CASE REPORT

Angiolipoma: rare cause of adult ileoileal intussusception

Ashwini K Esnakula,¹ Archana Sinha,² Marie Fidelia-Lambert,¹ Venkata S Tammana³

¹Department of Pathology, Howard University Hospital, Washington, DC, USA
²Department of Internal Medicine, Howard University Hospital, Washington, DC, USA
³Department of Gastroenterology, Howard University Hospital, Washington, DC, USA

Correspondence to
 Dr Ashwini K Esnakula,
 aesnakula@howard.edu

SUMMARY

Intussusception in adults is rare and more common in the paediatric population. Clinically, most adult patients have chronic non-specific symptoms due to partial obstruction. In contrast, most paediatric patients present with the classic triad of abdominal pain, vomiting and blood in stool. Adult intussusception is commonly associated with an organic aetiology, most likely a benign or malignant neoplasm as a lead point of intussusception. We describe a case of a 29-year-old woman with subacute presentation due to ileoileal intussusception secondary to a polypoid submucosal angiolipoma. Angiolipoma is a benign lesion composed of mature adipose tissue and thin-walled capillaries. The presence of thin-walled vessels differentiates it from a lipoma. Angiolipomas of the small intestine are extremely rare with very few reported cases. This case not only demonstrates an unusual benign lesion as a cause of intussusception, but also illustrates an atypical clinical presentation in adults with intussusception.

BACKGROUND

Intussusception represents abnormal telescoping of the bowel into the lumen of an adjacent segment, leading to obstruction. It is the most common cause of obstruction in infants and young children and is often idiopathic.¹ Intussusception in adults is rare and accounts for <5% of cases of intestinal obstruction; it is usually associated with an organic aetiology.² Clinically, the classic triad of abdominal pain, vomiting and blood in stool is seen in one-third of paediatric patients, but rarely in adults. In two-thirds of paediatric patients and most adult patients, intussusception presents with subacute to chronic non-specific symptoms secondary to partial obstruction. Owing to its high incidence in the paediatric population, intussusception is always a diagnostic consideration in this age group, but not in adult patients.³ We describe this interesting case of intussusception with a subacute presentation in a 29-year-old patient secondary to a rare benign lesion.

CASE PRESENTATION

A 29-year-old Caucasian woman presented to an outside institution urgent care facility with a 1-week history of episodic cramping abdominal pain lasting from 15 s to 1 min. At this time, abdominal and pelvic ultrasonography was negative; gastritis was suspected and the patient received supportive treatment. Two days later, the patient returned to emergency department at our institution with continuous pain of increased intensity.

The patient also had an episode of loose brown stool. The pain was localised to the periumbilical area with radiation to the right lower quadrant. Her last menstrual period was 2 weeks ago. On examination, diffuse abdominal tenderness was noted. Rebound tenderness was present in the right lower quadrant. Bimanual pelvic examination demonstrated no adnexal tenderness or masses. These findings were highly suspicious of small bowel obstruction.

Pelvic and transvaginal ultrasonography revealed normal right adnexa and uterus. The left adnexa was not well visualised. Dilated fluid-filled bowel loops and pelvic ascites were identified. Abdominal and pelvic CT scan revealed target-like appearance of a bowel segment on axial view (figure 1A) and the same segment was visualised as a sausage-shaped mass (figure 1C) on coronal view in the right lower quadrant. There was also an associated well-defined ovoid radiolucent lesion with the same attenuation as fat in the contiguous segment of bowel (figure 1B,D). These findings were classic for ileoileal intussusception. The small bowel loops proximal to the area of intussusception were dilated. Mild pelvic ascites was also noted.

The patient underwent an exploratory laparotomy. An intussuscepted segment of ileum was identified in the right lower quadrant. The bowel appeared to be viable, but a 4×2 cm intraluminal mass was palpated after reduction of the intussusception. The approximately 7 cm of ileum with mass was resected and an ileoileal anastomosis was performed.

