Journal of Korean Medical Science
Vol. 10, No. 2, 142-146 April 1995

Extensive Colonic Stricture Due to Pelvic Actinomycosis

Jin-Cheon Kim, M.D., Moon-Kyung Cho, M.D., Jung-Whan Yook, M.D.,
Ghee-Young Choe, M.D., In-Chul Lee M.D.

Department of Surgery, University of Ulsan College of Medicine and Asan Medical Center

A 36-year-old woman presented with a palpable tender mass at the left lower quadrant of the abdomen. She had suffered from constipation for five years and had a previous history of intrauterine device-use for one year. Preoperative barium enema and abdominopelvic CT showed a compatible finding of rectosigmoid colon cancer or left ovary cancer. She underwent segmental resection of the sigmoid colon along with the removal of left distal ureter, left ovary and salpinx. Pathologic examination revealed actinomycotic abscesses containing sulfur granules. Thereafter, she took parenteral ampicillin (50mg / kg / day) for one month and oral amoxicillin (250mg, tid) for 2 months consecutively. The patient has no specific problems for 6 months after surgical resection and long-term antibiotic therapy. This report may be the first of intrauterine device-associated pelvic actinomycosis involving both sigmoid colon and rectum extensively.

Key Words : *Pelvic actinomycosis, Colonic stricture*

INTRODUCTION

Actinomycosis is a progressive suppurative or granulomatous bacterial disease produced by anaerobic organism, actinomycetes. It is frequently presented with chronic inflammatory induration and multiple draining sinuses. Pelvic actinomycosis is relatively rare and generally associated with use of intrauterine device (IUD). This uncommon clinical condition and various clinical features of the disease make the diagnosis difficult. Actinomycosis needs to identify characteristic sulfur granules on histologic examination. Proper antimicrobial and surgical therapy is generally effective against these infections. A female patient with pelvic actinomycosis involving the rectosigmoid colon and genitourinary tract is presented.

Address for correspondence : *Jin-Cheon Kim, M.D., Department of Surgery, University of Ulsan College of Medicine and Asan Medical Center, 388-1 Poongnap-dong, Songpa-gu, Seoul, 138-040, Korea.*
Tel : 82-2-224-3489, Fax : 82-2-474-9027.

CASE REPORT

A 36-year-old woman was transferred to the Surgical Department, Asan Medical Center, Seoul, due to a palpable tender mass at left lower quadrant of the abdomen. One month prior to admission, she had found the mass incidentally. She had suffered from constipation for five years and used laxatives and cathartics regularly. Three weeks before admission, she had an abdominopelvic CT, barium enema and colonoscopy at the local clinic. The barium enema revealed a diffuse narrowing of the lumen and the thumb-printing appearance of mucosal thickening in the rectosigmoid colon (Fig. 1). The pelvic CT showed marked and diffuse thickening of the rectosigmoid colonic wall. A poorly defined mass with mixed cystic and solid nature was found in the left adnexa which was adherent to the sigmoid colon and obliterated the

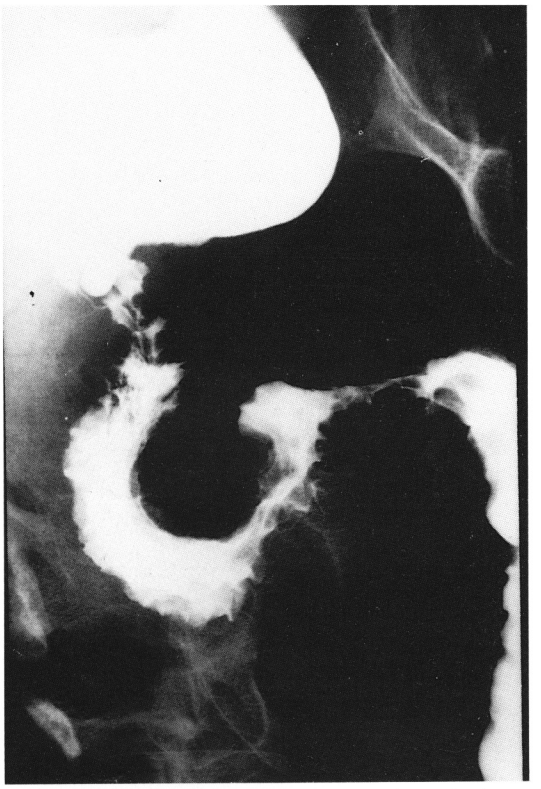

Fig. 1. Single contrast barium enema reveals marked narrowing of the rectosigmoid colon. The mucosal folds are thickened and thumb printing filling defects are noted in the rectosigmoid colonic wall indicating submucosal edema or infiltration.

