ADULT INTUSSUSCEPTION SECONDARY TO A GIANT ILEAL HAMARTOMA: A CASE REPORT

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Abstract

Significance

Adult intussusception is rare with an incidence of 2-3 cases per million per year. In contrast to children, it's presentation is subacute or chronic with predominant symptoms of partial intestinal obstruction. An identifiable lead point is found in majority of cases that often leads to surgical management.

Case Presentation

We present a case of a 39-year-old Filipino male who complained of 3-years history of recurrent periumbilical pain, characterized as intermittent, crampy, non-radiating, lasting for 30 minutes with a pain scale of 6/10. This was accompanied by episodic diarrhea and progressive weight loss (10kg). He was previously seen by 3 physicians and managed as Irritable Bowel Syndrome and Inflammatory Bowel Disease. Workups included: Normal CBC, ESR, CRP and fecalysis; CECT of Abdomen (2015): Hepatic hemangioma; EGD (2016): Antral Gastritis; Upper GI Series: segmental ileus. He already underwent 3 colonoscopies for a span of 3 years with the following results: colitis, cecum and ascending colon (2017); colitis, transverse colon (2018), Ilietis (2019). Treated with PPI, antibiotics, otolinium bromide, prednisone and mesalazine with only partial relief of symptoms. Physical examination revealed fullness over hypogastric area with tenderness on deep palpation.

Management

An initial impression of Ileal Crohn's disease was entertained. Magnetic resonance enterography was requested which revealed Ileo-colic intussusception with long segment telescoping of the terminal ileum into the descending colon. Patient was then referred to surgery for exploratory laparotomy. OR findings were a grossly visible ileoileal intussusception 20 cm from the ileocecal valve. Bowel segments were viable. On palpation, a 5.5 x 0.7 x 3.2 cm pedunculated polyp with a 4.0 x 3.0 x 1.5 cm cm stalk was noted. Segmental Ileal resection was done followed by end to end anastomosis. Histopathology showed hamartomatous polyp with gastric and pancreatic heterotropia and with focal hemorrhagic infarction.

Recommendation

A high index of suspicion and cross-sectional imaging plays a crucial role in the diagnosis of adult intussusception, especially in atypical cases of chronic, recurrent abdominal pain. Surgical exploration with the principle of resection without reduction remains the mainstay of treatment.

Keywords: case report, adult intussusception, giant ileal hamartoma

Introduction

Adult intussusception is rare with an incidence of 2-3 cases per million per year¹. It accounts for 1%-5% of all cases of intestinal obstruction in adults⁴. In contrast to children, it's presentation is subacute or chronic with predominant symptoms of partial intestinal obstruction. An identifiable lead point is found in majority of cases that often leads to surgical management.

Case Report

We present a case of a 39-year-old Filipino male who complained of 3-years history of recurrent periumbilical pain, characterized as intermittent, crampy, non-radiating, lasting for 30 minutes with a pain scale of 6/10. This was accompanied by episodic diarrhea and progressive weight loss (10kg). He was previously seen by 3 physicians at different institutions and managed as Irritable Bowel Syndrome and Inflammatory Bowel Disease. Workups included: Normal CBC, ESR, CRP and fecalysis; CECT of Abdomen (2015): Hepatic hemangioma; EGD (2016): Antral Gastritis; Upper GI Series: segmental ileus (Figure 1). He already underwent 3 colonoscopies for a span of 3 years with the following results: colitis, cecum and ascending colon (2017); colitis, transverse colon (2018), Ilietis (2019) – Figure 2. Treated with PPI, antibiotics, otolinium bromide, prednisone and mesalazine with only partial relief of symptoms. Patient opted to transfer to our institution for 4th opinion. Physical examination revealed fullness over hypogastric area with tenderness on deep palpation.

An initial impression of Ileal Crohn's disease was entertained. Magnetic resonance enterography was requested which revealed Ileo-colic intussusception with long segment telescoping of the terminal ileum into the descending colon (Figure 3). Patient was then referred to surgery for exploratory laparotomy. OR findings were a grossly visible ileo-ileal intussusception 20 cm from the ileocecal valve (Figure 4). Bowel segments were viable. On palpation, a 5.5 x 0.7 x 3.2 cm pedunculated polyp with a 4.0 x 3.0 x 1.5 cm stalk was noted almost completely obstructing the lumen (Figure 5). Segmental Ileal resection was done followed by end to end anastomosis. Histopathology showed hamartomatous polyp with gastric and pancreatic heterotropia and with focal hemorrhagic infarction (Figure 6). Tumor size was 5 cm in in widest dimension and was negative for dysplasia. Surgical margins were viable and all five recovered lymph nodes were unremarkable. Post-operative course was unremarkable and patient was discharged on the 5th post-operative day.



