# Pelvic actinomycosis in a postmenopausal patient with systemic lupus erythematosus mimicking ovarian malignancy: case report and review of the literature

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**Summary**

Pelvic actinomycosis is a chronic granulomatous suppurative disease caused by an anaerobic gram-positive organism *Actinomyces israelii* usually associated with intrauterine devices. Systemic lupus erythematosus is an autoimmune disorder associated with multiple primary and drug-related immunological defects that predispose patients to infections. The combination of both diseases in a postmenopausal patient is a rare occurrence.

A case of a pelvic mass in a 49-year-old postmenopausal patient with systemic lupus erythematosus treated with immunosuppressive therapy for two years is presented. The patient presented with lower abdominal pain to the gynecology clinic and was found to have a pelvic tumor. She had no history of intrauterine device use. Histopathologic examination of the laparotomy specimen revealed pelvic actinomycosis.

*Key words:* Actinomycosis; Systemic lupus erythematosus; Ovary.

## Introduction

Pelvic actinomycosis is a chronic granulomatous suppurative disease usually caused by an anaerobic gram-positive organism *Actinomyces israelii* [1-4]. The other related pathogenic organisms are *Actinomyces bovis, A. ericksonii, A. naeslundii, A. viscosus, A. odonlyticus.* The prevalence of human actinomycosis is estimated to be about 5/100,000 [5]. Pelvic actinomycosis is usually associated with an intrauterine device (IUD) and constitutes 3% of all human actinomycosis infections [6-9]. It classically manifests itself as a unilateral tubo-ovarian abscess [9] however, in many cases it presents as a bilateral ovarian tumor or retroperitoneal mass [11-14]. Pelvic actinomycosis can mimick pelvic or intra-abdominal malignancy [15-17].

We present a case of pelvic actinomycosis in a postmenopausal patient with systemic lupus erythematosus without any history of intrauterine device usage and discuss the treatment strategy.

## Case

A 49-year-old urban woman, married, Caucasian, gravida 2 para 2, was admitted to the gynecologic unit of our university hospital for pelvic pain and a pelvic tumoral mass discovered at abdominal sonography. She had never used an intrauterine device for contraception and had been in menopause for two years. She had not been having regular gynecologic examinations and a Papanicolaou smear had not been performed. On admission she was complaining of asthenia of one month's duration, moderate pelvic pain and a subfebrile fever especially at night. No intestinal or urinary disorder was noted. Two years before she had been admitted to the university rheumatology clinic for arthralgia. After clinical evaluation she was started on steroid therapy for systemic lupus erythematosus. During the course of the disease she was also administered additional immunosuppressive therapy including cyclophosphamide and azathioprine. Urinary examination revealed active sediments and proteinuria. Abdominal sonographic examination revealed bilateral grade 2 hydronephrosis. The complement 3 level was lower than normal and antinuclear antibody was 1/280 positive and granular in type. Pathologic examination of the renal biopsy specimen revealed diffuse proliferative lupus nephritis.

The general physical examination showed a malar rash and the patient complained of diffuse arthralgia. Her body temperature was 36.8°C on admission but during the observation period it was recorded as 38.0°C. Except for mild tenderness, no abdominal mass or evidence of peritonitis was noted on abdominal examination. On bimanual recto-vaginal digital examination the uterus was palpated as normal in size, however, the bilateral adnexal region and rectovaginal pouch seemed to be filled with a smooth tumoral lesion. Cervical motion tenderness was notable.

Biochemical findings showed an inflammatory process with a leukocyte count of 20,400/mm$^3$, anemia with a hemoglobin level of 8.8 g/dl, and a sedimentation rate of 48 and 105 mm at one-hour and two-hours, respectively. The vaginal sonographic examination showed a bilateral heterogeneous adnexal mass 60 x 50 mm in diameter without ascites.

A malignancy process could not been ruled out. During 24 hours of observation and laboratory evaluation, the patient's clinical condition deteriorated with worsening of pelvic pain and remarkable changes in abdominal examination findings. The patient was scheduled for exploratory laparotomy. During the procedure the abdominal cavity was observed to be full of

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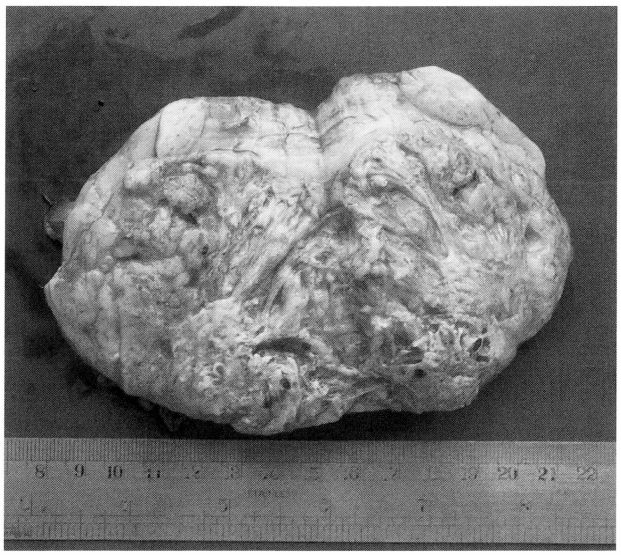

