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Research Article

Ileo-ileal intussusception in adult caused by Vanek's tumor: a report of five cases

Mohamed S. Belhamidi*, Abdelmounaim Aitali, Mohamed Tarchouli, Hicham Baba, Ahmed Bounaim, Khalid Sair

Department of General surgery, Mohamed V Military teaching hospital, Mohamed V- Souissi University, Rabat, Morocco

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***Correspondence:**
Mohamed S. Belhamidi,
E-mail: belbamidimohamedsaid@gmail.com

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ABSTRACT

Background: Inflammatory fibroid polyps (IFPs) (Vanek's tumor) are rare, benign lesions of the gastrointestinal tract. Its treatment is surgical. The debate focused on the issue of primary en bloc resection of intussuscepted bowel versus initial reduction followed by more limited resection. We report five cases of small bowel intussusception in adult caused by Vanek's tumor.
Methods: The retrospective study reviewed epidemiological data, clinical features, diagnosis and management of adults with a diagnosis of intussusception admitted to our department from August 2010 to April 2014. Only intussusception caused by Vanek's tumor was included in our study.
Results: There were 4 males and 1 female. Their average age was 53 years. All of them were admitted with abdominal pain. The abdominal exam revealed an abdominal distension in 4 patients and a palpable abdominal mass in 2 patients. One patient had a normal physical examination. CT demonstrated <<target lesion>>, a sign pathognomonic for ileo-ileal intussusception. All of the patients underwent surgical treatment by laparotomy for 4 patients and laparoscopy approach for 1 patient. 4 patients had an ileo-ileal intussusception; which was resected after partial reduction and followed by end-to-end anastomosis. In one case, we found an ileo-caeco-colic intussusception, which was resected and side-to-side ileo-colic anastomosis was performed. Diagnosis of Vanek's tumor was confirmed by immunohistochemical examination. Postoperative course was uneventful.
Conclusions: The recommended treatment of adult intestinal invagination is surgical resection. Discussion continues as to whether or not a reduction has to be done before a resection.

Keywords: Vanek's tumor, Intussusception, Surgical resection

INTRODUCTION

Inflammatory fibroid polyps are rare, non-neoplasic lesions that originate in the sub mucosa of the gastrointestinal tract. They were first described in the stomach by Vanek, in 1949. The aetiology of IFP is unknown. Surgical resection is the treatment of choice for this disease. The debate focused on the issue of primary en bloc resection of intussuscepted bowel versus initial reduction followed by more limited resection. We report five cases of ileo-ileal intussusception in adult caused by Vanek's tumor.

METHODS

The retrospective study reviewed epidemiological data, clinical features, diagnosis and management of all adults with a diagnosis of intussusception admitted to Department of General surgery, Mohamed V Military teaching hospital, Rabat, from August 2010 to April

2014. Only intussusception caused by Vanek's tumor was included in our study. An intussusception that involved only the jejunum or the ileum was considered an enteric intussusception and the intussusception that involved the ileum and colon was designated as an ileo-caeco-colic intussusception.

RESULTS

Demographics

The average age of the patients was 53 years, with a range of 22 to 62 years. Four (80%) were male and one (20%) was female.

Clinical presentation

Three patients had chronic symptoms and two had acute presentation (small bowel obstruction). Abdominal pain was the most common presenting complaint. It located in umbilical point in two cases and in right iliac fossa in one patient. Two patients had diffused abdominal pain. Cardinal signs of intestinal obstruction were found in two patients. All of the patients had abdominal distension in abdominal examination. In only two cases, palpable mass was revealed in the right iliac fossa and para-umbilic area.

Figure 1: Abdominal CT-scan showing ileo-ileal intussusception.

Preoperative diagnosis studies

A plain X-ray of the abdomen was carried out in four patients as an initial investigation, which showed a small bowel obstruction with multiple air fluid levels in two cases. It was normal in the other two patients. Ultrasonography was performed in one patient. It showed an image in rosette and pseudo-kidney and revealed a suspicion of ileal intussusception. Abdominal computed tomography demonstrated an image in target, a sign pathognomonic of bowel intussusception (bowel in bowel). Four patients had an ileo-ileal intussusception. In one patient, CT scan reported ileo-caeco-colic intussusception with polypoidal lesion as a lead point.

