An unusual form of Actinomycosis of the mandible with a resultant gross sequester in a 4-year-old child: a case report

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Mandibular osteomyelitis due to Actinomyces group is considered rare in the pediatric population. The initial complaint of the 4-year-old child described here was increased swelling of his cheek and pain. The patient was managed successfully by surgical treatment with antibiotic therapy.
J Clin Pediatr Dent 25(3): 237-239, 2001

INTRODUCTION

Actinomycosis is an uncommon chronic granulamatous infection caused by microorganism from the Actinomyces group, most often israelii and characterized by abscesses and sinus tract formation. The disease is especially seen in middle-aged men, whereas it is rare in children.[1] It can occur anywhere, but generally involves the cervicofacial, thoracic and abdominal regions. Although bone involvement is very rare, the disease more commonly involves the mandible.[1-4]

In this case report, we presented a rare case of actinomycotic osteomyelitis of the mandible of a 4-year-old child, whose clinical and radiological appearence was that of a non-specific osteomyelitis, and we aimed to discuss the etiology of the disease under the light of the literature review.

Case Report

A 4-year-old boy was referred to our department with a past history of tenderness on mastication, pain and slight swelling over the right mandibular deciduous molar region starting with a toothache four months earlier. A large sequester and hyperemic mucosa at its periphery were noted on clinical examination (Figure 1). Suppuration was also observed around the sequester.

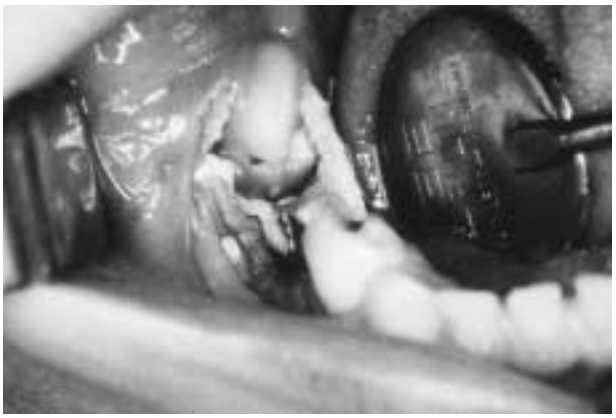

Figure 1. Preoperative clinical view of the case showing the sequester in posterior region of the mandible.

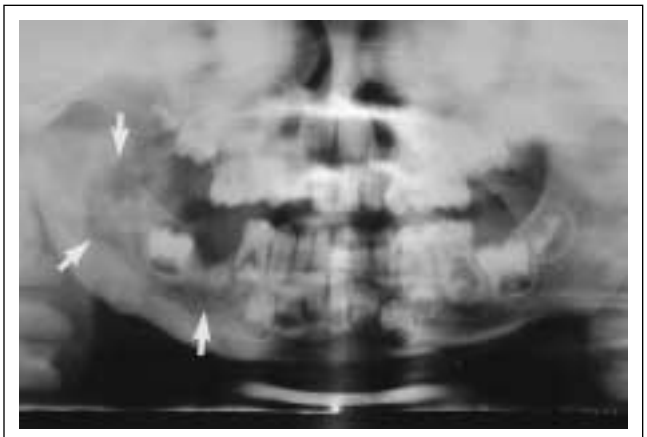

Figure 2. Preoperative panoramic radiograph of the case with radiolucent and sclerotic areas of the lesion (arrows).

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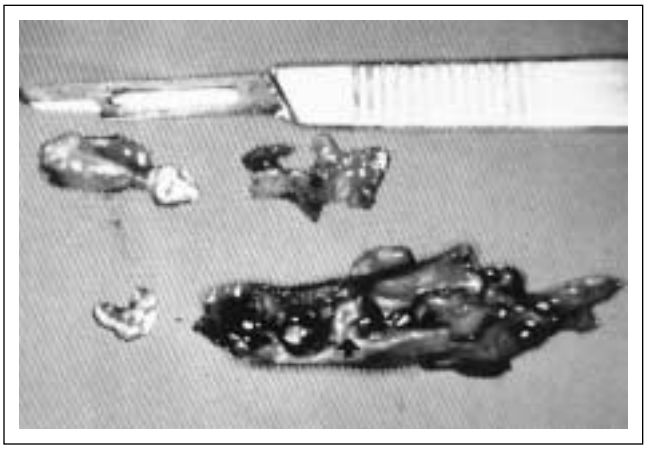

Figure 3. The large sequester together including the crown (arrow) of the first permanent molar within.

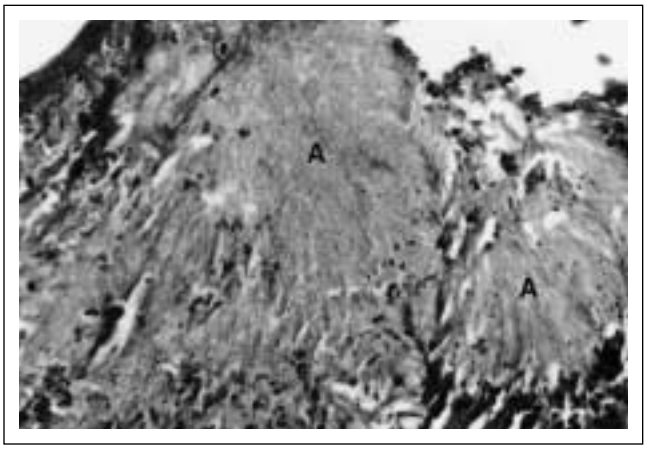

Figure 4. Photomicrograph of Actinomyces colonies (A) (Hematoxylin & Eosin stain, original magnification x 200).

