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ADULT ILEO-ILEAL INTUSSUSCEPTION: CASE REPORT AND LITERATURE REVIEW

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ABSTRACT: Adult intussusceptions is a rare entity accounting for only 5% of all intussusceptions and causes approximately 1% of all adult intestinal obstructions. Intussusception is an uncommon cause of abdominal pain in adults and poses diagnostic challenges for emergency doctors, due to its varied presenting symptoms and time course. Diagnosis is thus often delayed and results in surgical intervention due to the development of bowel ischaemia. We report a rare case of ileo-ileal intussusception in adult presented with acute intestinal obstruction.

INTRODUCTION: Intussusception was reported for the first time in 1674 by Barbette of Amsterdam. Intussusception, or 'introsusception' as it was named then, was later detailed in 1789 by John Hunter.^[1] In 1871, Sir Jonathan Hutchinson was the first to successfully operate on a child with intussusceptions. Intussusception is relatively frequent in children but is rare in adults. In contrast to pediatric intussusception, which is idiopathic in 90% of cases, adult intussusception has an organic lesion in 70% to 90% of cases.^[2] Adult intussusception can present with atypical symptoms of an acute, subacute, or chronic clinical entity, and timely diagnosis is often missed, leading to a delay in proper treatment.^[3] Although it is generally accepted that adult intussusception requires surgical resection because of the underlying pathology in the majority of patients, the extent of resection and the question of whether the intussusception should be reduced remain controversial. Intussusceptions occur when one segment of the gastrointestinal tract (intussusceptum) telescopes into the lumen of an adjacent distal segment of the gastrointestinal tract (intussusciens). Rarely a distal segment of the bowel telescopes into the lumen of the adjacent proximal segment which is known as retrograde intussusception. Intussusception is a relatively common cause of intestinal obstruction in children but a rare and uncommon clinical entity in adults. Adult intussusceptions is a rare entity accounting for only 5% of all intussusceptions and causes approximately 1% of all adult intestinal obstructions. Adult intussusception is usually caused by a tumour acting as the apex of the intussusception.^[4] In both small and large bowel intussusception, lipoma is the most common benign tumour. In adults, intussusception is commonly associated with underlying pathology. While the condition is clinically non-specific, the intestinal obstruction is found to be a common presentation. Adult patients mostly complain of obscure abdominal pain only. In adults, there is a lack of classical triad of abdominal pain, palpable abdominal mass and passage of 'red currant jelly' stools, but these are commonly found in children. Intestinal lipoma is an uncommon causation of adult intussusception and some lipoma may cause intussusception by acting as the lead point located in the ileum. The diagnosis of intussusception is readily suggested because of its pathognomonic appearance on computed tomography.^[5]

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On physical examination, he appeared in good general condition, was normothermic, and had a slightly distended abdomen, which however was soft and non-tender. Bowel sounds were exaggerated. On digital rectal examination, the rectum was found to be empty with the presence of rectal ballooning. Laboratory results were all within normal range. A provisional diagnosis of acute intestinal obstruction was made which is confirmed on plain X-ray of the chest and abdomen erect and supine which showed grossly dilated small bowel loops and multiple air fluid levels. A subsequent computerized tomography scan of abdomen and pelvis revealed a well-defined sausage shaped intraluminal mass in the terminal ileum, showing a target sign with proximal dilatation of jejunal, ileal loops with collapsed distal ileal loops suggesting of intestinal obstruction.

After resuscitating the patient with intravenous fluids and antibiotics, he was taken up for surgery. On exploratory laparotomy, an invagination of segment of ileum into distal segment of ileum of about 30 cm from the ileocaecal junction was present with proximally dilated and distally collapsed ileal loops (Figure 1).



Fig. 1: Intraoperative image showing ileo-ileal intussusception

There was no other organomegaly lymphadenopathy or free fluid. The intussusception was reduced by gentle traction and retrograde pressure from the apex. There was a smooth nodule on the lateral wall of the terminal ileum was identified. Rest of the gastrointestinal tract appeared normal. Resection of involved segment followed by end to end ileo-ileal anastomosis was performed. A polypoid mass, measuring 4× 3 cm was found which is acting as a lead point is identified and sent for histopathological examination (Figure 2).



Fig. 2: Intra operative image showing polypoid mass

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Post-operative period was uneventful. Patient was discharged in good general condition on 10th post-operative day.

DISCUSSION: Since its first description in 1674 by Barbette intussusception has been considered to be a disease of infancy and early childhood. Adult intussusception is distinct from pediatric intussusception. It is rare, the condition being found in less than 1 in 1300 abdominal operations and 1 in 100 patients operated for intestinal obstruction. The child to adult ratio is nearly 20:1.^[6] In contrast to intussusceptions in children where nearly 80% are idiopathic, a demonstrable etiology is found in nearly 90% of cases in the adult population.^[7] This necessitates resection in adults as against reduction in children.

Intussusception is a rare clinical entity in adults.^[8] The mechanism of intussusception is unclear.^[9] Male-to-female ratio is 1:1.3 and mean age of presentation is 54.4 years of adult intussusceptions. The adult intussusceptions are classified in three steps with aetiology, namely benign, malignant or idiopathic.^[10] Intussusception could be either idiopathic type or secondary type. In idiopathic type, intussusception occurs without a lead point lesion. In secondary type intussusception, mostly organic lesions have been implicated.

