

Original Article

Abdominopelvic Actinomycosis – Treatment Outcomes at the Chang Gung Memorial Hospital during the Past 16 years

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Key Words

Actinomycosis;
Abdominopelvis;
Radical resection;
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Purpose. Actinomycosis is uncommon and it causes various clinical pictures mimicking cancer, inflammatory disease, or diverticulitis, along with rarity of the disease, the diagnosis of actinomycosis is difficult. We draw out the optimal treatment modalities for the abdominopelvic actinomycosis in addition to proposed medical management (high-dose penicillin for a long and variable interval, 6 months to 1 year) or combined surgery with antibiotics treatment.

Methods. We review the previous cases of actinomycosis in the abdomen and pelvis. They were recruited from 1984 through 2001 at Division of Colon and Rectal Surgery, Department of Surgery in the Chang Gung Memorial Hospital (CGMH). Reviewing the charts, we recorded the clinical manifestations, image findings, white blood cell counts (WBC) and differential counts, surgical procedures, post-operative antibiotics treatment duration, interval of follow-up, and history of IUD use.

Results. In our series, the median follow-up period was 16 months (6-153 months). Eight patients (cases 1, 2, 4, 5 and 7-10) underwent radical surgery because the disease was mimicking malignancy morphologically and two patients (case 3 and 6) received prolonged antibiotic treatments after biopsy and bypass. Prolonged antibiotic treatments after radical resection of the lesions were not routine. No relapse even no prolonged antibiotics treatment during follow-up was identified.

Conclusion. In our cases, the standard long-term antibiotic treatment could be used in patients having no radical resection. But close follow-up by CT and clinical examination are important in such cases because disease relapse is possible. Surgical intervention alone can cure cases with infection by complete removal of the infection foci, and long-term antibiotic therapy is unnecessary.

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Actinomycosis is a chronic suppurative infectious disease caused by anaerobic gram-positive bacteria that can cause multiple abscess formation, draining fistulas, and the formation of dense granulation tissue.¹⁻³ Bradshaw,⁴ in 1846, proposed the first case of

abdominal actinomycosis. The actinomyces species are commensals and normally inhabit the oropharynx, gastrointestinal tract and female genital tract. Invasion of the bacteria has been noted at cervicofacia (50%), abdomen and pelvis (20%), and thoracic area (15%).

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Anterior abdominal wall or viscera invasion is rare.⁵⁻¹⁰ However, the clinical spectrum of abdominal presentation has changed. Abdominal actinomycosis was reported to occur after the destruction of the mucosa barrier or immunosuppression, as well as spontaneously or after surgical opening or perforation of bowel prior to the 1970s. The abdominal infection has been reported to mostly involve the cecum or appendix and it can simulate malignancy on clinical radiological examinations.^{3,11} However, the gynecological sources have been the prevalent origin of infection since the late 1970s, which was associated with the use of intrauterine devices (IUDs). Pelvic actinomycosis may simulate pelvic malignancy and it is often treated surgically because of difficult pre-operative diagnosis. If early diagnosis can be made before surgery or intraoperatively, removal of the intrauterine devices with antibiotic (Penicillin G) therapy for 12 months may be an effective treatment. In our cases, radical resections were done in eight of ten cases, but some did not have conventional antibiotic therapy as the routine treatment interval. We propose that the patients of abdominal actinomycosis undergoing radical resection, prolonged antibiotics therapy could be unnecessary.

Materials and Methods

From 1984 through 2001, we collected ten cases of abdomino-pelvic actinomycosis at our institute. They received radical resection, bypass surgery or drainage procedure with or without antibiotics treatment. A total of 10 women had surgical management for abdominopelvic actinomycosis. Nine of ten patients received elective surgery. One patient, case 7, received emergent surgery. The operative indications and pre-operative diagnosis were as followings: 1. pelvic mass lesion with suspected malignancy (case 1, 4, 8, 10); 2. colonic stricture with suspected malignancy (case 2, 5, 6, 9); 3. intra-abdominal abscess with fistula (case 3); and 4. para-renal abscess (case 7). The pre-operative evaluation modalities were computed tomography of abdomen (CT), colonoscopy, low gastrointestinal series (LGI), intravenous pyelography (IVP) or abdominal ultrasonography. The colonoscopy was arranged in case 1, 2, 6, 10 and

external compression with lumen stenosis was noted in these patients. LGI was performed in case 1, 2, 5, 6, 9, 10. One patient, case 8, received IVP for left hydronephrosis from pelvic mass. Another patient, case 7, received abdominal sonography for left flank pain. All patients received abdominal CT examination before surgical intervention. Preoperative colonoscopy biopsy of the lesions in cases 1, 2, 6, 10 showed negative for malignancy. Microscopically, colonies of actinomyces species (sulfur granules) were detected in all patients. By reviewing the charts, we recorded the clinical manifestations, image finding, white blood cell count (WBC) and differential counts, surgical procedures, post-operative antibiotics treatment duration, interval of follow-up, and history of IUD use. (Table 1)

