Rare presentation of idiopathic duodenoduodenal intussusception

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ABSTRACT

Enteroenteric intussusception is a condition in which the full-thickness bowel wall becomes telescoped into the lumen of distal bowel. Intussusception in adult occurs infrequently and varies from childhood intussusception, particularly in its presentation, aetiology and treatment. Duodenoduodenal intussusception is rare because the duodenum is fixed in the retroperitoneal position. It usually occurs secondary to tumour, lipoma, Brunner's gland hamartomatous polyp or adenoma. The diagnosis in adults is usually made at laparotomy, where presentation is with intestinal obstruction. In non-emergency presentation, it may be difficult to arrive at an accurate diagnosis as symptoms may be vague, self-limiting intermittent abdominal pain. Clinical examinations and investigations may not be conclusive and another working diagnosis such as irritable bowel syndrome would be made. We describe a case where a patient initially presented with symptoms mimicking pancreatitis but his symptoms persisted over the course of 2 weeks. When a laparotomy was performed, duodenoduodenal intussusception was discovered and confirmed with histopathology. In this case, a discernible leading point could not be identified.

KEYWORDS

Duodenal intussusception – Duodenoduodenal intussusception – Idiopathic duodenal intussusception

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Case report

A 19-year-old man with no previous medical illness had endured 2 days of intermittent generalised abdominal pain which radiated to the back. The pain was associated with absolute constipation, fever, nausea and vomiting. On examination, he was pyrexial at 38.2 degrees C, with diffuse abdominal tenderness and guarding at the right hypochondrium. Initial investigations revealed an elevated white cell count of 14,500 u/l and a serum amylase of 795 u/l with normal liver function tests. The patient was treated with intravenous antibiotics for a presumptive diagnosis of cholecystitis; abdominal ultrasonography was unremarkable. We changed our diagnosis to pancreatitis immediately upon noting the urine amylase level of 7320 u/l. As he had absolute constipation with intermittent vomiting, a nasogastric tube was inserted and it drained about a litre of bilious fluid for a few consecutive days.

During serial examination, the abdomen was soft, with mild tenderness at the epigastric region and no epigastric mass. A diagnostic oesophagogastroduodenoscopy was performed and a large bulge at the D2–D3 region was identified, suggestive of an external compression. We were unable to pass the scope beyond D2 owing to the narrowed lumen and bile reflux was observed. Parenteral nutrition

was initiated. An urgent contrasted computed tomography (CT) of the abdomen revealed a grossly distended stomach extending until the second part of duodenum with a beak-like narrowing at the third part of duodenum with collapsed distal bowel loops (Fig 1). It was eminent that jejunal loops had collapsed and malrotated to the right side, with whirl pattern of mesenteric vessels indicating malrotation. Our radiologist's impression was small bowel volvulus (D3). He was subsequently sent to theatre for a laparotomy, where an 8-cm segment of dusky and oedematous duodenum (from the ampulla of Vater until the proximal fourth part of duodenum) was identified (Fig 2). Multiple enlarged lymph nodes surrounding the duodenum and superior mesenteric vessels were detected but no malrotation of jejunal loops was identified on the table. A pyloric preserving pancreaticoduodenectomy (Fig 3) with jejuno-jejunostomy was performed. The postoperative period was uneventful and the patient made a steady recovery and was discharged a few days later. The histopathological report confirmed the picture of duodenoduodenal intussusception. No polyp, perforation or tumour was noted. Moreover, serial section of pancreatic tissues was unremarkable. We were unable to perform a follow-up on his condition as his employer had sent him back to his native country.

Figure 1 Axial cut of computed tomography of the abdomen (portal venous phase) showing a grossly distended stomach and a beak-like narrowing at D3.

Figure 2 Intraoperative view of the head of the pancreas, with ischaemic appearance; D2 and D3 are visualised in this image.

