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Case Report

Rare case presentation of intestinal intussusception due to leiomyosarcoma

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ABSTRACT

Intussusception is defined as one segment of bowel telescopes into an adjacent bowel segment, which can cause obstruction and even intestinal ischemia. Multiple complications such as intestinal obstruction, intestinal necrosis, and sepsis can occur as result if not treated early. Intussusception is much more common in the pediatric age group and rare in adults.in children it presents with the classic triad of cramping abdominal pain, bloody diarrhea, and a palpable tender mass. In children intussusception mostly likely occurs due to benign causes, whereas in adults cause of intussusception are more likely carcinomas, polyps, diverticulum, strictures, benign neoplasms, or postoperative condition. Most useful diagnostic tools are ultrasonography and computed tomography. We operated a case of adult intussusception due to leiomyosarcoma of small intestine which is even rare. on imaging, ileo-ileal intussusception with dilated proximal bowel loops were found. Exploratory laparotomy done in which large hard polypoidal mass causing ileo-ileal intussusception was found for which ileo-ileal resection & anastomosis done with normal bowel margin of 10 cm on both sides. Patient's post operative period was uneventful on histopathology, high grade leiomyosarcoma was detected.no clinical or radiological evidence of metastasis found.

Keywords: Intussusception, Leiomyosarcoma, Computed tomography, Malignancy, Resection & anastomosis

INTRODUCTION

Intussusception is a process in which a segment of intestine invaginates into the adjoining intestinal lumen.in which, proximal bowel segment telescopes into distal bowel segment in most cases. This condition is frequent in children and presents with the classic triad of cramping abdominal pain, bloody diarrhea and a palpable tender mass. However, bowel intussusception in adults is considered a rare condition, accounting for 5% of all cases of intussusceptions and almost 1%-5% of bowel obstruction.[1,2]

Generally, intussusception in children arise from an unknown cause. They might also arise due to infections, meckel's diverticulum, duplication cysts, polyps, appendicitis, hyperplasia of peyer patches, idiopathic, anatomical factors and altered motility. About 90% of intussusceptions in adults are caused by a definite underlying disorder such as carcinomas, polyps, diverticulum, strictures, benign neoplasms or postoperative condition: which are usually discovered intraoperatively.[3] However, neoplasm is the most common cause and is found in approximately 65% of adult cases.[4]

Malignant tumors are more common cause of intussusception than benign tumors in the colon, although the opposite is seen in the small bowel intussusception.[5] The most common symptoms of intussusception are abdominal pain, nausea, and vomiting; less frequent symptoms are blood-mucus in stool, weight loss, fever, lethargy and constipation. Symptoms are usually of long duration usually from several weeks to several months, although the patient may occasionally present with an acute abdomen and palpable abdominal lump.[6] Diagnostic modalities include plain X-ray abdomen, ultrasound imaging, contrast enhanced CT scan and

fluoroscopy (Barium study) are done. In pediatric age group where intussusception is idiopathic or no lead point is found, hydro or pneumoreduction is tried. But if it fails to relieve intussusception or where there is possibility of lead in both children or adult, surgical intervention should be done.in which simple reduction or resection of diseased bowel is done depends upon intraoperative findings.

We presented a rare case of ileo-ileal intussusception in an adult patient with intestinal obstruction caused by a rare mesenchymal malignant lesion of the distal ileum: high grade leiomyosarcoma.

CASE REPORT

A 58-year-old male is presented to emergency room with complaint of generalized abdominal pain since, 10 days which aggravates on food intake associated with vomiting since last 5-6 days, significant weight loss with anorexia over the past one-month, postprandial fullness and constipation since last 3 months. Patient is hypertensive since last 9 months and chronic tobacco chewer since, 40 years.

There was no past history of similar complaints. there was no significant personal and family history.pn physical examination, patient is vitally stable. On abdominal examination, abdomen is soft and distended. Blood reports are within normal range. Patient's Hb is 11.7 g/dl, WBC counts were 7890/22. X-ray abdomen is suggestive of air and fluid filled bowel loops noted in right lumbar and right iliac fossa region.