Results

All cases underwent laparotomy to confirm diagnoses. They were treated successfully either surgical interventions alone or concurrent penicillin-based medical treatment for the unresectability. The mean age of the 10 patients was 49.6 years old (31 y/o-67 y/o). The median followed period was 16 months (6-153 months). Most of patients presented with palpable abdominal masses and vague abdominal pain as secondary symptom. Six patients (Cases 1, 2, 4, 5, 9, and 10) had abdominal pain or painful masses, two had flank pain (Cases 3 and 7) and five had abdominal masses (Cases 1, 2, 6, 8, and 10). Five patients had colon stricture from external compression (Cases 1, 2, 5, 6, and 9). Four patients had IUD implantation, and another four had no detailed chart records. Eight patients (Cases 1, 2, 4, 5, and 7-10) underwent radical surgery (including total abdominal hysterectomy, salpingo-oophorectomy, anterior resection of colon and rectum, incision and drainage or nephrectomy) because the disease mimicking the malignancy morphologically. Case 3 had incisional biopsy with drainage. Case 6 had biopsy and bypass surgery. Patients of Case 3 and Case 6 received Penicillin G for 12 months and 18 months, respectively. The follow-up interval in case 3 and case 6 were 14 months and 153 months, respectively. No disease relapse was detected by CT ex-

Table 1. This table summarizes 10 patient's sex, age, clinical presentation, laboratory data, surgical procedure, post surgery antibiotics treatment interval and the use of IUD

No	Gender & Age	Symptom & Sign	Evaluation tools	Image finding	WBC	Surgery	Anti	IUD	Follow-up interval
1	48/F	Abdominal painful mass at LLQ; Body weight loss	Colonoscopy/ LGI/CT	S-colon extraluminal compression L't hydronephrosis	15100	ATH/BSO	nil	?	14 Ms
2	53/F	Abdominal painful mass at LLQ;	Colonoscopy/ LGI/CT	S-colon stricture L't hydronephrosis	12900	LAR. ATH/BSO	Tetra 2Ms	?	12 Ms
3	47/F	R't flank pain with abdominal abscess and sinus fistula	CT	R't hydronephrosis	13000	I/D and biopsy	PCN 3Ms	+	14 Ms
4	44/F	Low abdominal pain with difficult defecation	CT	L't Cul-de sac mass	16300	ATH/LSO	nil	+	18 Ms
5	55/F	Low abdominal pain with vaginal discharge	LGI/CT	Rectosigmoid colon stricture R't hydronephrosis Pelvic mass	11000	Tumor excision RSO/S-loop colostomy	nil	?	81 Ms
6	67/F	RLQ abdominal mass; Fever; body weight loss	Colonoscopy/ LGI/CT	S-colon stricture R't hydronephrosis	14200	Bypass/Biopsy	PCN 18 Ms	—	153 Ms
7	59/F	L't flank pain	Abdominal ECHO/CT	Pararenal abscess	15000	L't nephrectomy Colectomy	nil	—	55 Ms
8	43/F	Abdominal LLQ mass; body weight loss	IVP/CT	L't pelvic mass L't hydronephrosis	10800	ATH/BSO	PCN 1M	+	6 Ms
9	49/F	Irregular bowel movement and LLQ abdominal pain	LGI/CT	S-colon stricture L't pelvic mass	13500	ATH/BSO AR	PCN 12Ms	?	14 Ms
10	31/F	Abdominal painful mass at LLQ;	Colonoscopy/ LGI/CT	L't pelvic mass	15300	AR	PCN 3Ms	+	18 Ms

ATH: total abdominal hysterectomy; R/L/BSO: right/left/ Bil. salpingo-oophorectomy; LAR: low anterior resection; AR: anterior resection; I&D: incision and drainage; PCN: Penicillin G; Tetra: Tetracycline; IUD: intrauterine device; M: month; CT: computer tomography; LGI: low gastrointestinal series; IVP: intravenous pyelography

amination. Only one patient (Case 9) who underwent radical surgery also resumed antibiotic treatment for 1 year. Those undergoing radical surgery did not receive standard antibiotic treatment (for prophylaxis of 1 to 3 months interval).

