Ileocecal actinomycosis: A case report
and review of the literature

Takeyuki Ohmagari*, Toshio Miura**, Shigehiro Hashimoto*,
Takatoshi Shimoyama**, Tohru Nakagoe**, Ichiro Sekine***

*Department of Surgery, St. Francis Hospital
**First Department of Surgery, Nagasaki University School of Medicine
***Atomic Disease Institute, Nagasaki University School of Medicine

Received for publication, June 30, 1988

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. De-
spite the fact that infection accompanying an actinomycotic organism is relatively
rare, the possibility of such an infection should be kept in mind because the organ-
ism 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 anti-
biotics 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 exami-
nation. If the disease is recognized, the progno-
sis is good because antibiotic treatment is very
effective. In the present report, recent litera-
ture 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/mm³, 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. Anti-
biotics 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/mm³. 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°C.
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/mm³ with 71% neutrophils. Total serum protein levels was 7.6 g/dl. In liver function tests, normal level of bilirubin were detected with 0.5 total, 0.37 direct, GOT 27IU/l, GPT 38IU/l, Al-pase 13.8IU/l, ZTT 2.0K, Ch-E 0.79ΔpH, gamma-GTP 225 and LAP 426IU/l. Serum amylase was normal at 113 IU/l, urea nitrogen 12.8mg/dl, creatinine 1.3mg/dl and CPK 51IU/l. Serum electrolytes were Na 143mEq/l, K 4.0mEq/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
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. 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. 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, in others third, and in most second.

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 begin 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). 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 pre-operative 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 larger than that suspected preoperatively and was invasive to various abdominal 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 surrounding 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 non-surgical 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. Although 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 hemicolecctomy has been performed (22).

REFERENCES

9) Weese, WC and Smith, IM : A study of 57