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Clinical Features of Abdominopelvic Actinomycosis: Report of Twenty Cases and Literature Review

Myung-Min Choi,[1] Jeong Heum Beak,[1] Jung Nam Lee,[1] Sanghui Park,[2] and Won-Suk Lee[1]

Departments of [1]Surgery, [2]Pathology, Gil Medical Center, Gachon University of Medicine and Science, Seoul, Korea.

Purpose: Intrabdominal actinomycosis is difficult to diagnose preoperatively. This chronic infection has a propensity to mimic many other diseases and may present with a wide variety of symptoms. The aim of this study was to evaluate the characteristic clinical features with review of the literature. **Materials and Methods**: We retrospectively analyzed 22 patients with intrabdominal actinomycosis between January 2000 and January 2006. **Results**: There were two men and 20 women with a mean age of 42.8 years (range, 24 - 69). Twelve patients presented with masses or abdominal pain, whereas 3 patients presented with acute appendicitis. The rate of performing an emergency surgery was 50% due to symptoms of peritonitis. The mean size of tumor was 5.5 cm (range, 2.5 - 11.0). Sixty percent (n = 12) of female patients had intrauterine device (IUD). The average time to definite diagnosis was 10.6 days. **Conclusion**: Intrabdominal abdominal actinomycosis must first be suspected in any women with a history of current or recent IUD use who presents abdominal pain. If recognized preoperatively, a limited surgical procedure, may spare the patient from an extensive operation.

Key Words : Actinomycosis, surgery, intrauterine device

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Corresponding author: Dr. Won-Suk Lee, Department of Surgery, Gil Medical Center, Gachon University of Medicine and Science, 1198 Guwol-dong, Namdong-gu, Incheon 405-760, Korea.
Tel: 82-32-460-1234, Fax: 82-32-460-3009
E-mail: lws@gilhospital.com

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INTRODUCTION

Actinomycosis is a chronic, suppurative, and granulomatous disease caused by an anaerobic Gram-positive bacterium, *Actinomyces israelii*, manifesting itself as fistula, sinus, inflammatory pseudotumor, or abscess formation.[1] Humans are natural reservoirs and there is no documented person-to-person transmission of the disease, and it is commonly cultured from carious teeth, tonsilar crypts.[2] It is characterized by a tendency to feign malignancy due to its capacity to invade surrounding tissues and to form masses.[3] Therefore, there are multiple clinical presentations, often leading to misdiagnosis. The three main clinical forms of this disease are cervicofacial, thoracic, and abdominopelvic. The cervicofacial region accounts for 50% to 65%, followed by abdomen (20%).[4-6]

The disease usually shows an indolent course with clinical symtoms and signs that are not specific, resulting in delayed diagnosis. Actinomyces are sensitive to penicillin, but the duration of treatment varies from several weeks to months to achieve permanent recovery.[7-10] The aim of this study was to evaluate the characteristic clinical features with short literature review on the topic.

MATERIALS AND METHODS

Between January 2000 and January 2006, 22 patients with abdominopelvic actinomycosis were identified. Patient's demographic data and outcome are

