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Rare diagnosis of intestinal lipomatosis complicated by intussusception in an adult: A case report



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ABSTRACT

INTRODUCTION: Intestinal Lipomatosis consists of diffuse lipomas in various regions from the small to large bowel. They can remain asymptomatic or present with complications such as Intussusception.

DISCUSSION: Intestinal lipomatosis complicated by Intussusception is a rare occurrence that has not been well documented. Rare condition management is difficult to approach because of the customizability each scenario requires. We hope through sharing our approach this can serve as a rough template to physicians who find themselves in a similar scenario. Overtime, as more case reports and surgical approaches are recorded we can establish future advancements in surgery.

PRESENTATION OF CASE: We present the case of a 47 year-old male who arrived at the Emergency Department with a chief complaint of abdominal pain. A CT scan revealed ileocolic intussusception. An intramural lipoma of the terminal ileum served as the lead point. Exploratory laparotomy confirmed the Intussusception and a right hemicolectomy was performed to repair the affected area. Examination of the resected large bowel showed diffuse thickening of the mucosa in the area of the cecum confirmed to be submucosal lipomatosis on histological examination. Patient was discharged on the fifth post-operative day.

CONCLUSION: This case confirmed previous treatment modalities in the management of intussusception. It also corroborates the complication of intussusception with Intestinal lipomatosis. It teaches us the importance of keeping a wide differential when considering a diagnosis of bowel obstruction. Through imaging, surgical exploration, and pathological interpretation, this case, which began as a complaint of abdominal pain, concluded as a rare clinical entity.

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1. Introduction

This case report is in line with the SCARE guidelines for surgical case reports [12]. Intestinal lipomatosis, first described by Hellstrom in 1906, is a rare disease with an incidence at autopsy ranging from 0.04% to 4.5%. Lipomas can be found diffusely throughout the intestines with most being asymptomatic. Invasive management is not advised unless complications arise such as intussusceptions, obstruction, bleeding, or perforation leading to peritonitis [9,10]. Intussusception was amongst the most rare of complications [2]. We report the case of intestinal lipomatosis complicated by intussusception in a 47 year-old male, an extremely rare event.

Intussusception is the invagination of proximal bowel into distal bowel, otherwise known as ‘telescoping’ of the intestines. It is the most common idiopathic cause of bowel obstruction in certain pediatric populations (6–36mo) but very rarely occurs in adults [1].

For this reason the diagnosis can be overlooked. When found in an adult the cause is usually secondary to a pathological process such as neoplasm. With the need for early surgical correction as a form of treatment, it is important to keep the differential in mind when an adult presents with bowel obstruction.

2. Presentation of case

We present the case of a healthy 47 year-old Hispanic male, who presented to the emergency room with abdominal pain for 5 days. The pain started in the epigastric area and shortly thereafter, became diffuse. He admitted to nausea, vomiting, sweating, and chills, without fever. Laboratory findings showed a white blood cell count of 14,000 per microliter of blood. Upon physical examination the patient was in acute distress, anxious, and agitated. During examination of the abdomen there were no obvious lesions or discolorations. Bowel sounds were diminished and the abdomen was firm, distended, and tender to palpation. He expressed severe tenderness and guarding that was more pronounced at the right lower quadrant. Patient did not have frequency, urgency, dysuria, or hematuria, but did present with rectal bleeding. McBurney’s sign

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Fig. 1. Submucosal lipoma and ileocecal Intussusception.

was positive. A CT scan of the abdomen and pelvis was ordered to further investigate the severe abdominal pain. CT showed ileocolic intussusception and surgery was scheduled (Fig. 1).

Ileocolic intussusception with an intramural lipoma as the lead point was identified. Surgery was performed and the resected specimens, consisting of small and large bowel, were sent for histopathology. The small bowel measured 32 × 5 cm and large bowel measured 19 × 6 cm. The small bowel revealed a dark hemorrhagic surface, with an exophytic hard sub-mucosal nodule measuring 3 × 3 × 2 cm at 7 cm from the proximal margin. Upon opening the lumen, the small bowel revealed a hemorrhagic mucosa. The large bowel showed irregular nodular mucosa that was diffusely present in the cecum, which was confirmed by histological examination to be submucosal lipomatosis (Fig. 2).

Some diagnostic challenges were faced due to the fact that the patient was a poor historian relative to his severe pain. During the initial evaluation, McBurney's sign and an elevated WBC count were present on physical exam raising suspicion for appendicitis. This could have resulted in direct admission to the operating room without considering any type of imaging to suspect the intussusception. The CT scan of the abdomen and pelvis did not indicate any signs of diffuse intestinal lipomatosis (Fig 3).

