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Case Report

Intussusception in Adult: Case Series and Literature Review

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ABSTRACT

Adult intussusception is rare and is an infrequent cause of intestinal obstruction. The clinical presentation is often non-specific and preoperative diagnosis can be very challenging. The mainstay of treatment is surgical resection because of the associated pathological gut lesion in most cases. However, controversy still exists vis-à-vis the extent of bowel resection and whether or not reduction should be attempted before resection. We present five patients seen in our center and a literature review.

Keywords: Adult Intussusception, Case series, Review.

INTRODUCTION

Intussusception is the invagination or telescoping of a segment of bowel into an adjacent one. It is a condition found commonly in paediatric population and it is the most common cause of intestinal obstruction in infancy and young children. [1-3] Adult intussusception accounts for less than 5% of all cases of intussusception and is an infrequent cause of intestinal obstruction (1-3%) which may be acute, subacute or even chronic in nature. [4-6] The clinical presentation in adults is often non-specific and the diagnosis could be challenging as the classic triad of crampy abdominal pain, passage of red currant jelly-like stool and palpable abdominal mass seen in children are rare thus leading to delay in diagnosis. [5,7] The aetiology of adult intussusception may be primary (idiopathic) or secondary. The latter constitutes the majority and the lesion may either be benign or malignant.

Imaging techniques like ultrasound scan, computed tomography (CT) or magnetic resonance imaging (MRI) and endoscopy (small bowel enteroscopy and colonoscopy) can be helpful in the preoperative diagnosis. Also, upper gastrointestinal (GI) series and barium enema can be used depending on the clinical situation. However, the diagnosis in some instances is often made intraoperatively. The mainstay of treatment is surgical operation either open or laparoscopic.

CASE SERIES

We present five patients seen between September 2016 and August 2017 in our center and a literature review. The five cases are summarized in Table 1 below. Informed consent was obtained from the patients together with the institutional approval for publication. The duration of symptoms ranged from 3 to 92 days (mean 28.8 Days). None of the patients had abdominal CT as a result of non-availability of this facility in our setting.

Table 1: Summary of cases

Case	Sex/Age (year)	Presentation	Classification	Diagnosis	Operation	Pathology
1	F/70	Generalized peritonitis	Ileocolic	Intraoperative	Right hemicolectomy	Idiopathic
2	F/54	Partial small bowel obstruction	Ileoileal	Intraoperative	Small bowel resection+ primary anastomosis	Idiopathic
3	M/40	Acute intestinal obstruction	Ileocolic	Intraoperative	Right hemicolectomy	Idiopathic
4	M/30	Acute on chronic abdominal pain	Caecocolic	Ultrasound scan	Right hemicolectomy	Adenocarcinoma
5	F/32	Acute small bowel obstruction	Ileoileal	Ultrasound scan	Small bowel resection + primary anastomosis	Benign polyp

Case 1

A 70 year woman who presented with colicky central abdominal pain of 2 weeks duration. It was said to be moderate in severity and was on and off. There were 2 episodes of vomiting and a history of diarrhea after taking a local concoction she prepared herself. She passes flatus freely and no significant abdominal distension. A day before presentation the pain became severe and generalized with associated fever. There was no preceding history of change in bowel habit, weight loss, bleeding per rectum or previous abdominal operation.

Physical examination revealed an ill-looking elderly woman with a temperature of 38^{o}C, respiratory rate 26/m, pulse rate 108/m and BP 120/70 mmHg but not pale. There was marked right iliac fossa tenderness, rebound tenderness with guarding. Rectal examination was insignificant. A provisional diagnosis of ruptured appendix was made. Abdominal scan revealed marked probe tenderness at right lower quadrant with no other significant finding. Chest X-ray was essentially normal.

Intraoperative findings were: ileocolic intussception with gangrene of the bowel and perforation around the ileocaecal junction. She had right hemicolectomy and ileotransverse anastomosis

She developed superficial surgical site infection that was managed by daily dressing and was discharged home on the 13th day of admission. Histology did not revealed any underlying pathology

Case 2

A 54 year old woman who presented with intermittent colicky periumbilical abdominal pain of 4 weeks duration associated with occasional bilious vomiting. The pain did not shift and there was no diarrhea, constipation, abdominal distension or weight loss. Patient had subtotal hysterectomy for symptomatic fibroid 10 years prior to presentation

Physical examination revealed a patient who was in no distress and vital signs were stable. Abdomen was full with midline infraumbilical scar. Further examinations were unremarkable. Plain abdominal radiograph was also unremarkable. A clinical diagnosis of post-operative adhesive sub-acute intestinal obstruction was made. She was managed conservatively and the symptoms subsided for 3 days and patient was commenced on graded oral intake. However she developed worsening symptoms two days later which warranted exploratory laparotomy and the findings was ileoileal intussusception. She had small bowel resection and primary anastomosis and was discharged 7 days postop. Histology did not reveal any pathological lesion.

