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Case Report

An unusual cause of lower gastrointestinal bleed: angiomyolipomas, in an unusual location of the colon

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ABSTRACT

Angiomyolipomas are benign tumors derived from mesenchymal tissue. Majority of angiomyolipomas arise in the kidney. Extra renal angiomyolipomas are extremely rare and reported in the liver, nasal cavity, vagina, spermatic cord, skin, mediastinum and GI tract. Colonic involvement is very rare. Here we report the case of a 32 year old male patient who presented in the emergency department with complaints of Bleeding per rectum due to angiomyolipoma of ascending colon. The clinical presentation, operative procedure, pathological features and literature review are presented.

Keywords: Angiomyolipoma, Colon, HMB-45

INTRODUCTION

Angiomyolipomas are a form of mesenchymal hamartoma, composed of blood vessels, smooth muscle cells, and mature fat cells. The great majority of angiomyolipomas arises in the kidney and shows a female preponderance. Extrarenal angiomyolipomas are extremely rare and reported in the liver, nasal cavity, vagina, spermatic cord, skin, mediastinum and GI tract.[1] Colonic angiomyolipomas are rare benign tumors of the large intestine with particular histopathologic characteristics. Most of them were reported to be located in the left side of colon. Preoperative diagnosis is usually difficult and surgical excision is required. Inadequate resection may result in rapid local recurrence.

CASE REPORT

A 32 year old male patient presented in emergency department with 3 weeks complaints of Bleeding per rectum and pain abdomen. The pain was located over the right side of the abdomen with cramping characteristics. He had been well before with no underlying disease such as hypertension, renal, hepatic, or cardiac disease.

Patient was not an alcoholic or smoker. Physical examination showed that the patient was anemic, with hypotension. Abdominal examination revealed a vague tender mass in the right iliac fossa. Bowel sounds were normal. Digital rectal examination revealed blood staining of finger. Patient was resuscitated. Emergency Upper GI endoscopy was normal. Emergency USG abdomen revealed complex mass in the right iliac fossa. Blood investigations showed Hb 7 g/dl, WBC 15,000/cumm of which 60 % were polymorphs. His serum electrolytes, renal function tests and liver function tests were normal. Mean-while in the hospital he had another episode of hematochezia.

So further investigations were not done and the patient was taken up for emergency surgery after getting high-risk informed consent. Emergency exploratory

laparotomy was done and the abdomen was opened by a midline incision. Findings were multiple haemangioma looking lesions involving the terminal ileum, ileocaecal junction, caecum, ascending colon and it is mesentery (Figure 1 and 2). Remaining bowel was normal. There was no free fluid. Liver and other solid viscera were normal. Right hemicolectomy with ileo transverse anastomosis was done. The ileo transverse anastomosis was done in two layers in an end-to-end fashion. Postoperative recovery was uneventful except for wound infection which was treated by appropriate antibiotics, dressing and secondary suturing. The patient was discharged 20 days post-surgery. He was followed up in the outpatient department and had no tumor recurrence till now. Histology revealed an angiomyolipoma involving the Terminal ileum, ileocaecal junction, caecum, up to the whole of the ascending colon (Figure 3). Immunohistochemistry staining showed the proliferating smooth muscle cells were positive for HMB -45, desmin, vimentin, and smooth muscle actin .

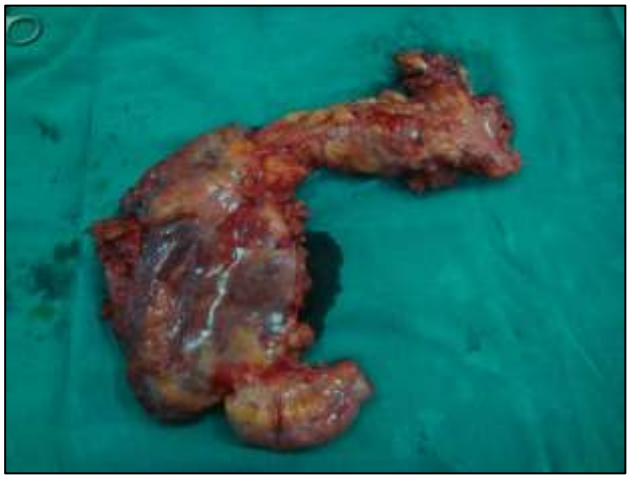

Figure 1: Gross appearance of the specimen.

