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Cervicofacial Actinomycosis in Children*

Michael E. Friduss, MD,[†] and Dennis R. Maceri, MD[‡]

Actinomycotic infections, once common in humans and cattle, are now rare causes of disease in man. This general group of organisms belongs taxonomically between the true bacteria and the fungi; however, the organisms behave clinically like true anaerobes. The organism, although phagocytized by the host cells, is not killed. Therefore, it is defined as a facultative intracellular parasite of the host. The protean manifestations of actinomycotic infections often mimic infectious processes, such as osteomyelitis or granulomatous disease, as well as neoplasia. It is therefore important for the surgeon to include actinomycosis in the differential diagnosis of cervicofacial masses in children as well as in adults. Being an anaerobe, the organism is difficult to culture and the diagnosis must be considered at the time of biopsy of a cervicofacial mass or when culturing a sinus tract.

A case of a 12-year-old girl seen at Children's Hospital of Los Angeles prompted the review of our experience with this disease. In this child, actinomycosis presented as an expansile mass in the mandible. From 1956 to 1986, five children were treated for cervicofacial actinomycosis. Detailed case analysis, pathophysiology, clinical characteristics, and current treatment recommendations are presented. (Henry Ford Hosp Med J 1990;38:28-32)

Since the advent of modern antibiotic therapy, cervicofacial actinomycosis is a rare infection (1). With the decline in frequency, the condition may not be included in a differential diagnosis and its presence not recognized. The pediatric literature has few reports regarding actinomycotic infection (2-5). At Children's Hospital of Los Angeles (CHLA) we recently managed a 12-year-old child with cervicofacial actinomycosis. The difficulty with diagnosis and management prompted a review of the cases at our hospital. From 1956 to 1986, four other children were treated for cervicofacial actinomycosis. Analysis of these cases is presented to illustrate important clinical aspects of the diagnosis and management.

Pathophysiology of Cervicofacial Actinomycosis

Actinomyces israelii is the causative bacteria for most human cases of actinomycosis. Recovery of this fastidious anaerobic organism is reported in fewer than 25% of infected sources (6). *Actinomyces* is a gram-positive bacillus with a variable morphologic appearance. When freshly cultured, the organism grows as branching filaments (Fig 1). However, when it is recovered from an infected source, increased septation leads to fragmentation into bacillary and coccoid elements of irregular diameter. These are indistinguishable from diptheroids (7). Sulfur granules, when present, are pathognomonic of actinomycosis (Fig 2). However, many authors have failed to demonstrate sulfur granules in purulent drainage or tissue specimens from documented actinomycotic infections (1,6).

Actinomyces species normally inhabit the oral cavity and can be found throughout the gastrointestinal tract. The three distinct forms of actinomycotic infection are cervicofacial, pulmonary, and abdominal, with cervicofacial being the most frequent.

Weese and Smith (8) found that over a 36-year interval 49% of cases were cervicofacial, 18% were pulmonic, and 23% were abdominal. Because the organism cannot penetrate mucosa, preexisting mucosal injury or dental disease is required for initial inoculation. Minor oral trauma, tic lesions, carious teeth, or the tonsillar fossae may provide initial portals for entry. *Actinomyces* spread by direct extension into the soft tissues of the head and neck (1). The infection may begin with an acute inflammation, as a fluctuant abscess, or as a less virulent granulomatous reaction. The Table lists the five cases of cervicofacial actinomycosis treated at CHLA. Cases 1 through 3, separately addressed herein, exemplify the protean manifestations of this disease.

Methods

Five patients with manifestations of cervicofacial actinomycosis were treated at CHLA between 1956 and 1986. The patients included three females and two males ranging in age from 10 months to 12 years. The diagnosis of actinomycosis was made at CHLA in four of the five cases and all patients were treated there. The records have been reviewed to determine the epidemiology, pathogenesis, microbiology, and treatment in these five patients.

