

CASE REPORT

Sacroiliitis preceding disseminated pelvic and peritoneal tuberculosis

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ABSTRACT

Mycobacterium tuberculosis is the infectious agent responsible for tuberculosis (TB), one of the most common infectious causes of death in adults worldwide. It's protean manifestations may mimic different diseases in almost all clinical specialties. Herein we present a case of untreated tuberculosis sacroiliitis (SI) that evolved to disseminated pelvic disease mimicking ovarian carcinoma with peritoneal carcinomatosis. The patient complained of low back pain three years ago which was investigated with osseous scintigraphy and magnetic resonance imaging of the sacroiliac region and confirmed a left asymmetric SI. She was oriented to sporadically use of non-steroid anti-inflammatory drugs by the general physician who suspected spondyloarthropathy. Despite this, the symptoms got worse and progressed to abdominal pain, especially in hypogastric region, chronic diarrhea, severe night sweats, menstrual irregularity, weight loss, dyspareunia and undulant fever during the last year. She was admitted at this point and submitted to an exploratory video laparoscopic, the histopathology findings were strongly suggestive of disseminated pelvic TB showing granulomatous chronic inflammatory process with central caseous necrosis. The clinical team decided to initiate TB treatment, after 9 months of completed therapy, she was asymptomatic. The diagnosis of peritoneal tuberculosis usually is a challenge and invariably demands peritoneal biopsy. Mycobacterium tuberculosis can reach peritoneal tissue through the blood, lymphatic system and by contiguity. This case report presents an unusual form of pelvic and peritoneal disseminated tuberculosis preceded by a SI misdiagnosed as spondyloarthropathy.

Key Words: Extrapulmonar tuberculosis, Sacroiliitis, Pelvic tuberculosis

1. INTRODUCTION

Tuberculosis (TB) is one of the most mimicker diseases of actuality, principally on developing countries. The spread of human immunodeficiency virus (HIV) and the use of new immunosuppressive drugs has incremented the morbidity and mortality of TB. TB incidence and drug resistance are

still rising.^[1] The respiratory tract is the most commonly involved system, especially lungs, however almost any part of the body may be involved. Because of this reason, every physician should be alert to the possibility that tuberculosis mimics some diseases of their own specialty.

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Extrapulmonary tuberculosis and miliary tuberculosis usually present in a subacute/chronic fashion, with or without constitutional symptoms like night sweats, weight loss, fever of unknown origin (FUO) and variable organ specific manifestations. One of the most involved extrapulmonary tuberculosis sites is the lymphatic system,^[2] but involvement of bones, joints, gastro-intestinal and genitourinary tracts is not uncommon.

When TB has disseminated spread, it may be confounded with neoplastic lesions or other infectious diseases such as brucellosis, especially at bones, joints or genital tract. These situations usually require surgical approach to yield a correct diagnosis.^[3,4] Herein we present an unusual case of disseminated pelvic tuberculosis which began as unilateral sacroiliitis (SI) with progressive spread to ovaries, colon and peritoneum.

2. CASE PRESENTATION

A 35-year-old woman was transferred to our tertiary care to investigate a three year history of back pain that progressed to abdominal pain and menstrual irregularity. The patient reported good health since she began with low back pain three years ago which was investigated with osseous scintigraphy and magnetic resonance imaging of the sacroiliac region and confirmed a left asymmetric SI (see Figure 1). She was oriented to sporadically use of non-steroid anti-inflammatory drugs by the general physician who suspected spondyloarthritis. Despite this, the symptoms got worse and progressed to abdominal pain, especially in hypogastric region, chronic diarrhea, severe night sweats, menstrual irregularity, weight loss, dyspareunia and undulant fever during the last year. She denied vaginal discharge or urinary complaints as so any comorbidities or previous disease. She denied alcoholism or smoking.



Figure 1. Left asymmetric sacroiliitis

On physical examination she was very emaciated and her vital signs are normal. The abdomen was tender but without

rebound and palpable masses. Bimanual vaginal examination revealed positive cervical excitation. The Gaenslen test was positive suggesting SI. The rest of physical exam was unremarkable.

The initial investigation workup was normal for electrolytes, renal and hepatic function, blood and urine cultures, chest X-Ray, complete blood count, except for discrete anemia of chronic disease, an elevated CA125 of 366.6 mg/dl. The HIV 1 and 2 serologies were negative. The tuberculin skin test was nonreactive and *Brucella mellitensis* serology was negative (see Table 1). She was submitted to transvaginal ultrasound that suggested possible left adnexal septate cyst confirmed by pelvic tomography which showed retro-uterine progression and gluteal muscles and large intestinal involvement. Colonoscopy revealed narrowing of sigmoidal lumen by extrinsic compression and little mucosal erosions at recto-sigmoid region which was biopsied.

