Huge Pelvic Mass, Cutaneous and Vaginal Fistulas, and Bilateral Hydronephrosis: A Rare Presentation of Actinomycosis with a Good Response to Conservative Treatment and with Long-term Sequelae of Renal Atrophy and Hydronephrosis

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SUMMARY

Objective: Actinomycosis with an extended pelvic abscess is an uncommon condition, which usually occurs coincident with the presence of an intrauterine contraceptive device (IUD) in the uterine cavity. The clinical picture of pelvic actinomycosis may vary between individuals, is often accompanied by complications, and is frequently misdiagnosed. Here, we report a case of pelvic actinomycosis, presenting as a huge pelvic mass and complicated by a vaginal fistula, a cutaneous fistula, and bilateral hydronephrosis, and we discuss the diagnosis and management of this patient.

Case Report: A 35-year-old woman was referred to our hospital with a huge pelvic complex mass and progressively worsening low abdominal pain. The tumor workup, which included a computed tomography (CT) scan, revealed an extended pelvic abscess and bilateral hydronephrosis. Both cutaneous and vaginal fistulas were also noted. Endometrial curettage and biopsies of the skin and vaginal lesions confirmed the diagnosis of actinomycosis. The patient underwent conservative treatment and recovered well, although the skin lesion only healed after 12 weeks of oral antibiotic treatment. At the 1-year follow-up, a CT scan showed sequelae including a mildly atrophic left kidney and left hydronephrosis.

Conclusion: In patients presenting with a pelvic mass and an IUD in the uterine cavity, the diagnosis of actinomycosis should be seriously considered. A detailed workup, including a CT scan, endometrial curettage and biopsies where possible, should be performed before surgery. Once diagnosis has been confirmed, conservative medical treatment should be attempted before considering laparotomy, to reduce the risk of complications. Despite successful treatment with antibiotics, long-term sequelae such as hydronephrosis and renal atrophy are possible in cases of extended pelvic actinomycosis. [*Taiwan J Obstet Gynecol* 2008;47(2):206–211]

Key Words: actinomycosis, cutaneous fistula, hydronephrosis, intrauterine device, sequelae, vaginal fistula

Introduction

Actinomycosis is the name given to an infection caused by the genus, *Actinomyces*, a group of anaerobic Gram-positive bacilli, which can be found in the normal microflora of the oral cavity, pharynx, gastrointestinal (GI)

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tract and genital organs in humans [1]. Actinomycotic infections usually present as localized, chronic, infiltrative granulomatous tumors of the cervicofacial region, oral cavity, respiratory system (including the bronchi), and GI system (including the liver) [2–5]. However, they are also known to cause extensive infiltrative infections, such as endometritis, tubo-ovarian abscesses or abdominal abscess, in women who have used long-standing intrauterine contraceptive devices (IUDs). The relationship between IUDs and pelvic actinomycosis has been established since the 1970s [6–9], and such IUD-associated infections can be sufficiently extensive to cause severe pelvic adhesions, GI obstruction, fistula formation, and hydronephrosis. However, the presentation of an infiltrative pelvic abscess, complicated by bilateral hydronephrosis and vaginal and cutaneous fistulas, has rarely been reported. Here, we report a case of a huge pelvic actinomycotic abscess with the above clinical presentation, which showed a good response to conservative antibiotic treatment. To the best of our knowledge, this is the first reported case of an individual with pelvic actinomycosis, with simultaneous cutaneous and vaginal fistulas and bilateral hydronephrosis, without bowel injury.

Case Report

A 35-year-old woman was referred to the emergency unit of our hospital from local clinics because of a 2-week history of progressively worsening low abdominal pain and vaginal bleeding. On arrival, she was noted to have a low grade fever (37.3°C) but otherwise stable vital signs. Pelvic examination showed an enlarged uterus with the vaginal cavity filled with a suppurative bloody discharge. A small pit was noted in the posterior fornix of the vagina, suggesting fistula formation (Figure 1). Physical examination also revealed an infiltrative erythematous area (about 8 × 10 cm) at the left lower abdominal wall, with no signs of peritonitis or GI obstruction. There were several perforation holes on the surface of the skin lesion (Figures 2–4). Ultrasonography revealed an infiltrating complex pelvic mass with no obvious margin, and an

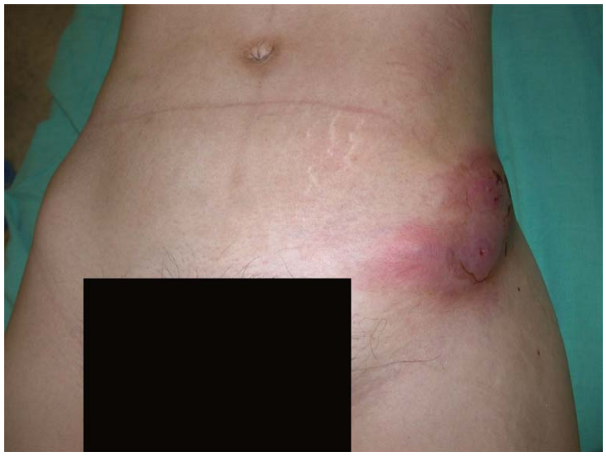

Figure 2. Skin lesion at left lower abdomen.

