Recurrent pelvic actinomycosis: a case report

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ABSTRACT

Background: Actinomycosis is a chronic granulomatous disease caused by an obligate anaerobic species from the genus *Actinomyces*.

Case Presentation: We describe the case of a 36-year-old woman, who presented with recurrent episodes of abdominal pain since 2002. These bouts of abdominal pain increased in severity over the last 4 years, and the patient was subsequently diagnosed to have actinomycosis.

Conclusion: Although intra-abdominal actinomycosis is very rare, it should be considered in the differential diagnosis of abdominal pain, particularly in any women using an intra-uterine device and presenting with abdominal pain or a pelvic mass. If actinomycosis is suspected preoperatively, appropriate handling and processing of cultures can increase the diagnostic yield and may save the patient from an extensive surgery.

Keywords: Actinomycosis, CT (computerized tomography), granulomatous, fungal, IUCD (intra-uterine contraceptive device).

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Background

Actinomycosis is a chronic granulomatous disease, caused by the genus *Actinomyces* species, which are obligate anaerobes. In the past, it was thought to be a fungal infection, but later the causative organisms of this disease were proved to be Gram-positive filamentous bacteria. Actinomycosis in humans is most commonly caused by the species *Actinomyces israelii* [1]. They are not considered remarkably virulent pathogens, but rather as opportunistic ones as the infection usually occurs only after the mucous membranes are disrupted. *Actinomyces israelii* is considered as a common commensal of the oropharynx, gastrointestinal tract, and the vagina [2,3].

The disease spreads by direct extension into surrounding tissues regardless of tissue planes through the formation of sinus tracts that can lead directly to the skin. There is no documented person-to-person transmission of this disease. Typically sulfur granules can drain from these tracts, but it is not a pathognomonic feature. This case of ours adds to the growing body of literature that actinomycosis should be considered in a list of differential diagnosis of abdominal pain, particularly in any women using an intra-uterine device and presenting to the clinic with abdominal pain or a pelvic mass.

Case Presentation

A 36-year-old single lady presented to King Edward Memorial Hospital, Mumbai, India with several episodes of abdominal pain, since July 2002, which had increased in severity for last 4 years. Physical examination elicited abdominal pain which was moderate-severe and not radiating to the legs. She had a past surgical history of tubectomy with no complications in 2002. USG abdomen and pelvis in 2006 showed an enlarged left ovary measuring 5 × 3 cm having a cyst of 3.5 × 3.5 cm s/o chocolate cyst. Laparoscopic removal of this ovarian cyst was done after a couple of months. Histopathology report was suggestive of fungal infection like *Actinomyces*. The patient was given IV broad antibiotic coverage followed by an oral course on discharge. The patient had subjective improvement, but the symptoms gradually returned in 2012 and she reported to the hospital by 2014. At that point, computerized tomography (CT) scan of the abdomen and pelvis showed a soft tissue mass of size 3.4 × 2.6 cm in left adnexa and another cystic lesion sized 5.5 × 3.2 cm was noted in the right iliac fossa Figures(1 – 3). The patient was again treated with broad antibiotic coverage for actinomycosis but failed to show any response. Ultimately, she underwent diagnostic laparoscopy followed by ovarian cystectomy, salpingectomy, and omental mass removal in February 2015. Slide preparation of the block dissection of pelvic adnexal mass was suggestive of an actinomycotic abscess. The patient was discharged from the hospital and was comfortable until August 2015. She returned with similar complaints as earlier soon, however, the gastrointestinal (GI) symptoms were mild. On physical exam, she had nodularity felt in the Pouch of Douglas. The

CT scan abdomen & pelvis revealed an ill-defined, infiltrative plaque-like lesion, located in the recto-uterine pouch, bilateral adnexal regions, and also involving peritoneal surfaces of these regions. Furthermore, there was involvement of distal half of sigmoid colon and the recto-sigmoid junction with encasement of almost entire uterus. The patient again underwent an exploratory laparotomy in November 2015, but surgery was abandoned since the mass was proven through naked eyes, to be plastered to the various organs and peritoneum. Patient consent was taken for use of patient-related information for possible publication. Unfortunately, the patient was lost to further follow-up.

