

Ileo-ileal Intussusception in an Adult Caused by Vanek's Tumour: A Rare Case Report

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ABSTRACT

Adult intussusception is relatively rare in patients with bowel obstructions. Adult invagination is mostly caused by benign tumours. Intussusceptions caused by inflammatory fibroid polyps (IFPs) are uncommon. IFPs are rare, localized, non-neoplastic lesions originating in the submucosa of the gastrointestinal tract. We are reporting a rare case of intestinal obstruction caused by an ileo-ileal intussusception, caused by an IFP, a rare but peculiar and always benign tumour.

Keywords: Intussusception, Vanek's tumour, Inflammatory fibroid polyp, Polyps

CASE REPORT

A 45-year-old lady presented with colicky abdominal pain, distension, vomiting and constipation, which she had since 3 days. There was no history of similar complaints in the past. She had undergone a tubectomy 17 years back. On examination, she was found to be comfortable but she was dehydrated. Her pulse rate was 94 beats per minute, with a blood pressure of 124/70mmHg. Abdominal examination revealed abdominal distension and tubectomy scar. Abdomen was soft, nontender, there was no palpable mass and there was hyperperistalsis. Digital rectal examination did not show the presence of faeces, or blood. Laboratory investigation showed neutrophilic leucocytosis, blood urea-50mg/dl, serum creatinine-0.8mg/dl. Serum electrolytes were within normal limits. A provisional diagnosis of an intestinal obstruction, probably caused by adhesion, was made. Patient was started on anti-biotics, intravenous fluids, and nasogastric decompression. Erect X-ray of abdomen revealed multiple air-fluid filled dilated small bowel loops. Abdominal ultrasonography (USG) showed dilated bowel loops. A contrast enhanced computed tomography (CECT) scan [Table/Fig-1] demonstrated an ileo-ileal intussusception with dilated proximal small bowel loops.

Laparotomy revealed an ileo-ileal intussusception [Table/Fig-2] with a dilated proximal small intestine. It was situated 50 cm proximal to the ileocecal valve. Intussusception was reduced and polyp was noted as lead point of intussusceptum [Table/Fig-3]. There was no evidence of any other polyps in the rest of the small bowel. We performed a segmental small bowel resection with extracorporeal end-to-end anastomosis. Post-operative specimen [Table/Fig-4] showed a firm, circumscribed, endoluminal 2x2.5cm polyp. Post-operative course was uneventful.

On microscopic examination [Table/Fig-5], the surface of the polypoid lesion was found to be covered by ulcerated mucosa, proliferating blood vessels, oedema and an eosinophilic infiltrate in a fibrous stroma. Foci of necrosis were noted. Features were those of an inflammatory fibroid polyp (also called as Vanek's tumour).

DISCUSSION

Adult intussusception occurs in only 1% of patients suffering from small bowel obstruction and 80% of such conditions are caused by benign tumours. [1,2]. This disease was first described to occur in the stomach by Vanek in 1949 [3]. Recently, a very interesting review of the literature on IFPs was published [4], which included 1000 cases of IFP, in which the characteristic of this disorder, the diagnostic and therapeutic strategies and microscopy have been described carefully. Only 5% of all intussusceptions occur in adults [5]. In 90% of adult cases, predisposing lesions can be found, but in the paediatric population, organic lesions are found in only 10% of the cases. In 63% of cases of small bowel intussusceptions, benign underlying lesions can be found, whereas in 58% of cases of large bowel intussusceptions, a malignant aetiology has to be expected [5].

Vanek's tumour is a rare, benign, non-encapsulated lesion, composed mainly of loose connective tissues, vessels and eosinophilic inflammatory cells. It was first described as polypoid fibroma by Konjetzny in 1920, then by Vanek in 1949 (so called Vanek's Tumour) and it was finally named as IFP in 1953 by Helwig and Rainer, indicating that its nature was probably "inflammatory". Synonyms include inflammatory pseudotumour, granuloma with eosinophils, polyp with eosinophilic granuloma. IFPs occur most commonly in the stomach (about 70% of cases), mainly in the gastric antrum



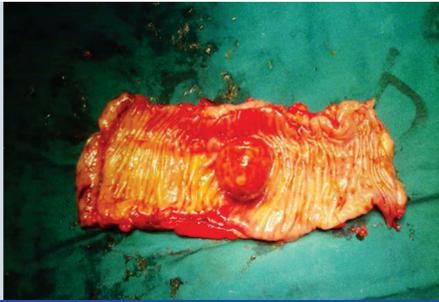
[Table/Fig-1]: CECT showing ileo-ileal intussusception and dilation of proximal small bowel loops with target sign



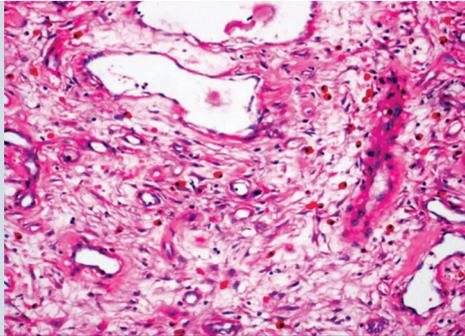
[Table/Fig-2]: Operative photograph showing ileo-ileal intussusception with dilatation of proximal segment



[Table/Fig-3]: Polyp as lead point of intussusceptum



[Table/Fig-4]: Specimen showed a firm, circumscribed, endoluminal 2x2.5cm polyp



[Table/Fig-5]: Histopathology of polyp showing necrosis, edema, inflammatory cells, proliferating blood vessels, fibroblasts (H & E, 100x)

and less frequently in the intestine. The symptoms are dependent on size and localization of the tumours in the gastrointestinal tract. Patients with IFPs in the small bowel are most likely to present with chronic episodes of colicky abdominal pain, lower gastrointestinal bleeding, anaemia and, more rarely, intestinal obstruction, caused by episodes of intestinal intussusception [6]. The occurrence of IFPs in the small bowel is rare, accounting for only 18% of all cases, and it is less frequent in the ileum, where these polyps rarely cause intussusceptions [4]. Recent review of cases of ileo-ileal intussusceptions caused by IFPs published in English, found only eight cases [7].

