Case Report

Severe pelvic actinomycosis with intrauterine device in situ and abdominal wall involvement treated with medication only: case report

H.J. Wang^{1,†}, Y.H. Wu^{1,†}, P.S. Huang^{1,2}

¹Department of Obstetrics and Gynecology, Taipei Medical University Hospital, Taipei City (Taiwan)
²Department of Obstetrics and Gynecology, School of Medicine, College of Medicine, Taipei Medical University, Taipei City (Taiwan)

Summary

Pelvic actinomycosis is a rare, granulomatous disease, caused by the gram-positive anaerobic bacteria, Actinomyces. It is usually associated with recent abdominopelvic surgery or intrauterine contraceptive device placement. The diagnosis is difficult because of the similarity with malignancy, inflammatory bowel disease, and tuberculosis. Although intrauterine device related actinomycosis is considered a well-documented diagnosis, there are intermittent case reports of unusual presentations. Appropriate image survey can aid clinicians in its diagnosis and treatment of pelvic actinomycosis and further prevent unnecessary invasive intervention. Here the authors present a case of suspected pelvic actinomycosis with abdominal wall involvement supported with images and describe the clinical manifestations. The disease with abdominal wall involvement and treated with empirical antibiotics administration only has yet to be reported.

Key Words: Actinomycosis; Pelvic infection; Intrauterine device; Computed tomography.

Introduction

Pelvic actinomycosis is a rare, chronic granulomatous suppurative infection, frequently associated with recent abdominopelvic surgery (such as pessary used during surgery), trauma, neoplasm, a perforated viscus or the use of intrauterine devices (IUD) [1, 2]. It is difficult to diagnose due to the non-specific symptoms like fatigue or lower abdominal pain, and the similarity with other clinical conditions such as gynecological malignancy, inflammatory bowel disease, tuberculosis, and nocardiosis [1-3]. As for the radiological findings, CT may show infiltrative nature of the disease with disruption of tissue planes and demonstrate one or more solid masses with focal low-attenuation areas or cystic masses with thickened walls [1, 2, 4]. Bacterial culturing is thought to be diagnostic, which is positive only in 50% of cases [1, 4, 5]. The diagnosis is usually made postoperatively; it has been estimated that fewer than 20 percent of cases received a preoperative diagnosis [2,

Here the authors report a case with IUD related pelvic actinomycosis. The patient had abdominal wall abscess diagnosed by CT and received medical treatment only without operation.

Case Report

Here the authors report a case of a 45-year-old Chinese female, (gravida 4 para 2 abortus 2), with a history of cesarean section and 15-year IUD placement. Her medical history was unremarkable except for hypertension, which was under medication control. She visited the emergency department due to progressing left lower quadrant pain for three weeks. She denied fever, dyspnea, nausea, vomiting, diarrhea, dysuria or abnormal vaginal discharge. Her vital signs were within normal ranges. Physical examination revealed local tenderness and redness over left lower quadrant of the abdominal wall. Per vaginal Examination showed cervical lifting pain with left adnexal tenderness. Lab investigation revealed leukocytosis (white blood cells, 12,190 cells/μL), anemia (hemoglobin 10.7g/dL), and elevated C-reactive protein level, 14.64 mg/dL. Contract CT (Figure 1) demonstrated an approximate 4.4×3.1cm-sized cystic, capsulated lesion with fine air content in left adnexa, in addition with diffuse thickening of pelvic wall muscles with fusiform appearance that was anteriorly bulging out to subcutaneous region. IUD in situ was also noted. Due to long-term IUD placement, pelvic actinomytosis was suspected initially, and then IUD was removed and cultured. Thereby, empirical intravenous cefmetazole (1,000 mg, every eight hours) and metronidazole (500 mg, every eight hours) were administered first. Then, clindamycin (900 mg, every eight hours) was added for better infection control. Diffuse pelvic wall tenderness and the margin of redness of the patient resolved gradually under antibiotics control. CT-guided percutaneous aspiration was going to be performed, but it was suspended due to her good response to medical treatment. Biochemistry analysis also indicated improving infection condition. IUD culturing showed no bacteria growth. After 14 days of hospitalization, she was discharged for outpatient follow-up with oral form of metronidazole and clindamycin use for

 $\overline{^{\dagger}Contributed}$ equally.

