



## Xanthogranulomatous salpingo-oophoritis with actinomycosis mimicking tubo ovarian tumor: a case report with review of literature

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### ABSTRACT

*Female genital Actinomycosis is relatively rare, although strongly related to long-lasting intrauterine contraceptive device (IUCD) application. Pelvic actinomycosis is difficult to diagnose pre-operatively and is diagnosed, in most cases, postoperatively on histopathological examination. Actinomycosis can mimic pelvic and abdominal malignancies.*

*A case report of a 25-year-old female patient with history of IUCD uses having tubo-ovarian mass is presented and the diagnoses of pelvic actinomycosis in relation to ovarian cancer are discussed. Clinicians must be aware of this rare association to spare women from this potential morbidity and its complications. Though not confirmatory, it may be possible, to diagnose actinomycosis on Papanicolaou smears. However, the finding of sulphur granules on histopathology from the sites other than the tonsils is considered pathognomonic. We suggest that routine and follow up cervical examinations are important for women who are IUCD users.*

**KEYWORDS :** Tubo-ovarian mass, intrauterine contraceptive device, Actinomyces.

### Introduction:

Actinomyces israeli, a filamentous, gram-positive bacillus, is a part of the microflora of the human oral cavity, gastrointestinal tract and vagina. Actinomyces israelii cannot penetrate through the normally intact mucosal barrier. [1,2] Pelvic actinomycosis is uncommon and often presents as a complication of an intrauterine contraceptive device (IUCD). Infection ascends from bacterial colonization of the cervix and ascends to the uterus, fallopian tubes, and the ovary. [2,3,4] Failure to diagnose Actinomycosis at an early stage may lead to tuboovarian abscess (TOA) and is often mistaken for other conditions such as diverticulitis, inflammatory bowel disease and malignant tumor, presenting a diagnostic challenge pre-operatively.[2,5] It is identified post-operatively on histopathology by the identification of characteristic sulphur granules in most cases.[4,5]

### Case history:

We report a rare case of tuboovarian mass in a 25 year female patient with para 2, who presented with pain in lower abdomen and weight loss for 4 months and history of IUCD use for 1 year. USG examination showed heterogenous lesion in left adnexal region measuring 12x9x6 cm with few cystic areas. CA-125 level was 170 U/L. A left salpingo oophorectomy was done and specimen sent for histopathological examination.

Gross examination showed a distorted globular tissue piece measuring 9x5x4 cm. Cut section was solid, creamish white in appearance with few small cystic and haemorrhagic areas. [Fig.1] Hematoxylin & Eosin section showed fibrotic tissue, macrophages, plasma cells and sulphur granules of Actinomyces [Fig. 2a & 2b]. Periodic Acid Schiff stain (PAS) was positive for these colonies [Fig. 2c]. Figure 2d showed necrotic and inflamed ovarian tissue with destroyed fallopian tube lining. AFB stain was negative. A diagnosis of Xanthogranulomatous Salpingo-oophoritis with Actinomycosis was made .The Patient was treated accordingly. She is on follow up and now improving in terms of clinical features and CA-125 level.

### Discussion:

Actinomycosis is a chronic suppurative granulomatous disease caused by anaerobic organisms from the genus *Actinomyces*. Although previously thought to be a fungal infection, these organisms are Gram-positive, filamentous, nonspore forming, anaerobic or microaerophilic bacilli. [6,7] These organisms are not considered particularly virulent, but rather as opportunistic ones, because infection usually occurs only after disruption of the normal mucous membranes. The disease spreads by direct extension into surrounding tissues regardless of tissue planes through the formation of sinus tracts that can

lead directly to the skin. Rarely, sulfur granules can drain through these tracts. [5, 8] Actinomycosis is usually associated with other bacterial types as well, which contribute by elaborating toxins, enzymes and/or by inhibiting host defenses. Thus, these copathogens increase the invasiveness of *Actinomyces*, and may cause treatment failure. After local establishment of actinomycosis, a slowly progressive hematogenous spread is possible. [6, 8] Most frequently affected locations include: cervicofacial (55%), thoracic (20%) and the pelvic region (15%).

