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Case Report

Solitary Juvenile Polyp Presenting as an Ileo-Ileal Intussusception in an Infant: A Case Report

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Abstract

Juvenile polyposis syndrome (JPS) was first described in 1964. It is a rare (approximately one in every 100,000 individuals) autosomal dominant disease that is characterized by the occurrence of several juvenile polyps in the gastrointestinal tract. Juvenile polyp is a specific type of hamartomatous polyp. The term 'juvenile' refers to the polyp histology rather than the age of onset of the polyp. An isolated juvenile polyp is not diagnostic of JPS and occurs in approximately 2% of children and adolescents. Here we present a case of a 7 month old baby boy who presented with recurrent vomiting and abdominal pain and on examination revealed RIF mass and tenderness. Clinically, the diagnosis of intussusception was suspected and confirmed on sonographic examination. Exploration of the abdomen revealed ileo-ileal intussusception. Further, Juvenile polyp was identified by the presence of cystically dilated glands lined by cuboidal to columnar epithelium, filled with secretions, abundance of edematous lamina propria with inflammatory cells on histopathological examination. The relevance of this case report lies on the fact that appearance of juvenile polyp in ileum is rare.

Keywords: Ileo-ileal intussusception, Juvenile polyp, Juvenile polyposis syndrome.

INTRODUCTION

Intussusception is the invagination of a portion of intestine into another portion of intestine, which causes bowel obstruction [1]. Intestinal intussusception is the common cause of bowel obstruction in infants and children, that increases their morbidity and mortality rate [2–4]. The primary type or idiopathic intestinal intussusception is mostly seen on infants in 95% of cases. In aged children, the intestinal intussusception occurs due to the secondary identifiable causes that can be either intraluminal or intramural pathologies such as ileal polyp, Meckel's diverticulum, lipoma, appendiceal stump, duplication of ileum, malignancies, etc. It is reported that, in infants and children the pathological lead points causing intussusception is about 1.5 to 14% of the cases and that tends to increase with age. However, the intussusception caused by the ileal polyp as a pathological lead point is very rare [1].

CASE REPORT

A 7-month old baby boy presented with recurrent vomiting and abdominal pain for one day duration. The child was born on elective c-section as the mother is type 1 diabetic. The birth weight was 3 kg, post-natal period were uneventful and weaning started at 6 months and vaccinated up to the age. Further, no family history of intestinal polyp or gastrointestinal (GI) malignancy was reported.

On clinical examination, there was a RIF mass and tenderness and a hyper pigmented macule over left leg. On further investigation by USG abdomen showed acute ileo-ileal intussusception.

Exploratory Laparotomy done with ileal resection and ileo-ileal end to end anastomosis. We received a specimen of intestinal segment which was everted out, one resected end cut opened, measuring 1.5 cm in length with a polypoid mass with smooth lobulated surface, measuring 3.5x1.5x1 cm appears to arising from the mucosa. The H&E stained sections showed; pedicle of the polyp with increased lamina

propria, and cystically dilated glands which filled with secretions. A diagnosis of juvenile polyp with changes of intussusception in the ileal segment was made.

DISCUSSION

The gastrointesintal polyps can occur anywhere in the alimentary canal and it is most commonly seen in children. The juvenile types of polyps are mostly benign and isolated lesions. The genetic changes are also an underlying cause of polyps in children, hence special care must have been needed to children with family history of colon cancer, multiple, recurrent, or adenomatous polyps [5].

It has been reported that the intestinal intussusception are more common in male children than female children. Nearly 75% of the cases occur within initial two years of life and more than 40% cases seen between the age of 3-9 months old. The classical triad of intussusception symptoms, i.e. abdominal pain, abdominal mass and rectal bleeding/mucoid stools were reported in 30 -60% of cases. In idiopathic intussusception where no pathological lead point is known are due to thickened bowel wall lymphoid tissues (Peyer's patches) or non-specific mesenteric lymphadenitis. However, the intestinal intussusception causes by ileal polyp as pathological lead point is very rare in children [1].

The juvenile polyps from the specimen we received are histologically characterized with cystically dilated glands lined by cuboidal to columnar epithelium and filled with secretions, abundance of edematous lamina propria with inflammatory cells. If dysplasia is found in a typical juvenile polyp, the pathological diagnosis of juvenile polyp can be easily made (Figure 1, 2 and 3).