Table 1. Laboratory tests at baseline

Laboratory Tests	Laboratory Results
Sodium (Na ⁺)	140 mmol/L
Potassium (K ⁺)	4.0 mEq/L
Calcium ion(Ca ²⁺)	1.20 mmol/L
Phosphor (P ⁻)	3.2 mg/dl
Magnesium (Mg ²⁺)	2.0 mg/dl
Aspartate Aminotransferase	25 U/L
Alanine Aminotransferase	20 U/L
Alkaline Phosphatase	100 U/L
Gamma Glutamyl Transferase (GGT)	20 U/L
Albumin	5.0 g/L
	Time: 12'
Prothrombin Time	Activity: 90%
	INR: 1.0
Creatinine	0.8 mg/dl
Urea	30 mg/dl
Urine Protein	80 mg/24h
Fasting blood glucose	80 g/dl
Red blood cell count	4.2 million/ μ l/mm
Hematocrit	33 %
Hemoglobin	10.8 g/dl
Platelet count	200.000/mm ³
Ferritin Level Blood	90 mcg/dl
HIV 1 and 2 serologies	Negative
CA125	366.6 mg/dl
Tuberculin skin test	Nonreactive
Brucella mellitensis serology	Negative

The patient was submitted to an exploratory video laparoscopic (VLP) surgery that, macroscopically, was suggestive of PC with multiple foci of implants on peritoneum which were biopsied. VLP also showed disseminated implants on small and large intestine with diffuse fluid collection in the

pelvis. The histopathologic findings of the peritoneal and large intestine biopsies were compatible with granulomatous chronic inflammatory process showing caseous necrosis (see Figures 2-3). Although the histopathologic findings were strongly suggestive of disseminated pelvic TB, the pathologist did not report it as such since the culture result and

bacilloscopies were negative. The clinical team decided to initiate TB treatment with rifampicin, pyrazinamide, ethambutol and isoniazid. After 4 days of therapy the fever and night sweats have improved. After 9 months of completed therapy, she was asymptomatic and a new magnetic resonance imaging of sacroiliac joint showed partial recovery.

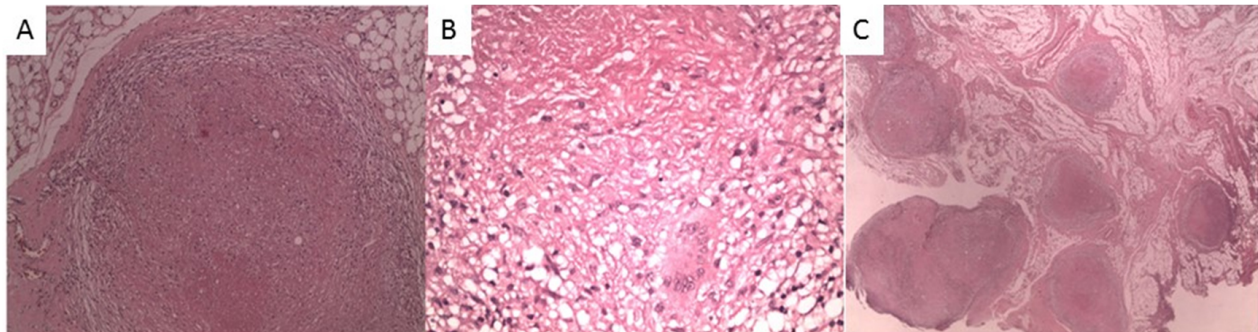


Figure 2. Peritoneal biopsies showing granulomatous chronic inflammatory process with central caseous necrosis: A) High power magnification showing central caseous necrosis B) High power magnification showing epithelioid histiocytes and a multinucleated giant cell C) Low power magnification showing granulomas with caseous necrosis.

