Case Report

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Acute intestinal obstruction revealing an undiagnosed jejunal sarcoidosis in a 59-year-old female

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Background: Sarcoidosis is a granulomatous illness with no known cause, defined histopathologically by noncaseating granulomas. While 90% of cases involve the lungs and mediastinal lymph nodes, clinically recognizable gastrointestinal sarcoidosis occurs in only 0.1–0.9% of patients, with small bowel involvement occurring in as few as 0.03% of all cases, and when it does occur, it is usually late in the disease course in patients with multisystem disease.

Case presentation: Herein, we report a case of 59-year-old patient who presented with a picture of complete jejunal obstruction and gastrointestinal bleeding. Imaging revealed jejunal thickening and mesenteric lymphadenopathy. Later, intra-operatively, it be found caused by a jejunal stricture and enlarged mesenteric lymph nodes secondary to an undiagnosed sarcoidosis. Postoperative complications included hepatic hematoma and gastrointestinal bleeding. She stabilized after multidisciplinary management. A 1-year follow-up showed mediastinal lymphadenopathy without pulmonary involvement.

Discussion: To provide individualized therapeutic care and follow-up, gastrointestinal sarcoidosis is a complex, possibly lethal condition that requires a multidisciplinary approach and early clinical suspicion. Without a prior diagnosis, sarcoidosis must be taken into account as a possible cause of acute intestinal blockage.

Conclusion: Our case of jejunal sarcoidosis is unique for the rarity of disease location, the lack of pre-existing diagnosed sarcoidosis, and the necrotized jejunal resected segment indicating bowel ischemia.

Keywords: abdominal pain, case report, intestinal obstruction, jejunal sarcoidosis

Introduction

Sarcoidosis disease is a multi-system disease with unknown etiology. Since the first discovery of sarcoidosis disease in 1899, studies started about it showing it affects people of all ethnicities and races and involves all age groups but it classically develops before 50 years of age peaking from 20 to 39[1]. Despite being the causes of sarcoidosis have remained unexplained, the list of potential causal agents has continuously enlarged[2]. Sarcoidosis is an uncommon systemic granulomatosis with an incidence estimated to be between 4.7 and 64/100 000 people[3]. The cardinal feature of sarcoidosis relies on the development and accumulation of granulomas which are formed and maintained by CD4+ T cells that interact with antigen-presenting cells[1].

It affects different organs but is almost mainly dominant in the lungs (90%) and lymph nodes (30%)[2]. Organ involvement with poor prognostic signs includes the heart and central nervous system[3]. However, digestive tract involvement is extremely rare accounting for only 0.1% of cases and of them, the stomach, particularly the antrum is the most to be involved, small bowel involvement occurs only in 0.03% of the cases[4]. Which usually when occurs, comes in patients after chronic multisystem involvement[5]. It is highly unpredictable to diagnose sarcoidosis after an initial presentation of intestinal obstruction.

Here, we report an unusual case report of a 59-year-old female who diagnosed intraoperatively with jejunal sarcoidosis after she presented with sign and symptoms of small bowel

HIGHLIGHTS

- Sarcoidosis is an uncommon but serious disease which affects different organs including the lungs, lymph nodes, central nervous system, and the heart.
- Sarcoidosis defined mainly by histopathological features including noncaseating granulomas.
- Gastrointestinal tract involvement of sarcodiosis is significantly rare presentation with highly challenging diagnosis.

