

Case Report

Pelvic actinomycosis mimicking an advanced ovarian carcinoma: a rare case report with review of literature

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ABSTRACT

Actinomycosis is a rare subacute to chronic granulomatous bacterial infection caused by actinomyces species. Pelvic actinomycosis is rare, often pretends to be malignant and therefore it is difficult to diagnose preoperatively. Here we discuss a case of a 45-years-old female who was operated for a pelvic mass thought to be malignant, but on histopathological examination it turned out to be a case of ovarian actinomycosis.

Keywords: Pelvic mass, Ovarian actinomycosis, Pelvic actinomycosis

INTRODUCTION

Actinomycosis is a rare subacute to chronic granulomatous bacterial infection caused by actinomyces species.^{1,2} These are gram positive, non-acid fast anaerobic bacteria normally constituting the human flora in the oropharynx, gastrointestinal and genital tract.³

Pelvic actinomycosis constitutes 3% of all human actinomycosis infections and occurs almost only in women.^{2,4} It may simulate pelvic malignancies or retroperitoneal tumours which often makes it difficult to diagnose preoperatively.²

Here, we report a case in which pseudotumor and pelvic masses resulting from actinomycosis were initially suspected to be malignant.

CASE REPORT

A 45-years-old female presented to the surgery department with 6 months long persistent, progressively worsening

lower abdominal pain. This was severe in nature, predominant in the right flank and right iliac region lasting for a few seconds and resolving spontaneously.

On physical examination she was afebrile with right flank and right iliac region tenderness with no rigidity or guarding. Pelvic examination revealed a tender, solid, fixed pelvic mass extending to the right pelvic side wall. Ultrasound scan revealed a right adnexal solid-cystic mass measuring 6.8×4×3 cm with masses in the infra-colic omentum and wall of the small bowel. Routine investigations showed microcytic hypochromic anaemia with leucocytosis (12.2×10⁹/l) with raised inflammatory markers (C-reactive protein 106 mg/l). Clinical diagnosis thought at this time was disseminated carcinoma with primary thought to be the right ovary.

Tumour markers were also largely normal, with only a slightly elevated CA-125 (65 IU/ml). The patient was operated, laparotomy revealed a right adnexal mass lesion with attached infracolic omentum and small deposits on small bowel, and again it was considered to be a malignant.

Tissue was sent for histopathological examination. We received a specimen of right adnexal mass measuring 7×4×3 cm, part of infracolic omentum measuring 3×3 cm and small deposits from the small bowel measuring 1.5×1 cm and left side salpingo-oophorectomy specimen (Figure 1).

Extensive sampling of the right adnexal mass showed numerous bacterial colonies showing radiating filaments with hyaline, eosinophilic club like ends, surrounded with dense mixed inflammatory cell infiltrate, giant cells, necrosis with small fragment of ovarian stroma (Figure 2). Sections from the infra-colic omentum and small bowel deposits showed similar microscopic findings. The diagnosis offered was ovarian actinomycosis with peritoneal spill. Her post-operative period was uneventful and was treated with antibiotics.