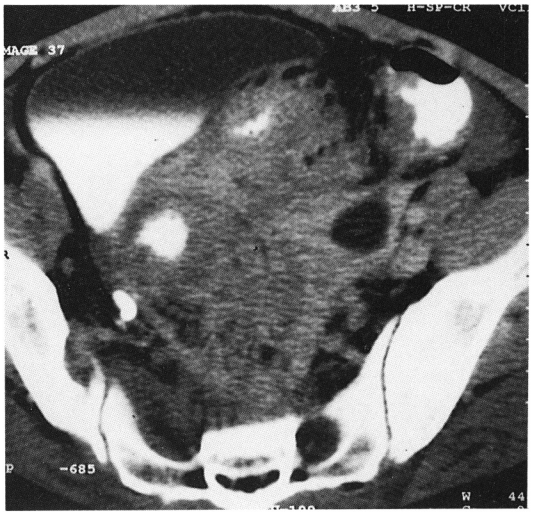

Fig. 2. Post-contrast enhanced pelvic CT image shows large ill-defined cystic and solid complexed mass lesion in the left adnexa, which is adherent to rectosigmoid colonic wall. The rectosigmoid colonic wall is markedly and diffusely thickened due to adjacent inflammatory lesions in the left adnexa.

left distal ureter causing hydronephrosis of the left kidney (Fig. 2). She had a previous history of an IUD for one year until three years before admission. Her family or social history was not contributory. She had suffered from severe constipation, tenesmus, back pain, lower abdominal pain and narrow-caliber stool. She had no weight loss. On physical examination, the patient appeared thin and weak. The abdomen was soft and flat, but a slight tender mass was palpable at the left lower quadrant. No abnormal finding was found on manual rectal examination. Vital signs revealed a normal range. Colonofiberscopic finding showed a severely stenotic rectosigmoid colon which was rigid as well as severely inflamed. Further advancement into the proximal sigmoid colon was not possible due to stenosis. Histologic finding revealed chronic inflammation only without evidence of neoplastic change. Transvaginal ultrasonography revealed an ill-defined, mixed echogenic mass, measuring 10 ×8cm in the left adnexa. Six days later, surgery was performed under the impression of rectosigmoid colon cancer or left ovarian tumor. There was small amount of ascites in the peritoneal cavity. The rectosigmoid colon, left ovary and left distal ureter were conglomerated to make an ill-defined, hard mass fixed to the posterior pelvic wall. Frozen biopsy from the fibrotic mass, adjacent mesenteric lymph nodes and rectal wall did not show any tumor cells. Because of severe stenosis and complete obstruction of the colon, a 25cm length of the sigmoid colon was resected combining the left distal ureter, left ovary and salpinx. Hartmann's procedure, transureteroureterostomy with double J-stent insertion and incidental appendectomy were performed. The mucosal surface of the large intestine showed an ulcerating tumorous lesion, measuring 7×5×4cm, which extended into pericolic fatty tissue. This ill-defined tumor obliterated the rectal lumen circumferentially. The rectosigmoid colon disclosed an extensive infiltration of inflammatory cells in the submucosa and muscle layer on histologic examination (Fig. 3). Numerous actinomycotic abscesses containing numerous sulfur granules with charac-

