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Inflammatory Myofibroblastic Tumor of The Colon Mimicking Inflammatory Bowel Disease, Followed by True Crohn's Disease: A Diagnostic and Therapeutic Challenge

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Abstract **Case Report**

Background: Inflammatory myofibroblastic tumor is a rare mesenchymal neoplasm of intermediate biological potential that most commonly affects the lung and pediatric population. Gastrointestinal involvement is uncommon, particularly in the colon. The association between IMT and inflammatory bowel disease, especially Crohn's disease, is exceedingly rare and poses significant diagnostic challenges due to overlapping clinical and radiological features. ***Case Presentation:*** We report the case of a 38-year-old man presenting with chronic diarrhea, weight loss, and obstructive symptoms. Imaging and colonoscopy revealed a stenosing cecal mass with terminal ileal involvement, initially suggestive of Crohn's disease or intestinal tuberculosis. Surgical resection was performed, and histopathological examination confirmed an inflammatory myofibroblastic tumor with clear margins. The postoperative course was favorable, with complete symptom resolution. Three years later, the patient developed recurrent diarrhea, and further evaluation revealed chronic active colitis with non-caseating granulomas, leading to a definitive diagnosis of Crohn's disease. Treatment with azathioprine resulted in sustained clinical and endoscopic remission. ***Conclusion:*** This case illustrates a rare and unusual temporal association between colonic IMT and Crohn's disease, with IMT preceding the diagnosis of inflammatory bowel disease. It underscores the importance of considering rare benign tumors in the differential diagnosis of mass-forming colonic lesions and highlights the need for long-term surveillance in patients with atypical inflammatory presentations. Improved awareness of this rare association may help prevent diagnostic delays and inappropriate therapeutic strategies.

Keywords: Inflammatory myofibroblastic tumor, Crohn's disease, Colonic neoplasm, Differential diagnosis, Chronic diarrhea, Surgical resection.

INTRODUCTION

Inflammatory myofibroblastic tumor (IMT) is a rare mesenchymal neoplasm characterized by spindle-shaped myofibroblastic cells accompanied by a prominent inflammatory infiltrate composed mainly of plasma cells and lymphocytes. Although IMT was initially described as a pulmonary lesion in children and young adults, extrapulmonary involvement has since been increasingly recognized, including rare occurrences within the gastrointestinal tract [1].

Gastrointestinal IMTs are uncommon, and colonic localization is particularly rare. When present, these tumors frequently manifest as mass-forming or stenosing lesions associated with nonspecific symptoms such as abdominal pain, diarrhea, or bowel obstruction, often mimicking inflammatory bowel disease or colorectal malignancy on imaging and endoscopy [1]. As a result, definitive diagnosis relies on histopathological examination supported by immunohistochemistry following surgical resection.

Crohn's disease is a chronic inflammatory bowel disease characterized by transmural inflammation, granuloma formation, and a relapsing–remitting clinical course, frequently involving the terminal ileum and colon. Chronic intestinal inflammation in Crohn's

disease is associated with activation and dysregulation of intestinal myofibroblasts, contributing to fibrosis and stricture formation [2].

The coexistence of IMT and inflammatory bowel disease is exceptionally rare and has been documented only in isolated case reports. Most published cases describe IMT arising in the context of long-standing inflammatory bowel disease, supporting the hypothesis that chronic inflammation may promote abnormal myofibroblastic proliferation. In contrast, IMT preceding the diagnosis of Crohn's disease has been only exceptionally reported.

We present a rare case of colonic inflammatory myofibroblastic tumor initially mimicking Crohn's disease, followed by the subsequent development of confirmed CD several years after curative surgical resection, highlighting the diagnostic complexity and the importance of long-term follow-up in atypical inflammatory presentations.

Inflammatory Myofibroblastic Tumor (IMT), previously called inflammatory pseudotumor and plasma cell granuloma, belongs to a class of rare spindle cell lesions showing a rather unpredictable biological behavior with occasional tendency toward invasion to the surrounding tissue and local recurrence. The lesion, as primarily described by Bahadori and Liebow in 1973, is a reactive/inflammatory process in the pulmonary system mostly occurring in children and young adults. Since then, many cases have been reported in older patients in addition to extra-pulmonary sites [11].

Case Report

We report the case of a 38-year-old man with no significant past medical history, who presented with chronic watery diarrhea (3–4 stools/day, diurnal and nocturnal) associated with weight loss and Koenig's syndrome evolving for a month since his admission.

