Case Report

A slow growing pelvic actinomycoses related abscess in a premenopausal patient mimicking genito-urinary malignancy - Case report and literature review

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Abstract: Actinomycosis in the pelvic region is an uncommon diagnosis. This infection in most cases is caused by Actinomyces israelii, a gram-positive anaerobic saprophyte bacterium, although other Actinomyces pathogens have also been reported. This bacterium is a normal inhabitant of the upper intestinal and genital tracts in humans. Pelvic actinomycosis is difficult to diagnose, and in many cases the preliminary suspicion is of a neoplastic process in the pelvic organs, as this infection may mimic pelvic and abdominal malignancies. We report a rare case of a 52-year-old female patient with a fixed pelvic mass within the vesico-vaginal fascia, located between the urinary bladder, uterine cervix and anterior vaginal wall. The diagnosis and treatment of pelvic actinomycoses related inflammatory disease are discussed in this report. Care providers should be aware of this rare infection that is slow growing and may mimic a malignant process, leading to morbidity that can be caused by unnecessary treatment.

Keywords: Actinomyces Israeli; Pelvic abscess; Intrauterine device; Infection.

INTRODUCTION

Actinomyces is a gram positive, non-spore-forming anaerobic microaerophilic rod. Actinomyces israelii causes most Actinomyces infections in humans, although other forms such as Actinomyces Odontolyticus, Actinomyces Viscosus, Actinomyces Meyeri, Actinomyces Gerencseriae, and Propionibacterium Propionicum have also been reported to cause infections in humans. Actinomyces israelii is a part of oral and genital tract flora with infections being reported in the oral-cervicofacial, thoracic, abdominal, pelvic, central nervous system, musculoskeletal regions as well as causing disseminated disease. Actinomycoses infections may also be polymicrobial, although in our case a single agent was suspected to cause the infection. We describe a rare case of a slow growing pelvic abscess, mimicking a uterine cervix malignancy in a premenopausal woman. The patient's history was remarkable for a longstanding neglected intrauterine device (IUD) of 15 years, which was removed two years prior to the diagnosis of an infection with Actinomyces.

CASE REPORT

A 52-year-old woman married and mother of three, was referred to the internal medicine emergency room (ER) due to severe abdominal pain and suspected urinary tract infection with bilateral moderate hydronephrosis that was diagnosed by ultrasound from an outpatient clinic.

During her evaluation in the ER the patient was found to have an elevated C-reactive protein (CRP) – 163 mg/L, leukocytosis 19.14 x 10^3/ul and neutrophilia 14 x 10^3/ul. The urinary culture which was obtained two weeks earlier was sterile.

She was admitted to the hospital and an abdominal computerised tomography (CT) scan revealed bilateral hydronephrosis and a cystic or necrotic space occupying lesion located between the uterine cervix and urinary bladder, anterior to the vaginal wall. A thick and enhanced bladder wall was noted, accompanied by significant stranding of the adjacent fat. The origin of the lesion was uncertain, raising possibility of a genito-urinary malignancy (Fig. 1).

With these findings the patient was sent for a gynecological consultation. She complained of lower urinary tract symptoms consistent with mixed urinary incontinence including stress incontinence, nocturia, frequency, urgency and dysuria that lasted for the past two months. No other complaints were reported by the patient. Her medical history was uneventful.

A gynecological transvaginal ultrasound examination demonstrated a non-cystic mass, which was also palpated through the anterior vaginal wall and measured 39x44 mm. It was mostly hypoechoic with hyperechogenic foci and surrounded by enhanced peripheral blood flow. However, the origin of the lesion was unclear. She was admitted to the gynecological ward. The vital signs were unremarkable: blood pressure 140/78 mm/Hg, temperature: 37.1°C, heart rate: 72 bpm. The laboratory findings were as follows: hemoglobin: 11.7 g/dl, white blood cells: 19.14 x 10^3/ul, neutrophils: 14.17 x 10^3/ul, C-reactive protein: 163.7 mg/dl. Other findings including the tumor markers such as CEA, CA19-9, CA15-3, CA 125, were within normal limits.

Due to these findings the patient underwent a gynecologic oncologist consultation including a colposcopy. The uterine cervix appeared to be normal.

Figure 1. – The CT scan revealed necrotic mass (*) between urinary bladder (B) and uterine cervix (U), surrounded by fat stranding. The bladder wall is markedly thickened.
Further evaluation included a cystoscopy which showed normal mucosa of the urinary bladder with no signs of malignant invasion. However, extrinsic compression probably due to the pelvic mass was noticed.

An abdominal MRI was performed. The study showed similar findings to those demonstrated on CT and ultrasound of a necrotic pelvic mass, localized in the vesicovaginal fascia, between the urinary bladder, uterine cervix and anterior vaginal wall, in a 52-year-old patient that two years previously had a neglected IUD removed.

The genus Actinomyces consists of several species such as Actinomyces Odontolyticus, Actinomyces Viscosus, Actinomyces Meyeri, Actinomyces Gerencseriae, and Actinomyces Israelli – which is the most common isolate in human disease. These bacteria are gram-positive, non-spore-forming bacteria, most of which grow at anaerobic conditions and tend to form branching filaments.

Actinomycotic disease is a rare diagnosis especially in developed countries. The overall annual incidence being 1:100,000 to 1:300,000, and due to unknown reasons, the rate is threefold higher in men. Most cases are seen in adolescents and middle-aged adults.

Actinomycotic infection may be associated with IUD usage. Chatwani and Amin - Hanjani 3 in their study including 1,520 women with IUD, showed that the colonization rate increases with the duration of IUD use, reaching an overall colonization rate of 11.4%. The authors suggested that patients with IUD should undergo annual cytological smears. Likewise, other data show that the incidence of IUD-associated pelvic actinomycosis is 1.65% to 11.6% of IUD users and infection is more common in women who have had an IUD for more than four years.

