Case Report

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Ileocecal Intussuception Caused by Inflammantory Fibrionid Polyp of Ileum: Case Report and Review of Literature

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ABSTRACT

Introduction: Inflammatory fibrinoid polyps or Vanek's tumor, are rear, uncommon, non-neoplastic cause of intussusception of gastrointestinal tract in adults.

Case Presentation: We report a 56-year-old woman with 30-day hystory of intermittent abdominal pain associated with flatulence. Abdominal examination showed diffuse tenderness to palpation, especially in the right lower quadrant of the abdomen with a muscular defense. By digito-rectal examination the presence of feces, mucus even blood was not detected. Abdominal X-ray examination of the abdomen showed obstruction of the small intestine with multiple aerocolic levels positioned centrally with no signs of pneumoperitoenum. Abdominal ultrasound showed a hypoechoic, circular tissue mass 90 x 50 mm in diameter at the end of the dilated terminal ileum, and also showed hyperechoic focal liver lesion in diameter 55 x 50 mm. After preoperative preparation urgent transumbilical laparotomy was done and the ileocecal intussusception was found. It is preformed right colectomy with ileo-transvezo anastomosis. The segment of bowel containing the lesion was resected. Macroscopically examination of resected segment, showed tissue lesion, diameter of 115 x 52 mm. Histopathological analysis showed lesion with spindle cells and an mixed inflammatory infiltrate with many eosinophils. Immunohistochemistry confirmed the diagnosis of ileal Vanek's tumor.

Conclusion: Although inflammatory fibroid polyps are rare, benign, uncommon, and non-neoplastic cause of intussusception in adults, in the case of intestinal obstruction the only solution is a surgical approach and resection of the intestinal segments involved.

Keywords: Inflammatory Fibroid Polyps; Vanek's Tumor; Ileocecal Intusscusception

Introduction

Inflammatory fibrinoid polyps (IFPs) or Vanek's tumor are rear, uncommon, localized and non-neoplastic cause of intussusception of gastrointestinal tract [1-5]. This lesion was first reported by Vanek in 1949 as eosinoflic submucosic granuloma on series of six cases, who were localized in a stomach [1,2,4,5]. These lesions have been referred to a variety of names, including eosinopltilic granuloma, granuloma with eosinophils, inflammatory pseudotumor, gastric fibroma with eosinophilic infiltration, eosinoplfilic gastroenteritis, polyp with eosinophilic granuloma [3,4]. IFPs can develop in various part of gastrointestinal tract, and they usually represent like solitar lessions [3-5]. The most common localization are gastric antrum and ileum rarely in the duodenum and jejunum [3,5,6,7-9]. These tumors affect both sexes and age groups with incidence pic between fifth and seventh decades, with similar male to female ratio [3,4,7,9,10]. The majorities of Vanek's tumors represents as asymptomatic lessions and were usually discovered as incidental findings during endoscopy or surgery performed for another reasons [10]. Usually, their size determines whether they are symptomatic or not [5,9,11]. The most common symptoms are usually associated with abdominal pain, dyspeptic symptoms, loss of weight, iron deficency anemia and intussusception [1,3,4,5,12]. Macroscopically inflammatory fibroid polyps appear as sessile or perpendicular polypoid lesions, usually of 0.2-20 cm in diameter [3]. Histologically they are mainly confined in the submucosa and characterized by vascular and fibroblastic proliferation together with an inflammatory response [1]. Endoscopic excision is the treatment of choice [3,6,11]. Only rarely caces require urgent surgical intervention. If this tumor arising below the Treitz ligament they can present with an acute abdomen usually like intussusceptions [3]. In these cases, exploratory laparotomy or laparoscopy are frequently recommended as the best treatment. The operation should be performed as early as possible in order to prevent the intussusceptions from leading to ischemia, necrosis and subsequent perforation of the invaginated bowel segment [3-4]. These tumors do not recure after complete surgical or endoscopic resection [5]. In this case report, we present a rare case of 56-year-old woman with ileocecal intussusception due to IFP in ileum.

