



Case Series

Pelvic actinomycosis: A confusing diagnosis[☆]

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ARTICLE INFO

Keywords:

IUD contraception
Actinomycosis
Gynecologic surgery
Case series

ABSTRACT

Introduction and importance: Actinomycosis is a rare chronic and suppurative infection caused by anaerobic Gram Positive bacteria: actinomycetes. Pelvic location is extremely rare, usually associated with history of IUD contraception and doesn't have specific signs. Pelvic actinomycosis diagnosis may be confused with pelvic gynecologic malignancies or abscess. We present a retrospective and descriptive study of twelve patients with pelvic actinomycosis diagnosed and managed in our department from January 2000 to December 2011.

Cases presentation: The patients' mean age was 47 years. 75% of them had a history of IUD for a mean period of 8,44 years. Pelvic pain was the most common complaint. In four cases, pre-operative clinical presumption was tubo-ovarian abscess. Gynecologic malignancies were suspected in 8 patients. Pelvic actinomycosis management was based on surgery and long-term antibiotic.

Clinical discussion: Pelvic actinomycosis is an extremely rare chronic infection, presenting 3% of human actinomycosis. Common clinical presentations include vaginal discharge, tubo-ovarian abscess and pelvic tumors mimicking gynecologic malignancies. It is difficult to diagnose. Association with IUD history was recognized. Management is based on surgery and long-term antibiotic administration.

Conclusion: Pelvic actinomycosis is an extremely rare chronic infection. This entity is difficult to diagnose. Accurate diagnosis can reduce complications and unnecessary surgeries, and can preserve fertility.

Case Series

1. Introduction

Actinomycosis is a rare chronic, suppurative and granulomatous disease. It is caused by actinomycetes, a Gram-positive anaerobic bacteria [1]. Pelvic actinomycosis location is extremely rare presenting 3% of all human actinomycosis [2]. The common clinical presentations include vaginal discharge, tubo-ovarian abscess and pelvic tumors mimicking gynecologic malignancies. The management of this infectious disease is based on surgery and long-term antibiotic therapy. The aims of this study are to describe clinical, biological, imaging and therapeutic features of pelvic actinomycosis. The work has been reported in line with the SCARE criteria [3].

2. Materials and methods

This is a retrospective and descriptive study enrolling twelve patients with pelvic actinomycosis diagnosed and managed in our departments from January 2000 to December 2011. The cases were identified from the pathological report and all data were retrospectively analyzed. All surgical procedures were performed by senior gynecologic surgeons.

3. Results

The patients' mean age was 47 years (ranging from 29 to 70 years). Nine patients (75%) had history of IUD for a mean period of 8,44 years (ranging from 3 to 13 years). Pelvic pain was the most reported complaint (n = 8), fever was present in 4 cases and vaginal bleeding was noticed in 3 cases.

[☆] The manuscript has not been previously published and is not being concurrently submitted elsewhere.

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<https://doi.org/10.1016/j.ijscr.2021.106387>

Received 6 August 2021; Received in revised form 2 September 2021; Accepted 2 September 2021

Available online 6 September 2021

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Fig. 1. Pelvic ultrasound imaging showing a mixed (solid and cystic) pelvic tumor.



Fig. 2. Transvaginal ultrasound imaging showing an intrauterine proliferation.

Clinical examination found pelvic mass in 4 cases. Lower-abdominal pain was discovered by palpation in 8 cases. All patients had transvaginal ultrasonography examination. It revealed complex pelvic tumor measuring meanly 7,7 cm (ranging from 5,4 to 14 cm) in 8 cases (Fig. 1). In four cases, transvaginal ultrasonography examination showed intrauterine heterogeneous proliferation (Fig. 2).

Basic laboratory examination was performed to all patients, it showed WBC's high level in 7 cases (from 13,400 to 27,600/ml), anemia was found in 10 cases. Blood CRP level, performed in 7 cases, was elevated. Its mean level was 85,71 U/l (it ranges from 34 to 138 U/l). CA-125 serum level was performed in 8 patients, it measured meanly 57,55 U/l (range, 35,12–91,51 U/l). Clinical, biologic and imaging features are reported in Table 1.

Nine patients underwent surgery. Intraoperative findings were:

- isolated tubo-ovarian abscess in 4 cases
- tubo-ovarian abscess extended to appendix and omentum in one case
- bilateral tubo-ovarian abscess in one case
- tubal abscess without ovarian extension in one case
- tubo-ovarian abscess with tubo-intestinal fistula in one case.

Unilateral salpingo-oophorectomy was performed laparoscopically in two cases. Open surgery was needed in 4 cases to perform:

- Total hysterectomy with bilateral salpingo-oophorectomy in two cases
- Right salpingo-oophorectomy with appendectomy and partial omentum removal in one case
- Hysterectomy, bilateral salpingo-oophorectomy and bowel resection in one case (to manage tubo-intestinal fistula).

Endometrial biopsy was performed in three patients for genital bleeding.

Histologic examination showed diffuse and chronic inflammatory reaction with giant multinucleated cells and intermixed actinomycotic colonies in 7 cases. It was unhelpful in 5 cases showing no specific inflammation, diagnosis was suggested by medical history and clinical presentation.

