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Ileocecal actinomycosis : A case report and review of the literature

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SUMMARY : A case of actinomycosis in the ileocecal area of a 44-years-old man which manifested as a painful lump in the right iliac fossa is reported. Clinical and laboratory findings suggested the diagnosis of perityphlitic abscess due to acute appendicitis. Upon a laparotomy, a malignant tumor was suspected and so a right hemicolectomy was performed. Gram staining of cecum tissues demonstrated gram positive pleomorphic filaments. Postoperative penicillin therapy was successful. Despite the fact that infection accompanying an actinomycotic organism is relatively rare, the possibility of such an infection should be kept in mind because the organism is known to be commensal in the oral cavity, lungs and intestinal tract.

INTRODUCTION

A case of ileocecal actinomycosis presenting as acute appendicitis with a lump in the right iliac fossa is reported. As the numbers of antibiotics and indications for their use have increased, actinomycosis has almost become a medical rarity in Japan as well as in western countries. Abdominal actinomycosis also is a rare condition which is difficult to diagnose, because of its close resemblance to other, more common conditions, and therefore diagnosis can only be established by histological examination. If the disease is recognized, the prognosis is good because antibiotic treatment is very effective. In the present report, recent literature findings are also reviewed and their clinical and radiological features are compared.

CASE REPORT

A 44-year-old man was urgently admitted on September 22, 1985 to the St. Francis Hospital because of ileocecal mass and right lower abdominal pain.

He began to complain of abdominal pain with nausea on August 1st, 1985. The white blood cell count was 12,800/mm^3, but no mass was palpable in the abdomen. A diagnosis of acute appendicitis was made and Cefmetazon 1,500mg per day was given intravenously for three days, followed by Cefaloxin, resulting in complete remission. Three weeks later, he again developed a pain in the right iliac region. Antibiotics were given with good results.

On September 21, he began complaining of the same abdominal pain, and consulted an other surgeon who pointed out an abdominal mass in the right iliac fossa with leucocytosis of 12,300/mm^3. Perityphlitic abscess was suspected and he was transferred to the Department of Surgery of St. Francis Hospital.

On admission, physical examination revealed a well-nourished man with a fever of 37.1℃. He had a regular pulse rate and blood pressure

of 156/80 mmHg. Breath sounds were normal and vesicular, and heart sounds were clear.

The abdomen was flat and soft, and a hard mass about 7 cm in diameter with moderate tenderness was felt in the right iliac fossa. There was localized tenderness with rebound tenderness at McBurney's point. Rectal examination revealed neither melena nor tenderness.

Initial laboratory data showed an Hb level of 14.7 g/dl and Ht of 44.0%. The white blood cell count was 8,600/mm^3 with 71% neutrophils. Total serum protein levels was 7.6 g/d ℓ. In liver function tests, normal level of bilirubin were detected with 0.5 total, 0.37 direct, GOT 27 IU/ℓ, GPT 38 IU/ℓ, Al-pase 13.8 IU/ℓ, ZTT 2.0 K, Ch-E 0.79 $\triangle$pH, gamma-GTP 225 and LAP 426 IU/ℓ. Serum amylase was normal at 113 IU/ℓ, urea nitrogen 12.8 mg/d ℓ, creatinine 1.3 mg/d ℓ and CPK 51 IU/ℓ. Serum electrolytes were Na 143 mEq/ℓ, K 4.0 mEq/ℓ, Cl 105 mEq/ℓ, and Ca 9.58 mEq/ℓ. Urinalysis was negative. Occult blood was found in the stools.

A plain abdominal X-ray showed a normal gas pattern. Barium enema taken on the day after admission showed a relatively soft mass shadow with mucosal irregularity of the cecum and fixed narrowing of the terminal ileum. The appendix showed irregularity (Fig. 1).

Abdominal ultrasonography showed a hypoechoic mass in the ileocecal region associated with an echogenic band which seemed to be the swollen wall of the appendix.

Two days later, abdominal pain and fever disappeared, and emergency operation was performed because perityphlitic abscess due to acute appendicitis was suspected.

During laparotomy, a large tumor mass without fluctuation was found infiltrating the wall of the cecum and invading the appendix, terminal ileum, omentum and parietal peritoneum. Right hemicolectomy with ileotransversostomy was carried out.

After the operation, treatment with antibiotics was started, resulting in complete remission from all symptoms.

