Colonic Diverticulitis in a 19-Year-Old Boy With Juvenile Idiopathic Arthritis: Surgical Implications of Chronic Immunosuppression

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A 19-year-old boy with a history of juvenile idiopathic arthritis (JIA) formerly on Humira, methotrexate 20 mg once weekly, prednisone 5 mg bid, and naproxen 500 mg bid for several years presented to the pediatric emergency room with a 1-day history of increasing left lower quadrant pain. He had severe debilitating JIA with diagnosis at age 5, and had been taking steroid therapy since diagnosis. More recently, several other agents were added to his immunosuppression regimen. He was seen 2 months prior for similar complaints and diagnosed with diverticulitis of the descending colon on computed tomography (CT) scan. At that time, he was discharged after emergency room evaluation with 14 days of antibiotics. On this presentation, he had left lower quadrant pain but no change in bowel habits, hematochezia, or melena. He had cushingoid features with short stature, truncal obesity with striae, moon facies, and a body mass index of 20.6 kg/m^2. His height was 4 ft 9 in. and weight on admission was 43.3 kg. He was afebrile and tachycardic. His abdomen was mildly distended and was tender in the left lower quadrant. The white blood cell count was 28,300/μL. A CT scan of the abdomen and pelvis with intravenous contrast showed diverticulitis again involving the descending colon and extensive air in the left retroperitoneum with minimal free fluid. There appeared to be small diverticula in the proximal transverse colon as well, though not in the rectosigmoid (Fig. 1).

He was taken emergently for exploratory laparotomy. Upon entry to the peritoneal cavity, there was no evidence of contamination. The descending colon was, however, indurated and adherent to the retroperitoneum. It was mobilized, and this revealed a pinpoint retroperitoneal perforation with pus but no fecal contamination. The descending colon was resected and a distal transverse colostomy performed with a Hartman pouch. On visual inspection and palpation, there were no other colonic diverticula; specifically, the sigmoid colon was spared.

On gross pathologic examination, there were multiple diverticula extending into the pericolonic fat, with no transmural perforation identified. Microscopically, 1 diverticulum was associated with transmural inflammation and an intramural abscess. The pericolonic fat was inflamed and fibrotic, and there was evidence of prior perforation (Fig. 2). This pathology is consistent with false diverticula that are typically seen in the adult population.

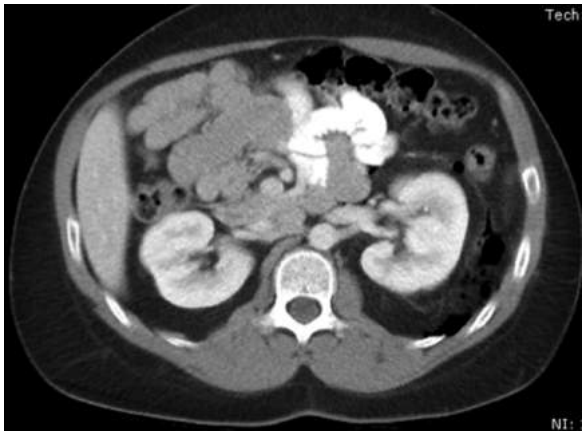

FIGURE 1. CT scan of the abdomen/pelvis with intravenous contrast demonstrating retroperitoneal free air and diverticulosis of the transverse colon.

He recovered uneventfully and was maintained on a stress-dose steroid course (initially hydrocortisone 50 mg IV followed by 30 mg q8hr, weaned to 20 mg, and then transitioned to oral prednisone). The patient was discharged home on postoperative day 9, but returned 5 days later with a large left retroperitoneal abscess. A percutaneous drain was placed by interventional radiology and removed a week later. He completed 3 weeks of IV antibiotics through a peripherally inserted central catheter line.

