CASE REPORT

Telescoping jejunum: a case of adult intussusception

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SUMMARY

Intussusception is the invagination of a proximal part of intestine into the adjacent distal part. Reported frequently in children, it forms a rare presentation in adults and can be difficult to diagnose pre operatively. Imaging modalities are increasingly being used for preoperative diagnosis with almost all cases in adults being secondary to an intestinal lesion. Intraoperative management of such lesions in adults was mostly reported to be done by en bloc resection without attempts at reduction, especially where a preoperative diagnosis of a benign lesion was not in hand. We present a case of a female patient with a background of active metastatic lung cancer presenting with symptoms and signs of high intestinal obstruction, found to have a jejunal intussusception on CT scan and managed conservatively initially, followed by definitive surgical management. Literature review of similar presentations, diagnostic and treatment modalities are discussed thereafter.

BACKGROUND

Intussusception forms a rare but important presentation in adults. Clinical presentation is usually vague but can be that of intestinal obstruction. Preoperative diagnosis can be accurately made using CT scans as the first-line imaging modality. Since most cases of adult intussusception are pathological, treatment is mostly surgical and involves resection of the effected segment without attempts at reduction if the lead point is considered malignant or if the bowel is ischaemic. No universal established guidelines exist for management; however, there is a generally agreed protocol for surgical treatment as seen in the literature. The aim of this case report was to highlight the importance of a preoperative diagnosis of small bowel intussusception (SBI) especially in adults and its subsequent surgical management.

CASE PRESENTATION

A 55-year-old woman presented to accident and emergency department of a district general hospital with complaints of colicky central abdominal pain and worsening nausea and vomiting for 3 days. Abdominal distension was also reported. She had a background of active lung cancer with likely metastasis in an adrenal gland and was receiving chemotherapy. The lung cancer had been identified as a non-small cell type with epidermal growth factor receptor (EGFR) mutation which was sensitive to EGFR tyrosine kinase inhibitor

1–4 on histology and DNA analysis and hence was deemed to have a good 3-year prognosis.

On examination, there was minimal tenderness in the epigastric region with mild upper abdominal fullness. Initial blood investigations showed a raised C-reactive protein level at 113 mg/L (normal value <5.0) and a normal white cell count. Abdominal X-ray was inconclusive for bowel obstruction. She was further investigated with a CT scan which showed duodenojejunal intussusception possibly secondary to a metastatic abdominal lymph node in keeping with her diagnosis of metastatic disease (figures 1 and 2).

In the light of her clinical picture, it was decided to manage her conservatively with nasogastric tube (NG) drainage, fluid resuscitation and total parenteral nutrition (TPN). She underwent a repeat CT on the third admission day to assess resolution of the pathology. The CT showed evidence of interval resolution but with subsequent recurrence at the same site. Conservative management was continued. On the sixth admission day, patient showed improvement in symptoms and was allowed to fluids by mouth. She underwent a gastrograffin follow through imaging which showed that obstruction was resolved and contrast was seen to flow into the distal bowel. It did not take long for the symptoms to recur after that, following which patient satisfaction and morale was deeply indented resulting in patient demand for a definitive management despite extensive counselling by the primary consultant on the challenges and risks of surgical management considering her morbidity.

OUTCOME AND FOLLOW-UP

On the 13th admission date, a decision was made after discussion with upper gastrointestinal surgeons to go for surgical management. She was taken to theatre on the 14th admission day and underwent a laparotomy. Intraoperatively, the stomach was found to be grossly distended and a very proximal jejunojejunal intussusception was identified. There was no evidence of a metastatic lymph node acting as the lead point (figures 3 and 4). The intussusception was subsequently reduced as a benign looking polyp was identified within the lumen of the invaginating jejunum as the lead point. The intussuscepting segment along with the polyp was resected and initial plan was for primary anastomosis with placement of nasojejunal (NJ) tube for drainage from the anastomosis site, due to unforeseen circumstances, a NJ tube was not available and decision was made to form a double barrel jejunostomy. Postoperatively, patient was strictly monitored and kept on TPN.

