

Case Report

Disseminated uterine actinomyces: A case report and review of literature

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ABSTRACT

Actinomyces - It mainly affects the cervicofacial and abdominopelvic organs. These bacteria are of low virulence and their spread requires breakdown or alteration of the normal tissue barrier, which can be due to any surgery, trauma, or infection. *Actinomyces* infection involving the reproductive organs may often mimic a neoplasm and lead to aggressive surgical management for the patient; it is therefore necessary to recognize the infection which can be effectively managed using a combination of pharmaceutical agents. Imaging such as computed tomography and magnetic resonance imaging aid in determining the extent of the disease. Histopathological diagnosis/culture is necessary to make the final diagnosis. We present a case of actinomyces of the uterus manifesting as a large uterine mass with dissemination to the pelvis and anterior abdominal wall.

Keywords: Actinomyces, Uterus, Computed tomography

INTRODUCTION

Actinomyces is a chronic infectious disease that is caused by a group of anaerobic, microaerophilic Gram-positive bacteria. These bacteria usually colonize the gastrointestinal tract and respiratory system. These infections many times present as infiltrative lumps and mimic malignancy. Radiological diagnosis is challenging and usually diagnosis is made by histopathological findings and culture reports. Risk factors for actinomyces are poor oral hygiene, diabetes mellitus, malnutrition, immunosuppression, and presence of foreign body.

Here, we describe the imaging findings in pelvic actinomyces and discuss imaging features differentiating pelvic actinomyces from a uterine neoplasm.

CASE REPORT

A 68-year-old women presented to our hospital with complaints of pain and lump in lower abdomen, discharge per vaginum, and off and on fever for 2 months. She also had complaints of constipation and mucoid discharge from the rectum. On general physical examination, she was a thin built lady with normal vitals. On abdominal examination, a large mass measuring $\sim 8 \times 8$ cm was seen in the right iliac fossa with local tenderness. On per speculum examination, her cervix looked normal, however, there was copious purulent discharge from os and threads of intrauterine copper device (IUCD) were seen coming out from the os. The patient belonging to low socioeconomic strata did not remember the placement of the IUCD and therefore did not follow-up for its removal on appropriate time. Per vaginal

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examination revealed an irregular fixed 12-week pelvic mass. Uterus was inseparable from mass. All fornices were full of mass and both parametrium appear infiltrated up to the pelvic wall. On per rectal examination, 8 × 8 cm mass was felt, projecting in the rectum, however, the mucosa of rectum was free.

Ultrasound abdomen revealed a bulky, enlarged uterus with diffuse and heterogeneous echotexture. Note was also made of poor endomyometrial delineation.

Computed tomography (CT) and magnetic resonance imaging (MRI) of the abdominopelvic region showed multiple poorly defined intramural lesions in uterus predominantly in the region of the fundus with associated serosal irregularity along the fundal surface. Endometrial cavity was poorly defined and showed and intrauterine device (IUD) *in situ* [Figures 1a-c and 2a-c]. A large peripherally enhancing collection with surrounding inflammatory changes was seen along the right anterolateral abdominal wall [Figure 1c]. Imaging findings raised the possibility of pelvic actinomycosis, in view of soft tissue extension of lesion with the absence of significant lymphadenopathy and ascites. Imaging morphology also suggested a possibility of malignant transformation of fibroid, a close differential of uterine actinomycosis. She was advised to get a positron emission tomography (PET) CT whole-body fearing sarcomatous degeneration of uterine fibroid. Whole-body PET CT was done

and a large Fluorodeoxyglucose PET avid heterogeneously enhancing mass lesion measuring 7.7 × 7.5 cm was seen in the uterine fundus; the uterus was inseparable from adjacent colon, raising the suspicion of colonic infiltration by the mass.

Furthermore, multiple FDG-avid serosal deposits [Figure 3a and b] were seen in the lower abdomen and pelvis, largest in right iliac fossa region of size ~6.1 × 4.6 cm. The lesion showed areas of necrosis and was seen invading right lateral abdominal wall muscle with subcutaneous fat stranding in the overlying soft tissues, suggestive of advanced malignancy.

She was then referred for ultrasound guided core biopsy of the abdominal implant. Ultrasound detected a solid cystic mass. Core biopsy was taken from the solid component and 100 mL straw colored purulent material was aspirated from the right iliac fossa mass and sent for aerobic and anaerobic culture sensitivity. Aerobic culture showed no bacterial growth. Anaerobic culture isolated *Bacteroides fragilis*, and no gram-positive or negative bacteria seen.

