



A RARE CASE SERIES OF ADULT INTUSSUSCEPTION IN RURAL AREA : CHALLENGE IN DIAGNOSING AND MANAGEMENT UPDATE

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ABSTRACT **Background:** Adult intussusception is a rare pathology representing 5-16% of all cases and are responsible for 1-5% of all bowel obstruction. The classic symptoms as found in children are often not present thus delaying diagnosis. The aim of this paper is to raise an awareness to the potential diagnosis and present a recent update in the management of adult intussusception. **Case:** We report 2 cases of adult intussusception with different clinical symptoms and leading point. Case 1, 53 years old man with a main clinical feature of progressing intermittent abdominal pain and no sign of obstruction and case 2, 35 years old woman presenting bowel obstruction. In both patients, laparotomy confirmed the diagnosis of ileoileal and ileocolic intussusception. Pathological finding was chronic colitis with *A. Lumbricoides* infestation and intact polypoid lesion of cavernous haemangioma in caecum. Both patient underwent resection without prior reduction and anastomosis. Early feeding was given 24h post surgery with good tolerance and was sent home after 5 days of hospitalization. **Conclusion:** Adult intussusception is a rare condition with non-specific clinical symptoms which may further delay the diagnosis thus giving rise to greater challenge especially in rural areas with low resources. Surgery is the definitive treatment for adult intussusception. Several recent literature show that reduction of intussuscepted bowel is considered safe in benign lesion and if there was no sign of bowel ischemia in order to avoid short bowel syndrome. Early feeding post operatively is considered safe according to ERAS.

KEYWORDS : Adult intussusception, Bowel Obstruction

INTRODUCTION

Intussusception was first reported by Barbet of Amsterdam in 1674, and in 1789 John Hunter present it in further detailed report as "introsusception". Intussusception is defined as invagination of a proximal portion of the bowel and its mesentery (intussusceptum) into a more distal segment (intussusciptiens).⁽¹⁾ This condition is common in children, but are a rare condition in adult 5-16% of all cases of intussusception. and responsible 1-5 % of all bowel obstruction.^(2,3) Unlike children with the "classic" triad of abdominal pain, vomiting, and currant jelly stools, the clinical presentation of intussusception in adult often non-specific that make delay for diagnosis and prompt treatment. Patients with intussusception may or may not be symptomatic and symptoms can be acute, intermittent, or chronic which make clinical diagnosis difficult until exploratory laparotomy was performed.⁽⁵⁾ The pathomechanism is thought to involve altered bowel peristalsis at the intraluminal lesion, which is then a lead point for the intussusception.⁽²⁾

With the advancement of science and technology, this has led to the development of few adjunct options to further assist in diagnosing adult intussusception such as plain abdominal radiography, abdominal ultrasonography, CT Scan, barium enema and MRI. CT Scan has become the gold standard for diagnostic tools to determine the causative factor of adult intussusception along with further management.^(3,4) Further recent studies also mention the use of milking procedure prior to resection of segmental intussusception to reduce the risk of short bowel syndrome after malignancy can be ruled out.⁽⁵⁾ The management does not end at the operative procedure, but further on to the postoperative phase.⁽⁶⁾ Physical condition and the operative procedure by itself possess a challenging phenomena especially in rural areas whereby nutrition is still a primary problem in those areas, therefore the lack of parenteral nutrition has to be done by early oral feeding.

Diagnosing adult with intussusception are challenging especially in rural area with limited resources and facilities but early diagnosis and treatment was important to reduce morbidity. The aim of this paper is to raise awareness to the potential diagnosis and recent update management of adult intussusception.

