Case Report

A rare case of combined ileal lipoma and heterotopic gastric mucosa as the leading pathological point for adult ileo-ileal intussusception

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ABSTRACT

Intussusception is defined as telescoping of one segment of bowel into another one. It is an uncommon cause of intestinal obstruction in adults with a reported incidence of 1 in 1300 abdominal cases presenting as obstruction. Intussusception as a cause of intestinal obstruction in adults is rare. There is invariably an underlying pathology which leads to intussusception in adults, some of the main causes of this are carcinomas, polyps, strictures, benign tumors, Meckel's diverticulum, and colonic diverticulum. Here we present a case of intussusception which had a subserosal lipoma as a lead pathological point, but the histopathological examination revealed dual pathology for the same. A subserosal lipoma with presence of heterotopic gastric mucosa was diagnosed in the ileum as the leading pathological point of ileo-ileal intussusception. This case highlights the importance of pathological examination to know the specific cause of intestinal obstruction.

KEYWORDS

Heterotopic gastric mucosa, intussusception, subserosal lipoma

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INTRODUCTION

Intussusception is defined as invagination of one segment of the bowel into an immediately adjacent segment.1 The main causes of intussusception in adults are carcinomas, polyps, strictures, benign tumors, Meckel's diverticulum, and colonic diverticulum.2

Benign tumors like lipoma are one of the pathological leading point of intussusception. It is a benign tumor of fat cells that are soft painless nodules that vary in size.3 They are encountered with decreasing order of frequency in the colon (65%-75%), small intestine (20%-25%), stomach, and esophagus. Their usual location in the small intestine is ileum (50%) while jejunum is the least common.4

When examining an intestinal lipoma, specifically, a small bowel lipoma, there are typically three types: inter muscular lipoma, subserosal lipoma, and submucosal lipoma. Submucosal lipomas are the most common, making up 90% of intestinal lipomas.5 Submucosal and subserosal lipomas tend to be the ones that cause intussusception, and large subserosal lipomas are prone to causing intestinal compression and volvulus. 6 Intussusception only represents 1% of all adult bowel obstruction cases with less than 4% of cases due to an intestinal lipoma.3 Small bowel lipomas can be either sessile or pedunculated and can have regular or lobulated contours. Their overlying mucosa can even have ulceration, especially when in the duodenum--the part of the small bowel exposed to the most gastric acid.7

Heterotopic gastric mucosa (HGM) can occur anywhere in the gastrointestinal tract; nevertheless, its occurrence in the small intestine is rare unless associated with remnants of vitelline duct (Meckel's diverticulum).8 Isolated, heterotopic gastric mucosa (HGM) is a focus of mature gastric tissue in a location outside the stomach, not associated with congenital anomalies such as Meckel's diverticulum or intestinal duplications. Isolated HGM is rarely found in the ileum and even more rarely it acts as pathologic leading point (PLP) for intestinal intussusception. 9

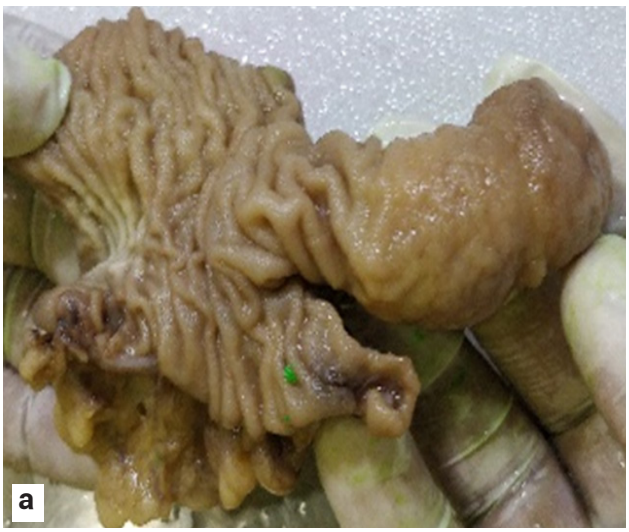
HGM may be present anywhere in the gastrointestinal tract from the mouth to the anus and can be found also in the airways, umbilicus, urinary bladder, and even in the scrotum. Heterotopic gastric mucosa is supposed to be of vitellointestinal tract origin and may cause intussusception as being a lead point in the ileum.10 A definitive diagnosis of heterotopic gastric mucosa is established by histopathological examination and it is important to differentiate heterotopia. It is usually clinically silent and surgical intervention can be considered in patients with complications such as gastrointestinal hemorrhage and intestinal obstruction.11 Heterotopic gastric mucosa comprises the gastric epithelium and fundal glands, as well as the chief and parietal cells. Intestinal ulceration due to peptic secretion of heterotopic gastric mucosa can cause recurrent inflammation.12