Histopathological evaluation of tissue sections showed fibro collagenous tissue with foci of dense acute inflammation, along with foci of mild lymphocytic infiltrate. Characteristic sulfur granules were noted in the histopathological sections [Figure 4a and b]. The bacterial colonies identified within

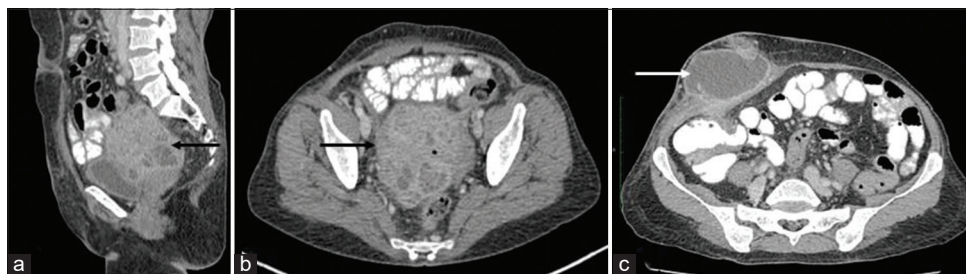


Figure 1: Contrast-enhanced (a) sagittal (b and c) and axial CT sections of pelvis show a bulky uterus, multiple intramural lesions (black arrows) with serosal irregularity with a peripherally enhancing collection (white arrow) in the right anterolateral abdominal wall with surrounding inflammatory changes.

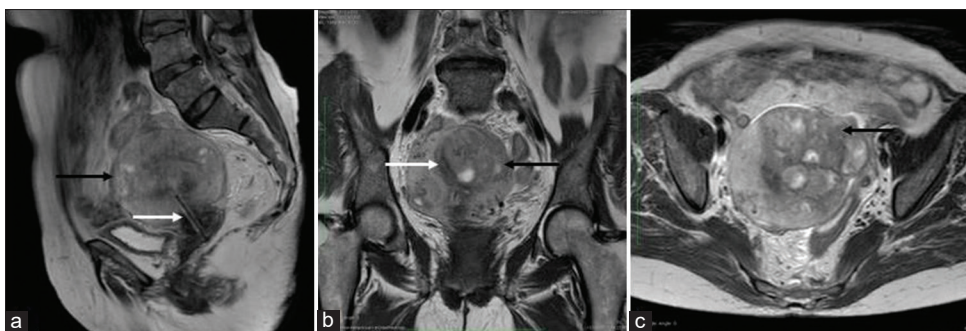


Figure 2: (a) T2-weighted sagittal, (b) coronal, and (c) axial magnetic resonance imaging images of pelvis showing a bulky enlarged uterus with multiple intramural lesions (black arrows) and serosal surface irregularity. Note a sharply delineated T2 hypointense line (white arrows) in the uterocervical region representing the intrauterine device.

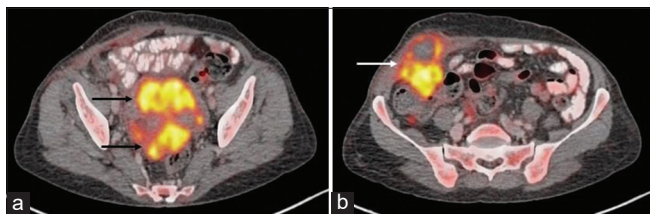


Figure 3: Computed tomography axial sections (a and b) of pelvis showing multiple FDG-avid enhancing lesions in the uterus (black arrows). Furthermore, note a an FDG enhancing lesion along the right anterolateral abdominal wall (white arrow).

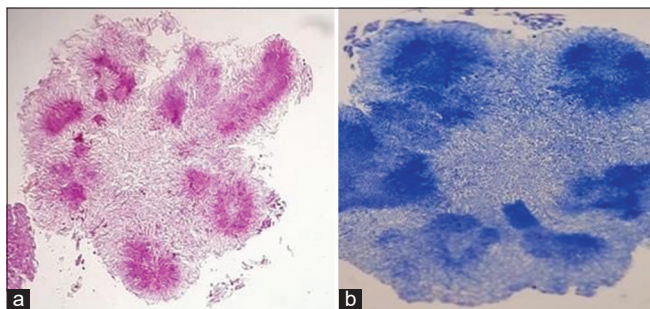


Figure 4: Photomicrograph-H and E $\times 200$ (a) and Gram stain $\times 200$ (b) show actinomycotic sulfur granules composed of numerous filamentous organisms with acute angle branching in a suppurative background. H and E: Hematoxylin and Eosin.

the specimen, morphologically favored actinomycosis. No granuloma was seen. Special stains for Acid fast bacilli (AFB)/Periodic acid schiff stain (PAS) were negative. No malignant cells could be identified.

Her IUCD was removed and its culture also revealed the same micro-organism, i.e., *Actinomyces*. As per the sensitivity, she was kept on injection piperacillin tazobactam 4.5 g IV once a day for 14 days, during which time her abdominal wall abscess ruptured with a sinus formation.

She was then shifted to injection ceftriaxone 2 g IV once a day for 30 days, following which her fever subsided, sinus healed and there was significant reduction in the size of the uterine mass. She is now on capsule augmentin 625 mg 3 times a day for the past 6 months and is presently asymptomatic. A follow-up CT of abdomen and pelvis done 6 months later confirmed the clinical findings, i.e., healing of the abdominal wall lesion and considerable reduction in size of uterus.