The ostomy was reversed 10 weeks following the initial hospitalization; however, he subsequently developed 4 intra-abdominal abscesses requiring multiple percutaneous drainage procedures during the following 4 months. He recovered from those episodes but returned with abdominal pain, and CT scan showed recurrent abscess with contrast extravasation. He was taken to the operating room for exploratory laparotomy and excision of a colonic fistula with loop ileostomy creation. He had an uneventful hospital course and was discharged home on a 7-day course of antibiotics with plans for ileostomy reversal.

DISCUSSION

Half of people older than 50 years have diverticulosis, and 10% to 15% develop colonic diverticulitis (1). In children, however, colonic diverticular disease is rare and there are few cases reported, all in association with pediatric syndromes.

Colonic diverticula are generally false diverticula as all 3 bowel wall layers are not present, in contrast to congenital diverticula, such as a Meckel diverticulum. False diverticula are defined as an outpouching of the mucosa and submucosa through the muscularis propria. This typically occurs at the penetration sites of the vasa recta through the circular muscle layer. Approximately 95% of diverticular disease is localized to the sigmoid colon, and one-third of those patients also have disease proximal to the sigmoid colon (2). This is a significant difference compared with what we report in our patient who had transverse colon diverticula and the sigmoid was spared.

In a sentinel article published in 1891, William Halsted identified the submucosal layer of the colon as the strongest layer. He did this by insufflating a segment of colon and observing that the submucosal layer was the only layer to maintain its integrity while the muscularis propria and mucosa disintegrated (3). As highlighted

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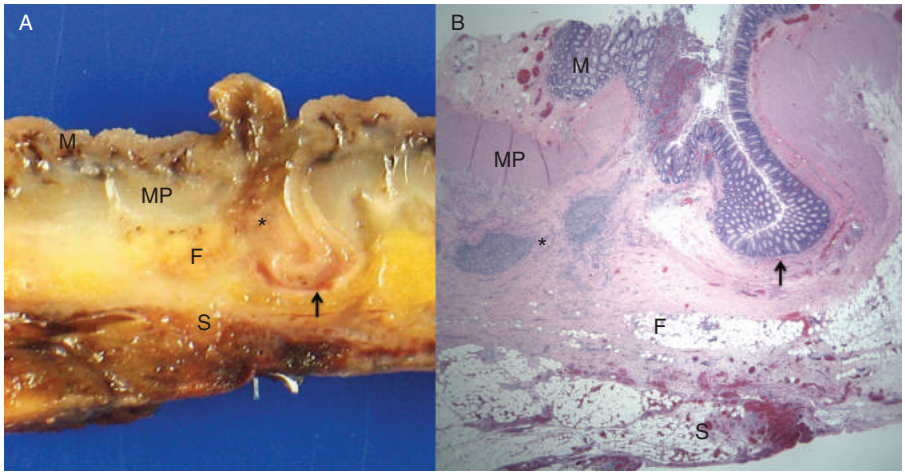

FIGURE 2. Gross and microscopic images of an inflamed diverticulum with evidence of perforation. A, Gross appearance of full-thickness section of colon demonstrating inflamed diverticulum, peridiverticular abscess, and serosal adhesions. B, The corresponding hematoxylin and eosin–stained section, micrograph taken at ×20 magnification. F = pericolonic fat; M = mucosa; MP = muscularis propria; S = serosa; asterix = peridiverticular abscess; arrow = diverticulum.

by Thomson et al (4), the collagen fibers in the submucosal layer become smaller and more densely packed as a person ages, which helps to explain why the incidence of diverticulosis in the aging population is more prevalent. These layers weaken over time and subsequently give way to the outpouching that defines the pathophysiology of this disease process. Thus, pediatric illnesses or syndromes that predispose patients to weakening of these essential layers may lead to diverticulitis.

JIA affects 1 to 22 children per 100,000 (5), and patients with JIA are typically on long-term steroid treatment or other immunosuppressant therapy. Nonetheless, there have been no reports of pediatric diverticulitis associated with JIA. Tyau et al (6) showed that in the immunocompromised adult population afflicted with acute diverticulitis, the risk of perforation was 43% compared with 14% in the non-immunocompromised population. Similarly, postoperative morbidity was higher (65%) in the immunocompromised population compared with non-immunocompromised patients (24%).

