# Journal of

## Gastroenterology and Hepatology Research

Online Submissions: http://www.ghrnet.org/index./joghr/doi:10.6051/j.issn.2224-3992.2015.04.492

Journal of GHR 2015 January 21 4(1): 1451-1454 ISSN 2224-3992 (print) ISSN 2224-6509 (online)

CASE REPORT

# A Case of an Ileal Inflammatory Fibroid Polyp Causing Intestinal Intussusception

Yukako Nemoto, Mikiko Kaneko, Jun Fujisaki, Yoshihisa Saida, Kei Takahashi, Iruru Maetani

Yukako Nemoto, Iruru Maetani, Division of Gastroenterology and Hepatology, Department of Internal Medicine, Toho University Ohashi Medical Center, Tokyo, Japan

Mikiko Kaneko, Jun Fujisaki, Physiological laboratory, Toho University Ohashi Medical Center, Tokyo, Japan

Yoshihisa Saida, Department of Surgery, Toho University Ohashi Medical Center, Tokyo, Japan

Kei Takahashi, Department of Surgical Pathology, Toho University Ohashi Medical Center, Tokyo, Japan

Correspondence to: Yukako Nemoto, MD, PhD, Division of Gastroenterology and Hepatology, Department of Internal Medicine, Toho University Ohashi Medical Center, 2-7-16, Ohashi, Meguroku, Tokyo, 153-8515 Japan.

Email: yukako.nemoto@med.toho-u.ac.jp

Telephone: +81-3-3468-1269 Fax: +81-3-3468-1251 Revised: December 13, 2014

Accepted: December 16, 2014 Published online: January 21, 2015

## **ABSTRACT**

Inflammatory fibroid polyp (IFP) usually appears as a solitary benign lesion, rarely located in the ileum. The clinical presentation varies according to its location, frequently they are mistaken as gastrointestinal (GI) neoplasia. Intussusception and obstruction are the most frequent initial symptoms when the polyp is located in the small intestine. A thirty nine years old man presented with upper abdominal pain. Though he prescribed anti biotic medicine, his intermittent pain continued. He presented to hospital two days later, and abdominal ultrasound (US) detected intestinal intussusception. Abdominal computed tomography (CT) and oral ileal barium study also revealed ileal mass. So he had partial ilectomy on the seventh hospital day. Pathology showed it IFP in small intestine. Intussusception secondary to IFPs of the small intestine is difficult to diagnose without recognition of its clinical and pathological characteristics. Abdominal US is useful in confirming an anatomical abnormality, histological examination establishes the final diagnosis

of IFPs. Abdominal US detected an intussusception with a mass lesion at its lead point.

© 2015 ACT. All rights reserved.

**Key words:** Inflammatory fibroid polyp; Intussusception; Ileal; Abdominal ultrasound; Computed tomography

Nemoto Y, Kaneko M, Fujisaki J, Saida Y, Takahashi K, Maetani I. A Case of an Ileal Inflammatory Fibroid Polyp Causing Intestinal Intussusception. *Journal of Gastroenterology and Hepatology Research* 2015; 4(1): 1451-1454 Available from: URL: http://www.ghrnet.org/index.php/joghr/article/view/932

## INTRODUCTION

IFPs are rare benign tumors of the GI tract with the gastric antrum being the most common site, followed by the ileum. Although the molecular pathogenesis of these lesions has been well characterized, their morphologic features often vary. IFPs are one of the rare benign conditions leading to intestinal obstruction in adults. Patients with intussusception present with either acute or chronic intermittent symptoms. The majority of adult intussusception occurs due to malignant processes. We report a case of IFP confined to the ileum which presented with acute symptoms and a repeated intussusception background history.

## **CASE REPORT**

A 39-year-old man was presented with acute abdominal pain and a two month history of intermittent upper abdominal pain. He had upper endoscopy at another clinic then, and was diagnosed reflux esophagitis. There was no history of previous abdominal surgery, smoking or alcohol consumption. He had taken a medicine of hypertension.

Laboratory analysis revealed 15,000 leukocytes with a prevalence of neutrophils, and CRP 3.1 mg/dL. Other parameters were within normal limits. He prescribed anti biotic medicine

#### Nemoto Y et al. Ileal inflammatory fibroid polyp

and went home. Two days later, he admitted to the hospital with continuous intermittent abdominal pain. On examination, he had a heart rate of 72, blood pressure 148/94 mmHg, and a temperature of 36.4°C. Generalized abdominal pain was found on abdominal examination without signs of peritoneal irritation. Bowel sounds slightly attenuated. Laboratory analysis revealed 6,600 leukocytes, hemoglobin 12.9 g/dL, and CRP 1.4 mg/dL. Abdominal radiology demonstrated a small bowel obstruction with a few air fluid levels in left upper quadrant (Figure 1).

