

## Case Report

# Actinomycotic hepatic abscess in woman with longstanding intrauterine contraceptive device

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## Abstract

We present a case of a 50 year-old female bearing an intrauterine contraceptive device for 20 years who was diagnosed with abdominopelvic actinomycosis with liver dissemination. The patient was successfully treated by a combination of surgical resection and a 3-month course of amoxicillin.

**Key words:** actinomycosis; intrauterine contraceptive device; hepatic abscess.

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## Introduction

Actinomycosis is a rare, chronic, granulomatous disease caused by anaerobic Gram-positive bacteria that normally colonize human mucosa. Due to its heterogeneous clinical manifestations, rarity, and non-specific imaging findings, the diagnosis of actinomycosis is usually delayed and based on the histological report of the surgically obtained tissue [1,2]. We present a case of abdominopelvic actinomycosis affecting the uterus, both adnexa, the rectum and appendix, with infiltration of the liver in a patient with an intrauterine contraceptive device (IUCD) that had not been replaced for 20 years.

## Case Report

The fifty-year-old female patient was referred to the Clinic for Digestive Surgery by her general practitioner. She was previously healthy, with history of bearing the same IUCD for 20 years. She had not visited a gynecologist for eight years.

She complained of abdominal pain, low-grade fever, malaise and 6-kilo weight loss in four weeks. Physical examination revealed a body temperature of 37.6°C and a large, fluctuating mass (10×cm) in the upper right quadrant. There was also a small fistula (6mm) with a minimal amount of odorous discharge. An additional mass located in the projection of the left

adnexa was also noted. The rest of her examination was unremarkable.

Laboratory analysis revealed a high erythrocyte sedimentation rate (115), leukocytosis (20.000) with neutrophilia (84%) and slightly elevated C-reactive protein (CRP) (21). Liver function tests and tumor markers (AFP, CEA, CA 19.9, HE-4) were normal. Serological tests for HBV and HCV infection were negative. Further clinical evaluation included chest X-ray and upper and lower endoscopy; all findings were unremarkable. There were no signs of free air or fluid on abdominal X-ray, while the presence of the IUCD was notable (Figure 1). The gynecologist confirmed the existence of a left-sided tubo-ovarian abscess accompanied by endometritis. His attempt to extract the ICDU failed. Both abdominal ultrasound (US) and computed tomography (CT) revealed a multilocular abscess (12 cm in diameter), located in the right lobe of the liver, between segments 4 and 8, with infiltration of the anterior abdominal wall (Figure 2).

She underwent operation during which the liver abscess located between segments 2, 4a and 8, measuring 15 cm in diameter, was observed and resected. There was also an inflammatory pseudotumor that infiltrated the enlarged uterus, both adnexa, the rectum and appendix, necessitating hysterectomy with bilateral adnexectomy, extraction of the IUCD,

appendectomy and anterior resection of the rectum with colorectal anastomosis. *Ex tempore* pathohistological analysis of tissue samples from the uterus and adnexa showed signs of chronic inflammation, but without any malignant cells.

There were no complications during the postoperative course. The patient was discharged from hospital eight days after surgery. The final pathohistological report recorded pelvic inflammatory disease, bilateral tubo-ovarian abscesses, chronic endometritis and cervicitis, periappendicitis, perirectal abscesses and fibrous adhesions, with presence of the sulfur granules characteristic of actinomycotic colonies. Histological examination of liver tissue demonstrated abscesses due to *Actinomyces* with typical sulfur granules, many histiocytes and rare multinuclear giant cells (Figure 3). Histological elements of malignant disease were not found. The patient received amoxicillin during three months and recovered completely.