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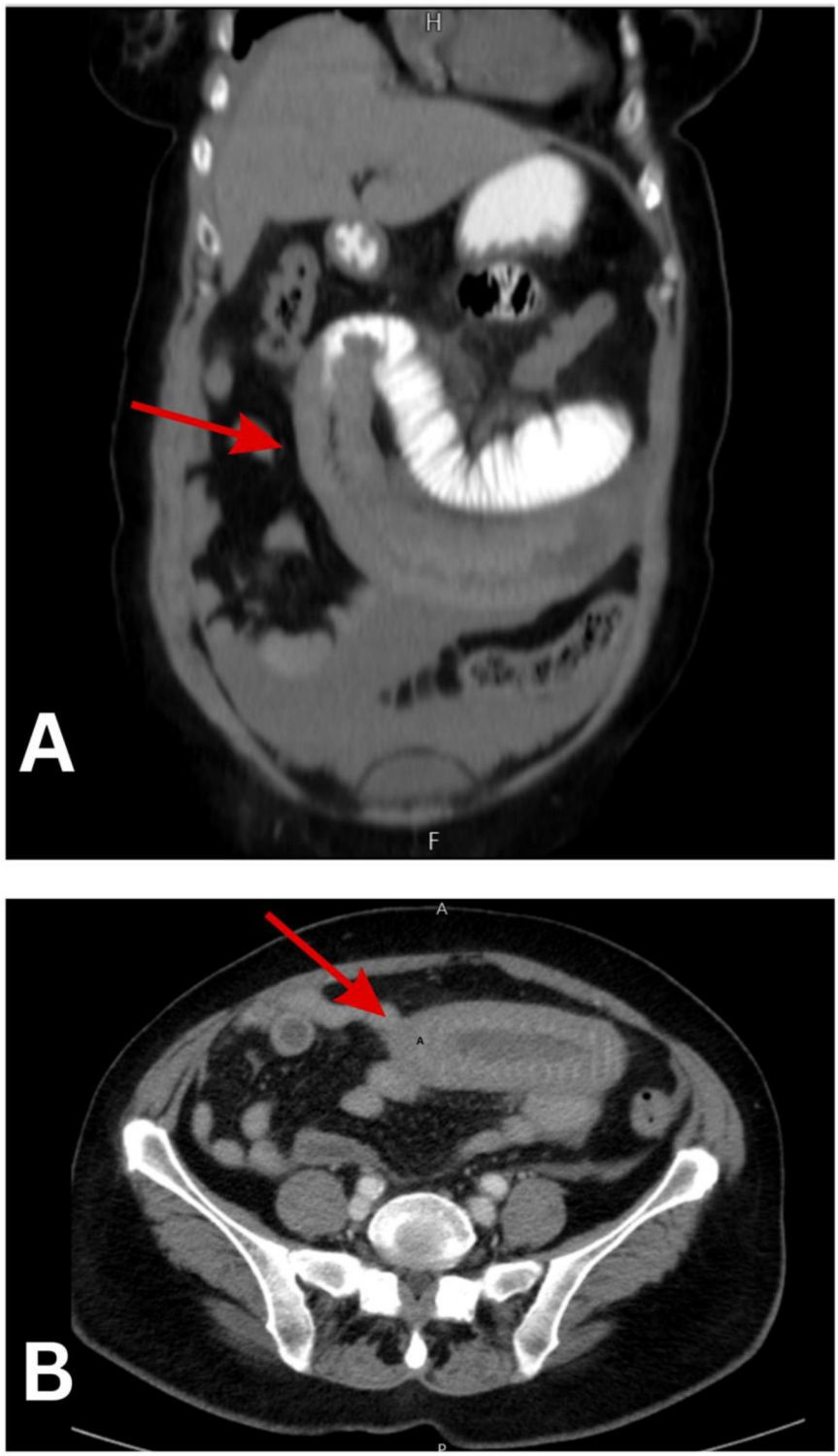


Figure 1. Contrast enhanced computed tomography (CT) scan of the abdomen and pelvis coronal (A) and axial (B) views show a segmental dilatation of the proximal part of the jejunum up to 4 cm in diameter, followed by a jejunal segment showing diffuse wall thickening that resulted in significant luminal narrowing with almost complete obstruction (Red arrows) and complete collapse of the distal small bowel loops and colon, with good wall contrast enhancement, and surrounding mesenteric fat stranding and enlarged mesenteric, para aortic and iliac lymph nodes as well as significant amount of abdominal free fluid.

obstruction. By reporting this unusual case, we highlight the rare presentation of abdominal sarcoidosis and the diagnostic challenges in identifying granulomatous diseases, in addition to confirm the importance of a multidisciplinary approach in managing surgical emergencies complicated by systemic conditions.

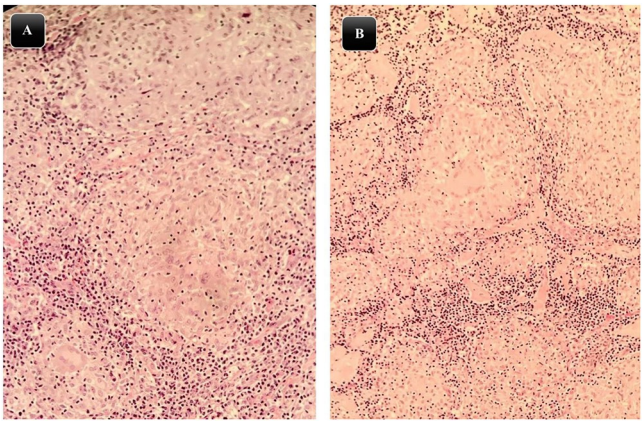


Figure 2. A and B) show H&E:20X: Compact Non-necrotizing granulomas with occasional Langhans giant cells.

