Case Report

A rare case of pelvic actinomycosis mimicking an ovarian malignancy

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Abstract

Actinomycosis is a rare chronic infectious disease caused by *Actinomyces* species. Orocervicofacial actinomycosis is the most common presentation (50%), followed by thoracic disease (15%–20%) and abdominopelvic disease (20%). It poses great diagnostic dilemma due to its variable presentations. We present a case of a 45-year-old female who presented with complaints of abdominal pain, abdominal fullness, constipation, weight loss and loss of appetite for the past 1 year. On examination, the patient was found to have palpable mass in lower abdominal region measuring about 10 cm × 12 cm, nontender, immobile mass. Patient was seen initially at another hospital, where ovarian malignancy was suspected in view of large size and radiological findings. Laparotomy was done but in view of adhesions and vascularity, multiple biopsies were taken and abdomen was closed. The patient was diagnosed to have actinomycosis based on histopathology and was treated accordingly. This case stresses on the diagnostic importance of clinical suspicion of actinomycosis in patients presenting with long-standing abdominal complaints, especially with history of previous surgeries.

Keywords: Actinomycosis, Frozen pelvis, Ovarian malignancy mimickers, Ovarian mass, Pelvic mass

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INTRODUCTION

Actinomycosis is a rare chronic infectious disease caused by *Actinomyces israelii*, an aerobic or microaerophilic Gram-positive bacteria present in the oral cavity, throughout the gastrointestinal tract, female genital tract and the bronchus.[1] *Actinomyces* has low virulence; consequently, disease occurs when the mucosal barrier has been compromised or in patients who are immunocompromised.[2] Diagnosis preoperatively is rarely made due to variable clinical presentations. The majority of cases are diagnosed after the specimen in question has been resected and examined histologically.[3] Abdominal actinomycosis develops after a localised inflammatory process, prolonged intrauterine device (IUD) use or recent abdominal surgery.[4] The appendix, cecum and colonic diverticulum are most affected. Its symptomatology imitates some malignant pelvic tumours, tuberculosis or nocardiosis, causing abscesses and fistulas.[2] The direct extension of *Actinomyces*, across the tissues, leads to the formation of multiple abscesses, abundant granulation tissue and sinuses.[5] The involvement of surrounding structures not only contributes to the insidious clinical

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course and delay of diagnosis but also may mimic a tumour. This report presents a case of intra-abdominal actinomycosis mimicking abdominal malignancy.

CASE REPORT

A 45-year-old housewife, resident of Visakhapatnam, presented with chief complaints of abdominal fullness, early satiety, loss of weight, constipation and lower abdominal pain which was of dull aching character for 1 year. Abdominal pain increased on taking food and relieved slightly on passing stools. There was no history of nausea, vomiting, fever, night sweats, shortness of breath and cough. There was no history of blackish discolouration of stools, or fresh blood in stools. There was no history of generalised weakness or easy fatigability. No history of similar complaint in the past. No history of diabetes mellitus, hypertension, asthma, epilepsy, malignancies and tuberculosis. The patient had normal menstrual history. She underwent tubectomy 10 years ago, and there was no history of usage of IUD. She consulted a local physician who prescribed her proton-pump inhibitors. As the pain persisted, she consulted a gynaecologist who felt a mass in the abdomen. The patient underwent laparotomy at a private hospital in Visakhapatnam, but resection was not done due to adhesions and the patient was referred to our hospital after obtaining open biopsy specimens along with histopathology blocks and sides. On examination, the patient was moderately built and nourished. Pallor was present; there was no icterus, or lymphadenopathy or pedal oedema. Vitals were stable. Examination of the abdomen revealed a firm-to-hard, nontender, immobile mass of size measuring 9 cm × 10 cm was palpated, in the left iliac fossa, left lumbar and suprapubic regions. Margins of the mass could not be delineated and lower border of the swelling was not palpable, suggestive of either pelvic extension of the mass or arising from the pelvis. Per speculum examination, the cervix was healthy. Per vaginal examination revealed a mass felt through the left and anterior fornices, and the uterus was not separately made out. Per rectal examination revealed a mass compressing the rectum from anterior aspect with nodular surface.

