Case report

Adult intussusception associated with mesenteric Meckel's diverticulum and antimesenteric ileal polyp

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SUMMARY

Meckel's diverticulum is the most common congenital abnormality affecting the gastrointestinal tract, affecting 4% of the general population. It is classically located on the antimesenteric border of the ileum within 100 cm of the ileocaecal valve. Complications may include haemorrhage, bowel obstruction, diverticulitis, perforation and malignancy. This report explores the case of intussusception in an adult, in association with a mesenteric Meckel's diverticulum and adjacent benign polyp. A 40-year-old man presented with acute abdominal pain, affecting the central abdomen and both flanks. CT imaging revealed small bowel intussusception, with either a Meckel's diverticulum or polyp acting as a lead point. Intraoperatively, the intussusception had already resolved; however, an inflamed outpouching was identified on the mesenteric border of the ileum, with a firm mass palpable within the bowel lumen. A 70 mm small bowel resection and primary anastomosis were performed. Histopathological analysis confirmed an inflamed Meckel's diverticulum as well as an adjacent diverticulum comprising a benign polyp.

BACKGROUND

Meckel's diverticulum (MD) takes its name from Johan Meckel, the German anatomist who first described its embryological pathogenesis in 1809. As stated by Charles Mayo (co-founder of the Mayo Clinic) in 1933, it is 'frequently suspected, often looked for and seldom found.'

MD is the most common congenital abnormality affecting the gastrointestinal tract and is present in 4% of the general population. The incidence of complications decreases with age, with only 4% of adults with MD developing symptoms.¹ These complications may include haemorrhage, bowel obstruction (through volvulus, herniation and intussusception), diverticulitis, perforation and malignancy.² MD is found equally in either sex; however, males are four times more likely to develop complications than females.³

MD arises from the failure of the omphalomesenteric duct to regress between the fifth and eighth weeks of gestation. It comprises a blind-ended outpouching composed of all five layers of the bowel wall. In 50% of cases, heterotopic tissue is present,^{1 2} with gastric mucosa being the most common. It is classically located on the antimesenteric border of the ileum within 100 cm from the ileocaecal valve and retains its own blood supply.² A small number of case reports have previously

reported MD affecting the mesenteric border, although debate exists over whether these should be classified as intestinal duplication cysts (IDCs).

This report examines a case of adult intussusception in the context of a mesenteric MD and adjacent benign polyp. We will compare our findings with previous literature with the aim of better informing our understanding of what defines an MD and the implications of the mesenteric location.

CASE PRESENTATION

A 40-year-old male teacher presented to the emergency department with a 1-day history of nausea and colicky abdominal pain, most severe in the central abdomen and both flanks, radiating towards the groin. He had suffered from similar milder occurrences of this pain over the previous 6 months. He was systemically well and denied any vomiting, change in bowel habit, bleeding per rectum or lower urinary tract symptoms. He suffered from mild asthma but otherwise had no significant past medical or surgical history. On examination, his abdomen was soft, with mild epigastric tenderness and no abdominal distension or hernias. His vital signs were all within normal limits.

INVESTIGATIONS

The blood results revealed a raised white cell count ($13.1 \times 10^9/L$), while other markers (C reactive protein (CRP), lactate, haemoglobin, lipase and liver function tests) were all within normal limits. An X-ray of his abdomen did not reveal any evidence of bowel dilatation, and a chest X-ray showed no evidence of subphrenic air. A urine dipstick was negative for leucocytes, nitrites and blood.

A provisional diagnosis of ureteric calculi was formed by the emergency physicians, and the patient was discharged to return the following day for a non-contrast CT of the urinary tract. No urinary tract abnormalities were detected; however, there appeared to be large established mid-small bowel intussusception 2 feet from the ileocaecal valve, with upstream small bowel dilatation up to 4 cm in diameter. This upstream bowel contained inspissated contents, indicating that this pathology may be subacute or long-standing. In light of these findings, he was referred onwards to the general surgical team.

Following surgical review, a contrast-enhanced CT of the abdomen and pelvis was performed, confirming a 13 cm area of small bowel intussusception 2 feet from the ileocaecal valve (figure 1).

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Figure 1 CT of abdomen pelvis revealing evidence of intussusception, with a target sign.