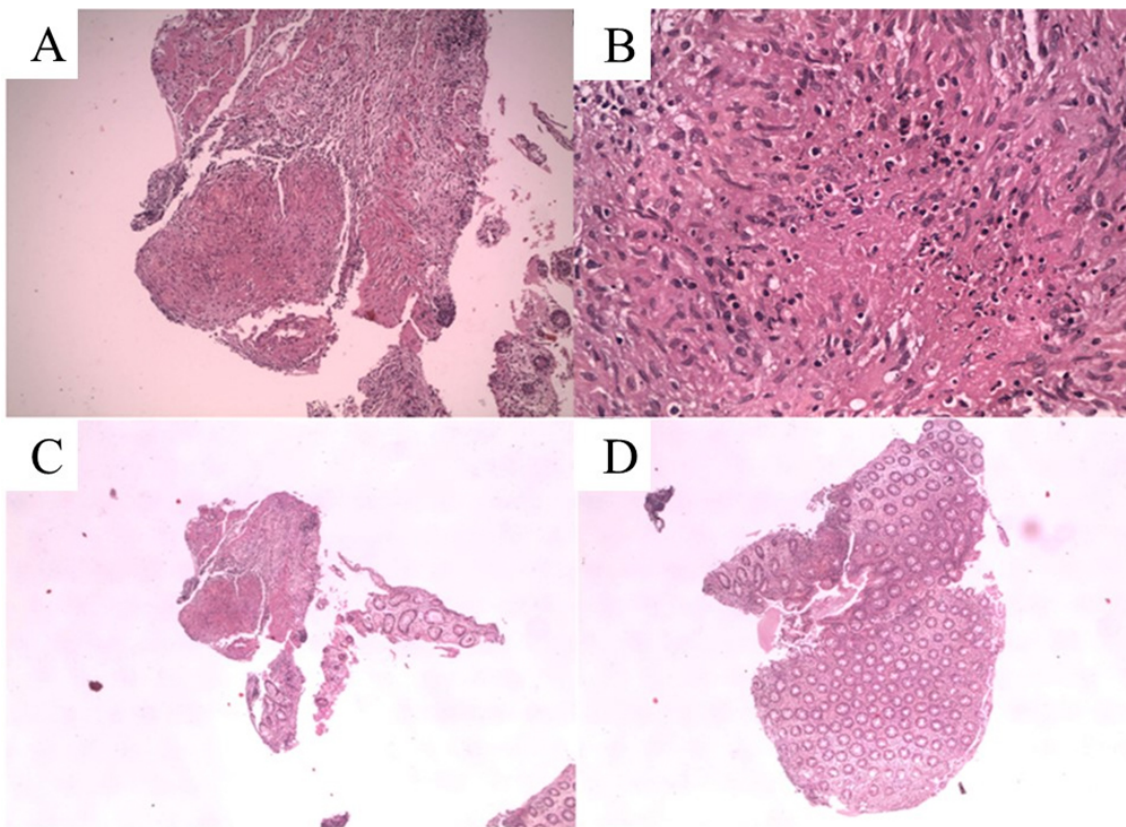


Figure 3. Large intestinal biopsies compatible with granulomatous chronic inflammatory process with central caseous necrosis

A: High power magnification showing architecture of the large intestine distorted by the presence of epithelioid granulomas with central caseous necrosis; B: High power magnification showing granuloma in the lamina propria of the large intestine fragment with central area represented by focus of caseous necrosis and cellular debris; C: Low power magnification showing the large intestine mucosa distorted by the presence of inflammatory infiltrate in the lamina propria; D: Normal fragment of large intestine.

3. DISCUSSION

The diagnosis of extrapulmonary TB is often difficult considering the protean clinical manifestations and the low degree of suspicion. It is especially difficult when there is no obvious previous history of contact with Koch bacillus, none respiratory symptom and no known immunosuppression like HIV infection.^[2,3] Approximately 10%-15% of tuberculosis presents with extrapulmonary involvement with higher indexes when considering immunocompromised patients.^[2]

This case illustrates a patient who initially presented with SI with posterior progression of her symptoms to the gynecologic and gastrointestinal tract. One of the most common extrapulmonary manifestations of tuberculosis is bone and joint involvement which may mimic some rheumatologic diseases. Poncet's arthritis,^[4] TB arthritis,^[5] peripheral septic arthritis,^[6] TB spondylodiscitis and SI are some forms of presentation of extrapulmonary tuberculosis.^[7-10] Even without any pathognomonic radiologic sign of TB SI, nuclear resonance showing subchondral bone marrow edema and irregularity of articular surface on an asymmetric distribution confirms unilateral SI.^[11,12] Asymmetric SI without any other findings suggestive of spondyloarthropathy should be promptly investigated for infectious disease.^[13] The chronic presentation of asymmetric SI pointed the clinical team in front of two possible infections: brucellosis and TB. Despite the macroscopic aspect of peritoneal carcinomatosis found on VLP, SI is not a habitual manifestation of cancer.