Various genetic syndromes such as Williams-Beuren syndrome, Marfan syndrome, cystic fibrosis, and Ehlers-Danlos syndrome have been associated with colonic diverticular disease in children. In Ehlers-Danlos and Marfan syndromes, changes in connective tissue may predispose to colonic diverticula, whereas in Williams-Beuren syndrome, the accelerated aging process and defect in the elastin gene increase the likelihood of developing diverticular disease during adolescence (7). Some of the same factors may contribute to the occurrence of diverticular disease in the adolescent with JIA, although this, and other disorders requiring long-term immunosuppression, has not been reported in association with pediatric diverticulitis. It is possible that the long-term high-dose immunosuppressive therapy used for JIA management may have contributed to the development of diverticulitis in this patient. In addition, this predisposed him to the perioperative infectious complications, even at a greater delay than most similar patients.

The discussion of immunosuppressant therapy and perioperative steroid treatment is also relevant to pediatric patients with inflammatory bowel disease. This remains a complex topic and the present literature regarding the associated risks of perioperative immunomodulation varies with regard to postoperative complications. Schaufler et al (8) showed that preoperative immunosuppression and steroid therapy are common for pediatric patients with colitis undergoing colectomy and, in fact, was not correlated with an increase in infectious complications. In their cohort of 51 patients, indications for colectomy were fulminant colitis in 26% and medically refractory chronic disease in 74%. Conversely, Kennedy et al (9) showed that for children undergoing total proctocolectomy with ileal pouch anal anastomosis for chronic ulcerative colitis, those treated with infliximab preoperatively were twice as likely to experience postoperative complications such as anastomotic leaks, pouchitis, and wound infection compared with those who were not on immunomodulation therapy. In a systematic review of postsurgical morbidity, Yang et al (10) demonstrated that Crohn disease patients receiving infliximab preoperatively had only a moderately increased risk of early postoperative complications, specifically infectious complications. These studies demonstrate that the impact of immunomodulation therapy on surgical outcomes is still uncertain.

CONCLUSIONS

As more children with rheumatologic and immunologic disorders are maintained on long-term high-dose immunosuppression, particularly those with connective tissue disease, they may also be at a higher risk for complicated diverticulitis. This patient illustrates that this can occur with conditions other than those described previously. The disease may not involve the sigmoid colon, which is more typical in the adult patient. Acute diverticulitis should be considered in the differential for any such patient presenting with abdominal pain. These patients are also at a higher risk for perioperative infectious complications, which may prolong hospital course and lead to readmissions and multiple subsequent procedures.

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Gastric Burkitt Lymphoma in a Teenager

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A 15-year-old teenager was referred for abdominal pain and weight loss. In the last few months, the patient was evaluated by a surgeon at the local hospital. Initial endoscopy (EGD) revealed several gastric ulcerations without the presence of *Helicobacter pylori* organism. Therapy with proton pump inhibitor was initiated. Repeated (×2) EGDs showed continuous gastritis and the patient referred to our facility.

The initial physical examination was normal with the exception of mild epigastric pain. The patient denied using nonsteroidal anti-inflammatory drugs, alcohol, or smoking. Patient acknowledged 10-lb weight loss. On EGD, gastric biopsies confirmed significant gastritis and negative *H pylori* infection. Esophagus and duodenum were normal. The patient continued with sucralfate and proton pump inhibitor for another few months. A follow-up EGD showed no mucosal improvement, but big fundal ulcer was identified. Multiple biopsies from the rim of the ulcer and stomach showed gastritis and negative *H pylori* organism. Abdominal computed tomography scan was normal except for gastric wall thickening with no lymphadenopathy. Complete blood cell count, aminotransferases, and gastrin levels were normal. Clinically the patient felt better and attended school regularly. A follow-up EGD showed a worsened, nonbleeding deep fundal ulcer with a white base (0.5 cm × 0.5 cm) (Fig. 1A). Multiple biopsies from the ulcer rim and ulcer base were obtained. A single biopsy from the ulcer base showed lymphocyte proliferation suggestive of lymphoma (Fig. 1B). Lymphocytes staining was positive for CD20 (Fig. 1C), CD10, and bcl-6 and was strongly positive for Ki-67. Immunoperoxidase antibody staining was not detected against CD3, CD10, CD21, CD43, bcl-2, MUM-1, and MYC. Fluorescence in situ hybridization (FISH) for Epstein-Barr virus was diffusely positive in the neoplastic cells. Molecular analysis indicated