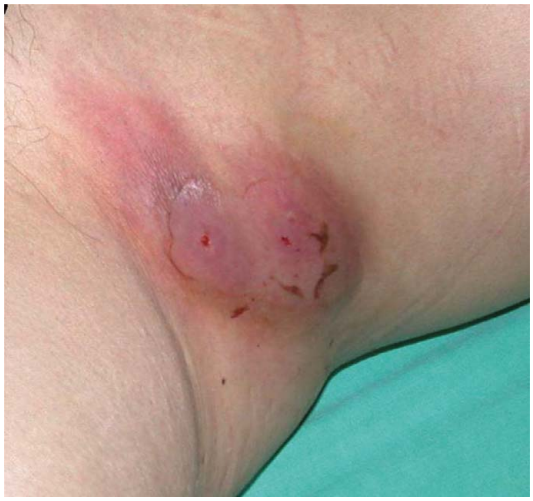

Figure 3. Skin lesion of left lower abdomen showing typical signs of infection.

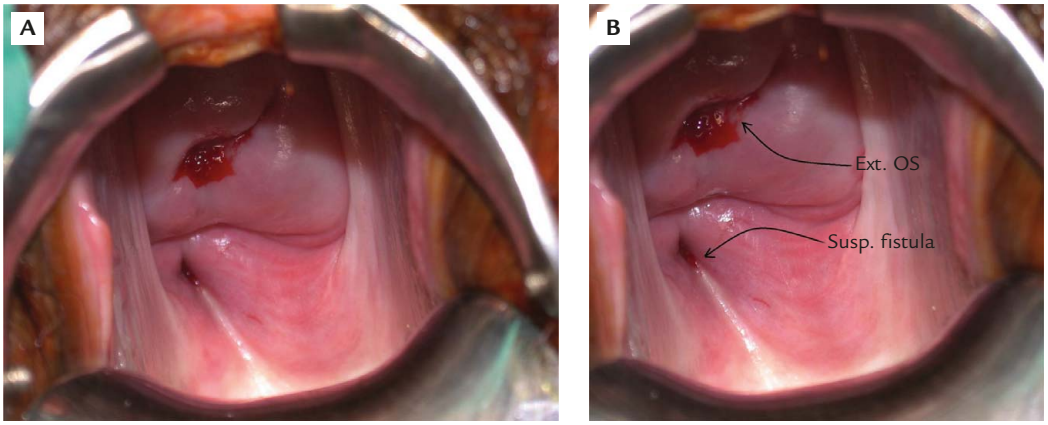

Figure 1. (A) Direct view of external os and vagina. (B) The arrows indicate the external os and the fistula tract of the vagina. Ext. OS = external os; Susp. fistula = suspected fistula.

IUD in the uterine cavity. On arrival, the laboratory data for this patient showed mild anemia (hemoglobin 7.9 g/dL), mild leukocytosis (white blood cell count 12,700/μL) without urinary tract infection, elevated C-reactive protein (15.1 mg/dL), and otherwise normal serum biochemistry. The serum CA125 level was also within normal limits (24.62 U/mL). Some granulation tissue protruding through the vaginal fistula was taken for pathologic examination.

Due to a suspected pelvic mass with suspicious abscess formation and abdominal wall and vaginal invasion, the patient was admitted for detailed workup. She underwent a computed tomography (CT) scan, which unexpectedly revealed marked bilateral hydronephrosis and hydroureter (Figures 5 and 6), as well as an ill-defined heterogeneous infiltrative mass with cystic and soft tissue parts occupying the cul-de-sac, both sides of the perirectal fascia, both sides of the parametrium, and the left side of the retroperitoneum. The lesion was also noted to invade the right psoas and iliopsoas muscles and to extend to the left abdominal wall, through a fistula tract (Figures 7 and 8).