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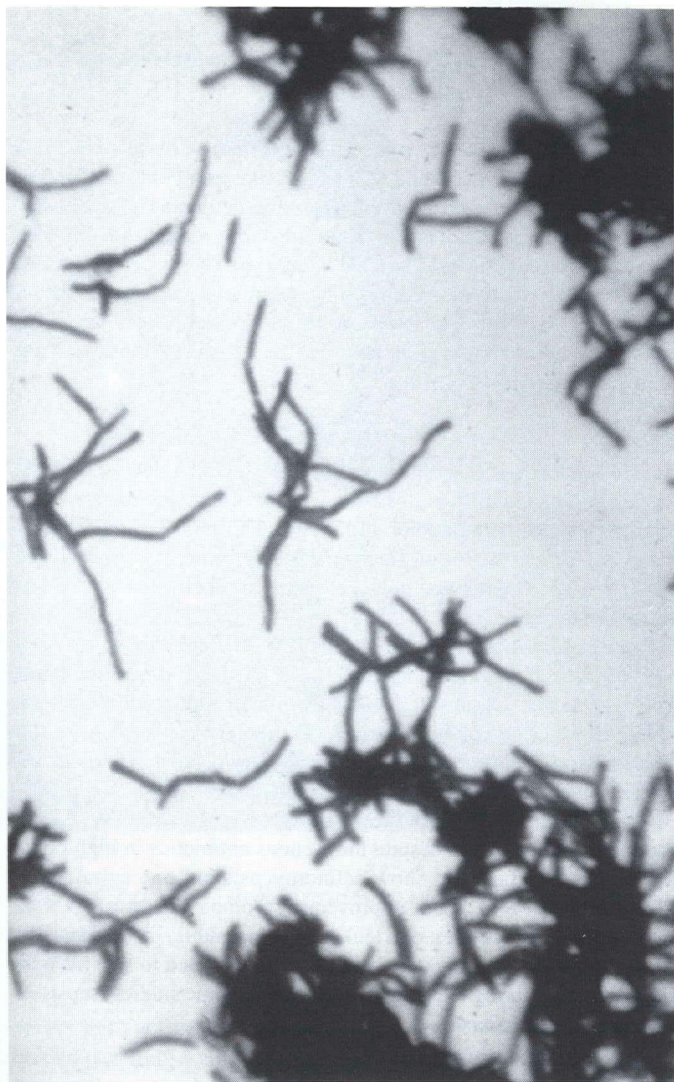


Fig 1—Photomicrograph of cultured *Actinomyces israelii*.

Case Reports

Case 1

A 12-year-old girl first presented to the CHLA emergency room with a three-year history of intermittent pain and swelling of the right mandible. There was no history of oral trauma or dental disease. Diffuse enlargement and tenderness of the mandibular angle was associated with marked trismus. Radiographs revealed soft tissue swelling and sclerosis of the ramus (Fig 3). The only significantly abnormal blood test was an elevated sedimentation rate. Computed tomography (CT) revealed a calcified cyst in the angle of the mandible (Fig 4A) and a heterogeneous expansile process extending into the condyle (Fig 4B). Antibiotics given orally and nonsteroidal anti-inflammatory medications provided transient improvement, but cessation of antibiotic therapy was followed by recurrent symptoms. Intraoral open biopsy was performed with curettage of the calcified cyst and abnormal bone. Cultures for bacteria, acid-fast bacilli, and fungi were negative. Initial improvement was followed by a return of swelling, pain, and trismus one month later. Repeat CT revealed vertical enlargement of the right mandibular ramus with increased lucency compared to the previous study. At reoperation the mandibular ramus disclosed diffusely thickened bone with a soft central core. Although no sulfur granules were seen, cultures of the spec-

Table
Summary of the Five Patients with Actinomycosis from the Children's Hospital of Los Angeles

Case	Sex	Age	Presentation	Sulfur Granules	Gram Stain (Hyphae)	Culture	Surgical Procedures
1	F	12 yr	Mandibular mass	—	+	+	4
2	F	5 yr	Neck mass	+	+	+	11
3	M	10 mo	Mandibular mass with ulceration	—	+	—	4
4	F	4.5 yr	Neck mass	+	+	—	2
5	M	10 mo	Recurrent branchial cleft cyst	+	+	—	6