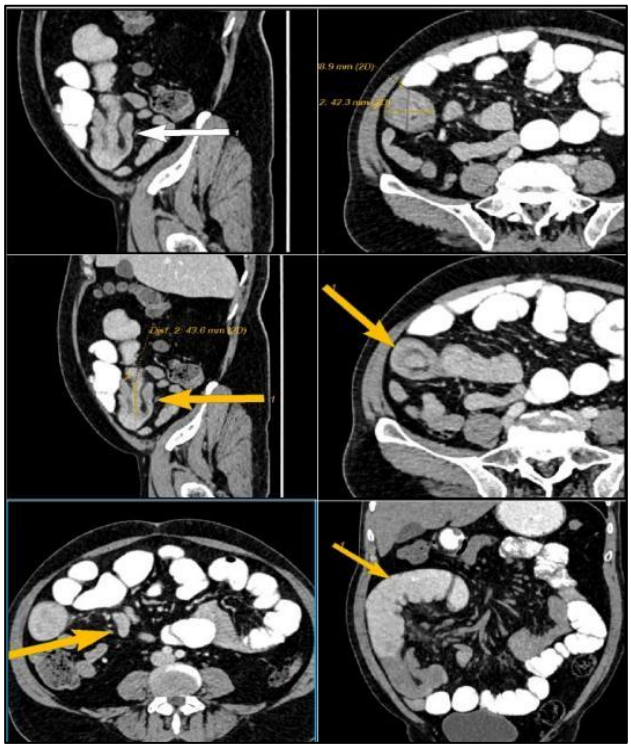

Figure 1: Computed tomography of abdomen.

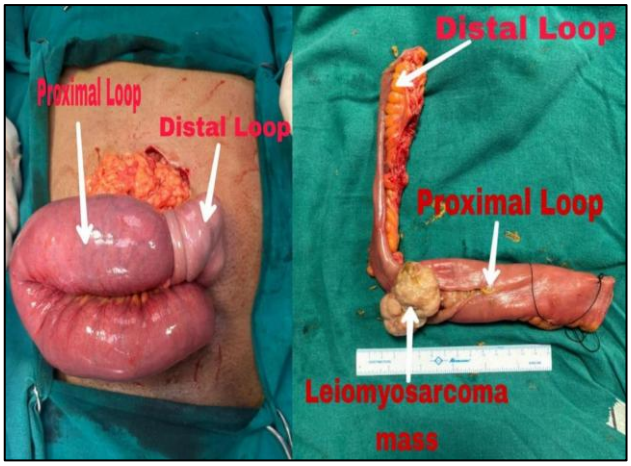

Figure 2: Intussusception due to leiomyosarcoma growth in ileum.

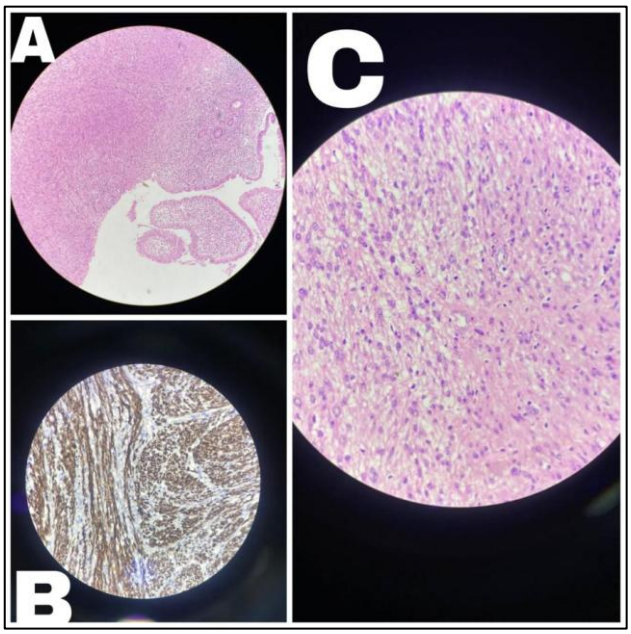

Figure 3: Mucosal involvement (A) nuclear pleomorphism with (B) mitosis (C) Desmin positive.