Discussion

Actinomycosis is an infectious disease caused by the Gram-positive, nonspore-forming anaerobic bacterium, *actinomyces israelii* and less commonly, *actinomyces bovis*, or *actinomyces naeslundii*. The disease was considered to originate by contiguous spread from the intestinal tract during appendectomy, colonic surgical procedure or tissue trauma resulted from any causes because of its inability to penetrate mucosa. The destruction of the mucosal barrier by trauma, operation, endoscopy and immunosuppression were rec-

ognized as a predisposing factor for the penetration of the actinomyces species.^{5,6,9} There has been a dramatic change in the incidence since the 1970s. In 1973, Henderson¹² described the increasing association of pelvic actinomycosis in use of intrauterine devices (IUD). The probability of colonization increases with the duration of use of the IUD. In our series, 4 out of 10 cases had IUD implantation but the duration was not available in chart record. The antibiotics with removal of the IUD may lead to complete resolution if the diagnosis is recognized pre-operatively.¹³ However, the rarity of the disease with diverse manifestations makes the pre-operative diagnosis difficult, especially when intra-peritoneal or retro-peritoneal organs are involved (Figs. 1 and 2). Under the biopsy available by intraoperative or pre-operation CT guided biopsy and microscopy examination, the colonies of actinomyces species (sulfur granules) (Fig. 3) can be detected with surrounding inflammatory infil-

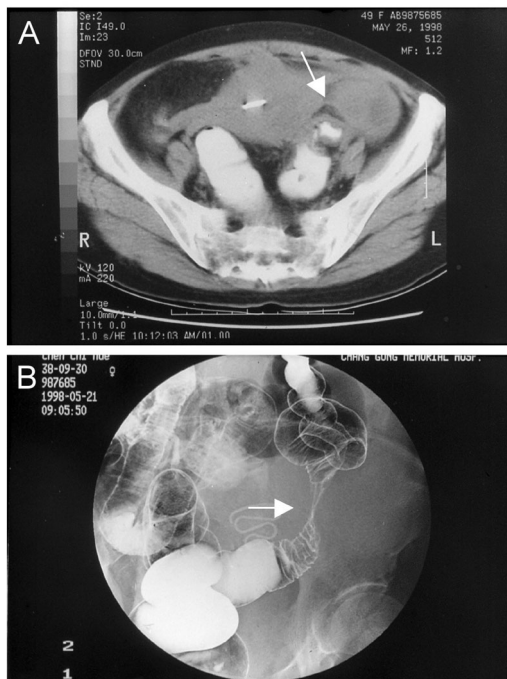


Fig. 1. This patient, Case 9, has irregular bowel movement with dull abdominal pain over left low quadrant. A: Mass lesion with lumen stenosis over junction of S-colon and distal D-colon at abdominal CT examination; IUD at uterus; B: Mucosa lesion with luminal stenosis (apple-cord) at barium enema, double contrast examination.



Fig. 2. This patient, Case 10, has dull abdominal pain over left low quadrant. A: Mass lesion with luminal stenosis at S-colon; B: Mucosa lesion suspicion with luminal stenosis (apple-cord) at barium double contrast examination.

tration.^{14,16} However, it has been seen in pus in only 50% of cases. Actinomyces are also difficult to be cultured because they are fastidious bacteria and sensitive to most antibiotics.²⁵ In our series, no Actinomyces were cultured but sulfur granules were found in specimen. It is sufficient for patients with long-term medical treatment to avoid unnecessary resection, but the dilemma is difficult in obtaining definite diagnosis in time.

Long-term penicillin therapy for actinomycosis has been established, the routine treatment interval from 6 months to 1 year is still variable and it should not be standardized as noted by Eastridge et al²⁴ in 1972. Sudhakar et al. had report of short-term use of antibiotics in treating actinomycosis, but the duration depends on the disease course and response. However, patients with bulky thoracic and /or abdominal disease should not receive short course antibiotics therapy unless surgical debulking is performed.¹⁵ Despite good responses to high dose long-term penicillin

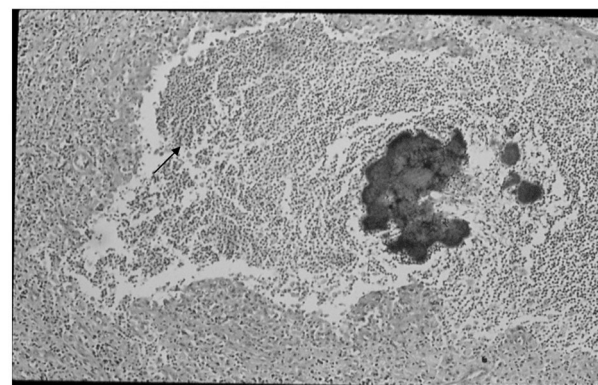


Fig. 3. H&E stain, 400x; the colonies of actinomyces species, sulfur granules, can be detected with surrounding inflammatory infiltration.

