ORIGINAL RESEARCH PAPER
Volume - 12 | Issue - 03 | March - 2023 | PRINT ISSN No. 2277 - 8179 | DOI : 10.36106/ijsr
INTERNATIONAL JOURNAL OF SCIENTIFIC RESEARCH

ILEO-ILEAL INTUSSUSCEPTION IN AN ADULT DUE TO INFLAMMATORY FIBROID POLYP: A CASE REPORT

Surgery

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ABSTRACT

Intussusception is a common cause of acute abdomen in the pediatric population, but it is rare in case of adults. They present with features of intestinal obstruction which include abdominal pain, distension, vomiting, constipation. Radiological investigations like plain abdominal X-rays, barium studies and CT (computed tomography) may help in reaching a diagnosis. Intussusceptions occurring in adults usually have an underlying etiology, which may be benign or malignant. Therefore, surgery is the mainstay of treatment with resection of the involved part. Adult intussusceptions involving the small bowel are mostly benign, and among the benign causes, inflammatory fibroid polyp (IFP) is a rare cause. IFPs are also known as Vanek's tumor, inflammatory pseudotumor, granuloma with eosinophils, eosinophilic granuloma, etc. They are benign, usually solitary lesions arising from the sub mucosa of the gastrointestinal tract. We report a case of intestinal obstruction due to an ileo-ileal intussusception in a 48-year-old female, caused by an IFP.

KEYWORDS

Intussusception, Intestinal obstruction, Inflammatory fibroid polyp

INTRODUCTION:

Intussusception occurs when the proximal segment of the gastrointestinal tract invaginates into the adjacent distal segment, causing obstruction.[1,2] It occurs more commonly in children, and although adults can rarely present with it.[3] Adult intussusception accounts for 5% of all bowel intussusceptions, 1-5% of all bowel obstructions in adults[6,7], 0.08% of all abdominal surgeries and 0.003-0.02% of all hospital admissions.[10]

In children, 95% of the cases are idiopathic. However, in adults, 80-90% have an identifiable etiology, which may be benign or malignant.[8] Inflammatory fibroid polyps (IFPs), or Vanek's tumor, is a rare benign cause.[11] They are polypoidal growths originating in the sub-mucosa of the GIT.[12,13] They occur most commonly in the antrum of stomach (66-75%), followed by small intestine (18-20%) and colorectal region (4-7%).[15]

We report an unusual case of intestinal obstruction due to ileo-ileal intussusception caused by an inflammatory fibroid polyp (IFP), which was confirmed by histopathological examination.

Case Report:

A 48-year-old female presented to the emergency department with history of abdominal distension and colicky abdominal pain, inability to pass flatus and stool, and vomiting. On examination, patient was dehydrated and afebrile. Abdomen was distended with stepladder peristalsis; increased bowel sounds and a palpable mass in the peri-umbilical region. Her blood pressure was 106/62 mmHg, and pulse rate of 68 beats/min.

Patient was admitted in the surgery ward, kept nil by mouth and started on intravenous fluids, antibiotics, and nasogastric tube and urinary catheter were inserted. Her leukocyte count was 11200, Hb was 12.7 g%, and albumin was 3.3. Erect abdominal X-ray revealed dilated bowel gas shadows. Abdominal ultrasound showed thickened bowel wall measuring 9.3mm and 11mm. CECT revealed features of small bowel loop obstruction with long segment narrowing of distal ileal loop with internal mesenteric component (Ileo-ileal intussusception).

She underwent exploratory laparotomy on the next day. Intra-operatively, an ileo-ileal intussusception of approximately 20 cm length in the ileum was found with dilated proximal bowel loops. Upon its reduction, an intramural obstructing polypoid mass was found as the lead point, approximately 120cm from the ileocecal junction. Resection of the segment containing the mass with 5cm margins on each side was done, followed by ileo-ileal end-to-end anastomosis, and specimen sent for histopathological examination.

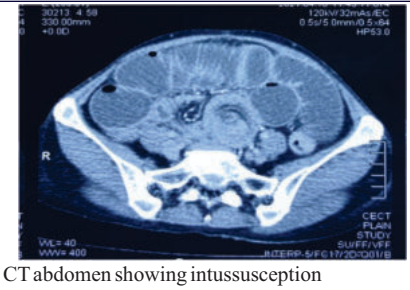

Figure 1: CT abdomen showing intussusception

Figure 2: Intra-operative picture of ileo-ileal intussusception

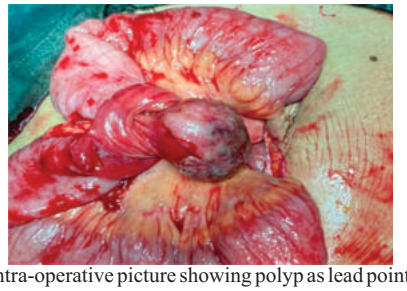

Figure 3: Intra-operative picture showing polyp as lead point

On gross examination, the resected segment measured 12.5cm and its cut section showed a large ulcerated pedunculated polyp measuring 3.5cm X 3cm X 2.5cm with areas of congestion.