Case 3

A 40 year old man who presented with anorexia, colicky central abdominal pain and bilious vomiting of 3 days duration. There was associated constipation, abdominal distension and fever. There was no history of passage of blood or mucus per rectum and no weight loss. He has had 2 similar episodes of the abdominal pain within 5 months of presentation. There was no past history of abdominal operation.

Physical examination showed an acutely ill-looking man with a pulse rate 98/m, respiratory rate 22/m and a temperature of 38.9^{0}C. The blood pressure was normal. The abdomen was distended

with tenderness at right iliac fossa and no mass was palpable. The plain abdominal radiograph demonstrated multiple air-fluid levels mainly at the central abdomen. Patient had laparotomy after resuscitation and the intraoperative finding was ileocolic intussusception. He had a right hemicolectomy with primary anastomosis. The histology of the specimen did not reveal any pathology.

Case 4

A 30 year old man who presented with colicky periumblical pain of 3 months duration. The pain later shifted to the right iliac fossa. There was associated anorexia and occasional vomiting but no fever. There was no abdominal distension, change in bowel habit or passage of bloody or mucoid stool andno previous abdominal surgery. On examination, there was a palpable mass about 6x7cm at the right iliac fossa which was confirmed by abdominal scan. The patient was admitted and managed conservatively and discharged home on the 5th day following improvement. He was seen at surgical outpatient department a week later and was advised to continue with antibiotics. However, patient defaulted and presented at a private hospital where he had appendicectomy with a marginal relief of symptoms. Six weeks after this operation, the symptoms recurred and he was then referred back to our facility. The repeat scan this time was diagnosed as intussusception. The exploratory laparotomy confirmed a caeco-colic intussusception with a 5x6cm mass in right iliac fossa. He had a right hemicolectomy and primary bowel anastomosis. Histology of specimen showed infiltrating colonic adenocarcinoma (AstlerColler B2).He is presently on chemotherapy.

Case 5

A 32 year old woman with colicky central abdominal pain of 6 days duration, bilious vomiting, constipation and fever of 2 days. There was no bleeding per rectum, weight loss, anorexia or previous operation. She delivered a baby 3 months prior to presentation.

The physical examination revealed an acutely ill-looking woman in painful distress. She was febrile (38.7^{0}C) with pulse rate 98/m, respiratory rate 20/m and BP 130/75 mmHg. The abdomen was full but no tenderness or guarding. An ill-defined mass was palpated extending from umbilicus to the right iliac fossa which was confirmed to be an intussusception on abdominal ultrasound scan. Rectal examination was unremarkable.

Findings at laparotomy confirmed a small bowel intussusception with the apex of intussusceptum about 25cm from ileocaecal junction (Figure 1). Ileal resection and primary anastomosis were performed and the recovery was uneventful. She was discharged home within a week and subsequent histology confirmed a benign polyp as the lead point.

Figure 1: Ileo-ileal intussusception with gangrene of the intussusceptum

DISCUSSION

Intussusception occurs infrequently in adults and accounts for about 1-3% of intestinal obstruction. Patients often have a wide range of non-specific and intermittent symptoms over a varied period of time making diagnosis to be difficult. There may be acute presentation within few days in one-half of patients while the other half presents with chronic symptoms that may span weeks, months or even years. [5,8,9]

Abdominal pain is the most common presenting symptom in up to 90% of patients. [10-12] Other non-specific symptoms include nausea, vomiting, fever, diarrhea, constipation, abdominal distension, gastrointestinal bleeding and weight loss. [13]

Some of the symptom complex may even masquerade as peptic ulcer disease. [14] In complicated cases with bowel gangrene or perforation, features of generalized peritonitis supervene leading to a more diagnostic dilemma. Late presentations, delays in making definitive diagnosis or instituting treatment are risk factors for the latter complications with attendant morbidity and mortality. [15]

Intussusception can be primary (idiopathic) or secondary. Unlike in children where the aetiology is largely idiopathic in more than 90% of cases, [16,17] idiopathic intussusception accounts for about 8-20% of cases in adult population. [5] However, our experience in this series showed that three were idiopathic as no lesion was found during histological examination of the specimen.

According to the locations, intussusceptions are classified into: entero-enteric, colo-colic, ileo-colic, and ileo-cecal. [18] Other rare ones include gastro-duodenal and colo-anal types.