The diagnostic reasoning behind this case came from the information gathered from the CT scan of the abdomen. Differential diagnoses included acute appendicitis, small and large bowel obstruction, and colorectal carcinoma. Due to the intussusception

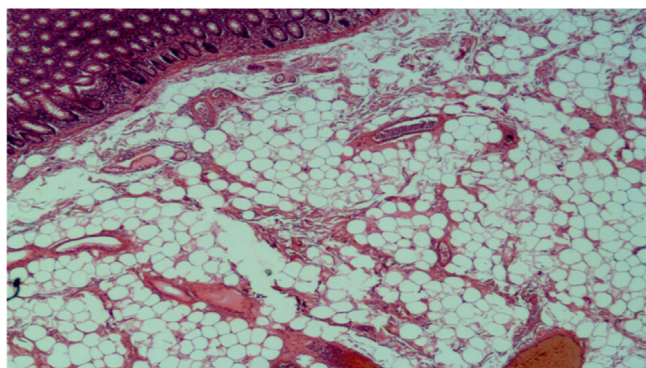


Fig. 2. Cecum, Submucosal Lipomatosis.

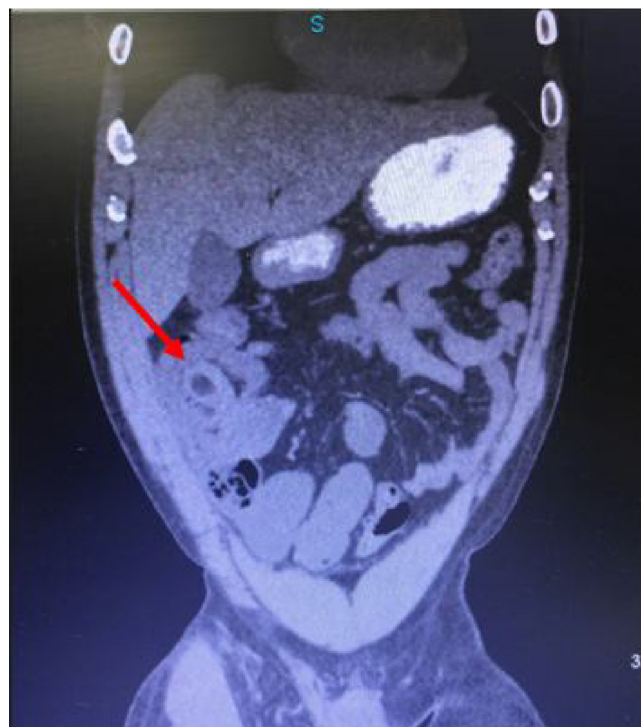


Fig. 3. Coronal view of submucosal lipoma and ileocecal intussusception.

the adult complication may present with acute, chronic, or intermittent symptoms. Whether it is acute or chronic, the presentation of adult intussusception is similar to that of small and large bowel obstruction. The three symptoms of tenderness, abdominal mass and rectal bleeding are rare. A prognostic characteristic that pointed to a benign etiology was the submucosal lipoma serving as the lead point rather than a malignant etiology.

The patient was placed on NPO and a foley was ordered. Lactated ringers solution was infused at 125 cc/h. Protonix at 40 mg IV daily for gastrointestinal prophylaxis. Pneumatic compression stockings were placed bilaterally on the legs for DVT prophylaxis and low molecular weight heparin was given at 40 mg subcutaneously. Dilaudid and Zofran were given as needed for pain and nausea (Fig. 4).

The surgical procedures performed included a diagnostic laparoscopy converted to open laparotomy, and a right hemicolectomy. An ileocecal intussusception up to the level of the distal ascending colon was found along with a dilated and ischemic cecum with impending perforation. The appendix also appeared to have chronic inflammatory changes. A right hemicolectomy was performed with ileotransverse colon anastomosis.

Reductions prior to surgical resection have been done in other cases of adult intussusception. In this case it was avoided because of the risk of perforation as bowel segments were severely inflamed and distended. Post operative instructions included admission to ICU and gastrointestinal specialist consult. Supportive care was implemented.

Post surgical progress of the patient was uneventful. Patient was sent to the Intensive Care Unit for 48 h following surgical procedure, extubated with IV hydration, pain management, gastrointestinal, and DVT prophylaxis. IV antibiotics were given to prevent any type of infection. Pathology report confirmed intussusception with area of necrosis and submucosal lipoma at the level of the terminal ileum. Large bowel partial resection with intestinal lipomatosis was also confirmed. Patient was discharged home on post-operative day 5 and was seen in the office for follow-up without any complications.

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