Published: 15 April 2020

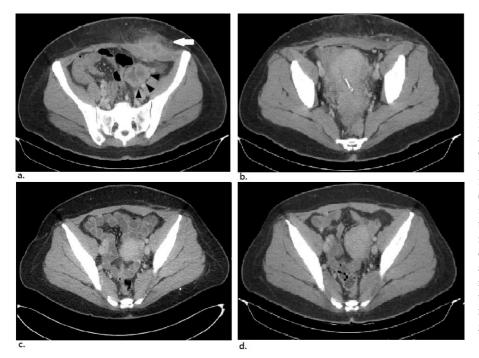


Figure 1. — (A) Diffuse thickening of left pelvic wall muscles with fusiform appearance that is anteriorly bulging out to subcutaneous region (arrow). A septate capsulated mass lesion with fine air content in the left adnexa (arrowheads). (B) Intrauterine device in situ. (C) Image with contract on the third month. Although the left adnexal mass had decreased, irregular wall of the mass and the fatty stranding of adjacent subcutaneous region are still noted. (D) Image with contract on the sixth month. A small soft tissue nodule in the pelvis, suggesting the granulation after medical treatment.

four months and laboratory testing revealed resolution of infection (C-reactive protein level, 0.03 mg/dL). Transvaginal sonography, performed on the third month after discharge, showed that the left side tubo-ovarian abscess disappeared. Abscess reduction was observed by the CT image on the third and the sixth month after discharge (Figure 1).

Discussion

Pelvic actinomycosis is characterized by slow-growing, granulomatous inflammatory response, followed by extensive reactive fibrosis and necrosis, abscesses, draining sinuses, and fistula. It is difficult to diagnose due to the non-specific symptoms including: fatigue, unintentional weight loss, lower abdominal pain, palpable mass, abnormal vaginal discharge, and related bowel obstruction [1, 5, 7]. Laboratory abnormal findings may show anemia, leukocytosis, and elevated ESR [4, 8]. The diagnosis is usually made postoperatively, since most patients undergo exploratory laparotomy for a suspected neoplasm [8]. CT seems to be helpful to define the extent of affected lesions and extramucosal involvement.

In the present case, the authors performed CT which revealed left side adnexal mass with pelvic abscess. Nevertheless, inhomogeneous pelvic masses and the infiltrative nature of vague interface with adjacent colon or bladder may sometimes be confused as malignancy or other inflammatory diseases [1].

In the suspected cases whom were examined with CT, pelvic actinomycosis was suggested preoperatively at the rate of 66% [4]. One possible route of contagion of pelvic actinomycosis is through IUDs, which has a traumatizing effect on endothelium by causing erosion and facilitates the

growth of microorganisms through wires that are left in the exocervix [3, 5]. Additionally, the IUD changes the carbohydrate metabolism in endometrial cells, developing more inflammation [3].

It is clearly associated with the duration of the IUD use; therefore, it is recommended that an IUD be replaced every five years [5]. In 2010, Jeong-Hoon et al. reviewed 16 cases of pelvic actinomycosis [4], which were diagnosed by surgery and histopathological studies. Fifty out of the 16 (93.8%) patients had IUD use, and eventually all the patients were cured with surgery and antibiotics. Timely removal of an IUD is important in cases suspected with IUD-associated actinomycosis [5, 9] as in the present case. Echo or CT-guided percutaneous aspiration may relieve the severity of the disease and is also a good method for collecting material for culturing [2]. Bacterial culturing is thought to be diagnostic, and the typical microscopic findings include necrosis with yellowish sulfur granules and filamentous Gram-positive fungal-like pathogens, which are only found in 50% of cases [1, 4, 5].

In the present case, IUD culturing showed no bacteria detected. Therefore, histopathological examination is the definitive diagnostic method, which is generally conducted after surgical intervention [1-3]. For uncomplicated cases, the standard antimicrobial treatment is intravenous penicillin G (10 to 20 million units per day divided every four to six hours) for a prolonged period, which requires hospitalization up to 4-6 weeks, and followed by oral form of penicillin (2 to 4 g/day) or amoxicillin for 2-12 months [2, 3, 8].

