CASE REPORT

Rare case of adult jejunojejunal intussusception secondary to angiolipoma

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SUMMARY

A woman in her mid-50s presented with a 3-month history of upper abdominal pain. Initial examination using ultrasound was unremarkable and the patient was sent home. The patient returned 8 days later and CT imaging revealed intussusception as the cause of her symptoms. The involved bowel was surgically reduced and transected with the lead point found to contain a 3 cm mass. Histological examination revealed the mass to be an angiolipoma. To the best of our knowledge, this is the first reported case of adult jejunojejunal intussusception secondary to angiolipoma.

BACKGROUND

Intussusception is more often seen in children and uncommonly found in adults. The incidence of adult intussusception stands at 2–3 cases per 1 000 000 per year and is responsible for about 2% of all bowel obstructions.^{1 2} In children, it is usually idiopathic, but in adults it is usually due to a lesion. Angiolipoma is a benign lesion composed of adipose tissue and thin-walled blood vessels, usually located on the arms and trunk in subcutaneous tissues. We present the first reported case of adult jejunojejunal intussusception secondary to angiolipoma.

CASE PRESENTATION

A woman in her mid-50s with a medical history of diabetes mellitus, hypertension and hypothyroidism status post thyroidectomy, initially presented to the emergency department with abdominal pain of 3 months duration. The abdominal pain was intermittent, described as crampy, and located in the right and left upper abdominal quadrants. An ultrasound was performed for suspected cholecystitis, which was unremarkable, and the patient was sent home. Eight days later, the patient returned to the emergency department with worsening abdominal pain. The patient denied fever, weight loss, changes in bowel habits and urinary symptoms. Her surgical history included hysterectomy and thyroidectomy for thyroid cancer. The patient's last colonoscopy and oesophagogastroduodenoscopy were 4 years ago. Examination revealed a tender upper abdomen without rebound tenderness or guarding. The rest of the examination was unremarkable.

INVESTIGATIONS

Biochemical and haematological investigations were within normal limits. Initial ultrasound was unremarkable. CT scan revealed intussusception involving approximately 30 cm of mid-small bowel with a portion of central mesenteric root pulled into the

intussusception (figure 1). There was also the possibility of an occult mass at the lead point. Wall thickening was non-specific and may have been related to intussusception with underlying inflammatory/infectious enteritis or bowel ischaemia. No pneumatosis, free air or free fluid was seen. No adenopathy was identified and the proximal small bowel was not dilated.

DIFFERENTIAL DIAGNOSIS

Although intussusception is a common occurrence in children, it is rare in adults. When it does occur in adults, it is usually due to a mass or lesion. Moreover, angiolipomas are benign tumours often found in subcutaneous tissues, but rarely found in the gastrointestinal tract. With this information in mind, the final diagnosis in this situation was fairly surprising.

The patient initially presented with upper abdominal pain and there was a suspicion of a gall-bladder process. When the lab values and ultrasound were unremarkable, the patient was sent home, only to return 8 days later with the same symptoms. At this point, a bowel obstruction was the main suspect, especially since the patient had previously had a hysterectomy. Adhesions that developed following the surgery could have been the cause of the obstruction. CT scan finally revealed that this was not the case. Instead, the patient had intussusception secondary to a mass, later found to be an angiolipoma.

Although intussusception is rare in adults, it should be considered in the presence of abdominal pain and may be secondary to a mass.

TREATMENT

The patient went to the operating room and the operation was performed under general anaesthesia. The incision was placed over the skin and subcutaneous tissue. The patient had extensive adhesions that were reduced and released from the anterior abdominal wall. A dilated mass with venous congestion compatible with intussusception was found in the mid-jejunum (figure 2). The involved bowel was reduced by milking the proximal portion while retracting the distal portion without complication. The lead point of the involved small bowel contained an umbilicated lesion about 2 cm in diameter with a thickened 5 cm mass. Hyperactive peristaltic movement of the bowel seemed to be inducing the intussusception. Mesenteric inflammation was present with inflammatory lymph node enlargement. The bowel was transected proximal to the lead point and

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Figure 1 (Left) Non-contrast CT coronal view: The full extent of the jejunojejunal intussusception is evident in this image from lead point (*) to distal involvement. (Right) Non-contrast CT axial view: Thickened bowel wall with visible lead point (*) involving the jejunum, suggestive of intussusception.

approximately 60 cm distal. The reason for the extensive bowel resection was due to venous congestion and questionable viability of the involved intestine. Wider resection would pose less risk for future complications.

OUTCOME AND FOLLOW-UP

Pathological examination revealed a 3 cm polypoid mass. When bisected, the mass expressed a bright yellow colour without haemorrhage or necrosis. Microscopic examination was performed and the final diagnosis was angiolipoma (figure 3).

The postoperative course was uneventful and the patient was discharged on the ninth postoperative day due to a slow recovery.

DISCUSSION

Intussusception is the telescoping of a portion of bowel into an adjacent segment that can lead to obstruction and ischaemia. It is most commonly seen in children where it is idiopathic and easily reduced. In contrast, the occurrence in adults is rare and is usually secondary to a causal lesion in 90% of cases.³

Figure 2 Intraoperative finding of intussusception (*) involving approximately 30 cm of the jejunum.

Incidence of adult intussusception stands at 2–3 cases/1 000 000 per year and is responsible for about 2% of all bowel obstructions.^{1 2} In children, presentation usually involves the classic triad of abdominal pain, vomiting and blood in stool.⁴ Owing to the fact that adults rarely present with this clinical triad, diagnosis is more challenging. CT of the abdomen is the preferred imaging study with a sensitivity of 71.4–87.5% and a specificity of 100%.⁵ An appearance of a ‘sausage-shaped’ mass in the longitudinal axis or a ‘target’ mass in the transverse axis is virtually pathognomonic.² Treatment in children usually involves barium or air enema for reduction. This may be attempted in adults with idiopathic intussusception but in adults as a population, intussusception is usually secondary to a lead point. Therefore, enemas in this population are rarely successful and can be potentially dangerous. Treatment in adults usually involves surgical intervention that requires reduction, if possible, and resection if necessary. There is debate over whether to reduce or not to reduce prior to surgical resection but the general consensus is that if at all possible, reduction can aid in saving as much viable bowel as possible.⁵

Angiolipomas are benign tumours that are variants of lipomas that contain adipose tissue and thin-walled blood vessels. They commonly occur in young adults, and are usually located on the arms and trunk in subcutaneous tissues.² They are rarely in the gastrointestinal tract and rarely greater than 2 cm in diameter.^{2 6} Patients with gastrointestinal angiolipomas can be asymptomatic or present with obstruction and bleeding. Treatment involves surgical excision.²

Figure 3 Histopathology showing lipid vacuoles and vasculature consistent with angiolipoma.

Based on our literature review, we found only 28 cases that involved angiolipomas in the gastrointestinal tract.^{2 4 6–29} Of the articles reviewed, only four cases presented with intussusception secondary to angiolipoma.^{2 4 6 7} However, this is the first reported case involving jejunojejunal intussusception secondary to angiolipoma.

Learning points

- Consider intussusception in the presence of abdominal pain.
- In adults, intussusception may be secondary to a mass or lesion.
- Surgical intervention is necessary for resolution in the presence of an underlying lead point.

Competing interests None declared.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

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