Figure 1. Upper GI Series showing non-dilated air filled bowel loops with delayed transit of contrast through the small intestines may be suggestive of segmental ileus



Figure 2. Colonoscopy showing non-specific ileitis. A.) terminal ileum and B.) ileocecal valve



Figure 3. Magnetic Resonance Enterography showing ileo-colic intussusception with long segment telescoping of terminal ileum into cecum, ascending, transverse and descending colon. A.) coronal view and B.) axial view



Figure 4. OR findings: on opening, noted a grossly viable intussusception around 20 cm from the ileocecal valve



Figure 5. Postoperative gross specimen showed a 5.5 x 0.7 x 3.2 cm pedunculated polyp with a 4.0 x 3.0 x 1.5 cm stalk



Figure 6. Histopathology showing a hamartomatous polyp with gastric and pancreatic heterotropia and with focal hemorrhagic infatrction. A.) Small intestinal mucosa showing congestion and edema B.) Polypoid mass is composed of fibroadipose tissues and tubular to cystically dilated pyloric glands and nests of pancreatic acinic C.) Nests of pyloric glands and pancreatic acini with prominent arborizing bands of smooth muscle D.) Higher magnification of the glands which show basally located nuclei with abundant eosinophilic cytoplasm

Discussion

Intussusception refers to the invagination or telescoping displacement of a proximal segment of bowel (intussusceptum) into an adjacent distal segment (intussuscipiens). It accounts for 1% to 5% of all cases of intestinal obstruction in adults¹. In contrast to children, small bowel intussusception in adults is associated with an underlying pathologic process that serves as the lead point in 70-90% of cases. This includes benign tumors (e.g. polyps, lipoma, Peutz–Jegher type polyps), neoplasms (e.g. adenocarcinoma, lymphoma), postoperative changes, adhesions and miscellaneous conditions like lymphoid hyperplasia and Meckel's diverticulum².

Preoperative diagnosis in adult cases is infrequent due to its varying presentation, but may manifest with acute, intermittent or chronic symptoms. The mean duration of symptoms is 7 days, usually presenting clinically from 2 weeks to several months in most cases, and others reaching up to 5 years⁴. Clinical presentation of intussusception is nonspecific. The predominant symptoms are those of partial intestinal obstruction, where the nature of abdominal pain is periodic, intermittent and crampy. Other signs and symptoms such as nausea, vomiting, constipation, fever, hematochezia, diarrhea, weight loss and a palpable abdominal mass are less frequent.

In terms of diagnosis, CT scan is the most useful imaging option with a diagnostic accuracy of 58% to 100%. Characteristic findings include an early target mass with enveloped, eccentrically located areas of low density, which may appear as target sign or sausage shaped mass. More importantly, it defines the location, nature of the mass, its relationship to surrounding tissues and staging in the cases of suspected malignancy⁴. Ultrasound can also be used in diagnosis where the classic target sign on the transverse view can be seen². The classic features of intussusception on plain film of the abdomen are the air crescent sign and presence of a soft tissue mass with decreased colonic air which are neither sensitive nor specific¹.

Surgical exploration remains the mainstay of treatment. The principle of resection without reduction is well established. The high likelihood of malignancy in colonic intussusception justifies resection without reduction. In small bowel intussusception, a more selective approach may be feasible, although resection is advocated unless a benign lesion has been previously confirmed. But in the majority of cases, the inability to differentiate benign from malignant etiologies, signs of bowel ischemia and the possibility of perforation should be considered⁴.

Although the preoperative diagnosis was unclear, the critical aspect of diagnosis for this case was our decision to repeat the cross-sectional imaging, even though the patient already had previous negative work-ups the previous years. This is due to the fact that the hamartoma was just small and grew over time. Only a few cases of intussusception secondary to a hamartomatous polyp have been reported in the literature.

Conclusion

A high index of suspicion and cross-sectional imaging plays a crucial role in the diagnosis of adult intussusception, especially in atypical cases of chronic, recurrent abdominal pain. A correct and timely diagnosis is necessary to prevent the complications of bowel infarction. Surgical exploration with the principle of resection without reduction remains the mainstay of treatment.

References

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