Section: General Surgery

Case Series

ADULT INTUSSUSCEPTION DUE TO ANGIOMYOLIPOMA: A CASE SERIES

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ABSTRACT

Adult intussusception is an uncommon cause of intestinal obstruction and, unlike paediatric cases, is usually associated with an underlying pathological lead point. Intestinal angiomyolipoma (AML), a benign mesenchymal tumour composed of adipose tissue, smooth muscle, and thick-walled blood vessels, is a very rare extrarenal lesion that may present as adult intussusception. Its clinical manifestations are nonspecific, while radiological findings frequently mimic more common lesions such as lipomas or gastrointestinal stromal tumours, making preoperative diagnosis difficult. Definitive diagnosis relies on histopathological examination demonstrating the characteristic triphasic morphology. We present a series of 12 adults with intestinal angiomyolipoma presenting as small bowel intussusception. Preoperative imaging consistently suggested lipoma, whereas histopathology confirmed angiomyolipoma in all cases. All patients underwent bowel resection with primary anastomosis and had favourable postoperative outcomes.

INTRODUCTION

Adult intussusception refers to a situation where one part of the gastrointestinal tract gets folded into another. 5% of all intussusception cases occur in adults and intestinal obstruction reported in 1% of all and is associated with atypical presentation.1 Unlike in children, adult intussusceptions have pathological lead points, usually malignancies. In colonic intussusceptions, malignancy is common, while benign processes such as lipomas are common in small bowel cases.2,3 The symptoms include abdominal pain, nausea, vomiting, and obstruction, making diagnosis difficult. Computed tomography (CT) scans are used for their high sensitivity in identifying pathology and lead points. Surgery should be done because of the presence of pathological processes.2,4,5

Angiomyolipoma (AML), a mesenchymal tumour consisting of fat, smooth muscle, and blood vessels, belongs to the PEComa group and displays immunoreactivity towards melanocytic and smooth muscle proteins.6 Extrarenal AMLs are rare with most AMLs arise in the kidney, often in association with tuberous sclerosis complex. Rare cases of extrarenal AMLs have been reported in the liver, retroperitoneum, adrenal gland, skin, and pelvic region. Gastrointestinal AML is extremely rare and can cause intussusception or intestinal obstruction. It poses diagnostic difficulties due to its similarity to other types of tumours, such as gastrointestinal stromal tumours and lipomas.7 Due to the heterogeneous content of AMLs on CT scans, diagnosis before surgery is difficult. A definitive diagnosis relies on histopathological and immunohistochemical evaluations performed post-surgery.8

AMLs display fat cells, smooth muscles, and vascular structures, while immunohistochemistry highlights positivity for specific stains such as HMB-45.8 In cases where there is a risk of bowel ischaemia, surgery is necessary for the successful treatment of adult intussusceptions resulting from angiomyolipomas. Intestinal angiomyolipoma causing adult intussusception is extremely rare, with limited literature describing its clinical, radiological, and pathological features. We present a series of cases highlighting the surgical and histopathological findings of this uncommon entity.