Ultrasonography is suggestive of bowel within bowel appearance showing target sign measuring approximately 38×52 mm (AP×TR) noted in right iliac fossa, with intussusceptum being one of the ileal loops and intussuscipiens being one of the small bowel loops (ileal). Bowel loops show preserved vascularity. Wall thickness of ileal loop (intussuscipiens) measures approximately 4.5 mm, minimally edematous.

Above findings suggest possibility of ileo-ileal intussusception. Mild free fluid noted in interbowel region in right iliac fossa. Contrast enhanced CT scan of abdomen shows an ideal loop in the right iliac fossa shows intra-luminal herniation of its proximal segment into its distal segment giving bowel-within-bowel

appearance suggests ileo-ileal intussusception, no evident lead point is identified.

Length of intussusception is 43 mm. Diameter of intussusception is 42×39 mm. Resultant dilatation of proximal ileal loops and jejunal loops is noted having maximum transluminal diameter about 4.8 cm. Few enlarged & sub centimeter sized non-necrotic mesenteric lymph nodes are noted in paraumbilical region, left hypochondriac region and in right iliac fossa; largest 21×14 mm in size (Figure 1).

DISCUSSION

Patient was operated on next day, in which exploratory laparotomy was done, in which approx. 45 cm proximal from ileocecal junction, proximal ileal loop is found telescoping into distal loop. intussuscepting part of ileal loop lumen relieved approx. 6×4×3 cm sized hard polypoidal mass felt inside lumen. Enlarged lymph nodes found in mesentery of ileum. Approximately 25 cm of part of distal ileum 35 cm proximal from ileoceal junction resected and sent for histopathological examination with 10 cm of normal bowel is resected on both side of polypoidal mass, then ileo-ileal anastomosis done. Specimen opened and 6×4×3 cm polypoidal mass found inside lumen of ileal loop (Figure 2). Patient's postoperative period went uneventful and patient was discharged on 9th post operative day. Patient came regularly in follow-up; oncology opinion was taken and suggested no active intervention required at present and follow up after 6 months suggested.

Histopathology is suggestive of high grade leiomyosarcoma proximal and distal margin is free from tumor. Mucosa is involved, tumor is just adjacent to serosa lymphovascular invasion is not seen, pathological staging: T1N0MX, tumor shows desmin-positive, CD-117/C-KIT-negative, DOG-1-negative, s-100-negative, ALK/CD246 - negative, KI 67-30% (Figure 3).

It is generally believed that masses in the bowel or lumen act as an irritant and provoke abnormal peristaltic movement, which may lead to the telescoping of one bowel segment over the adjacent segment. the prolapsing part of the bowel is described as the intussusceptum, while the distal segment of bowel receiving the intussusceptum is described as the intussuscipiens. Intussusception appears as a complex soft-tissue mass consisting of the outer intussuscipiens and the central intussusceptum. any tumor acting as the lead point of an intussusception may be outlined distal to the tapered lumen of the intussusceptum.

the classic pediatric presentation of acute intussusception is a triad of cramping abdominal pain, bloody diarrhoea and a palpable tender mass. But also present with nausea, vomiting (green from bile), pulling legs to the chest. Pain is intermittent because the bowel segment transiently stops contracting. Later signs include rectal bleeding, often with "red currant jelly" stool, and lethargy. physical examination may reveal a "sausage-shaped" mass. children may cry, draw their knees up to their chest, or experience dyspnea with paroxysms of pain.[7]

Classic pediatric presentation is rare in adults. The presenting symptoms are nonspecific and the majority of cases in adults have been reported as chronic, consistent with partial obstruction. Nausea, vomiting, gastrointestinal bleeding, change in bowel habits, constipation or abdominal distension, lethargy, intermittent colicky abdominal pain are the nonspecific symptoms and signs of intussusception.