Case Report 1

A man in his mid- 50s initially presented to the outpatient department with abdominal pain of 1 month duration. The abdominal pain was intermittent, described as abdominal pain and located at the right quadrant and migrating to the flank. The Patient has already been treated with several analgesic and gastric medication but failed to

alleviate the symptoms. The patient also complained of fever and vomiting. The patient denied weight loss, changes in bowel habits and urinary symptoms. On physical examination the patient was in moderate discomfort; holding his abdomen. On palpation there was distended abdomen, tenderness at right lower quadrant and suprapubic region. The patient has normal bowel sounds. No masses were palpable. His medical history revealed no remarkable feature. His body temperature was 37,7 degrees celcius. Laboratory findings were as follows: hemoglobin level 9,5 g/dL; white blood cell count 16.600; platelets 488.000. Urinalysis revealed leukocyturia. Abdominal radiography was normal. An ultrasound was performed and revealed an intraluminal mass 'bull's eye' sign which is suggestive intussusception. He underwent explorative laparotomy. Under general anesthesia, the abdominal cavity was entered through a midline incision. An ileoileal intussusception was found located 260 cm from Treitz ligament and proximal segment was 60 cm from the ileocaecal valve. There were also enlargement of mesenteric lymphnode. Segmental resection without reduction of the intussusception was performed at the fundus followed by primary anastomoses with continous Polyglycolic Acid (PGA) 3/0 and secured with Lembert suture. The resected segment of the ileum measured 29 cm and three intraluminal living ascaris were evacuated. Macroscopic ileum specimens measuring length of 7,3 centimeters was sent for pathologic evaluation. Microscopic evaluation revealed part of mucosal erosion with necrosis, extravasation of eritrosit and vascular dilatation. There was no sign of malignancy and the final histopathology diagnosis was Chronic Colitis. By 24 hours postoperative procedure, we revealed a positive flatus and bowel movement therefor was started on a liquid diet. If these were tolerated well thus it was changed to normal diet. The patient was discharged after the fifth day when there was no sign of abdominal distension and was able to tolerate a normal diet. The patient scheduled and was met at the outpatient department and showed positive outcome and recovery.

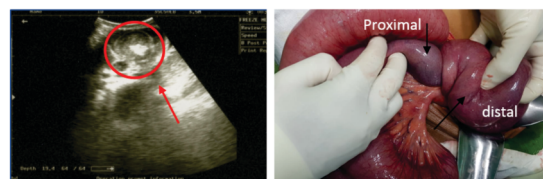


Fig 1. Abdominal Ultrasonography Fig 2. Finding at laparotomy of ileoileal intussusception showing "bull's eye" sign of ileoileal intussusception

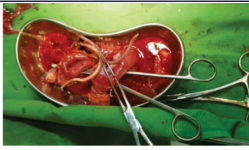


Fig 3. Three pieces of living *A. Lumbricoides* on Resected Segment of Ileoileal Intussusception



Fig 4. Macroscopic finding of ileum was sent for pathological evaluation

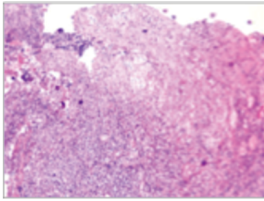


Fig 5. Histopathology Finding Showing Chronic Colitis

Case Report 2

This 32-year-old woman presented to the emergency room with main complaint of abdominal distention for 2 days. She also present symptoms of profuse vomiting and lethargy. She has a History of crampy, and progressing abdominal pain since 1 week located at the umbilicus and watery stool mixed with bloody mucus since 3 days. She did not have fever, hematemesis, and weight loss except nausea during 3 weeks. On physical examination, the patient was lethargic in moderate discomfort. On palpation there was distended abdomen, tenderness in all quadrants of the abdomen. There was an absent bowel sound, but positive meteorite sign. No masses were palpable. Vital signs were normal. Laboratory findings were as follows : hemoglobin level 12 g/dL; white blood cell count 7.200; platelets 386.000; Abdominal radiography showed dilated colon and air fluid level. The patient underwent an emergency laparotomy. Upon exploration, there was telescoping of ileum, 330 cm distal to the ligament of Treitz. Segmental resection without prior reduction of the intussuscepted ileum until 2/3 transverse colon was performed and followed by end to end ileotransverse colonic anastomosis with continuous suture with PGA 3/0 and secured with Lembert suture. A polypoid lesion, measuring 3 centimeters in diameter was noted at the caecum and was proposed to be the leading point of intussusception. There was also several mesenteric lymphadenopathy. Histologically, this unencapsulated polypoid lesion consisted of multiple various blood-filled spaces which were lined by thin endothelial cells and separated by scant connective tissue stroma. The final diagnosis is cavernous hemangioma causing intussusception of the ileum. By 24 hours postoperative procedure, we revealed a positive flatus and bowel movement therefor was started on a liquid diet. If these were tolerated well thus it was changed to normal diet. This patient was discharged without immediate complications on the sixth day. The patient scheduled and was met at the outpatient department and showed positive outcome and recovery.



