



## Case Report

## Management of Patients with Huge Pelvic Actinomycosis Complicated with Hydronephrosis: A Case Report

Pin-Kuei Fu<sup>a,b</sup>, Che-An Tsai<sup>c\*</sup><sup>a</sup>Division of Chest medicine, Department of Internal Medicine, Taichung Veterans General Hospital, Taichung, Taiwan.<sup>b</sup>School of Chinese Medicine, China Medical University, Taichung, Taiwan.<sup>c</sup>Division of Infection, Department of Internal Medicine, Taichung Veterans General Hospital, Taichung, Taiwan.

Actinomycosis is an uncommon, chronic, granulomatous disease caused by several species of the genus *Actinomyces*, a Gram-positive, filamentous bacterium that normally colonizes mucosal areas. Actinomycosis can be mistaken for malignant tumors, and in most cases the diagnosis is delayed or missed entirely until surgery. Actinomycosis in the abdomen or pelvis mostly results from prolonged use of an intrauterine device. We report the case of a 40-year-old female who presented with decreased urine production, poor appetite, marked loss of body weight and intermittent lower abdominal pain for 3 months. Abdominal computed tomography indicated a large infiltrative pelvic mass that was complicated by bilateral hydronephrosis, bladder compression and small bowel adhesions. Despite the elevated levels of cancer antigen 125, we suspected pelvic actinomycosis because of a 15-year history of an intrauterine device. The diagnosis was confirmed by histopathological examination of soft tissue obtained from a laparotomy biopsy. We successfully treated the patient with prolonged antibiotics instead of surgical eradication. Abdominal computed tomography obtained 1 year later showed almost complete resolution of the pelvic inflammatory mass.

**KEYWORDS:** huge pelvic actinomycosis, hydronephrosis, intrauterine device, prolonged antibiotics

### Introduction

Actinomycosis is an uncommon, chronic, granulomatous disease caused by several species of the genus *Actinomyces*,

a Gram-positive, filamentous bacterium. Infection usually occurs after disruption of the mucous membranes and these organisms have been isolated from the mouth, bronchi, gastrointestinal tract and female genital tract.<sup>1</sup> In the female genital tract, intrauterine devices (IUD) can traumatize the endometrium, causing erosions that create a favorable environment for *Actinomyces*.<sup>1–3</sup>

When a woman presents with a pelvic tumor accompanied by increased levels of cancer antigen 125 (CA-125), lower abdominal pain and loss of body weight, malignancy is typically the initial diagnosis. We describe a female patient with an IUD who presented with lower abdominal and pelvic actinomycosis, complicated by bilateral hydronephrosis, that was diagnosed during a scheduled diagnostic laparotomy and biopsy. The huge pelvic mass

\*Corresponding author. Division of Infection, Department of Internal Medicine, Taichung Veterans General Hospital, 160 Section 3, Taichung-Kang Road, Taichung 407, Taiwan.

E-mail: [lucky-sam@yahoo.com.tw](mailto:lucky-sam@yahoo.com.tw)

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almost completely resolved after a long period of antibiotic treatment instead of major surgical eradication. This case emphasizes the need to consider pelvic actinomycosis in a female patient with a history of prolonged IUD implantation, presenting as an infiltrated soft tissue mass arising from the tubo-ovarian region. The patient showed a good response to prolonged treatment with antibiotics, even with such a huge complicated pelvic actinomycosis.

## Case Report

A 40-year-old woman (gravida 2, para 2) was admitted with a 6-month history of lower abdominal pain, intermittent fever and decreased urine production. Her lower abdominal pain was intermittent, localized below the umbilicus, and accompanied by elevated body temperature (above 38°C), poor appetite, marked loss of body weight (about 7 kg over the preceding 3 months), general malaise, decreased urine production and changeable bowel habits. On examination, her body temperature was 38.6°C, pulse rate was 98 beats/min, and blood pressure was 130/80 mmHg. Physical examination showed pale conjunctiva, hypoactive bowel sounds, non-pitting edema of the lower left leg, knocking pain in the bilateral flank and tenderness over the left side below the umbilicus.

