Bilateral Hydronephrosis and Hydroureters as First Manifestation of Abdominal Actinomycosis: A Case Report

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Obstructive uropathy is a commonly encountered medical condition in an emergency room. Many intraabdominal lesions, such as infections, inflammations, stones or tumors, may cause it. We report a case of abdominal actinomycosis in a 41-year-old female who had been pathologically proved pelvic actinomycosis, which may be related to the use of an intrauterine contraceptive device (IUCD) with initial manifestation of obstructive uropathy. Her abdominal computed tomography (CT) scan revealed an unusual clinical feature consisting of an infiltrative and irregular retroperitoneal soft-tissue mass encasing the adjacent ureters resulting in bilateral hydronephrosis and hydroureters. Abdominal actinomycosis is an uncommon infectious disease that must be included among the differential diagnoses of infiltrative intra-abdominal disorders, especially in the patient with an IUCD. Therefore, all emergency physicians should be alert to any patient with an IUCD in situ in the emergency department (ED). Definite diagnosis is generally based on the histopathologic identification of the actinomycotic sulfur granules, culture of the Actinomyces or both.

Key words: actinomycosis, contraceptive device, hydronephrosis, hydroureters

Introduction

In 1878, Israel first described human actinomycosis. Abdominal actinomycosis is a rare and chronic granulomatous disease caused by Actinomyces, which is gram-positive, slow growing, anaerobic, non-acid-fast and filamentous bacteria. Actinomyces are normal inhabitants of the upper respiratory, gastrointestinal and female genital tracts and rarely causes diseases. Diseases only occur when mucosal barriers are breached and the bacteria gains access to deeper tissues. Because of the nonspecific nature of the illness, symptoms may persist from months to years before the diagnosis is established. Clinical presentations are so insidious and slow that it is difficult to make an early accurate diagnosis. Therefore, the correct diagnosis is rarely diagnosed preoperatively, with many patients undergoing a total abdominal hysterectomy and bilateral salpingo-oophorectomy for what was presumed to be ovarian cancer. In 1973, Dr. Henderson described the relationship between the IUCD and pelvic actinomycosis. The majority of cases are usually secondary to the presence of damaged tissue, especially with the prolonged usage of an IUCD for more than 3 years. The rate of infection increases with the length of time the IUCD has been in place. Definite
diagnosis is generally based on the histopathologic identification of the actinomycotic sulfur granules, culture of the Actinomyces or both. Abdominal actinomycosis complicated with an obstruction of the genitourinary tract is rare. Herein, we report an interesting case with initial presentation of obstruction of the genitourinary tract.

Case Report

A 41-year-old female had a history of pulmonary tuberculosis (TB) diagnosed in 1993, with an incomplete course of anti-TB drugs for 2 months. She received an IUCD implantation about 5 years ago. She suffered from left lower abdominal pain, poor appetite, left lower leg edema, body weight loss of 6 kg for 4 months and fever accompanied with chills and dysuria for a week prior to her admission to our hospital. She was taken to a local clinic 4 days where anemia, pyuria and impaired renal function were found through a series of examinations. Abdominal ultrasound showed gall bladder stones, bilateral hydronephrosis and a huge pelvic mass about 11×6×11 cm

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of an undetermined nature. Because malignancy of the genitourinary tract could not be ruled out, she was referred to our ED for further and more in-depth examinations on February 17, 2005.

Upon her arrival to our ED, the patient had blood pressure of 138/80 mm Hg, pulse rate of 88 beats/min, body temperature of 36.8°C and respiratory rate of 18 times/min. She had alert consciousness, pale in appearance, clear breathing sound, regular heartbeats, lower abdominal tenderness and bilateral flank knocking pain. Laboratory data showed white blood cell counts (WBC) 15000/mm

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with neutrophils 82.7% and lymphocytes 11.6%, hemoglobin 5.6 g/L, mean corpuscular volume 80.7 fl, platelet cell counts 448×10