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Case Presentation

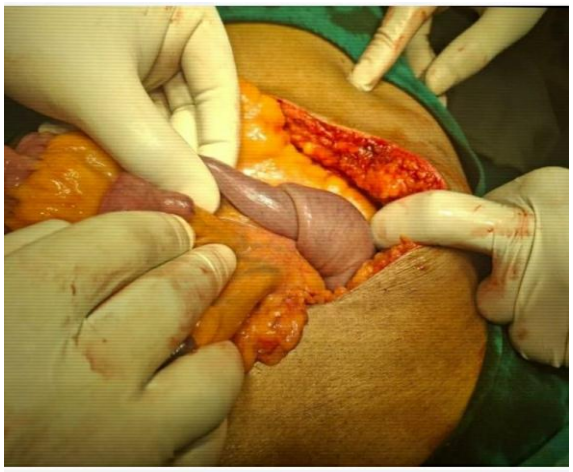

Figure 1: Dissection of ileal segment showing Ileoileal intussusception

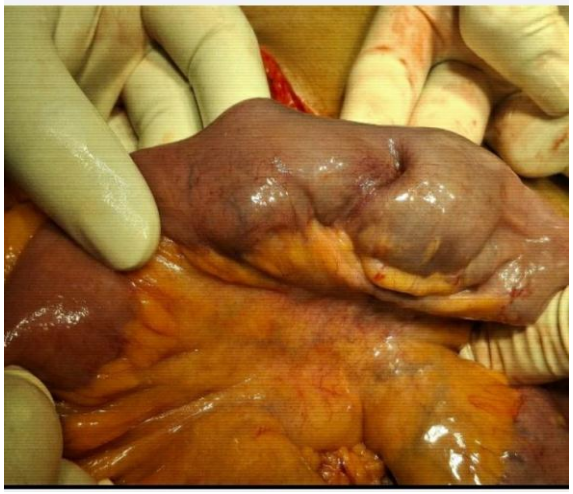

Figure 2: Manual reduction of edematous bowel segments

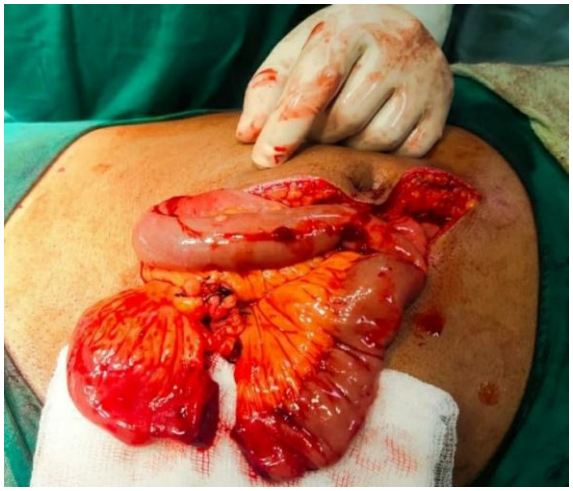

Figure 3: Segmental resection of 5-10cm of edematous distal ileum

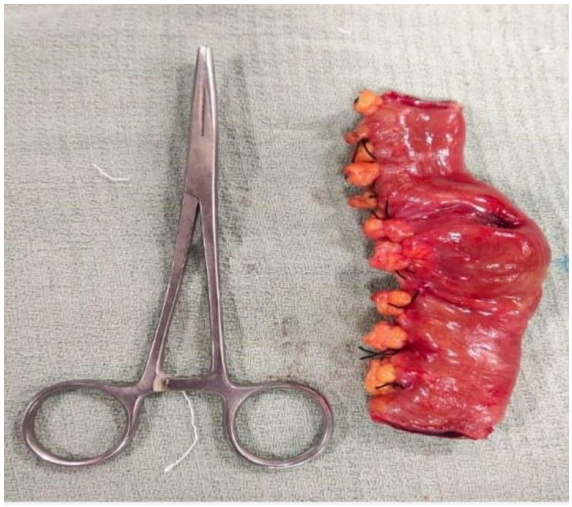

Figure 4: Resected ileal specimen of size 8*2.5cm approximating the length of a curved artery forceps

Case 1

A 54-year-old man presented with 20 days of chronic lower abdominal pain. His vitals were stable. He appeared pale and moderately nourished. Abdominal and rectal examinations were unremarkable. Blood tests showed haemoglobin (Hb) levels of 10.7 g and a Total count of 10700 cells/cu. mm, Platelet 3.6 Lakhs, Creatinine 0.6, LFT(N). Ultrasound revealed distal small bowel intussusception, submucous lipoma, and hepatic cysts in both liver lobes. The patient had no comorbidities. CECT suggested an ileal lipoma with ileoileal intussusception and small bowel obstruction, a 3.4x3.1 cm polypoidal lesion. Exploratory laparotomy with ileal resection and ileoileal end-to-end anastomosis was performed. The postoperative period was uneventful. Histopathology revealed a well-circumscribed nodular lesion measuring 2.9x1.5x2cm. The external surface was grey-brown and nodular. The cut surface was grey-yellow and grey-white with haemorrhagic areas and firm in consistency. Microscopy revealed congestion and haemorrhage of the ileal mucosa. Submucosa had clusters of mature adipocytes with thick-walled vessels, bundles and fascicles of smooth muscles, and dense inflammatory infiltrate.

