Case Report

Diffuse Intestinal Lipomatosis Presenting as Recurrent Intussusception: A Case Report

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Abstract

Ileo- ileal intussusception is a rare case of intestinal obstruction in adults, often secondary to a pathological lead point. Diffuse intestinal lipomatosis is characterized by multiple lipomas within the intestinal wall and is an uncommon entity contributing to intussusception. We present a case of an adult male with ileo-ileal intussusception due to diffuse intestinal lipomatosis, highlighting its clinical presentation, diagnostic approach and surgical management.

Keywords: ileo-ileal intussusception; diffuse intestinal lipomatosis; surgical resection; histopathology; diagnosis

Introduction

Intussusception in adults is rare, constituting only 1-5% cases of bowel obstruction.[1] Unlike pediatric cases, where idiopathic causes predominate, adult intussusception often has an underlying pathology such as tumors, adhesions, or inflammatory conditions.[2, 3, 4] Diffuse intestinal lipomatosis is a rare, benign condition with an incidence of 0.04-4.5% on autopsy, characterized by diffuse proliferation of mature adipose tissue in the submucosal layer of the intestine.[5] This condition may present with non- specific symptoms such as abdominal pain or altered bowel habits but in rare cases intussusception causing intestinal obstruction is also documented.[6] Due to its rarity, there is limited literature on its prevalence, diagnosis and management in adults.

Case Report

A 24 year old male presented to the emergency department with complaints of severe abdominal pain for two days associated with multiple episodes of vomiting and non-passage of stools and flatus since one day. In present episode, the pain was around umbilicus, at times radiating to back. It was insidious in onset, progressively increasing in intensity, colicky in nature and accompanied by persistent abdominal cramps which was not relieved by analgesics. There were around 7-8 episodes of projectile, bile stained vomiting, each occurring at a gap of 2-3 hours associated with history of on and off constipation for the last 5 days and absolute constipation since one day. Patient also gave history of decreased urinary output from last 2 days. He had anorexia and weight loss of approximately 5 kgs in last 2 months. There was no associated history of fever, diarrhoea, melena, hematochezia, trauma or any other chronic illness.

On physical examination patient was conscious, alert, dehydrated and afebrile with pulse rate PR-109/minute, BP- 100/60 mmHg, RR- 20/minute and distended abdomen. On palpation there was generalised abdominal tenderness with guarding and rigidity but no organomegaly. On auscultation bowel sounds were present but decreased in intensity and frequency. Other systemic examinations were normal. Routine investigations were done including chest Xray and abdominal ultrasound. On chest Xray, free gas under the diaphragm was observed and USG abdomen revealed left renal calculus of approx.4 mm size and dilated small gut loops -? Intestinal obstruction. Because the patient presented with peritonitic features and free intraperitoneal air, emergency surgery was indicated. No CT scan was performed due to the need for immediate laparotomy.

The patient was admitted and emergency exploratory laparotomy was performed. Approx. 500 ml hemorrhagic fluid was present in peritoneal cavity. Whole of jejunum and proximal ileum were dilated with collapsed distal ileum, caecum and colon. The small bowel (ileum) at the site of obstruction was edematous, thick walled, having jumbled up gut loops and gangrenous patches. Adjoining mesentery was also thick walled having enlarged lymph nodes. Small bowel resection with end ileostomy was performed and resected specimen was sent to the pathology department.

On gross examination, multiple sessile, well-defined, homogeneous, non-encapsulated nodules with a soft-rubbery texture were observed along with perforation measuring 1cm in diameter proximal to intussusception. These nodules varied in size, ranging from 0.3 to 5.5 cm in diameter, and were found projecting into the lumen of the gut and on the serosal surface. They were present throughout the gut, including at the site of intussusception.

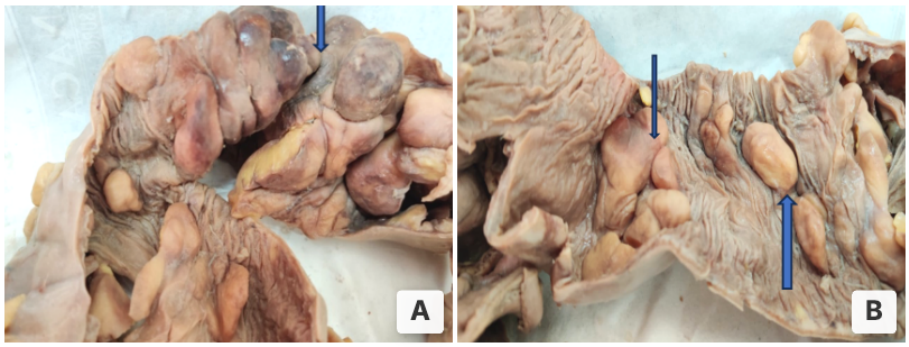

Figure 1: A: Resected ileal segment showing diffuse mural thickening and multiple submucosal nodules. B: Serosal surface demonstrating scattered yellow nodules consistent with lipomatous proliferation.

