Pediatric Cervicofacial Actinomycosis: A Case Report

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ABSTRACT

Cervicofacial actinomycosis affects many soft tissue and bony structures in the head and neck, and has both granulomatous and suppurative features. Pathogenesis of actinomycosis is still unclear, but trauma provides a portal of entry for the infection. It usually presents as a diffuse swelling with multiple sinus tracts containing macroscopic colonies of the organism known as "sulphur granules." Cervicofacial actinomycosis in children is rare. This article reports a case of actinomycosis in a 10-year-old-boy overlying the left ramus of the mandible. (*J Dent Child.* 2004;71:87-90)

Keywords: Actinomycosis, Mandible, Child

ctinomycosis is an uncommon, chronic suppurative and granulomatous disease¹⁻³ caused by a filamentous bacterium.^{4,5} Actinomyces israelii is the main causative organism^{3,5-7} in man although other species, Actinomyces naeslundi, Actinomyces bovis, Actinomyces viscosus, Actinomyces odontolyticus, Arachnia propionica, and Bifido-bacterium dentium are occasionally isolated.^{1,2,6,8} Actinomycosis may be classified anatomically as: cervicofacial, pulmonary, or ileocaecal.^{1,7} Actinomycosis is a relatively infrequent infection in the cervicofacial region,4 despite actinomyces present in the normal oral flora.^{2,5} Cervicofacial actinomycosis is known to affect many soft tissues and bony structures in the head and neck region⁹ and usually presents as a persistent swelling with multiple sinuses.⁶ It is not clear how infection becomes established, but the bacteria must be inoculated deep into the tissues where the necessary anaerobic conditions prevail for growth of actinomycetes.⁴ Its portal of entry is usually through disruption of the mucosal barrier after trauma or dental manipulation.2,4,6-8,10,11

The presence of these bacteria as an oral commensal^{2,5} and the rarity of actinomycotic infection reflect the organism's low virulence or invasion.² The infrequency of actinomy-

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cotic infections indicates that inoculation at the time of oral trauma is not the only causative factor for infection.² Indeed, in some cases infection occurs after minor oral trauma, in others infection does not follow even after significant laceration of the maxillofacial area. Schwartz and Wilson reported 2 cases of cervicofacial actinomycosis following orthognathic surgery.¹² A review of published literature revealed a case of intramasseteric actinomycosis associated with needle tract inoculation.⁴

Actinomycosis, a pathologic condition that has both granulomatous and suppurative features, most often presents in 2 clinical forms.² One is chronic, slowly progressive with indurated infiltration and multiple abscesses and fistulas. The other is more acute and rapidly progressive, with fever and a sore, fluctuating swelling. The more commonly presenting form is the chronic, slowly extending one. The chronic form typically presents as a persistent and very firm swelling which is frequently dark red or purplish. As the lesion grows, multiple abscesses develop, breaking through the skin in several areas and causing a characteristic pattern of multiple sinuses. A yellowish purulent discharge from a sinus tract can usually be detected, as can the well-known sulphur granules.^{6,10,13} These are collections of microcolonies of the actinomycetes.7 A definitive diagnosis is usually based on the bacteriologic examination of the discharge.5,10,13

Actinomycosis is more commonly found in young healthy adults.² In the literature, the age range changes from 15 to 50. Most authors report a mean age of 40 years.¹¹ This disease very rarely affects children.^{2,14} The literature reports a predilection for males and a male:female ratio of 3:1 or 4:1.² The mandible is affected more frequently than the maxilla.²

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Figure 1. Panoramic radiograph of the patient demonstrates the absence of the dental focus for this severe infection.

Cervicofacial actinomycosis usually is localized in the submandibular area or around the angle of the jaw.^{3,11}

In this case, the authors report cervicofacial actinomycosis overlying the left ramus of the mandible in a 10-year-old-boy.

CASE REPORT

A 10-year-old-boy was referred to the authors' oral surgery clinic with the chief complaint of painful laceration and persistent swelling overlying the left ramus of the mandible. The swelling had been present for approximately 3 months. His parents stated that the child had a similar lesion 1 year ago at the same region. He had received IM clindamycin antibiotic therapy for 1 week. The lesion had disappeared and treatment stopped. There was no history of extraction of a tooth or dental trauma. The patient had a left submandibular lymphadenopathy, but otherwise was in good general health. The radiographic (Figure 1) and clinical examination revealed absence of a dental focus for this severe infection. All teeth were vital with no evidence of periapical pathosis. Oral hygiene was good.

Clinical examination revealed a firm, raised swelling involving the left masseteric muscle (Figure 2). There was a sinus tract accompanying purulent discharge (Figure 3), but no sulfur granules were found. CT scans revealed diffuse involvement of left ramus and angle of the mandible and a localized 5 cm \times 2 cm mass infiltrating the masseteric muscle (Figure 4). The patient was hospitalized. The purulent discharge from sinus was submitted for the bacteriologic examination and cultured for actinomyces species. Bacteriologic analysis was negative. Finally, an intramasseteric biopsy was taken from the most specific part of the lesion.

