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Case Report

JEJUNOJEJUNAL INTUSSUSCEPTION BY A SARCOMATOID CARCINOMA OF JEJUNUM – A RARE CASE REPORT

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ABSTRACT

Background: Jejunojejunal intussusceptions are not common in adults and it usually has a pathological lead point. The lead point being a sarcomatoid carcinoma is extremely rare.
Case presentation: We present a case of a sarcomatoid carcinoma of the jejunum in a 49-year-old male patient with features suggestive of small bowel obstruction. Radiological imaging revealed jejunojejunal intussusception with left sided pleural effusion and expansile lytic lesion in left sacral vertebrae. The patient underwent surgical resection of the jejunum and pathology revealed jejunal sarcomatoid carcinoma. He died 5 weeks after surgery.
Conclusion: Sarcomatoid carcinoma of the jejunum has a worse prognosis. Treatment modality remains a challenge but surgery is still considered optimal treatment.

INTRODUCTION

Intussusception is the telescoping of proximal segment of gastrointestinal (GI) tract into the adjacent distal segment(1). Adult intussusception constitute only 5% of all intussusception cases and majority of cases have pathological lead point in the form of neoplasm, polyp, Meckel's diverticulum(2). Due to the associated risk of malignancy, about 70 to 90% cases require surgical treatment (3).

Sarcomatoid carcinoma is a very rare biphasic tumorwith both mesenchymal and carcinomatoid component (4,5). It has been described in various organs like salivary gland, thyroid, breast, respiratory and gastrointestinaltract(6). The clinical presentation of sarcomatoid carcinoma of gastrointestinal tract includes abdominal pain, obstructive symptoms, anemia and GI bleeding (7,8). Literature review showed that only 18 cases of sarcomatoid carcinoma of the jejunum have been reported(4).Surgical treatment remains mainstay therapeutic approach, as neither chemotherapy nor radiotherapy has shownany improvementin survival (8,9). The tumor hasworse prognosis compared to other small intestinal tumorsdue to its metastatic potential and aggressive course (10).

We report the case of a 49-year-old male patient withjejunojejunal intussusception caused by sarcomatoid carcinoma of the jejunum.

CASE REPORT

A 49-year-old man presented with abdominal pain and vomiting of five days duration. He also complained of constipation for two days. One year back, he had been operated for carcinoma of left side of tongue and completed 26 cycles of radiotherapy. He complained of left lower limb pain and inability to walk, three months after completing radiotherapy.

General physical examination of the patient was inconspicuous. Abdominal examination revealed abdominal distention, tenderness and exaggerated bowel sounds. Laboratory parameters revealed an elevated total leukocyte count and raised ESR. Abdominal Xray showed dilated small bowel loops. Chest X- ray showed left sided pleural effusion. For further assessment, an abdominal and chest computed tomography (CT) was done which showed short segment of jejunojejunal intussusception, left sidedpleural effusion and an expansile lytic lesion at left sacral ala extending upto third sacral vertebra (Figure 1,2).

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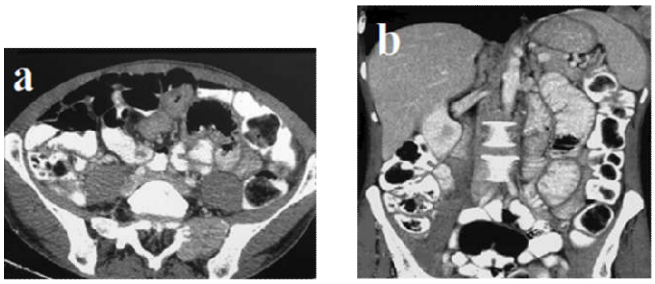

Figure 1 Abdominal CT scan showing a short segment jejunojejunal intussusception (a).(b).

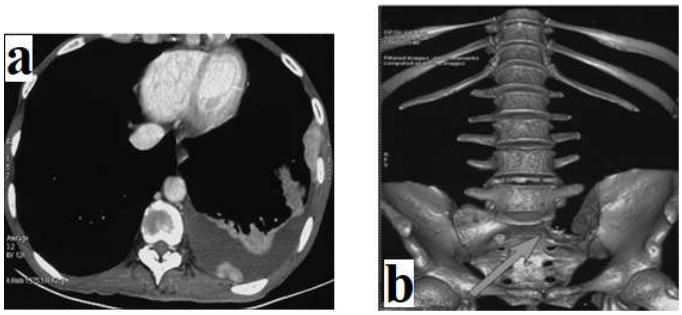

Figure 2 Chest CT scan showing left sided pleural effusion (a); 3D CT reconstruction reveals an expansile lytic lesion at sacral ala extending tothird sacral vertebra (b).

The patient was diagnosed with small bowel obstruction due to jejunojejunal intussusception and exploratory laparotomy was performed. During operation, ajejunojejunal intussusception 20 cm from DJ flexure was identified. The lead point of intussusceptum being a mass 3.8 x 4 cm with induration over anterior wall (Figure3). Jejunal resection and anastomosis was performed. Unfortunately, the patient died 5 weeks after surgery.