Macroscopic examination of the resected specimen revealed an elevated lesion 7cm in diameter in the serosal side of the cecum. The overlying mucosa including ileocaecal valve was edematous and showed a shallow ulcerated lesion above the mass (Fig. 2). A longitudinal section through that area showed the presence of a large, ill-defined mass in the wall of the cecum. The tumor was seen to infiltrate the wall of the terminal ileum. The lesion was mostly

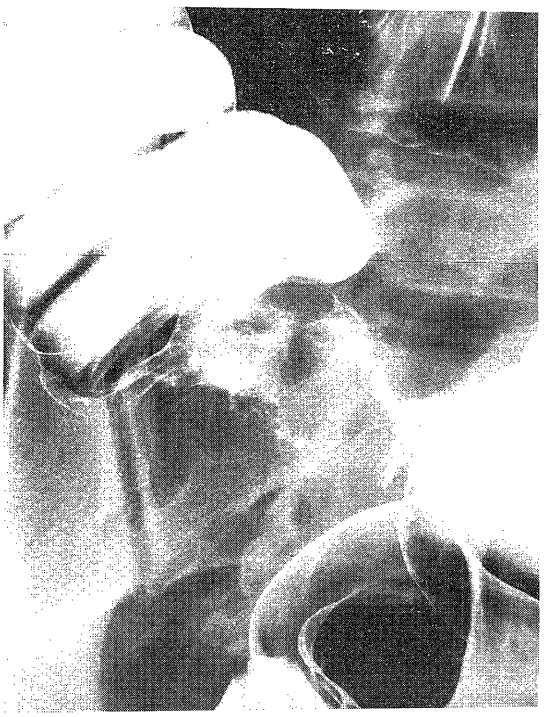

Fig. 1 Barium enema showing narrowed segment of the terminal ileum and irrequrality of the cecum.

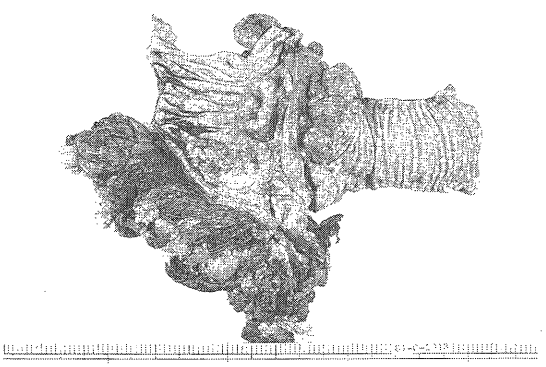

Fig. 2 Macroscopic view of the resected specimen

grayish-white with some small yellow spots.

Microscopic examination disclosed signs of chronic non-specific inflammation in the mucosa and submucosa. The submucosa was scarred by fibrous bands and contained a moderately increased inflammatory infiltrate. The serosa was considerably thickened. It showed extensive inflammation with areas of granulation tissue, more fibrous zones, and areas containing a heterogeneous inflammatory infiltrate composed of polymorphs and mononuclear elements. Many small abscesses were scattered in the serosa. In these abscesses, irregular colonies of Actinomyces (sulfur granules) were found (Fig 3).

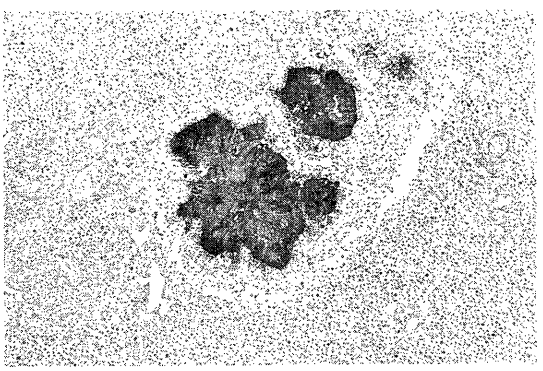

Fig. 3 Sulfur granules are enveloped by the purulent exudate.

In the wall of the appendix, a similar inflammatory reaction was present. No evidence of malignancy was found.

DISCUSSION

Abdominal actinomycosis is an uncommon clinical condition [8]. Actinomycosis is a chronic, progressive and suppurative infection, characterized by the formation of multiple abscesses, draining sinuses, abundant granulation and dense fibrous tissue [5].