There was no radiological evidence of bowel ischaemia or perforation. The lead point was postulated to be either an MD or benign polyp.

DIFFERENTIAL DIAGNOSIS

In contrast to children, where intussusception is usually idiopathic, in adults, an underlying cause is found in 90% of cases.⁴ The most common aetiologies of intussusception in an adult are a benign polyp, malignancy (particularly adenocarcinoma or lymphoma), MD and Crohn's disease.

TREATMENT

Following surgical review, the patient was made nil by mouth and treated with intravenous fluids and analgesia. A nasogastric Ryles tube was attempted to permit drainage of the small bowel; however, the patient was unable to tolerate this. His symptoms of nausea and pain improved following initial presentation and had almost fully resolved prior to surgery.

Forty-eight hours following presentation to the emergency department, the patient underwent a laparoscopy. Intraoperatively, an outpouching of bowel was identified on the mesenteric ileal border, 60 cm proximal to the ileocaecal valve (figure 2). No evidence of intussusception was found during surgery. Inflammatory changes affecting both the outpouching and the adjacent proximal and distal small bowel were noted, with no evidence of ischaemia or perforation. Based on visual evidence of bowel wall inflammation, the procedure was converted to laparotomy

Figure 2 Laparoscopic view of the Meckel's diverticulum.

Figure 3 Intraoperative view of the Meckel's diverticulum located on the mesenteric border of the bowel.

in order to permit small bowel resection. On manual examination, a firm mass could be palpated within the lumen of the small bowel, adjacent to the outpouching (figure 3). A 70 mm length of small bowel resection was resected, and a side-to-side ileoileal anastomosis constructed.

Histopathological examination concluded that the features of the outpouching (30 mm long and up to 12 mm in diameter) were consistent with an inflamed MD. There were no definite features of heterotopic tissue throughout the specimen. Microscopically, this tissue showed features of both acute and chronic inflammation, with no features of dysplasia or malignancy. The firm intraluminal mass was concluded to comprise a benign polyp (18×18×20 mm) located within a separate diverticulum adjacent to the MD.

OUTCOME AND FOLLOW-UP

The patient had an uneventful immediate postoperative course; however on day 3, postoperatively, he developed vomiting and central abdominal pain. These symptoms worsened over 3 days, and his white cell count and CRP rose to 16 and 368, respectively. A contrast-enhanced CT of the abdomen and pelvis was therefore performed to rule out an anastomotic leak. This found marked diffuse small bowel dilatation proximal to the anastomosis; however, no evidence of mechanical obstruction or perforation. He was treated conservatively for postoperative ileus, and his symptoms resolved within 5 days. The patient was seen 1 month later in the general surgery clinic, where he was found to be making a good recovery. Unfortunately, he required laparoscopic adhesiolysis 12 months postoperatively, following presentation with small bowel obstruction secondary to adhesions.

DISCUSSION

MD is a vestigial remnant of the omphalomesenteric duct, which provides nutrients to the midgut during embryonic development. MD is formed by the failure of this duct to regress and involute between the fifth and eighth weeks of gestation. It comprises all five layers of the bowel wall, possesses its own blood supply and is invariably within 100 cm proximal to the ileocaecal valve. While classically located on the antimesenteric border of the ileum, the mesenteric variant was first described by Chaffin *et al* in 1941⁵ and has since been the subject of several case reports.^{6–17} Some experts contend that these cases should actually be classified as IDCs.

A much rarer congenital anomaly, IDCs can occur anywhere in the gastrointestinal tract. Although the exact embryological aetiology is unknown, none of the current theories support involvement of the omphalomesenteric duct.¹⁸ Around half of IDCs occur on the mesenteric border of the ileum.⁸ Unlike MD, they rarely communicate with the bowel lumen and share their blood supply with the surrounding bowel. In this case, no evidence of a separate blood supply was found; however, a previous case series of 776 patients found as few as 10% of MD with an identifiable distinct blood supply.¹⁹ The presence or absence of heterotopic tissue is unhelpful in distinguishing between these two congenital abnormalities since 16%–33% of IDCs have heterotopic tissue compared with 55% of MD.⁸ Kurzbart *et al* provided compelling evidence for the classification of mesenteric MD as a separate entity to IDC. In their case report, a fourth lumen was found in the transected umbilical cord of an infant. Intraluminal contrast injection and X-ray imaging revealed a patent omphalomesenteric duct. When this infant underwent an umbilical hernia repair 3 months later, this duct had spontaneously regressed; however, an MD attached to mesentery was found and resected.¹⁰ This proved a mesenteric variant of the MD could result from the failure of omphalomesenteric duct regression.