Acute presentation appears as sudden increase in abdominal pain, nausea, vomiting, abdominal distention, palpable abdominal lump or blood in stool/melena.[9] This occurs due to intestinal obstruction and inflammatory changes ranging from thickening to ischemia of the bowel wall which occurs due to incorporation of mesentery into the intussusception. Thus, venous return is compromised, resulting in edema and further restriction of blood flow. Eventually, arterial supply to the bowel is interrupted, lead to ischemia and necrosis of involved bowel segment.

The intestinal mucosa is extremely sensitive to ischemia because it is farthest away from the arterial supply. ischemic mucosa sloughs off, leading to the heme-positive stools and subsequently to the classic "currant jelly stool" (a mixture of sloughed mucosa, blood, and mucus). if untreated, transmural gangrene and perforation of the leading edge of the intussusceptum may also occur.

Generally, intussusception in children arise from an unknown cause. They might also arise due to infections, meckel's diverticulum, duplication cysts, polyps, appendicitis, hyperplasia of peyer patches, idiopathic, anatomical factors and altered motility. About 90% of intussusceptions in adults are caused by a definite underlying disorder such as carcinomas, polyps, diverticulum, strictures, benign neoplasms or postoperative condition. which are usually discovered intraoperatively.[3]

Lead points include meckel diverticulum, colonic diverticulum, enlarged mesenteric lymph node, benign or malignant tumors of the mesentery or of the intestine including lymphoma, polyps, ganglioneuroma, hamartomas associated with peutz-jeghers syndrome, mesenteric or duplication cysts, metastasis from various primary sites such as the lung or breast, submucosal hematomas which can occur in patients with henoch-schönlein purpura and coagulation dyscrasias, ectopic pancreatic and gastric mass/tissue, postoperative intussusceptions (inverted appendiceal stumps, sutures and staples along an anastomosis, intestinal hematomas secondary to abdominal surgery) or hematomas secondary to abdominal trauma, foreign body (including indwelling catheters such as feeding jejunostomy tube),

hemangioma, kaposi sarcoma, post-transplantation lymphoproliferative disorder (PTLD) and cystic fibrosis.[10-12] Intussusceptions have been classified according to their locations into four categories that is entero-enteric, confined to the small bowel, colo-colic, involving the large bowel only, ileo-colic, defined as the prolapse of the terminal ileum within the ascending colon and Ileo-cecal, where the ileo-cecal valve is the leading point of the intussusception, appendiceal intussusception, all these types of intussusceptions occurs due to causes/factors mentioned above.[5,6,9]

Gastric intussusception is a rarely documented condition that occurs secondary to a mobile gastric tumor that prolapses into the small bowel mainly duodenum.[6] Various gastric lesions including adenoma, leiomyoma, lipoma, hamartoma, inflammatory fibrinoid polyp, adenocarcinoma, and leiomyosarcoma can serve as lead points.[7] Rectal intussusception is a concentric invagination of the entire rectum that progresses toward the anal canal but does not protrude through the anus.[13] Ileocolic >ileoileocolic >ileoileal >colocolic. In which ileocolic intussusception is most common (75-95%), presumably due to the abundance of lymphoid tissue related to the terminal ileum and the anatomy of the ileocecal region. In children there is a strong predilection for the ileocolic intussusception.

Laboratory investigation is usually not helpful in the evaluation of patients with intussusception. After obtaining a thorough history and performing a careful physical examination, obtain plain radiographs of the abdomen with the patient in the supine and upright positions. Which may demonstrate an elongated soft tissue mass (typically in the upper right quadrant in children) with bowel obstruction (and therefore air-fluid levels and bowel dilation) proximal to it. There may be an absence of gas in the distal collapsed bowel, perforation may also be identified.