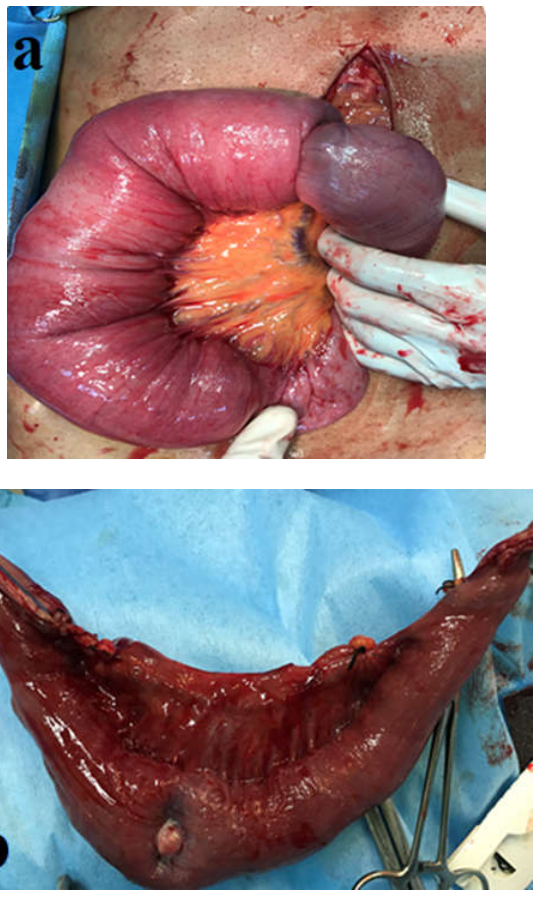

Figure 3 (a) Jejunojejunal intussusception 20 cm from DJ flexure; (b) Tumor over anterior wall of jejunum.

Pathological examination revealed the tumor with a maximum diameter of 4 cm and the tumor had invaded from the mucosa upto the serosa. Both the resected margin were free of neoplasm. Histologically, the tumor was characterized by atypical spindle-shaped cells. Immunohistochemically, the tumor cells were diffusely positive for vimentin and focally positive for cytokeratin- AE1/AE3. On other hand, the tumor cells were negative for desmin, CD 117 and CD34. Our final diagnosis was sarcomatoid carcinoma of jejunum with bony and pleural metastases.

DISCUSSION

Malignancies of small intestine are rare compared to other gastrointestinal tract malignancies and comprises less than 5% of all GI cancers(7). Majority of small intestinal tumors include adenocarcinoma, neuroendocrine tumors, stromal tumors and lymphomas(10). Sarcomatoid carcinoma of small intestine is an extremely rare type having both carcinomatous and sarcomatous features(11).Small intestinal sarcomatoid carcinoma primarily occurs in ileum, followed by jejunum and duodenum (4,12). In 1973, “Dikman and Toker described this tumor and named it enteroblastoma”(13). It normally occurs in elderly patient with a male to female ratio of 1.5:1(7,14). Patients may present with abdominal pain, intestinal obstruction, abdominal mass, GI bleeding and anaemia (15). The present case report is the first of its kind to describe sarcomatoid carcinoma presenting as intussusception.

Histologically, sarcomatoid carcinoma typically exhibits a biphasic pattern of epithelial and mesenchymal cells (16). The present case was predominantly composed of atypical spindle cells. Sarcomatoid carcinomas usually exhibit positivity for cytokeratin and vimentin(14), as was found in this case. Other intestinal spindle cell tumors express positivity for CD34 , CD117and desmin, while these tumor markers are negative in intestinal sarcomatoid carcinomas (17).

There is a limited data regarding most appropriate treatment but surgery is considered as first line of treatment,as there is poor response to chemotherapy and radiotherapy(10).The tumor itself is rapidly progressive and high metastatic potential and hence, patients usually present in an advanced stage at the time of diagnosis(5).Reid-Nicholson *et al* reportedthat “70% of the patients died within 2 months to 3-yearfollow-up”(14).

CONCLUSION

Sarcomatoid carcinoma of jejunum is a very rare malignancy with high mortality. Treatment modality remains a challenge but surgery still holds superior. As the tumor presents at an advanced stage, early diagnosis and surgical resection would have better prognosis on patient’s survival. The present case adds more insight on this rarer cancer entity. It also emphasizes the need of considering sarcomatoid carcinoma as a differential diagnosis in a rapidly progressing small intestinal tumor.

Abbreviations

CT: Computed tomography, CD: Cluster of differentiation, DJ: Duodenojejunal flexure, ESR: Erythrocyte sedimentation rate, GI: gastrointestinal

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Ethics approval: It was not required for the imaging, diagnosis and study of the case, in our institution.

Consent: Informed consent was obtained from the patient.

Conflicts of interest: None

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