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CASE REPORT

Inflammatory Fibroid Polyps Causing Intussusception in Adult Patients: Two Case Reports and Review of Literature Focusing on Radiological Features

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ABSTRACT

Inflammatory fibroid polyp is a rare benign lesion originating from the submucosa of the gastrointestinal tract. Inflammatory fibroid polyp in the small bowel commonly presents with intussusception and bowel obstruction. We report two cases of inflammatory fibroid polyps as rare causes of intussusception in adult patients, and review the literature on imaging features of inflammatory fibroid polyps. Due to its well-defined and smooth outline as well as homogeneous appearance and low attenuation on computed tomography, diagnostic difficulty may be encountered as it can be easily missed for a complicated intussusception complex. Lead points must be carefully looked for in adult patients presenting with intussusception.

Key Words: *Gastrointestinal tract; Intestinal polyps; Intussusception; Multidetector computed tomography*

中文摘要

成人炎性纖維性息肉致腸套疊：兩宗病例報告和放射影像學特徵的文獻回顧

李芷茵、袁銘強

炎性纖維性息肉源自胃腸道的粘膜下層，是一種罕見的良性病變。小腸中的炎性纖維性息肉通常以腸套疊及腸梗阻起病。本文報告兩名病因為罕見的炎性纖維性息肉的腸套疊成人患者，並回顧炎性纖維性息肉影像學特徵的文獻。由於炎性纖維性息肉分界清晰、輪廓光整、電腦斷層掃描圖像性質均勻、低衰減，診斷可能有困難；而且可能因解剖輔助的腸套疊體部而漏診。診療腸套疊的成人患者必須仔細尋找導引點。

INTRODUCTION

Inflammatory fibroid polyp (IFP) is a rare, benign lesion originating from the submucosa of the gastrointestinal tract. It is most often found in the stomach, followed by the small bowel, and, rarely, in the large bowel, duodenum, gallbladder, and oesophagus.[1,2] Clinical symptoms may vary according to the site and size of the lesion. The most common acute presentation of an

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IFP in the small bowel is intussusception with bowel obstruction.[1,2] We report two cases of IFPs as rare causes of intussusception in adult patients, and review the literature on imaging features of IFPs, with special focus on computed tomography (CT).

CASE REPORTS

Case 1

In May 2011, a 56-year-old Chinese woman, with a medical history of hypertension and hysterectomy, initially presented with diffuse abdominal pain for the past few weeks. The abdominal pain was associated with vomiting and diarrhoea; however, there were no systemic symptoms of fever, chills, or rigors.

Physical examination revealed a vague abdominal mass in the right lower quadrant of the abdomen with tenderness and guarding on palpation. Laparoscopy, therefore, was performed for suspected acute appendicitis with abdominal mass. A normal appendix was found on laparoscopy; however, inflamed ascending colon wrapped by omentum without obvious mural mass or serosal lesion was also noted. Diagnosis of diverticulitis of the ascending colon was made. Laparoscopic adhesiolysis and appendicectomy were performed. Histological examination of the appendix revealed incidental carcinoid tumour. The diverticulitis was treated with antibiotics. Subsequently, her abdominal pain subsided and she was discharged.

She was readmitted 1 day after discharge for colicky periumbilical abdominal pain, vomiting, and diarrhoea. Once again, she was afebrile and haemodynamically stable. Physical examination revealed tenderness over the abdomen. Abdominal radiograph did not reveal dilated bowel loops. White cell count was mildly raised to 13.4 x 10^9 /l (reference range, 3.9-10.7 x 10^9 /l). Contrast-enhanced CT showed multiple concentric rings in the proximal colon, with invagination of transverse colon into the ascending colon, caecum and terminal ileum, compatible with intussusception (Figure 1). A 4-cm round well-defined, homogeneous, non-enhancing, low-density intraluminal lesion of 16 Hounsfield Unit (HU) was seen near the splenic flexure as a lead point (Figure 2). There was no internal calcification or fat density.

Emergency laparotomy confirmed ileocolic intussusception caused by a submucosal mass in the terminal ileum about 15 cm from the ileocaecal valve, measuring 4 cm x 5 cm x 5 cm, which was invaginated into the distal transverse colon near the splenic flexure. The segment of ileum with intussusception and the small bowel tumour were resected. Histopathological examination revealed IFP.

Figure 1. Case 1: Intussusception with invagination of bowels shown on computed tomography (arrows).