Discussion

The most common site of human actinomycosis is the cervicofacial region, which is known to be affected in about 40%–50% of cases. About 15% of the cases occur in the thorax [4]. Pelvic actinomycosis occurs almost always in women only. It may conceivably mimic pelvic malignancies or even retroperitoneal tumors [5] and therefore diagnosing it generally is problematic. Almost 85% of cases of pelvic actinomycosis occur in women, who have had an intra-uterine contraceptive device (IUCD) *in situ* for more than 3 years [6]. *Actinomyces israelii* infects 1.65%–11.6% of IUCD users [7]. Due to its invasion of surrounding tissues and the formation of masses, it is often confused with an ovarian neoplasm [8–10]. Usually, the symptoms include lower abdominal pain, cachexia, vaginal discharge, and nausea [11]. As the disease advances, tubo-ovarian abscess formation occurs. This may lead to a "frozen pelvis," mimicking pelvic cancer [11].

The gold standard for diagnosis is culture from the tissue, pus, or IUCD itself. *Actinomyces* species is a slow-growing, anaerobic bacterium that requires 2–3 weeks to culture and with a failure rate of more than 50% [12]. Therefore, most diagnoses are made histologically with the presence of sulfur granules, which are yellowish particles with a mycelium-like structure containing clusters of neutrophils and *Actinomyces*. Sulfur granules may be scant or absent in some patients [13] and their presence is not pathognomonic of *Actinomyces* infection [12].

Peabody and Seabury [14] endorsed abscess drainage along with high-dose antibiotics as the principal therapy for actinomycosis. *Actinomyces* species are known to be susceptible to a broad range of beta-lactam agents and, when given with beta-lactamase inhibitors, they are deemed as the first choice [15]. Smith et al. [16] described that ciprofloxacin and tetracyclines showed inadequate performance against *Actinomyces* species.

Although there are contrasting opinions in the literature about dosage and duration of antibiotic treatment, antibiotics can medically treat uncomplicated actinomycosis [15]. The antibiotics have poor penetration into the fibrous tissues and so a long course of treatment is required. In advanced and complicated actinomycosis, there are abundant avascular spaces are present due to severe tissue reactions. In such cases, medical therapy may be less effective, resulting in a longer duration of antibiotic treatment, regardless of the site of actinomycosis. It was interestingly noted that two patients in one series (9.0%) who were treated with IV antibiotics after surgery for less than 4 weeks did not recur during the last 34 months of follow-up. Therefore, the

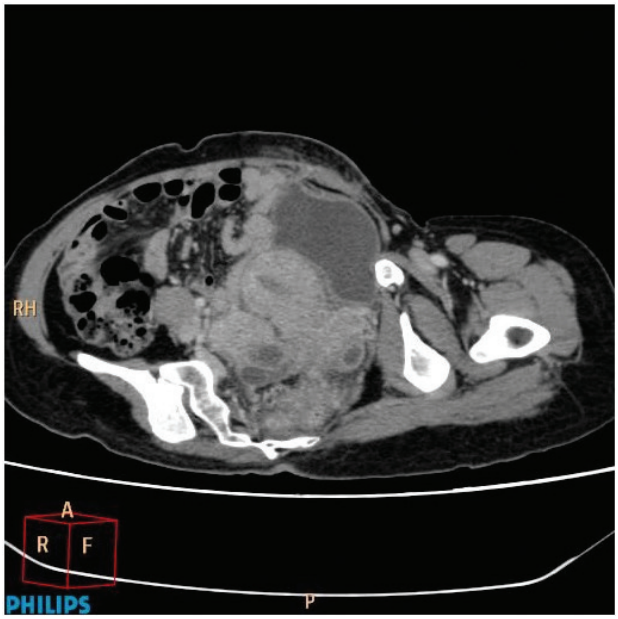

Figure 1. *Heterogeneously enhancing soft tissue lesion is seen in bilateral adnexa with non-enhancing thick-walled collections in bilateral adnexa.*

