PELVIC ACTINOMYCOSIS MIMICKING A LOCALLY ADVANCED PELVIC MALIGNANCY - CASE REPORT

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PELVIC ACTINOMYCOSIS MIMICKING A LOCALLY ADVANCED PELVIC MALIGNANCY - CASE REPORT (Abstract): We present the case of a former user of an intrauterine contraceptive device (IUD) for 10 years, diagnosed with a bulky, fixed pelvic tumor involving the internal genital organs and the recto sigmoid, causing luminal narrowing of the rectum, interpreted as locally advanced pelvic malignancy, probably of genital origin. Intraoperatively, a high index of suspicion made us collect a sample from the fibrous wall of the tumor mass, large Actinomyces colonies were thus identified. Surgery consisted in debridement, removal of a small amount of pus and appendectomy, thus avoiding a mutilating and useless surgery. Specific antibiotic therapy was administered for 3 months, with favorable postoperative and long-term outcomes. Pelvic actinomycosis should always be considered in the differential diagnosis of pelvic tumors in women using an IUD. The association of long-term antibiotic treatment is essential to eradicate the infection and prevent relapses. Keywords: ACTINOMYCOSIS, INTRAUTERINE DEVICE, PELVIC TUMOR.

Actinomycosis is a chronic infection most commonly caused by strains of Actinomyces israelii, a gram-positive, anaerobic, filamentous, non-sporulating bacterium (1,2). It causes a chronic suppurative inflammation with formation of multiple abscesses, sinuses and recurrent fistulas and abundant granulation and dense fibrous reaction tissue. Pelvic actinomycosis is found in about 3% of cases (3). Although the abdominal form of disease can extend directly to the pelvis, pelvic actinomycosis is mainly associated with intrauterine contraceptive devices (IUD) (4). The clinical presentation is mostly nonspecific, mimicking a tumor process (5,6). Moreover, the radiological findings are also nonspecific, making an accurate diagnosis difficult (7). This explains why patients often undergo extensive surgery involving multiple organ resections, with a correct diagnosis being made postoperatively by anaerobic cultures and histopathologic examination. We present the case of a female patient with pelvic actinomycosis diagnosed as locally advanced ovarian cancer in which the diagnosis was made intraoperatively, thus avoiding useless extended resectional surgery.
The diagnostic difficulties and treatment strategies are discussed.

**CASE REPORT**

A 51-year-old female patient with altered general status was admitted for investigations and treatment of a pelvic tumor mass. The onset of symptoms was insidious, over 4 months, with abdominopelvic pain associated with abnormal vaginal bleeding and a tendency to constipation, low grade fever, anorexia, marked physical asthenia and weight loss of 8 kg. From her personal history we found that the patient had been using an IUD for 10 years, being removed two months earlier. Physical examination on admission revealed a hard, palpable, imprecisely delimited mass in the hypogastrium. Laboratory tests showed leukocytosis (13,800/μL) with mild neutrophilia and anemia (hemoglobin level 10.9 g/dL). C-reactive protein levels were within normal range. Other laboratory tests including tumor markers were all within normal range, except CA 125 which was slightly elevated 19.80/mL. Vaginal examination revealed the presence of a pelvic mass occupying the vaginal vault, encompassing both parameters and an immovable cervix. Uterine curettage showed only an unspecific inflammatory process. Colonoscopy identified rectal extrinsic stenosis of approx 3 cm, at 10 cm from the anal verge.

Pelvic magnetic resonance imaging (MRI) showed a tumoral mass of 74x84/75 mm that included the posterior vaginal wall, extending to the right piriformis and levator ani muscles, involving the medial and lower mesorectum (fig. 1).