Treatment

Four patients underwent a laparotomy through a midline incision. Laparoscopic resection was carried-out in one patient. An abdominal exploration found an ileo-ileal intussusception in four patients, which was situated between 15 cm and 30 cm proximal to ileo-caecal valve. The other one patient had on ilio-caeco-colic intussusception. The affected ileum segment with ileo-ileal intussusception was reduced and a limited resection was performed followed by end-to-end anastomosis. An ileo-caecal resection, with not reduction, was performed in case with ileo-caeco-colic intussusception. The faecal stream was re-established by side-to-side ileo-colic anastomosis. In all of the five cases, histopathological analysis of excised specimen showed a lesion with a variable cellularity, formed by spindle cells; immunohistochemistry was positive for CD34. Based on these pathologic and immunohistochemical findings, a diagnosis of inflammatory fibroid polyp was made. Post-operative course was uneventful.

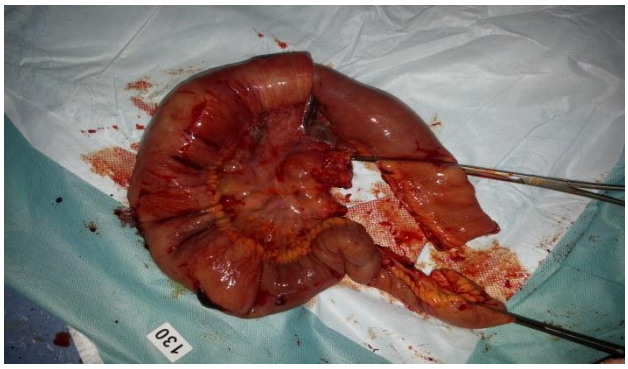

Figure 2: Intussuscepted segment resected.

DISCUSSION

Adult intussusception occurs in only 1% of patients suffering from small bowel obstruction. Intussusception caused by inflammatory fibroid polyps is uncommon.[1] IFPs (Vanek's tumor) are rare, benign, non-neoplasic lesions, composed mainly of loose connective tissues, vessel, and eosinophilic inflammatory cells. Konjetzny first described this lesion as polypoid fibroma in 1920, then by Vanek in 1949 as a gastric sub-mucosal granuloma with eosinophilic infiltration.[2] It was finally named as IFP in 1953 by Helwing, indicating that its nature was probably inflammatory.[3] IFPs affect both sexes and its typical presenting age is the 5th to 7th decades of life.[4] In our cases, there were 4 males and one female with a mean age of 42 years.

The aetiology remains unknown, but it could be a consequence of chronic irritation and inflammation or a consequence of extreme reaction of the body to an intestinal trauma.[1]

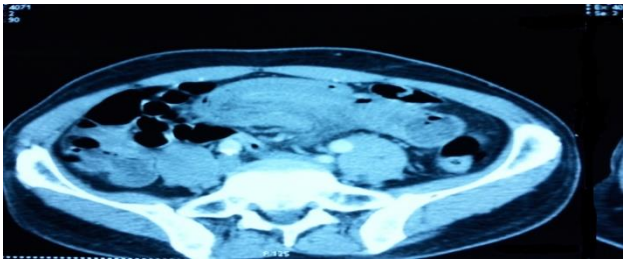

Figure 3: Intussusception with bowel invagination showing a pseudo-kidney appearance.

The most common site of IFP is the gastric mucosa accounting for 70%. Of other gastrointestinal sites affected, the small bowel is the most common; accounting for 23%, with ileum predominating.[5] In all our patients, no gastric localisation was found on the endoscopy performed postoperatively. The symptoms are dependent on size and localization of the tumour. The most common presenting sign and symptom of an adult patient with intussusception caused by IFP in the small bowel is colicky abdominal pain (71% to 90% of patients). Nausea and vomiting, bleeding, diarrhoea, constipation or a change in bowel habits is other non-specific findings at the time of presentation. Palpation of an abdominal mass during clinical examination is only reported in the less than 50% of patients. Clinically, ileo-ileal intussusception presents either as acute pathology with signs and symptoms of abdominal obstruction or as a more chronic pattern with recurrent colicky abdominal pain. Ultrasound is the primary imaging modality of choice or the first-line examination for intussusception, and the classic imaging features include the target, bull's eye or doughnut sign in the trans- verse view and the pseudo-kidney, sandwich, or hayfork sign in the longitudinal view.[6]

Figure 4: Operative image showing ileo-ileal intussusception with dilatation of proximal segment.