Examination of the ileal segment revealed a 4.3×2.5 cm ovoid semipedunculated polyp in the lumen. The surface of the polyp showed areas of haemorrhage and fibrinous exudate (figure 2A). Serial sections revealed a tan yellow homogeneous mass arising from the submucosa (figure 2B). Microscopically, the ulcerated lobulated mass demonstrated extensive mucosal inflammation and superficial granulation tissue (figure 2C). The mass consisted of lobules of mature adipose tissue with scattered collections of thin-walled capillaries some of which contained fibrin thrombi (figure 2D). These findings were consistent with an angiolipoma.

OUTCOME AND FOLLOW-UP

The patient's postoperative course was uneventful and she was discharged 4 days after surgery.

DISCUSSION

This case represents a rare cause of ileal intussusception due to an angiolipoma. Angiolipomas are

To cite: Esnakula AK, Sinha A, Fidelia-Lambert M, et al. *BMJ Case Rep* Published online: [please include Day Month Year] doi:10.1136/bcr-2013-008921

Figure 1 Abdominal CT scan, showing target-like appearance of bowel segment on axial view (A) and sausage-shaped mass on coronal view (C). A well-defined ovoid radiolucent lesion contiguous with the intussuscepted bowel noted on both axial (B) and coronal views (D).

benign neoplasms composed of mature adipose tissue and small thin-walled capillaries containing occasional fibrin thrombi. The presence of numerous thin-walled capillaries differentiates an angiolipoma from a lipoma. Clinically, angiolipomas are multiple and commonly identified in a subcutaneous location on the upper extremities and trunk of young patients. Approximately half of these lesions are painful, another distinguishing feature from lipomas, which are painless.⁴ Angiolipomas of the gastrointestinal tract (GIT) are extremely rare, whereas lipomas are the most common benign mesenchymal neoplasms of the GIT. Eighteen cases of angiolipomas have been reported in the literature. Including our case only seven cases have been reported in the small intestine. Of these seven, only three cases have been

associated with intussusception.^{5 6} In the other cases, a major symptom was anaemia secondary to bleeding.^{6 7} Angiolipomas of the GIT have similar histopathological features to angiolipomas located in subcutaneous soft tissue.

This case also exemplifies the subacute clinical presentation of intussusception in adults. Her initial symptom of intermittent cramping pain is probably secondary to incomplete obstruction, with automatic correction of intussusception. The patient's clinical symptoms worsened when the intussusception was no longer automatically reducible, leading to complete obstruction. This clinical course is common in adults and often leads to a delay in diagnosis.³ Cramping abdominal pain is the most common symptom in patients with adult intussusception. Other

Figure 2 An ovoid ulcerated polypoid mass in the lumen of the small bowel (A). Serial cross-sections revealing predominantly yellow adipose tissue (B). Lobulated submucosal mass with mucosal ulceration (C). Mature adipose tissue with scattered collections of thin walled capillaries and occasional fibrin thrombi in the vessels (black arrows) (D, $\times 200$).

common symptoms include nausea, vomiting and abdominal distension, which are associated with partial obstruction. Less than 20% of patients have initial presentation of an acute abdomen.⁸

Ninety percent of adult cases have an organic aetiology as a lead point of intussusception.³ A neoplastic process represents the cause of intussusception in approximately two-thirds of these cases. The neoplasm could be either benign or malignant. Up to 60% of colonic intussusceptions are secondary to malignancy, but less than one-third of small bowel intussusceptions are due to malignancy.⁸ Most common non-malignant lesions associated with intussusception are lipomas, Peutz-Jeghers and adenomatous polyps. Idiopathic intussusception in adults accounts for less than 10% of cases and are more common in small intestinal cases.³ Non-neoplastic causes of intussusception include anatomic anomalies like Meckel's diverticulum, adhesions secondary to previous surgeries and inflammatory conditions such as Crohn's disease and celiac disease.^{2 3 8}