Laboratory tests indicated a white blood cell (WBC) count of  $15.0 \times 10^9/L$  with 82.7% neutrophils, a hemoglobin level of 5.6 g/dL, and a platelet count of  $448.0 \times 10^9/L$ . The C-reactive protein (CRP) level was 16.0 mg/dL and the erythrocyte sedimentation rate (ESR) was 110 mm/hr. Urine examination indicated pyuria and a urine culture yielded *Escherichia coli* 3 days later. The liver function test was normal, but impaired renal function (creatinine = 2.2 mg/dL) was noted.

Pelvic ultrasound indicated a large pelvic mass with a blurred and irregular margin. Computed tomography (CT) of the abdomen showed an IUD in the endometrial cavity, a distended bladder, and a well-enhanced, infiltrating soft tissue mass ( $9.6 \times 7.2 \times 7.6$  cm) over the lower abdomen and pelvis. The mass was adhered to the uterus and regional bowel loops and encased the bilateral lower ureter, causing bilateral hydroureter and hydronephrosis (Figure 1). Several tumor markers were tested, and increased levels of CA125 (97.17 U/mL; normal range, <35.0 U/mL), but normal levels of alpha-feto protein,

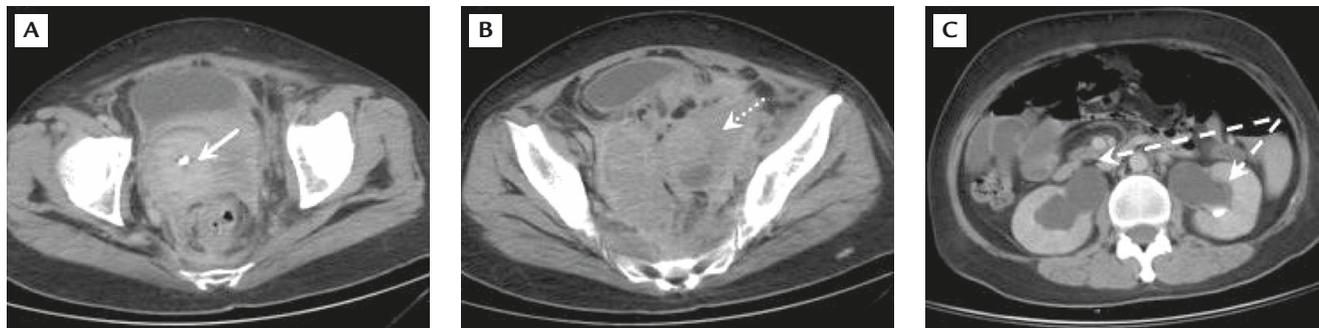
carcinoembryonic antigen and CA15-3 were found. Bilateral percutaneous nephrostomy was performed for obstruction relief.

Further gynecological inquiries revealed that the patient had been using an IUD for 15 years and had recently experienced prolonged menstruation (about 14 days) and increased vaginal discharge. A Pap smear was negative for malignancy and a cervical culture yielded *E. coli*. The IUD was removed and cultured. It was positive for *Bacteroides fragilis* and *E. coli*. We performed an open biopsy and found a firm and irregular mass (about  $7 \times 6$  cm) with adhesions to the intestine during laparotomy. After staining with Periodic acid-Schiff, acid-fast, Gram stains, and Gomori methenamine silver, pathologic sections revealed sulfur granules (hematoxylin and eosin stains) that were compatible with a diagnosis of actinomycosis (Figure 2).

After confirmation of actinomycosis, we administered intra venous penicillin G (3 million U; q6 h) and clindamycin (300 mg, q6 h). Defervescence occurred 8 days later. Her renal function and clinical condition also improved. She was discharged after an 8-week course of penicillin via intravenous injection. She was prescribed oral penicillin V (400,000 U, q6 h) for 7 months, followed by amoxicillin (500 mg, q8 h) for the next 6 months. CT scans of the abdomen, performed 3 months, 9 months and 2 years after diagnosis, showed almost complete resolution of the pelvic inflammatory mass (Figure 3).