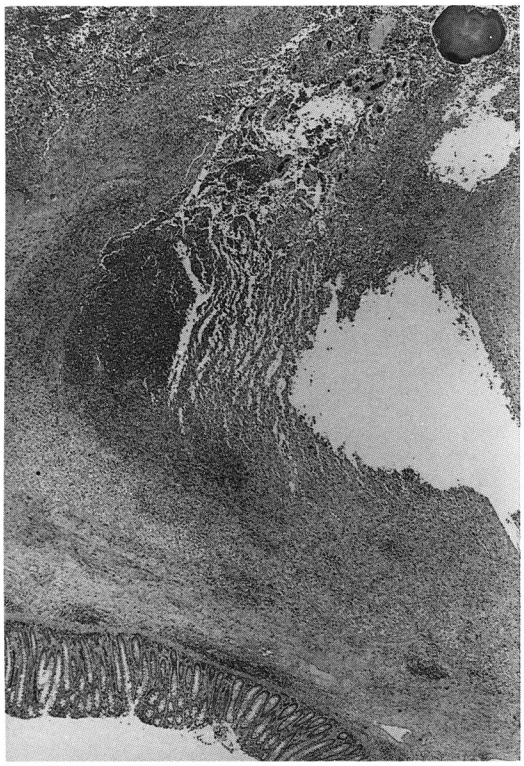

Fig. 3. Infiltrates of acute and chronic inflammatory cells with abscess formation are prominent in the subserosal and proper muscle layer, which extend to submucosa. A sulfur granule is also identified in the subserosal layer (right upper corner)(H & E,×20).

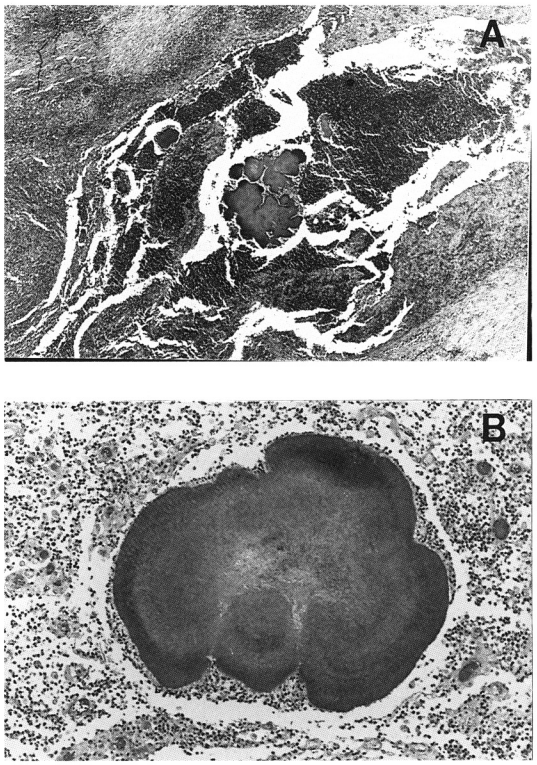

Fig. 4. Photomicrograph shows actinomycotic abscesses containing sulfur granules with radiating filaments in fibrotic serosa of intestine and pericolic fatty tissue(A)(H & E, × 100). A magnified view of the characteristic sulfur granule (B)(×400).

teristic radiating filaments were identified in fibrotic serosa of intestine and pericolic fatty tissue (Fig. 4 A & B). However, both aerobic and anaerobic culture failed to detect organisms. Because penicillin G was not available, ampicillin (50mg/kg/day) was administered intravenously for the first 4 weeks postoperatively, followed by oral amoxicillin (250mg, tid) for 2 months. Postoperative course was generally uneventful. Six months after resective surgery, she is nearly completely free of symptoms at the present time and waiting for a restorative surgery for the colon.

DISCUSSION

Actinomycosis is a chronic suppurative and granulomatous bacterial infection. The causative agent is Actinomyces israelii, showing gram-positive, non-spore-forming anaerobic microaerophilic which may be normal flora in the mouth, bowel and female genital tracts (Sullivan and Goldworthy, 1940). Some stimulus is thought to make this organism become pathogenic. In the past, this organism has been misclassified as fungus, however, the species is closer to bacterium than fungus. Collection of organism and debris may look like a granule of elemental sulfur in the draining lesion. Microscopic examination of the sulfur granule reveals a central core of intertwined branching filaments making radiating clublike projection at the periphery of the granule (Brewer et al., 1974). These sulfur granules suggest actinomycosis, but are not always conclusive unless found in the viscera (Robby and Vickery, 1970).