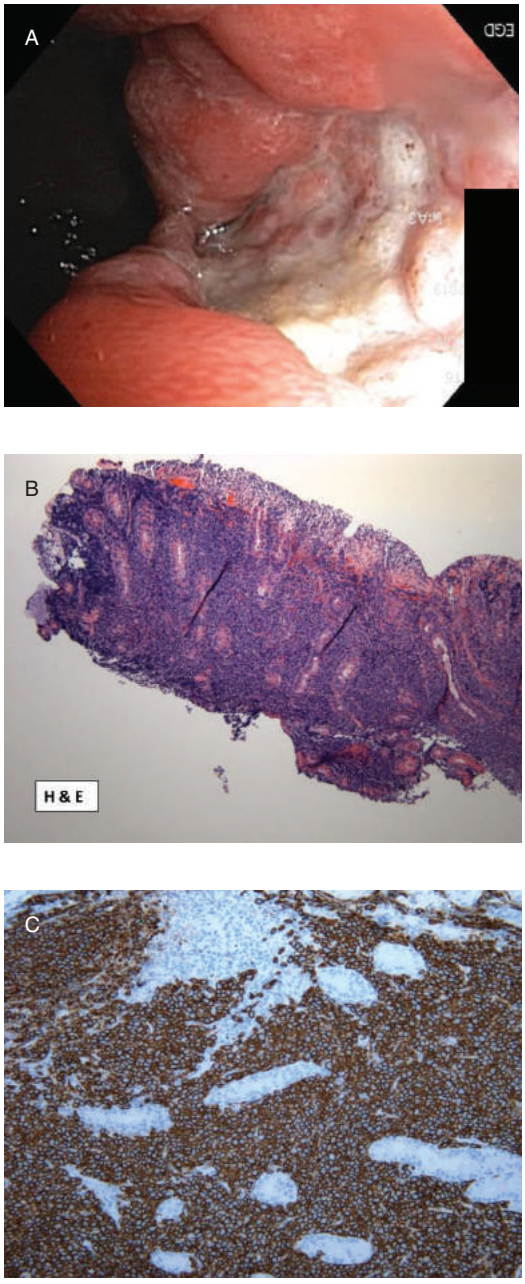

FIGURE 1. A, Deep gastric ulcer in the fundus. B, Gastric biopsy showing extensive lymphoid infiltrate within mucosa with gland destruction (H&E stain, original magnification ×10). C, Gastric biopsy illustrating that the destructive infiltrate is of B-cell lineage (CD20 immunohistochemistry, original magnification ×20).

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MYC/IGH fusion [t(8;14)] in 100% of nuclei. No abnormalities of BCL2, BCL6, or IGL were observed. The diagnosis of Burkitt lymphoma was established. Referral to the hematology–oncology team was made. Blood examination for uric acid and lactate dehydrogenase were normal. Bone marrow aspiration and spinal tap showed no extra gastric involvement. ECHO study showed normal heart. Positron emission tomography was positive for diffuse hyperactivity and thickened gastric mucosa. The patient was classified as a stage 1 localized non-Hodgkin Burkitt lymphoma. The patient started 43-day treatment protocol consisting of intravenous cyclophosphamide, adriamycin, vincristine, and oral prednisone (CHOP therapy) (1). His posttreatment imaging studies and recent endoscopy reveal no evidence of disease.