## Discussion

Actinomycosis is a slowly progressive infection, most commonly caused by the anaerobic Gram-positive filamentous bacteria, *Actinomyces israeli*. The most common forms of actinomycosis are cervicofacial, thoracic and abdominogenital.<sup>4</sup> Symptoms of abdominal and pelvic actinomycosis are typically indolent and nonspecific. The most common clinical presentations are fever (60%), abdominal pain (85%) and body weight loss (44%).<sup>5</sup> Typical findings from laboratory tests include: increased WBC count, anemia, increased CRP, ESR, CA-125 and CA-199.<sup>6</sup> When complicated by severe hydronephrosis, renal function deteriorates, presenting as increased blood urea nitrogen and creatine levels.<sup>7</sup> Because the clinical manifestations of actinomycosis are many and the diagnosis is commonly



**Figure 1.** Computed tomography of the abdomen on admission. (A) An intrauterine device in the endometrial cavity and (B) a huge soft tissue mass in the pelvis, complicated with (C) bilateral hydronephrosis.

confused with a neoplasm, clinicians need to be more aware of this disease in order to minimize unnecessary surgical interventions, morbidity and mortality.

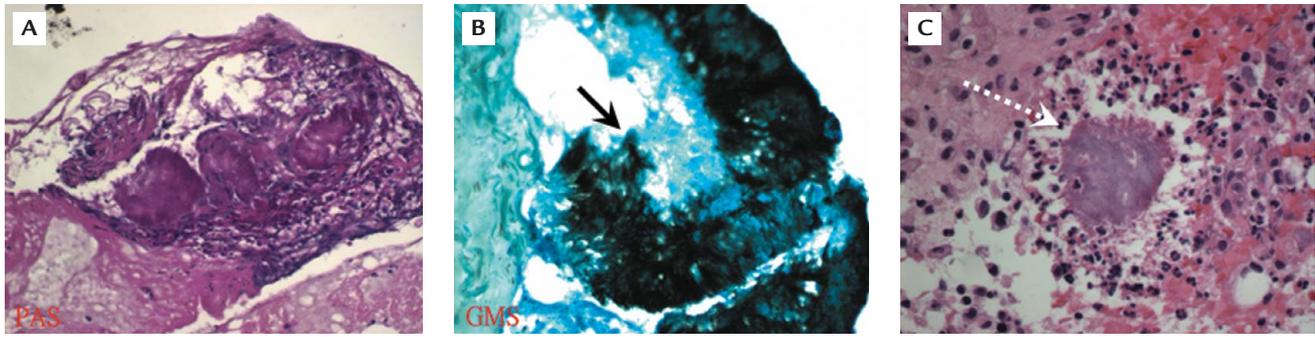
Actinomyces-induced pelvic inflammation most commonly occurs via the ascending route from the uterus in association with IUD.<sup>1,2,5,8-10</sup> Mali B *Actinomyces israeli* is isolated from 10% of asymptomatic IUD users and 25% of symptomatic IUD users during routine vaginal examination.<sup>3</sup> Lee and Hawnaur reviewed the incidence of actinomycosis in IUD users and found that it was between 8–16% in most reports, and rises slightly after 2 years of IUD use.<sup>11,12</sup> Because this patient had most of the aforementioned abnormalities and a 15-year history of IUD implantation, she was at high risk of suffering from pelvic actinomycosis.

Imaging of abdominal and pelvic actinomycosis typically does not help with diagnosis because the disease usually presents as an abscess or a mass lesion and is mistaken for a tumor. Diagnosis is usually made after surgery, confirmed by pathologic examination and identification of sulfur granules in the pus or soft tissue mass.<sup>13</sup> Before confirmation of the diagnosis, there should be differential diagnoses of abdominal tuberculosis, ovarian cancer, rectal cancer and other malignancies.<sup>14</sup> Therefore, the most difficult factor with this uncommon disease is to make a correct diagnosis before a major operation is performed. CT- or ultrasound-guided aspiration or needle biopsy may be useful to obtain a correct diagnosis.<sup>12,13</sup> We performed an open biopsy on this patient because the lower abdominal mass was adhered to the uterus and regional bowel loops, increasing the risk associated with CT-guided biopsy.

