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Unusual Presentation of Sarcoidosis as Partial Small Bowel Obstruction (SBO) Secondary to Jejunal Stricture, with Peritoneal Involvement.

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Abstract

Sarcoidosis involving the small bowel is extremely rare. We present an unusual case of gastrointestinal sarcoidosis in a 29 years old male presenting as partial small bowel obstruction secondary to a jejunal stricture. Diagnosis was confirmed by histopathology through diagnostic laparoscopy, and clinical improvement was noted with corticosteroids treatment

Introduction

Sarcoidosis is a multisystem inflammatory disease that is characterized by the presence of noncaseating granulomas on histopathology. Pulmonary sarcoidosis is the most common manifestation. Gastrointestinal (GI) sarcoidosis itself is rare with a reported prevalence of less than 1%, with the stomach being the most frequently affected organ.[1]

Case Summary

A 29-year-old African American male with no prior medical history presented with intermittent right lower quadrant abdominal and flank pain for one-year duration associated with weight loss, intermittent nausea, and vomiting. No change in bowel movements. No reported shortness of breath, cough, or fever. Physical exam was significant for mild tenderness to palpation of the right flank, otherwise remainder of exam was unremarkable. Initial laboratory work was pertinent for haemoglobin level of 11.6 g/dl, platelets of 510 k/ul, WBC of 6 k/ul, erythrocyte sedimentation rate of 130, and C reactive protein of 69.

Initial chest x-ray (CXR) showed a trace right pleural effusion. Abdominal x-ray showed small bowel stricture without evidence of obstruction. Computed tomography (CT) of abdomen and pelvis with intravenous contrast showed a possible SBO, along with mesenteric and inguinal lymphadenopathy. This was followed by a small bowel series which showed a stricture likely located in the jejunum. An upper endoscopy was performed that only revealed evidence of gastritis; however, the small bowel stricture was not reached. A colonoscopy showed no pathology. CT guided left inguinal biopsy was performed, results showed no abnormal clonal B cells or atypical T cell population. During his hospital stay, patient developed an enlarging right sided pleural effusion. A thoracentesis was performed, pleural fluid analysis showed lymphocyte predominant exudate, negative bacterial culture and AFB smear, ADA level of 23, and flow cytometry was negative for clonal lymphoid expansion. Repeat CT chest and abdomen after one week showed reaccumulating moderate right sided pleural effusion, ascites not present on prior examination, along with peritoneal enhancement and mild nodularity of the peritoneal surface. A chest tube was placed, and patient underwent paracentesis and diagnostic laparoscopy. Peritoneal fluid, omentum, and peritoneal mass samples were sent for analysis. Histopathological examination showed extensive non-necrotizing granulomatous inflammation with adjacent small focal areas of necrosis. AFB and fungal stains on the biopsy were negative. Peritoneal culture grew Propionibacterium acnes. Bronchoscopy was also performed. AFB stain and cultures on bronchioalveolar lavage of the right lower lobe were negative, lymph node biopsy from the carina showed normal mucosa.

Patient was started on a three-day course of high dose steroids and began to show improvement in his symptoms. Patient endorsed tolerating oral intake and having an improved appetite with regular bowel movements and no abdominal pain, nausea, or vomiting. Repeat CT chest and abdomen showed trace ascites and a nearly resolved right sided pleural effusion. Chest tube was eventually removed after two weeks without recurrence of pleural effusion. Patient was discharged on oral prednisone with outpatient follow up. An autoimmune workup subsequently came back negative.

Serum protein electrophoresis was consistent with a chronic inflammatory pattern. Quantitative immunoglobulin was sent and ruled out common variable immunodeficiency (CVID). Tissue transglutaminase IgA and IgG antibodies, and endomysial IgA antibodies were negative, ruling out Celiac disease.

Discussion

Sarcoidosis is a granulomatous disease of unknown etiology, characterized by noncaseating granulomas on tissue biopsy. Greater than 90% of patients with sarcoidosis have lung involvement. GI involvement is rare and occurs in less than 1% of patients.[2] A recent report in the American Journal of Gastroenterology showed that 80% of GI sarcoidosis involves the proximal GI tract (up to the duodenum) with gastric involvement being the most common.[3] According to the American Thoracic Society 2020 Practice Guidelines, the diagnosis of sarcoidosis is based upon three major criteria: a compatible clinical presentation, the finding of non-necrotizing granulomatous inflammation in one or more tissue samples, and the exclusion of alternative causes of granulomatous disease. Sarcoidosis involving the jejunum is quite rare. A review of databases showed only very few cases reported of those the clinical presentation was variable and included the following: massive haemorrhage from a jejunal sarcoid lesion, acute small bowel obstruction at the level of the jejunum in a patient with inactive pulmonary sarcoid, involvement of the terminal ileum with some jejunal involvement, nonspecific granulomatous inflammation of the stomach and duodenum, and acute small bowel obstruction with ophthalmology, pulmonary and joint sarcoid.[3, 4] Of interest, latent infection with Propionibacterium acnes has been proposed to be linked to development of sarcoidosis secondary to a hypersensitivity reaction against the microorganism.[5] Given the extremely rare involvement of the small intestine with GI sarcoidosis, jejunal sarcoid can be hard to diagnose especially in the absence of any pulmonary or other extrapulmonary manifestations. Delay in diagnosis and mislabeling GI presentation of sarcoidosis as inflammatory bowel disease have been reported in the literature before. Diagnosis tend to be confirmed after appearance of systemic sarcoidosis manifestations over the course of years.[6] Small bowel sarcoidosis should be sought in the differential diagnosis of unclear gastrointestinal clinical presentation, especially in younger populations with recurrent GI symptoms with negative initial workup.

While imaging might be in favor of the diagnosis with presence of diffuse mesenteric and retroperitoneal lymphadenopathy, tissue biopsy and pathology is needed to confirm the diagnosis. Treatment tend to be heterogenous and depends on the clinical presentation and the degree of GI involvement, and varies from watchful waiting, to steroids with or without other immunosuppressive agents, and surgical in some cases.[3] More data and follow up of these cases is needed to establish the prognosis of small bowel sarcoidosis, however the prognosis of GI sarcoidosis is usually favourable, and the main cause of death remains cardiac or pulmonary involvement.[7]

Conclusion

This case reminds physicians that sarcoidosis of the small bowel, though rare, should be considered as a cause of SBO even in patients without pulmonary or other extrapulmonary manifestations of sarcoid.

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