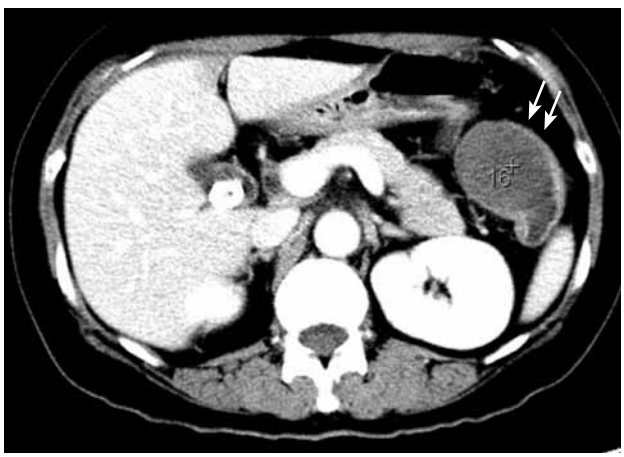

Figure 2. Case 1: Histopathologically confirmed inflammatory fibroid polyp which is round, smooth, homogeneous with low attenuation on contrast computed tomography (16 HU) [arrows].

Case 2

In August 2012, a 63-year-old Chinese woman with good past health presented with abdominal pain shifting from the epigastric region to the lower quadrant for a few days. She had no systemic symptoms of fever, chills, or rigors.

On admission, she was afebrile and haemodynamically stable. Physical examination revealed generalised abdominal tenderness. Laboratory tests including white

blood cell count, and liver and renal function tests were normal.

Oesophagogastroduodenoscopy was performed which showed chronic gastritis. Abdominal radiograph showed dilated small bowel loops. Contrast-enhanced CT showed multiple concentric rings in part of the ileum in the right lower quadrant compatible with intussusception. A 2.5-cm round well-defined, homogeneous, non-enhancing, low-density intraluminal lesion of 16 HU was seen as a lead point (Figures 3 and 4). There was no internal calcification or fat density.

Figure 3. Case 2: Round, well-defined, homogeneous, non-enhancing, low-density intraluminal lesion of 16 HU is seen as a lead point in lower abdomen (arrows).

Figure 4. Case 2: A coronal computed tomography image showing the same lesion within the small bowel (arrows).

Emergency laparotomy confirmed ileoileal intussusception caused by a submucosal mass at about 1 m from the ileocaecal valve measuring 3 cm x 2 cm x 2 cm. An oedematous small bowel segment of around 10 cm was noted adjacent to the point of intussusception. The segment of ileum with intussusception and the small bowel tumour were resected. Histopathological examination revealed IFP.

DISCUSSION

IFPs were first described by Vanek[3] in 1949 as a "gastric submucosal granuloma with eosinophillic infiltration". Various terms have been used to refer to this lesion, including gastric eosinophilic submucosal granuloma, Vanek's tumour, eosinophilic granuloma, haemangiopericytoma, myxoma, inflammatory pseudotumour, fibroma with eosinophilic infiltration, and polypoid myoendothelioma.[1,2] The term "inflammatory fibroid polyp" was introduced in 1953 by Helwig and Ranier,[4] and is now widely used and accepted.

The aetiology of IFPs is uncertain.[1,2] Various aetiologies have been proposed, including allergic, neural (hyperplasia), muscular, inflammatory (in the form of granulation tissue), and genetic.[5] Macroscopically, IFPs are submucosal, sessile or pedunculated polypoid lesions, usually solitary and are rarely multiple. Histologically, IFPs consist of masses of loose fibrous connective tissue arising in the submucosa, with the presence of an inflammatory infiltrate often dominated by eosinophils. Immunohistochemistry is the definitive complementary study. The spindle cell component of IFPs shows almost 100% positivity for CD34 and vimentin.[6]

Intussusception in adults is frequently caused by serious underlying disease, with 70% to 90% of cases having a demonstrable cause based on discharge diagnosis or surgical results.[7] Therefore, preoperative imaging is important for diagnosis and identifying the underlying cause of adult intussusception before proceeding to surgery. One benign cause of intussusception with or without small bowel obstruction in small bowels is IFP.

In the 1990s, the radiological appearance of IFPs was reported to be non-specific. CT was performed in only one case and did not furnish any unique diagnostic information.[8] As there is increasing use of cross-sectional imaging to determine the underlying cause and lead point of intussusception, more cases

of IFP causing intussusception with CT features were reported. However, no particular literature focused on or summarised the imaging features of IFP itself. Therefore, we performed a literature search of PubMed databases and Google scholar using the search terms "inflammatory fibroid polyp" and "intussusception" for articles in English published between January 1990 and August 2013, focusing on review of imaging, in particular CT features of IFPs causing intussusception.