DISCUSSION

Of all non-Hodgkin lymphoma (NHL), Burkitt lymphoma in children represents approximately 20% to 30% (2); however, localized gastric Burkitt lymphoma is extremely rare in this population. In the present study, we present a primary Burkitt lymphoma that affected the gastric mucosa without the association of *H pylori* infection. In spite of multiple endoscopic evaluations of the gastric mucosa, the diagnosis was made by a lymphoid aggregation found in only 1 biopsy that was taken from the base of the gastric ulcer.

Burkitt lymphoma of the gastrointestinal tract is a rare presentation in children. In a review article of 317 cases with NHL, only 3 cases were reported with Burkitt lymphoma (3). The incidence of primary Burkitt lymphoma in the gastrointestinal tract is usually located in the cecum or small intestine while primary gastric lymphoma is extremely rare, estimated at <2% (2,4). In our case, the lymphoma was localized to the deep mucosa (in situ) at the base of the ulcer without any protrusion into the gastric lumen or involvement of other distant locations (3). Indeed, without the persistent follow-up endoscopies, the tumor in our case would likely have been missed until his disease had advanced.

The rate of gastrointestinal non-Hodgkin lymphoma (GI-NHL) in children is low and the risk factors for its development are not known. In large review studies of GI-NHL in adults, the tumors were increased in male sex, stomach localization, and the diffuse large B-cell lymphoma was the most common pathology (5,6). Burkitt lymphoma was documented in only a few patients (5). The risk factors associated with the development of GI-NHL in adults may include *H pylori* infection (5,7), celiac disease, inflammatory bowel disease, and underlying immunosuppression (5). In our case *H pylori* was negative and the patient had no history of either of those conditions. The positive Epstein-Barr virus staining and the large gastric ulceration were the only clues to the diagnosis of our patient.

The treatment and prognosis of Burkitt lymphoma are dependent on the spread of the disease with an excellent prognosis in patients with localized disease. Our investigation revealed no spread of the disease outside of the gastric mucosa, suggesting a stage 1 disease (defined as extranodal involvement or involvement of a single anatomic area (nodal), excluding the abdomen and mediastinum). The estimated 4-year event-free survival in children with stage 1 disease is around 98% and 4-year overall survival is approaching 100% (8).

In conclusion, we present a rare case of a teenager with low-stage (stage 1) localized Burkitt lymphoma of the stomach, which was histologically described as ''in situ.'' The high index of suspicion and persistent investigation lead to the right diagnosis and excellent prognosis for the patient.

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Focal Foveolar Hyperplasia: A Rare Cause of Upper Gastrointestinal Bleeding in Infancy

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Except for hypertrophic pyloric stenosis, gastric masses in infants are uncommon. Authors describe a case of a young infant who were presented with melena and anaemia because of a giant gastric polyp histologically proved to be focal foveolar hyperplasia (FFH). Since present paediatric literature is limited to only 7 case reports, these provide little insight on FFH; a review of paediatric literature is performed.

A 1-year-old-boy was admitted to our department for melena. The child was well until 3 days before the admission when he had 2 episodes of nonbilious vomiting. Physical examination revealed a pale child. Laboratory data were as follows: haemoglobin 8.5 g/dL, mean corpuscular volume 81.9 fL, elevated transaminases and lipases, and normal bilirubin and inflammatory indices. Faecal occult blood test was strongly positive.

Abdominal ultrasound (US) showed a thickness of the gastric antrum wall extending beyond the pylorus (Fig. 1), terminating in a round structure below the gallbladder. Upper gastrointestinal (GI)

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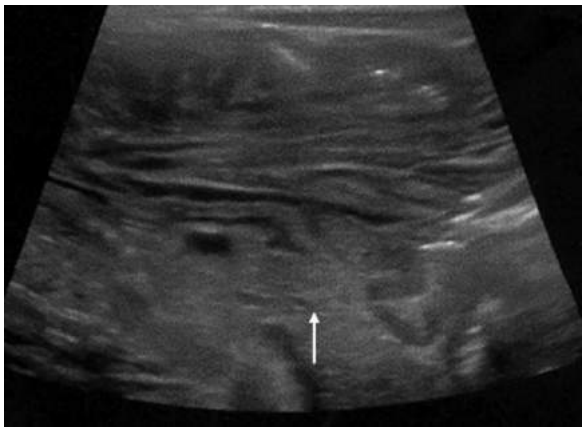

FIGURE 1. US scan shows a thickened wall in the antral-pyloric region (arrow) extending beyond the pylorus.

contrast study revealed a massive polypoid filling defect in the antrum. Transit time through gastroduodenal junction was slightly prolonged.