Case 2

A 45-year-old woman presented with 24-day history of chronic lower abdominal pain. Her vital signs were stable, and she was moderately nourished. Abdominal and rectal examinations were unremarkable. Blood tests showed haemoglobin (Hb) levels of 10 g and a total cell count of 8300 cells/cu. mm, Platelet 4.0 Lakhs, Creatinine 0.6, LFT(N). Ultrasound revealed distal small bowel intussusception and a submucous lipoma. The patient had no comorbidities. CECT revealed an ileal lipoma with ileocolic intussusception and small bowel obstruction, measuring 4.4x2.1 cm polypoidal lesion. Exploratory laparotomy and ileum resection with ileocolic anastomosis were performed. The post-intervention period was uneventful. A 4 cm mass

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showed triphasic (adipocytes, smooth muscles, and blood vessels) morphology.

Case 3

A 52-year-old man presented with a 20-day history of abdominal distention. Vitals were stable. He appeared pale and moderately nourished. Abdominal and rectal examinations were unremarkable. Blood tests showed haemoglobin (Hb) levels of 10.5 g and a Total count of 10000 cells/cu. mm, Platelet 3.5 Lakhs, Creatinine 0.6, LFT (N). Abdominal ultrasonography revealed small bowel intussusception, likely in the jejunum. He had no comorbidities. CECT suggested jejunal lipoma with intussusception and small bowel obstruction, with a 2.5x2.1 cm lesion. Exploratory laparotomy and jejunum resection with jejunojejunal anastomosis were performed. The postoperative period was uneventful. The resected 2.5 cm mass showed triphasic morphology (adipocytes, smooth muscles, and blood vessels).

Case 4

A 39-year-old woman presented with 20 days of abdominal distention. Her vital signs were stable, and she appeared pale and moderately nourished. Abdominal and rectal examinations were unremarkable. Blood tests showed haemoglobin (Hb) levels of 9.5 g, with a total cell count of 9000 cells/cu. mm, platelets 2.5 Lakhs, creatinine 0.6, LFT (N). Abdominal ultrasonography indicated small bowel intussusception, probably in the ileum. The patient had no comorbidities. CECT suggested an ileal lipoma with ileoileal intussusception and small bowel obstruction, with a polypoidal lesion measuring 4.8x4.1 cm. An exploratory laparotomy was performed with ileal resection and ileoileal end-to-end anastomosis. Postoperative recovery was uneventful, except for two days of minimal purulent discharge. The resected 5 cm mass showed triphasic morphology (adipocytes, smooth muscles, and blood vessels).

Case 5

A 61-year-old man presented with 10 days of abdominal pain. His vitals were stable. He appeared pale and moderately nourished. Abdominal and rectal examinations were unremarkable. Blood tests showed haemoglobin (Hb) levels of 12.5 g and a total cell count of 8000 cells/cu. mm, Platelet 3.5 Lakhs, Creatinine 0.6, LFT(N). Ultrasound indicated small bowel intussusception, likely in the jejunum of the patient. He had no comorbidities. CECT suggested jejunal lipoma with intussusception and obstruction, with a 4.5x4.1 cm polypoidal lesion. Exploratory laparotomy and jejunal resection with end-to-end anastomosis were performed. The postoperative period was uneventful. The resected 4.5 cm mass showed triphasic cell morphology (adipocytes, smooth muscles, and blood vessels).

Case 6

A 33-year-old man presented with a 10-day history of abdominal distention. Vitals were stable. Examination showed that he was well-nourished, and abdominal and rectal examinations were unremarkable. Blood tests: Hb 13 g, total cell count 7000 cells/cu. mm, platelet 3.5 lakhs, creatinine 0.6, LFT normal. Ultrasound indicated small bowel intussusception, likely in the jejunum. CECT suggested jejunal lipoma with intussusception and obstruction, with a 2.9x2.7 cm polypoidal lesion present. Exploratory laparotomy and jejunal resection with end-to-end anastomosis were performed. Postoperative period was uneventful. The 3 cm mass that was resected showed triphasic cells (adipocytes, smooth muscles, and blood vessels).