Disease in humans is most commonly caused by *A. israelii*, which is even considered as a common commensal of the oropharynx, the gastrointestinal tract, and the vagina. [7,8] *A. israelii* infects 1.65% to 11.6% of IUCD users, and infection is more common in women who have had an IUCD use in situ longer than four years.[4,5] However, in this case there was a history of IUCD use for 1 year only. The infection spreads by contiguity often mimicking the characteristics of a malignant neoplastic process. [5, 7]. In this case also, considering the large tubo ovarian mass on USG, clinical features and high CA-125 level, a provisional diagnosis of ovarian tumour was made and salpingo oophorectomy was done. However, histopathological examination showed presence of sulphur granules which was PAS positive, thus, confirming the diagnosis of Actinomycosis. [Figure 2,3,4]

When pelvic actinomycosis occurs, it usually causes endometritis, salpingo-oophoritis, or tubo-ovarian abscess and a solid mass in the adnexal region might be palpable, suggesting a pelvic malignancy. Ultimately, extension to the abdominal wall or deep pelvic structures can occur, complicating the situation further. [5,9,10].

Diagnosis of actinomycosis is difficult because of the insidious nature of the infection. Usually, diagnosis is difficult pre-operatively, even after fine-needle aspiration cytology [11,12]. It can be confirmed by culture. However, it is often difficult to culture *Actinomyces*. In fact, the detection rate of *Actinomyces* in patients with pelvic actinomycosis is as low as 2%. [13] The finding of sulfur granules from any other site than the tonsils is considered pathognomonic [5, 12]. Similarly in this case, presence of sulphur granules along with plasma cells and inflammatory infiltrate, ruled out malignancy. Fiorino reviewed the literature in 1996 and found 92 cases in 63 reports describing actinomycotic pelvic inflammatory disease simulating ovarian cancer. [11] Although rarely diagnosed, it's important to consider it in the differential diagnosis of ovarian cancer. [14]

Some authors raise the importance of Papanicolaou smears in the early diagnosis of Actinomyces who are IUCD users. [11, 15] But, the

late presentation along with complications and tubo ovarian mass mimicking malignancy, often leads to surgical interventions. We suggest that routine and follow up Papanicolaou smear examinations are important for females who are IUCD users. In a patient who is an IUCD user, sudden appearance of symptoms such as abdominal pain, weight loss along with a positive Papanicolaou smear for Actinomyces raises the possibility of Pelvic actinomyces. A response to antibiotics along with the removal of IUCD can be an early approach to management, preventing surgical interventions. [8,15]

### Conclusion

Pelvic actinomycosis should always be considered in patients with tubo ovarian mass especially in those using IUCD. Presence of sulphur granules on histopathology from tubo ovarian mass is considered pathognomonic. Surgeons must be aware of this rare association to spare women from its potential morbidity and complications. A routine and follow up papanicolaou smear examinations is promising to some extent in early diagnosis, in case of Chronic IUCD users.



Figure 1 (Gross): Tubo ovarian mass, cut section creamish white.

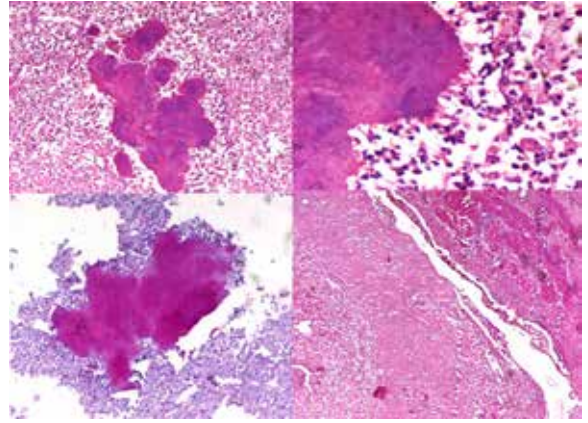


Figure 2a (10 X): sulphur colonies surrounded by macrophages & plasma cells.

Figure 2b (40 X): sulphur granules of Actinomyces.

Figure 2c (PAS): PAS positive sulphur colonies of Actinomyces.

Figure 2d (40 X): necrotic and inflamed ovarian tissue with fallopian tube lining.

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