DISCUSSION

Actinomycosis infection is a rare infection affecting the pelvic organs and is seen to be strongly associated with a long-standing IUD use.^[1] Early detection of the infection not only significantly minimizes morbidity but also avoids unnecessary surgical intervention.^[2] Various forms of actinomycosis are known, most common being the cervicofacial form, (31–65%),

followed by thoracic (15–33%), abdominal (10–36%), and pelvic (approximately 3%) forms.^[3] Actinomycosis infection tends to be more common in men, however, the abdominopelvic form is 5 times more common in women, reason largely attributed to the use of IUDs for birth control.^[4,5] According to the literature, up to 70–80% of the patients infected with *Actinomyces* had used IUDs for a minimum of 5 years.^[6] Our case also had presence of long-standing IUD.

Actinomyces infection cannot cross an intact mucosal barrier, and therefore, its spread requires disruption of the normal mucosal barrier. In our case, the main culprit for mucosal barrier breakdown was a foreign body, i.e., IUCD. Once the mucosal barrier is breached, the bacteria incites a characteristic granulomatous response, resulting in abscess formation, necrosis, and reactive fibrosis.^[7]

A patient of pelvic actinomycosis usually presents with non-specific symptoms which include lower abdominal pain, palpable mass, and weight loss. Similar findings could be seen in our patient. Although the imaging findings of CT/MRI also tend to be variable and non-specific, they play a pivotal role in determining the extent of disease. CT/MRI of pelvic actinomycosis involving the uterus shows a bulky, enlarged uterus with ill-defined solid-cystic mass(es), having heterogeneous contrast enhancement. The lesions may infiltrate the surrounding mesenteric fat/organs and spread across tissue planes. There can be associated bowel wall thickening which is inflammatory in nature. On MRI, the lesions are usually hypointense on T1-weighted sequences and of intermediate signal intensity on T2-weighted sequences. Cystic areas appear bright on T2-weighted images. The entire gamut of clinicoradiological findings often leads to misdiagnosing pelvic *Actinomyces* as a neoplastic process. However, Actinomycotic infection should be considered in the three main clinical circumstances:^[8] Chronic disease process having extensive spread along tissue planes and mass-like appearance; drainage of an abscess by a sinus tract, which may close spontaneously or after the antibiotic course and improvement after course of antibiotic treatment.

Additional features like paucity of nodal enlargement and minimal to no ascites on imaging also favor the diagnosis of an infective process rather than a neoplastic entity. Pelvic actinomycosis shows avid FDG uptake, similar to a malignant neoplasm on FDG PET/CT,^[9] adding no additional value in differentiating pelvic actinomycosis from malignancy.

Final diagnosis of pelvic actinomycosis requires identification of sulfur granules on histopathological examination or culture of actinomycosis.^[1,10]

The mainstay of treatment for pelvic actinomycosis is penicillin along with removal of IUD.^[11] Other antibiotics such as ceftriaxone used in our case may be administered in cases not responding to penicillin or for patients allergic to penicillin. Surgical management is usually reserved primarily for drainage

of very large abscesses. Medical management alone can show dramatic response and marked improvement in patient's clinicoradiologic picture as evidenced in our case also.

CONCLUSION

The non-specific clinical as well as radiological picture in cases of pelvic actinomycosis often poses as a diagnostic challenge. However, in an appropriate clinical scenario, pelvic actinomycosis must be included in the differential diagnosis by a radiologist when encountering a large infiltrative pelvic mass extending along tissue planes, particularly in the setting of an IUD, with the absence of nodal enlargement and ascites. Medical management with antibiotics is the mainstay of treatment, with the role of surgical intervention limited to drainage of very large abscesses. Early identification of pelvic actinomycosis can ensure its prompt treatment with minimal complications.

TEACHING POINTS

1. Diagnosis of actinomycosis infection should be favored over uterine malignancy in view of paucity of lymph nodes and no/minimal ascites.
2. The mainstay for uterine actinomycosis is antibiotic therapy and not surgical management.
3. Cause of mucosal barrier disruption should be looked for in actinomycosis infection.

MCQs

1. Which of the following imaging findings favor diagnosis of pelvic actinomycosis over uterine malignancy?
 - a. Extensive lymphadenopathy
 - b. No/minimal ascites
 - c. No spread of disease along soft tissue planes

Answer Key: b

2. What is the treatment for pelvic actinomycosis?
 - a. Radical hysterectomy
 - b. Antibiotic therapy using penicillin
 - c. Antibiotic therapy using penicillin with IUCD removal
 - d. None of the above

Answer Key: c

3. What is the gold standard for diagnosis of actinomycosis infection?
 - a. Sulfur granules on histopathological examination
 - b. Culture of actinomycosis
 - c. CT/MRI imaging alone
 - d. a and b

Answer Key: d

Ethical approval

Institutional Review Board approval is not required/waived-off.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Conflicts of interest

There are no conflicts of interest.

Use of artificial intelligence (AI)-assisted technology for manuscript preparation

The authors confirm that there was no use of artificial intelligence (AI)-assisted technology for assisting in the writing or editing of the manuscript and no images were manipulated using AI.

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