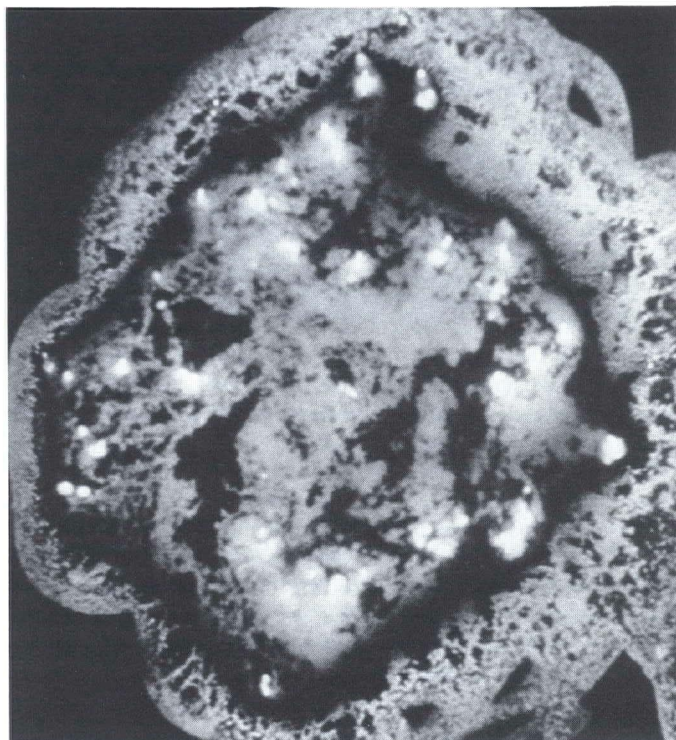


Fig 2—Photomicrograph (low power view) of sulfur granule. Note central dense mass with periphery of radiating mycelial elements.

imen grew *Actinomyces* and *Veillonella* species. High dose intravenous therapy with penicillin G (12 mU/day) was given. Despite an initial response, she continued to have recurrent pain and trismus with an elevated sedimentation rate. A third surgical procedure demonstrated a soft, paste-like marrow cavity and a thickened outer cortex. Cultures were negative. For six additional weeks she was treated with parenteral antibiotics via a Hickman catheter. Hyperbaric oxygen treatments were also given. When the symptoms still failed to resolve, the lesion was subjected to further debridement. At surgery the surface of the mandible was covered with a 1 mm layer of soft vascular cancellous bone. The lateral cortex was removed from the sigmoid notch to the lateral incisor and the mandible was reconstructed with a bone graft from the iliac crest. After another six-week treatment with parenteral antibiotics, the patient remains well and has no further evidence of recurrent disease.

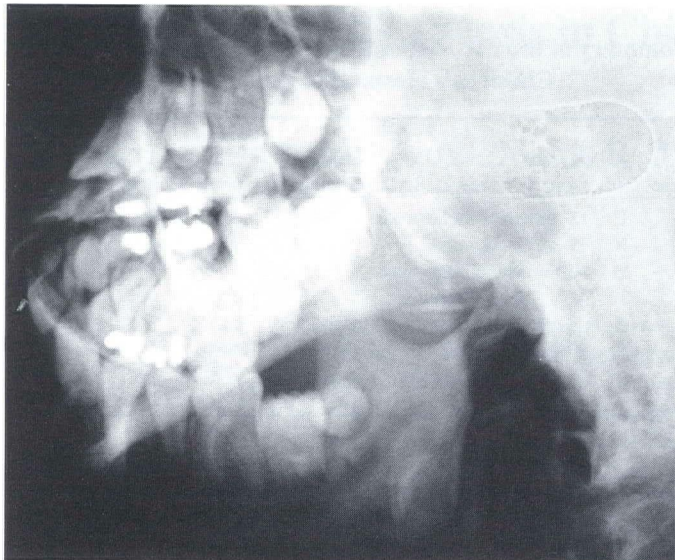


Fig 3—Mandible radiograph (Case 1, oblique view) showing soft tissue swelling and sclerosis of the right mandible.