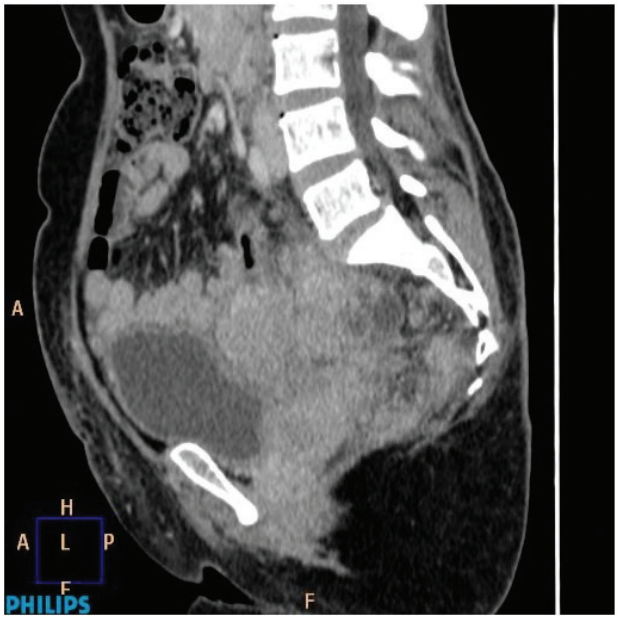

Figure 2. *Heterogeneous soft tissue is seen involving the presacral space and recto-uterine space.*

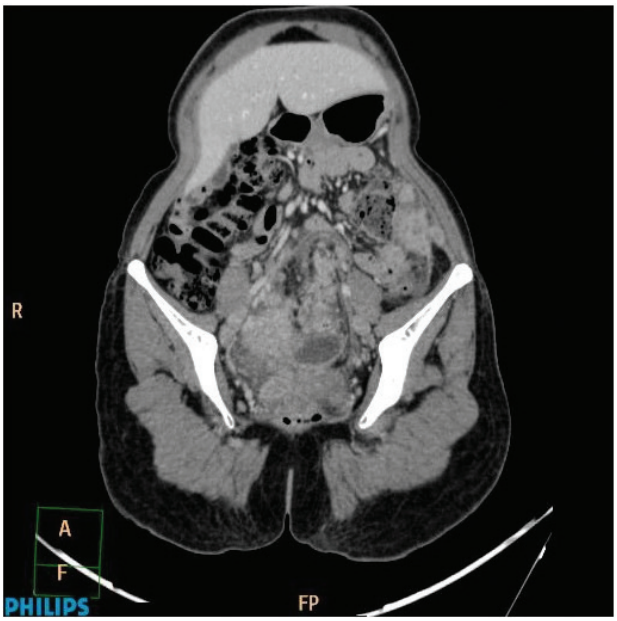

Figure 3. *Pelvic tubo-ovarian mass is seen with a heterogeneously enhancing lesion along the parietal peritoneal wall on the left side.*

clinical impact of surgical resection followed by short-term antibiotics merits, further study [15]. However, no complicated case like ours exists in the prior published literature.

Conclusion

Although intra-abdominal actinomycosis is very rare, it should be considered in the differential diagnosis of recurrent abdominal pain and pelvic mass particularly in women with a history of IUCD use. If actinomycosis is suspected preoperatively, appropriate handling of cultures will increase the diagnostic yield, which may obviate the need for an extensive surgery. Furthermore, early empirical treatment with appropriate antibiotic coverage for suspected actinomycosis is another practical solution.

Acknowledgement

None.

List of abbreviations

CT	Computerized tomography
IUCD	Intra-uterine contraceptive device

Consent for publication

Informed consent was taken from the patient.

Ethical approval

Ethical approval is not required at our institution to publish an anonymous case report.