Intussusceptions are classified according to location into: enteric, colonic, and ileocaecal or ileocolic.^[11] Enteric and colonic intussusceptions are those that are confined to the small and large intestine, respectively. Ileocolic intussusceptions are defined as those in which ileum prolapses through the ileo-caecal valve into the colon, and these constitute 15% of all intussusceptions. The ileo-caecal valve and the appendix preserve their normal anatomical position, and the organic lesion is usually in the ileum. Our case was one where a part of terminal ileum telescoped into the distal ileum and the whole intussusception then went through the ileo-caecal valve into the caecum and up to the ascending colon.

Small intestinal tumors are rare, accounting for 1-2% of all gastrointestinal tract tumors.^[12] Among these, benign tumors are still more rare and account for approximately 30% of all small bowel tumors. The lipomas are rare benign tumors, representing 2.6% of nonmalignant tumors of the intestinal tract. The incidence of intestinal lipomas has been reported between 0.15% and 4.4%. Intestinal lipomas usually occur in older persons, with a slightly increased incidence in females. After gastrointestinal stromal tumors, lipomas constitute the second most common benign-tumor group.^[13] Most occur in colon which constitutes from 65% to 75% of cases in comparison with small intestine which constitutes from 20% to 25%. In the small bowel terminal ileum is the commonest site for lipomas.

Although they are usually asymptomatic, lipomas larger than 2 cm may cause bowel obstruction, intermittent nonspecific abdominal pain, diarrhea, or bleeding. Furthermore, some lipomas by forming a lead point may cause intussusception, as well. Adult intussusceptions present with nonspecific obstructive symptoms like nausea, vomiting, and abdominal pain. Other symptoms may also be present such as melena, weight loss, fever, constipation, diarrhea, and abdominal mass.^[7] In 20% to 50% of cases of adult intussusception, the etiologic agent is a malignancy.^[9]

Since the clinical picture is vague, varied, and nonspecific, preoperative diagnosis of adult intussusception is rare and essentially radiological. Plain skiagram of the abdomen may reveal

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features of acute intestinal obstruction. On barium enema colonic lipomas appear as circular, ovoid, well demarcated, and smooth radiolucent masses (because of presence of fat). They show "squeeze sign" due to their fluctuation in size and shape.^[14]

Ultrasonography is often used to evaluate suspected intussusception as it is cheap, readily available, and noninvasive. The classic features include the "target and doughnut sign" on transverse view and the "pseudokidney sign" in longitudinal view. The major disadvantages are operator dependency and difficulty in image interpretation in presence of air, which is often present in cases of obstruction. The preoperative diagnostic accuracy of ultrasonography is 78.5%. In cases of palpable abdominal mass, the diagnostic accuracy of ultrasonography is even better 86.6%.

CT scan has been reported to be the most useful imaging technique, with a diagnostic accuracy of 58%–100% and a specificity of 57–71%.^[15,16] On CT, lipomas are seen as homogenous masses, well-circumscribed, ovoid, or round with sharp margins. In addition, they demonstrate characteristic attenuation values between –40 and –120 HU typical of the fatty composition. The CT findings of intussusception are a mass-like lesion, including the inner intussusceptum, an eccentric fat density mass that represents the intussuscepted mesenteric fat, and the outer intussusciens, and this appears as a "target" or a "sausage" mass according to imaging plane.^[17] CT is excellent in revealing the site, level, and cause of intestinal obstructions and in indicating possible bowel ischaemia. It can give additional information, such as metastasis or lymphadenopathy, which may indicate an underlying pathology.^[15] Endoscopy can show a smooth yellow surface with a pedunculated or sessile base or either the "cushion sign" or "naked fat sign".^[16]

In view of the uncertain aetiology and diagnosis and high incidence of malignancy (approaching 50%), the treatment of intussusception in adults is invariably surgical resection. However, the extent of bowel resection and the manipulation of the intussuscepted bowel during reduction remain controversial.^[8,10,15] In contrast to pediatric patients, where intussusception is primary and benign, preoperative reduction with barium or air is not suggested as a definite treatment for adults.^[18] The theoretical risks of preliminary manipulation and reduction of an intussuscepted bowel include (1) intraluminal seeding and venous tumor dissemination, (2) perforation and seeding of microorganisms and tumor cells to the peritoneal cavity, and (3) increased risk of anastomotic complications of the manipulated friable and edematous bowel tissue.^[15,19] Moreover, reduction should not be attempted if there are signs of inflammation or ischemia of the bowel wall and at age above 60 years.^[19] However, several others believe that the risks are theoretical, and gentle reduction should be attempted in selected cases to avoid unnecessary resection of healthy bowel.^[20] Endoscopic resection of colonic lipomatous polyps and laparoscopic resection of benign bowel tumors causing ileal and/or ileo-colic intussusception has a role in very selected settings.^[15,20]

CONCLUSION: Adult intussusception is a rare but well-recognized condition. A high index of suspicion and early diagnosis with a CT scan will identify patients requiring emergent surgery and thus prevent serious complications such as haemorrhage, intestinal gangrene, perforation and obstruction. This case, as well as a review of the literature, showed that a missed initial diagnosis

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of intestinal intussusception in adults can delay proper treatment and cause serious consecutive complications. Therefore, early surgical treatment is needed regardless of the etiology.

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