Computed tomography (CT) confirmed a large oblong mass, with inhomogeneous enhancement after contrast medium, extended through the pylorus up to the base of the duodenal cap causing mild dilatation of the main pancreatic duct, the common bile duct, and gallbladder over distension.

At upper GI endoscopy a giant polypoid lesion arising from the anterior gastric wall was detected. Because endoscopic polypectomy was considered not feasible owing to the mass size, at the end of endoscopy the child was taken to the operative room for surgery.

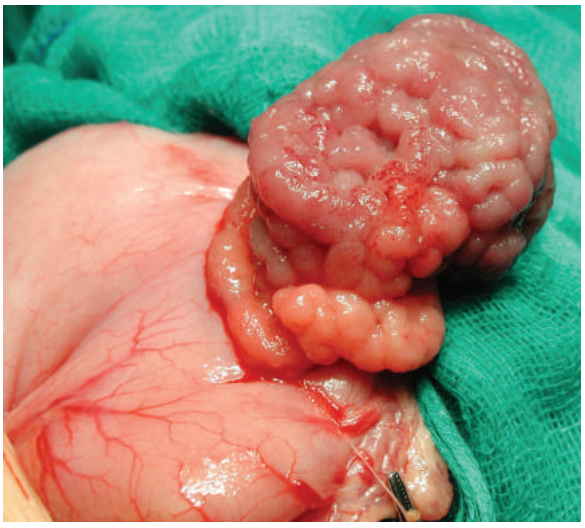

FIGURE 2. Laparotomy reveals a polypoid mass with haemorrhagic areas on the surface.

At laparotomy a large polypoid mass, of enlarged tortuous mucosal–type tissue, measuring 3 cm × 4 cm × 4 cm, with haemorrhagic areas on the surface, was found and resected (Fig. 2). Histological analysis revealed mucosa related to antral-angular area with no architectural distortion of glands and presence of FFH. In the lamina propria, some glandular structures were surrounded by mixoid stroma and muscle bundles belonging to muscularis mucosae, whereas other glands showed cystic ectasia filled with several foamy histiocytes ($CD68^{+}$) (Fig. 3). Considering