Retrograde intussusception is another surgical entity which is seen infrequently. It may occur as a rare complication of gastric operations such as gastrojejunostomy, Billroth II gastrectomy, and Roux-en-Y anastomosis. [19] Its occurrence in other sites without prior surgery has also been described. [20] The exact mechanism for this is not entirely clear. However, many reasons have been postulated vis-à-vis motility disorder from the divided bowel, long afferent loop, jejunal spasm with abnormal motility, increased mobility of the efferent loop, adhesions leading to the intussusception of a more mobile segment into a fixed segment, increased intra-abdominal pressure and retrograde peristalsis. [19,21]

In adults with intussusception, 90% of it occurs in the small or large bowel while the remaining 10% involve the stomach or a surgically created stoma. The small bowel has been found to be the single most common site. [5,6,13,22] About 30% of cases of intussusception occurring in the small intestines are malignant while on the other hand up to 60-66% of intussusception occurring in the large bowel is more likely to have an underlying malignant aetiology. [4,6,22]

The diagnosis of intussusception in adults can be challenging and definitive diagnosis may be on the operating table. Plain abdominal X-rays are neither sensitive nor specific for intussusception and therefore not usually helpful as it may only show a soft tissue mass with or without features of bowel obstruction or perforation. [4,6] This is usually our first line diagnostic tool in patients with suspected acute intestinal obstruction. Three of our patients had this which revealed multiple air –fluid levels with no clear cut aetiology.

Ultrasonography in a skillful and experienced hand is diagnostic of intussusception but is less sensitive than CT. Ultrasonography may show a "target" or "doughnut" or bull's eye sign on the transverse view and the "pseudo-kidney" or "hay-fork" sign in longitudinal section. [4,23]

Abdominal CT is currently considered the modality of choice in adults because of its high diagnostic accuracy of 58–100%. [5,18] CT defines the location, the nature and the relationship of the lesion to surrounding tissues. The finding of a heterogeneous "target" or "sausage-shaped" soft-tissue mass with layering effect is virtually pathognomonic. [18] Kim et al reported that CT can facilitate distinguishing between intussusception without a lead point from that with a lead point which may be of value in determining the appropriate treatment and has the potential to reduce the prevalence of unnecessary surgical interventions. [24]

Upper GI series could also be used and may show a "stacked coin" or "coiled-spring" sign, while a barium enema examination may show a "cup-shaped" filling defect or "coiled-spring" sign in colocolic or ileocolic intussusception.[12] However, Azar et al reported that the accuracies of upper GI contrast series and

barium enema were 21 and 54% respectively. [5]

Colonoscopy and small bowel enteroscopy may be valuable in evaluating intussusception with subacute presentation or chronic large bowel obstruction. [25,26] Its main benefits are confirmation of intussusception, localization and biopsy to ascertain the diagnosis of the underlying organic lesion and hence decide on the possible treatment.

Surgical resection is the mainstay of treatment of adult patients with intussusception and the approach could either be through laparotomy or laparoscopy. Immediate exploratory laparotomy and bowel resection without reduction was the norm before the advent of diagnostic modalities. [5] However, recent studies recommend a more selective approach to bowel resection which takes into consideration the part and the severity of the bowel affected and pathologic characteristics of the underlying lesion whether benign or malignant.

Despite the fact that most surgeons favour surgical intervention in adult intussusception because of the underlying structural anomalies and the high incidence of malignancies, controversy still exists vis-à-vis the extent of bowel resection and whether or not reduction should be attempted before resection. There are concerns that bowel manipulation and reduction can lead to intraluminal seeding of malignant cells, gut perforation and increased risk of anastomotic complications of the friable and oedematous bowel. [12,27] Reduction should not be attempted if signs of bowel compromise are present. [18] Since most small bowel intussusception are benign, reduction could be attempted in order to allow for more conservative resection and avoid short bowel syndrome. [4] In this series all our patients underwent surgical resection and primary anastomosis with no attempt at reduction because of bowel oedema and perforation.

En bloc resections using appropriate oncological principles are recommended for colonic disease because there is a higher risk of malignancy as the underlying aetiology. [28,29] Nonoperative reduction is not advisable in adults because the majority of cases are associated with pathological lesion with a relatively high incidence of malignancy. [7]

CONCLUSION

Intussusception is an infrequent cause of intestinal obstruction in adults and preoperative diagnosis of this condition could be challenging. Definitive diagnosis may at times have to wait for laparotomy and surgical resection is the mainstay of treatment. Attempted reduction could be made if bowel wall is not compromised or malignancy is not suspected in small bowel intussusception while en bloc resections using appropriate oncological principles should be done in large bowel intussusception because of higher risk of malignancy.

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