Comments

This case report illustrates the difficulty in diagnosis of actinomycosis. Intermittent swelling of the mandible responded transiently to oral antibiotic therapy for nearly three years. The differential diagnosis of the expansile intramandibular mass in this child, which includes tumor, fibrous dysplasia, infection, and intrinsic bone diseases, requires biopsy for culture and histologic analysis.

The patient's course emphasizes an important point in the management of actinomycotic infection. Once diagnosis of actinomycosis is established and parenteral antibiotics are administered in high dose, exacerbation of symptoms is an indication for open debridement. Antibiotic therapy alone may fail to control infection in bone because avascularity limits attainment of effective antibiotic levels (9). Therefore, in the management of advanced actinomycotic infection, open debridement is essential.

Case 2

A 5-year-old girl presented to CHLA because of left-sided otalgia and fever for ten days which had progressed to swelling over the left side of her neck. Her temperature was 39°C (102.2°F), and she had a 12 cm, warm, fluctuant mass. Dental evaluation was negative. Incision and drainage of the abscess yielded 60 mL of pus which on periodic acid-Schiff stain was shown to contain variable filaments and sulfur granules. The child responded to high dose penicillin therapy given for three weeks. Cultures of the pus grew gamma-streptococci but were negative for *Actinomyces* species.

During the next eight years, the patient was hospitalized 11 times for recurrent abscess of the left side of her neck and had a total of 11 surgical drainage procedures. At each successive surgery of the abscess cavity, more extensive local invasion and destruction was demonstrated. Despite negative cultures, sulfur granules and gram-positive hyphal elements were found with every examination. During the tenth hospitalization, the *Actinomyces* was isolated and identified by gas chromatography. After the tenth drainage procedure, she was given chronic oral tetracycline therapy which suppressed the acute symptoms. Several months later, she failed to take her antibiotics for one week and a recurrent abscess developed. During the subsequent hospitalization, a pharyngocutaneous fistula was demonstrated between the left pyriform

sinus and the abscess cavity. Chronic oral tetracycline therapy was maintained and the fistula ultimately closed spontaneously.

Comments

This case report illustrates the prolonged morbidity that can occur with actinomycotic infection. This patient required 11 hospitalizations for surgical drainage and intravenous antibiotic therapy. Despite treatment with as much as 420 mU of penicillin during a single hospital stay, the inflammatory process continued to expand. The abscess eventually eroded into the pyriform sinus resulting in a pharyngocutaneous fistula. More aggressive surgical intervention with wide local debridement early in the disease might have shortened the duration of illness and prevented the subsequent fistula.

Case 3

This 14-month-old boy presented to CHLA with a three-month history of an inflammatory mass over the angle of his right mandible. He had fallen two months earlier and incurred a minor laceration of his mandibular gingiva. Initial therapy with antibiotics given orally resulted in minimal reduction in size of the mass, but the lesion ulcerated and began to drain.

Examination disclosed ulcerations from the condyle to the symphysis of the right mandible and smears of the drainage contained hyphal elements and sulfur granules. Despite treatment with high intravenous doses of penicillin for one week, drainage persisted and a submental abscess formed. Incision and drainage yielded negative cultures but the wet mount from surgery revealed hyphal elements.

During the next seven months progressive ulceration developed about the neck and face despite intravenous antibiotics in high doses. Repeat cultures failed to show actinomycosis but one culture grew *Klebsiella ozaenae*. Lack of improvement prompted hematologic evaluation with demonstration of a leukocyte abnormality. A tetralium dye test revealed that the patient's white blood cells failed to lyse bacteria. Unfortunately, during the course of the hematologic studies, sepsis developed and the patient died.