On microscopy, these polypoidal nodules showed diffuse expansion of submucosa by lobules of mature fibroadipose tissue with thinned out and atrophic ileal mucosa. Capsule was not seen. Sections examined from site of perforation

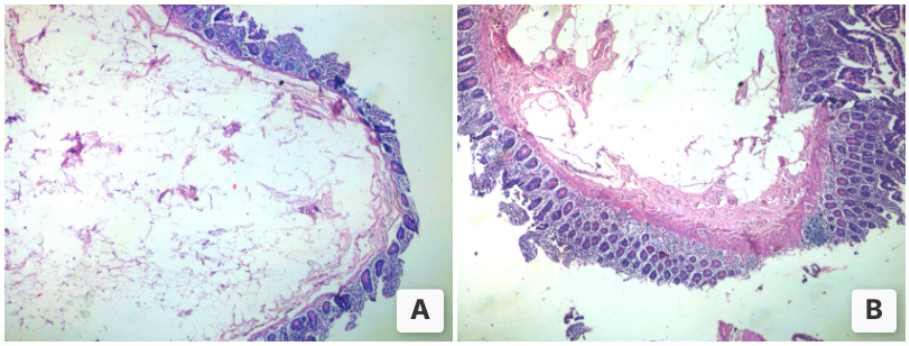

Figure 2: Microscopic features of intestinal lipomatosis. A: Submucosa expanded by mature adipocytes with overlying thinned mucosa (H&E, 40×). B: Higher magnification showing non-encapsulated adipose proliferation without atypia (H&E, 100×).

revealed mucosal ulceration, transmural congestion, edema, infiltration by mononuclear inflammatory cells, focal necrosis and extensive suppurative serositis. Histopathological findings were consistent with ileo-ileal intussusception with gut perforation secondary to diffuse intestinal lipomatosis.

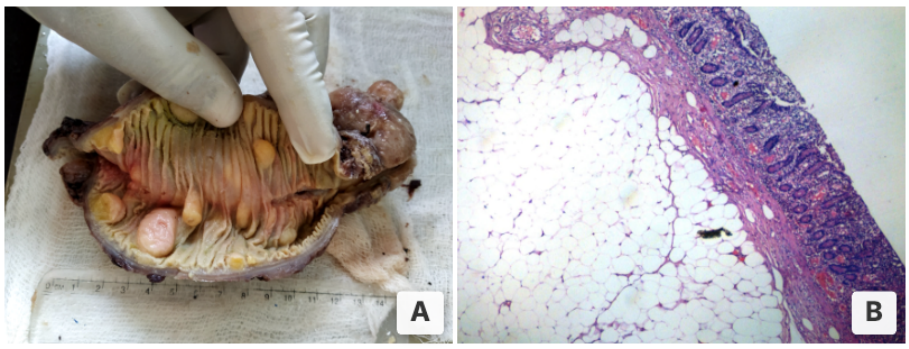

Figure 3: Second resection specimen. A: Gross specimen of remaining segment of ileum showing multiple uniform submucosal nodules consistent with residual diffuse lipomatosis. B: H&E stained sections at 10× showing submucosal lipomatous proliferation.

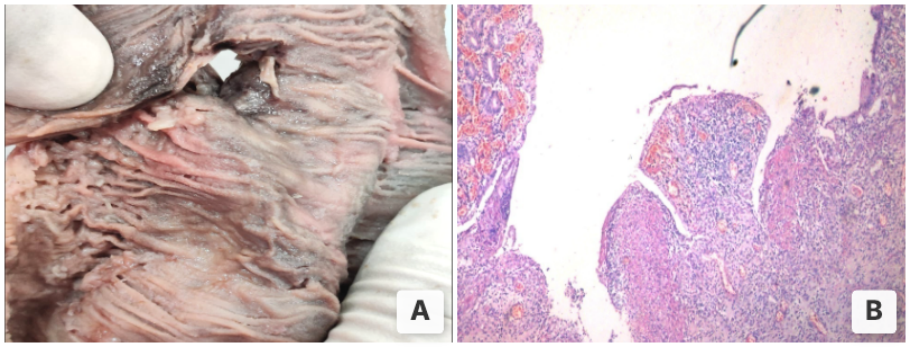

Figure 4: A: Intestinal mucosa revealing an area of perforation. B: H&E stained sections at 20× showing mucosal ulceration, transmural congestion, edema, infiltration by mononuclear inflammatory cells and focal necrosis.

