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Inflammatory Myofibroblastic Tumor Presenting as Colorectal Intussusception: A Case Report

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Abstract

Inflammatory Myofibroblastic Tumors (IMTs) are characterized by proliferative myofibroblastic spindle cells accompanying inflammatory infiltration. This case report presents a rare case of inflammatory myofibroblastic tumor presenting with colorectal intussusception in a 22-year-old female. Despite ALK-1 (Activin receptor-Like Kinase-1) negativity in the tumor, SMA (Smooth Muscle Actin) and vimentin positivity were observed. The tumor was surgically excised, and no recurrence was observed during the 6-month follow-up. Favorable outcomes can be achieved with early diagnosis and optimal treatment. No cases of IMT presenting with colorectal intussusception have been reported in the literature, making our case a rare occurrence.

Keywords: Inflammatory myofibroblastic tumor; Colorectal invagination; ALK-1

Introduction

Inflammatory Myofibroblastic Tumors (IMTs) are characterized by proliferative myofibroblastic spindle cells accompanying inflammatory infiltration [1]. IMTs are rare tumors that can occur at any age, although they are most commonly seen in children and young adults [2]. They are most frequently localized in the lungs, followed by extrapulmonary sites such as the liver, pancreas, intestines, and bones. Intestinal involvement by IMT is relatively rare, and invagination caused by IMT is even rarer [3]. Clinically, radiologically, and histologically, they resemble malignant neoplasms. Although the pathogenesis involves infectious, autoimmune, reactive, and neoplastic processes, the etiology remains unclear [4]. Herein, we present a case of inflammatory myofibroblastic tumor diagnosed in a 22-year-old female presenting with abdominal pain and nausea/vomiting resulting in colorectal intussusception.

Case Presentation

A 22-year-old female patient presented to the emergency department with complaints of constipation and abdominal bloating for a month, along with abdominal pain, nausea, vomiting, and inability to pass gas or stool for the past 3 days. The patient had no known medical history. Physical examination revealed distended abdomen with tenderness but no rebound tenderness. Rectal examination revealed an empty rectum ampulla. Abdominal X-ray showed colonic-type air-fluid levels. Abdominal CT (Computed Tomography) scan with intravenous contrast revealed invagination of the sigmoid colon into the rectum (Figure 1).

Rectosigmoidoscopy revealed a mass lesion at the mid-upper rectum location obliterating the lumen, with smooth borders and mobilizable with manipulation.

Laparoscopic exploration revealed invagination of the distal sigmoid colon with its mesentery into the rectum, with the mass being the lead point. The patient underwent laparoscopic segmental sigmoid colon resection with colorectal anastomosis (Figure 2).

Final pathology revealed a tumor with widespread mixed lymphoplasmacytic cell infiltration involving the mucosa, lamina propria, muscularis propria, and subserosa, characterized by vesicular nuclei, prominent cytoplasm, spindle-shaped cells, and absence of significant atypia and mitosis. Perineural tumor spread was observed, but lymphovascular invasion and tumor necrosis were absent. The tumor measured 5 cm × 4 cm × 3.5 cm, and no macroscopic tumor perforation was seen. Twenty-two reactive lymph nodes were found in the mesentery of the resected colon segment. Immunohistochemical examination showed positive staining for SMA and vimentin, while desmin, c-kit, CD 34, S100, DOG1 (Delay of Germination 1), beta-catenin, IgG, IgG4, and

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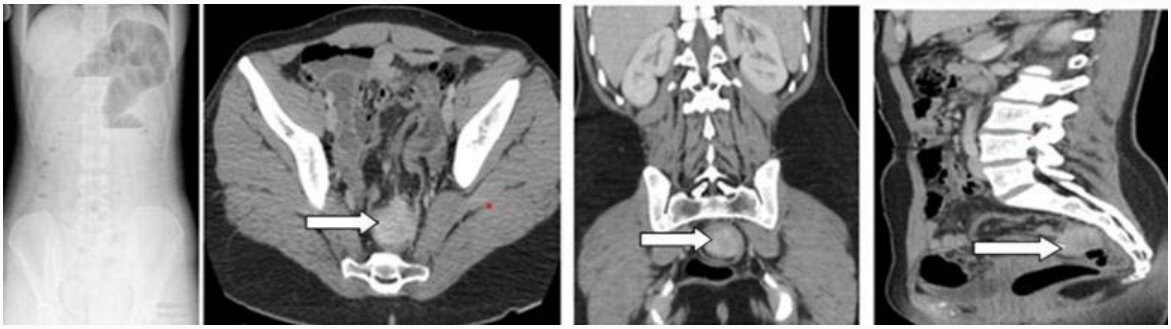

Figure 1: Standing direct abdominal radiography and computed tomography section.

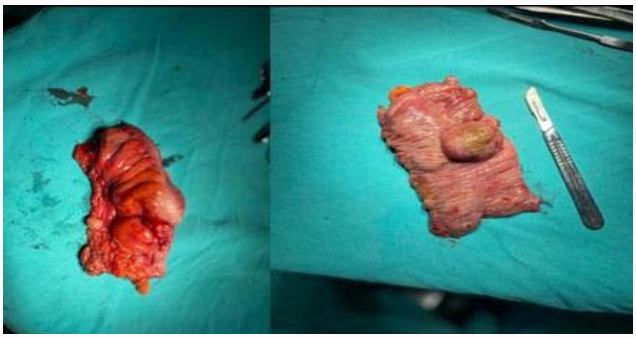

Figure 2: Macroscopic view of the pathology.

ALK were negative. Based on the histopathological findings and immunohistochemical examinations, the tumor was considered consistent with inflammatory myofibroblastic tumor.

During 6-month follow-up, no evidence of recurrence or metastasis was observed.

Discussion

Inflammatory myofibroblastic tumors are rare benign tumors that can present with varied clinical manifestations depending on the organ involved, including the lungs, liver, pancreas, bones, and oral cavity [5]. Intestinal involvement by IMT is relatively rare, and IMT-related intussusception is even rarer [3]. Causes of intussusception in adults often include adhesions, polyps, inflammatory bowel disease, gastrointestinal tumors, and leiomyosarcoma [6]. Intussusception due to inflammatory myofibroblastic tumor is extremely rare. Our case presented with ileus secondary to intussusception, manifesting as abdominal pain, nausea, vomiting, and inability to pass gas or stool. Histopathological examination and immunohistochemical studies are essential for the diagnosis of IMTs [7]. ALK translocation and ALK positivity are observed in approximately 50% of cases, especially in young patients [8-10]. However, ALK negativity does not exclude the diagnosis of IMT, as it was in our case. Vimentin and SMA are generally strongly positive, while desmin, S-100, and CD117 are usually negative [7,8,10]. Our case showed parallel results with SMA and vimentin positivity and desmin, S-100, and CD117 negativity.

Conclusion

This case report presents a rare case of inflammatory myofibroblastic tumor presenting with colorectal intussusception in a young female. Although ALK-1 was negative in spindle-shaped cells in our case, SMA and vimentin positivity were observed. The lesion was surgically excised, and no recurrence was observed during the 6-month follow-up. Early diagnosis and optimal treatment can lead to favorable outcomes in these tumors through diagnostic evaluation and a multidisciplinary approach. To our knowledge, no cases of IMT presenting with colorectal intussusception have been reported in the literature, making our case unique.

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