Three major clinical presentations of this disease include the cervicofacial, thoracic and abdominal regions. Actinomyces israelii infection almost always involves the cervicofacial area, dental abscess or

trauma (Brewer et al., 1974). The disease may last for months or years, but not be fatal. Thoracic actinomycosis is thought to result from aspiration of infected particles or may develop as a result of burrowing of hepatic abscesses through the diaphragm. The abdominal form frequently occurs after appendectomy, appendiceal abscess or traumatic perforation of the bowel. The infection in the cecal area usually comes from appendectomy and results in an indurated, firm, pericecal mass. It may progress into the formation of indolent sinuses draining to the abdominal skin if untreated. Although rare, over 250 cases of pelvic actinomycosis have been reported in the English literature (Stevenson, 1957 ; Brady et al., 1964).

The pathogenesis of the pelvic actinomycosis is thought to be either an ascending infection from the lower genital tract or a spread from an intestinal lesion (Henderson, 1973). Recent reports have documented an increased incidence of pelvic actinomycosis in women using IDUs (Duguid et al., 1980). IUDs placed for a long period elicit an inflammatory response on the surface of the endometrium (Sagiroglu and Sagiroglu, 1970 ; Gupta et al., 1971 ; Segal et al., 1975). The most probable route of infectious spread appears to be a patent tubal structure (Luff et al., 1978). Actinomyces can be identified in approximately 10% of asymptomatic IUD-users in the routine vaginal examination and can be in as many as 25% of IUD-users with symptoms (Gupta and Woodruff, 1982 ; Muller-Holzner et al., 1990). The risk of infection increases with the longer duration of IUD-use. Intestinal obstruction due to IUD-associated genital actinomycosis is very rare. However, when IUD-users complain of abdominal pain, menstrual irregularity, adnexal mass or symptoms suggestive of intestinal obstruction, the physician must always consider the possibility of actinomycosis (Valicenti et al., 1982). Less than 10 cases of rectal stricture due to IUDs-associated pelvic actinomycosis have been reported from both domestic and world literature (Ratliff et al., 1986 ; Lee et al., 1989). Furthermore, sigmoid colon is shown to be spared from pelvic actinomycosis because of its anatomical location and redundancy. This rare case of a sigmoid stricture probably came either from the retroperitoneal or rectal spread of a prolonged pelvic actinomycosis. Anorectal involvement may result from secondary extension of an abdominal infection or may arise primarily in anorectal region. The anorectal form of actinomycosis may begin from the anal crypt resulting in fistula or abscess formation (Brewer et al., 1974 ; Alvarado-Cerna and Bracho-Riquelme, 1994).

Infection with Actinomyces israelii is endogenous and usually spreads via the alimentary canal rather than by lymphatics or hematogenous routes. However, further spread may occur through vascular or lymphatic cannels. The only tissue which seems to escape consistently is the surface epithelium (George et al., 1968). It is rarely transmitted from man to man by accidental inocculation. Actinomyces israelii can remain dormant in avasular tissue for a long latent period. This organism is shown to be incapable of penetrating into the intact mucosa. Infection is postulated to be preceded by inoculation at a break in the mucosal barrier due to disease, trauma or the presence of a foreign body.

Diagnosis is seldom made preoperatively because of no reliable or specific clinical manifestation and the relative infrequency of the disease. It should be suspected in case of an indolent mass with fistulae or chronic sinus. It is characterized by chronic inflammatory induration and multiple draining sinus formations. The histologic demonstration of sulfur granules in the drainage fluid from fistulae or surgically resected specimen is highly suggestive of actinomycosis, but is not pathognomonic. Diagnosis can be confirmed by culture of causative organism. Agglutination reaction or complement fixation test is not satisfactory (Pheils et al., 1964). In this case, the preoperative barium enema and abdominopelvic CT were compatible with rectosigmoid colon cancer or left ovary cancer. Although the preceding culture of drainage for aerobe and anaerobe failed to reveal any growth of microorganisms, histologic examination revealed actinomycosis.