Case Report

56-year-old women was referred for examination, with 30-day hystory of intermittent abdominal pain associated with flatulence. In the available medical records, fecal occult blood test is positive, and heteroehogen focal liver lesion was observed in abdominal ultrasound. In the physical examination pain sensitivity in the ileocecal area without muscular defense is dominated. After examination we indicated hospital testing for complete diagnosis (laboratories, abdominal ultrasound, gastroscopy, colonoscopy, etc.). Eleven days after the examination, the patient comes as an emergency case because of severe abdominal pain, nausea and urge to vomit. Abdominal examination showed diffuse tenderness to palpation, especially in the right lower quadrant of the abdomen with a muscular defense. Digito-rectal examination is not detected the presence of feces, mucus and blood. Laboratory analysis showed only WBC 10.3 Hgb 98 g/l, CRP 90, 1181 amylase, lipase 22 U/l. Other laboratory parameters were within normal limits. Abdominal X-ray examination of the abdomen showed obstruction of the small intestine with multiple aerocolic levels positioned centrally with no signs of pneumoperitoenum (Figure 1). Abdominal ultrasound showed a hypoechoic, circular tissue mass 90 x 50 mm in diameter at the end of the dilated terminal ileum. Abdominal ultrasound is also showed hyperechoic focal liver lesion in diameter over 55 x 50 mm. After preoperative preparation urgent transumbilical laparotomy is done and the ileocecal intussusception was found (Figure 2). The right hemicolectomy with ileo-transvezo anastomosis is performed. Macroscopically examination of resected segment at the junction of the small intestine to the colon, showed defect in the wall which the protruding tissue, diameter of 115 x 52 mm (Figure 3). The tissue has a solid structure, with mottled reddish fields. The segment of bowel containing the lesion was resected, and continuity of the bowel was restored by end-to-end anastomosis. Microscopically it is a submucosal proliferation, with newly created network of blood vessels, predominantly constructed from stromal elements in a very edematose stroma. There was a mixed inflammatory infiltrate (neutrophils, lymphocytes, and plasma cells) with the presence of focal tissue eosinophilia. The stromal elements contained in the «onion skin" schedule and in some places on the surface there are ulcerative proliferation. On immunohistochemistry, immunophenotyping detected positive immunoreactivity for CD 34, negative for CD 117, actin-alpha SMA, desmin, S-100 protein, and Bcl-2. Proliferative status Ki-67 was less than 0.1%. Described histological figure corresponds with inflammatory fibroid polyp or Vanek tumor. The postoperative course complicated with intermittent fever up to 39 °C, and the infection of wound with the genus Klebsiella, which were treated with antibiotics according to the antibiogram. On the eleventh day of hospitalisation the patient was discharged in good general condition. Additional diagnostics performed with ambulante regimen. Standard biochemical parameters (blood test results, urea, hepatogram, immunology, virusology, tumor markers) were within normal ranges. Hyperechoic focal liver lesion in the liver, 54 x 47 mm in diameter was diagnosed as a hemangioma of the liver on MRI scan of the abdomen. The patient in next six months after the surgery is without problems. Follow-up colonoscopy performed 12 months after surgery showed no signs of any pathology within the site of resection.

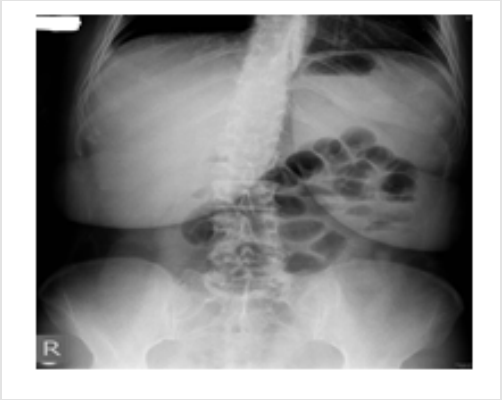

Figure 1: X ray of abdomen shows obstruction of the small intestine with multiple aerocolic levels positioned centrally with no signs of pneumoperitoenum.

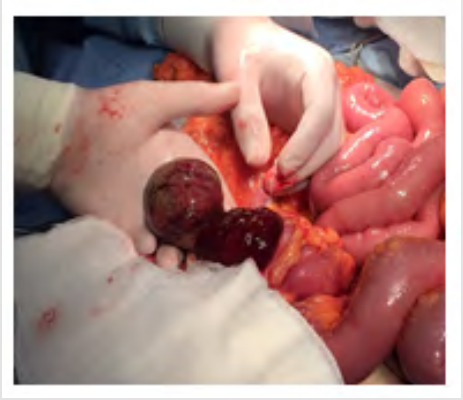

Figure 2: The view of IFP of ileum before resection.

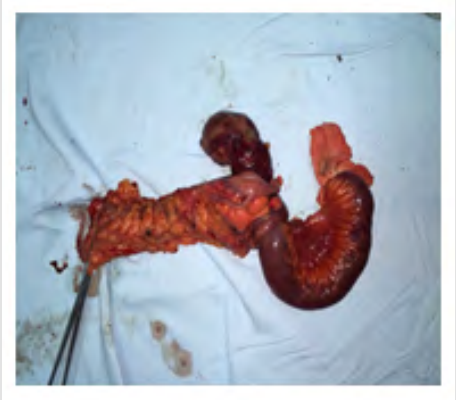

Figure 3: Resected ileocecal intussusception and IFP of ileum after resection.

Discussion

Review of literature showed 85 cases of intussusception caused by Vanek's tumor [13]. The patients were aged from 4 to 81 years, half of which were women (mean 46 + 17.5 years). In relation to the localization of intussusception in 63 was in the ileum, jejunum in 17, three had a colon and two ileo jejunal localization [13]. In our patient intussusception were caused by IFP in the terminal ileum. Vanek's tumor in our patient was size 115 x 52 mm in diameter and is the largest on this localization.

