

Pelvic Actinomycosis: A report of two cases with different presentations

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SUMMARY

Actinomycosis is a type of bacterial infection which manifests in a subacute and chronic manner. It is characterized by a contiguous spread with suppuration and granulomatous inflammation. *Actinomyces* is a normal flora inhabiting the oral cavity, gastrointestinal and urogenital tract. The commonest type of infection is the cervicofacial followed by abdominal and thoracic type. Pelvic actinomycosis is a subtype of abdominal actinomycosis and it is commonly associated with intrauterine contraception device (IUCD) use. The most challenging aspect in management of actinomycosis is to reach the diagnosis early. In cases diagnosed early, removal of IUCD with antibiotic treatment may lead to complete remission averting operative measure and its complications. We report two cases of pelvic actinomycosis with different presentations. The first case was diagnosed after patient was operated for suspected gynaecological malignancy while the second case was diagnosed with Papanicolaou smear from routine cervical cancer screening.

Keywords: Actinomycosis; pelvic; papanicolaou smears, intrauterine device; antibiotic

INTRODUCTION

Actinomycosis is a suppurative disease and often associated with granulomatous inflammation. The causative agent is an anaerobic filamentous-like gram-positive bacteria named *Actinomyces israelii*. It is an uncommon infection that manifests in a subacute and chronic manner whereby patients may present with fistula, sinus and abscess formation.¹ Occasionally, the clinical presentation mimics malignancy characterized by extensive contiguous spread regardless of tissue planes.² *Actinomyces israelii* is human's commensal flora residing in oral cavity, gastrointestinal and urogenital tract.¹ It acquires its pathogenicity through invasion of breached local tissue. There are three types of actinomycosis which are the cervicofacial, thoracic, and abdominal pelvic. Cervicofacial actinomycosis is the commonest type which accounted for approximately 50% of the cases, followed by the thoracic and abdominal type with around 20% of the cases for both.³ Pelvic actinomycosis is a subtype of abdominal actinomycosis and the cases reported in literature so far are limited.¹ The following two cases of pelvic actinomycosis will demonstrate the heterogeneity of its clinical presentation from being asymptomatic to full blown infection with giant size tumour.

CASE 1

Madam LGY is a 39 years old teacher who came to review her recently taken cervical cancer screening result. The Papanicolaou (Pap) smear reported presence of organism resembling *Actinomyces* albeit absence of early signs of cervical cancer. On further questioning, she had infrequent yellowish per vaginal discharge for two months. However, she denied abdominal pain, fever, dyspareunia and sexual

promiscuity. Per speculum examination revealed moderate amount of yellowish discharge with normal cervix and bedside pelvic ultrasound found intrauterine contraceptive device (IUCD) in-situ which she had forgotten about and three years past its due date for removal. IUCD was removed and Madam LGY was treated with stat dose of intramuscular Ceftriaxone 500mg and tablet Azithromycin 1g. Her symptoms resolved during her next review and IUCD for culture & sensitivity (C&S) found *Actinomyces* growth while other laboratory tests excluded sexually transmitted diseases. Diagnosis of pelvic actinomycosis was made and she was treated with oral phenoxymethylpenicillin for six months. She was asymptomatic during treatment and repeated Pap smear was normal.

CASE 2

Madam NA is a 52 years old housewife who presented with complaint of worsening abdominal pain for one week preceded by gradual abdominal distention for three years. She had history of retained IUCD for ten years and it was removed three years ago. Abdominal examination revealed a pelvic mass measured up to 20 weeks in size which was reported to be left ovarian malignancy with local infiltration from computed tomography (CT) scan (Figure 1). Exploratory laparotomy performed found left lateral pelvic and retroperitoneal tumor infiltrated mid-sigmoid colon (Figure 2A & 2B). Total abdominal hysterectomy with bilateral salphingo-oophorectomy and omentectomy were performed and Madam NA was discharged well after four days of monitoring in the ward. Histopathological examination (HPE) found *Actinomyces* infection (Figure 3) in the left ovary and surrounding tissues confirming the

diagnosis of pelvic actinomycosis rather than ovarian malignancy. She completed six months of oral phenoxymethylpenicillin and remained asymptomatic throughout follow up. Her repeated CT scan after completion of antibiotic was reported to be normal.

Figure 1: Heterogeneous mass mixed solid and cystic component at left lower abdomen and pelvis from left ovary.