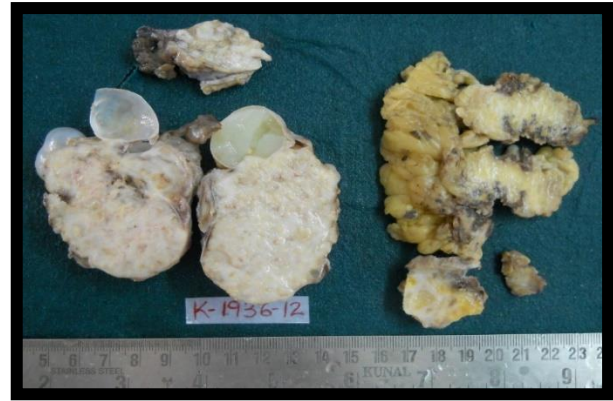
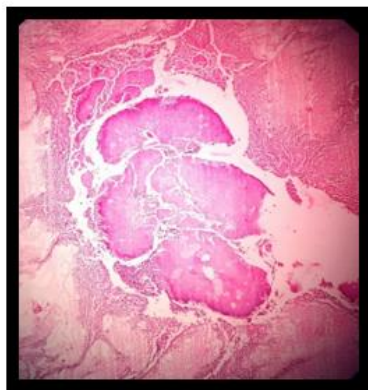
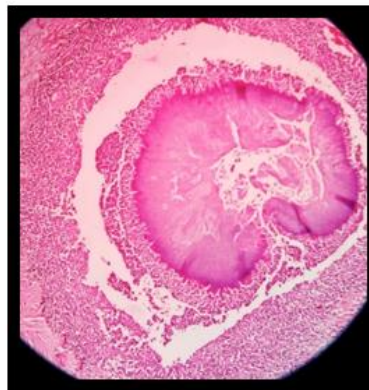


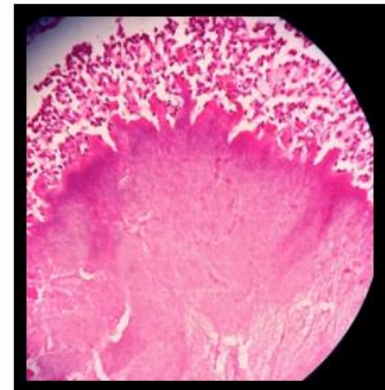
Figure 1: Grey-white solid-cystic mass from the right side, part of infracolic omentum, nodule from the wall of small bowel and left side salpingo-oophorectomy.



40x



100x



400x

Figure 2: Scanner, low power and high-power view showing bacterial colony showing radiating filaments with hyaline, eosinophilic club like ends, surrounded with dense mixed inflammatory cell infiltrate with foci of ovarian tissue also noted.

DISCUSSION

Human actinomycosis was first described in 1878 by Israel and Wolff who first isolated actinomyces in culture.⁴ Actinomyces are closely related to Nocardia species, and both were once considered fungi because of their branching filaments but are currently classified as bacteria.⁵ Actinomycosis in humans is most commonly caused by the gram positive, non-acid fast, filamentous bacteria, *A. israelii*. These organisms are not considered particularly virulent pathogens, but rather as opportunistic ones, because infection usually occurs only after disruption of the mucus membrane and their ability to secrete proteolytic enzymes, disrupt tissue planes and compress surrounding tissue makes their appearance similar to a malignant process.^{3,6-8} Based on anatomical site of infection they are classified into the cervicofacial region, abdominopelvic region, thoracic region and might involve the central nervous system (CNS).^{4,6} Pelvic actinomycosis constitutes 3% of all human actinomycotic

infections. *A. israelii* infects 1.65% to 11.6% of intrauterine device (IUD) users and is more common in women who have had an IUD use in situ for more than four years.⁶

The presence of the IUD strings is thought to allow ascension of the Actinomyces spp into the uterus where focal necrosis of the endometrium by the IUD is the breach required for disease to occur.³ Papanicolaou smears are useful for the evaluation of patients with pelvic actinomycosis associated with an IUD.^{1,9} Some researchers have reported that 53-80% of women who had Actinomyces on Papanicolaou smears actually had symptoms.⁹ The patient may present with non-specific symptoms such as fever, weight loss, abdominal pain, abdominal mass and are often misdiagnosed as a gynaecological malignancy.^{9,10}

Ovarian actinomycosis is rarer because the structure of the ovary is resistant to surrounding inflammatory disease. It

has been assumed that bacteria enter the ovary when its surface is broken by the process of ovulation.^{8,9}