Case presentation

A 59-year-old female with a past medical history of diabetes mellitus, hypertension, and hypothyroidism, and a past surgical history of open cholecystectomy, open appendectomy, aortic and mitral valve replacement due to rheumatic heart disease on warfarin anticoagulation, presented to the emergency department with a 2-day history of generalized abdominal pain associated with nausea, vomiting, and constipation. On physical examination, she was conscious, alert, and oriented. Her vital signs were normal except for tachycardia of 125 bpm. Her abdomen was distended with generalized tenderness and no evidence of organomegaly. Bowel sounds were hyperactive. Initial imaging study with abdominal x-ray revealed a concerning single proximal air-fluid level with a distal almost airless abdomen. A contrast-enhanced computed tomography (CT) scan of the abdomen and pelvis showed signs of a jejunal segment of diffuse wall thickening that resulted in significant luminal narrowing and resultant small bowel obstruction (Fig. 1A,B).

During her evaluation in the emergency department, she developed rectal bleeding with tachycardia and orthostatic hypotension, her hemoglobin dropped from 9.1 to 6.9, so she was transferred to the intensive care unit for close observation, correction of her INR via IV vitamin K and started on intravenous fluids and blood transfusion, warfarin was switched to heparin infusion, with follow up CBC and coagulation profile.

She was diagnosed as a case of complete jejunal obstruction with gastrointestinal bleeding, prepared for surgery on the next day, and an open laparotomy was done that revealed 1.5 L of free blood in the abdominal cavity, about 20 cm of thickened jejunal segment with patches of necrosis and bleeding 30 cm from the ligament of treitz, thickened and congested mesentery of the previous described jejunal segment, and enlarged lymph nodes. So, she underwent resection of the obstructed jejunal segment with side-to-side anastomosis.

Histopathologic examination of tissue biopsy revealed ischemic necrosis of the bowel segment and lymph node biopsy showed multiple compact and coalescent non-necrotizing granulomatous and Langerhans's giant cells (Sarcoid-type granulomas), PAS and Ziehl Neelsen Stains were negative compatible with sarcoidosis (Fig. 2).

Retrospectively, the patient's records also included previous abdomen and Lumbosacral CT scans 1 and 2 years before this admission that showed the presence of multiple Paraaortic enlarged lymph nodes (Fig. 3).

Her postoperative course was complicated by subcapsular hepatic hematoma surrounding the right lobe measuring 3 cm, upper gastrointestinal bleeding, and hemoglobin drop, which required blood transfusion due to the heparin infusion that was started on the postoperative day 0 as she had a high risk for a stuck metallic valve. Then on postoperative day 13, she was started on Warfarin bridging with heparin, hemoglobin levels and the subcapsular hepatic hematoma were stable upon her serial follow-up during hospitalization. Thereafter, she was discharged to follow up with the rheumatology outpatient clinic for further evaluation of the sarcoidosis.

The patient returned 1 year later for the rheumatology clinic, she was doing well, off complaints, asymptomatic. Follow-up Chest CT scan showed multiple enlarged mediastinal lymphadenopathy with no parenchymal pulmonary involvement (Figure 4).