Abdominal US displayed gross thickening of presumed small intestine (ileum/ileum) 57×53×41 mm with target sign (Figure 2).

On examination, a palpable mass in the left upper quadrant was noted. Abdominal CT was performed, which demonstrated a target-shaped soft tissue mass pathognomonic intussusception (Figure 3).

Oral ileal barium study revealed a leading part shadow, a diameter of 40 mm that the surface was smooth in the ileum (Figure 4).

Though intussusception was resolved with hands, his symptoms worsened.

An open surgery was performed with the diagnosis of sub-acute intestinal obstruction on the seventh hospital day. An intussusception with a mass lesion at its lead point approximately 250 cm proximal to the ileocaecal valve was found. Intussusception was spontaneously reduced during exploration. Limited edema at the lead point of the bowel was the only sign of the intussusception. A partial resection was performed to the affected bowel segment. Macroscopic examination revealed a polypoid lesion, the inside is uniform with white (Figure 5).

Histopathologic examination showed that the polyp was confined to the submucosa with no extension into the muscularis propria. Associated with the lesion there was localized mild predominantly chronic inflammatory cell infiltrate. Spindle shape or stellar lightformed mesenchyma system cells appeared mainly in the submucosal layer, and inflammatory cell permeation to mix lymphocytes and plasma cells, acidophiles were outstanding. Immunohistochemistry performed on this lesion was negative for S100, desmin and  $\alpha$ -SMA, effectively excluding a neurogenic tumor, gastrointestinal stromal tumor and a submucosal leiomyoma. Spindled cells within the submucosa expressed CD34 (weak to intermediate expression). The cells with positive for C-kit suspected leucocytes that was scattered in the nodes (Figure 6).

These features favor an ileal IFP, which was responsible for the intussusception and subsequent small bowel obstruction.

The anemia recovered during postoperative follow-up. The patient had an excellent post-surgery recovery and was discharged home on the twelfth hospital day. He has remained well and symptom-free until 5 years after discharge.



**Figure 1** Abdominal radiology showed a small bowel obstruction with a few air fluid levels in left upper quadrant.

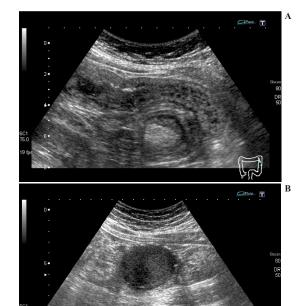


Figure 2 Abdominal US revealed the target sign in the transverse view (A) and a lead point with oval shaped, surface smooth, uniform parenchyma mass (B).

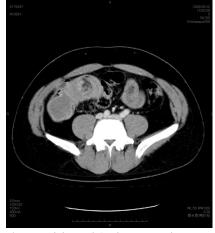


Figure 3 Abdominal CT demonstrated a target-shaped soft tissue mass pathognomonic intussusception.



**Figure 4** Oral ileal barium study revealed a leading part shadow, a diameter of 40 mm that the surface was smooth in the ileum.



Figure 5 Macroscopic examination revealed a polypoid lesion; the inside is uniform with white.

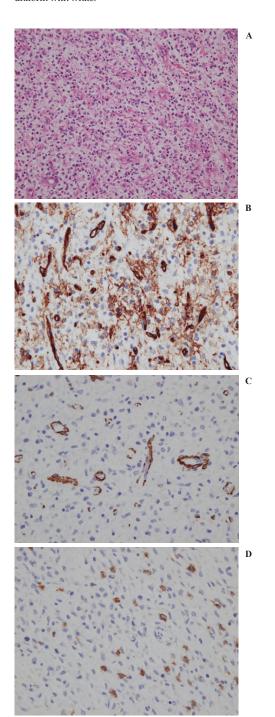


Figure 6 Immunohistochemistry of HE (A), CD34 (B),  $\alpha$ -SMA (C), and C-kit (D).

# **DISCUSSION**

IFPs are rare, idiopathic peudotumorous lesions of the gastrointestinal tract, which were first described by Vanek in 1949 as an eosinophilic submucosal granuloma<sup>[1]</sup>. The term IFP first proposed by Helwig and Ranier in 1953 for gastric polyps has gained acceptance for similar lesions throughout the gastrointestinal tract<sup>[2]</sup>.

IFPs can develop in many different locations in the gastrointestinal tract. The most common site is the gastric antrum (66%-75%), followed by the small bowel (18%-20%), colorectal region (4%-7%), gallbladder (1%), esophagus (1%), duodenum (1%), and appendix (<1%)<sup>[3]</sup>. However, the ileal segment is the most common site where these polyps cause intussusception<sup>[4]</sup>.