In the current case, all of the patients presented at the emergency department with abdominal pain and vomiting, two of them had paroxysmal pains. Two patients out of 5 presented signs of intestinal obstruction and two patients had a palpable mass in the right iliac fossa. Ultrasound is the primary imaging modality of choice or the first-line examination for intussusception, and the classic imaging features include the target, bull's eye or doughnut sign in the trans- verse view and the pseudo-kidney, sandwich, or hayfork sign in the longitudinal view.[6]

Figure 5: Excised specimen, opened to illustrate ileo-ileal intussusception caused by polypoid mass.

Ultrasonography enables the diagnosis or exclusion of intussusceptions with a sensitivity of 97.9% to 98.5%, a specificity of 97.8% to 100%, a positive predictive value of 86.6% and a negative predictive value of 99.7%. However, obesity and the presence of a large amount of air in the distended bowel loops can limit the image quality and diagnostic accuracy. CT imaging from a group of 136 adult intussusceptions was used to diagnose surgical entero-enteric intussusceptions. Using the criteria of length > 3.5 cm, the imaging diagnosis yielded a mean sensitivity of 100% and a specificity of 57.3%. Using a measured axial diameter >3 cm, the mean sensitivity and specificity were 100% and 32.9%, respectively. On CT, a bowel-within-bowel configuration, which is suggested by mesenteric fat and vessels compressed between the walls of the small bowel, is pathognomonic of intussusceptions.[7] In contrast to ultrasonography, CT is unaffected by the presence of gas in the bowel lumen. Therefore, we suggest that all patients presenting with an intestinal obstruction should undergo a CT scan as a routine diagnostic tool.[8] In our case, ultrasound was not indicated as first examination as it is limited by abdominal distension. In all our patients, the diagnosis was obtained by an abdominal CT scan, which showed pathognomonic signs of intussusception. The cause of intussusception was identified in only one patient.

Histologically, an inflammatory fibroid polyp is distinguished by a localized proliferation of mononuclear spindle-shaped cells with an inflammatory infiltrate. Immunohistochemical staining shows positivity for CD34, suggesting that these polyps may develop from primitive perivascular or vascular cells.[9] Negative staining to S100 protein and CD 117 (KIT gene product) distinguishes an inflammatory fibroid polyp from a neurogenic tumour or gastrointestinal stromal tumour (GIST).[5] In our case, the diagnosis of IFP was confirmed by immunohistochemical analysis.

The appropriate management of adult intussusceptions remains controversial, with the debate mainly focused on the issue of primary en bloc resection versus initial reduction followed by more limited resection. There are several areas of controversy here, including the question

of intraoperative reduction; the extent of the resection and the approach taken for possible reduction and subsequent resection; and the possibility of potential tumour seeding of the peritoneal cavity must be taken into consideration. Chang et al demonstrated that 76.1% of all leading lesions are benign in Asian adult intussusception, and IFPs are believed to have no malignant potential. So we believe that reduction of adult intussusception has a place, especially of the small bowel, where the risk of overt malignancy is lower.[10] Theoretically, this surgical reduction before resection may permit more limited resection.[8] The likelihood of cancer in ileo-colic and colo-colic intussusceptions is 43% - 100%.[11] It is advisable for colonic intussusception to be left unreduced and resected as a single mass, obeying standard oncological principles.[10] In our case, all patients with small bowel intussusception underwent partial surgical reduction before segmental resection of the ileum. In one patient, who had an ileo-caeco-colic intussusception, a right hemi colectomy without reduction was performed. The surgery should be performed as early as possible to prevent the intussusceptions from leading to ischemia or necrosis and the subsequent perforation of the invaginated bowel segment. When surgery is delayed and intestinal perforation with peritonitis occurs, there is a considerable increase in morbidity and mortality. With current technological advances, laparoscopic surgery may have a role in the management of adult intussusception. The role of laparoscopic surgery in the literature has mainly been manipulation and de-rotation of the associated volvulus, and especially in intra-corporeal reduction. Hand-assisted laparoscopic surgery may better facilitate these manoeuvres, as well as bowel mobilization, if required. Intra-abdominal resection may be simple or complex, ranging from the stapling of intussuscepting IFP post-reduction to more complicated or even colonic cases; otherwise, it may be more prudent to perform extracorporeal resection via a minimal and convenient laparotomy access.[12,13] In our cases, laparoscopic approach was performed in one patient. In the literature, no case of recurrence was reported.

CONCLUSION

An intussusception is a rare cause of adult intestinal obstruction caused by Vanek's tumor. CT scan is useful in confirming anatomical abnormalities, but final diagnosis will be made, based on histopathological analysis. The recommended treatment of adult intestinal invagination is surgical resection. Discussion continues as to whether or not a reduction has to be done before a resection.

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