OPEN ACCESS
Saudi Journal of Pathology and Microbiology
Abbreviated Key Title: Saudi J Pathol Microbiol
ISSN 2518-3362 (Print) |ISSN 2518-3370 (Online)
Scholars Middle East Publishers, Dubai, United Arab Emirates
Journal homepage: http://scholarsmepub.com/sjpm/

Case Report

Ileocecal Intussusception Due to Heterogenous Cecal Polyps; One of Which Is Unusual; a Case Report

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DOI:10.21276/sjpm.2019.4.7.6 | **Received:** 04.07.2019 | **Accepted:** 14.07.2019 | **Published:** 23.07.2019

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Abstract

Background: Inflammatory fibroid tumor (IFT) is uncommon tumor in large bowl. In this patient it was the unusual cause for large bowel intussusception. More over the coexistence of mucinous adenocarcinoma arising from adenomatous polyp is a rarity previously unreported in the world literature. ***Case presentation:*** Here, we present the case of a 65 year old female who complained of lower abdominal pain with rectal bleeding occasionally. Imaging studied confirmed the diagnosis of intussusception of ileocecal type. After resection of intestines, two sessile polyps in caecum were detected. Histology of the polyp's revealed one showing classical features of IFP and the other a malignant growth arising from tubular adenoma with deep invasion upto serosal coat. Unusually IFP lacks CD 34 stromal cells. ***Conclusions***: Intussusception of ileocecal type due to sessile polyps in caecum is a rare surgical event. The heterogenous nature of these polyps being the cause for this unusual case is previously unreported.

Keywords: Inflammatory fibroid polyp, intussusception, caecal polyps: IFP and Adenoma with malignant transformation.

BACKGROUND

As such, intussusception is not a rare condition; upto 1954 itself approximately 750 cases had been reported; the first case of which was operated in a child by Sir. Jonathan Hutchinson in 1871. According to Azar *et al*. [1], intussusception in adults is much less common; representing 5% of all cases of intussusception, and 1% of all bowel obstruction. The over all incidence of this condition in adults has been estimated to be around 2-3cases per 1,000,000 populations. In children, it is mostly due to idiopathic causes while in adults, malignancy is more responsible for it and incidence as high as 65% was reported in the literature [2, 3]. Athanasius etal succinctly [4] described this condition as " an internal prolapse of proximal bowel and its mesenteric fold within the lumen of distal bowel as a result of overzealous or impaired peristalsis; further obstructing free passage of intestinal contents." Alternatively it is defined as the invagination of the gastrointestinal tract into the lumen of the contiguous distal segment. Wikipedia classifies intussusception into ileocleal (4%), ileocolic or ileocecal (77%), ileo-ileo colic (12%), Colocolic (2%), multiple (1%), retrograde (0..2%) and other (2.8%) types. In an extensive review of 34 cases of intussusception in adults, adenomatous polyps were responsible for 3 cases and carcinoma in 7 cases (carcinoma of small bowel 2 cases, carcinoma of large lowel in 5 cases) (Stubenbord and THorbjarnarson [5]. In this unusual case, two unrelated polyps in cecum were responsible for intussusception of large bowel; this collision being unreported previously.

CASE PRESENTATION

A 65 year old lady was admitted in a hospital for lower abdominal pain, lasting for three months. The pain became acute at the time of admission and the signs and symptoms suggested intestinal obstruction. She had rectal bleeding on few occasions also. Radiological and ultra sound findings suggested ileocolic intussusception. Though the polyps were not visualized, the patient was prepared for radical surgery. On operation, ileocolic intussusception was confirmed and two cecal polyps were detected. Both of them are sessile and situated close to one another. One was soft while the other was firm and ulcerated and covered with necrotic material. The proximally situated polyp measured 5.5cmsx4.0x4.5cms. Histologically this polyp

showed proliferation of small blood vessels in submucous coat; smooth muscle elements and few inflammatory cells including many eosinophils. However the mucosa was completely destroyed and replaced by a thick layer of eosinophils which were mature and not mixed with other inflammatory cells. Macroscopically and microscopically, the polyp was proved to be IFP (Figure1 and 2). The other polyp, measured same as above and situated distally was frankly malignant. At its top it retained features of tubular adenoma and as it extended deeper it assumed intestinal adenocarcinomatous features with deep invasion. The malignant glands were surrounded by mucinous pools. Mucinous adenocarcinomatous elements were present in the serosal aspect also. (Figure:3) Few lymph nodes were dissected nearby and they were free from metastases.

DISCUSSION

IFP is an uncommon submucosal lesion; which is benign and histologically distinctive. It was first described by Vanek in 1949 in the stomach and the present term was coined by Helwig and Ramier in 1943 [6]. The diagnosis is based upon mostly by finding out CD34 positive cells encircling small blood vessels in the submucosa. They were usually arranged in a whorled pattern and there are few case reports elaborating the absence of such arrangement [7]. Secondly the presence of significant number of eosinophils surrounding them is considered as a reliable histological marker of this condition [8]. In our patient, eosinophils encirclement was not seen but they formed a thick layer of more than 4cms in the substance of polyp. Such layering may mimick eosinophilic malignancy but the absence of other blastic cells in their midst ruled out such a possibility. Eosinophilic colon is not a possibility because of its histology.

Immunochemistry of IFP is a helpful procedure in its diagnosis; c-kit negativity rules out GIST and smooth muscle and neural tumors also can be ruledout by immunochemical methods [9]. Its histology mimicks inflammatory myoblastic tumor; composed mostly of myofibroblasts, plasma cells and lymphocytes; which are also positive for smooth muscle actin, desmin and anaplastic lymphokinase, and cytokeratin but negative for CD34; [9] While inflammatory myofibroblastes tumor is common in children, IFD is an adult based tumor.

The first case of colonic intussusception due to IFD was reported by Robert De La Plaza *et al*. in 1999 [10]. Till that time even though 23 cases were reported in colon; none of them was responsible for intussusception. Even in DeLa plazas case, IFP was solitary in a 63 yr old male admitted with a complaint of abdominal pain for two months as in our patient and with a radiological finding revealing a mass effect in the inferior hemiabdomen with shifted dilated small bowel loops. Even though our patient had severe pain in lower abdomen along with melena indicating bowel cancer, preoperative imaging results were inconclusive.

While IFP is covered with necrotic material due to ulceration, it was firm and fibrous on examination; on the other hand the other polyp was friable and mucoid and showed the evolution of mucinous adenocarcinoma from preexisting tubular adenoma. Colonic mucosa bearing IFP and the other malignant polyp is unusual except in one case wherein such a possibility occurred but this was in stomach (Ref-11) PDRGA mutation is incriminated in IFP while in familial adenomatous polyposis, the inherited gene defect was APC, MUTYH.

Abbreviations

IFP: Inflammatory fibroid polyp

Acknowledgments: nil relevant
Funding: nil relevant

Author's contribution

1. NK conceptualized the idea of publishing the clinical material in a journal and was mainly responsible for preparing the manuscript. She collected references and incorporated them in the text.
2. S.B.responsible for elaborating pathologic description and performing intricate manoevres in this study.
3. SR. Collaborated with main author in the preparation of this article and acted as a guide

Ps: All three participants read the article before dispatching the same.

Competing interests

The authors declare that they have no competing interests.

Consent for publication

Written consent was obtained from the patient for the publication of the case report.

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