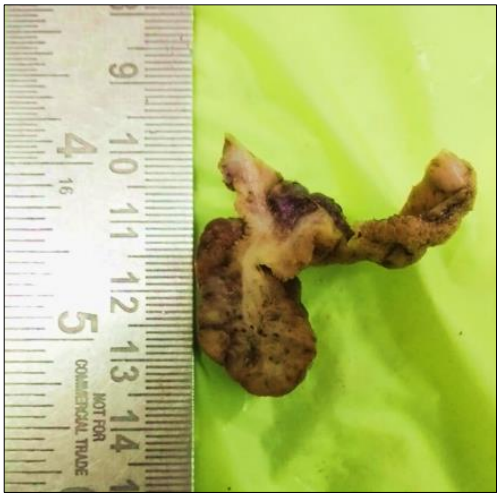

Figure 1: Resected specimen of ileum with polyp

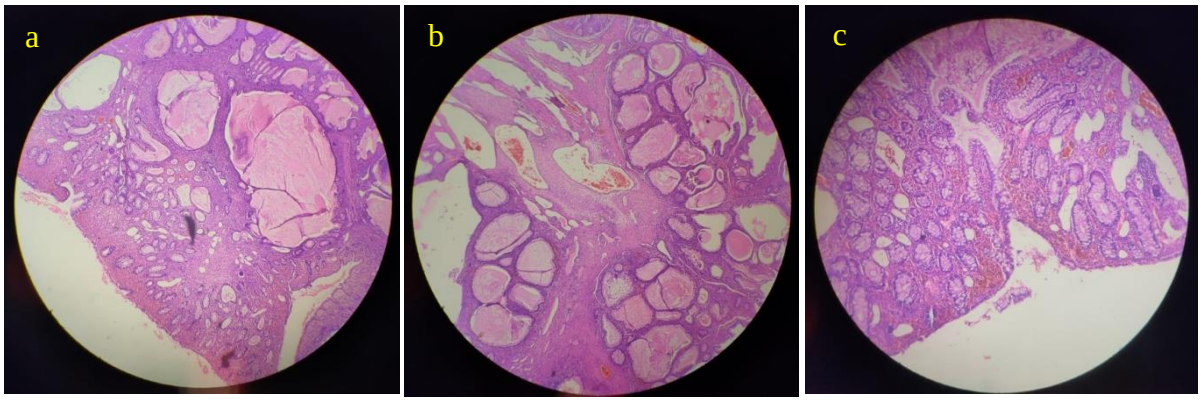

Figure 2: H and E stained sections of ileal polyp a) Cystically dilated glands filled with secretions (4x), b) Thickened muscularis mucosa(4x) and c) Areas of haemorrhage (4x).

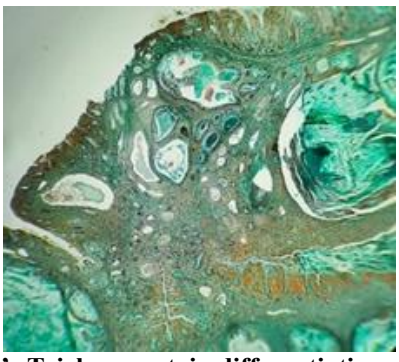

Figure 3: Ileal polyp in Masson's Trichrome stain differentiating muscle fibers and collagen (4x).

Germline mutations in SMAD4 or BMPR1A have been reported in 50-60% of patients with Juvenile polyposis syndrome (JPS). The JPS belongs to the group of hamartomatous polyposis, that includes Peutz-Jeghers syndrome (caused by germline STK11 mutation) and Cowden syndrome (caused by germline PTEN mutation). It is difficult to differentiate these hamartomatous polyposis without genetic mutational analysis. Hence, understanding germline gene mutation is important for accurate diagnosis of JPS [6].

CONCLUSION

In conclusion, juvenile polyp is the most frequent colonic polyp seen in children. Traditionally it has been described as single and located in recto sigmoid area. But in the present case we found a juvenile polyp in ileum and it act as a pathological lead point to cause an intussusception.

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Conflicts of interest: There are no conflicts of interest.

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