Intussusception was defined as intussusception of the proximal part of the small intestine along its mesentery in an adjacent segment, which leads to disorders of bowel function, obstruction and possible vascular compromise [13]. Intussusception were described for the first time in 1674 from Barbette, and later in 1789 from Hunter [13]. According to localization is divided into enteroenteric, ileocolic (ileocecal) and colocolic. Etiological were divided into primary (idiopatic) or secondary (benign or malignant) [13]. Only 1- 5% of cases of small-bowel obstruction were caused by intussusception [13]. In the small intestine intussusception usually caused by benign tumor in 90% of cases (hamartoma, lipoma, adenoma, IFP, adhesions, trauma, lymphoid hyperplasia) [3]. In the colon more frequently is malignant etiology 43-80% (adenocarcinomas, adenomas, lipomas, endometriosis) [3,4,12,14]. In one case it is described as a rare form of double invagination intussusception of the small bowel caused with IFP [10]. IFPs are a rare, idiopathic pseudotumors of gastrointestinal tract which were first described by Vanek in 1949 as a submucosal eosinophilic granuloma [15]. Vanek in the first plan put submucosal inflammatory nature and origin of the tumors [1]. The term IFP suggested by Helwing and Ranier 1953 for polyps of the stomach is extended to all of the same lesions in the GI tract [2,4,8,9,10,14]. In 2008, the neoplastic nature of IFP became evident by the detection of activating PDGFRA mutations in these tumors. The gene PDGFRA is located at chromosome 4q12 [3,7,16,17,18]. IFP is the most common in the antrum of the stomach (66-75%), small intestine (18-20%), colorectal (4-7%), and rarely in the bile duct, esophagus, duodenum [3]. Macroscopic peduncular or sessile (0.2-20 cm in size) originating from the submucosa, they usually represent like solitar lessions, rare like multiple lesions [2,9,14,15]. The mucosa is usually ulcerated or faint. Etiopathogenesis is unknown (trauma, allergic reactions, genetic, bacterial, physical, chemical damage, chronic irritation and inflammation, or a consequence of extreme reaction of the body to an intestinal trauma, H. pylori infection) [3-5,7,13,14,16]. Clinical presentation is usually asymptomatic and symptomatic when depend on the localization (pain, obstruction, other GI symptoms are rare) [2,8,9,10,14]. The localization in the esophagus or in the upper part of the stomach can cause symptoms of gastro-esophageal reflux or dysphagia [11]. The ileal segment is the most common site where these lesions cause intussusceptin [13,14]. There is one report which described intussusception resulting from double invagination (with antegrade with retrograde bowel loops) due to IFPs [10]. Tissue is microscopically composed from mononuclear spindle cells that form the concentric structure [1-3,5,10,12,15]. Inflammatory infiltration includes blood

vessels, eosinophilis, lymphocytes, macrophages, mast cells [1-3,5,8,9,13,15,16-19]. Microscopic differential diagnosis is relating to other changes on the spindle cells (inflammatory fibrosarcoma, gastrointestinal stromal tumor, eosinophilic gastroenteritis) [2,3,5,6,15,20,21]. Immunohistochemistry GIST and IFP are positive for CD34 and vimentin, but GIST is positive for CD117 and the IFP does not [7,13,14,17]. Some authors supposed that FP (fibroplastic polyps in colon) represent an initial stage of Vanek's tumor [17].

An accurate diagnosis is based on a good medical hystory and physical examination, imaging techniques, abdominal X-ray examination, ultrasonography (US, EUS), CT, MRI, enteroclysis, endoscopic procedures, endoscopy capsule [3,15]. Ultrasonography has a sensitivity of 98-100% and specificity of 88-89% for the diagnosis of intussusception [3,4,10,14,18]. CT is the most sensitive screening for confirmation of intussusception, with diagnostic significance in 58-100% of cases [7,10,12-14,17,18]. MR correctly diagnosed cause of obstruction in 95% cases [3]. Modalities of solutions are the primary resection block, or initial reduction followed by limited resection [3]. The laparoscopic approach is still controversial because high risk of tumor rupture or bleeding, which can result in peritoneal spreading. But there are reports of successful and safe treatment even of large tumors with this method [11]. Recent studies suggest an initial reduction of the visible outside of the small intestine before resection [3,11,13,14]. Reduction can be attempted if there are signs of inflammation or ischemia of the bowel wall and malignancy is not suspected [12,20]. In ileocolic and colocolic intussusception in adults' possibility of cancer can be expected in 43-100% of cases, however, recent studies in these cases suggests primary resection. Surgical resection is recommended treatment of adult intestinal invagination of the intestinal segments involved, which was performed in our case, and this was curative with no longstanding complication [17,21-27].

Conclusion

Small bowel intussusception in adults is rare and often challenging to diagnose owing the elusive, non-specific symptoms. Vanek's tumours arising within the small bowel rarely present with obstruction or intussusception. The optimal surgical management of adult small bowel intussusception varies between reduction and resection. Reduction can be attempted in small bowel intussusceptions provided that the segment involved is viable and malignancy is not suspected. CT is the most sensitive screening for confirmation of intussusception. In the presence of a lead point lesion but no preoperative tissue diagnosis, surgical intervention in the form of bowel resection without reduction is advisable. Although inflammatory fibroid polyps are rare, benign, uncommon and non neoplastic causes of intussusception, in the case of intestinal obstruction the only solution is a surgical approach and resection of the intestinal segment involved.

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