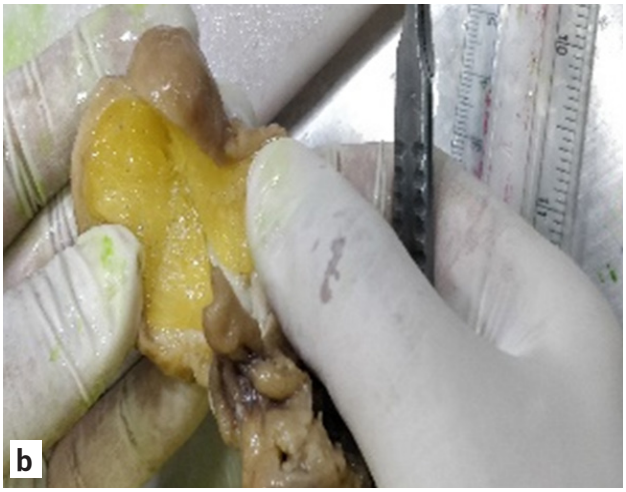
Here, we present a case of adult ileo-ileal intussusception with two leading point present concomitantly. A subserosal lipoma with overlying gastric heterotopic mucosa as a leading cause of adult small intestinal intussusception.

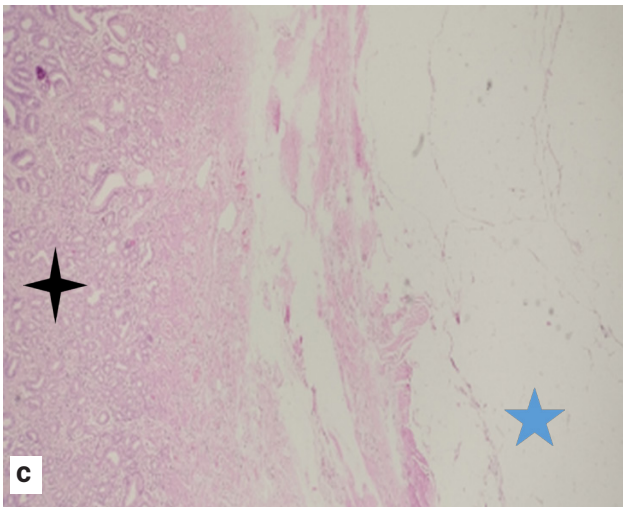
CASE REPORT

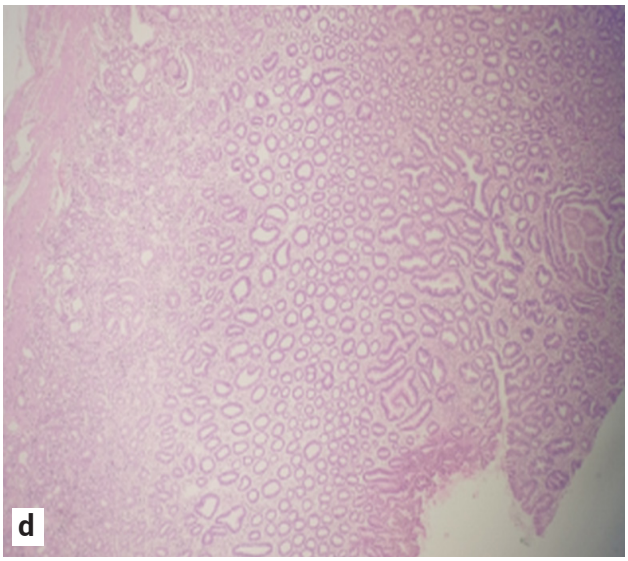
A 23 years old male presented to this hospital with complaints of pain abdomen which was acute on onset, without any aggravating and relieving factors. The pain subsided with analgesics and was admitted for operation and relieve of intestinal obstruction. Past history was insignificant. On examination, physical examination was normal. His vitals were within normal. All other systemic examination was within normal limits. CT scan showed an ileo-ileal intussusception was noted in right lower quadrant in distal ileum. An approximately 4.7x2.4 cm2 sized fat density lesion with thick enhancing capsule is noted within the intususceptum as leading point. Rest of the findings were normal. Exploratory Laparotomy with resection anastomosis for ileo-ileal intussusception was done. Operative finding was that of ileo-ileal intussusception with large intraluminal lipoma. Ileo-ileal intussusception anastomosis was done. Gangrenous changes or perforation or collection are not noted.