Figure 1. — Macroscopic appearance of the right ovarian mass.

purulent fluid, to have a normal sized uterus and bilateral ovarian abscesses of approximately 3 x 4 cm and 4 x 5 cm each with dense adhesions to the pelvic side wall, sigmoid colon, anterior rectal wall, posterior uterine wall and the pouch of Douglas.

After extensive adhesiolysis, a total abdominal hysterectomy with bilateral salpingo-oophorectomy was performed with difficulty. The intraoperative frozen section pathologic examination confirmed a chronic inflammation process without any evidence of malignancy. Purulent fluid was collected for culture for aerobic and anaerobic media which was later reported to be negative for *Actinomyces israelii.* Figure 1 demonstrates the macroscopic appearance of the right ovarian mass. Figure 2 demonstrates the microscopic appearance of chronic endosalpingitis. Figure 3 demonstrates the typical microscopic appearance of sulphur granules. In the postoperative period, the patient received empiric therapy with intravenous teicoplanin 1 gram/day for one week for polymicrobial coverage. After the definitive histological examination confirmed the diagnosis of bilateral actinomycosis salpingo-ovarian abscess, the patient's therapy was replanned and she was scheduled to receive 3 gram/day ampicillin parenterally for one month followed by the same dose orally for six months. Estrogen replacement therapy was initiated. The patient was discharged on the 15th day after surgery without any postoperative complications.

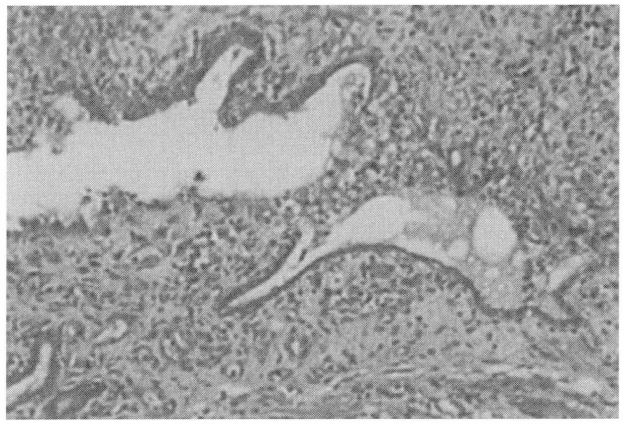

Fig. 2

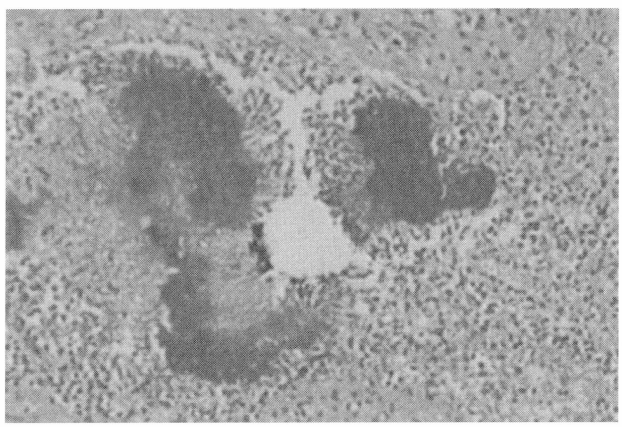

Fig. 3

Figure 2. — Microscopic appearance of chronic endosalpingitis (H&E x 10).

Figure 3. — Typical microscopic appearance of sulphur granules (H&E x 10).

## Discussion

Pelvic actinomycosis is a chronic granulomatous suppurative disease usually caused by an anaerobic gram-positive organism *Actinomyces israelii.* Several studies have reported the high frequency of *Peptostreptococcus* and *Bacteroides fragilis* associated with *Actinomyces israelii* infection [1-4]. Although it was reported by some authors [18] to be a rare disease whose pathogenesis is poorly understood, pelvic actinomycosis should be considered in the differential diagnosis of a pelvic mass. Extensive disease may lead to complications like visceral obstructions which result in a high morbidity [19-22]. Nasu *et al.* [19] reported a case of pelvic actinomycosis involving the uterus, sigmoid colon, urinary bladder, and right ureter. In their report, right hydronephrosis and hydroureter were recovered successfully after surgical and medical therapy. Delay in surgical and medical therapy may result in the permanent sequelae of the involved organ [21, 22]. Bercovich *et al.* [21] reported a case of ureteral obstruction secondary to pelvic actinomycosis which was not released despite stenting, prolonged antibiotic therapy, and debridement. The patient later underwent ureteral resection and reconstruction.