treatment that has been reported by some authors, the cure is rarely achieved without an operation to eradicate the inflammatory process. Turnbull et al¹⁷ reported a rectosigmoid stricture in a patient who had

the correct diagnosis after biopsy and whose symptoms resolved completely after antibiotic treatment. However, it might be a temporary response according to Dayan's article. Dayan et al³ presented one case with long-term high dose penicillin treatment that resolved under CT examination 1 year after initiation of treatment. However, the disease reappeared and the surgical intervention was still required to achieve a cure. In 37 cases of actinomycosis treated at the John Hopkins Medical Center, the cure rate rose to 88% after combining a long-term regimen of penicillin with wide surgical excision.¹⁸ In our series, most patients received radical surgery for lesion resection. Two patients received biopsy with bypass of intestinal tract and incisional drainage, and they received subsequent long term antibiotics. Despite no prolonged usage of antibiotics in the eight patients undergoing radical surgery for actinomycosis may achieve a cure, we do not confirm that the relapse of disease may ensue as well as the recurrence in other studies. Especially, these patients had either colonic stricture with suspected malignancy (case 2, 5, 6, and 9) or pelvic mass lesion with suspected malignancy (case 1, 4, 8, and 10). It has been suggested that treatment consists of appropriate antimicrobials complemented by operation. The surgical managements have the role in the disease because the disease may imitate other diseases such as inflammatory bowel disease (Crohn's disease) or cancer. When abdominal actinomycosis can not be demonstrated, surgical resection of lesions simulating malignancy via laparotomy is indicated. Colonic resections or tumor excision also may be necessary in cases either for relief of bowel obstruction.²³ Putman et al reported 122 cases in Mayo clinic,² 84% of patients in this series underwent emergent operation for acute inflammation or perforated bowel lesions. The majority of the patients underwent surgery for abscess drainage or to rule out cancer. The surgical excision of infected or necrotic tissues also has improved the action of antibiotics.^{17,19-22} Wang et al. reported clinical manifestation of actinomycosis in Southern Taiwan.²⁵ Their treatment for cervicofascial, thoracic and pelvic actinomycosis included antibiotics and surgery. Ten cases received pre-operative prophylactic antibiotics and post-operative adjuvant antibiotics were used as different interval (range from 3 to 21 days). Eleven

cases received surgery only without post operative long-term antibiotics usage, and there was no recurrent lesion within 6 months of follow up. Based on a review of the literatures, it is possible to achieve a cure after radical resection even no routine long-term antibiotic treatment. The introduction of antibiotics as an adjuvant role was reported by Putnum et al,² and the prognosis of actinomycosis dramatically changed from 16% to 95%.

Conclusions

In our cases, the standard long-term antibiotic treatment can be used in patients having no radical resection. But close follow-up by CT and clinical examination are important in such cases because disease relapse is possible. Surgical intervention alone can cure cases with infection by complete removal of the infection foci and antibiotics therapy is unnecessary.

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原 著

腹部及骨盆腔放線菌臨床治療結果— 長庚紀念醫院 16 年病例統計

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目的 放線菌感染為一少見的感染，因為其罕見及多樣的表現使得術前診斷十分困難。本文探討腹部及骨盆腔放線菌感染不同於過去臨床上高劑量，長時期的盤尼西林臨床治療方式及結果。

方法 在本文收集了林口長庚醫院大腸直腸外科自 1984 年至 2001 年腹部及骨盆腔放線菌感染病例共 10 例；紀錄所有病例的臨床症狀，子宮內避孕器使用病史，醫學影像，血液生化，外科手術術式，術後抗生素使用時程及術後追蹤狀況。

結果 十位病例平均追蹤時間為 16 個月，其中八位病患因臨床徵狀疑似大腸直腸或後腹腔惡性腫瘤併周邊器官局部侵犯因而接受廣泛性腫瘤切除手術。一位病患接受切片生檢及手術引流；另一位病患則接受切片生檢及阻塞繞道手術。兩位因有無法手術切除的病灶，分別於接受切片引流及阻塞繞道後接受為期 12 及 18 個月的抗生素治療。接受廣泛切除的病例於術後並未接受常規性長期的抗生素治療；這些病例在後續追蹤也沒有復發的徵象。

結論 在本文，未接受廣泛切除的病例於切片生檢確診後可接受常規性長期的抗生素治療，但因放線菌感染在未接受廣泛切除的病例有復發之機會，應密切接受電腦斷層及理學檢查追蹤。透過手術廣泛的完全移除感染病灶可達到痊癒的治療效果，常規性長期的抗生素治療在這類病患並非絕對需要。

關鍵詞 放線菌、腹部及骨盆腔、廣泛切除、子宮內避孕器。