Laboratory studies demonstrated a white blood cell count of 8800/mm^3, haemoglobin 9.2 g/dL, platelet count of 160,000 mm^3 and mean corpuscular volume of 79.9 FL (normal 82–99 FL). Liver function tests, renal function tests, blood glucose levels and complete urine examination all were within the normal limits. Contrast-enhanced computerised tomography (CECT) abdomen axial images showed large ill-defined heterogeneously enhancing solid cystic mass lesion in both the adnexae displacing urinary bladder anteriorly and sandwiching uterus [Figure 1]. Histopathological examination of open surgical biopsy

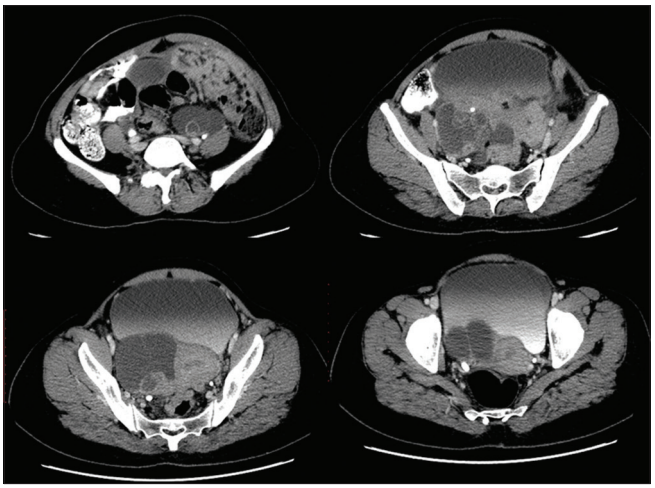

Figure 1: CECT abdomen axial images showing large ill-defined heterogeneously enhancing solid cystic mass lesion in both the adnexae displacing urinary bladder anteriorly and sandwiching uterus. Tiny speck of calcification is seen in the right side mass. Both ovaries are not visualised separately from the mass lesion. The omentum is thickened and showing nodularity. CECT = Contrast-enhanced computed tomography

specimen showed granular eosinophilic material surrounded by filamentous structures (Splendore-Hoeppili phenomenon) [Figure 2], suggestive of actinomycosis. Biopsy was negative for acid-fast staining and positive for Periodic Acid–Schiff staining. The patient was started on high-dose intravenous benzylpenicillin 4 million units, 6th hourly with a plan to continue it for 4 weeks and later switched over to oral penicillin therapy for 12 months more for complete resolution. However, the patient developed hypersensitivity reaction to penicillin after 2 weeks of treatment and hence managed with intravenous doxycycline and ceftriaxone for 2 more weeks and later switched over to oral doxycycline and cotrimoxazole. The patient was followed-up with CECT of the abdomen after 2 months of treatment, which showed a significant decrease in size of the growth, and the patient symptomatically improved and gained weight. The patient is being treated with the above

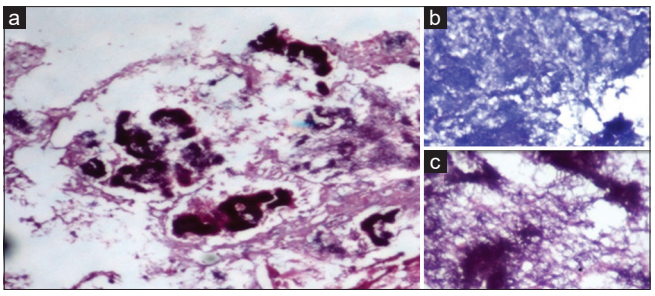

Figure 2: Photomicrograph of biopsy obtained from of pelvic mass showing granular eosinophilic material surrounded by filamentous structures (*Splendore Hoeppli phenomenon*) (Hematoxylin and eosin ×40) (a), negative for 1% acid-fast staining (×1000) (b) and positive for Periodic Acid Schiff staining (×400) (c)

antibiotics for about 2 years, and there has been a significant maximum regression of mass though incomplete. Hence, we planned to continue the patient on antibiotics further until complete resolution to prevent relapse.