The mean duration of intravenous antibiotic therapy was 6 months. All patients had amoxicillin- clavulanic acid, metronidazole and gentamycin as first line intravenous antibiotic switched to oral route based on penicillin V as 2-4 g/day. At 9 months-follow-up, all patients had positive outcome.

4. Discussion

Actinomycosis is a chronic infectious disease caused by actinomycetes, a gram-positive anaerobe [1]. Female genital tract infection is usually the result of ascending infection from uterine cavity. The microorganism became pathogenic when uterine mucous membrane is distorted specially by an IUD. The association IUD- Actinomycosis was established

Table 1
Clinical, biologic and imaging findings. **Table 1:**

| Cases | Age (years) | IUD (years) | Complaints | WBC X1000/ml | CRP mg/l | CA125 U/l | Transvaginal ultrasonography | Tumor size cm | Pre op presumption |
|-------|-------------|-------------|----------------------|--------------|----------|-----------|---------------------------------------|---------------|-----------------------------------|
| 1 | 46 | 13 | Pelvic pain Fever | 19,9 | 69 | – | Cystic | 5,4 | TOA |
| 2 | 42 | 07 | Pelvic pain | 27,6 | 79 | 35,12 | Pelvic solid tumor | 6,2 | Ovarian cancer |
| 3 | 29 | 03 | Pelvic pain Fever | 13,4 | 133 | – | Cystic | 6,9 | TOA |
| 4 | 46 | No IUD | Pelvic pain | 19,2 | 90 | 77,8 | Pelvic mixed tumor (solid and cystic) | 14 | Ovarian cancer |
| 5 | 54 | 09 | Pelvic pain | 8,7 | – | 91,51 | Pelvic mixed tumor (solid and cystic) | 10,5 | Ovarian cancer |
| 6 | 61 | 14 | Vaginal bleeding | 7,6 | – | 75 | Intrauterine proliferation | 03 | Endometrial cancer |
| 7 | 45 | 04 | Pelvic pain Fever | 17,6 | 138 | – | Cystic | 4,7 | TOA |
| 8 | 32 | No IUD | Vaginal bleeding | 10,1 | – | 52 | Intra uterine proliferation | 2,7 | Hyperplasia or Endometrial cancer |
| 9 | 29 | No IUD | Pelvic pain Fever | 18,4 | 57 | – | Cystic | 6,2 | TOA |
| 10 | 56 | 05 | leucorrhoea | 6,7 | – | 48 | Intra uterine proliferation | 3,1 | Endometrial cancer |
| 11 | 54 | 09 | Pelvic pain | 16,6 | 34 | 42 | Pelvic mixed tumor | 7,7 | Ovarian or digestive cancer |
| 12 | 70 | 12 | Vaginal bleeding | 9,3 | – | 39 | Intrauterine proliferation | 3,6 | Endometrial cancer |

firstly in 1970 [4,5]. Actually, this association is well documented [6,7].

Fiorino's study of 92 cases of pelvic actinomycosis noticed this association with 8 years of IUD history. The mean age of diagnose was 37 years (range, 20–77 years), 65% of the patients had less than 40 years. Clinic presentation is much diversified and no specific. The most common symptom was abdominal and pelvic pain (85%) and fetid vaginal loss in 24% of cases [6,8]. In laboratory examination, a high level of WBC is noticed with anemia which been revealed in 70% of cases [6,8]. Ultrasonography and other radiologic examination show no specific or suggestive signs, they reveal mostly a solid or cystic mass with thickened wall mimicking ovarian malignancy [2,9–11]. The diagnosis is based on bacteriologic or pathologic examination [1,12]. Bacteriologic culture must be in anaerobic environment with examination in Gram coloring what allows to spot gram positive bacillus [11,13,14]. Bacteriologic determination of this bacillus is so hard because of its sensibility to oxygen, it's difficult and slow growth in culture and its association with other anaerobic germs. Bacteriologic identification is made only in 50% of cases [15].

Once pelvic actinomycosis is diagnosed, IUD if it exists, must be removed and penicillin administration must be started [16,17]. Tetracycline, clindamycin and erythromycin can be used in case of allergy to penicillin [6].

Surgery consists on abscess incision and drainage with excision and removal of fibrosis and infected tissues. Intravenous administration of 10–20 millions of penicillin G for 2 weeks followed by oral route of penicillin V (2–4 g/day) is recommended [18] for several months due to avascular and necrotic lesions and anaerobic nature of the actinomycotic infection [19].

Without surgical approach, recurrence and complications may occur as extension to pelvic organs, digestive or cutaneous fistula and systemic dissemination [20].

5. Conclusion

Pelvic actinomycosis is an extremely rare chronic infection, presenting 3% of human actinomycosis. It is difficult to diagnose. Accurate diagnosis can reduce complications and unnecessary surgeries and may preserve fertility. Management strategy is based on long-term administration of antibiotics and surgery to avoid complications and recurrence.

CRedit authorship contribution statement

Mohamed Aymen Ferjaoui and Ramzi Arfaoui: surgery and study design.

Slim Khedhri, Mohamed amine Hannechi and Kais Abdessamia: manuscript redaction.

Khaoula Samaali, Wissal Fezai and Marwa Salhi: Data collection.

Monia Malek and Khaled Neji: Manuscript supervisors.

Declaration of competing interest

None.

No source of funding to declare.

IRB exemption was obtained before submission.

Provenance and peer review.

Not commissioned, externally peer-reviewed.

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