Fig 6. Abdominal X-ray revealed distended bowel loops and air fluid level



Fig 7. Macroscopic presentation of resected segment of intussusception showing a polypoid lesion

DISCUSSION

There are few reports of intestinal intussusception in adults, and the reported series are small and cover long time periods. Most common initial signs include abdominal pain, vomiting, distention, and occasionally palpable masses. An acute condition often occurs, accounting for 5% of intestinal occlusions seen at emergency rooms, but may sometimes have a subacute or chronic nature causing intermittent abdominal pain and vomiting. According to several literature, the most common symptoms is just acute abdominal pain.^(7,8) Several studies reported that both acute and chronic pain is even higher in intussusception, so diagnoses are often delayed^(8,9). Intussusception classified by location (enteroenteric, ileocolic, ileocaecal, or colocolic), and cause (benign, malignant, idiopathic). Further, intussusception in an adult can classified also on the basis of whether a lead point is present.⁽⁹⁾ While, an intussusception without a lead point is tends to be transient. Transient intussusception of the small bowel has been reported in adults with celiac disease and Chron's disease. Ileoileal or colonic intussusception is uncommon in infants but is frequent in adult. Childhood intussusception is idiopathic in 95% cases, while up to 90% of occurrence in adults have a leading point, which is defined as a well definable pathological abnormality such as neoplasm.⁽¹⁰⁾ The most common benign lesions in the small bowel include adhesions, Meckel's diverticulum, submucosal lipoma and polyps. While malignant lesions in the small bowel account for 20-50% of all intussusceptions and are usually due to metastases from tumors such as melanoma, lymphoma, and so on. Traditionally, Resection without prior reduction has been the traditional treatment of choice due to the significant risk for malignancy in most series.^(11,12)

In this paper, we describe 2 case of adult intussusception with different presentation and leading point. In first case, we describe an unusual case of ileoileal intussusception as an initial manifestation of Chronic colitis. There was ileoileal intussusception with attributable chronic colitis, mesenteric lymphadenopathy, and *Ascaris Lumbricoides*. Adult intussusception in the setting of inflammatory bowel disease (IBD) is a rare phenomenon.^(13,14) Some literature speculate that intussusception was secondary to inflammatory edematous mucosa and spasm, which led to impaired contraction of the bowel and allowed unbalanced peristaltic forces to rotate the intestinal wall inwards and initiate the invagination. In case two, there was an ileocolic intussusception with the lead point of intussusception was a small bowel cavernous hemangioma. Small bowel hemangioma accounts for 5-10% of all benign neoplasms of the small intestine⁽¹⁵⁾. These tumors may be solitary or multiple, and more frequent in jejunum.⁽¹⁶⁾ Intussusception secondary to cavernous haemangioma is extremely rare in an adult. Hemangioma are classified as a cavernous, capillary, or mixed type, and the cavernous type is the most common and very difficult to diagnose preoperatively. Malignant change is quite unusual. Most patients having small bowel hemangioma presented with abdominal pain and intestinal bleeding with acute or chronic anemia.^(15,17) In this case, after post resection procedure it was revealed a haemangioma in an understructable form. In many cases with small bowel haemangioma, there are frequent signs of abdominal pain and intestinal bleeding, but in this specific case, the bloody mucous faeces was thought to be from the necrotic part of the specific bolwe and not from the haemangioma itself.