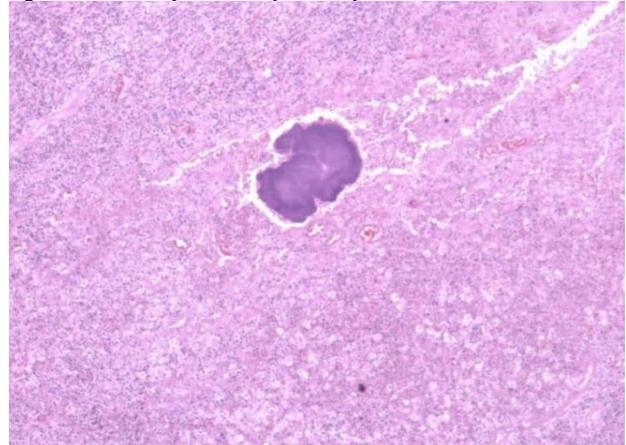
Figure 2A: Specimen post-operatively (Total abdominal hysterectomy with bilateral salpingo-oophorectomy and omentectomy with adhered sigmoid colon).



Figure 2B: Granulomatous changes noted from left ovary.



Figure 3: *Actinomyces* colony in ovary.



DISCUSSION

Diagnosing pelvic actinomycosis is a challenge for healthcare providers because patient may present in different and atypical manner. Madam LGY's diagnosis was confirmed via IUCD C&S which was ordered after *Actinomyces* was found in routine Pap smear. Finding from the latter investigation was neither diagnostic nor predictive of actinomycosis because *Actinomyces* is a common flora in female genital tract². It is supported by a review article that 0.3% of 20,000 routine Pap smear which detected *Actinomyces*-like organisms, only half of them had a positive culture result⁴. Cervical culture is the gold standard for *Actinomyces* identification but Pocius et al opined that it is not useful for clinical practice⁵. Therefore, they suggested that IUCD C&S to assist in diagnosing and guiding management of pelvic actinomycosis.

Madam NA presented with suspected gynaecological malignancy and her diagnosis was confirmed from HPE result obtained post-operatively. It concurred with a review by Wong et al and other case reports that actinomycosis commonly presented with clinical, laboratory and imaging results suggesting malignancy¹. Only less than 10% of cases were diagnosed pre-operatively by which CT scan is the most useful modality (albeit not specific) as compared to other investigations^{1,6,7}. Contrary to that, Madam NA's CT scan missed the diagnosis and she underwent major operation. If it was detected before operative measure, administration of antibiotic coupled with regular radiological monitoring may produce cure while avoiding potential surgical risk such as injury to surrounding intra-abdominal and reproductive organs, post-op infection and anaesthetic risks^{1,2}.

Of note, the two cases had a similarity which was history of retained IUCD which is a common predisposing cause. Multiple cases were reported with similar coincidence, commonly with the extended usage of IUCD averaged around seven years⁸⁻¹⁰. However, these findings did not amount to either an association or causative relationship because case reports generally have a low evidence level. On further literature search, studies to establish association of IUCD use with abdominopelvic actinomycosis were also scarce. Thus, healthcare providers should be alert to remove IUCD on time and also

have high index of suspicion of actinomycosis if patient has symptoms suggestive of abdominal cancers coupled with extended use of IUCD. Generally, removal of IUCD is not mandatory for women with clinical features suggestive pelvic inflammatory diseases (PID) or Pap smear with incidental finding of *Actinomyces* species⁵. However, Madam LGY's IUCD was removed as recommended by guideline due to presence of both factors¹¹. This recommendation is due to the fact that *Actinomyces* flourishes well on foreign bodies (IUCD) and becomes pathogenic which may result in worsening of actinomycosis infection.

Actinomyces is known to be sensitive to penicillin. Intravenous penicillin G for about six weeks, followed by oral penicillin for four to six months are recommended for large abdominal lesion with abscesses or sinus tracts¹. If patients are allergic to penicillin, tetracycline and macrolides group are suggested as an alternative antimicrobial.¹ Antibiotic recommendation for pelvic actinomycosis confirmed via IUCD C&S without abdominal mass was not specific. It was stated that high dose antibiotic for minimum of eight weeks and consultation with microbiologist are recommended.¹¹ The duration of antibiotic for treatment is controversial, ranging from four weeks to six months but majority practiced or recommended the longer duration of therapy.^{1-3,10} Study to determine antibiotic duration is required to prevent unnecessary long term antibiotic usage causing resistant organisms and side effects. Development of antibiotic resistance as is known is a leading cause of preventable deaths worldwide.¹² However, both of our patients responded well with six months of oral penicillin and repeated investigations showed disease clearance without any complications or recurrences.

CONCLUSION

Pelvic actinomycosis is a rare chronic suppurative infection with a diverse clinical presentation which includes massive abdominal distension, pelvic inflammatory disease or sometimes asymptomatic altogether and detected accidentally through a routine Pap smear. Due to similarity with cancer presentation, surgery is often performed. Therefore, high index of suspicion is required to diagnose it especially patient who has extended use of IUCD. If diagnosed early, management with removal of IUCD and

long term antibiotics may avert surgical intervention and its associated complications.

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