Concerning long hospitalization time, some studies reported successful therapy with oral or intramuscular form of antibiotics before surgery, which is more convenient for outpatient management. Onal et al. reported three cases with pelvic actinomycosis successfully treated with daily intramuscular ceftriaxone as an adjunctive therapy to surgery and short term IV penicillin [8]. As for the patients with extensive abscesses and obstructive symptoms, invasive intervention, such as drainage or resection of infected and necrotic tissues should be considered. Although the complication associated with percutaneous drainage of pelvic abscesses, such as damage to adjacent organs (the bladder, uterus or colon), puncture site pain, nerve injury, and hematoma [6], rarely occur, it is an alternative method to treat deep pelvic abscess for cases with poor response to antibiotics, and the duration of antimicrobial therapy could be reduced [3]. Early identification and treatment of actinomyces, as well as elaborate preoperative evaluation could have helped to reduce treatment-related morbidity and a overall improve patient outcomes and safety [7].

The present case was interesting in several aspects. After initial inspection of the patient, pelvic actinomycosis was suspected, so the IUD was removed promptly. Then, empirical antibiotic treatment was administered. On the other hand, the severity of the disease observed from CT, which revealed septate pelvic mass with subcutaneous region invasion was very similar to pelvic malignancy. However, at the request of the patient, the authors conducted a relatively conservative treatment and subsequently it was proved to be effective. Thus, they avoided unnecessary percutaneous abscess drainage or surgical intervention.

Although the medical treatment of choice commonly suggested is parenteral antibiotic use for at least four weeks, along with oral form afterwards for more than two months, the present patient received intravenous antibiotic therapy only for two weeks, and a significant improvement in symptoms was detected. During the periodic follow-up examinations, laboratory testing, and imaging studies of the patient showed no sign of recurrence. CT is also used for monitoring the radiologic response to treatment during follow-up examinations.

Conclusion

Pelvic actinomycosis should always be considered in patients with pelvic abscess and the prolonged use of IUD. It is difficult to give a precise diagnosis preoperatively due to the characteristics of the disease having similarity with malignancy and other inflammatory disease. Detailed history taking, laboratory investigation and image survey may assist in the diagnoses of actinomycosis. The medical treatment of choice commonly suggested is parenteral antibiotic use for at least four weeks. In the case of an event when empirical conservative antibiotics provided no relief, percutaneous drainage of pelvic abscesses or surgical intervention is still indicated.

References

- [1] Yang S.S., Im Y.C.: "Severe abdominopelvic actinomycosis with colon perforation and hepatic involvement mimicking advanced sigmoid colon cancer with hepatic metastasis: a case study". BMC Surg., 2018, 18, 51.
- [2] Kim M., Ko J., Lee C.: "Pelvic actinomycosis with abundant ascites, pleural effusion, and lymphadenopathy diagnosed with endometrial biopsy and treated with medication only". *Taiwan J. Obstet. Gy*necol., 2014, 53, 588.
- [3] Garcia-Garcia A., Ramirez-Duran N., Sandoval-Trujillo H., Romero-Figueroa M.D.S.: "Pelvic Actinomycosis". Can. J. Infect. Dis. Med. Microbiol., 2017, 2017, 9428650.
- [4] Bae J.H., Song R., Lee A., Park J.S., Kim M.R.: "Computed tomography for the preoperative diagnosis of pelvic actinomycosis". J. Obstet. Gynaecol. Res., 2011, 37, 300.
- [5] Valour F., Senechal A., Dupieux C., Karsenty J., Lustig S., Breton P., et al.: "Actinomycosis: etiology, clinical features, diagnosis, treatment, and management". *Infect. Drug Resist.*, 2014, 7, 183.
- [6] Inatomi A., Tsuji S., Amano T., Kobayashi M.: "Successful treatment of pelvic actinomycosis using transgluteal drainage: A case report". J. Infect. Chemother., 2016, 22, 567.
- [7] Sawtelle A.L., Chappell N.P., Miller C.R.: "Actinomyces-Related Tubo-Ovarian Abscess in a Poorly Controlled Type II Diabetic With a Copper Intrauterine Device". *Mil. Med.*, 2017, *182*, e1874.
- [8] Onal E.D., Altinbas A., Onal I.K., Ascioglu S, Akpinar M.G., Himmetoglu C., Sardan Y.C.: "Successful outpatient management of pelvic actinomycosis by ceftriaxone: a report of three cases". *Braz. J. Infect. Dis.*, 2009, 13, 391.
- [9] Pillai M., Van de Venne M., Shefras J.: "Serious morbidity with long-term IUD retention". J. Fam. Plan. Reprod. Health Care, 2009, 35, 131.

Corresponding Author:
PEI-SHEN HUANG, M.D.
250 Wuxing St.
Taipei 11031 (Taiwan)
e-mail: muse caffeine@hotmail.com