Comments

This child's course demonstrates the important relationship between host defense mechanisms and pathogenicity of ordinarily commensal oral bacteria. The actinomycotic infection advanced despite high dose antibiotic therapy and surgical debridement. Further evaluation demonstrated compromised host immunity, a leukocyte abnormality which undoubtedly contributed to the patient's rapid decompensation. Whether the actinomycosis altered the immune status or the spreading infection was a consequence of depressed host immunity cannot be stated.

Discussion

These case reports illustrate the protean clinical manifestations of cervicofacial actinomycosis in the pediatric patient. The most common presentation of actinomycosis is almost subclinical with the initial focus interpreted as "nonspecific" and treated with oral penicillin. If the antibiotic administration resolves the infection without the diagnosis being established, the true incidence of the disease cannot be determined (1). Many antibiotics, sulfonamides, aminoglycosides, macrolides, and tetracyclines may be effective against actinomycosis, but penicillin is the antibiotic of choice (9).

At times the presenting signs and symptoms may be confused with tumors of the jaw and surrounding soft tissues. Such was the case in case 1. Weese and Smith (8) reported that 23% of the

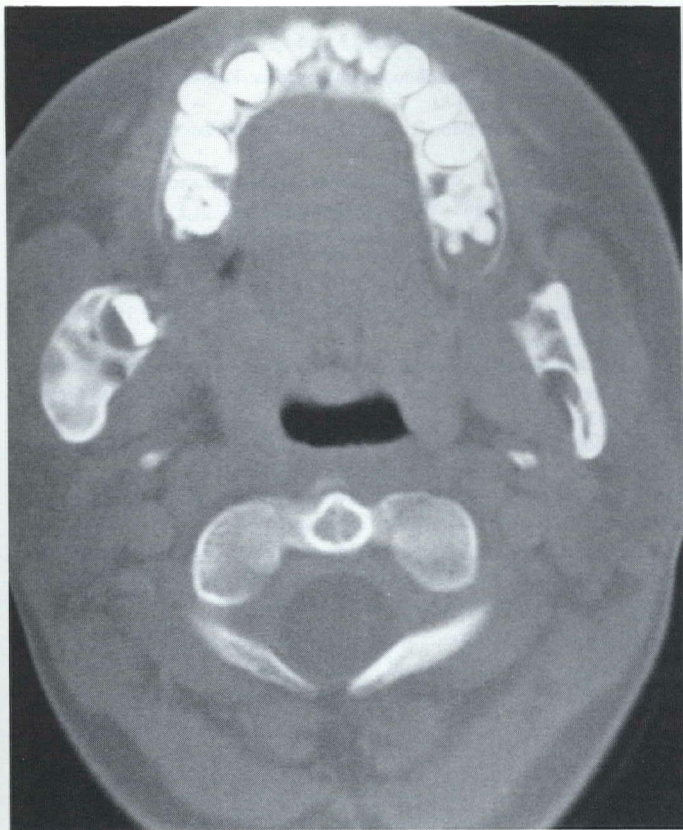


Fig 4A—CT (Case 1) demonstrates a calcified cyst in the angle of the right mandible.

