

Pelvic Actinomycosis Mimicking Ovarian Malignancy Causing Unilateral Hydronephrosis

Cemil KAYMAZ¹, Bahadır SAATLI¹, Özgür BİGE¹, Meral KOYUNCUOĞLU², Aykut KEFİ³, Uğur SAYGILI¹

Izmir-Turkey

Pelvic actinomycosis is a chronic granulomatous suppurative disease caused by *actinomyces israelii* and usually associated with long-standing use of an intrauterine device. Pelvic actinomycosis can cause tuboovarian abscess, ureteral and bowel obstruction in some rare cases mimicking an ovarian tumor. We report a pelvic actinomycosis case with elevated Ca 125 level and obstructed ureter mimicking an ovarian tumor. The preoperative imaging methods and Ca 125 level indicated an ovarian malignancy, but frozen section diagnosis was benign, and overtreatment was avoided. Pelvic actinomycosis, although is a rare granulomatous suppurative disease, in case of long-standing use of an intra-uterine device and ovarian mass, it should be in the mind of the surgeon as a differential diagnosis to avoid excessive surgery.

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Pelvic actinomycosis is a chronic granulomatous suppurative disease caused by *actinomyces israelii* and usually associated with long-standing use of an intrauterine device [IUD].¹

Patients with pelvic actinomycosis usually present with a unilateral or bilateral tubo-ovarian abscess.¹ The disease may cause pelvic inflammatory disease, abdominal wall abscess, and bowel obstruction by the effect of inflammatory process and adhesions or compression.^{1,2,3} Ureteral obstruction and hydronephrosis due to pelvic actinomycosis is rare.^{3,4} Besides these manifestations pelvic actinomycosis can mimic pelvic and abdominal malignancies.⁵

In this case report, we report a woman with pelvic actinomycosis mimicking ovarian carcinoma and causing unilateral ureteral obstruction and hydronephrosis. And relationship between ureteral obstruction and pelvic actinomycosis was discussed.

Case Report

A forty eight years old patient was attended to gynecology department because of having nausea, vomiting, abdominal discomfort and gross hematuria. Her obstetrical history

¹Dokuz Eylül University, School of Medicine Departments of Obstetrics and Gynecology,

²Pathology,

³Urology, İnciraltı, İzmir, Turkey

Address of Correspondence Cemil Kaymaz
Dokuz Eylül University, Faculty of
Medicine Department of Obstetrics &
Gynecology
İnciraltı, İzmir, Turkey

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included three normal vaginal delivery and five first trimester abortions. She had been using a copper bearing IUD for 10 years and it was removed 1 year ago. From her medical history it was learned that she underwent mitral valve replacement 4 years ago, and she has been taking oral anticoagulant therapy since then. Her gynecologic examination revealed a solid, fixed, tender mass in right adnexia approximately 6x5 centimeters in size, the size of the uterus and left adnexia were normal. She did not have any vaginal discharge. The pap smear of the patient was normal. Her body temperature was 38.2 °C. Her serum biochemistry revealed disturbed renal functions (blood urea nitrogen 49.6 mg/dl, serum creatinine 2.5 mg/dl) and elevated liver enzymes (AST 75 UI/ml, ALT 50 UI/ml). Tumor markers were negative except Ca 125 elevated to 59.7 UI/ml. Abdominopelvic ultrasonography demonstrated hydronephrosis in the right kidney, semisolid, right adnexial mass in 7.6x4.9 centimeters in size and pelvic free fluid. Magnetic resonance imaging revealed a right adnexial heterogeneous mass in 7x8.5 cm in size, omental cake, and multiple implants on pelvic peritoneal surfaces. Performed rectosigmoidoscopy was normal. Cystoscopy was performed and double J stent was inserted into right ureter, no suspicious lesions were seen on the mucosal surface of bladder and ureteral orifices. After insertion of double j stent renal function tests returned to normal levels (blood urea nitrogen 18.9 mg/dl, serum creatinine 1.08 mg/dl). Oral anti-coagulant therapy was replaced by low molecular weight heparin therapy before operation. The coagulation parameters of the patient were as follows on the day of operation: INR 1.22, activated partial thromboplastin time 23.67 sc, prothrombin time 15.04 sc.

With all these signs an ovarian tumor was suspected and laparotomy was performed. At laparotomy a right adnexial mass, multiple and dense intestinal adhesions were observed.

