CASE REPORT

Adult jejunojejunal intussusception in the face of jejunal adenocarcinoma: two infrequently encountered entities

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SUMMARY

Adult intussusception and small bowel adenocarcinoma are rarely encountered together. Intussusception should be considered in the differential diagnosis of adult patients presenting with abdominal pain, especially those with unremitting symptoms. Concomitant anaemia should lower the threshold for suspicion of underlying malignancy. Jejunal adenocarcinoma represents a rare, but possible aetiology.

BACKGROUND

The invagination of a portion of the gastrointestinal tract into an adjacent bowel loop is termed intussusception.¹ Despite being the most common cause of intestinal obstruction in children, it remains to be a relatively rare disease in adults representing 0.003–0.02% of hospital admissions and 5% of bowel obstructions.¹ A pathological lead point is usually demonstrated in 70–90% of adult cases that may be benign or malignant.¹ Malignant causes constitute about one-third of lead points in small bowel intussusceptions.² Primary malignancies of the small bowel are also rare and represent only 1–2% of gastrointestinal cancers.³ They are most commonly adenocarcinomas and occur in the jejunum in up to one-third of cases. In our search, we encountered only five reported cases of jejunojejunal intussusception caused by adenocarcinoma and, to the best of our knowledge, these have been the only cases reported in the English literature since 1944. This emphasises the rarity in reporting of these two conditions occurring together.

CASE PRESENTATION

A 60-year-old woman with a body mass index of 31.8 presented with intermittent, crampy, non-radiating, moderate-severe epigastric pain of several hours duration. There was nausea as well as multiple episodes of vomiting of greenish/yellowish vomitus. There was neither fever, chills, dysuria, constipation, bloody stools nor history of weight loss. Medical history included benign gastric and colonic polyps, and intermittent symptoms similar to the current complaints, over the past 2 years, attributed to gastro-oesophageal reflux disease and gastritis. Surgical history included hysterectomy. Family history was positive for metastatic colon cancer. Drug history was non-contributory.

On examination, the patient was in painful distress. Vital signs revealed a blood pressure of 178/63 mm Hg, heart rate of 101 bpm and a

temperature of 36.9°C. Abdominal examination revealed a distended abdomen with epigastric tenderness, but no signs of peritonitis. Abdominal auscultation revealed hypoactive bowel sounds. Laboratory tests revealed an elevated white blood cell count at 12 000 cells/U/L, low haemoglobin at 9.8 g/dL and normal carcinoembryonic antigen, serum glucose, transaminases, bilirubin, amylase and lipase. The patient continued to have intractable vomiting and abdominal pain, despite antiemetics and analgesics. Abdominal ultrasound was unremarkable. Abdominal and pelvic CT with contrast revealed proximal jejunal intussusception with enlarged mesenteric lymph nodes and high-grade small bowel obstruction (figures 1 and 2). The patient was admitted for initial conservative management of small bowel obstruction. On postadmission day 1, the patient had complete resolution of symptoms. Follow-up abdominal CT with contrast was unremarkable for intussusception, intestinal obstruction and masses. Upper gastrointestinal series with small bowel follow through revealed a 2.6 cm submucosal lesion along the lateral margin of the proximal jejunum with resolution of the jejunal loop intussusception and small bowel obstruction (figure 3). Owing to the patient's history of recurrent symptoms, microcytic anaemia and presence of a jejunal mass suggestive of being the lead point, operative intervention was warranted.

DIFFERENTIAL DIAGNOSIS

Adult intussusception (AI) presents with non-specific symptoms that overlap with many other gastrointestinal, genitourinary and gynaecological conditions making the diagnosis challenging.

TREATMENT

Laparoscopy revealed a strictured area of the jejunum with a mass lesion, approximately 10–12 cm distal to the ligament of Treitz (figure 4). Owing to technical difficulties, the procedure was converted to open laparotomy and approximately 15 cm of the jejunum was resected with the surrounding lymph nodes. Intraoperative frozen section biopsies of the distal mass and mesenteric lymph nodes were positive for adenocarcinoma. Primary end-to-end jejunojejunal anastomosis was undertaken and the resected specimen was sent for histopathological examination. Gross examination of the specimen revealed a 4.5×4.5 cm ulcerated, raised, tan mucosal lesion. Microscopic examination

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Figure 1 CT of the abdomen (axial view) showing Target sign of jejunojejunal intussusception (white arrow).

revealed moderately differentiated adenocarcinoma invading the muscularis propria of the jejunal segment, with clear margins. One of four excised lymph nodes was positive for metastasis.

OUTCOME AND FOLLOW-UP

The postoperative course was uneventful and the patient was discharged on postoperative day 11. She was placed on an outpatient based adjuvant chemotherapy course of five fluorouracil and oxaliplatin. At 2 months postoperatively, the patient was doing well and was to continue to follow-up.

DISCUSSION

AI is a relatively rare clinical entity. A lead point is usually identified in 70–90% of cases with reported rates of malignancy ranging between 25–30% in the small bowel,⁴ and 63–68% in the large bowel.^{5–6} Malignant lesions of the small bowel are

Figure 2 CT of the abdomen and pelvis with contrast (coronal view) showing abrupt discontinuation of contrast at the area of jejunojejunal intussusception (white arrow).