BRADSHAW [1] is credited with the first description, in 1846, of a patient with abdominal actinomycosis. BOLLINGER [3] had initially described the development of hard masses that occurred in the jaw bones of cattle and from which he noted binding mycelia. ISRAEL [2] noted granules obtained from human autopsy material which contained the same mycelia as those described by BOLLINGER [3] in cattle. The diagnosis of actinomycosis was first made in living man by PONFICK [4] in 1879.

The incidence of actinomycosis has been declining in recent years since the use of antibiotics has become widespread.

The infective agent is Actinomyces israelii, a gram-positive filametous anaerobic bacterium, normally residing in the mouth.

There is no discernible sex predilection, most cases occurring in adolescents and middle-aged individuals [5]. It causes an endogenous and non-contagious inflammation in hypoxic tissue. This is the reason why cervicofacial localization is most frequent after a trauma in that region.

Thoracic actinomycosis is an infection of the lung, probably due to aspiration, whereas the pleura, mediastinum and chest wall are probably involved by contiguity.

Abdominal actinomycosis shows a predilection for the right iliac fossa and has frequently followed operations for acute appendicitis or drainage of abscesses in this region.

The frequency with which abdominal actinomycosis occurs as compared with the cervicofacial and thoracic forms of the disease varies. In some studies, it ranks first [6], in others third [7] and in most second [8,9].

We have made reference to the 53 cases of actinomycosis reported during the past five years in Japan (1981-86), and 298 cases in the

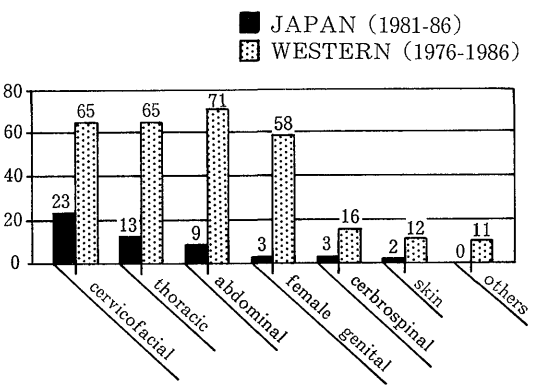

Fig. 4 Reported cases of actinomycosis in literature ; Sites of infected organs

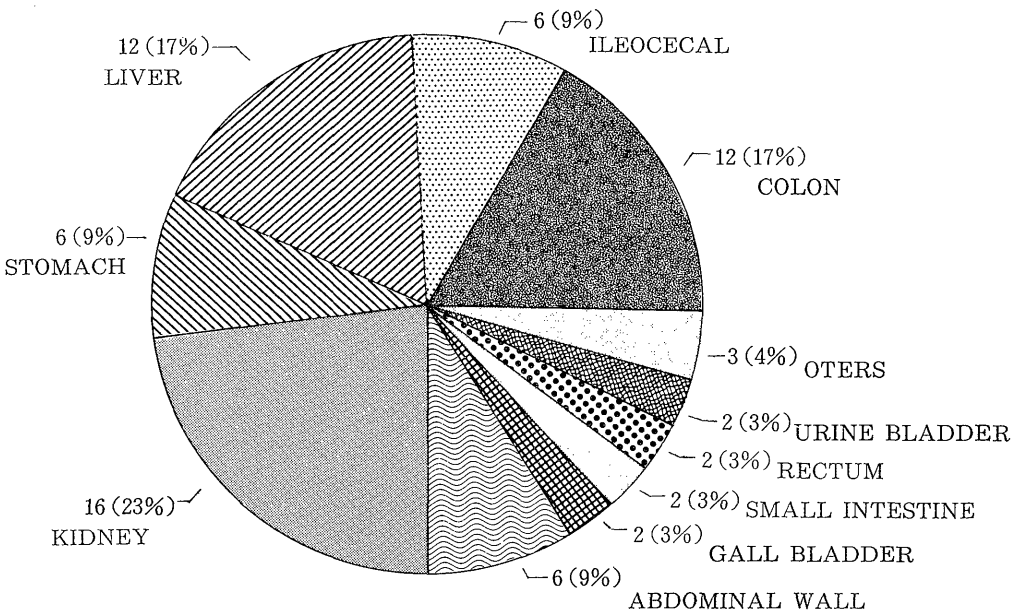

Fig. 5 Sites and cases of abdominal actinomycosis ; 69 case in English literatures (1976-1986)

English literature (1976-86), respectively. As Table 1 indicates, among the collective cases in the Japanese literature, cervicofacial actinomycosis ranks first at 54.8%, followed by thoracic involvement at 23.8%, abdominal at 11.9 %, female genital organs at 4.8% and others at 4.8%, while among those in the English literature, abdominal actinomycosis ranks first at 25.8%.