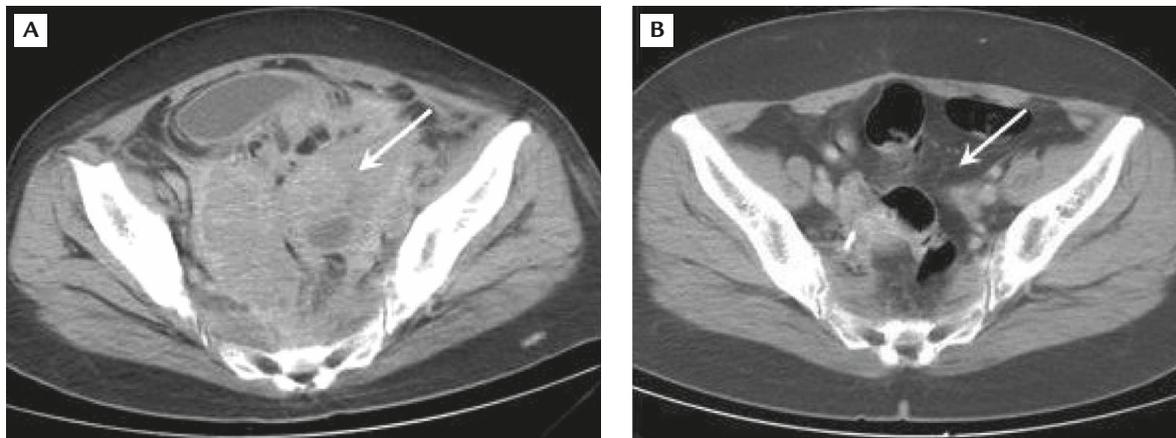
After pelvic actinomycosis is confirmed, administration of penicillin, as well as IUD removal, is the rule.<sup>15,16</sup>

High-dose penicillin, administered over a prolonged period, is the cornerstone of therapy for actinomycosis. Current recommendations include an initial course of penicillin G at 20 million IU/day for 4 weeks, followed by oral penicillin V at a dose of 2–4 g/day for 12 months.<sup>16</sup> Surgery should be considered if malignancy cannot be definitely excluded, or if removal of persistent sinuses, abscess drainage, or excision of necrotic tissue is necessary.<sup>12,16</sup> Although there was a huge pelvic mass, close to 9 cm in diameter and complicated with bilateral hydronephrosis, in our patient, we explored the opportunity to avoid unnecessary surgical operation. Prolonged intravenous penicillin for 8 weeks was prescribed due to the more severe pelvic infection in this patient. Oral penicillin V for 7 months and amoxicillin for another 6 months were then prescribed until the soft tissue mass was completely resolved. The patient responded very well to conservative therapy. We also suggest that CT scans could be used to monitor the response to therapy and inform clinicians as to the required duration of treatment.

In summary, early and accurate diagnosis of actinomycosis is challenging because it is an indolent, slowly-progressive infection, and the clinical presentations are many and commonly confused with a neoplasia. Some clues may be helpful for early recognition of this disease, including the prolonged use of an IUD. Laboratory test results showing elevated WBC counts, anemia, increased ESR, CRP and CA-125 are common findings in actinomycosis. If the abdominal CT scan shows a unilateral or bilateral tubo-ovarian abscess, or mass, accompanied by renal/urinary tract distention, hydronephrosis or topical intestinal obstruction, a differential diagnosis of actinomycosis should be considered. Once diagnosis is established,



**Figure 2.** Confirmation of the branching, filamentous *Actinomyces* bacterium. The bacterium was confirmed after staining with (A) Periodic acid-Schiff and (B) Gomori methenamine silver (GMS) and hematoxylin and eosin. The GMS stain shows the grey-black staining of actinomycosis. (C) The sulfur granules are composed of filamentous *Actinomyces* with a radial arrangement at the periphery and surrounded by a rim of lymphocytes.



**Figure 3.** Comparison of the abdominal computed tomography scans (A) on admission and (B) 2 years after diagnosis of actinomycosis, showing almost complete resolution of the huge pelvic inflammatory mass.

this infection can be successfully treated with high-dose, long-term penicillin, even for a huge pelvic mass. An awareness of this disease will expedite diagnosis and treatment and minimize unnecessary surgical interventions, morbidity, and mortality.

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