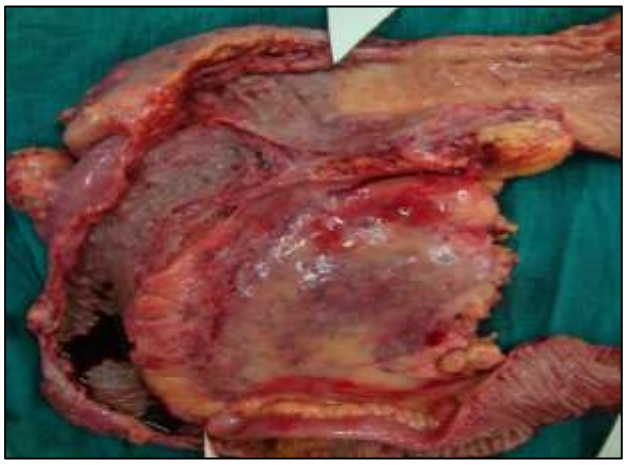

Figure 2: Cut opened colon by angiomyolipoma specimen showing the involvement of term: ileum, caecum and ascending colon.

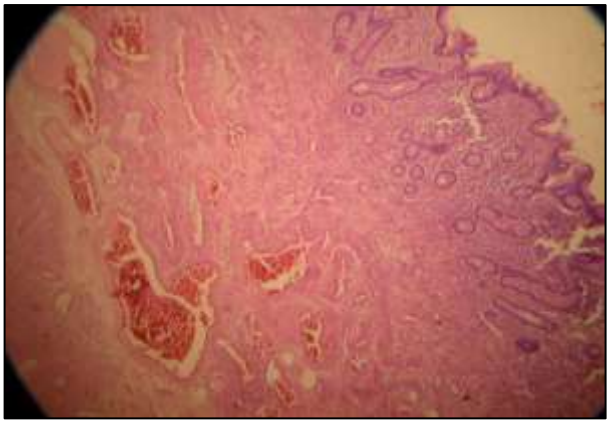

Figure 3: Microscopic examination of the specimen of the tumor consisting of three components: mature fat cells, blood vessels, and smooth muscle cells.

DISCUSSION

Angiomyolipomas are histologically benign tumors derived from mesenchymal tissue. They occur sporadically or as a part of tuberous sclerosis complex in 45 - 80 %of cases.

Tuberous sclerosis, a multisystemic disease with autosomal dominant inheritance, with occasional association with the triad of epilepsy, mental retardation, and adenoma sebaceum. The great majority of angiomyolipomas arise in the kidney. Extrarenal presentations are very rare and are most commonly found in the liver.[1] Colonic involvement is rare. Most of the colonic angiomyolipomas present as small mucosal polyps, but this case presented as a large lesion.

Preoperative diagnosis is difficult. Combination of CT, ultrasound and MRI increases the diagnostic rate to 60%. Surgical excision is the treatment of choice. The recurrence rate is high in cases of inadequate resection, but when the tumor can be removed completely, the prognosis is excellent, with a nearly 100 % cure rate. Grossly, they are grayish-yellow with a lobulated appearance and vary in size. Microscopically, angiomyolipomas are composed of three components smooth muscles, adipose tissues and blood vessels. The contents of these components varied in proportion, especially in adipose tissue, and range from less than 10 percent to more than 50 percent of the tumor.[2] Immunohistochemically, these tumors usually are cytokeratin and CD-34 negative and are positive for HMB 45, vimentin, smooth muscle actin and desmin.

Only few cases of colonic angiomyolipomas have been reported in the literature.[1-8] All cases occurred in middle-aged men, and most were polypoid or pedunculated. All were restricted to the colon and left sided. Most of them were removed endoscopically, rarely surgery was indicated.

Probably this may be the first reported case of angiomyolipoma of the Ascending colon involving the ileocaecal junction. Moreover this case differs from other reported cases in having a varied presentation (emergency presentation as bleeding per rectum) and having diffuse colonic involvement.

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