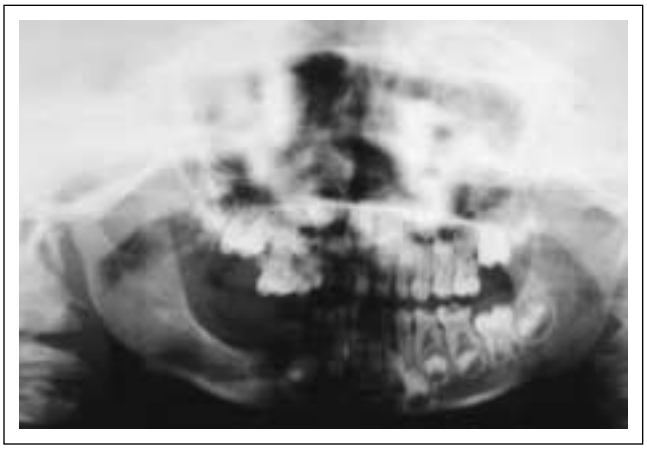

Figure 5. Postoperative panoramic radiograph (after 1 year) of the case showing signs of bone healing.

Radiological examination showed bony changes with osteolytic and osteosclerotic areas extending from the mandibular decidious first molar to midramus (Figure 2). There were no soft tissue involvement. The source of the infection was unknown.

Alpha hemolytic Streptococci were isolated from the pus. Previously, he had been treated with antibiotics on and off. The pain had disappeared after which the second deciduous molar tooth was spontaneously lost. It was stated that tiny fragments of bone were exfoliated from the area. 'Sulfur granules' were not noted at the time of examination. Treatment with ampicillin and sulbactam, 50mg/kg per day was initiated. Alpha hemolytic Streptococci, which were sensitive to ampicillin and sulbactam were isolated from the pus. Therefore, the patient was put on these antibiotics.

After the acute symptoms subsided, a large sequestre together with two small pieces of sequestra were removed surgically (Figure 3). Surgical decortication was not necessary. The surgical site healed with secondary intention. On histopathological examination, trabeculae of necrotic woven bone enclosing Actinomyces colonies with chronically inflamed bone marrow and a number of partly resorbed bony sequestra, nonspecific inflamatory cell infiltrates, vascular proliferations and granulation tissue were seen (Figure 4).

Actinomycosis of the mandible was diagnosed and the same antibiotic added with metronidasole, 25mg/kg per day was continued for a month.

The postoperative healing was uneventful (Figure 5) during the time which the patient has been followed-up for nine months. There was some subperiosteal new bone formation around the mandibular body on the right.

DISCUSSION

Occasionally anaeorobic pathogens can be etiological agents in oral infections. This is especially true with children because of their tendency to insert soiled fingers into their mouths and play with things which are often contaminated with anaeorobic pathogens such as Peptostreptococci.

Characteristically Actinomyces produce a subacute tissue reaction with connective tissue fibrosis and early diagnosis with vigorous antibiotic therapy may prevent sinus tract formation.[3] Absence of any draining sinus tract formation, although not diagnosed initially, may partly be explained by the use of potent antibiotics at different time intervals by the patient. Presence of sulfure granules are suggestive of Actinomyces infection and therefore require an anaeorobic culture for confirmation.[1,3,4]

However, in the case presented we failed to see any sulfur granules in the pus, with clinical and radiological features leading to the provisional diagnosis of nonspecific osteomyelitis, therefore anaeorobic culture did not seem to be absolutely necessary at the time. However, histopathological diagnosis of Actinomycotic osteomyelitis proved that anaerobic culture should be performed in every case which is characterized by pus formation.

Actinomycosis of bone is a rare disease. Actinomycotic inflamation of the mandible results from the presence of Actinomyceteceae resident in the oral cavity, especially when the normal composition of the microbial flora is disturbed and as a chronic infection it leads to localised pathological changes in bone. Contrary to non-specific bacterial inflamation of bone, the pathomechanism of Actinomycosis is unknown.

The typical clinical picture should be confirmed by bacteriological and histopathological examination, which are decisive for final diagnosis, but the results of both examinations are not always conclusive.[4] In our case, histopathological examination led to the diagnosis of Actinomycosis of the mandible. On the other hand, it was not clear whether the Actinomycosis was the primary infection or the secondary infection to a pre-existent non- specific osteomyelitis of the mandible. In the present case, the history of tooth infection in the related area support the idea that the latter possibility is more probable than the former.

Systemically antibiotic treatment is the basic form of therapy against Actinomycosis. Duration of treatment depends on the general condition of the patient and on the time of remission of pathological changes. Routine administration of antibiotics as recommended in the literature could be applied from 2 to 3 months.[1] Treatment with penicillin or tetracycline for about 6 weeks after radiological healing is usually required. Further therapy with metronidasole is recommended for 2-4 weeks to kill the associated flora.[4,5]

In our case, when the histopathological examination revealed an Actinomycotic infection, antibiotic treatment was continued. In keeping with the literature, the antiobiotic therapy was administerated from three weeks preoperatively until ten weeks postoperatively.

In conclusion, Actinomycosis of bone is a rare disease of vague pathomechamism usually affecting the mandible. Further study is required to understand if mandibular Actinomycosis is a primary disease process or a secondary infection to a primary non-specific osteomyelitis resulting in pathogen transformation of Actinomyces present in the flora. Diagnosis is based on clinical observation confirmed by bacteriological and/or histopathological studies. However the results of both examinations are ot always conclusive. The basic method of treatment is an intensive and prolonged therapy with antibiotics, especially with penicillin, and routine surgical procedures.

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