The pelvic peritoneum was thickened and all the surfaces were fragile to any manipulation. The omentum was edematous but no obvious infiltration was observed. Purulent drainage was observed from the mass. Total abdominal hysterectomy and bilateral salphingoophorectomy was performed and a drain was inserted to douglas. Right adnexial mass send to frozen section daignosis was reported as benign. In the post operative intensive care unit approximately 500 cc hemorrhagic drainage was observed. In spite of multiple blood transfusions the hemodynamic parameters of the patient deteriorated, hematocrit and hemoglobin levels decreased dramatically (hemoglobin 5.7 g/dl, hematocrit 16%). Intraabdominal bleeding was suspected and laparotomy was performed four hours later the formal operation. In second laparotomy pelvic hemorrhage was observed but there was no bleeding from any major pelvic arteries or veins. After the ligation of hypogastric arteries bleeding was controlled, and the hemodynamics of the patient was normal at the end of the operation.

Antibiotherapy with ceftriaxone and metronidazole was started empirically. The patient was discharged from the intensive care unit on the second post operative day. The result of the aerobe and anaerobe cultures taken from abscess material was negative. The pathology result of total abdominal hysterectomy and bilateral salphingoophorectomy specimens was reported as the adnexial mass containing bacterial groups compatible with actinomycosis and abscess focuses with chronic inflammation signs. . The antibiotherapy was changed to clindamycine 3x600 mg intravenously. The patient was discharged from hospital on postoperative 11th day without any medical or operative complications. The patient used oral amoxicillin and clavulonic acid for three months after her discharge. The right ureteral double j stent was taken three months later. The patient was free of symptoms six month within the operation.

Discussion

Pelvic actinomycosis is a rare complication of intrauterine devices.^{1,6} The classical scenario involves a patient with a unilateral or bilateral tuboovarian abscess, however the vast majority of patients are diagnosed as bilateral ovarian tumor or retroperitoneal mass.^{1,6,7,8} Pelvic actinomycosis is caused by a filamentous, gram-positive, anaerobic bacteria that is the part of the endogenous microflora of the human and animal oro-pharyngeal cavities and become pathogenic when they enter the peritoneal cavity.⁹ More than 50% of the actinomycosis infections occur in the cervicofacial region and only about 20% of the cases are abdominal infections. Other sites of infection include the thorax, liver, pancreas, bones, bone marrow, inguinal lymph nodes, testis, lacrimal duct, kidney, adrenals and breast.¹⁰ Classical actinomycosis infection is characterized by granulation tissue, severely dense fibrosis,

multiple small abscesses and draining sinuses. This process, producing a hard mass in the pelvis and retroperitoneum may compress the ureters and the adjacent intestinal loops like any intra-abdominal tumor.¹¹

Preoperative diagnosis of pelvic actinomycosis is difficult.^{1,6} Imaging methods are not diagnostic and usually suggest a neoplasm or an inflammatory process. Patients with correct preoperative diagnosis are less than 10%.¹² The best treatment modality for pelvic actinomycosis is antibiotic treatment and patients should undergo limited surgery when necessary.^{1,3,6} The treatment of actinomycosis is based on 18 to 24 million units of penicillin intravenously for 2 to 6 weeks, followed by oral therapy with penicillin or amoxicillin for 6 to 12 months.¹³ Erythromycin, doxycycline and clindamycine are other suitable alternatives.¹⁴

To our knowledge, only a few cases of ureteral obstruction caused by actinomyces infection in IUD users have been reported in the literature.^{3,4,15} This condition is related to mass effect and proper surgical and antibiotic therapies improve the ureteral obstruction in most cases, as in our case. When the obstruction is severe, placing ureteral stents usually relief obstructive symptoms. In our case actinomyces infection caused an ovarian mass that obstructed the ureter. Obstruction in the ureter was solved by a double j stent and laparotomy with total abdominal hysterectomy and bilateral salphingoophorectomy was the surgery. In laparotomy although purulent discharge from the mass was seen it was not possible to make differential diagnosis of a tuboovarian abscess and for this reason right adnexia was send to frozen section diagnosis. After frozen section diagnosis was reported as benign and further surgery was not performed. Although omental cake was seen in magnetic resonance imaging, omentectomy was not performed because, in laparotomy omentum was only edematous and the frozen section diagnosis was benign. Omentectomy could be performed in cases with macroscopic omental involvement.¹⁶ We suggest sending the tumor for frozen section diagnosis in cases with a pelvic tumor with long standing usage of IUD, because it can not always be possible to differentiate a malign tumor from a benign pelvic mass caused by a granulomatous disease.