Ultrasonography is considered a useful tool for the diagnosis of intussusception, both in children and in adults, found to have a high sensitivity and specificity in the detection of ileocolic intussusception. Ultrasonography has a false-negative rate approaching zero and is a reliable screening tool for children at low risk for intussusception.[14,15] ultrasonographic finding includes target sign (also known as the doughnut sign), pseudo kidney sign, crescent in a doughnut sign, absent blood flow in the intussusceptum (a contraindication to enema reduction), free fluid within the intussusceptum, a lead point within the intussusceptum mass. CT has become the modality of choice for assessment of acute abdomen in adults, and thus most frequently detects intussusception. The appearance of intussusception on CT is characteristic and depends on the imaging plane and where along the bowel the images are obtained. CT scan findings are not reliable as ultrasonographic imaging.in CT imaging, lead point/mass lesion might be missed but can give details about bowel wall viability, changes of ischemia and length of intussusception.it also helps to identify other possible pathology which helps in correlation. A contrast enema remains the gold standard, demonstrating the intussusception as an occluding mass prolapsing into the lumen, giving the "coiled spring" appearance (barium in the lumen of the intussusceptum and in the intraluminal space) and has the potential to be therapeutic. the main absolute contraindication for an enema is perforation, which needs to be treated surgically. Flexible endoscopy of the lower GI tract is considered invaluable in evaluating cases of intussusception presenting with subacute or chronic large bowel obstruction.[16] confirmation of the intussusception, localization of the disease and demonstration of the underlying organic lesion serving as a lead point are the main benefits of endoscopy.

treatment regimen is slightly different in pediatric and adult age group for intussusception.in pediatric patients, where intussusception is primary and benign, preoperative reduction with barium (hydro reduction) or air (pneumoreduction) is tried before surgical management. Reduction techniques are useful in idiopathic intussusception where there no identifiable lead point. Patient with lead point mostly require surgical intervention in both age groups. The presence of peritonitis and any evidence of perforation revealed on plain radiographs are the only 2 absolute contraindications to an attempt at nonoperative reduction with a therapeutic enema. If non-surgical methods fail, symptoms persist or rectal blood is present, surgical management is evident. In which entry into the abdomen is through a right paraumbilical incision. deliver the intussusception into the wound and attempt nonoperative reduction. milking the intussusceptum out of the intussuscipiens is important. sustain gentle manual pressure rather than pulling out the intussusceptum to avoid risk of iatrogenic perforation. If manual reduction is not possible or perforation is present, perform a segmental resection with an end-to-end anastomosis.

laparoscopy can be performed in all cases of intussusception. reduction of the intussusception, confirmation of radiologic reduction, and detection of lead points have all been reported.[18] Theoretical risks of preliminary manipulation and reduction of an intussuscepted bowel include intraluminal seeding and venous tumor dissemination, perforation and seeding of microorganisms and tumor cells to the peritoneal cavity and increased risk of anastomotic complications of the manipulated friable and edematous bowel tissue as in adult age group, malignant lead point is possible.[19,20] so precaution should be taken. Reduction should not be attempted if there are signs of inflammation or ischemia of the bowel wall.[21] Therefore, in patients with ileo-colic, ileo-cecal and colo-colic intussusceptions, especially those more than 60 years of age, due to the high incidence of bowel malignancy as the underlying etiologic factor, formal resections using appropriate oncologic techniques are recommended, with the

construction of a primary anastomosis between healthy and viable tissue.[22] If frozen section facility is available or possibility of malignant mass is there, minimum 10-15 cm of normal bowel is resected on both side of tumor/malignant lead point. In post operative period, after patient is tolerating diet and prepared for discharge, further treatment should be planned as per histopathology report.

CONCLUSION

As leiomyosarcoma itself is a rare malignant tumor of gastrointestinal tract, intussusception due to leiomyosarcoma growth is even rare. Thus, we presented a case of the same and elaborate its management. surgical management still remains mainstream treatment for the same. Postoperatively, chemotherapy-radiotherapy depends upon patient's postoperative condition. Follow up CT scan, pet CT is advised once patient becomes better.

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