Treatment consists of long-term antibiotic therapy and adequate surgery, including incision and drainage of abscess, removal of persistent sinuses, and excision of necrotic or infected tissue if possible. Penicillin and tetracyclin are both effective, while antifungal agents are not effective. Penicillin therapy should consist of at least one million units of penicillin G daily. A tetracyclin regimen of 500mg four times a day for 8 to 12 weeks is effective in most cases. Antimicrobial therapy should be continued until all signs of inflammation disappear and this may take from several months to one year or more. A high concentration of antibiotics is needed to penetrate the fibrotic area and the colony of the organism itself. Follow-up observation to ensure complete healing is

mandatory.

REFERENCES

Alvarado-Cerna R, Bracho-Riquelme R. *Perianal actinomycosis-a complication of a fistula-in-ano. Dis Colon Rectum 1994; 37: 378-80.*

Brady HH, Doughterty CM, Mickal A. *Actinomycosis of the female genital tract. Obstet Gynecol 1964; 23: 580-3.*

Brewer NS, Spencer RJ, Nichols DR. *Primary anorectal actinomycosis. J Am Med Assoc 1974; 228: 1397-9.*

Duguid HL, Parratt D, Traynor R. *Actinomyces-like organisms in cervical smears from women using intra-uterine contraceptive devices. Br Med J 1980; 1: 534-7.*

Georg LK, Coleman RM, Brown JM. *Evaluation of an agar gel precipitin test for the serodiagnosis of actinomycosis. J Immunol 1968; 100: 1288.*

Gupta PK, Malkani PD, Bhasin K. *Cellular response in the uterine cavity after IUD insertion and sturctural changes of the IUD. Contraception 1971; 4: 375-84.*

Gupta PK, Woodruff JD. *Actinomyces in vaginal smears. JAMA 1982; 247: 1175-6.*

Henderson SR. *Pelvic actinomycosis associated with an intrauterine devices Obstet Gynecol 1973; 41: 726-32.*

Keebler C, Chatwani A, Schwartz R. *Actinomycosis infection associated with intrauterine contraceptive devices. Am J Obstet Gynecol 1983; 145: 596-9.*

Lee YJ, Kim HK, Choi CS, Lee JJ, Kim HK, Park YE. *Rectal stricture due to pelvic actinomycosis. J Kor Surg Soc 1989; 36: 541-5.*

Luff RD, Gupta PK, Spence MR, Frost JK. *Pelvic actinomycosis and the intrauterine contraceptive device. Am J Clin Pathol 1978; 69: 581.*

Muller-Holzner E, Gschwendtner A, Abfalter E, Solder E, Schrocksnadel H. *Actinomycosis and long-term use of intrauterine devices. Lancet 1990; 336: 939.*

Pheils MT, Reid DJ, and Ross CF. *Abdominal actinomycosis. Br J Surg 1964; 51: 345.*

Ratliff DA, Carr N, Cochrane JPS. *Rectal stricture due to actinomycosis. Br J Surg 1986; 73: 589-90.*

Robby SJ, Vlckery AL Jr. *Tinctorial and morphologic properties distinguishing actinomycosis and norcardiosis. N Engl J Med 1970; 282: 593-6.*

Sagiroglu N, Sagiroglu E. *The cytology of intrauterine contraceptive devices. Acta Cytol 1970; 14: 58-64.*

Segal SJ, Atkinson LE. *Changing concepts of intrauterine contraception. Prog Gynecol 1975; 6: 381-99.*

Stevenson AE. *Actinomycosis of ovaries and fallopian tubes. J Obstet gynecol Br Commonw 1957; 64: 345-7.*

Sullivan HR, Goldworthy NE. *A comparative study of anaerobic strains of actinomyces from clinically normal mouths and from actinomycotic lesions. J Pathol Bacteriol 1940; 51: 253-61.*

Udagawa SM, Portin BA, Bernhoft WH. *Actinomycosis of colon and rectum. Dis Colon Rectum 1974; 17: 687.*

Valicenti JF, Pappas AA, Craber CD, Williamson HO, Fowler Willis N. *Detection and prevalence of IUD-associated Actinomyces colonization and related morbidity. A prospective study of 69925 cervical smears. JAMA 1982; 247: 1149-52.*