Table 1. Summary of 22 Patients with Intrabominal Actinomycosis

Patient no.	Sex	Signs and symtom	Time to diagnosis	Presence of IUD	Initial leukocyte count (mm^3)	Antibiotics	Risk factors	Recurrences
1	F	Painful mass	15	No	8,690	IV penicillin	None	None
2	F	Abdominal pain	7	No	9,340	IV penicillin	None	None
3	F	Abdominal pain	6	No	11,890	IV penicillin	None	None
4	F	Abdominal pain	18	Yes	16,500	IV penicillin	None	None
5	F	Fever	4	No	18,330	IV penicillin	None	None
6	F	Abdominal pain	15	No	11,750	IV penicillin	None	None
7	F	Abdominal pain	8	Yes	22,900	IV penicillin	None	None
8	F	No symptom	3	Yes	13,570	IV penicillin	None	None
9	F	Abdominal pain	7	No	5,950	IV penicillin	None	None
10	F	Painful mass	14	No	9,440	IV penicillin	None	None
11	F	Abdominal pain	15	No	8,990	IV penicillin	Liver cirrhosis	None
12	F	Abdominal pain	17	No	15,300	IV doxycline	None	None
13	F	Abdominal pain	7	Yes	20,940	IV ciprofloxacin	None	None
14	F	Fever	12	Yes	7,400	IV penicillin	None	None
15	F	Abdominal pain	9	No	14,560	IV penicillin	None	None
16	F	Abdominal pain	19	No	16,210	IV penicillin	None	None
17	F	Abdominal pain	17	Yes	7,650	IV penicillin	None	None
18	M	Abdominal pain	13	N / A	4,180	IV penicillin	DM	None
19	M	Abdominal pain	10	N / A	20,490	IV penicillin	None	None
20	F	Abdominal pain	8	No	5,310	IV penicillin	None	None
21	F	Painless mass	4	Yes	16,970	IV penicillin	DM / Hypertension	None
22	F	Abdominal pain	6	Yes	14,480	IV penicillin	None	None

IUD, intrauterine device.

summarized in Table 1. The clinical data including age, gender, mass size, preoperative diagnosis, presence and duration of intrauterine device (IUD) were retrospectively analyzed. Intrabdominal mass assessments consisted of physical examination, colonoscopy, ultrasonography, and abdominopelvic CT scan.

RESULTS

The clinical details of these patients are presented in Table 2. There were two men and twenty women with a mean age of 42.8 (range, 24 - 69) years. Twelve patients presented with masses or abdominal pain, whereas three patients presented with acute appendicitis (Table 3). Among the twenty two patients, only two patients presented with a colonic mass mimicking colon cancer. Fifteen patients (68.2%) had leukocytosis with a mean WBC count of 12,765 mm^3 (range; 4,180 - 22,900 mm^3). None of the patients presented with small bowel or colon obstruction. However, emergency surgery rate was 50% due to peritonitis symptoms. A preoperative abdominal CT scan

Table 2. Patients' Characteristics

	Intrabdominal actinomycosis (n = 22)
Age (yrs)	
Mean (range)	42.8 (24 - 69)
Gender: M / F	2 : 20
IUD (n = 20)	
Yes	12 (60%)
No	8 (40%)
Emergency vs. elective operation	
Yes	11 (50%)
No	11 (50%)
WBC, mm^3	
Mean (range)	12,765 (4,180 - 22,900)
GI obstruction	
Yes	0 (0.0%)
No	100 (0.0%)
Mass size, cm	
Mean (range)	5.5 (2.5 - 11.0)

IUD, intrauterine device; WBC, white blood cell; GI, gastrointestinal.

Table 3. Pre-Existing Diagnosis before Intrabdominal Actinomycosis

Pre-existing diagnosis	No. of patients (n = 22), %
Diverticulitis	2 (9.0)
PID	2 (9.0)
Pelvic mass	5 (22.8)
Tubovarian abscess*	5 (22.8)
Appendicitis	3 (13.6)
Lymphoma	3 (13.6)
Endometriosis	1 (4.5)
Pelvic abscess	1 (4.5)

*One patient with combined sigmoid colon fistula. PID, pelvic inflammatory disease.

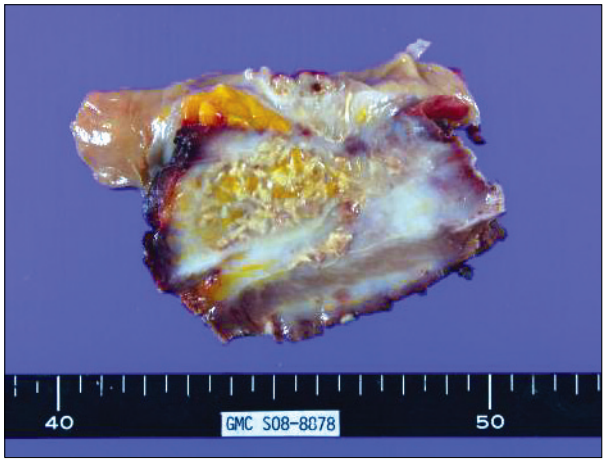

Fig. 1. 10.3 × 9.3 cm ovoid mass on the serosal surface of the cecum and ascending colon with ulceration. The cut surface demonstrates typical light gray color with necrosis.