Case 7

A 47-year-old woman presented with chronic lower abdominal pain for 10 days. Her vitals were stable. The patient was moderately nourished. Abdominal and rectal examinations were unremarkable. Blood tests: Hb 10 g, total cell count 8500 cells/cu. mm, Platelet 3.0 Lakhs, Creatinine 0.6, LFT(N). Ultrasound revealed distal small bowel intussusception and submucous lipoma. She had no comorbidities. CECT revealed an ileal lipoma with ileocolic intussusception and small bowel obstruction, a polypoidal lesion measuring 4.2x4.1 cm. Exploratory laparotomy and resection of the ileum with ileocolic anastomosis were performed. Post-intervention was uneventful. The 4.2 cm mass showed triphasic (adipocytes, smooth muscles, and blood vessels) morphology.

Case 8

A 55-year-old man had chronic lower abdominal pain for 15 days. His vitals were stable. He appeared pale and moderately nourished. Abdominal and rectal exams were unremarkable. Blood tests revealed haemoglobin (Hb) of 10.7 g and a Total count of 10000 cells/cu. mm, Platelet 3.6 Lakhs, Creatinine 0.6, LFT(N). Abdominal ultrasound revealed distal small bowel intussusception and submucous lipoma. The patient had no other comorbidities. Contrast-enhanced computed tomography (CECT) revealed an ileal lipoma with ileoileal intussusception and small bowel obstruction, measuring 3.8x3.5 cm polypoidal lesion. Exploratory laparotomy and ileal resection with ileoileal end-to-end anastomosis were performed. Post-intervention was uneventful. Histopathology revealed a well-circumscribed nodular lesion 3.9x3.5x2cm. The external surface was grey-brown and nodular. The cut surface was grey-yellow and grey-white with haemorrhagic areas and was firm. Microscopy revealed congestion and haemorrhage of the ileal mucosa. Submucosa had clusters of mature adipocytes, thick-walled vessels, smooth muscle bundles, and dense inflammatory infiltrate.

Case 9

A 24-year-old woman presented with a 7-day history of abdominal distention. Vitals were stable. She appeared pale and moderately well-nourished. Abdominal and rectal examinations were unremarkable. Blood tests showed haemoglobin (Hb) levels of 8.5 g and a total count of 10000 cells/cu. mm, platelets 3.5 Lakhs, creatinine 0.6, LFT(N). Ultrasound indicated small bowel intussusception,

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likely jejunum. The patient had no comorbidities. CECT suggested jejunal lipoma with intussusception and small bowel obstruction, a 2.0x2.1 cm polypoidal lesion. Exploratory laparotomy and jejunal resection with end-to-end anastomosis were performed. Post-intervention was uneventful. The 2 cm mass showed triphasic cell morphology (adipocytes, smooth muscles, and blood vessels).

Case 10

A 68-year-old man presented with 10-day abdominal pain. Vitals were stable. He appeared pale and moderately nourished. Abdominal and rectal examinations were unremarkable. Blood tests showed haemoglobin (Hb) levels of 9.5 g and a total cell count of 9000 cells/cu. mm, Platelet 2.5 Lakhs, Creatinine 0.6, LFT(N). Abdominal ultrasound indicated small bowel intussusception, likely of the ileum. He had no comorbidities. CECT suggested an ileal lipoma with ileoileal intussusception and small bowel obstruction, with a 5.0x4.8 cm polypoidal lesion. Exploratory laparotomy and ileal resection with ileoileal anastomosis were performed. The postoperative period was uneventful, except for 2 days of minimal purulent discharge. The resected 5 cm mass showed triphasic cell morphology (adipocytes, smooth muscle cells, and blood vessels).

Case 11

A 41-year-old female had chronic lower abdominal pain for 5 days. Her vitals were stable, and she was moderately nourished. Abdominal and rectal exams were unremarkable. Blood tests: Hb 10 g, total count 7500 cells/cu. mm, Platelet 4.0 Lakhs, Creatinine 0.6, LFT (N). Ultrasound showed distal small bowel intussusception and submucous lipoma. She had no comorbidities. CECT revealed an ileal lipoma with ileocolic intussusception and small bowel obstruction, with a 3.6x3.1 cm polypoidal lesion. Exploratory laparotomy and ileum resection with ileocolic anastomosis were performed. Post-intervention was uneventful. The 3.6 cm mass exhibited triphasic morphology (adipocytes, smooth muscles, and blood vessels).