The *in situ* IUD and extensive pelvic mass suggested the possibility of pelvic actinomycosis. Surgical interventions,

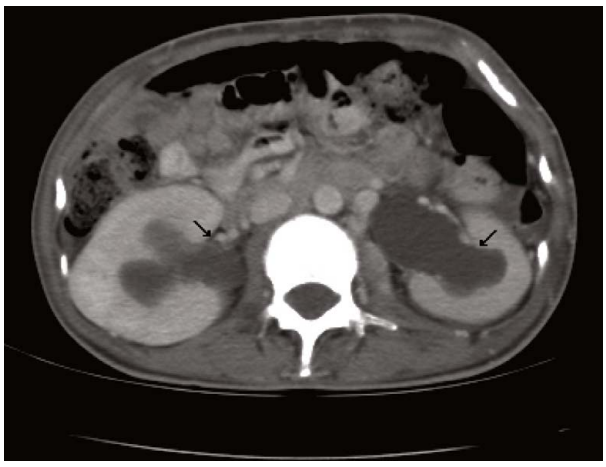

Figure 6. Computed tomography scan with contrast. The arrows indicate the hydroureter.

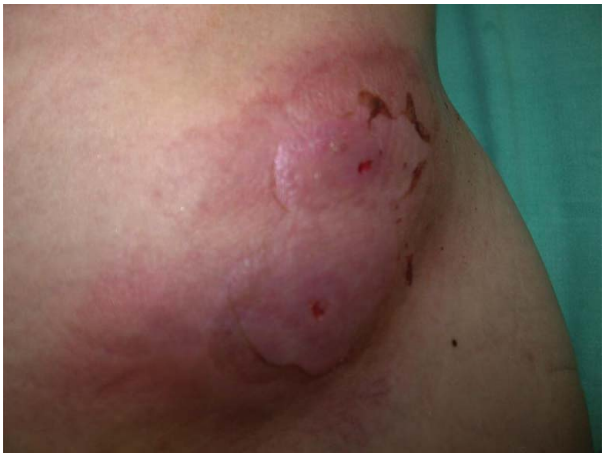

Figure 4. There are two perforation holes, indicating cutaneous fistula formation.

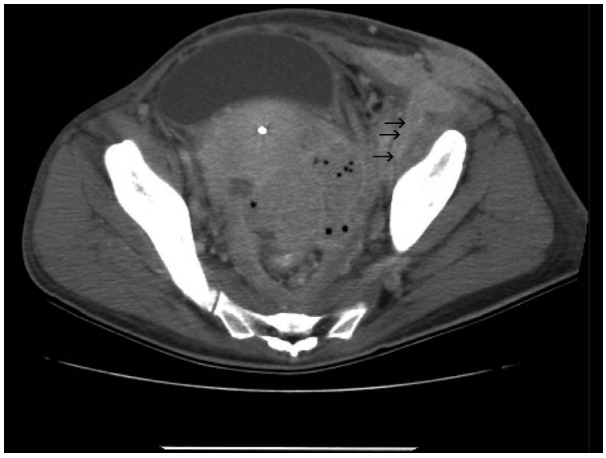

Figure 7. Computed tomography scan. The arrows indicate the fistula tract towards the left lower abdominal wall.

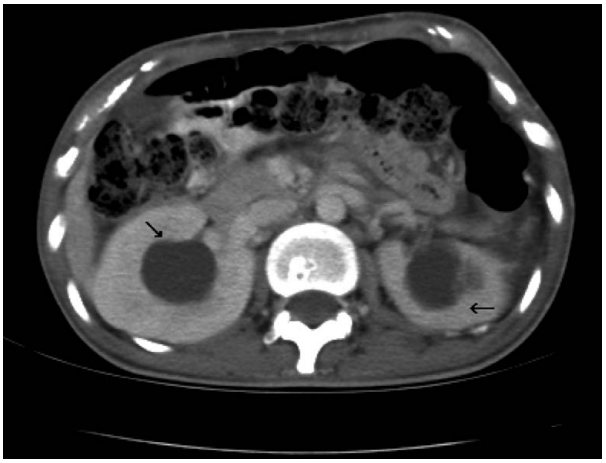

Figure 5. Computed tomography scan with contrast. The arrows indicate bilateral hydronephrosis.

