A Rare Case of Perforated Tubular Ileal Duplication in 72-Year-Old Male

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Abstract Enteric duplication is a rare anomaly, which is common in pediatric population, and it is very rare in adults. Enteric duplication can occur anywhere from mouth to anus. Ileal duplication is the most common (30 %) followed by duodenum, stomach, jejunum, colon and rectum. Though most cases are presented within the first 2 years of life, some may present at later age. Here we report a case of a 72-year-old male presented to us in emergency as acute abdomen, which was presumptively diagnosed as ca rectosigmoid, but later the patient developed perforative peritonitis. On exploratory laparotomy he was diagnosed as a case of perforated ileal duplication cyst. Though these lesions are very rare, the importance of enteric duplication cysts lies in the fact that they can mimic many disease conditions and may be left untreated.

Keywords Ileum · Intestinal duplication · Perforation

Introduction

Duplication cysts are a rare group of anomalies which vary in their presentation. Often they present in a very nonspecific way like vague pain in the abdomen or lump in the abdomen. Rarely, they present as intestinal obstruction, perforative peritonitis, malignancy, melena, intussusception and constipation. There are different theories regarding their origin but none are proven yet. Enteric duplication can occur along the entire length of alimentary tract. They are located along the mesenteric border of the intestine and are lined by smooth muscle. Heterotopic gastric mucosa often lines

duplication cyst, and its inflammation is responsible for melena, perforation and intestinal obstruction. Enteric duplication can be a diagnostic dilemma for those who are involved in its management. Often diagnosis is made on the operating table as in our case.

Case report

A 72-year-old Indian male came to us with history of pain in the abdomen for 3 months, and vomiting and loose motions for 10 days. On admission, it was found that the patient had tenderness in the left iliac fossa and hypogastric region, and he was febrile with raised leukocyte count. X-ray erect abdomen was normal. On ultrasonography, it was found that the patient had thickened wall of the rectosigmoid junction along with few thickened ileal loops. Patient's CT scan abdomen was done, which showed rectosigmoid wall thickening and the hypoechoic mass attached to the rectum on the right side. Barium enema showed the apple core sign likely to be due to malignancy, but on colonoscopy all large colon found to be normal.

Decision was taken for elective laparotomy, but a day before elective surgery the patient developed severe pain in the abdomen with rigidity. X-ray erect abdomen showed air under the right side of the diaphragm, so emergency laparotomy was done.

During exploration, we found ruptured small bowel with inflammatory bowel loops adhered to the rectosigmoid region in the left iliac fossa. On separating adhesions, we found interesting finding: a 10 cm duplicated ileal loop, which was perforated. On opening its anterior wall, it was found to be communicating both proximally and distally (Figs. 1, 2 and 3) Segmented bowel resection and end-to-end anastomosis was performed. Patient's postoperative recovery was uneventful.

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Fig. 1 Ileal duplication (arrow) with perforation on exploratory laparotomy

Discussion

Reginald Filtz [1] first used the term ‘intestinal duplication’ in 1844. Intestinal duplication anomalies are generally common in pediatric population within the first 2 years of life [2]. About 2/3 cases are detected within the first 2 years. This finding is very rare in adult population.

Several theories are proposed for the pathogenesis of intestinal duplication. According to split notochord theory, splitting of the notochord is a primary event during embryological growth responsible for duplications. Bentley and Smith in their landmark article described the concept of the split notochord and found a spectrum of disorders that share a common embryogenesis, which includes enteric duplication along with vertebral anomalies [3]. Other theories are persistent vacuolation theory, faulty separation of the endoderm and notochord, localized forms of twinning, etc [4].

Duplication cysts can be found anywhere from mouth to anus but is most common in the small intestine. Ileal duplication is the most common (30 %) followed by duodenum, stomach, jejunum, colon and rectum [5]. There are two forms

Fig. 2 Wall of intestinal duplication is cut opened to show common sharing of wall (arrow) between duplication and normal intestine; Ryle's tube is within normal ileum

Fig. 3 Diagrammatic representation of ileal duplication

of duplications: spherical or cystic duplications and tubular duplications. Cystic duplications (75 %) are more common than tubular duplications (25 %) [6]. These hollow structures possess a well-developed smooth muscle wall and are lined by intestinal mucosa. Often heterotopic tissues like gastric mucosa or pancreatic tissue are present in the wall of them. The length of tubular duplications varies from few centimeters up to as long as the whole length of the small intestine. Tubular duplication often communicates with the main intestine proximally or distally or at both ends like in our case. These duplications share common wall and blood supply.

Intestinal duplications are very difficult to diagnose preoperatively. They present with myriad of symptoms. The symptoms are often related to location of duplication. Most cases are detected during early years of life; as far as we know there are very few cases in the literature which are diagnosed at old age in adults. The most common clinical presentation in adults is vague abdominal pain and dyspepsia. Other less common presentations are intestinal obstruction, perforative peritonitis, malignancy [7], melena [8], intussusception and constipation. Acid secreting heterotopic gastric mucosa often causes peptic ulcer disease, which leads to perforation and peritonitis.

Diagnosis of intestinal duplication is often a challenging task. Most of the time it is not possible to diagnose preoperatively or misdiagnosis is made like in our case and diagnosis is made on the operating table. Various radiological modalities like ultrasound and CT scan are helpful in making diagnosis. Ultrasound is important in case of cystic duplication but is often inconclusive in tubular duplications [9, 10]. Barium enemas can be helpful in few cases but most of the time is not much helpful. ^{99}Tcm -pertechnetate scanning is a reliable ^{99}Tcm -pertechnetate, noninvasive method to diagnose ectopic gastric mucosa [11, 12].

Most authors believe that treatment is required in symptomatic patients and those who present with complications. Intestinal resection and anastomosis is required in all cases and include adjacent normal intestine because of common sharing of bowel wall and of blood supply [13, 14]. Postoperative outcomes are often favorable. Though there are some

case reports of malignancies in ileal duplication, these findings are rare [2].

To conclude, ileal duplications are rare anomalies in adults, which are often undiagnosed preoperatively and can present as perforative peritonitis, intestinal obstruction, bleeding and malignancy.

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