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Specialty

Internal Medicine

References

1. Bennhoff DF. Actinomycosis: diagnostic and therapeutic considerations and a review of 32 cases. Laryngoscope 1984; 94(9):1198–217; https://doi.org/10.1288/00005537-198409000-00013
2. Sehouli J, Stupin JH, Schlieper U, Kuemmel S, Henrich W, Denkert C, et al. Actinomycotic inflammatory disease and misdiagnosis of ovarian cancer. A case report. Anticancer Res 2006; 26(2C):1727–31.
3. Kim YJ, Youm J, Kim JH, Jee BC. *Actinomyces*-like organisms in cervical smears: the association with intrauterine device and pelvic inflammatory diseases. Obstet Gynecol Sci 2014; 57(5):393–6; https://doi.org/10.5468/ogs.2014.57.5.393
4. Weese WC, Smith IM. A study of 57 cases of actinomycosis over a 36-year period: a diagnostic failure with good prognosis after treatment. Arch Intern Med 1975; 135(12):1562–8; https://doi.org/10.1001/archinte.1975.00330120040006
5. Westhoff C. IUDs and colonization or infection with *Actinomyces*. Contraception 2007; 75(6):S48–50; https://doi.org/10.1016/j.contraception.2007.01.006
6. Schmidt WA. IUDs, inflammation, and infection: assessment after two decades of IUD use. Hum Pathol 1982; 13(10):878–81; https://doi.org/10.1016/S0046-8177(82)80046-4
7. McCormick JF, Scorgie RD. Unilateral tubo-ovarian actinomycosis in the presence of an intrauterine device. Am J Clin Pathol 1977; 68(5):622–6; https://doi.org/10.1093/ajcp/68.5.622
8. Akhan SE, Dogan Y, Akhan S, Iyibozkurt AC, Topuz S, Yalcin O. Pelvic actinomycosis mimicking ovarian malignancy: three cases. Euro J Gynaecol Oncol 2008; 29(3):294–7.
9. Choi MH, Hong DG, Seong WJ, Lee YS, Park IS. Pelvic actinomycosis confirmed after surgery: single center experience. Arch Gynecol Obstet 2010; 281(4):651–6; https://doi.org/10.1007/s00404-009-1157-2
10. Kumar N, Das P, Kumar D, Kriplani A, Ray R. Pelvic actinomycosis mimicking: an advanced ovarian cancer. Indian J Pathol Microbiol 2010; 53(1):164–5; https://doi.org/10.4103/0377-4929.59216
11. Lely RJ, van Es HW. Case 85: pelvic actinomycosis in association with an intrauterine device. Radiology 2005; 236(2):492–4; https://doi.org/10.1148/radiol.2362031034
12. Wong VK, Turmezei TD, Weston VC. Actinomycosis. BMJ 2011; 343: d6099; https://doi.org/10.1136/bmj.d6099
13. Brown JR. Human actinomycosis. A study of 181 subjects. Hum Pathol 1973; 4(3):319–30; https://doi.org/10.1016/S0046-8177(73)80097-8
14. Peabody JW Jr, Seabury J. Actinomycosis and nocardiosis. A review of basic differences in

therapy. Am J Med 1960; 28:99–115; https://doi.org/10.1016/0002-9343(60)90226-6
15. Kaya E, Yilmazlar T, Emiroğlu Z, Zorluoğlu A, Bayer A. Colonic actinomycosis: report of a case and review of the literature. Surg Today 1995; 25(10):923–6; https://doi.org/10.1007/BF00311761
16. Smith AJ, Hall V, Thakker B, Gemmell CG. Antimicrobial susceptibility testing of *Actinomyces* species with 12 antimicrobial agents. J Antimicrobial Chemotherapy 2005; 56(2):407–9; https://doi.org/10.1093/jac/dki206

Summary of the case

Patient (gender, age)	1	Female, 36
Final diagnosis	2	Pelvic actinomycosis
Symptoms	3	Abdominal pain
Medications (Generic)	4	Antibiotics
Clinical procedure	5	Computerized tomography
Specialty	6	Infectious diseases