Duncan et al.[10] reported that actinomycosis in general, and the abdominal form in particular, may be significantly decreasing in incidence in western countries. However, the incidence of actinomycosis is difficult to estimate.

Abdominal actinomycosis has a predilection for the terminal ileum, cecum and appendix. The appendix is by far the most common intra-abdominal organ involved [6)11)12)], followed by much less common involvement of the colon, stomach, liver, gallbladder, pancreas, small bowel, anorectal region, pelvis, abdominal wall and other less common sites. How Actinomyces gains access to the abdominal organs to begins its characteristic chronic suppurative inflammatory course remains a matter of controversy [13)]. The most plausible theory seems to be that only when the normal barrier of an intact gastrointestinal mucosa has been destroyed by a disease process or by trauma can the organism penetrate into adjacent tissue and become pathogenic, as in the oral cavity [12)].

The clinical aspects of abdominal actinomycosis are multiple and variable [14)5)]. Although actinomycosis generally presents as a chronic, localized inflammatory process associated with fever and leucocytosis, with the absence of a draining sinus, the diagnosis is often not suspected. There is usually a latent interval of days to weeks between the onset of symptoms and previous clinical presentation which often involves perforation or previous surgical procedures, and a persistent draining sinus, particularly following operation for a perforated viscus [8)]. It should be remembered that in the initial stage of the disease process, it may be impossible to clinically distinguish abdominal actinomycosis from other disease processes [15)16)].

Localization to the appendix may very well simulate acute appendicitis with or without perforation. A palpable mass in the right iliac fossa may simulate an inflammatory

process or carcinoma of the cecum [17) 8)].

In general, the diagnosis is based on culture or microscopic examination, typical yellow " sulfur granules " may be seen in the pus.

The same type of colony may be seen in colonoscopic biopsy specimens. However, a preoperative diagnosis of ileocecal actinomycosis is almost impossible, except in rare cases of abdominal fistula from which pus is available for culture.

Radiographic studies frequently suggest a malignant tumor or an ulcer. As in our case, barium enema examination may show a mass with extrinsic compression and narrowing of the cecum, suggestive of submucosal tumor. Angiographic examination has been used in a case of actinomycosis of the abdomen [18)], although the angiograph obtained did not show any changes that could be regarded as specific for actinomycosis.

Both CT scan and ultrasonic examination may also be useful modalities for differential diagnosis from perityphlitic abscess due to acute appendicitis. SILVERMAN [19)] *et al.* demonstrated the CT appearance of actinomycosis in the neck, in which a thickwalled enhancing mass with a low attenuation center was identified between the muscle. LINGER *et al.* [20)] reported a case of pelvic abscess caused by actinomycosis in which the diagnosis had been established preoperatively by transvaginal fine needle aspiration.

In most cases, the endoscopic findings have been suggestive of a circumscribed and ulcerated cecal carcinoma. It was not possible to make the diagnosis on the basis of endoscopy alone, since no pus was available and the biopsies were not specific due to the submucosal localization of the abscesses.

In most cases, as in ours, the surgeon found a mass that was large than that suspected preoperatively and was invasive to various abdomimal organs. Macroscopic examination of the operatively specimen revealed the presence of an ulcerated submucosal tumor.

Since the tumor is usually ill-defined and infiltrative to the surrouding tissues, some form of malignant proliferation, such as leiomyosarcoma, is sometimes considered. The is not like those usually seen in these types of tumor. A correct diagnosis is only established by Gram-staining.

Prior to the advent of antibiotics, surgical treatment was probably the most effective modality in the management of abdominal actinomycosis, in spite of the multitude of nonsurgical therapeutic measures that were also used [13)]. If a diagnosis of actinomycosis is established through any means, antibiotics including penicillin should be considered as the first choice [12) 21)]. Surgical treatment in the antibiotic era still remains essential in many instances of abdominal actinomycosis. Althogh surgery is generally limited to incision and drainage with curettage of abscess cavities or sinus tracts, a wide, aggressive approach may be required, given certain conditions. In most cases, a right hemicolectomy has been performed [22)].

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