Case 12

A 36-year-old man presented with 7 days of acute lower abdominal pain. His vital signs were stable, and he was moderately nourished. Abdominal examination showed umbilical tenderness, and the rectal examination was unremarkable. Blood tests reported haemoglobin (Hb) of 12 g and total cell count of 8000 cells/cu. mm, Platelet 2.0 Lakhs, Creatinine 0.6, LFT(N). Ultrasound revealed distal small bowel intussusception and submucous lipoma. No comorbidities. CECT revealed an ileal lipoma with ileocolic intussusception, small bowel obstruction, and a 4.1x3.5 cm polypoidal lesion. Exploratory laparotomy and ileum resection with ileocolic anastomosis were performed. Post-intervention was uneventful. Bowel resection revealed a 4 cm mass with triphasic morphology (adipocytes, smooth muscles, and blood vessels).

DISCUSSION

Adult intussusception is rare, accounting for 1-5% of intestinal obstructions, usually with an identifiable pathological lead point.[1] In our series of 12 patients with intussusception due to intestinal AML, ages ranged from 24 to 68, with a slight male predominance, differing from previous reports, which mostly affect females. Yan et al. reported PEComa tumors in middle-aged women, while Yokota et al. described a 40-year-old woman with colonic PEComa-associated intussusception.[9,10]

Ultrasonography and CECT showed enteroenteric, ileoileal, ileocolic, or jejunojejunal intussusception with intraluminal lesions of 2-5 cm, often misinterpreted as lipomas. Yan et al. noted PEComa tumors lack specific radiological traits, while Qian et al. described them as broad-based or polypoid masses mimicking other tumors.[9,11] Yokota et al. reported a 34 mm ileocecal mass initially considered as a lipoma, GIST, or leiomyoma.[10] AML's heterogeneous composition of AML makes the diagnosis difficult. Adipose tissue resembles lipoma, spindle-cell proliferation mimics leiomyoma, and enhancing soft tissue components simulate GIST. CT imaging can identify intussusception and lead points but is insufficient for a definitive preoperative diagnosis.

Most of our patients had chronic or intermittent abdominal pain and distension lasting 5-24 days, and only one patient had acute pain. Similar nonspecific symptoms were noted by Moges et al.[12] The predominance of vague symptoms suggests intermittent bowel telescoping and delayed vascular compromise in our patient. These findings highlight the need for suspicion of pathological lead points in adults with recurrent obstructive symptoms.

The ileum was the most commonly involved site in our series, the intussusception patterns included ileoileal, ileocolic, and jejunojejunal involvement. Yan et al. and Bagheri et al. found the colon to be the most common site for PEComa tumors.[9,13] The predominance of small bowel involvement in our series may reflect the greater mobility of the small intestine, making these lesions more likely to serve as lead points for intussusception.

All lesions showed AML's triphasic morphology of mature adipose tissue, thick-walled blood vessels, and smooth muscle bundles, confirming the diagnosis of classic angiomyolipoma. Yan et al., Qian et al., and Yokota et al. also reported positivity for HMB-45, Melan-A, and smooth muscle actin.[9-11] HMB-45 positivity supports melanocytic differentiation, distinguishing AML from similar tumours, such as GIST, leiomyoma, lipoma, and liposarcoma. Histopathological evaluation confirmed the diagnosis in all cases. Although immunohistochemistry has been reported as useful in previous studies, it was not performed in the present series.

All patients underwent exploratory laparotomy with bowel resection and primary anastomosis, resulting

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in favourable outcomes with no mortality and minor complications. Similar outcomes have been reported by Moges et al. and Yokota et al.[12,10] Surgical resection is preferred for adult intussusception because of the risk of neoplasia, ischaemia, and malignancy. Given the rarity of intestinal angiomyolipoma, further case accumulation and multicentre studies are needed to better define its diagnostic characteristics, management strategies, and long-term outcomes.

CONCLUSION

Intestinal angiomyolipoma is a rare cause of adult intussusception that presents diagnostic challenges due to its nonspecific clinical and radiological features. Most patients experience intermittent abdominal pain and bowel obstruction, with imaging often suggesting lipoma or other tumours. Diagnosis depended on postoperative histopathological identification of the triphasic morphology of angiomyolipoma. Surgical resection with primary anastomosis yielded favourable results. Awareness is essential for the early recognition and management of adult intussusception with pathological lead points.

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