Figure 3 Picture from UGI series with small bowel follow-through showing a 2.6 cm filling defect representing a submucosal lesion along the lateral margin of the proximal jejunum (white arrow). UGI, upper gastrointestinal.

commonly metastatic rather than primary lesions.² Interestingly, a retrospective review published in 2011, representing the largest data set in the USA to date, revealed combined rates for primary and metastatic malignant causes of only 8% and 7.72%, for colonic and enteric intussusceptions (CI and EI), respectively.⁷ These rates were lower compared to those in previous reports, suggesting a more selective role for surgical resection as treatment for AI.⁷ Table 1 summarises the clinical and pathological findings of our case and the five previously reported cases of jejunojejunal intussusception secondary to jejunal adenocarcinoma.^{1–8–11}

AI presents with non-specific symptoms that overlap with many other gastrointestinal, genitourinary and gynaecological conditions making the diagnosis challenging. Symptoms tend to be chronic and constitute mainly abdominal pain, nausea and vomiting, and—less commonly—a palpable mass and bloody stools. Preoperative diagnosis ranges from 30% to 70%.^{2–12} Abdominal CT represents the most accurate diagnostic modality with the possibility of demonstrating lead points, vascular compromise and possible associated complications such as intestinal obstruction or local spread.¹³ Other modalities such as X-ray,

Figure 4 Image taken during laparoscopy showing an area of jejunal stricture (white arrow).

Table 1 Clinicopathological findings in reported cases of jejunojejunal intussusception caused by jejunal adenocarcinoma

Author/year	Presentation	Onset—course of symptoms	Diagnostic modalities/findings	Intraoperative findings	Microscopic examination	TNM staging
Elmoghrahi <i>et al</i> /2016	Abdominal pain, nausea, vomiting, anaemia	Chronic—intermittent for 2 years	CT—Intussusception UGI series and small bowel follow through—Jejunal mass	Jejunal stricture	Moderately differentiated adenocarcinoma	T2N1M0
Burgers <i>et al</i> /2014	Abdominal pain, nausea	Acute—3 days	CT—intussusception	Intraluminal tumour	Adenocarcinoma	T2N0 (M-not reported)
Okolo <i>et al</i> /2010	Abdominal pain	Acute—3 days	Abdominal ultrasound—Intussusception	Mural tumour mass	Moderately differentiated adenocarcinoma	T2N0 (M-not reported)
Andriessen <i>et al</i> /2008	Abdominal pain, nausea, vomiting, anaemia	Chronic—intermittent for 2 years	CT—thickened small bowel wall, narrowing, polypoid mass Jejunoscopy—semicircular growing tumour	n/a	Moderately differentiated adenocarcinoma	T3N1Mx
Zeebregts <i>et al</i> /2004	Abdominal pain, vomiting, absent defaecation	Subacute—intermittent for 5 days	CT—intussusception with evidence of a lead point	n/a	Moderately differentiated adenocarcinoma	T3 (N,M-not reported)
O'Donoghue <i>et al</i> /1944	Abdominal pain, nausea	Chronic—intermittent for 2 years	Intraoperative	Firm constricting infiltrating mass	Papillary adenocarcinoma	n/a

n/a, not applicable; UGI, upper gastrointestinal.

ultrasound and barium studies have been less reliable in achieving a diagnosis.¹³ In established cases of CI, colonoscopy may play a role either preoperatively or intraoperatively in identifying a causal lesion.⁵

Surgery represents the mainstay of treatment, however, with continuing controversy on the choice of reduction-resection versus en bloc resection. Many authors have advocated surgical en bloc resection without reduction due to the presumed higher incidence of malignant causal lesions especially in CI, and the risk of bowel perforation due to possible associated ischaemia and oedema.⁶ As outlined earlier, the recently published large dataset suggested that there may be justification for non-surgical management given the presence of considerably less malignant aetiologies than found by previous studies.⁷

Despite the fact that small bowel comprises 90% of the length of the alimentary tract, malignancies of the small bowel represent a rare clinical entity.¹⁴ They are predominantly of the adenocarcinoma subtype. In descending order of frequency, they may occur within the duodenum (48.4%), jejunum (32.5%) or ileum (19.2%).³ Patients may remain asymptomatic for months or even years. Most common clinical manifestations in patients with jejunal adenocarcinoma are abdominal pain and gastrointestinal bleeding resulting in anaemia.¹⁴ In a study by Goh *et al*,⁶ anaemia represented an independent preoperative predictor of malignancy in patients presenting with intussusception. Definitive preoperative diagnosis of small bowel adenocarcinoma is present in only 35–72% of patients. Although upper gastrointestinal (UGI) endoscopy and upper GI series have excellent diagnostic accuracy within the duodenum, the jejunal and ileal subtypes may be missed. Push-type jejunal endoscopy may enhance accuracy of detecting jejunal lesions. CT scans have an overall diagnostic accuracy of 45%, with higher sensitivity in more advanced stages of cancer. Surgical intervention is mandatory unless the tumour is unresectable due to extensive lymph node, liver or peritoneal metastasis. Small bowel adenocarcinoma is radio resistant; however, chemotherapy is used for stage C and D cancers with similar treatment regimens to those used for col carcinoma.³

Learning points

- ▶ Adult intussusception is a rare entity.
- ▶ Small bowel tumours are a rare but possible aetiology of adult intussusception.
- ▶ A high index of suspicion is necessary for diagnosis of intussusception in adults presenting with abdominal pain, especially those with unremitting symptoms.
- ▶ Concomitant anaemia in adult patients presenting with intussusception should lower the threshold for underlying malignant aetiology.

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