The diagnosis of intussusception in adults can be aided by imaging. The CT scan is the most sensitive method in diagnosing intussusception. Ultrasonography is a good screening tool and is more sensitive in children. The target or doughnut sign with alternating hyperechoic and hypoechoic areas, corresponding to telescoping fat and bowel wall in the lumen of bowel, is diagnostic on ultrasonography. In the CT scan, intussusception can be visualised as a target-like appearance or sausage-shaped mass based on the direction of the x-ray beam to the long axis of the bowel segment. These findings are secondary to alternating high and low attenuation of bowel wall and mesenteric fat in the intussuscepted bowel segment. We observed both of these characteristic signs in the abdominal CT scan of our patient. The organic aetiology, angiolipoma in our case, was also evident as a radiolucent ovoid lesion. Another radiological appearance of intussusception is reniform or bilobed mass, secondary to bowel wall and mesenteric oedema, observed in cases of complete obstruction.⁹

Owing to high risk of associated malignancy, conservative management such as radiological decompression is not favoured in adult patients. Surgical resection is always preferred in colonic intussusception because they are more commonly associated with malignancy. In small bowel intussusceptions, reduction is an option, only if the bowel is viable and no neoplastic process is identified.^{2 3 8}

Learning points

- ▶ Intussusception in adults is rare and often associated with an organic aetiology.
- ▶ A benign or malignant neoplasm is the most common aetiology of adult intussusception.
- ▶ Ultrasonography is very useful in diagnosing intussusception in paediatric population; and CT is preferred in the diagnosis of adult intussusceptions.
- ▶ Angiolipomas of the gastrointestinal tract (GIT) are uncommon and are a rare cause of small bowel intussusception.
- ▶ Surgical resection of GIT intussusceptions is preferred in most adult cases because of the strong association with neoplasms.

Contributors AKE contributed by preparing the draft of the manuscript. AS and VST gathered the clinical and radiological information and were involved in drafting and reviewing the manuscript. MF-L was involved in revising the pathological findings and in the discussion for portion of the case report.

Competing interests None.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

REFERENCES

- 1 Wehlmiller SN, Buonomo C, Bachur R. Risk stratification of children being evaluated for intussusception. *Pediatrics* 2011;127:e296–303.
- 2 Azar T, Berger DL. Adult intussusception. *Ann Surg* 1997;226:134–8.
- 3 Marinis A, Yiallourou A, Samanides L, et al. Intussusception of the bowel in adults: a review. *World J Gastroenterol* 2009;15:407–11.
- 4 Fletcher CDM. Diagnostic histopathology of tumors. 3rd edn. Philadelphia: Elsevier Health, 2007.
- 5 Kaneko T, Karasawa Y, Inada H, et al. An adult case of intussusception due to inverted Meckel's diverticulum accompanied by angiolipoma. *Nihon Shokakibyo Gakkai Zasshi* 1996;93:260–5.
- 6 Aminian A, Noaparast M, Mirsharifi R, et al. Ileal intussusception secondary to both lipoma and angiolipoma: a case report. *Cases J* 2009;2:7099.
- 7 Della Volpe N, Bianco L, Bonuso C, et al. Rare ileal localisation of angiolipoma presenting as chronic haemorrhage and severe anaemia: a case report. *J Med Case Rep* 2008;2:129.
- 8 Haas EM, Etter EL, Ellis S, et al. Adult intussusception. *Am J Surg* 2003;186:75–6.
- 9 Baleato-Gonzalez S, Vilanova JC, Garcia-Figueiras R, et al. Intussusception in adults: what radiologists should know. *Emerg Radiol* 2012;19:89–101.

Copyright 2013 BMJ Publishing Group. All rights reserved. For permission to reuse any of this content visit <http://group.bmj.com/group/rights-licensing/permissions>.

BMJ Case Report Fellows may re-use this article for personal use and teaching without any further permission.

Become a Fellow of BMJ Case Reports today and you can:

- ▶ Submit as many cases as you like
- ▶ Enjoy fast sympathetic peer review and rapid publication of accepted articles
- ▶ Access all the published articles
- ▶ Re-use any of the published material for personal use and teaching without further permission

For information on Institutional Fellowships contact consortiasales@bmjgroup.com

Visit casereports.bmj.com for more articles like this and to become a Fellow