3. Flexible sigmoidoscopy #3 complete resection of the heterotopic mucosa.

underlying HGM contributing to esophageal adenocarcinoma have been reported.[11]

A systematic review of HGM in the rectum revealed that resection of the lesion was the definitive treatment in 83% of the cases.[6] Surgical resection was the treatment of choice until the 1990s, but since then, endoscopic mucosal resection has been adopted more and more into practice.[12,13] Orise gel has recently been recalled by US Food and Drug Administration because of noted increase in foreign-body reactions.[14] This gel was used before the recall in this particular patient. Other techniques including saline lift are effective in inducing submucosal lift for effective resection.[15] Other endoscopic modalities used included argon plasma coagulation[16] and endoscopic submucosal dissection.[6,16] To the best of our knowledge, this is the youngest reported case to have undergone EMR for resection of her HGM in the rectum.

In conclusion, HGM in the rectum should be considered in children with rectal prolapse and painless rectal bleeding. Considering the risk of serious complications in the pediatric population and malignancy in adults, its complete excision is recommended. Endoscopic resection can be an effective treatment option in young children with HGM in the rectum.

DISCLOSURES

Author contributions: S. D'Sa: primary author, editing and creating the manuscript. C. Mziray-Andrew: editing and reviewing of manuscript. P. Porayette: editing and reviewing of manuscript, manuscript mentor, and is the article guarantor.

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Informed consent was obtained for this case report.

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