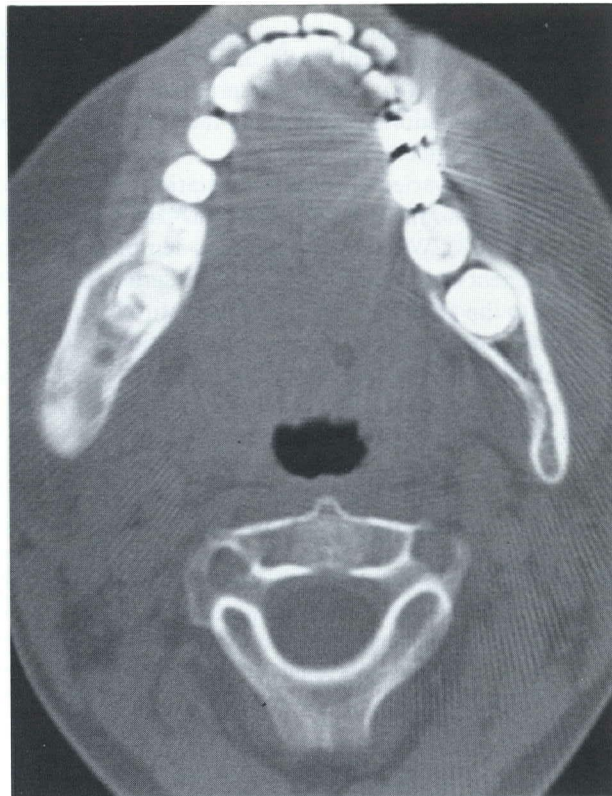


Fig 4B—CT (Case 1) demonstrates the expansile heterogeneous intramandibular mass of the right mandible.

patients with actinomycosis were admitted to the hospital with a presumptive diagnosis of cancer. The differential diagnosis also includes acute cervical adenitis with or without suppuration (cases 2 and 4), granulomatous lesions of bone, and, infrequently, congenital branchial cysts (case 5). In each of the cases presented, the protracted illness was characterized by periods of remission and exacerbation that were related to the use of antibiotics. These episodes should increase the suspicion of infection with actinomycosis (cases 1 and 2).

Actinomyces species were demonstrated in cultures from only two of our five patients, confirming the difficulty of isolating the bacteria. Data from the Armed Forces Institute of Pathology reviewed by Brown (6) disclosed that the causative organism was cultured in only 24% of the cases, with no growth found in 46% and polymicrobial flora reported in the remaining 30%. Sulfur granules, thought to be pathognomonic of actinomycotic infections, were present in three of our five patients. As Brown (6) has emphasized, the absence of sulfur granules does not rule out the diagnosis of actinomycosis. A positive culture is diagnostic of actinomycosis, whereas when the culture is negative sulfur granules or gram-positive hyphae must be looked for to establish the diagnosis. Certain cases may require an empiric trial of penicillin without precise bacteriologic diagnosis.

Factors that permit this usually commensal organism to invade and destroy soft tissue and bone are unknown. Our study confirms the polymicrobial nature of *Actinomyces* infection re-

ported by others (1). Multiple organisms were isolated by culture from each patient. The secondary pathogens are thought to facilitate the spread of actinomycosis by modifying tissue resistance. For example, streptococcal infection stimulates release of hyaluronidase and collagenase which impair tissue barriers and other host defense mechanisms. Because of the polymicrobial nature of the actinomycotic infection, antimicrobial therapy must be planned to cover both the primary as well as secondary pathogens (10).

Altered host immunity may play a role in the pathogenesis of actinomycosis. The patient in case 3, who developed progressive necrosis of the mandible and who eventually died of sepsis, was shown to have a phagocytic defect of the leukocytes. A report of a patient with the acquired immunodeficiency syndrome and actinomycosis suggests also that altered host defenses may facilitate invasion by actinomycotic organisms (11).

Cervicofacial actinomycosis is characterized by multiple recurrences and prolonged morbidity. Patients in our series had from two to 11 surgical procedures. Case 2 had 11 acute exacerbations of her actinomycosis during an eight-year period despite multiple high dose courses of penicillin and surgical debridement. These cases are the rule, not the exception, and indicate that treatment should include prolonged intravenous administration of penicillin in high dosage, as well as aggressive (but not deforming) surgical debridement. The presence of the sulfur granule may strengthen the case for aggressive surgical treat-

ment. Some suggest that the sulfur granule offers a protective shell to the organism, preventing the antibiotic from achieving adequate tissue levels (6). Local ischemia secondary to acute and chronic inflammation may also limit the ability of antibiotics to eradicate the infection.

In summary, the varied clinical presentations and difficult bacteriologic analysis make actinomycosis a challenge to the diagnostician (4,6,8). When the infection is demonstrated, aggressive treatment is recommended to prevent serious complications. Prior to the antibiotic era the only useful treatment for actinomycotic infection was surgery. With antibiotic therapy, surgical debridement may not be required. However, in this series of patients, aggressive surgical debridement along with high dose antibiotic therapy proved to be essential.

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