IFPs affect both sexes and all age groups, with a peak incidence in the 5th and 6th decades of life. Although most of the cases are sporadic, a familial relationship has also been described. The lesions have always been recorded as solitary polyps. The aetiology remains unknown, but it could be a consequence of chronic irritation and inflammation or a consequence of extreme reaction of the body to an intestinal trauma. Histologically, they arise from the submucosa and are characterized by vascular and fibroblast proliferations and an inflammatory response, dominated by eosinophils [7,8].

The lesions seem to lack a malignant potential and recurrence of the polyps has been reported only once [9]. Conventional abdominal radiography may show dilated bowel loops. Abdominal USG is a generally available technique in an emergency. Obesity and distended bowel loops decrease image quality. Computed Tomography (CT) scan has become the key-modality for the diagnosis of intussusception.

Histopathological differential diagnoses include spindle cell lesions such as inflammatory fibrosarcomas, spindle-cell carcinoids, and

gastrointestinal stromal tumours (GISTs). Immunohistochemical staining shows positivity for CD34, as in our case, suggesting that these polyps may develop from primitive perivascular or vascular cells. Negative staining for S100 protein and CD 117 (KIT gene product) distinguishes an inflammatory fibroid polyp from a neurogenic tumour or a gastrointestinal stromal tumour (GIST) [10-12].

An ileal inflammatory fibroid polyp causing chronic ileocolic intussusception can mimic a caecal carcinoma [13].

Resection is the treatment of choice for adults presenting with intussusceptions. It is important to differentiate between small bowel and colonic intussusceptions. Discussion continues as to whether or not a reduction has to be done before a resection. The high number of malignant causes of colonic intussusceptions makes en-bloc resection the best option, due to the lower risk of perforation/spillage. In case of small bowel intussusceptions, a reduction, followed by an elective resection, can be done

CONCLUSION

An intussusception is a rare cause of adult intestinal obstruction. It is generally caused by a definable intraluminal pathology. Although inflammatory fibroid polyps are seen very rarely in adults, they are among the probable diagnoses that should be considered in obstructive tumours of the small bowel. CT scan is useful in confirming anatomical abnormalities, but final diagnosis will be made, based on histopathological analysis. Surgical intervention is the rule in treatment and resection is the definitive method.

REFERENCES

- [1] Stewart D, Hughes M, Hope WW. Laparoscopic-assisted small bowel resection for treatment of adult small bowel intussusception: a case report. *Cases J.* 2008;1:432.
- [2] Chiang JM, Lin YS. Tumor spectrum of adult intussusception. *J Surg Oncol.* 2008;98:444-7.
- [3] Vanek J. Gastric submucosal granuloma with eosinophilic infiltration. *Am J Pathol.* 1949;25:397-411.
- [4] Wysocki AP, Taylor G, Windsor JA. Inflammatory fibroid polyps of the duodenum: a review of the literature. *Dig Surg.* 2007;24:162-8.
- [5] Begos D G, Sandor A, Modlin I M. The diagnosis and management of adult intussusception. *Am J Surg.* 1997;173:88-94.
- [6] Barussaud M, Regenet N, Briennon X, De Kerviler B, Pessaix P, Kohneh-Sharhi N, et al. Clinical spectrum and surgical approach of adult intussusceptions: a multicentric study. *Int J Colorectal Dis.* 2006;21:834-9.
- [7] Mohamud SO, Motorwala AS, Daniel AMR, Tworek JA, Shehab TM. Giant ileal inflammatory fibroid polyp causing small bowel obstruction: a case report and review of the literature. *Cases J.* 2008;1:341.
- [8] Kolodziejczyk P, Yao T, Tsuneyoshi M. Inflammatory fibroid polyp of the stomach. A special reference to an immunohistochemical profile of 42 cases. *Am J Surg Pathol.* 1993;17:1159-68.
- [9] Yakan S, Caliskan C, Makay O, Deneçil AG, Korkut MA. Intussusception in adults: clinical characteristics, diagnosis and operative strategies. *World J Gastroenterol.* 2009;15:1985-9.
- [10] Nonose R, Valenciano JS, da Silva CM, de Souza CA, Martinez CA. Ileal intussusception cause by Vanek's tumour: a case report. *Case Rep Gastroenterol.* 2011;5:110-16.
- [11] Tennika M, Jacobs, Andreas L. Lambrianides. Inflammatory fibroid polyp presenting as intussusception. *J. Surg. Case Rep.* 2013;2. rjt005 doi:10.1093/jscr/rjt005.
- [12] Singhal M, Singh P, Misra V, Dhingra V, Bhatia R. Inflammatory fibroid polyp of small intestine: Report of two cases with review of literature. *JCDR.* 2010;4:3241-4.
- [13] Naveen Gara, John S Falzarano, Whitney ML Limm, Thomas S Namiki, Laurie KS Tom. Ileal inflammatory fibroid polyp causing chronic ileocolic intussusception and mimicking caecal carcinoma. *World J Gastrointest Oncol.* 2009;1:89-92.

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FINANCIAL OR OTHER COMPETING INTERESTS: None.

Date of Submission: **Jul 05, 2013**
Date of Peer Review: **Oct 07, 2013**
Date of Acceptance: **Oct 27, 2013**
Date of Publishing: **Dec 15, 2013**