Based on histopathological findings, a definitive diagnosis of actinomycosis was made. The histopathological examination showed that the lesion consisted of a mass of granulomatous inflammatory response with a central area of suppurative necrosis. The center of necrosis contained a dense collection of fibrous tissue, with several wellformed colonies of filamentous micro-organisms resembling actinomyces (Figures 5A, 5B). The treatment protocol was focused on high doses of penicillin therapy parenterally until the lesion subsided. The patient was treated with IV penicillin G at doses of 10 million units daily for 12 days. When the swelling and purulent discharge disappeared, the patient was discharged and 800 mg IM penicillin was prescribed twice a day for 3 weeks. The treatment was continued by the administration of 1.2 million units of penicillin every 3 weeks for 18 months. The patient was followed

for 5 years, and no signs of recurrence were noted. The sinus tract healed without scarring (Figures 6A, 6B).



Figure 2. Pretreatment extraoral appearance of the patient with a serious swelling overlying the left ramus of the mandible.



Figure 3. Close view of the actinomycotic lesion with sinus tract.



Figure 4. CT scans revealed the diffuse involvement of left ramus and angle of the mandible and localized a mass infiltrating masseter muscle.

DISCUSSION

Therapy of actinomycosis includes surgical incision, drainage, and excision of all sinus tracts, combined with antibiotic therapy.^{2,10} Actinomycosis produces a massive fibrotic reaction in the involved lesion that surrounds the necrotic center.² This results in a hypovascular tissue with a low oxidoreductive potential and low penetration of drugs. Treatment should, therefore, be focused on surgical manipulations and high-dose long-term antibiotic regimen.^{2,10} A large number of different antibiotics have been proposed for the treatment of cervicofacial actinomycosis such as penicillin, erythromycin, tetracyline, lincomycin, minocycline, clindamycin, first-generation cephalosporins, ampicillin, and chloramphenicol.^{8,15}

Penicillin treatment remains the norm in dealing with actinomycosis in nonallergic patients. The drug of second choice is tetracyline, with minocyline or doxycyline being preferred in the allergic patient.¹⁵ Patients with severe infection or the bone of the jaws affected, should receive IV penicillin G at doses of 12 to 20 million units daily in 4 equal doses while the patient is hospitalized. IV treatment lasts until the disease is clinically cured.² The treatment should be followed by a long-term course of penicillin V at fractioned doses for a period of 3 months. The reason for the prolonged administration of the antibiotics is to prevent the recurrence of the infection.8,10 Actinomycosis can recur after months or years of apparent cure.10

It is, therefore, of primary importance to follow patients. The authors followed their patient for 5 years, and no recurrence was evident. In the present case, inaccurate diagnosis of the early lesion resulted in the low dose and short-term antibiotic treat-

ment that led to recurrence of the actinomycotic lesion.

Because the site of injection is highly fibrotic and avascular, some authors have recommended the use of hyperbaric oxygen in the treatment of cervicofacial actinomycosis.²

Clinical manifestations of the disease are confusing because they often mimic other disease processes.¹⁶ Diagnosis may be difficult due to a general lack of familiarity with the disease and the fastidious nature of the organism in culture.¹⁶ When clinical manifestations of actinomycosis are present and there is a history of persistent soft tissue infection accompanying sinus tract with exudate, actinomycosis should be included in differential diagnosis of neoplasms and chronic suppurative and granulomatous lesions of the head and neck region,¹⁷ even though no sulphur granules have been identified.⁴

An absolutely certain diagnosis requires isolation of the bacterium.¹⁸ Hence, the exudate must be cultured for actinomyces species to perform appropriate treatment.⁴



Figure 5A. Histopathologic section presenting actinomycotic colony between bone trabeculae (H&E ×40).



Figure 5B. Histopathologic pictures revealed several well–formed colonies of filamentous microorganisms with surrounding inflammatory infiltrate (H&E ×250).



Figures 6A and 6B. Posttreatment extraoral appearance of the patient without scarring.

However a thorough search of the literature reveals that negative cultures are found in 50% to 76% of the cases of actinomycosis in which a histologic diagnosis is established.¹⁸ Histologic diagnosis rests on the observation of sulphur granules (the mycelia-like microcolonies).

CONCLUSIONS

Bacteriologic analysis may often be negative. Therefore, histologic examination of a tissue biopsy is required for accurate and timely diagnosis of actinomycosis.¹⁸ In the present case, a certain diagnosis was made on histologic examination of the biopsy after the bacteriologic analysis was found negative.

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