The diagnosis of pelvic actinomycosis is challenging with only 10% of cases diagnosed preoperatively.³ Ultrasound and CT scan are the most commonly used imaging modalities; however, the findings are usually nonspecific therefore do not help much. CT findings in women with abdominopelvic actinomycosis show predominantly solid masses with focal areas of reduced attenuation or thick-walled cystic masses.¹¹ Magnetic resonance imaging (MRI) shows relatively low signal intensity on T2 weighted sequences in association with extensive pelvic infiltration that is atypical for a malignant ovarian tumour.³ Thus, the diagnosis is done on pathological, serological, and bacteriological examinations.⁸

Abdominal actinomycosis makes up 20% of the cases.¹⁰ It is the most indolent and presents with variable clinical findings depending on the primary site of involvement and the duration of the disease. Virtually all organs have been reported to be compromised, appendix and colon are the most commonly involved intrabdominal organs and the exact explanation for the route of infection for these organs remains controversial.^{5,11} Patients who have had acute appendicitis particularly with perforation accounts for 65% of the cases, other predisposing factors include gastrointestinal perforation, previous surgery, neoplasia, and foreign bodies in the gastrointestinal tract or genitourinary tract, with or without erosion through the mucosal barrier.¹⁰ Nonspecific clinical and radiological findings makes the diagnosis difficult.

Various studies have reported the unusual presentations of actinomycosis. Ponfick in 1882 reported the first case of actinomycosis in the CNS.¹² Ravindra et al in 2018 reported 17 cases of CNS actinomycosis. Less than 5% of Actinomyces infections involve the CNS and are reported to occur as brain abscess, meningitis/meningoencephalitis and actinomycetoma in 67%, 13% and 7% respectively.⁴

Raymond in 1987 stated actinomycotic CNS lesions most frequently resulted from primary sources of infection in the lungs (27%) or the cervicofacial region (20%). Abdominal and pelvic primary foci were less common.¹² Risk factors include dental caries, recent tooth extraction, head trauma, gastrointestinal tract surgery, chronic otitis, mastoiditis, sinusitis, chronic osteomyelitis.¹²

Musculoskeletal infections are usually caused by spread from adjacent soft tissue (75% of cases), but can also be from local trauma (19%) or haematogenous spread (3%). The facial bones, especially the mandible, are the most common sites of bone disease.¹⁰ Diagnosis of actinomycosis is difficult because of the insidious nature of the infection and the non-specific symptoms.^{6,7} On microscopy there are clusters of bacteria, producing the classical macroscopic appearance of yellow 'sulphur granules': rounded basophilic masses with eosinophilic terminal clubs on staining with H and E. This 'sulphur

granule' appearance is not unique to Actinomyces species, but can be differentiated from other bacterial infections with a similar appearance by the observation of filamentous branching bacteria at the periphery on Gram, Gomori-methanamine-silver and Giemsa stains. Gram stains are usually more sensitive than culture, which can be insensitive in up to 76% of cases as Actinomyces spp is a fastidious anaerobic bacterium that requires 2-3 weeks to culture with a failure rate of more than 50%.^{3,7} Therefore, most diagnoses are made histologically by direct isolation of the organism from pus, tissue or from sulphur granules. However, the failure rate of isolation is high (>50%) for various reasons including previous antibiotic treatment, overgrowth of concomitant organisms, or inadequate methodology.¹⁰

Prolonged courses of antibiotic therapy are the cornerstone of treatment for actinomycosis. In vitro studies show sensitivity of actinomycetes to a wide range of antimicrobials, although they are resistant to cephalexin and metronidazole. Penicillin is a common first line agent for the treatment of actinomycosis and the development of penicillin-resistance is low. Antibiotics are typically given for at least 3 months, although complete resolution has been reported with shorter courses. Surgical excision of necrotic tissue and drainage of abscesses may also be beneficial, particularly in the context of refractoriness to medical therapy. Resolution of infection should be confirmed by repeat CT scan.⁷

CONCLUSION

Surgeons should always consider pelvic actinomycosis as a differential in patients with pelvic mass especially in those using IUD and who have a history of appendectomy, tonsillectomy or dental infection to avoid unnecessary surgery and delay in appropriate treatment.

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