Microscopic examination revealed extensive areas of ulceration covered with acute inflammatory exudate, edematous sub-epithelium with numerous proliferating capillaries and scattered fibroblasts like spindle cells in a collagenised background. Moderate mixed inflammatory cell infiltration with predominantly eosinophils is seen.

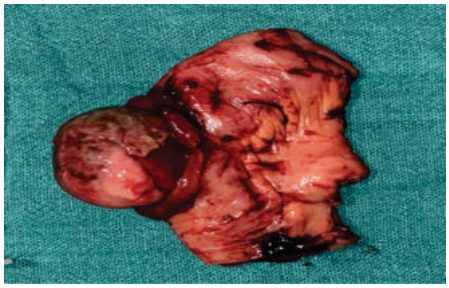

Figure 4: Resected specimen with the polyp

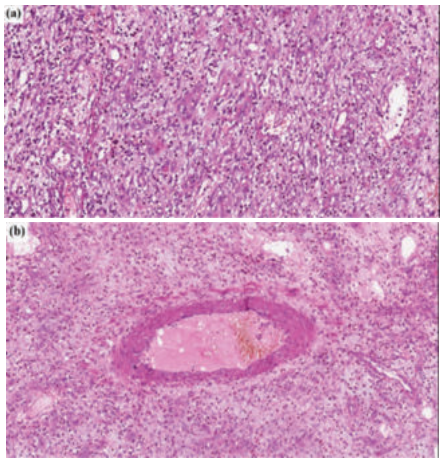

Figure 5(a) &5(b): Histopathological examination of the polyp showing fibroblasts and inflammatory cells

The post-operative period was uneventful for the patient. She improved steadily over the course of her hospital stay and was discharged a few days later. She was followed up in the outpatient department a few weeks later and had no complaints. She was eventually discharged from follow-up.

DISCUSSION:

Intussusception was first described by Barbette of Amsterdam in 1674.[1] The first surgery was done on a child with intussusception by Sir Jonathan Hutchinson in 1871.[2] Intussusception of the bowel refers to a process in which a segment of bowel containing a part of its mesentery telescopes into the lumen of the distal bowel segment adjacent to it.[4] Intussusception is frequent among children, being the second most common cause of acute abdomen after acute appendicitis.[5] In children, it occurs around 6-18 months of age with male to female ratio of 3:1.[6] However, it is rare in adults, and the overall incidence in adults is estimated at 2-3 cases/1,000,000 population/year.[8] The ratio of pediatric to adult intussusception cases is approximately 20:1.[9,10]

95% of the causes of intussusception in children is idiopathic. However, in adults, almost 90% are secondary to an identifiable pathologic lesion that serves as a lead point. Two-thirds of the cases with identifiable lead point is attributed to benign/malignant neoplasms, out of which 50% is due to a malignant cause; while the remaining maybe due to infections, post-operative adhesions, Crohn's disease, Meckel's diverticulum.[3]

More commonly occurring in the small intestine, 50-75% of small bowel intussusceptions are due to benign causes, like lymphoid hyperplasia, lipomas, leiomyomas, hemangiomas, polyps, and inflammatory lesions, while malignant causes include metastasis, leiomyosarcomas, adenocarcinomas, GIST and lymphomas.[3] Adult intussusception of the colon occurs in 20-25% of all cases and most likely have a malignant cause, most common being adenocarcinoma of the colon, while the most common benign cause is lipoma.[3]

According to location, intussusception is classified into: (i) Entero-enteric (involving only small bowel), (ii) Colo-colic (involving only large bowel), (iii) Ileo-colic (when the terminal ileum invaginates into the ascending colon), and (iv) Ileo-cecal (when the ileo-cecal valve acts as the lead point). The proximal segment is called "intussusceptum" while the distal segment is referred to as "intussuscipiens."[5]

The exact mechanism of intussusception remains unclear. It is believed to involve altered bowel peristaltic activity due to some lesion in the wall, or an irritant within its lumen, which forms the leading edges for the intussusception. An area of constriction above and relaxation below the stimulus is produced following ingestion of food and subsequent peristalsis, thereby telescoping the lead point into the distal segment. This leads to complications like intestinal obstruction, bowel ischemia & necrosis, and sepsis.[9]

The classical triad of intermittent colicky pain, a palpable sausage-shaped tender mass and "red currant jelly" stool in pediatric intussusception is rare in adults. Clinical features include abdominal pain, vomiting, bloating/distension, constipation, bloody diarrhea, and altered bowel habits. On examination, abdomen may be distended with hypoactive or decreased bowel sounds, a mass may be palpable in some cases (24-42%), and in late stages, there may be peritonitis with hypotension and tachycardia.