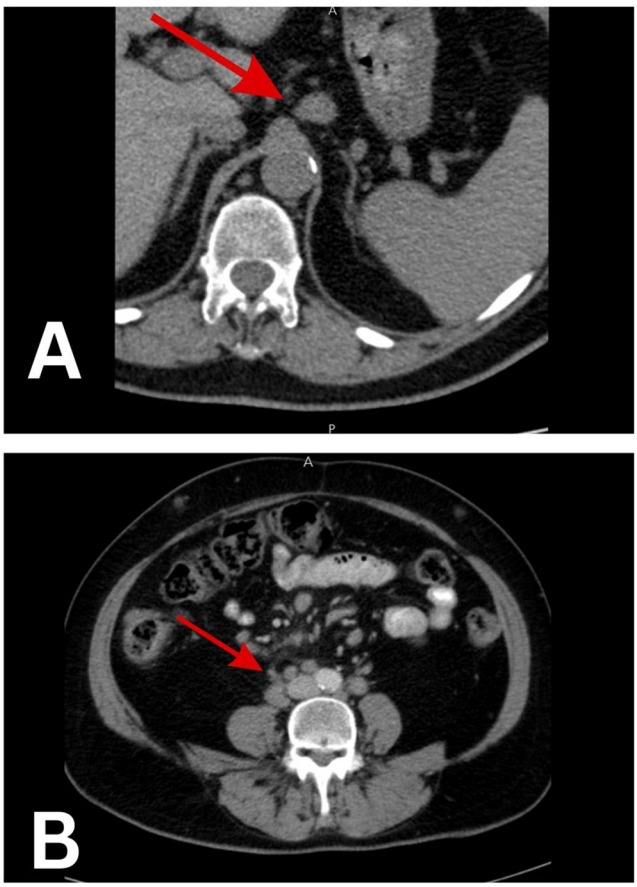
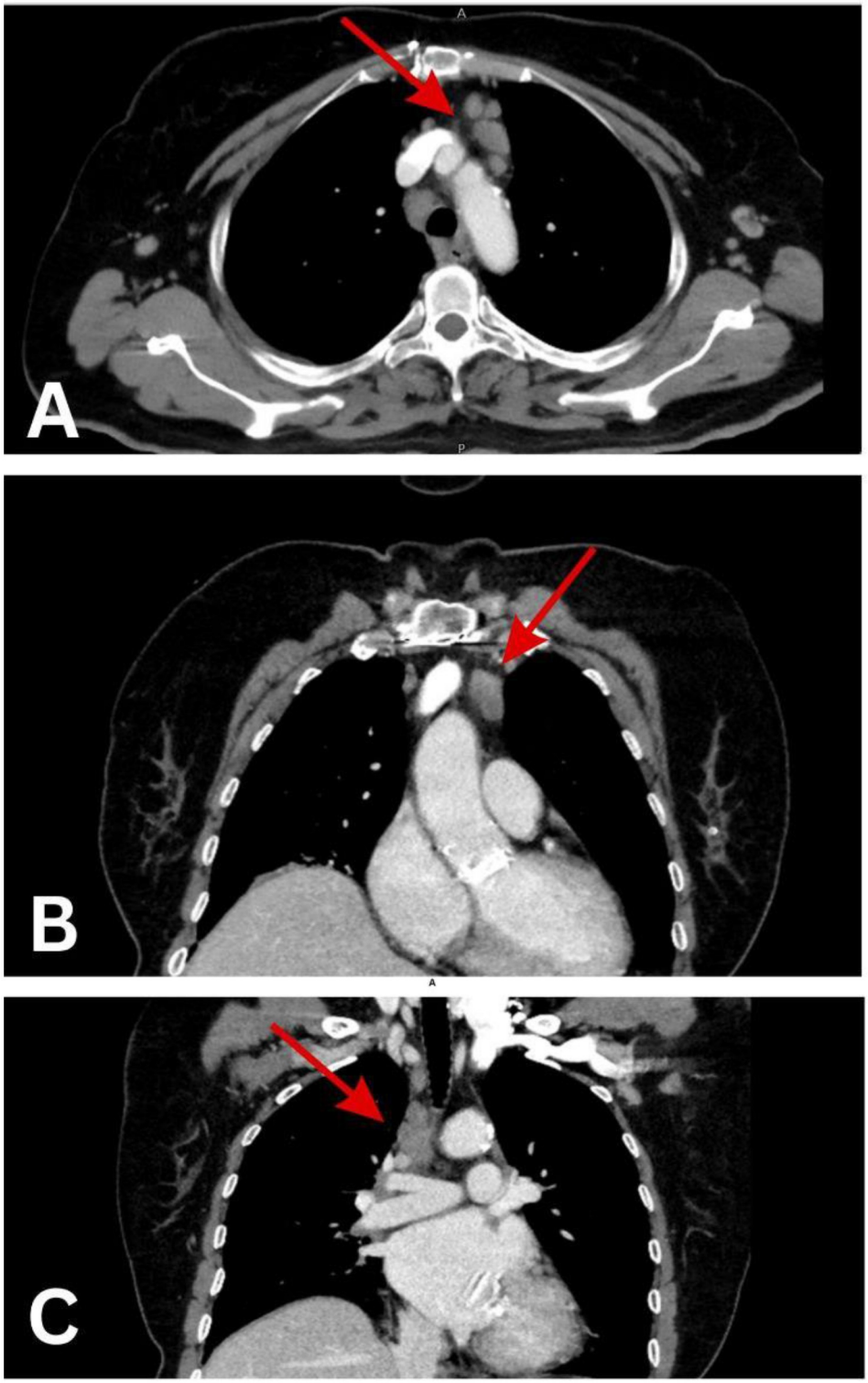


Figure 3. Lumbar (A) and stone protocol (B) unenhanced CT scans done 1 and 2 years respectively prior to the presentation showing multiple enlarged paraaortic lymphadenopathy (red arrows) with no signs of bowel wall thickening.