**Discussion**

Actinomycosis is a chronic, suppurative and granulomatous infection, characterized by the presence of extended necrosis, abscess and sinus formation, and reactive fibrosis. It is caused by anaerobic or microaerophilic Gram-positive bacteria of *Actinomyces* species. Depending on the primary site of infection, actinomycosis can be cervicofacial (more than 50%), thoracic (15%) and abdominopelvic (20%), although there can be overlap [3-5]. The liver is rare as a primary site of infection, accounting for only 15% of abdominal infections and 5% of all actinomycotic infections. [5,6]. The liver becomes affected by hematogenous

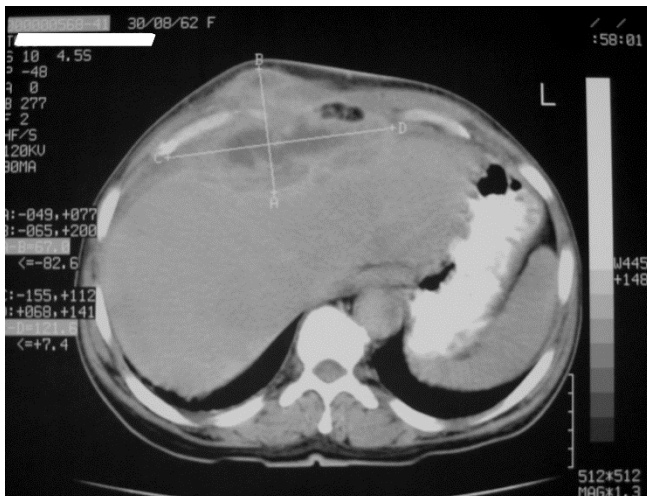
**Figure 1.** Abdominal X-ray with clearly visible IUCD.



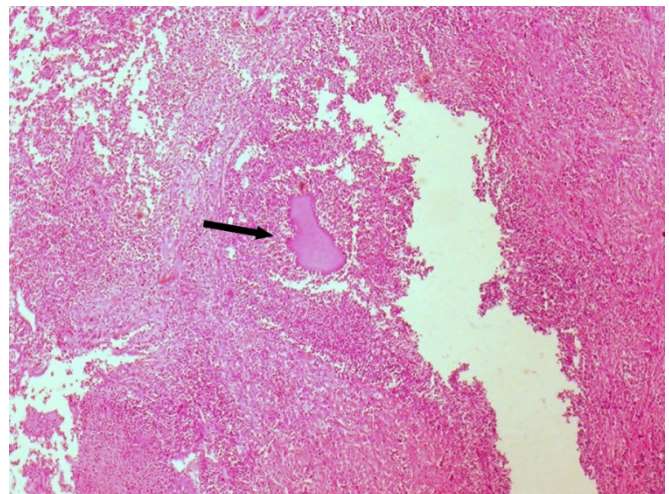
dissemination from a distant lesion or direct spread of infection from surrounding tissue. The formation of a hepatic abscess takes from four days to 18 months [7]. To our knowledge, there are few reports of liver involvement in patients with abdominopelvic actinomycosis and a long-standing IUCD presence [8,9].

Previous abdominal surgery or trauma (perforation and injuries of the intestine and anterior abdominal wall), the presence of foreign bodies in the

**Figure 2.** Abdominal CT revealed multilocular abscess (12 cm in diameter) in the right lobe of the liver with infiltration of the anterior abdominal wall.



**Figure 3.** Histological preparation of liver tissue showing the abscess due to *Actinomyces*.



abdominopelvic region (biliary stent, IUCD) and other comorbidities (hematologic diseases, renal failure, diabetes) usually predispose to the development of abdominopelvic actinomycosis [7,10]. The long-term presence of an IUCD is the most important risk factor for development of actinomycosis in women [3,11-14]. Since *Actinomyces* colonize human urogenital and digestive tracts, IUCDs are often colonized with these bacteria; antibiotics are not indicated in such cases [15]. *Actinomyces spp.* infects 1.65% to 11.6% women with IUCDs [3].

Symptoms and signs are often non-specific: low-grade fever, weight loss, abdominal or pelvic pain, fatigue lasting 2-5 weeks [4]. Partial or complete bowel obstruction and hydronephrosis with difficulty in urinating have also been described [10]. Physical examination usually reveals a palpable tumor mass, sometimes with spontaneous cutaneous fistula [16].