Laboratory investigations include raised white cell counts and increase in acute phase reactants (CRP, thrombocytes).[3] Radiological investigations help in the diagnosis. Plain abdominal films, being the first investigation, usually reveal signs of intestinal obstruction which may include dilated loops of bowel with absence of colonic gas, multiple air fluid levels, "air crescent sign" (intraluminal air trapped between walls of the intussusceptum and intussuscipiens). However, they lack sensitivity and specificity.[3] Upper GI contrast studies may reveal "stacked coin" or "coiled spring" appearances.[3]

Ultrasonography features include "target" or "doughnut" in the transverse plane, and "pseudo-kidney" sign in the longitudinal view.[9] It has a sensitivity of 98-100%[3] and a specificity of 88%. Its disadvantages include possibility of masking of features by gas-filled bowel loops and operator dependency.[9] Abdominal CT is the most sensitive with a reported diagnostic accuracy of 58-100%. The pathognomonic feature is a bowel-within-bowel appearance, "target" or "bulls-eye" appearance; and "sausage-shaped" lesions as a concentric hyperdense double ring. CT can also determine the location of the lesions serving as the lead point, nature of the lesion, relationship of the lesion to surrounding structures and any vascular compromise in the wall.

Other investigations include MRI (magnetic resonance imaging), but it is not routinely done. MRI features include "bowel-within-bowel" or "coiled-spring" appearance. Besides these, endoscopic approaches like lower GI endoscopy, enteroscopy and capsule endoscopy have been employed in selected cases. A lower GI endoscopy can be useful in cases involving the large bowel. It helps in confirming the intussusception, localizing the disease and demonstrating the underlying lesion serving as the lead point. On capsule endoscopy, it has been reported that intussusception appears as a mass lesion of the small bowel. However, some consider that it is contraindicated in presence of obstructive symptoms. A double balloon enteroscopy can visualize up to 70-150 cm of the small bowel. Lastly, a diagnostic laparoscopy (DL) may help if diagnosis cannot be established preoperatively.[16]

Inflammatory fibroid polyp (IFP) is a rare, benign, usually solitary lesion that arises from the submucosa of the GIT.[12] It was first described in the stomach by Vanek in 1949 as an eosinophilic submucosal granuloma.[12,14] Hence, it is also known as Vanek's tumor. In 1953, Helwig and Ranier introduced the term Inflammatory Fibroid Polyp (IFP) as it showed fibrous and vascular tissue with an inflammatory infiltrate, including eosinophils.[17] IFPs are also known as inflammatory pseudotumor, granuloma with eosinophils, eosinophilic granuloma, hemangiopericytoma, and polypoid myoendothelioma.[18] IFPs affect both sexes equally and all age groups, with peak incidence in the 5th-6th decades.[19] They are mostly sporadic and solitary[19], but rarely, metachronous lesions have been reported in familial cases. They seem to lack a malignant potential and recurrence reported only once till date.

The etiology of IFPs is unknown, but it is believed to be due to chronic irritation and inflammation, or reaction to intestinal trauma, or some variant of eosinophilic gastroenteritis. IFPs can occur at different locations, mostly in the gastric antrum (66-75%), followed by small intestine (18-20%), large intestine (4-7%), duodenum, esophagus and gall bladder (1%), and in the appendix (less than 1%).[12] They are usually asymptomatic or incidentally diagnosed during other

procedures.[12] If symptomatic, patients present with abdominal pain (if the polyp is present in the stomach), or with obstruction in case of small intestinal polyps.[12]

Grossly, IFPs are either pedunculated or sessile, and project into the lumen, with usually pale and ulcerated mucosal surface, measure 2-5cm in diameter, however reports of giant IFPs upto 12.5 cm have been found.[20,21] Microscopically, it consists of mononuclear, spindle-shaped cells arranged in whorls around blood vessels or mucosal glands with inflammatory infiltrates (eosinophils, lymphocytes, macrophages, and collagen containing matrix).[20] On immunohistochemistry, IFPs are variably positive for CD34, vimentin but they are negative for CD117, which differentiates it from GIST.[11] Differential diagnosis of IFPs include Gastro-Intestinal Stromal Tumors (GIST), inflammatory myofibroblastic tumor and schwannoma.

The mainstay of treatment in case of adult intussusception is surgery, due to significant rates of underlying pathology and high incidence of malignancy. Unlike in pediatric population, preoperative reduction with barium/air is not a recommended treatment in adults.[6] Surgery may be performed as an open surgery (laparotomy) or laparoscopically. There has been controversy as to whether reduction should be done intra-operatively. Reduction carries the risk of dissemination of malignant cells and possibility of perforation if the bowel is ischaemic.[2] Selective reduction and resection could be done in some cases, especially in intussusception of the small intestine, where risk of malignancy is lower, but only if the bowel is viable and easily reducible.[13] Reduction can be done by milking it from distal to proximal direction, thus allowing it for resection. However, colonic intussusception is preferred to be left unreduced and resected as a single mass obeying standard oncologic principle.[22] The role of laparoscopic surgery has mostly been in manipulation and de-rotation of associated volvulus, intra-corporeal reduction of the intussusception and resection. It is done only in selective cases.

CONCLUSION:

Intussusception is an uncommon cause of bowel obstruction in adults. It is mostly associated with an underlying pathology, serving as the lead point. In the small intestine, benign lesions are common, and one of the rare benign causes are IFPs. CT scans are useful in making a diagnosis of intussusception, but definitive diagnosis of IFP is made only on histopathological examination. Surgical intervention is the treatment of choice and resection is the definitive method of treatment in case of intussusception due to IFPs.

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