The patient had a colonoscopy showed an infiltrative stenosing caecal lesion and abdominal CT-scan revealed a caecal mass with ileal wall thickening with marked infiltration of the adjacent mesenteric fat, containing multiple infra- and juxtacentimetric lymph nodes. Histology suggested chronic inflammatory colitis with poorly defined granulomatous features, not excluding Crohn's disease or intestinal tuberculosis. Tuberculosis GeneXpert was negative.

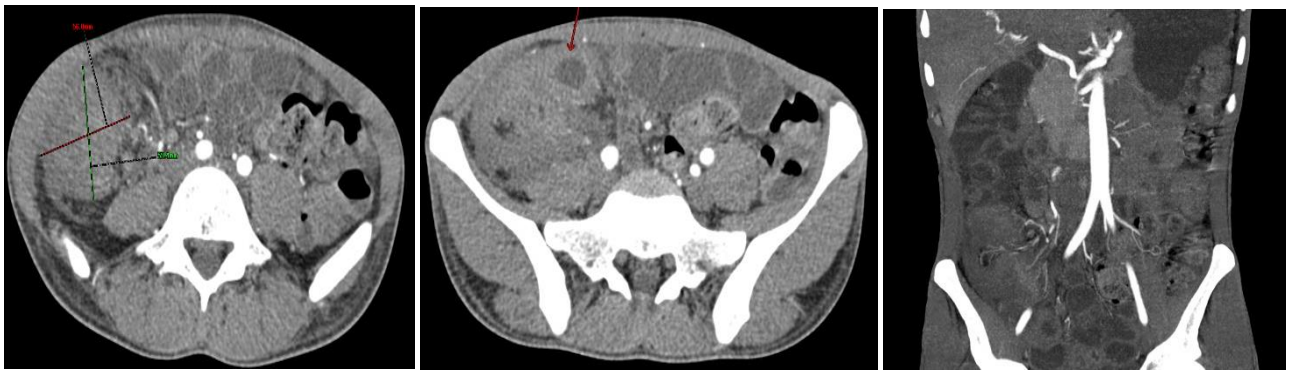

Figure 1: Abdominal CT-scan of the patient showing a cecal mass measuring 56 × 46 × 59.4 mm, relatively well circumscribed, demonstrating heterogeneous enhancement following intravenous contrast administration with marked infiltration of the adjacent mesenteric fat, containing multiple infra- and juxtacentimetric lymph nodes. With an irregular circumferential thickening of the terminal ileum, with a maximum wall thickness of 7 mm, showing a target-like enhancement pattern after contrast injection

The patient underwent a right hemicolectomy in May 2019. Histopathological examination concluded to an inflammatory myofibroblastic tumor, it was composed of a proliferation of spindle-shaped cells arranged in the hyaline material with chronic inflammatory cells, composed mainly of plasma cells and lymphocytes. Immunohistochemically, tumor cells were positive for smooth muscle actin, and vimentin, and negative for desmin, CD117 (c-kit), anaplastic lymphoma kinase-1with clear margins and no lymph node involvement, supported by immunohistochemistry.

Postoperative outcome was favorable, with complete resolution of symptoms.

3 years later, diarrhea recurred. Abdominal CT-scan showed segmental colonic wall thickening, and colonoscopy revealed continuous left-sided colitis extending from the rectum to the splenic flexure, with a sessile rectal polyp. Histology demonstrated chronic active colitis with non-caseating epithelioid granulomas, and focal crypt architectural distortion with areas of preserved mucosa without malignancy. Upper endoscopy showed mild chronic gastritis with rare *Helicobacter pylori* and no duodenal involvement.

The diagnosis of Crohn's disease was retained and the patient was started on azathioprine at a dose of 2.5 mg/kg/day, with good clinical, biological, and endoscopic response during follow-up.

DISCUSSION

This case highlights the rare association of IMT and IBD. Chronic diarrhea is a frequent but nonspecific symptom that may reflect a wide spectrum of diseases, ranging from infectious and functional disorders to inflammatory and neoplastic conditions. In this context, distinguishing Crohn's disease from rare benign tumors such as IMT remains a significant clinical challenge.

CD is a chronic inflammatory bowel disease characterized by transmural inflammation, segmental involvement, and a relapsing–remitting course. Typical manifestations include diarrhea, abdominal pain, weight loss, and systemic inflammatory features, while complications such as strictures, fistulas, and abscesses may develop over time [1]. However, as illustrated in this case, the initial presentation of CD can overlap and probably be masked by other gastrointestinal disorders, particularly when radiological and endoscopic findings are inconclusive or misleading.