Discussion

Bowel intussusception is the telescoping of a proximal segment of the gastrointestinal tract into an adjacent distal segment. While intussusception is the number one cause of bowel obstruction in children, adult intussusception is rare, accounting for only 5% of all intussusceptions.⁵ While 95% of intussusceptions in children are idiopathic, 80–90% of intussusceptions in adults have an identifiable cause.⁵ The most common site for adult intussusception is the small bowel, while the least common types are coloanal and gastroduodenal intussusception.¹ Duodenoduodenal intussusception is very rare in adults, as the duodenum is fixed in the retroperitoneum. It is almost always associated with a lead point such as lipoma, hamartomatous polyp, Brunner's gland hamartoma or hyperplasia.⁵

Figure 3 Frontal view of the gross pancreaticoduodenectomy specimen; D1 up to the proximal jejunum can be seen, together with the head of the pancreas. The duodenum is ischaemic in appearance.

The clinical symptoms of duodenoduodenal intussusception are non-specific and may easily be misinterpreted.¹ Epigastric pain, abdominal mass, gastrointestinal bleeding and enteral obstruction may be present.⁵ The biliary and pancreatic systems are generally not involved.⁵ In our reported case, although the serum and urine amylase levels were raised, both the CT and the histopathology sections of the pancreas were unremarkable, likely indicating slight pancreatic inflammation, owing to the pressure effects of duodenoduodenal intussusception. Because of its rarity and non-specific elusive presentations, the clinical diagnosis is often delayed and challenging.⁵ Various imaging modalities have been used to help in establishing the diagnosis. However, the diagnosis is frequently confirmed only during surgical intervention.⁵

CT seems to be the most reliable investigation in making a preoperative diagnosis, with a sensitivity of 71.4–87.5% and a specificity of 100% in the prospect of diagnosis of intussusception.⁵ Abdominal CT findings that might indicate the presence of small bowel intussusception include the 'target' or 'doughnut' signs, as well as a sausage-shaped mass or

pitchfork image.¹ Additionally, CT helps in identification of the lead point lesions when present, and other associated malignancies. In our reported case, CT did not show the pathognomonic findings of intussusception. It did, however, help to localise the pathological part of the small bowel. Abdominal ultrasonography may also be used to diagnose small bowel intussusception.¹ The classical ultrasonography findings include ‘target’ or ‘doughnut’ signs on transverse view, the ‘pseudo-kidney’ sign on oblique view and the ‘trident’ sign on longitudinal view.⁵ However, it is operator dependent and quality of the views may be affected by gas in distended loops of bowel and the patient’s body habitus (obesity).⁵ Barium studies have also been used to diagnose bowel intussusceptions in adults with long-term nonspecific abdominal pain. However, these studies are contraindicated in patients suspected to have bowel obstruction.⁵ In addition, upper gastrointestinal endoscopy may be used in establishing the diagnosis, and to exclude obvious obstructive growths.⁵

Small bowel intussusception in adults commonly requires surgical resection of the affected bowel.¹ This is due to the significant rate of other pathologies associated with bowel intussusception in adults. However, for idiopathic intussusceptions, some authors advise for a simple reduction, after excluding bowel ischemia and perforation.⁵ In our reported case, the duodenal segment was ischaemic so a resection was performed. The accessibility of intraoperative histopathological tissue diagnosis facilities may help in determining the extent of surgical resection. However, up to 60% of adult intussusception cases are emergency operations, where such a facility is not always accessible.⁵

In conclusion, duodenoduodenal intussusception is a challenging diagnosis because of its rarity and nonspecific

presentations. It should be a differential diagnosis in patients presenting with nonspecific abdominal pain, gastric outlet obstruction, pancreatitis, upper gastrointestinal bleed and obstructive jaundice. A high index of suspicion coupled with the appropriate imaging might help in establishing a diagnosis preoperatively.⁵ An abdominal CT is likely the investigation of choice, owing to its high sensitivity and specificity.¹ Treatment of intussusception in adults is typically surgical and it should not be delayed, as it can become complicated with bowel obstruction, bowel ischaemia or bleeding.⁴ Where there is no discernible lead point lesion, and there are no signs of bowel ischemia or perforation, a simple reduction may suffice.⁵

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