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Figure 1 Coronal section CT scan showing intussusception.

She was allowed oral fluids on the second postoperative day with NG tube in situ on free drainage. Specimen analysis revealed multiple other polyps in the resected segment of the jejunum which indicated the presence of metastatic disease in the bowel. All polypoid lesions showed similar morphology of ulcerated malignant epithelial tumours invading the submucosa. Immunohistochemically, the tumours were positive for AE1/AE3, CK7, carcinoembryonic antigen and thyroid transcription factor 1. Some areas of neuroendocrine differentiation were positive for synaptophysin and CD56. Further analysis revealed deletions in

Figure 2 Axial section of CT scan showing pathognomonic 'target' sign.

Figure 3 Intussuscepting segment of jejunum with the lead point.

the exon 19 of the EGFR gene which are known to convey good response to tyrosine kinase inhibitors. Final histology report confirmed the polypoid lesions to be metastatic lung combined adenocarcinoma and high-grade neuroendocrine carcinoma.

Postoperative recovery was steady but satisfactory. The new findings prompted a multidisciplinary meeting for further management. It was decided in the patients best interest that further management be only conducted along the lines of palliation. Her fluid and nutritional needs were optimised and stoma output was aimed at keeping to a minimum with plans for long-term TPN. This approach led to a good clinical recovery.

DISCUSSION

Intussusception forms one of the important causes of abdominal pain and occurs when a proximal portion of the bowel called intussusceptum, invaginates into an adjacent distal portion termed the intussusciens. It is frequently seen in paediatric patients presenting with a specific set of symptoms, but it can occur at any age.¹ In the adult patient, it forms a rare presentation accounting for 1%–5% of all cases of bowel obstruction.^{1–4} Mostly idiopathic in children, in adults it almost always (90%) occurs secondary to a pathology of the intestines; however, idiopathic cases have been reported in the literature.^{1–7} The pathology of the intestines causing this form of obstruction is most commonly a lesion within the lumen, acting as a lead point which pulls the proximal portion into the distal with normal peristalsis.⁵ Lesions of the intestinal tract acting as a lead point can be benign, inflammatory or metastatic.^{4–6} Most lead points in SBI are benign lesions including but not limited to adenomas, lipomas, leiomyomas, haemangiomas and fibromas.³ Malignant

Figure 4 Intussuscepting segment of jejunum.

lesions in SBI account for 15%–30% of cases; however, in cases of large bowel intussusception (LBI), malignant lead points are more commonly reported in the literature accounting for 65%–80% of cases.^{13 5 8 9} Literature also reports cases of SBI secondary to diffuse small bowel disease such as coeliac disease.¹⁰

Similar to our case, clinical presentation in such patients varies and abdominal pain is universally reported as the most common symptom.^{2 4-6} The classical clinical triad of paediatric intussusception, that is, red currant stools, abdominal pain and palpable mass, is rarely seen in adults.^{8 9} Literature review revealed that most cases of adult intussusception present with vague and chronic symptoms; however, intestinal obstruction is a frequent presentation and will depend on the size and location of the causing lesion.^{1 3 4 7 8} Intussusception in adults is classified into four categories based on the location, namely, enteroenteric, ileocolic, ileocaecal and colocolic, with the enteric variety being far more common in adults accounting for as much as 88% as reported in the literature.^{4 6-8} Our case was that of the enteroenteric variety.