This tumor was centered by the ampulla recti, with probable invasion of the anterior rectal wall. Another mass of 52x40/52 mm, of unknown origin, located on the right and posterior surface of the right broad ligament was noted. Considering the suspicion of right ovarian tumor, a triple contrast CT was performed identifying gas bubbles in

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**Fig. 1.** Pelvic magnetic resonance imaging showed a tumoral mass of 74x84/75 mm that includes the posterior vaginal wall and extends to the right piriformis and levator ani muscles involving also medial and lower mesorectum. A second mass of 52x40/52 mm, located to the right and posterior of right broad ligament is noted.
the uterine cavity. The presence of right obturator and left external iliac and interaortico-cavous enlarged lymph nodes was also noted (fig. 2).

![Fig. 2. Pelvic CT scan showing an inhomogeneous mass involving internal genital organs without separation from rectum and enlarged pelvic lymph nodes.](image)

A left basal pulmonary nodule of 5 mm with uncertain features was identified. A preoperative diagnosis of locally advanced pelvic malignancy of unknown origin, was made and surgery was indicated. Exploration of the peritoneal cavity revealed a bulky, hard, irregular, fixed, infiltrating abdominopelvic mass involving the uterus, annexes, rectum, small bowel loops and vermiform appendix. During dissection a small amount of pus was exteriorized and collected for cytology and bacteriology exam. At that moment a sample of tumor/abscess wall was removed and sent for fresh-frozen examination which found multiple areas of polymorphous inflammation with abscess formation with large, central, filamentous *Actinomyces* colonies. Surgical debridement, appendectomy, peritoneal lavage and drainage were performed. Pathology established the final diagnosis of pelvic actinomycosis (fig. 3).

![Fig. 3. Abscesses with large, central, filamentous *Actinomyces* colonies (a. HE x25; b. HEx100).](image)
High doses of intravenous penicillin were commenced immediately. The postoperative course was uneventful with the patient being discharged on the 7th postoperative day. The antibiotic therapy was continued with high-dose penicillin for 4 weeks followed by oral penicillin (Augmentin 3g/day) for a further 6 months. A three months’ follow-up exam by MRI showed almost normal postoperative appearance of the pelvic region and no clinical complaints (fig. 4).

**Fig. 4.** Three months postoperative – MRI exam almost normal of pelvic region

**DISCUSSION**

Actinomycosis is a rare infectious disease, its true incidence being difficult to define because many cases with mild infection are probably eradicated through the common use of antibiotics. The abdominopelvic location is responsible for about 20% of human cases (1,2,8). It is generally accepted that actinomycosis has become a less frequent disease, unlike abdominopelvic actinomycosis which has an increasing prevalence due to the increasing use of IUDs. The first case of abdominopelvic actinomycosis associated with IUD was reported by Henderson in 1973. The incidence of *Actinomyces israelii* in IUD users is differently reported by various authors, ranging from 1.6% to 20% (1,4,6) and it increases with duration of IUD use, the mean duration of usage in patients with actinomycosis being about 8 years (7,9). These devices cause prolonged trauma to the endometrium, inducing areas of necrosis in the presence of pre-existing pelvic inflammatory disease, providing a favorable environment for the development of actinomycosis. Disruption of the mucosal barrier of cervical mucus allowing the ascension of germs or detachment of small fragments from the device which could act as foci of infection are other supposed mechanisms (6,7). The main problem related to actinomycosis is the difficulty of preoperative diagnosis, this disease being rightly called the great mimicker (4,7,8). The clinical and imaging features often mimic malignant lesions, inflammatory diseases, tuberculosis, fungal infections, endometriosis (5,9). The most common presenting features, as in our case, are abdominal pain, weight loss and physical weakness associated with abnormal vaginal
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bleeding. The tumor related symptoms are often in the foreground with a poorly delimited, fixed and painful pelvic mass accompanied by altered general status, cachexia, and sometimes fever. The biological inflammatory syndrome is constant and the CA 125 is sometimes elevated, as in our case (1,2,4,8). Extension to adjacent organs is possible, especially to the bladder and recto sigmoid with a tendency to fistula formation (9,10). Retroperitoneal involvement is common, causing ureteral and/or vascular compression and sometimes hydro nephrosis or thrombophlebitis (11,12). The lymph nodes are not usually involved, and spread into the peritoneal cavity is exceptional (4,7). The presence of a pelvic tumor mass with frozen pelvis appearance, with rectal stenosis and interaortocaval, left external iliac and right obturator enlarged lymph nodes, created confusion, pleading for the neoplastic nature of the lesion in our case.