Actinomyces israelii is a saprophyte found in various organs and in particular situations such as in mucosal lesion or in areas with a low oxygen level it becomes pathogenic. This

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Figure 2. – A sagittal T2 weighted MRI shows the abscess (*) between urinary bladder (B) and uterine cervix (U). Uterine cervix is unremarkable.

Figure 3. – CT scan obtained during imaging guided abscess drainage showed the needle placed into the abscess (*), avoiding damage of the rectum and major pelvic neuro-vascular structures.
condition allows the penetration of actinomyces through the mucosa and initiates an inflammatory process leading to the formation of abscesses and pseudo tumors. The abscesses tend to grow slowly and become symptomatic when they apply pressure on adjacent structures, form fistulas or perforate.

Apart from IUD, some predisposing events for abscess formation may include previous bowel surgery and endoscopic manipulation, perforated gastric ulcers, loss of gallstones after laparoscopic cholecystectomy, trauma, diverticulitis, pancreatitis and immunocompromised patients.

Actinomyosisc abscess has been reported to affect the colon, ileum, ovaries, vulva, liver, abdominal wall, pancreas, greater omentum, retroperitoneum, kidney and abdominal wall. However, to the best of our knowledge, this is the first report of an actinomyces abscess in the vesicovaginal fascia or the uterine cervix.

The disease is characterized by an infiltrative and granulomatous inflammation, which may result in multiple abscesses, and sinuses that contain Sulphur granules. However, this is not always a pathognomonic sign, because other species such as Staphylococci, Nocardia, Aspergillus and Streptomyces can also form Sulphur granules.

The differential diagnosis, in patients who present with an abdominal or pelvic mass, depends on the patient’s gender and age and includes: ovarian lesions, leiomyoma, ectopic pregnancy, pelvic inflammatory disease, appendicitis, diverticulitis, inflammatory bowel disease, and tuberculosis. A diagnosis of a malignant tumor of various structures is also frequently made. The ileocecal site is the most frequently affected in patients with abdominal actinomycosis. The involvement of the vesicovaginal fascia located between the urinary bladder, uterine cervix and anterior vaginal wall, in this case made a diagnosis of genito-urinary malignancy even more reasonable.

Imaging findings of actinomycosis may vary greatly. Lesions may be single or multiple, and they may look like tumors, sometimes with foci of necrosis, or may present as abscesses. This behavior of the pathogen makes it very challenging to reach the correct diagnosis prior to an invasive intervention and as most cases of actinomycosis are based on tissue diagnosis.

Preoperative diagnosis aided by aspiration or biopsy of the lesion as was done in our case, may avoid surgical intervention and unnecessary morbidity in patients affected by actinomycosis. Actinomyces is a slow growing pathogen and in many cases the bacteriologic analysis is uninformative. In our case there were no positive cultures from aspirated pus, however histopathologic findings, which were positive for branching filaments consistent with Actinomyces infection and Actinomyces like bacterial colonies, as well as a history of neglected IUD helped us in reaching the correct diagnosis.

In such cases an antibiotic treatment rather the operative approach is required.

The cornerstone of the treatment for actinomycosis is prolonged antimicrobial treatment with penicillin. The recommended antibiotic regimen is Penicillin G (18-24 million units/day) for 2-6 weeks. After several weeks of parenteral penicillin treatment, a prolonged treatment with amoxicillin for a 6-12-month period is advised, with or without surgical drainage for bulky disease.

Where penicillin allergy exists, treatment with tetracycline, clindamycin, or doxycycline has been reported.

In cases, when there is no benefit, and general health conditions continue to deteriorate after a week of antibiotic treatment or it is not possible to exclude a potential malignancy, surgery may also be required.

In the case presented here, the combination of a long-standing IUD heavily colonized with Actinomyces, together with a probable mucosal damage caused by the traumatic extraction of an IUD, gave the pathogen an ability to cause an infection outside the uterine cavity. The long interval between IUD removal and the development of symptoms, points out to the unique infectious clinical syndrome caused by this organism. This highlights the importance of thorough history taking and specific questioning regarding long standing IUD presence in all patients with unexplained sub-acute symptoms and abnormal findings on pelvic imaging. The issue of whether to treat asymptomatic women with a histological finding of actinomyces colonies in the presence of IUD is still debatable, but in the presence of symptoms of endometritis – IUD removal and a short course of penicillin treatment is recommended. In our case, the patient had no clinical symptoms of endometritis at the time of IUD removal and the first evidence of actinomyces infection appeared two years later. We believe that antibiotic treatment given in the presence of actinomyces to prevent deterioration to a more serious infection is still a matter of clinical judgment, taking into account the time elapsed from the insertion of the IUD, and the traumatic potential of the extraction procedure.

In fact, pelvic actinomycosis is a rare chronic suppurrative disease caused by Actinomyces israelii and related species, that may result in an infiltrative mass lesion, which is often indistinguishable from malignancy at the beginning of the patient’s evaluation.

In the majority of cases, a high index of suspicion is needed in order to reach a diagnosis. The diagnosis should be supported by a thorough anamnesis, imaging, bacteriology and histopathological findings, as in some cases only imaging and bacteriological findings are not enough.

The most challenging task for the management of actinomycosis is to reach a correct diagnosis before surgical intervention, as an accurate preoperative diagnosis is rare. However, if the diagnosis is promptly achieved, the gold standard treatment is based on prolonged high-dose antibiotic therapy, in particular penicillin, rather than a surgical approach.

DISCLOSURE STATEMENTS

There was no conflict of interest, informed patient consent was obtained.

REFERENCES


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