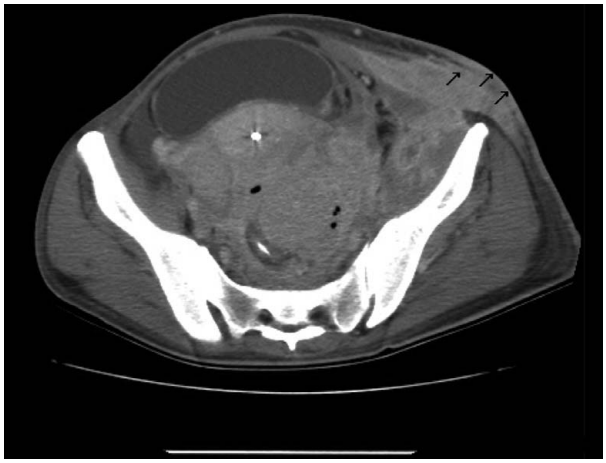

Figure 8. Computed tomography scan. The arrows indicate the skin lesion at the left lower abdominal wall.

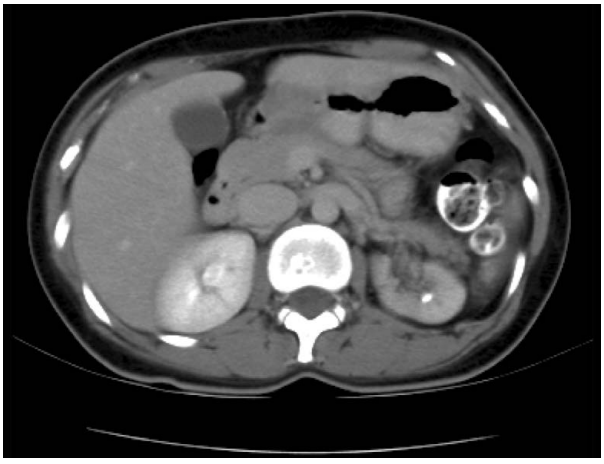

Figure 9. Computed tomography scan at 1-year follow-up. Note the enhancement difference between the bilateral kidneys.

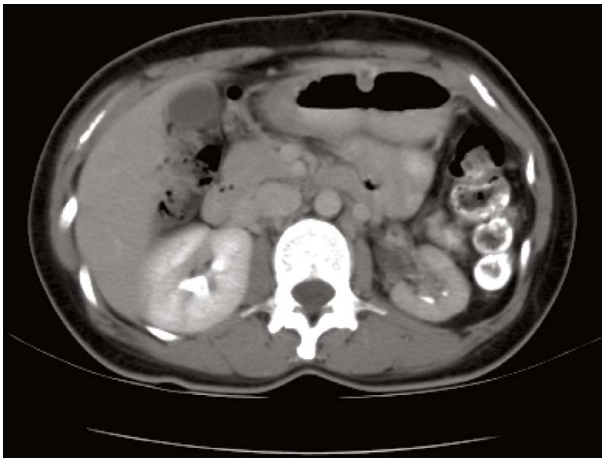

Figure 10. Computed tomography scan at 1-year follow-up, showing mildly dilated left renal pelvis.

including an exploratory laparotomy, were not undertaken, and instead, diagnostic endometrial curettage, cutaneous lesion biopsy, and ultrasound-guided pelvic mass biopsy were performed. The IUD was also removed during the operation. Pathology reports confirmed the diagnosis of actinomycotic infiltration and were negative for malignancy. Microbiologic cultures from the abdominal and vaginal fistula tracts produced negative results.

The patient underwent conservative treatment with parenteral antibiotics. She was given aqueous penicillin 10 million IU every 6 hours, which was changed to Augmentin 1 g orally every 12 hours after 3 days. The fever subsided following antibiotic treatment. The abdominal wall lesion shrank progressively but did not heal until week 12 of oral antibiotic treatment. This patient underwent follow-up ultrasonography after 3 months of oral antibiotics. At this time, the right-sided pelvic abscess was reduced and the left-sided pelvic mass had shrunk to half its original size. Oral antibiotic treatment was continued for a further 3 months, until there was no detectable residual left pelvic infiltrating mass. Because of personal reasons, the follow-up CT scan was delayed until 1 year after the primary treatment, at which time it showed mild left kidney atrophy with reduced contrast enhancement (Figures 9 and 10). The left kidney measured 7.31 × 3.91 cm. A detailed anatomic and functional work-up of the left kidney was advised but was refused by the patient because of personal reasons (she was an immigrant).