Brucellosis is an infection disease caused by gram-negative bacteria *Brucella sp.* The most common agent to cause infections in humans is *Brucella melitensis* with variable cases of other species. It has protean manifestations frequently with constitutional symptoms and a predilection to gastrointestinal and osteoarticular manifestations.^[14,15] Gonadal manifestation of brucellosis is very rare, with few cases described in the literature.^[15] Brucellosis mainly affects the axial skeleton.^[14] On the contrary of vertebral involvement, SI secondary to brucellosis has no specific signs that could help on distinction between tuberculous SI and SI due to brucellosis. The undulant fever pattern, despite not specific, is characteristic of the disease together with profound night sweats with typical odor.

Confirmation of SI due to brucellosis is difficult with different sensitivity and specificity of serology and cultures. So, depending on the clinical context and epidemiologic risk factors suggestive to *Brucella melitensis* infection, an empirical therapy can be indicated.^[14,15]

Despite ovarian TB usually presents together with peritoneal involvement, SI due to TB generally occurs without other involved sites.^[8,9] Balci *et al.* reported a case of a 32-year-old

female admitted with pelvic pain and abdominal distension. Pelvic sonography revealed diffuse pelvic ascites and a simple cystic lesion measuring up to 3 cm × 4 cm in the right adnexal area. The pelvic CT scan showed diffuse fluid within the abdomen, nodules on the peritoneum, peritonitis carcinomatosis, and a simple cystic lesion on the right adnexal. At laparoscopic exploration, there were diffuse peritoneal ascites and a peritonitis carcinomatosis-like appearance. Biopsies were taken from peritoneal nodules, and frozen sections were examined. The pathology was concordant with tuberculosis granuloma. However there was neither involvement of the sacroiliac joint nor intestinal involvement.^[16]

There are two clinical forms of tuberculous peritonitis: "wet peritonitis" characterized by the presence of ascites and "dry peritonitis" associated with minimal or no ascites.^[17] The presence of ascites may facilitate the diagnosis, being a source for culture and providing material for biochemical analysis such as dosage of adenosine deaminase (ADA). Unfortunately, ascitic fluid culture for TB has low sensitivity (20% to 50%) and takes around 30 to 45 days to obtain results, delaying the treatment, and, despite good sensitivity and specificity of adenosine deaminase (100% and 95% respectively), in cases of dry peritonitis it is of little value on diagnosis of TB.^[18]

The diagnosis of peritoneal tuberculosis in cases of dry peritonitis usually is a challenge and invariably demands peritoneal biopsy.^[3,19] Mycobacterium tuberculosis can reach peritoneal tissue through the blood, lymphatic system and by contiguity. This case report presents an unusual form of pelvic disseminated TB started by a SI misdiagnosed as spondyloarthropathy. Gosein MA *et al.* reported a 44 year old female presented with a history of abdominal distension, weight loss, decreased appetite and low grade fever. Ultrasound scan showed moderate ascites with a septated cystic left adnexal mass. CT scan showed smooth peritoneal thickening. Biopsies were taken by laparotomy for tissue diagnosis, leaving the uterus and ovaries intact. Histopathologic examination revealed caseating granulomas with epithelioid histiocytes and Langhans-type giant cells. Again there was no involvement of the sacroiliac joint in this patient.^[20]

The association of gynecologic masses and peritoneal spread may be confounded by a gynecologic neoplasm showing peritoneal carcinomatosis. TB may involve the gynecologic tract by many forms like ovarian tuberculomas, cervical lesions and chronic salpingitis.^[21] Symptoms like infertility, dyspareunia, menstrual irregularity, ascites and abdominal mass present on variable fashion. Constitutional complaints may or may not be present. Fever of unknown origin and night sweats are good clinical clues to the diagnosis. The

measurement of CA125 lacks specificity. It can be elevated with variable conditions such as endometriosis, TB and ovarian tumors.^[22] Therefore, it is not uncommon to misdiagnose peritoneal TB as peritoneal carcinomatosis, especially in the presence of an ovarian mass. Despite the findings of computed tomography and macroscopic evaluation of an expert surgeon suggesting peritoneal carcinomatosis, the confirmation of the diagnosis by histopathologic examination is necessary.^[3,23]

Some cases of diffuse pelvic TB with peritoneal involvement mimicking peritoneal carcinomatosis were described in the literature. Nevertheless, the present case report has a peculiar characteristic: the clinical presentation of SI preceding disseminated pelvic and peritoneal TB.

CONFLICTS OF INTEREST DISCLOSURE

The authors declare no conflict of interest.

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