Adult Jejunoileal Intussusception Due to Submucosal Angiolipofibroma and Lipoma: A Rare Phenomenon

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Abstract

Intussusception in adults is highly uncommon and accounts for only 5% of all reported case. Small bowel intussusception is much less common than the ileocolic type, with jejunoileal intussusception being amongst the most rare. Benign tumors of the small bowel are rare clinical entities. These often remain asymptomatic throughout life. Despite comprising 75% of the length and 90% of the surface area of the gastrointestinal tract, the small bowel harbors relatively few primary neoplasms and fewer than 2% of gastrointestinal malignancies. We report a case of 28 year old female with jejunal submucosal angiolipofibroma and lipoma that became symptomatic due to intermittent obstruction episodes and caused intestinal obstruction due to intussusception. Involved part of jejenum was resected and end-to-end anastomosis was done and the patient's postoperative period was uneventful. In this case report, the diagnosis and management of intussusception due to benign tumour are discussed along with a literature review.

Keywords: Intussusception; Small intestines; Obstruction; Polyp; Neoplasm.

Introduction

Intussusception is highly uncommon in adults and accounts for only 5% of all reported cases. It is more commonly secondary to an identifiable bowel lesion in 90% of cases, whereas 10% have no discernable cause[1,2]. Lipomas constitute about 10% of the gastrointestinal benign tumours[3] and there are limited scattered cases in the literature of intestinal lipomas presented with bleeding[4], and bleeding and intussusceptions[5]. Symptoms due to obstruction in adults tend to be chronic or intermittent and include pain, constipation, weight loss, or a palpable abdominal mass at physical examination. In adults, intussusceptions may be ileocolic, colocolic, enteroenteric and there is no anatomic predilection. The lead points of adult intussusceptions that involve the colon are usually malignant (carcinoma, lymphoma), whereas those

that involve the small bowel tend to be benign (lipoma, polyp, Meckel diverticulum, from lymphoid hyperplasia secondary to viral infection). The clinical presentation of patients with intussusceptions also differs in these two age groups. Children present acutely with colicky abdominal pain, vomiting, and bloody stools that look like currant jelly, and often a palpable mass. Symptoms in adults tend to be more chronic or intermittent and include pain, constipation, weight loss, or a palpable abdominal mass at physical examination[6]. Diagnostic imaging plays an important role in the diagnosis of the condition. Ultrasonography and computed tomography (CT) are the most commonly used imaging techniques. Here we report a case of small intestinal (jejunal) Angiolipofibroma and Lipoma which presented with intestinal obstruction caused due to intussusception and review some aspects of diagnosis and treatment.

Case Report

A 28 years old female patient was admitted to the emergency department with a history of pain abdomen, fullness and nausea with few episodes of vomiting for 4-5 days. He gave history of intermittent abdominal pain, distension and nausea aggravated by eating and on-off constipation for last 2 years, since then patient is on stool softener. History of loss of appetite is also present, which is associated with loss of weight from 72 to 50 kg in last 2 years. There was no past history of any previous operation. There was no family history of gastrointestinal disorders or neoplasms. Bowel sounds were slightly increased. On physical examination, the abdomen was without palpable masses, tenderness or rigidity. Mild abdominal distension was noticed. Digital examination showed that the rectum was empty of stool. Examination of the other systems was normal. Plan X-ray abdomen and results of routine laboratory tests were within normal limits. MSCT-Volume scan done using IV contrast with 0.6 mm Multiplanar Protocol Abdomen showed small gut intussusception in the left paraumblical region along with an ovoid intrasmall gut area of reduced density (-95hu). On exploratory laparotomy, Intussusception was found. Apex was formed by multilobulated mass in jejunum (Submucus Lipoma). The affected segment of jejunum was resected with an end-to-end anastomosis. The patient's post-operative period was uneventful and was discharged on 5th postoperative day. Pathology report microscopy & impression of jejunum segmental resection suggestive of two submucosal Angiolipofibroma and one Lipoma, jejunum largest lipoma measuring 2.5 cm in greatest dimension with mature adipose and fibrous tissue and thick and thin walled vessels in varying proportions. Focal pyloric metaplasia in overlying mucosa but no evidence of granuloma or malignancy seen.

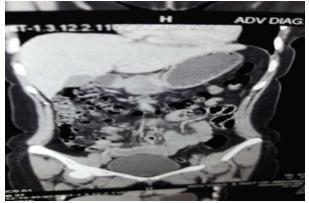


Fig. 1: Showing CT Abdomen Showing Intussusception



Fig. 2: Intussusception- prior to resection.