In our case a second laparotomy was needed to control hemorrhage. This complication was related with patients being on medication by low molecular weight heparin therapy and the fragility of the tissues secondary to long standing infection. For this reason maximum attention should be paid on homeostasis these cases. Less frequently, myocardial or endocardial infection occurs, either via extension from the pericardium or by initial hematogenous seeding of the endocardium.¹⁷ In our case there was no clinical suspission of myocardial or endocardial infection.

Pelvic actinomycosis usually presents a diagnostic dilemma to the clinician. It is important to consider this diagnosis in any woman with a history of IUD usage who presents with a pelvic mass. Frozen section is helpful in presence of an elevated Ca 125 and a pelvic tumor when it is difficult to make a decision whether the mass is malign or a benign condition caused by a granulomatous infection. In addition, as shown in our case, pelvic actinomycosis should be considered in the etiology of ureteral obstruction due to pelvic mass when an IUD is involved.

In summary pelvic actinomycosis should be kept in mind while evaluating a patient with long standing usage of IUD who represents an adnexial mass causing ureteral obstruction with an elevated Ca 125 level. Because pelvic actinomycosis is a benign condition frozen section diagnosis should be made to avoid overtreatment.

References

1. Fiorino AS. Intrauterine contraceptive device-associated actinomycotic abscess and actinomycetes detection on cervical smear: review. *Obstet Gynecol* 1996;87:142-149.
2. Pearlam M, Frantz AC, Floyd WS, Faro S. Abdominal wall actinomycetes abscess associated with an intrauterine device: a case report. *J Reprod Med* 1991;36:398-402.
3. Nasu K, Matsumoto H, Yoshimatsu J, Miyakawa I. Ureteral and sigmoid obstruction caused by pelvic actinomycosis in an intrauterine contraceptive device user. *Gynecol Obstet Invest* 2002;54: 228-231.
4. Jackson AE, Parry JRW, Shah JR. Ureteric obstruction secondary to pelvic actinomycosis. *Br J Urol* 1988;62:85-87.
5. Sehouli J, Stupin JH, Schlieper U, Kuemmel S, Henrich W, Dencert C, Dietel M, Lichetegenner W. Actinomycotic inflammatory disease and misdiagnosis of ovarian cancer. A case report. *Anticancer Res.* 2006;26:1727-31.
6. Hamid D, Baldauf JJ, Cuenin C, Ritter J. Treatment strategy for pelvic actinomycosis: case report and review of the literature. *Eur J Obstet Gynecol Reprod Biol* 2000;89:197-200.
7. Hoffmann M, Roberts W, Solomon P, Gunasekarin S, Cavanagh D. Advanced actinomycotic pelvic inflammatory disease simulating gynecologic malignancy: a report of 2 cases. *J Reprod Med* 1991;36: 543-545.
8. Dogan NU, Salman MC, Gultekin M, Kucukali T, Ayhan A. Bilateral actinomycetes abscesses mimicking pelvic malignancy. *International Journal of Gynecology and Obstetrics* 2006;94:58-59.
9. Davies M, Keddie NC: Abdominal actinomycosis. *Br J Surg* 1973;60:18-22.
10. Weese WC, Smith IM: A study of 57 cases of actinomycosis over a 36-year period. *Arch Intern Med* 1975;135:1562-1568.
11. Berardi RS: Abdominal actinomycosis. *Surg Gynecol Obstet* 1979;149:257-267.
12. O'Connor KF, Bagg MN, Croley MR, Schabel S. Pelvic actinomycosis associated with intrauterine devices. *Radiology* 1989;170:559-560.
13. Mandell, Douglas, and Bennett's Principles and Practice of Infectious Diseases. Philadelphia, USA: Elsevier Inc, 2005;vol(2):2931.
14. Martin MV. Antibiotic treatment of cervicofacial actinomycosis for patients allergic to penicillin: A clinical and in vitro study. *Br J Oral Maxillofac Surg* 1985;23:428-435.
15. Butterworth RJ, Davies DGL, Hart AJL. Actinomycosis causing ureteric obstruction. *Br J Urol* 1990;66:548-549.
16. Atad J, Hallak M, Sharon A, Kitzes R, Kelner Y, Abramovici H. Pelvic actinomycosis. Is long-term antibiotic therapy necessary? *J Reprod Med.* 1999 Nov;44(11):939-44.
17. Lam S, Samraj J, Rahman S, Hilton E. Primary actinomycotic endocarditis: case report and review. *Clin Infect Dis* 1993;16:481-485.