

Case Report

Thrombocytosis and small bowel perforation: unusual presentation of abdominopelvic actinomycosis

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Abstract

Intrauterine devices (IUD) are frequently used as a family planning procedure in developing countries because they are easy to administer and governmental policies support their use in many countries. It is recommended that IUDs be removed or replaced after 10 years, but longer use is common, especially in developing countries. In some cases, rare infections such as pelvic inflammatory diseases, pelvic tuberculosis, or abdominopelvic actinomycosis related to IUD can develop.

Pelvic actinomycosis is a rare disease and is often diagnosed incidentally during surgery. In recent years, there has been an increase in actinomycotic infections mostly due to long-term usage of IUD and forgotten intravaginal pessaries. It usually develops as an ascending infection. It is usually associated with non-specific symptoms such as lower abdominal pain, menstrual disturbances, fever, and vaginal discharge. The disease is sometimes asymptomatic. The rate of accurate preoperative diagnosis for pelvic actinomycosis is less than 10%, and symptoms and imaging studies sometimes mimic pelvic malignancy. This report details a case with abdominopelvic actinomycosis associated with an IUD presenting with highly elevated thrombocyte count and small bowel perforation with abscess formation.

Key words: abdominopelvic actinomycosis; intrauterine device; small bowel perforation; thrombocytosis

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Introduction

Actinomycosis is a chronic suppurative and granulomatous infection caused by an anaerobic non-spore forming Gram-positive bacteria of the genus *Actinomyces*. The most commonly seen type is *A. Israelii*. Normally the mucous membranes of the oral cavity, bronchus, gastrointestinal and genital tracts contain *Actinomyces* as commensal flora elements, and these do not pass through the mucosa. There are some predisposing factors, such as endoscopy, any upper respiratory or oral cavity operation, immunosuppression, and chronic inflammatory disease that may cause this bacteria to pass through mucosal barriers [1]. *Actinomycoses* infections generally occur in the cervicofacial region (50%), abdomen (20%), and thorax (15%) [2]. Abdominopelvic actinomycosis has increased in frequency in recent years and has been associated with bowel perforation, abdominal surgery, and trauma [3].

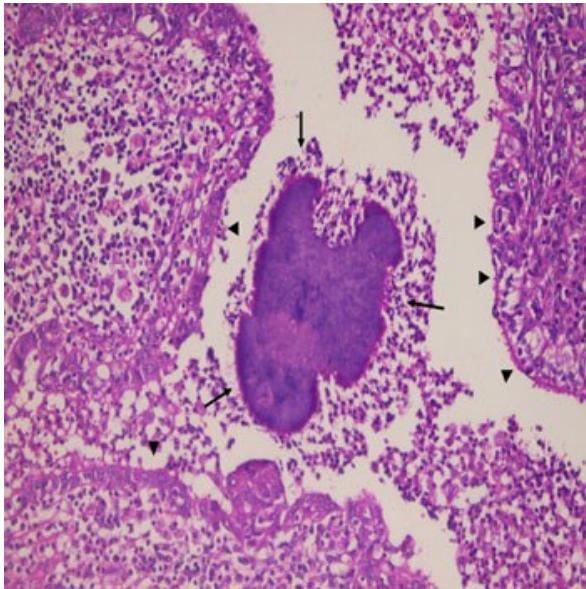
There is a relation between actinomycosis infection risk and IUD usage [4]. After 24 months of use, colonization rates increase [5]. Plastic intrauterine devices are more prone to infection, so removing or replacing a plastic device with a copper one decreases the risk of infection [6].

Infections related to IUD usage are mostly seen in developing countries and rural areas of developed countries. True incidence has not been well established; different forms of presentations in case reports can be found in the literature.

Case Report

A 49-year old woman was referred to the haematology department because of an elevated thrombocyte count. She had also had dull left lower abdominal quadrant pain for twenty days. She had been using a copper IUD for 12 years that had not been replaced. Her past medical history revealed one normal vaginal delivery.

Figure 1a The tubal epithelium (short arrows) in the tubal lumen, and the actinomycotic granules surrounded via inflammatory cells (long arrows), Hematoxyline and Eosin dye,x200.

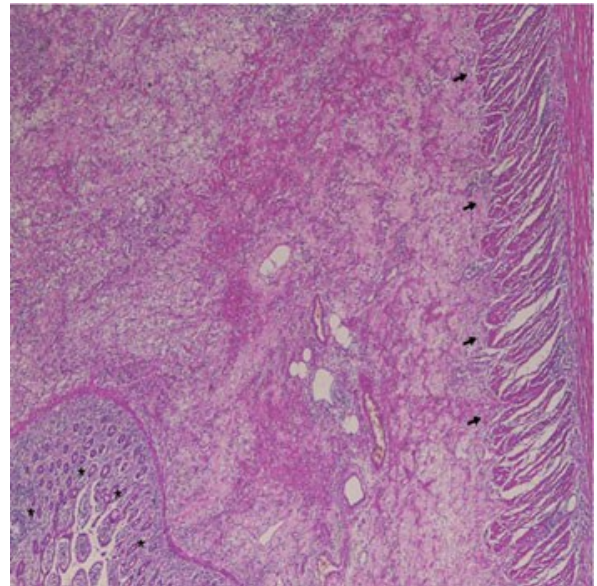


Her axillary temperature was 36.7°C. On pelvic examination, she had malodorous vaginal discharge. On her left lower abdominal quadrant she had a palpable tender and fixed adnexal mass. Laboratory examination revealed mild leucocytosis with a white blood cell count of 17,200/ μ L, an elevated thrombocyte count of 960000 K/ μ L, C-reactive peptide was found elevated to 230mg/L, and her Ca-125 level was moderately elevated to 58 IU/mL. Her cervicovaginal smear result was normal, no organism was isolated, and no malignancy or cervical intraepithelial lesion was seen. Her pelvic ultrasound examination revealed a 9 x 8 cm adnexal heterogenous mass.