IMTs are rare benign mesenchymal tumors composed of spindle-shaped myofibroblasts associated with a prominent inflammatory infiltrate. Although they are more commonly described in pediatric populations and pulmonary locations, gastrointestinal involvement—particularly colonic or rectal localization—has been increasingly reported [2–4]. IMTs may present with nonspecific symptoms such as abdominal pain, diarrhea, bleeding, or bowel obstruction, closely resembling inflammatory bowel disease. Several case reports have documented IMTs masquerading as CD, leading to diagnostic delays or inappropriate medical therapy [3–5].

The diagnostic approach to chronic diarrhea requires a stepwise and multimodal strategy integrating clinical evaluation, laboratory testing, imaging, endoscopy, and histopathological assessment. Cross-sectional imaging techniques, including computed tomography and magnetic resonance enterography, play a supportive role by identifying transmural bowel wall thickening, skip lesions, or mass-like abnormalities. However, imaging findings alone lack specificity and must be interpreted in conjunction with endoscopic and histological data. Ileocolonoscopy with targeted biopsies remains central to diagnosis, allowing direct visualization of mucosal lesions and histopathological confirmation. The presence of transmural inflammation and non-caseating granulomas strongly supports a diagnosis of Crohn's disease, although granulomas may be absent in many cases and can occasionally be mimicked by other inflammatory conditions [6].

Accurate differentiation between Crohn's disease and IMT has major therapeutic implications. CD typically requires long-term immunosuppressive or biologic therapy, whereas IMTs are generally managed with complete surgical excision, which is often curative and does not necessitate prolonged immunomodulatory treatment [4,7]. Misdiagnosis may therefore expose patients either to unnecessary surgery or to prolonged immunosuppression with potential adverse effects. This underscores the importance of maintaining a broad differential diagnosis in patients presenting with atypical features or refractory disease courses.

Management strategies for CD are tailored according to disease severity, location, behavior, and patient-specific factors. Corticosteroids remain effective for induction of remission but are unsuitable for long-term use due to their well-documented adverse effects, including metabolic disturbances and osteoporosis [8]. Immunomodulators such as azathioprine are frequently employed as steroid-sparing agents for maintenance therapy and have demonstrated efficacy in reducing relapse rates. In recent years, biologic therapies targeting tumor necrosis factor and other inflammatory pathways have significantly improved outcomes, promoting mucosal healing and sustained remission in many patients with moderate-to-severe disease [9].

Despite advances in medical therapy, surgical intervention remains unavoidable in a substantial proportion of patients with Crohn's disease, particularly those with stricturing or penetrating phenotypes. In the present case, progressive ileal strictures refractory to medical management necessitated segmental bowel resection. Surgical intervention in CD aims to manage complications rather than cure the disease, as postoperative recurrence is common. Consequently, surgery must be integrated into a comprehensive long-term management plan that includes postoperative surveillance and appropriate medical therapy to reduce recurrence risk [10].

Long-term management of CD extends beyond disease control to encompass nutritional status, psychosocial well-being, and quality of life. Chronic symptoms, repeated hospitalizations, and the unpredictable nature of disease flares substantially impact mental health and social functioning. Increasing evidence highlights the high prevalence of anxiety, depression, and psychosocial distress among patients with inflammatory bowel disease, emphasizing the need for multidisciplinary care models that incorporate psychological support [5]. In this patient, sustained remission following combined medical and surgical management translated into a meaningful improvement in quality of life, although continued vigilance for disease recurrence remained essential.

This case also underscores the importance of longitudinal assessment in chronic inflammatory

diseases. The evolution of symptoms, therapeutic responses, and complication profiles over time provides valuable insight into disease behavior and informs clinical decision-making. Personalized treatment strategies, guided by clinical, radiological, endoscopic, and histological findings, are essential to optimizing long-term outcomes in Crohn's disease [7].

CONCLUSION

In conclusion, this case illustrates the diagnostic complexity of chronic diarrhea and the necessity of distinguishing Crohn's disease from rare mimickers such as inflammatory myofibroblastic tumors. A comprehensive diagnostic approach and a multidisciplinary management strategy are essential to avoid misdiagnosis and inappropriate therapy. Integrating medical, surgical, nutritional, and psychosocial interventions remains fundamental to improving long-term outcomes in patients with Crohn's disease.

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