Discussion

Sarcoidosis is a granulomatous disease of no clear etiology where histopathological examination is characterized by noncaseating granulomas. While 90% of cases involves the lungs and mediastinal lymph nodes[6], clinically recognizable gastrointestinal



Figures 4. A, B and C) show follow up coronal and axial Chest CT scans one year after presentation revealing multiple enlarged mediastinal lymphadenopathy up to 2 cm in the retrocaval region.

sarcoidosis occurs only in 0.1–0.9% of patients, with small bowel involvement being as rare as 0.03% of all cases, that when happens usually occurs late in the disease course in patients with multisystem disease, our patient who had an acute episode of small bowel obstruction interestingly unveiling sarcoidosis diagnosed after histopathological examination of a resected necrotized 20 cm segment of jejunum and mesenteric lymph nodes revealed noncaseating granuloma[6].

When gastrointestinal sarcoid pathology occurs, It is usually caused by granulomatous organ infiltration or by mechanical compression caused by an enlarged lymph node[7]. Sarcoidosis-related intestinal manifestations are non-specific, The most common manifestation being nonbloody diarrhea associated with colicky abdominal pain[4]. It ranges from pain mainly in the epigastric and periumbilical regions, chronic diarrhea causing weight -loss and malabsorption, protein-losing enteropathy to even intestinal obstruction[5]. Other unusual features include mesenteric venous insufficiency caused by pressure from enlarged involved lymph nodes, megaloblastic anemia from terminal ileum involvement[4] which all could be found in multiple intestinal pathologies, i.e.. Crohn's disease, mycobacterial, fungal, and parasitic infections, which led to delayed diagnosis and mislabeling the gastrointestinal manifestations as inflammatory bowel disease where the diagnosis of sarcoidosis is made late after the appearance of systemic manifestations over years. In addition, initial clinical and endoscopic findings can be highly suggestive of gastrointestinal malignancy. Nevertheless, the definite diagnosis of sarcoidosis-related small intestinal impairment can only be established after histopathological examination showing noncaseating granulomas[5]. Our patient was lucky enough to be diagnosed with intestinal sarcoidosis before she had to wait for the development of multiple system involvement.

Tissue biopsy is always required to confirm the diagnosis, even if radiological features might be in favor of the diagnosis showing retroperitoneal or mesenteric lymphadenopathy[6].

The treatment of GI sarcoidosis is determined by symptomatology and disease activity; asymptomatic individuals can be watched without active therapy, whereas symptomatic patients require active therapy[4]. Corticosteroids and other immunosuppressive agents, and surgical treatment should be considered in individuals who have significant organ involvement and granulomatous inflammation on tissue biopsy[4]. Moreover, the treatment of sarcoidosis-related small intestine involvement is still debatable. Previous authors have proposed that steroids are to be considered in individuals who have symptomatic lesions of the intestine and when associated with multi-visceral and severe sarcoidosis. The recommended initial dose for steroid treatment is 20–40 mg daily, with gradual dose reductions. It has been found that steroid medication improves intestinal gastrointestinal symptoms in 66% of individuals[5].

The prognosis is usually favorable, and the main cause of death remains cardiac or pulmonary involvement. Small bowel sarcoidosis should be sought in the differential of unclear GI clinical presentation, especially in younger adults with recurrent symptoms[6].

A review of the literature revealed very few cases only reporting jejunal sarcoidosis of those the clinical manifestations showed high variability including jejunal sarcoid lesion with massive lower gastrointestinal hemorrhage, a patient with inactive pulmonary sarcoidosis presenting with acute small bowel obstruction at the level of the jejunum, terminal ileum involvement with associated mild jejunal involvement, granulomatous inflammation of the stomach and duodenum, and acute small bowel obstruction associated with ophthalmic, pulmonary and joint sarcoidosis[8].

Conclusion:

Sarcoidosis of the GI tract is a diverse, potentially fatal illness that necessitates a multidisciplinary approach and early clinical suspicion in order to establish tailored treatment care and follow-up. Sarcoidosis must be considered as a possibility to cause sudden intestinal obstruction without previously established diagnosis.

Our case of jejunal sarcoidosis is unique for the rarity of disease location, the lack of pre-existing diagnosed sarcoidosis, and the necrotized jejunal resected segment indicating bowel ischemia.

Ethical approval

Approval for this study was provided by the Ethical Committee of our institution (Al-Quds University, Jerusalem, Palestine).

Consent

Written informed consent was obtained from the patient for publication and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

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Author's contribution

K.S., M.H., and H.A. contributed to writing the first draft. Y.N. revised and edited the draft and added the radiology part. O.A. and B.J. supervised the project. All authors contributed to the article and approved the submitted version.

Conflicts of interest disclosure

Not applicable.

Research registration unique identifying number (UIN)

Not applicable.

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Data availability statement

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