Given the location of the abnormality, its communication with the bowel lumen and macroscopic appearance, it is this author's view that our case was more consistent with a mesenteric MD than IDCs.

The mesenteric variant of the MD has been hypothesised to result from a short vitelline artery.¹³ Sarioglu-Buke *et al* proposed that this may lead to a mesodiverticular band adjoining the diverticulum to the surrounding mesentery, which pulls the diverticulum away from the antimesenteric border during embryological development. Other authors have suggested that adhesions between the vitelline duct and ileal mesentery underlie this anomaly.²⁰

Among current literature, only one case of mesenteric MD has resulted in intussusception⁶. Intussusception makes up 4%–14% of all complications arising from MD.⁴ Its pathophysiology is not well understood, but some authors propose that the presence of gastric mucosa may result in abnormal peristaltic movement.²¹ However, as in Abbas *et al*'s case report, our patient's specimen showed no histopathological evidence of gastric mucosa, and therefore, the underlying process causing intussusception is unclear. A complicating factor in our case was the presence of an inflamed benign polyp adjacent to the MD, occupying its own diverticulum. While MD is thought to be responsible for 4% of all cases of adult intussusception,²² benign neoplasms make up 29%.²³ This raises the question of whether it was indeed the MD that acted as the lead point for intussusception or whether it was the polyp, thus making the MD a red herring.

This case highlights limitations in current operative management of suspected MD. Small bowel resection was required due to the presence of inflammatory changes noted intraoperatively and risk of bowel wall ischaemia. However, had this MD been found incidentally during laparoscopy—for example in an appendectomy—it is highly likely that a laparoscopic diverticulectomy would have been performed. This would have resulted in the persistence of the adjacent polyp and thereby increased the risk of future complications including intussusception and malignancy. An alternative approach would be to extend a port site incision to allow manual examination of the area of interest. In this case, a palpable firm intraluminal mass suggested the presence of this secondary pathology. In addition, this author proposes that when examining the length of the small bowel in order to exclude MD (eg, in a negative

appendectomy), examination of the mesenteric as well as the antimesenteric border is necessary.

While transmural ischaemia and intussusception in the context of MD are absolute indications for resection,^{22 24} controversy still exists over the management of incidental MD. Some authors support resection in all cases due to the risk of complications, while others favour management guided by risk factors such as age and sex.^{1 2}

This case also highlights the difficulty in preoperative diagnosis of MD. Technetium scanning is one of the most sensitive and specific imaging modalities in preoperative diagnosis of MD. This relies on increased uptake of the radionuclide by gastric mucosa compared with other tissues. In this case, the lack of gastric mucosa would have increased the risk of a false negative result.²⁵

In summary, this case exposes the complexities in distinguishing an MD from an intestinal duplication cyst. While both require resection when symptomatic, there is no consensus on how these congenital abnormalities should be managed if asymptomatic. It also highlights how a mesenteric MD can be missed, in not examining the mesenteric border of the small bowel and in relying on technetium imaging to detect heterotopic tissue.

Learning points

- Despite several features which distinguish Meckel's diverticulum from intestinal duplication cysts, some authors believe these to be a spectrum of the same pathology.
- In order to exclude a Meckel's diverticulum intraoperatively, the mesenteric border of the small bowel should be studied.
- Meckel's diverticula lacking in gastric tissue are more likely to return a false negative result from a technetium radionuclide scan.

Contributors AKM was the primary author, was responsible for drafting and editing the case report, did research into similar cases, and submitted the case report. FS was responsible for data collection for the case report and edited and drafted the case report. NT was responsible for the planning and conception of the case report, obtained photographic images used in the case report, edited and drafted case report, did research into similar cases, and contributed to discussion. AR was responsible for the planning and conception of the case report, obtained photographic images used in the case report and edited and drafted case report.

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