A total of 62 articles involving 76 cases of intussusception due to IFPs were identified. Together with our two cases, a total of 78 cases were included. Presenting age ranged from 17 to 78 years (mean, 50.6 years). Most of the lesions measured approximately 3 to 4 cm in size. There were two rare, larger ones, measuring up to 11 cm.[9,10] Regarding the site of the lesions, 57 tumours were located in the ileum, 19 in the jejunum, and 2 in the colon.

In the past, IFPs with intussusceptions were commonly diagnosed by endoscopy or laparotomy without preoperative imaging, due to clinical emergency. Among the 78 cases, 27 (35%) were diagnosed without imaging. Fluoroscopic contrast studies (enteroclysis) were also used previously to identify the site and cause of intestinal obstruction. Barium enema examination, now rarely performed in acute situations, can demonstrate a 'cup-shaped' filling defect, which is a characteristic finding of intussusception. Luminal defect can be seen if there is underlying mass as a lead point. Among the 78 cases, fluoroscopic contrast studies had been performed in only six (8%) cases.

Ultrasonography is a non-invasive tool commonly used to demonstrate intussusception. Classical imaging features include the 'cresent-in-doughnut sign',[11] and the 'pseudokidney sign'.[12] Ultrasonography had been performed in 11 (14%) of the 78 cases. However, despite the non-invasiveness of ultrasonography, studies stated that ultrasound was mostly reliable in paediatric cases. CT is mandatory in adults to make the diagnosis, determine the underlying cause, find a lead point, and evaluate complications.[13] It is also the most sensitive technique for diagnosis and investigation of the location, type of lesion, any vascular compromise, and extent of the tumour.[13] Abdominal CT is helpful in distinguishing between an intussusception with a lead point and one without a lead point. An intussusception with a lead point will present with a greater cross-sectional diameter, longer length of intussusception, and may be associated with proximal bowel obstruction.[14] Identification of a

Table . CT characteristics of IFPs causing intussusception in small bowels in literature, with lead point identified from description or CT image(s) in the report.

Reference	Sex	Age (years)	IFP location	IFP size (cm)	Shape	Margin / outline	Density in contrast study (HU if available)	Homogeneity
Bays et al[17]	F	54	Ileum	3.5	-	Smooth	Soft-tissue density	-
Bays et al[17]	M	17	Jejunum	3	Round	Smooth	Low fluid to soft-tissue density	Homogeneous
Coulier et al[18]	M	58	Ileum	6	Pear-shaped	Smooth	Low fluid density	Homogeneous
Dicle et al[16]	M	22	Ileum	-	-	Well-defined	Low fluid to soft-tissue (17 HU)	-
El Hajj and Sharara[19]	F	52	Jejunum	3.5	Round	Well-defined	Soft-tissue	-
Gara et al[20]	F	76	Ileum	5.5	-	-	Low fluid density	-
Korkmaz et al[21]	M	30	Jejunum	3	Round	Well-defined	Soft-tissue density	-
Lasithiotakis et al[22]	F	52	Ileum	-	Round	Smooth	Low fluid density	Heterogeneous
Present study (case 1)	F	56	Ileum	5	Round	Well-defined, Smooth	Low fluid density (16 HU), non-enhancing	Homogeneous
Present study (case 2)	F	63	Ileum	3	Round	Well-defined, Smooth	Low fluid density (16 HU), non-enhancing	Homogeneous
Miyata et al[23]	F	64	Jejunum	4.5	-	-	Soft-tissue density	-
Nonose et al[24]	F	56	Ileum	4.5	Round	Smooth	Low fluid to soft-tissue attenuation	Homogeneous
Patel et al[25]	F	64	Jejunum	4.5	-	-	-	-
Rehman et al[10]	F	46	Jejunum	11	Round	Smooth	Low fluid to soft-tissue attenuation	Homogeneous
Ruffolo et al[26]	F	44	Ileum	4	Round	Smooth	Medium soft-tissue attenuation	Homogeneous
Savargaonkar et al[27]	F	42	Ileum	2.5	-	-	-	-
Yoon et al[15]	M	39	Ileum	2.5	Round	Smooth	High soft-tissue attenuation	Heterogeneous
Zager et al[28]	M	36	Ileum	3.5	-	-	-	-