The patient was discharged on the 5th post-operative day and asked to report after 3 weeks for ileostomy closure. At 3-week follow-up, the patient reported mild intermittent abdominal cramps. Ileostomy-site endoscopy demonstrated multiple smooth submucosal nodules within a 10-cm ileal segment, with normal proximal mucosa. Since symptoms persisted and nodules suggested residual disease, segmental resection of 15 cm of ileum proximal to the stoma was performed, followed by end-to-end ileo-ileal anastomosis. The resected specimen was sent to pathology department which revealed similar polypoidal nodules on mucosal surface, 0.2 to 2.5 cm in diameter. Microscopy of polyps revealed similar submucosal lipomatous proliferation.

After both the surgeries, we did follow-up for period of 3 years and patient continued to be symptom free without recurrence of lesion. Written informed consent was obtained from the patient for publication of this case report and images.

Discussion

Intussusception in adults is an infrequent clinical entity with an incidence of 0.002-0.003%. Enteric intussusception represents approximately 40–45% of these cases. Unlike pediatric intussusception, which is often idiopathic, 90% of adult cases have a pathological lead. Lipomas are the cause of intussusception in adults in 66.7% of cases.[7] Mostly these lipomas are solitary but few cases of diffuse adipose tissue infiltration of submucosa without tumor formation, known as lipomatosis have also been reported. This can be differentiated from lipoma by lack of encapsulation.[8, 9]

Intestinal lipomatosis also known as lipohyperplasia is an exceedingly rare condition accounting for 0.04-4.5% of autopsy cases with an unclear etiology.[10] The pathogenetic mechanism have not been described yet but several etiological factors have been suggested including embryonic displacement if of adipose tissue, congenital predisposition disturbance of fat metabolism, chronic inflammatory conditions such as inflammatory bowel disease, infection, hamartomatous syndromes and alcohol consumption.[11] It usually occurs in fourth decade of life with no sex predominance or predilection for particular site. The majority of cases are asymptomatic or present with mild symptoms however complications such as intussusception, obstruction, or perforation can occur.

The primary diagnostic modality is CT, which differentiates lipomatosis from other pathologies.[12] Intestinal lipomatosis is seen as well-defined homogeneous intramural hypodense areas on multidetector CT. It can be differentiated from true lipoma as they present as an asymmetric mass, whereas lipomatosis manifests as symmetric enlargement. The recurrence of symptoms three weeks postoperatively, along with persistent submucosal nodules on endoscopy, supports the diagnosis of diffuse lipomatosis rather than a solitary lipoma. The need for a second resection highlights the segmental and multifocal nature of the disease.

Several conditions may mimic diffuse lipomatosis clinically or radiologically. True submucosal lipomas are the closest mimickers; however, they typically present as solitary, well-circumscribed, encapsulated lesions rather than the diffuse, non-encapsulated adipocytic proliferation seen in lipomatosis.[13, 14, 15] Hamartomatous polyposis syndromes, such as Peutz–Jeghers syndrome, may produce multiple intraluminal polyps causing intussusception, but these lesions display characteristic arborizing smooth muscle cores and are accompanied by mucocutaneous pigmentation, which were absent in this case.[16] Pneumatosis intestinalis may superficially resemble multiple mural nodules, yet consists of gas-filled cysts rather than adipose tissue and shows a distinct radiolucent pattern on imaging.[17, 18]

There is no consensus regarding ideal management. Resection of the affected segment is essential to ascertain the nature of lesion and to rule out any malignant pathology. Once the diagnosis of lipomatosis is confirmed, surgical resection is the treatment of choice for symptomatic cases, as it alleviates symptoms, prevent complications and recurrence.

Conclusion

This case highlights the importance of considering diffuse intestinal lipomatosis as a rare cause of intussusception in adults. In emergency settings where CT cannot be performed, diagnosis relies on intraoperative findings and histopathology. Early recognition and surgical management are crucial for favorable outcomes. Future studies should explore the pathogenesis and management approaches for improved patient care and outcomes.

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