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/ mm

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, sodium 142 mEq/L, calcium 8.2 mg/dL, blood urea nitrogen 17 mg/dL, creatinine 2.2 mg/dL, albumin 2.9 g/dL, alkaline phosphatase 96 U/L, glutamic-oxalacetic transaminase 17 U/L, glutamic-pyruvic transaminase 11 U/L, lactate dehydrogenase 151 U/L, creatine kinase <28 U/L, blood sugar 124 mg/dL, prothrombin time 15.3 sec, activated partial prothrombin time 33.4 sec, and C-reactive protein 15.9 mg/dL. Urine routine showed WBC 3+/HPF and negative for nitrite. An IUCD was noted by the abdominal plain film. The blood transfusion with packed red blood cells was given for severe anemia. The abdominal computed tomography (CT) revealed an infiltrative and irregular enhanced retroperitoneal soft-tissue mass with the adhesion of regional bowel loops and the encasing of the adjacent ureters which resulted in bilateral hydroureters and hydronephrosis in the lower abdomen and pelvis (Figure 1). In addition, adhesion of the uterus with an IUCD in place was also noted (Figure 2). We consulted a gynecologist under the suspicion of an ovarian tumor. Initially, it was excluded by the gynecologist due to the clinical data we received and the pelvic sonography pointing toward infection. Bilateral percutaneous nephrostomies with tube insertion were performed for obstructive uropathies on the next day after her admission to our hospital. She then was admitted to our ward for further survey and management.

Empiric cephalosporin and gentamycin were initially prescribed under the impression of urinary tract infection, but the urine culture showed no growth of bacteria. The urine cytology, acid-fast stain and serum tumor markers, including alpha-fetoprotein, carcinoembryonic antigen and carbohydrate antigen-153, were all negative. The IUCD was removed on the third day of her admission, and the culture of the IUCD yielded Bacteroid fragilis and Escherichia coli. Due to the uncertain diagnosis, an exploratory laparotomy with tissue biopsy was done one week after her
Fig. 1  An infiltrative and irregular enhanced retroperitoneal soft-tissue mass in the lower abdomen and pelvis with adhesion of regional bowel loops and encasing of the adjacent ureters (A) resulting in bilateral hydronephrosis and hydroureters (B)

Fig. 2  An infiltrative and irregular enhanced soft-tissue mass in the lower abdomen and pelvis with adhesion of the uterus (A) and an intrauterine contraceptive device (IUCD) in place (B)
admission. Actinomycosis, resulting in a chronic infiltrative inflammatory mass, was pathologically proved by the presence of the actinomycotic sulfur granules within bacteria (Figure 3). Penicillin at a dose of 3 million units every 6 hours was prescribed for 18 days beginning on February 24, 2005. The patient was then transferred to a nearby hospital to complete a 4-week therapeutic course. A 2-year follow-up period was uneventful and the patient gradually recovered. The follow-up abdominal CT scans on November 26, 2005 and March 23, 2007, respectively showed gradual resolution and almost complete resolution of the retroperitoneal infiltrative mass lesion.

**Discussion**

Actinomycosis is an uncommon chronic, suppurative infection caused by the gram-positive anaerobic bacterium, *Actinomyces israelli*. It belongs to the order of actinomycetales, which have a prokaryotic cell type, and reproduce by fragmentation or spore formation. Initially, it was thought to be a fungus due to these morphologic features, but it is actually a bacterium. Although the organisms are normal flora of the oral cavity, gastrointestinal tract and female genital tracts, they rarely seem to be pathogens. Diseases occur only when mucosal barriers are breached and the bacteria gain access to deeper tissues\(^1\). Actinomycosis was first described in 1878. *Actinomyces spp.* are difficult to cultivate, particularly if the patient has been treated with antimicrobials before culture. Two weeks or longer are required for isolation. Yellow or orange granules, called sulfur granules, can sometimes be seen in affected tissues. However, the lack of these granules does not exclude infection. These granules are only found in 50 percent of the cases, though the presence of sulfur granules is considered to be an important finding, which is compatible with our case. The three major sites of involvement are the cervicofacial (60%), abdominopelvic (25%), and thoracic (15%) regions\(^3\).