Regardless of the location, when it invaginates, the intussusceptum takes its mesentery with it causing constriction of venous and lymphatic flow ultimately making it oedematous and not only obstructing the flow of intestinal contents causing intestinal obstruction but also leading to strangulation and eventual ischaemia.^{4 8}

In the acute setting, when it presents as intestinal obstruction, diagnosis is difficult based on the clinical examination alone. X-rays are of limited value in the preoperative setting and only give a hint towards the site of obstruction.^{2 4 5} Ultrasonography (USG) is too operator dependent and requires a

lot of experience at the sonographers end in order for it to be of value in the acute setting since obstruction can make identifying the pathognomonic 'target' or 'donut' sign difficult, and hence USG also has limited value in making preoperative diagnosis.^{2 4 9} Literature review supports CT scanning in the acute setting as the mainstay for diagnosis which can also identify the lead point with sensitivity range reported from 58% to 100% and is increasingly being used in the recent years as the first imaging modality.^{1 2 4-6} Other investigation including endoscopic evaluation and contrast X-rays, which maybe contraindicated in the acute setting, has been reported but with lower level of sensitivity when compared with CT scans and their use maybe dictated based on the clinical setting.^{4 5 8 9} In our case, the CT was not able to pick up the lead point but it did, however, correctly outline the location of the SBI.

Even though there are no clear established guidelines for treating SBI or LBI, there seems to be a consensus found in the literature for optimum management. Unlike for paediatric cases, where conservative management or simple reduction alone will be sufficient to treat the intussusception, adult cases will almost always require surgical management via a laparotomy.^{1-3 6-9} At this point, some reported studies warrant initial attempts at reduction provided a preoperative or intra operative diagnosis of a benign lead point is established and the bowel is not ischaemic.^{1 5 7 8} Based on the likelihood of benign and malignant aetiologies being the cause in SBI and LBI, another consensus seen in literature is that of en bloc resection in cases of colocolic intussusception without attempts at reduction intra operatively. This is reasoned by the fact that due to the high proportion of the lead point being malignant in nature in LBI, reduction can be detrimental as it can seed malignant cells intraluminally or systematically following perforation of the friable bowel.^{1 3 5-9} Since most cases of SBI can be secondary to a benign lesion, some studies in the literature warrant initial attempts at reduction to reduce the segment of the bowel that will need resection.^{3 5 7} Like in our case, since the lead point was identified as a high benign looking lesion, initial reduction followed by resection was done. Despite this, many studies advocate en bloc resection for all adult cases of intussusception due to the chances of the lead point being malignant and due to the fact that non-viable bowel secondary to oedema and/or ischaemia maybe discovered intra operatively which can lead to complications with anastomosis.^{3 6} In cases where resection is carried out, it is recommended to meet the standards of oncological resection.¹

Cases of SBI secondary to diffuse bowel pathology, most prominently reported in the literature to be coeliac disease, require early recognition and high index of suspicion as it presents with transient intussusception and requires a non-operative approach for management and so should always be considered as the cause in adult cases of SBI with the right clinical picture to avoid unnecessary surgery.¹⁰

Recently, minimally invasive surgery has been reported in the literature for the definitive diagnosis and management for such cases, but it depends largely on the clinical setting and the expertise at hand.⁵ Literature supports the idea for catering management to each clinical setting in terms of primary versus delayed anastomosis as was done in our case.

Our case presented with technical difficulties since the patient had a significant comorbidity with limited surgical options to begin with. But towards the end, definitive surgical management proved to be the right way to go for the patient and went in line with the published literature. The decision for surgery was further advocated by the good prognosis from the point of view of the primary malignancy. The case report not only highlights

the importance of obtaining a timely diagnosis but also signifies the value of early definitive management as delay in a definitive solution not only makes the surgical option technically demanding but also affects patient's morale and decreases confidence in the health services and its providers.

Learning points

- Cases of small bowel intussusception form a rare but important cause of adult bowel obstruction.
- Most cases will be secondary to a pathological lead point for which a preoperative diagnosis should be sought to dictate management.
- Management largely depends on patient factors, site of the involved segment and availability of a preoperative diagnosis and should be catered to each clinical setting.
- Malignant causes of intussusception in adults require en bloc resection with or without primary anastomosis.

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