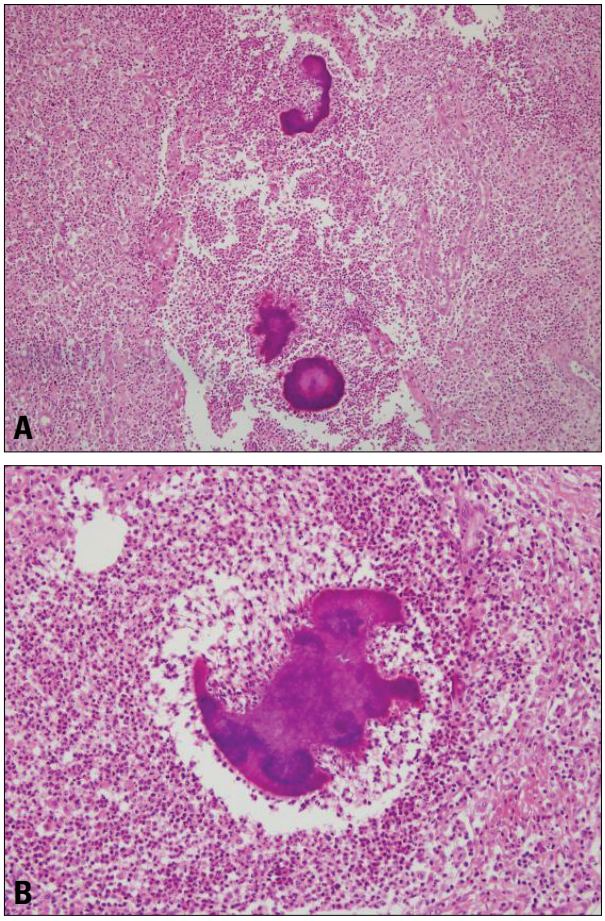

Fig. 2. (A) A actinomycotic abscesses containing sulfur granules with radiating filaments (H & E, × 100). (B) A magnified view of the characteristic sulfur granule (H & E, × 200).

or ultrasonography was done in all patients and detected intrabdominal mass or abscess but failed to give a definite diagnosis. The median operative time was 140 (range, 90 - 420) minutes and the median blood loss was 250 (range, 150 - 800) mL. The mean size of tumor was 5.5 (range, 2.5 - 11.0) cm. Sixty percent (n = 12) of female patients had IUD. The patients had been wearing IUD for an average of 7 years, and 15% had been wearing an IUD for 3 years or less. Confirmation of the diagnosis of actinomycosis was done by histology in all cases. Microscopically, each of the specimens showed chronic inflammatory reactions with sulfur granules (Figs. 1 and 2). None of the patients underwent percutaneous biopsy. There were no cancer cells found in all patients. The average time to definitive diagnosis was 10.6 days (range, 4 - 19 days).

After a median follow up of 37.5 months (range, 6.6 - 23.1 months), recurrence was not seen in any patients. The antibiotic of choice was IV penicillin, however, one patient was given ciprofloxacin due to penicillin allergy. The duration of treatment was 3 months in twelve patients, 6 months in five patients, 4 months in two patients and 1 month in two patients who refused to continue.

DISCUSSION

Actinomycosis was first diagnosed in a live patient by Ponfick in 1879.[11] It is a chronic suppurative disease caused by an anaerobic, filamentous Gram positive bacteria.[12] Actinomycosis Israelii is a constant part of the microflora in the human oral cavity, gastrointestinal and genital tracks.[12,13] The organism is unable to cross normal mucosal barrier, therefore, opportunistic infections can occur only in context of underlying local disease such as trauma, surgery, or a foreign body which is significant enough to penetrate this barrier. Once the organisms have penetrated the mucosa, spread by continuity seems to be the primary method of intrabdominal propagation. Lymphatic and hematogenous spread is uncommon,[14,15] although there is a hematogenous and lymphatic spread with nodal involvement reported.[16]

Clinically, the disease follows an indolent course and the initial presentation usually includes lower abdominal pain and fever with or without a palpable mass as was seen in our series. Since symptoms and signs are not specific, the diagnosis is often delayed and only 10% of cases are diag-

nosed preoperatively.[16] In our series, only one patient (4.5%) was diagnosed before surgery.