IFPs are usually asymptomatic and typically identified during endoscopic procedures and laparotomy. When they are symptomatic, the clinical symptoms depend on the location and size of the tumor<sup>[4]</sup>. Abdominal pain is the main symptom in patients with lesions in the stomach. Intussusception and obstruction are the most frequent initial symptoms when the polyp is located in the small intestine. Adult intussusception is a very rare condition, according for 1% of all adult bowel obstruction and occurs in only 5% to 16% of all intussuscepted cases<sup>[5]</sup>. About 70% to 90% of intussusception cases are due to benign or malignant neoplasms as a lead point and IFPs, lipomas and adenomas are the benign causes of intussusception<sup>[6]</sup>. The preoperative diagnosis of intussusception is controversial. An accurate diagnosis is based on a good medical history, thorough physical examination, and specific imaging modalities, such as X-ray, US, CT, magnetic resonance imaging (MRI), enteroclysis, endoscopic procedures, angiography, and capsule endoscopy. US has a sensitivity of 98%-100% and specificity of 88%-89% for diagnosing intussusception. On the other hand, CT has a diagnostic accuracy of 58%-100% for confirming intussusception. In our case, both US and CT detected intussusception, but only US showed it with a mass lesion at its lead point. In the recent reports, colonoscopy, US, and CT are the methods used to detect IFP lesions[7].

However, most cases are only diagnosed during surgery. At the time of diagnosis, most IFPs usually measure between 2 and 5 cm in diameter. IFPs usually present in the sixth or seventh decade<sup>[4]</sup>.

Histologically, IFPs were centered within the submucosa in all resection specimens, but mucosal extension was found in 89%<sup>[8]</sup>. There was a rare case report of IFP that the infiltrating tumor cells disrupted the muscularis mucosa above the tumor cells and the muscularis propria below the tumor cells, and extended into the subserosa<sup>[9]</sup>. The tumors varied in both cellularity and degree of vascularity. However, the characteristic feature of perivascular onion skinning was present in only 54% of the cases. Although the majority of IFPs expressed CD34, 14% were negative. No associated dysplasia or malignancy was seen.

# CONCLUSION

Intussusception secondary to IFPs of the small intestine are difficult to diagnose without recognition of its clinical and pathological characteristics. Abdominal US is quite useful in confirming an anatomical abnormality, histological examination establishes the final diagnosis of IFPs.

## CONFLICT OF INTERESTS

There are no conflicts of interest with regard to the present study.

# **REFERENCES**

- 1 Vanek J. Gastric submucosal granuloma with eosinophilic infiltration. Am J Pathol 1949; 25: 397-411
- 2 Helwig EB, Raner B. Inflammatory fibroid polyps of stomach. Am J Pathol 1952; 28: 535-536
- 3 Akbulut A: Intussusception due to inflammatory fibroid polyp: A case report and comprehensive literature review. World J Gastroenterol 2012; 18: 5745-5752
- 4 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: 110-116
- 5 Eisen LK, Cunningham JD, Aufses AH Jr: Intussusception in adults: institutional review. J Am Coll Surg 1999; 188: 390-395
- 6 Karamercan A, Kurukahvecioglu O, Yilmaz TU, Aygencel G, Aytac B, Sare M: Adult ileal intussusception: an unusual emergency condition. Adv Ther 2006; 23: 163-168

- 7 Sulu B, Günerhan Y, Kösemehmetoğlu K: A rare ileal tumor causing anemia and intussusception: inflammatory fibroid polyp. *Turk J Gastroenterol* 2014; 25: 116-117
- 8 Liu TC, Lin MT, Montgomery EA, Singhi AD. Inflammatory fibroid polyps of the gastrointestinal tract: spectrum of clinical, morphologic, and immunohistochemistry features. *Am J Surg Pathol* 2013; 37: 586-592
- 9 Bae JS, Song JS, Hong SM, Moon WS. An unusual presentation of an inflammatory fibroid polyp of the ileum: A case report. *Oncol Lett* 2015; 9: 327-9. Epub 2014 Nov 5

Peer reviewers: Francis Seow-Choen, Professor, Seow-Choen Colorectal Centre, 290 Orchard Road. Paragon #06-06, 238859 Singapore; Marco Bustamante, MD, PhD, Endoscopy Unit, University Hospital La Fe, Bulevar Sur, sn, Valencia 46017, Spain; Michael Kew Lim, Department of Colorectal Surgery, Christchurch Hospital, Riccarton Avenue, Christchurch 8011, New Zealand.