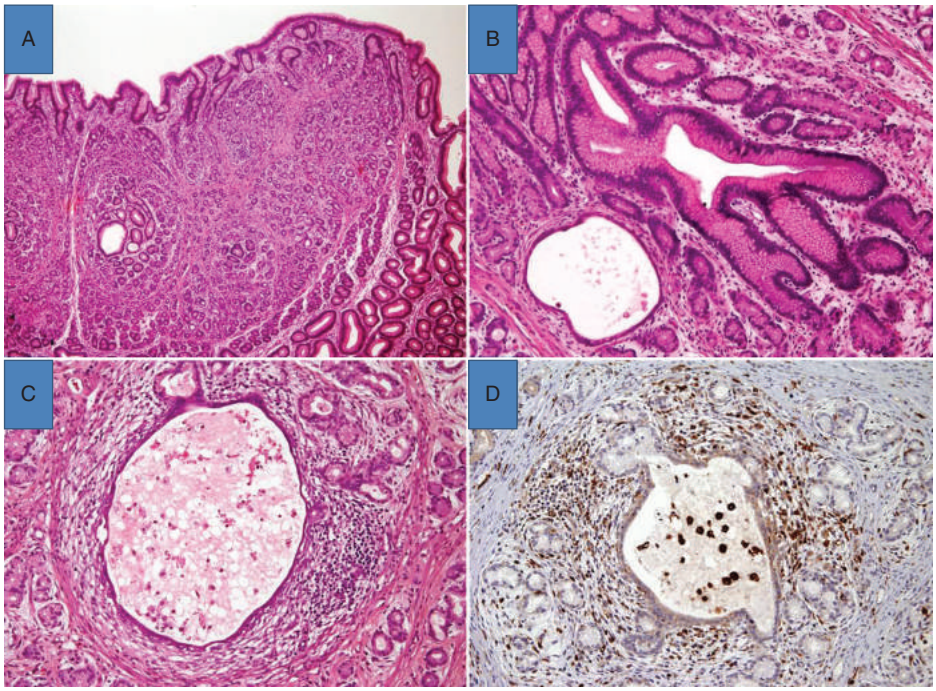

FIGURE 3. Photomicrograph of FFH. Mucosa related to the antral-angular region with no architectural distortion of the glands and the presence of focal hyperplasia limited to the foveolae (A and B: H&E). In the lamina propria, some glandular structures were surrounded by muscle bundles belonging to muscolaris mucosae, whereas others glands showed cystic ectasia filled with several foamy histiocytes (C: H&E; D: immunostain for CD68). FFH = focal foveolar hyperplasia.

TABLE 1. Review of the studies on paediatric FFH

References	Patient sex, age	Symptoms	Diagnostic tools	Diagnosis	Location	Size	Therapy	Outcome
Katz et al (1)	Boy, 7 wk	Vomiting Failure to thrive	GI contrast study Endoscopy	FFH	Greater gastric curvature extending into the lumen of antrum and pylorus	2.4 cm × 1.2 cm × 0.7 cm	Partial antrectomy, Billroth1 gastroduodenostomy	Symptoms resolution
McAlister et al (5)	Boy, 31 wk	Emesis Intermittent abdominal distension Weight loss	GI contrast study Sonography Endoscopy	FFH	Antrum	—	Surgical resection	Vomiting resolution Dead for bronchospasm, neurological deterioration
Holland et al (3)	Boy, 6 wk	Vomiting and weight loss in Pierre-Robin syndrome	GI contrast study Sonography Endoscopy	FFH	Antrum	—	Surgical resection Pyloroplasty Nissen fundoplication	Symptoms resolution
	Girl, 13 wk	Intermittent projectile gastric vomiting	Sonography GI contrast study	FFH	Antrum-pylorus	—	Surgical resection	Symptoms resolution
Morinville et al (4)	Boy, 7 wk	Gastric vomiting	Sonography GI contrast study Endoscopy with biopsy	FFH	Greater curvature side of the pyloric channel	1 cm	Surgical resection	Symptoms resolution (necessity of hydrolysate formula and soy formula)
Epifanio et al (6)	Boy, 42 wk	Vomiting Feeding refusal Weight loss Slight dehydration	GI contrast study Sonography and colour-Doppler Endoscopy with biopsy	FFH	Antrum-pylorus	—	Endoscopic polypectomy in 2 stages	Symptoms resolution
Perme et al (7)	Girl, 41 wk	Nonbilious gastric vomiting in 22q11.2 microdeletion	Sonography	FFH and hypertrophic pyloric stenosis	Antrum Pylorus	—	Pyloromyotomy	Symptoms resolution after surgery and interruption of Prostaglandin therapy
Our case	Boy, 1 y	Melena Anaemia	Sonography GI contrast study CT Endoscopy	FFH	Antrum	3 cm × 4 cm × 4 cm	Surgical resection	Symptoms resolution

CT = computed tomography; FFH = focal foveolar hyperplasia; GI = gastrointestinal.

morphological features, diagnosis was consistent with polypoid mucosal fold characterised by FFH.

Postoperative course was uneventful. At US performed before discharge, dilatation of the main pancreatic duct and of the common bile duct was no more detected. At 1-year follow-up the patient was healthy, with satisfactory weight gain, normal blood tests, and unremarkable upper GI endoscopy findings.