Actinomycosis is rarely diagnosed at the initial presentation. The majority of cases are diagnosed after surgery, based on clinical, radiological and histological findings, since this infection often imitates a malignant tumor [17,18]. In addition, pyogenic abscesses, echinococcal cysts, cystic liver tumors, and amoebic abscesses are included in the differential diagnosis [7].

Biochemical results are usually normal, with the exception of anemia, leukocytosis, and often elevated CRP and erythrocyte sedimentation rate [4,7,19]. Mildly increased alkaline phosphatase, lower albumin levels and elevated tumor markers (AFP, CA 19.9) are occasionally found in patients with hepatic actinomycosis [7,20].

Imaging findings are non-specific. Abdominal US, CT and magnetic resonance imaging (MRI) imaging show single (66%) or multiple (33%) liver masses/abscesses [20]. US shows hypoechoic, heterogeneous and sometimes poorly defined lesions [20]. CT and MRI reveal nodular hepatic lesions, sometimes multilocular, lobulated, with undefined boundaries. CT can also reveal hypodense, limited liver lesions with peripheral rim enhancement corresponding with abscesses. Sometimes there is a partly cystic or solid inflammatory pseudotumor that infiltrates abdominopelvic structures, usually without lymphadenopathy and ascites. Since the sigmoid colon is involved in 50% of cases, thickening of the bowel wall (up to 8.3 cm in length) is one of the most common CT findings [3,4,7,21]. In some cases, a non-homogeneous infiltration of the abdominal wall and retroperitoneum has been described, which confirms the infiltrative nature of the disease [11,17,22]. MRI of hepatic actinomycosis is characterized by low-signal

intensity on T1 and high-signal intensity on T2 weighted images, while a tubo-ovarian abscess demonstrates lower signal intensity on T2 weighted sequences [7,23]. PET-scan can verify the hepatic focus of increased metabolic activity resembling a metastatic tumor (SUV 11,6) [21].

Colonoscopy and gynecological examination are recommended as a part of patient evaluation to exclude diverticulitis, appendicitis, other granulomatous diseases (Crohn's disease, tuberculosis), and malignancy [3]. The IUCD should be removed (if possible) and antibiotics should be introduced [24]. Surgical resection is often required.

Since actinomycosis lacks typical clinical features, the diagnosis is based on microbiological and/or histological examination of the affected tissue. Liver specimens can be obtained by ultrasound-guided percutaneous puncture, laparoscopic exploration or laparotomy [7]. Sometimes material obtained by percutaneous liver biopsy shows only a nonspecific organizing abscess with plasma cell infiltration, without confirmation of diagnosis [25].

Typical histological findings reveal the presence of necrosis with yellow sulfur granules of 1-2 mm in diameter, representing microcolonies of bacteria surrounded by lymphocytes, plasmocytes, histiocytes, and epithelium cells [15]. Bacterial confirmation is challenging and usually is accomplished in only 50% of cases, since culturing requires immediate specimen transport, prolonged anaerobic incubation and special equipment [25,26]. Identification of *Actinomyces* is possible using 16S rRNA gene sequencing [27].

Many patients require surgical treatment due to their clinical presentation or to inadequate preoperative diagnosis [15]. Intraoperative findings correspond to the neoplastic/inflammatory tissue in the pelvis that involves bowel (ascending colon, cecum, distal ileum, appendix), uterus and adnexa. The tumor mass can occupy the retroperitoneal space, sometimes adhering to the abdominal wall [3].

Actinomycosis can be treated successfully with a prolonged course (6-12 months) of antibiotics (such as penicillin G, amoxicillin, ampicillin, or clindamycin). Although long-term antibiotic therapy has been traditionally recommended, treatment can be reduced to a three-month course in those who have had surgical treatment [1,15].

## Conclusion

Actinomycosis of the liver should be considered in women with palpable abdominal masses and long-term use of IUCD, especially in the presence of a cutaneous

fistula. Diagnosis is based on histological findings and/or bacterial confirmation [2]. Surgical resection and antibiotics can lead to complete recovery in treated patients [1,15].

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