Abbreviations: CT = computed tomography; IFP = inflammatory fibroid polyp.

lead mass that is separated and distinct from bowel loops at CT scan serves as a reliable radiological indicator of intussusception with a lead point and has the potential to reduce the number of unnecessary surgeries.[10] In one recent report, removal of the IFP was performed by double-balloon enteroscopy instead of surgery.[15]

Among the 78 cases, 39 (50%) had undergone preoperative CT. Interestingly, the lead point was mentioned in the CT findings or seen in CT images in less than half of the cases (n=18/39, 46%). The CT findings are summarised in the Table.[10,15-28] In both of our cases, the IFPs were seen as lead points within the intussusceptum. Both had smooth and round outlines, homogeneous and low attenuation, without significant contrast enhancement. There was no internal calcification. These findings are consistent with those from previous studies. Dicle et al[16] reported the low Hounsfield Unit of IFP (17 HU) and suggested that this was related to the high water content of these tumours.[16] We also demonstrated similar findings in our cases. We propose that due to the low attenuation similar to that of fluids and homogeneous appearance, IFPs are easily missed within the complicated intussusception complex, causing difficulty in detection.

In the 1990s, when CT was not used frequently, IFPs were often mistaken as other benign or malignant mural lesions.[8] We believe that lack of internal fat or calcification and lack of contrast enhancement may help in differentiating IFPs from other benign or malignant mural lesions that commonly cause intussusception such as leiomyomas, lipomas, or metastases. The smooth outline, homogeneity in attenuation as well as lack of adjacent inflammatory or infiltrative changes of IFPs may also be feasible in differentiating from malignant or inflammatory causes of intussusception.

Magnetic resonance imaging (MRI) was used in one of our cases to demonstrate intussusception due to IFP, by revealing bowel-in-bowel appearance and an intermediate, intense-signal polypoid mass as the lead point on breathing-independent HASTE (half-Fourier–acquired single-shot turbo spin echo) images.[29] MRI is also believed to be useful in the diagnosis and identification of the lead point. Further studies or reports are required to characterise the MRI features of IPFs in future.

CONCLUSION

IFP is a rare benign lesion in the gastrointestinal tract. When present in the small bowel, it commonly causes intussusception with bowel obstruction, and clinically, may be confused with neoplastic polyps. Due to its well-defined and smooth appearance as well as homogeneous and low attenuation on CT, diagnostic difficulty may be encountered as it can be easily missed within the complicated intussusception complex. Lead points must be carefully looked for in adult patients presenting with intussusception.

REFERENCES

1. Wysocki AP, Taylor G, Windsor JA. Inflammatory fibroid polyps of the duodenum: a review of the literature. Dig Surg. 2007;24:162-8. cross ref
2. Akbulut S. Intussusception due to inflammatory fibroid polyp: a case report and comprehensive literature review. World J Gastroenterol. 2012;18:5745-52. cross ref
3. Vanek J. Gastric submucosal granuloma with eosinophilic infiltration. Am J Pathol. 1949;25:397-411.
4. Helwig EB, Ranier A. Inflammatory fibroid polyps of the stomach. Surg Gynecol Obstet. 1953;96:335-67.
5. Morales-Fuentes GA, de Ariño-Suárez M, Zárate-Osorno A, Rodríguez-Jerkov J, Terrazas-Espitia F, Pérez-Manauta J. Vanek's polyp or inflammatory fibroid polyp. Case report and review of the literature. Cir Cir. 2011;79:242-5, 263-7.
6. Hasegawa T, Yang P, Kagawa N, Hirose T, Sano T. CD34 expression by inflammatory fibroid polyps of the stomach. Mod Pathol. 1997;10:451-6.
7. Kim YH, Blake MA, Harisinghani MG, Archer-Arroyo K, Hahn PF, Pitman MB, et al. Adult intestinal intussusception: CT appearances and identification of a causative lead point. Radiographics. 2006;26:733-44. cross ref
8. Harned RK, Buck JL, Shekitka KM. Inflammatory fibroid polyps of the gastrointestinal tract: radiologic evaluation. Radiology. 1992;182:863-6. cross ref
9. Akbulut S, Sevinc MM, Cakabay B, Bakir S, Senol A. Giant inflammatory fibroid polyp of ileum causing intussusception: a case report. Cases J. 2009;2:8616. cross ref
10. Rehman S, Gamie Z, Wilson TR, Coup A, Kaur G. Inflammatory fibroid polyp (Vanek's tumour), an unusual large polyp of the jejunum: a case report. Cases J. 2009;2:7152. cross ref
11. del-Pozo G, Albillos JC, Tejedor D, Calero R, Rasero M, de-la-Calle U, et al. Intussusception in children: current concepts in diagnosis and enema reduction. Radiographics. 1999;19:299-319. cross ref
12. Anderson DR. The pseudokidney sign. Radiology. 1999;211:395-7. cross ref
13. Baleato-González S, Vilanova JC, García-Figueiras R, Juez IB, Martínez de Alegría A. Intussusception in adults: what radiologists should know. Emerg Radiol. 2012;19:89-101. cross ref
14. Tresoldi S, Kim YH, Blake MA, Harisinghani MG, Hahn PF, Baker SP, et al. Adult intestinal intussusception: can abdominal MDCT distinguish an intussusception caused by a lead point? Abdom Imaging. 2008;33:582-8. cross ref
15. Yoon DW, Lee BJ, Lee JH, Park JJ, Kim JS, Bak YK, et al. A case of giant inflammatory ileal polyp removed by double-balloon enteroscopy. Clin Endosc. 2012;45:198-201. cross ref
16. Dicle O, Erbay G, Haciyanli M, Bora S. Inflammatory fibroid polyp presenting with intestinal invagination: sonographic and correlative imaging findings. J Clin Ultrasound. 1999;27:89-91. cross ref