![Fig. 3](image)

Fig. 3 Chronic inflammatory lesion with lymphocytes infiltration and actinomycotic sulfur granules within bacteria (A). Actinomyces colonization on Gomor’s M. Silver stain (B)
In the early stage, the disease may be difficult to distinguish from other clinical conditions such as acute appendicitis, inflammatory bowel disease or malignant tumor, as was the presentation in our case. Because of the nonspecific nature of the illness, symptoms may persist from months to years before the diagnosis is established. In our case, the clinical course before definite diagnosis lasted for 4 months. Preoperative diagnosis of abdominal actinomycosis is difficult. Laboratory tests only reveal normocytic, normochromic anemia, leukocytosis, and an elevated erythrocyte sedimentation rate. Radiological examination is not useful; however, a CT scan may be helpful in revealing an infiltrative and dense heterogeneous contrast-enhanced mass. Although the operative view was supported, a definitive diagnosis of actinomycosis was made, indicating the sulfur granules in the resective samples after surgery.

Abdominal actinomycosis with obstruction of the genitourinary tract is rare. The majority of cases are secondary to the use of IUCD. Actinomycosis must be considered in any woman with a history of IUCD use who has abdominal pain, pelvic mass or obstructive uropathy in ED. The rate of infection increases with the length of time the IUCD has been in place. The diagnostic problem of pelvic actinomycosis associated with the use of IUCD is that this disease can simulate pelvic malignancy. The definite diagnosis should be provided by culture result from the abscess and the bacterium can also be detected on histological sample from the removed IUCD. However, our case was not compatible with such a diagnosis.

There are two parts in treatment of abdominal actinomycosis: surgical and medical. Surgery usually is required because of difficulty in diagnosis and therapeutically for debridement of necrosis, for incision and drainage of abscesses. However, surgery very seldom resolves the condition and thus has to be combined with medical treatment. If no allergy exists, high-dose and long-term use of penicillin is recommended to eradicate actinomycosis. At present medical treatment with 10 to 20 million units of penicillin G intravenously daily is recommended for a period of four to six weeks, followed by oral phenoxyethyl penicillin, 4 to 6 g daily. Because of the low penetration in the fibrosis and the tendency to recur, it is recommended that oral therapy be continued for a period of 6 to 18 months or until after complete disappearance of the lesions or until stabilization of the lesions for a period of six weeks. Good alternatives for patients who are allergic to penicillin are tetracycline, chloramphenicol, erythromycin, clindamycin, and imipenem.

Summary

Abdominal actinomycosis is an uncommon infectious disease that must be included among the differential diagnoses of infiltrative intra-abdominal disorders, especially in the patient with an IUCD. The use of an IUCD remains an important risk factor. Therefore, all emergency physicians should be alert to any patient with IUCD in situ in the ED. The definite diagnosis is made with a pathological examination of the specimen demonstrating the presence of sulfur granules. Large doses of penicillin are required from weeks to months for patients.

References

3. Levine LA, Doyle CJ. Retroperitoneal actino-
Actinomyces and hydrenephrosis

腹部放線菌病以雙側腎盂積水
和輸尿管積水為最初表現：一病例報告

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阻塞性尿路病變在急診室是經常碰到的情況。它可能經由多種腹腔內病變所引起，像是感染、發
炎、結石、或是腫瘤。我們報告一例經由骨盆腔病理切片證實以阻塞性尿路病變為初步表徵之腹部放線
菌病之四十一歲女性，這可能和子宮內避孕器的使用有關。腹部電腦斷層掃描顯示一個不尋常的影像，
包括一浸潤性的不規則後腹膜腫塊圍繞鄰近的輸尿管而導致雙側腎盂積水和輸尿管積水。腹部放線菌病
是不常見的感染性疾病，然而腹部內有浸潤性的異常時，特別在有子宮內避孕器的病人，必需列入鑑別
診斷。因此，所有急診醫師對於急診有子宮內避孕器的病人應警覺。確定診斷則要病理切片組織中顯示
放射菌的含硫顆粒，培養出放射菌或是兩者皆有。

關鍵詞：放線菌病，避孕器，腎盂積水，輸尿管積水

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