Discussion

Actinomycosis is often considered to be an opportunistic infection that manifests itself in the cervicofacial (50%), abdominal (20%) and thoracic (15%) areas. Pelvic actinomycotic abscesses are known to be related to the long-term use of an IUD [6,9]. *Actinomyces* colonies are usually present in the removed IUD, though the presence of *Actinomyces* in the genital organs or an IUD does not necessarily indicate an infective disorder. The estimated prevalence of actinomycosis is low [1], even in HIV-positive patients [10]. However, IUD-related pelvic actinomycotic abscesses are often reported. Some pelvic actinomycoses appear as wide-spreading and extensive infiltrating abscesses. GI tract involvements, including GI obstruction [11], bowel fistula formation, bleeding and extensive adhesions, are the most common complications [12,13]. Other reported complications of pelvic actinomycosis include abdominal wall abscesses and fistula formation [14], vaginal involvement, and unilateral or bilateral hydronephrosis [15–17]. Such complications have rarely been recorded simultaneously. We believe this patient to be the first reported case of primary pelvic actinomycosis presenting simultaneously as abdominal and vaginal fistulas, and bilateral hydronephrosis.

The preoperative diagnosis of actinomycosis can be very difficult. Most pelvic masses are treated as suspected ovarian tumors, and thus receive surgical interventions, with the diagnosis being confirmed postoperatively, based on the pathologic findings [1,11]. In our case, the pelvic mass also presented initially as a huge pelvic tumor and surgical intervention was therefore considered. The differential diagnosis of actinomycotic abscesses and pelvic tumors, such as ovarian cancer, is often difficult; they show similar infiltrative and solid or complex components on ultrasound and rarely cause marked signs of infection, such as high fever, leukocytosis, pus formation or sepsis. Exploratory laparotomy is commonly conducted but can lead to massive bleeding

and severe complications, requiring GI resection or repair [18].

In our patient, the preoperative diagnosis of actinomycosis was confirmed by ultrasound-guided biopsy, abdominal wall biopsy, and biopsy of the vaginal fistula. Fistula formation in both the vagina and abdominal wall was helpful in establishing the correct diagnosis in this patient. In most cases of ovarian or pelvic tumors, preoperative needle biopsy is not recommended because of the possibility of tumor dissemination, but this policy also restricts the opportunity to confirm the actinomycotic infection preoperatively, and so avoid unnecessary complications. However, in our case, tissue could be obtained from the two fistula tracts, thus allowing an early diagnosis. Furthermore, ultrasound-guided needle biopsy of the pelvic mass excluded the possibility of malignancy. We therefore administered antibiotic treatment with a good clinical response. If the tumor size, hydronephrosis and infective signs fail to respond well to medication, surgical intervention should still be considered.

The important role of CT scans in confirming the diagnosis of pelvic actinomycotic abscesses has been demonstrated [19,20]. In our case, the primary physical examination revealed no costophrenic angle knocking pain, and there were no symptoms, signs or complaints concerning the urinary tract, such that CT scanning was required to identify the severe bilateral hydronephrosis. The CT scan also helped to visualize the spreading tract of the abscess, and thus helped to confirm the consistency and accuracy of the biopsy results. In some cases, if an ultrasound-guided biopsy is not achievable, a CT-guided biopsy may be considered [21].

In this case, we conducted diagnostic endometrial curettage and removal of the IUD to confirm the diagnosis and exclude the possibility of malignancy. The pathology report confirmed the presence of *Actinomyces* in the uterine cavity. However, as the "original" site of *Actinomyces* colonization, the uterine cavity showed much less inflammation than the other spreading lesions. Extensive pelvic abscesses without signs of endometritis may sometimes delay an accurate diagnosis, and this should be kept in mind.

The recommended primary management of actinomycosis is antibiotic treatment. Penicillin G 20 million IU/day parenterally is advised. However, because there was some pus coating on the abdominal lesion, we also administered metronidazole and gentamicin to avoid secondary infection. After primary parenteral antibiotics, Augmentin treatment was continued because of its broad spectrum. There have been at least two reports in the literature of cases of pelvic actinomycoses that recurred weeks [22] or even months [23] after removal of the IUD, and regular image follow-up is therefore strongly recommended.

This patient had severe bilateral hydronephrosis but normal serum renal function tests. We consulted the urologist concerning management of the hydronephrosis; primary double-J tube insertion was not recommended because of possible complications, such as perforation. The hydronephrosis improved after conservative treatment, but the 12-week follow-up ultrasound and 1-year follow-up CT scan still showed signs of left hydronephrosis. Mild left renal atrophy and left hydronephrosis were confirmed as long-term sequelae. Early double-J tube insertion may be helpful in preventing such long-term sequelae, but may also lead to severe complications such as renal loss. There is no single recommended treatment for patients with pelvic actinomycosis and hydronephrosis. Renal failure has been reported [24], and the management of primary hydronephrosis with the aim of preventing long-term sequelae, as well as the optimal timing of interventions, still presents a significant challenge in the treatment of such cases. Serial follow-up and careful consideration of individual cases are strongly recommended.

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