DISCUSSION

Actinomycosis is an uncommon chronic granulomatous disease caused by *A. israelii*, slow-growing filamentous Gram-positive anaerobic bacteria that are commensals in human oropharynx, gastrointestinal tract (GI) (especially appendix and colon) and urogenital tract. Orocervicofacial actinomycosis is the most common presentation (50%), followed by thoracic disease (15%–20%) and abdominopelvic disease (20%).[6] A history of appendicitis, especially if ruptured, is the most common predisposing condition leading to abdominopelvic actinomycosis.[7] Other predisposing factors include a previous history of surgery, GI tract perforation, GI or genitourinary foreign body (particularly IUDs in women) and neoplasia.[6] Pelvic actinomycosis has recently become more prevalent and is associated almost exclusively with women who use IUDs (our patient had no history of using IUD). The ability for actinomycosis to secrete proteolytic enzymes, disrupt tissue planes and compress the surrounding tissue makes their appearance similar to a malignant process. Furthermore, due to the slow-growing nature of *Actinomyces*, the nonspecific clinical presentation and subsequent extensive spread before diagnosis, the diagnosis is often overlooked as occurred in our case. Abdominopelvic actinomycosis constitutes 20% of total cases of actinomycosis cases and may mimic malignancy, tuberculosis and other pelvic inflammatory diseases.[6] In pelvic actinomycosis, symptoms are typically indolent; fever, weight loss, abdominal pain and abnormal vaginal bleeding or discharge are the most common. The earliest stage of disease – often endometritis – commonly progresses to pelvic masses or a tubo-ovarian abscess. Unfortunately, because the diagnosis is often delayed, a 'frozen pelvis' mimicking malignancy or endometriosis can develop by the time of recognition.[8] Clinical presentation as well as imaging findings often misdiagnoses it as neoplasm resulting in delayed diagnosis in most of the cases. CA125 levels may be elevated, further contributing to misdiagnosis.[8] Misdiagnosis of abdominopelvic actinomycosis has earned the infection the title of 'the great pretender' and a 'greatest imitator' in clinical practice.[9] While actinomycosis commonly co-exists with Gram-negative organisms and other anaerobic bacteria such as *Actinobacillus* sp., *Eikenella* sp. and *Bacteroides* sp., single-agent therapy is usually successful. Although therapy must be individualised, the intravenous administration of 18–24 million units of penicillin daily for 2–6 weeks, followed by oral therapy with penicillin or amoxicillin (total duration, 6–12 months), is a reasonable guideline for serious infections and bulky disease.[9] The total duration of therapy though remains controversial. A prolonged treatment regime has been recommended due to the poor penetration of antibiotic into the inflammatory fibrotic tissue, especially when the disease is bulky as in our case. Ceftriaxone, tetracycline, minocycline, erythromycin and clindamycin can also be used to treat *Actinomyces* in patients allergic to penicillin.[8] We treated our patient with penicillin initially, later with sulfamethoxazole-trimethoprim and doxycycline in view of penicillin allergy. Antibiotics can cure very extensive pelvic disease. The role of surgery in the management of actinomycosis includes biopsy to exclude malignancy, resolving obstruction of the surrounding organs like ureters, etc. Furthermore, surgery is often required for diagnosis by obtaining tissue for histopathological examination and culture as in our case which proved the diagnosis. Regardless, the prognosis of pelvic actinomycosis is excellent, and the treatment outcome with antibiotics is usually complete resolution though needs prolonged therapy and follow-up.

Despite advanced imaging techniques, actinomycosis escapes the diagnosis unless histopathology and microbiologic tests are conducted. It is important to suspect infections like actinomycosis in women who present with a history of prolonged nonspecific symptoms and a presumed ovarian malignancy in imageology and a history of previous surgeries. This will spare patients from unnecessary, repeated extensive surgery and reduce morbidity and mortality.

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Conflicts of interest
There are no conflicts of interest.

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