A computerized tomographic (CT) scan of the abdomen and pelvis revealed a 12.3 x 11.4 cm pelvic mass containing cystic cavities, which displaced the bladder anteriorly. The mass was thought to arise from the right adnexa. The mass abutted both the uterus and sigmoid colon with a loss of normal fat planes. The preoperative diagnosis was pelvic abscess.

At the time of surgery, the patient had a frozen pelvis. At the right side of the uterus, there was an abscess formation containing loops of small intestine, the right fallopian tube and right ovary. A loop of small intestine was spontaneously perforated from one side and extended through this abscess formation. At the left side of the uterus there was also a tubo-ovarian abscess. The uterus, left fallopian tube, and left ovary were densely adherent to the sigmoid colon, and a

Figure 1b shows dense active chronic inflammation, starting from the serosal surface, deepening through muscular layer (arrow), than submucosal layer (space between stars and arrows), and reaching the bowel mucosa (stars). H&E,x100



second conglomerated mass was noted. The abscesses were drained and adhesiolysis was performed. After the uterus and fallopian tubes had been dissected in the pelvis, a total abdominal hysterectomy and salpingo-oophorectomy was performed. The perforated small intestinal loop was resected and an end-to-end anastomosis was performed. Postoperatively, there were no complications. The thrombocyte level decreased to 603,000 K/ μ L on the third day, and to 2500,00 K/ μ L on the tenth day of hospitalisation. Parenteral penicilin was used in the early postoperative period for 10 days; oral penicillin was then used for nearly six months. Pathologic examination reported actinomycotic infection in the abscess with sulphur granules and a full thickness abscess formation involving the small intestine. Figure 1 shows the histopathologic findings.

The patient was well and she was discharged from hospital on the tenth day, without fever and with normal bowel activity; there were no complications reported in the following six months.

Discussion

Preoperative diagnosis of actinomycosis is very difficult. The symptoms are non specific, and there are no specific tests available for the diagnosis. Most cases are identified incidentally by hystopathologic examination of specimens. Bae *et al.* revealed that CT studies could only diagnose 56.3% of cases [7]. Actinomycotic infections may represent malignancy

[8,9], tuberculosis, Crohn's disease, diverticulitis, or appendicitis, and pelvic magnetic resonance imaging (MRI) may be helpful in distinguishing between these etiologies [10]. Figure 2 shows the CT scan of the conglomerated mass composed of uterus, ovary, bowel, and abscess in the patient.

Thrombocytosis can be the initial presenting symptom in a variety of solid tumors including endometrial, ovarian, vulvar, and cervical cancers [11]. Koo *et al.* studied the predictors associated with the severity of pelvic actinomycosis, but they found no evidence of high thrombocyte counts in their study [12].

The patient in this case study presented with pelvic pain. Her complete blood count revealed an extremely high thrombocyte count mimicking a hematologic disorder. In the operation, a small bowel perforation was found. This is, to the best of our knowledge, the first case of pelvic actinomycosis presenting with thrombocytosis and small bowel perforation. Valade *et al.* studied thrombocytosis in trauma patients; they found that the most common cause of reactive thrombocytosis was infectious process, and they reported that reactive thrombocytosis appeared to be a good prognostic factor [13]. In the present case, an elevated thrombocyte count was also considered to be a reactive process to infection. The patient was treated successfully with surgery and antibiotic therapy. Mazarelo *et al.* reported a rare case of pseudohyperkalemia due to thrombocytosis in a tubo-ovarian abscess case; however, the patient in the current study had normal blood potassium levels [14].

Pelvic actinomycosis usually spreads extensively to adjacent organs. Lee *et al.* found that the sigmoid colon is the most involved site in a study of 18 abdominopelvic actinomycosis with gastrointestinal tract involvement [15]. Devendra and Chen presented a pelvic actinomycosis case with acute abdominal pain. They found an absence of planes between the bowel abscess and overlying omentum [16] due to the infiltrative tissue damage. Woody induration is the fibrotic inflammatory response to actinomycosis described by Goodman *et al.* [17]. This inflammation and absence of surgical planes are often misdiagnosed as a frozen pelvis [3].

In the present case, the uterus, left fallopian tube, left ovary, and sigmoid colon were densely adhered by the abscess, and a frozen pelvis was observed. The small intestine was ruptured and seemed to be a conglomerated mass with the ovary and fallopian tube on the right side.

Figure 2: Pelvic CT of the patient showing conglomerated mass in pelvic cavity



The exact treatment protocol of pelvic abscess due to IUD is not well established, but removal or replacement of the old and infected IUDs can be a reasonable approach. Dense desmoplastic reaction of the bacteria leads to difficult drug penetration; in cases of actinomycosis, long period of high-dose antibiotic therapy is required [18]. Surgical treatment should be considered for those with dense abscesses and obstructive symptoms. During surgery, infected and necrotic tissues must be removed, which will cause a decrease in bacterial load and recurrence rates. There is a risk of recurrence after many years, so periodic gynecologic examinations and imaging studies should be performed [19]. Penicillin is the first drug of choice. The parenteral route can be chosen for serious cases. When the clinical treatment is successful, oral treatment can be continued. The treatment should continue until the disease is cured [20].

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