In the Pathological department, a segment of ileum with released intussusception was received. Part of ileum 6.0 cm in length. On further cut opening, a mucosa covered sessile polyp was identified measuring 5x 1.5 cm. Cut section of polyp was solid, fatty smooth area. It was 3 cm and 1 cm from both resected end. The mucosa around the polyp is flattened while rest of the mucosa is grossly unremarkable.









(Fig a: Gross of lead point polyp, b: Cut section of polyp revealing yellow soft and lobulated fatty tissue, Black arrow) Fig c: H & E revealing subserosal lipoma Asterik: mucosa, star: subserosal fatty tissue of lipoma, d: Mucosa with heterotopic gastric mucosa-10X magnification)

Section from ileal polyp shows a polypoidal tissue lined by heterotopic gastric mucosa comprising of chief cells and mucus secreting cells. Some of the glands were cystically dilated. Focal area of surface ulceration was also noted. Rest of the glands show intestinal mucosal lining with villous architecture and reactive atypia. Numerous congested thin walled blood vessels and mixed inflammatory infiltrates are noted in the lamina propria. Subserosal lobules of mature adipose tissue separated by fibrous septa with capsulation was noted beyond the muscularis propria suggestive of subserosal lipoma. Sections examined from rest of the ileal mucosa were within normal histologic limit. There was no evidence of malignancy. The histopathological diagnosis was subserosal lipoma with overlying heterotopic gastric mucosa, consistent with clinical evidence of Intussusception. The post-operative period of the patient was uneventful and was discharged shortly, free of disease.

DISCUSSION

Intussusception was firstly described by Paul Barbette in 1674, and the Scottish surgeon James Hunter coined the term "intussusception" in 1793.13 Adult intussusception is a rare condition, with an incidence of 2/10,00, 000 cases per year worldwide. 14

Intestinal lipomas are benign, non-epithelial, intestinal tumors with an exceptionally rare localization at the ileum. 15 If present, 90% are seen in submucosal and 10% in subserosal location. Mostly located in the colon, but they can be found in the esophagus, small intestine, and rarely in the stomach. 16 Small intestinal lipomas produce symptoms more often than colonic lipomas, irrespective of the size of the tumor.17

Usually, they present as a solitary, sessile or pedunculated polypoidal mass emerging from the submucosa without any lesion of the mucosa and may also affect the sub serous layer and epiploic appendices. Imaging methods can contribute greatly to the diagnosis. The treatment of intussusception in adults is almost always surgical, depending mainly on the size of the lipoma, location, preoperative diagnostic confirmation, or the presence of complications.18

Clinical presentation of HGM varies and depends on the location and size of the heterotopic tissue. The most common complication of HGM is intestinal mucosal ulceration with gastrointestinal bleeding. It can also form an intraluminal mass causing intestinal obstruction. Rarely, it can serve as a lead point for intussusception. Several cases of intussusception led by HGM in children have been reported in the literature. However, this event is exceedingly rare in adults.8 A pre-operative diagnosis of HGM in the small bowel also appears both inconvenient and offers little benefit.19 So, a high level of suspicion is required to diagnose this entity, However, the diagnosis can be challenging preoperatively as the clinical features are those of intestinal obstruction until and unless associated with Meckel's diverticulum. However,

this case was diagnosed preoperatively as intussusception with intraoperative finding of lipoma as pathological lead point. Microscopic examination proved that there was dual pathology.

In summary, intussusception is a rare cause for intestinal obstruction in adults. Our case represents an exceedingly rare cause for intussusception in adults where HGM in the ileum was associated with subserosal lipoma serving as a dual pathology existing together and giving rise to a lead point for ileo-ileal intussusception, and was treated successfully with surgical resection. The diagnosis of dual pathology being confirmed in the histopathology only.

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