Imaging diagnosis of actinomycosis is difficult. Frequently, the seemingly aggressive nature seen on CT or MRI might lead to the suspicion of a malignant disease. Lee describes the main radiological findings in abdominal actinomycosis. Each radiological finding may mimic an abdominopelvic tumor process, therefore a correct differential diagnosis is essential. (5,9,13). The CT scan typically shows a solid, inhomogeneous mass, or a cystic mass with thickened wall surrounded by inflammatory infiltrate in the adjacent fat (5,9). The MRI may help in diagnosis, but has a low specificity (5,13). Recently PET scan has been used to diagnose infectious and inflammatory diseases, but it is unfortunately not specific for actinomycosis. It could however be used to evaluate the efficacy of antibiotic therapy (13).

In terms of histopathology a characteristic chronic inflammatory response is found, with pus production and abscess formation centrally and granulation tissue in the periphery. In this atmosphere of destroyed and damaged PMNs, bacterial colonies of filamentous Actinomyces are identified. Confirmation may also be obtained by fine needle aspiration or biopsy under radiologic guidance or during surgical exploration (1,2). Unfortunately, many authors report highly variable rates (1-50%) of isolation of Actinomyces species from the histologic material (1,2,4,14,15,16). In our case, the intraoperative fresh frozen examination identified large filamentous colonies of Actinomyces. The literature also reports the presence of Gram-positive "sulfur granules", with a mycelium-like structure, positive on PAS and Grocott staining’s, which are unfortunately non-specific (1,4). The morphologic picture can be attenuated by prior antibiotic treatment thus histological tests should be carried out before the initiation of antibiotic treatment (2,4).

The presence of Actinomyces without apparent pelvic infection does not require IUD removal and antibiotic treatment, but many authors believe that a vaginal smear positive for Actinomyces is an indication for antibiotic treatment to eradicate the infection or recommend IUD replacement at 4 years (6).

Standard treatment is based on intravenous penicillin G for 2-6 weeks, followed by oral penicillin V (2-4 g/day) for 6-12 months due to the limited penetration into the fibrous tissue and tendency to recurrence (1,8). The risk of developing penicillin resistance is cited as very low. In preg-
nant women, erythromycin can be safe, and in patients who are allergic to penicillin, doxycycline, clindamycin and erythromycin are adequate (14,15,17). In case of concomitant intraabdominal infection with anaerobic bacteria, the combination of amoxicillin and clavulanic acid with metronidazole, or clindamycin can be used. However, the literature shows a high rate of recurrence after antibiotic therapy alone (2,15). Definitive indications for surgical treatment are: abscesses that are not amenable to percutaneous drainage, extensive necrotic tissue, local complications such as compression or stenosis, to rule out a malignant disease, or when medical treatment is inefficient. Complete removal of infected tissue is rarely possible, with a high risk of recurrence. It is very important to make or suspect the diagnosis intraoperatively, because most often a pelvic malignancy is suspected and patients undergo major surgery. (8,14,17).

The prognosis is favorable with long-term antibiotic treatment. Some authors have shown that a combination of complete surgical resection followed by short-term antibiotic treatment is also an effective therapy (1,17). However, severe complications due to local or generalized spread of infection have been reported, justifying the prolonged surveillance (14,15).

**CONCLUSIONS**

In this case pelvic actinomycosis pelvic, rare infection associated with the use of an IUD over a long period of time, mimicking a locally advanced pelvic malignancy. Therefore, actinomycosis should be included in the differential diagnosis in women with presumed pelvic cancer and a history of long-term use of IUDs. The primary goal of surgery is to obtain a clear evidence for histopathological diagnosis and surgical treatment should be limited to debridement and drainage. Essential to eradicate the infection and prevent recurrences is the association of the specific antibiotic treatment.

**REFERENCES**

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