17. Bays D, Anagnostopoulos GK, Katsaounos E, Filis P, Missas S. Inflammatory fibroid polyp of the small intestine causing intussusception: a report of two cases. Dig Dis Sci. 2004;49:1677-80. cross ref
18. Coulier B, Maldague P, Broze B, Gielen I. Ileal inflammatory fibroid polyp causing ileocolic intussusception. JBR-BTR. 2008;91:149-52.
19. El Hajj II, Sharara AI. Jejunojejunal intussusception caused by an inflammatory fibroid polyp. Case report and review of the literature. J Med Liban. 2007;55:108-11.
20. Gara N, Falzarano JS, Limm WM, Namiki TS, Tom LK. Ileal inflammatory fibroid polyp causing chronic ileocolic intussusception and mimicking cecal carcinoma. World J Gastrointest Oncol. 2009;1:89-92. cross ref
21. Korkmaz O, Yilmaz HG, Yilmaz F. Intussusception due to inflammatory fibroid polyp located at ileum: report of two cases. Turk J Med Sci. 2007;37:181-4.
22. Lasithiotakis K, Grisbolaki E, Filis D, Athanasakis I, Zoras O, Chalkiadakis G. Ileocolic intussusception precipitated by diagnostic colonoscopy: a case report. Surg Laparosc Endosc Percutan Tech. 2012;22:e161-3. cross ref
23. Miyata T, Yamamoto H, Kita H, Yano T, Sunada K, Sekine Y, et al. A case of inflammatory fibroid polyp causing small-bowel intussusception in which retrograde double-balloon enteroscopy was useful for the preoperative diagnosis. Endoscopy. 2004;36:344-7. cross ref
24. Nonose R, Valenciano JS, da Silva CM, de Souza CA, Martinez CA. Ileal Intussusception Caused by Vanek's Tumor: A Case Report. Case Rep Gastroenterol. 2011;5:110-6. cross ref
25. Patel A, James A, Thomas DM. Inflammatory Fibroid Polyp causing Jejuno-Jejunal Intussusception. Gastroenterology Today. 2008;18:87-91.
26. Ruffolo C, Scarpa M, Bassi D, Angriman I. Inflammatory fibroid polyp causing intestinal obstruction following restorative proctocolectomy for ulcerative colitis. Dig Surg. 2009;26:285-6. cross ref
27. Savargaonkar P, Morgenstern N, Bhuiya T. Inflammatory fibroid polyp of the ileum causing intussusception: report of two cases with emphasis on cytologic diagnosis. Diagn Cytopathol. 2003;28:217-21. cross ref
28. Zager JS, Shaw JP, Kaufman JP, DeNoto G. Three cases of small bowel intussusception in relation to a rare lesion: inflammatory fibrous polyps. Dig Surg. 2001;18:142-6. cross ref
29. Balci NC, Radjazi S, Polat H. Adult intussusception secondary to inflammatory fibroid polyp: demonstration by MRI. Eur Radiol. 2000;10:1708-10. cross ref