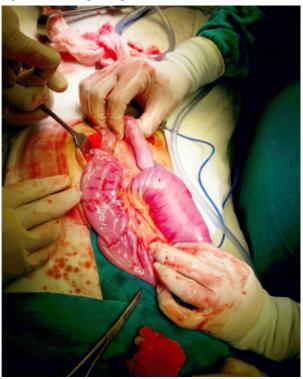


Fig. 3: Showing typical intussusception- on table look



 $Fig\ 4\ \text{Bowel wall is cut away showing the mass}$

Discussion

Intussusception is relatively rare in the adult population, and this along with the vague clinical features, makes diagnosis difficult. Intussusception occurs when a segment of bowel, the intussusceptum, invaginates into the lumen of the more distal bowel, the intussuscipiens. Intussusception in adult patients may be caused by intraluminal, mural or extraluminal lesions [7,8]. The primary mechanism by which intussusception is thought to occur is when an intraluminal mass is pulled forward by peristalsis and drags the attached bowel wall segment with it. Pedunculated tumors, such as adenomatous polyps or lipomas, are the classic examples of this group [9-11]. Adult intussusception is rare and usually associated with neoplasms, of which up to 77% are malignant [12]. Polyps, lipomas, fibroma and leiomyoma are known to be the commonest type benign tumours in the small bowel. Lipomas account for 10-12% of all benign tumours of the small intestine. Ileum is the commonest site of the small bowel lipomas, followed by jejunum and duodenum. Their size ranges between 1 and 6 cm but they may occasionally reach a size up to 30 cm. Small intestinal lipomas are usually single and they are multiple in about 10% to 15% of cases are often considered in distinctive diagnosis. Therefore, because of these characteristics, they are mostly removed to exclude the diagnosis of malignant lesions[13-15]. The majority of lipomas are submucosal (90%), although they can also be subserosal or intramuscular. Adult intussusception is not easily diagnosed because patients usually present with non-specific vague symptoms such as abdominal pain, the most common symptom. Other symptoms include nausea, vomiting, and possible bleeding from the rectum [16]. Approximately 50% of patients will have had symptoms for more than one month prior to an acute exacerbation of symptoms that leads to diagnosis [17,18]. The physical findings are also non-specific and are not consistent with an acute abdomen. Early diagnosis of intussusception may prevent the necrosis of the bowel and, in some cases, even save the patient's life [16]. As symptoms are vague, diagnostic imaging plays the main role in diagnosis. . Barium enema and endoscopy are not useful for the diagnosis of jejunal or ileal lipomas, although they are basic measures for investigating colonic or duodenal lipomas. Many imaging modalities are used for diagnosis, such as radiographs, ultrasonography, CT and magnetic resonance imaging. The most commonly used are ultrasonography and CT scan. The classic appearance of an intussuscepted bowel on a sonographic image in a transverse plane is called the "doughnut sign" or a "target lesion" and represents several concentric rings of the bowel. Usually there is a thick hypoechoic rim with an echogenic area in the middle. The hypoechoic rim represents an edematous bowel wall, and the echogenic center corresponds to intussuscepted mesenteric fat. Sometimes, within the echogenic area in the center, an additional anechoic spot may be seen, which is believed to represent a collection of fluid in the apex of the intussusceptum [7,19]. The longitudinal appearance of intussusception usually appears as multiple parallel lines, the so-called "sandwich appearance" or "pseudo-kidney sign". The lines demonstrate bowel walls and their layers. The major limitation of ultrasonography for evaluating acute obstructive symptoms is the presence of air in the bowel, which leads to poor transmission and difficulties in image interpretation. Like sonography, CT scanning can be used to identify the intussusception; however, the underlying cause can still be difficult to determine. Although intestinal lipomas are rare, they should be kept in mind when evaluating the adult patient with intermittent abdominal symptoms. They should be removed because they can cause symptoms such as obstruction or bleeding and usually a histological evaluation is indicated in intestinal mass to exclude the possibility of malignancy. Endoscopic removal entails a risk of perforation or bleeding due to submucosal origin of the majority of the lipomas. The size of the stalk is of greater importance than the diameter of the lipoma itself when patient is evaluated for endoscopic resection. Laparoscopic resection is also a viable alternative to open excision in selected cases. In our patient the intussusception was diagnosed with abdominal CT. Our patient went on to surgery, where the CT findings were confirmed.

Consent

Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

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