The most difficult task in the management of pelvic actinomycosis is to reach a diagnosis before a surgical approach. In many cases the diagnosis of a pelvic abscess is obtained during surgical exploration. The detection of *A. israelii* by Papanicolaou smears has been reported [23, 24]. The isolation of *A. israelii* from IUD or bacteriological samples by routine bacteriological cultures of anaerobic organisms is usually difficult because of its exclusive anaerobic nature and a slower growth rate compared to other anaerobes [27]. Moreover, actinomycetes belong to the normal flora of the female genital tract. Therefore the identification of these organisms in the vagina or cervix by any laboratory technique, including.

Papanicolaou smears, immunofluorescence or cultures are not diagnostic or predictive of any disease [18]. When

preoperative diagnosis is not certain, diagnosis can be established after careful histologic examination of specimens obtained from hysterectomy and bilateral salpingo-oophorectomy [19, 25]. High suspicion and a timely thorough evaluation by a nonsurgical approach followed by prolonged antibiotic therapy may eliminate the need for extensive surgical surgery and may help in maintaining future fertility. Tru-cut needle biopsy [29] and transcutaneous computed tomography-guided core needle biopsy [30] of an abscess seem to be more accurate in isolating *A. israelii* than bacteriological samples. Radiologic studies are able to identify the presence of a mass but are not able to discern malignancy from a pelvic abscess or other tumoral lesions [19, 26-28].

The correct time-scale for antibiotic therapy is still unknown. Some authors report that at least one month is necessary to clear most of the infection and inflammation from the pelvic cavity [11, 13, 27, 28]. This is essential to minimize the risk of bowel or urinary injuries during exploratory surgery. Penicillin is the antibiotic of choice for treatment of actinomycosis infection whereas metronidazole seems to be the most successful in treating the associated infection of anaerobic gram-negative organisms [15]. Antibiotic treatment for actinomycosis has also been reported in pelvic abscesses in the absence of peritonitis [16], but a surgical approach is almost always necessary [15-27]. This is usual when pain is still present or if there is obvious sepsis even after appropriated antibiotic therapy or evidence of peritonitis. Similarly, the presence of compression on the urinary tract or bowel is an indication for surgical treatment. Even in asymptomatic patients the low possibility of malignancy or the potential risk of recolonization by other organisms is one more element in favor of surgical treatment.

Preoperative antibiotic therapy seems to be the best therapeutic strategy in developed countries with high-level medical care. In underdeveloped countries with poor medical care it may be difficult to obtain benefit from preoperative antibiotic therapy for one month. Otherwise, the very high morbidity of an initial surgical approach and the efficiency of penicillin therapy, which is a basic and cheap antibiotic, indicates the advantage of preoperative antibiotherapy. Evidence strongly suggests that removal of the intrauterine contraceptive device of a patient with a positive culture is not necessary and that, in the absence of evidence of pelvic infection, antibiotics are not required [18].

Theoretically, bilateral salpingo-oophorectomy could be sufficient but the intensity of pelvic inflammation, multiple ovarian abscesses and endometriosis indicate, in the majority of cases, more extensive surgery consisting of both bilateral salpingo-oophorectomy and total hysterectomy. Debulking surgery is associated with a very high risk of perforation to nearby structures especially the bowel, ureter, and bladder [9-17].

A laparoscopic approach would initially indicate unilateral salpingo-oophorectomy with an intraoperative histological examination to eliminate the possibility of malignancy and then a transverse laparotomy to clear the pelvic cavity. Even after appropriated antibiotic therapy, laparoscopic treatment is difficult because of the possibility of extensive inflammation and dense adhesions.

The optimal treatment for pelvic actinomycosis without perforative complications should consist of high-dose intravenous antibiotic therapy with penicillin and metronidazole continuing up to normalization of biological parameters. Then, consolidation enteral antibiotic therapy is proposed for one month before surgery which is necessary to free the pelvis from residual abscess, urinary or bowel compression. Theoretically, bilateral salpingo-oophorectomy is sufficient but extensive pelvic inflammation often leads to both bilateral salpingo-oophorectomy and total hysterectomy. This scheme can minimize the risk of mutilating debulking surgery with better control of inflammatory infection. Radical and reconstructive surgery may be required in patients with permanent sequelae [21, 22].

High suspicion, thorough evaluation and timely surgical approach followed by prolonged antibiotic therapy may help to improve patient morbidity and decrease the frequency of permanent sequelae caused by untreated pelvic actimycosis.

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