Each of the above cases has manifested with a different clinical features. There was no obstruction sign in case 1 as compared to obstruction which was present on case 2. This approach to diagnosis of intussusception in adults and children is different. Diagnosing intussusception in adults is challenging due to varied presenting symptoms and time course. Frequently, patients may present with vague, chronic, and non specific symptoms as the above cases. Abdominal pain is the most common presenting complaint, being present in 71-91% cases.^(18,19) Chronic abdominal pain may be regularly be missed during emergency ward examination/admission which are often being given analgesics without relief of symptoms thus requiring futher and frequent visits to the emergency ward. Other common symptoms that patients may report are nausea, vomiting, changes in bowel habit, abdominal distention, and hematochezia, which varies in their timelines occurrence, being either acute, intermitent, or chronic, and further complicating the accuracy of the diagnosis. When intussusception is caused by a malignancy or an accompanying malignancy, features such as weight loss, melena, or palpable abdominal mass must be present.^(8,11)

Several adjunct may assist in the diagnosis of an adult intussusception. There are plain abdominal film, barium studies, abdominal

sonography, abdominal computerized tomography (CT), Magnetic resonance imaging.^(20,21) Typically, clinicians will use plain abdominal radiography and ultrasonography as their first imaging modality. Plain abdominal radiography will showing sign of bowel obstruction. While ultrasonography showing “target sign” or “doughnut like” shaped on transverse view and “pseudo-kidney sign” or “Hayfork sign” on longitudinal view.^(22,23) Although varies in diagnostic imaging, Abdominal CT scan provides the best accurate diagnostic rate in adult intussusception. It's accuracy reportedly ranges from 58-100% according to various studies.⁽³⁾ CT often demonstrates an early target mass with the fascial planes around the mass retained. This can include bowel thickening and the characteristic mass with layering effect (area of high density with curvilinear low-density zone). Several literature show that CT Scan also help differentiate between lead point and non-lead point intussusception.^(4,24)

The treatment of adult intussusception remains controversial. Most authors agree that surgery should be performed because of the high incidence of organic pathology. The controversial issue is, if surgery was done, is whether the intussusception should be reduced or not. This maneuver may help clearly define the limit of resection, and in specific cases where the casue is benign, the possibility of resecting the affected segment could be dismissed. But the problem is when there was malignant pathologic, reducing intussusception segment may lead to tumor seeding.⁽⁵⁾ Moreover, manipulation could manifest for further perforation and the seeding of microorganisms, and tumor to peritoneal cavity after exposing and handling the ischemic, friable, and edematous bowel tissue. This make preoperative diagnosis of intussusception become more important and challenging to choose an appropriate management for adult intussusception to reduce the morbidity rate.^(5,25) In general, several literature review and case report show that, the cases of colocolonic intussusception should be resected en-bloc without reduction, since there is a high risk of malignancy. Actually, entero-enteric intussusceptions account for the majority of cases in adults and small intestinal lesions are more often benign and it is reasonable to attempt reduction first unless there are sign of bowel ischemia or a suspected malignancy.⁽¹⁹⁾

Because of the limited resource of parenteral nutrition it was decided to start oral liquid feeding within 2 hours after surgery. feeding after surgery, where the diet initiated by filtrate liquid within 24 h after surgery. Over the next 24 h, the liquid diet was replaced by a normal diet in case tolerance was desirable and was continued if no vomiting or other complicaions were observed.⁽²⁶⁾ This condition differs from traditional method of diet given in post resection bowel anastomosis where late feeding were given after 4-5 days. Currently as suggested by ERAS, early feeding can lead to significant clinical efficacy such as, shorter time of ileus resolution after surgery, shorter overall hospital length of stay, and greater patient satisfaction with the treatment process.^(6,27,28)

CONCLUSION

Adult intussusception is uncommon in adult and is a rare cause of intestinal obstruction. Although uncommon, this diagnosis should be considered in patient with chronic abdominal pain. Several adjunct such as abdominal ultrasonography may be useful to assist diagnosing, but CT Scan is the gold standard because of high sensitivity and specificity, and its availability to distinguish a lead point, and also a malignant lesion. In adult, surgery is recommended for adult intussusception, and few recent literature suggest that reduction prior to bowel resection are allowable if lesion are benign.

In rural areas, diagnosing and management of adult intussusception become challenging because of low medical modalities (ex. Diagnostic tools and equipments). In this case, an en bloc resection was done because we can't eliminate malignancy. Other challenging factor postoperatively is the absence of parenteral nutrition thereby prompting to start early oral feeding as ERAS protocol, whis this will assist in an expeditied recovery.

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