DISCUSSION

FFH is a nonneoplastic mucosal gastric polyp first described in 1985 (1). Normally gastric epithelium is formed by glands and crypts covered by foveolar cells (2). FFH is characterised by elongation and dilation of gastric foveolae that become tortuous, giving papillary appearance to the intervening crests, finally resulting in a broad-based polyp. Redundant mucosa produces a sausage-shaped structure that arises from a narrow, elongated base, commencing in the distal portion of the antrum and sometimes extending through the pylorus to the base of the duodenal cap (3).

FFH is rare in children. To the best of our knowledge, up to now, only 7 cases have been reported in paediatric literature (Table 1) (1,3–7). Patients mean age at diagnosis was 21 weeks with male-to-female ratio of 5:2.

As lesions were located mostly in the gastric antrum, nonbilious vomiting and weight loss were the main presenting symptoms, which are typical of gastric outlet obstruction but not pathognomonic of FFH. Including our case, GI-tract bleeding (presenting with emesis and melena) was observed only in 2 studies, and the bleeding can be related to mucosal erosions owing to intussusception through the pylorus (Table 1). Furthermore, in our patient, because of the large size of the protruding polyp up to the second duodenum, obstruction of papilla of Vater occurred with dilatation of the main pancreatic duct and of the common bile duct together with gallbladder over distension and abnormalities of liver function tests.

US appearance of FFH shows typical pattern characterised by a thickened wall in the antral-pyloric region extending into the pyloricduodenal lumen (5,6), keeping in mind this pattern can be useful for a right preoperative diagnosis. Upper GI tract endoscopy is mandatory to better visualise the mass, its origin, extension, and size, to perform its resection and to rule out other possible causes of upper GI bleeding. Regarding morphological differential diagnosis, benign and malignant gastric neoplasms were considered in our case.

Among benign lesions (adenomyosis, hyperplastic polyp, and inflammatory fibroid polyp) no architectural abnormalities of gastric wall or of mucosal layer were found in our case. Considering malignant processes, glandular epithelium did not show cytological atypia related to adenomatous polyps or adenocarcinomas. Also muscular layers were normal without any feature related to smooth muscle–derived neoplasms (leiomyomas or leiomyosarcomas). Neither any characteristics of vascular proliferation, lymphoproliferative disease, teratomas, or ectopic tissues were found.

All of the patients, except 1, underwent resection of FFH (6 by surgery, 1 by 2-stage endoscopy). Size of FFH was reported only in 2 patients (1,4), beyond our case. After removing the mass, symptoms disappeared in 6 patients, whereas in 1, presenting cow's-milk protein hypersensitivity, symptoms disappeared after resection of the lesion associated with introduction of hydrolysate formula and soy formula. In 1 case, reported by Perme et al (7), FFH appeared simultaneously with infantile hypertrophic pyloric stenosis. Both entities were present as an adverse effect of prostaglandin E1 therapy administered because of the presence of cardiovascular abnormalities in 22q11.2 microdeletion syndrome. In this case, symptom resolution occurred after pyloromyotomy was performed, whereas FFH resolved spontaneously after cessation of prostaglandin treatment.

We believe that we must consider the role of endoscopic resection for these patients instead of surgery. As FFH arises from gastric mucosa, the risk of perforation at endoscopic resection is lower than that for other gastric polyps arising from submucosa. Incomplete endoscopic resection will probably not increase the risk of local recurrence as, although the aetiology of infantile FFH is unknown, it has been supposed that it may represent a hyperregeneration of gastric mucosa under injurious influence (8) as cow's-milk hypersensitivity (4) and prostaglandin therapy (7,9).

Eventually our case focuses on some aspects: although FFH is uncommon, paediatric gastroenterologist and surgeons should consider FFH in differential diagnosis above all in infants who present with GI bleeding; US may be an invaluable tool to identify characteristic pattern of FFH, avoiding more invasive examination. If a correct preoperative diagnosis is made, size of the lesion could not be more considered a contraindication to FFH endoscopic resection.

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