Intrabdominal actinomycosis can appear as an abdominal mass of ambiguous benignity and can mimic a malignant tumor. For example, actinomycosis of the colon or the greater omentum is a rare differential diagnosis of colonic carcinoma or peritoneal tumor.[17-19] The pathogenesis of abdominal actinomycosis is not yet well understood. There are two possibilities suggested that can affect intrabdominal organs: through blood-borne infection or by swallowing.[20] Actinomyces can normally inhabit colon, predominating in areas of stagnation i.e. the cecum and appendix. Actinomyces requires injury to the normal mucosa to penetrate and cause disease. Predisposing factors may include appendicitis and diverticulitis, gastrointestinal perforations, previous surgery, foreign bodies, or neoplasia.[21] Thus, intrabdominal actinomycosis should be included in differential along with other inflammatory diseases such as ulcerative colitis, Crohn's disease, tuberculosis, diverticulitis and pelvic inflammatory disease.

Pelvic actinomycosis has recently become more prevalent and is associated almost exclusively with women who use IUDs.[16] Orogentital tract is thought to be an important mode of acquiring this type of infection in lower genital tract.[22] In the uterine cavity, the microorganisms are apparently confined to the superficial layers of the mucosa, a fact perhaps related to its cyclic shedding. The most likely route of spread with subsequent development of pelvic abscess appears to be through patent fallopian tubes. The use of IUD may increase the risk of infection through injury to the normal uterine mucosa.[13,23] Pelvic actinomycosis associated with the use of IUDs can mimick pelvic malignancy.[24] For such reason, it is often surgically excised. However, if a diagnosis can properly be made preoperatively, antibiotic treatment and removal of IUD may lead to complete remission, avoiding unnecessary surgery. In our series, none of the patients with IUDs was diagnosed of actinomycosis preoperatively. Fiorino[25] reported 92 patients with actinomycotic abscess in 63 case reports. In this study, the average duration of IUD implantation was 8 years, and only 16% had been using an IUD for less than 3 years. Likewise, the average duration of IUD was 7 years and only 15% of those patients had IUD for less than 3 years in our study. Although IUD is strongly correlated with intrabdominal actinomycosis, a definite duration of IUD implantation and the risk of developing actinomycosis infection has not yet been established.

The modern principle of therapy for actinomycosis began with Peabody and Seabury in 1960,[26] who recommended abscess drainage in combination with high dose antibiotics. Smith et al.[27] reported that ciprofloxacin and tetracyclines showed poor performance in antimicrobial susceptibility testing of actinomyces species. Actinomyces species appear to be susceptible to a wide range of beta-lactam agents and, when combined with beta-lactamase inhibitors, they should be regarded as agents of first choice.[27]

Uncomplicated actinomycosis can be medically treated by antibiotics, although there are differing opinions in the literature about dosage and duration of antibiotic treatment.[28,29] A prolonged treatment course is required because of the poor penetration of antibiotics into the fibrotic tissues. Thus, when there are more avascular spaces present due to severe tissue reactions, medical therapy may be less effective, resulting in longer duration of antibiotic treatment, regardless of the site of actinomycosis. Interestingly, two patients in our series (9.0%) who were treated with iv antibiotics less than 4 weeks did not recur during the last 34 months of follow-up. Hence, the clinical impact of surgical resection followed by short-term antibiotics merits for further study.

Even though intrabdominal actinomycosis is very rare in its frequency, it should be included in a list of differential diagnosis, especially in any women with a history of IUD use who presents with abdominal pain or a pelvic mass. If actinomycosis is suspected preoperatively, appropriate handling of cultures will increase the diagnostic yield which may spare the patient from an extensive surgery.

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