Pediatric Maxillary Osteomyelitis: A Case Report of a Rare Entity

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Maxillary osteomyelitis is a rare disease, especially in the pediatric population. We present a case of maxillary osteomyelitis in an eight- year- old girl with favorable outcome. Diagnosis was based on magnetic resonance imaging as well as on direct inspection intra operatively. Treatment should be based primarily on clinical signs (e.g. loose teeth). Teeth should not been extracted if healthy.

***Keywords**: maxillary osteomyelitis, loose teeth, children.*

INTRODUCTION

Osteomyelitis is defined as inflammation of the bone, beginning as an infection of the medullary cavity[1]. Typically, the Haversian systems become rapidly involved in the disease process as well as the surrounding periosteum. In the pre-antibiotic era, osteomyelitis was more common but today osteomyelitis of the jaws and especially of the maxilla is a rare disease and even rarer in children[2]. Well known contributing factors to osteomyelitis are diabetes mellitus, human immunodeficiency viral infection, and other immunocompromising diseases or therapies (e.g. chemotherapy or innate defects of the immune system)[3]. In clinical practice, osteomyelitis of the jaw is classified into three different forms: acute, primary chronic and secondary chronic inflammation[4]. We report a case of an acute maxillary osteomyelitis in an otherwise healthy eight year old girl.

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Case Report

An eight year old girl presented to the maxillofacial surgeon with erythematous swelling of the right cheek and lower lid accompanied by impaired vision of the affected eye. Several days before presentation, tooth 54 had been treated for deep caries with pulpectomy and dental filling. A thorough investigation revealed swelling of the oral vestibule and pus exiting from the gingival pockets of seven teeth. The girl was hospitalized with suspected diagnosis of abscess formation at the canine fossa. Inflammatory markers (C–reactive protein level (CRP) 131 mg/l, leukocytes 16.12 G/l) were significantly elevated. An antibiotic therapy with intravenous (i.v.) cefuroxime (Hikma, Terrugem, Portugal) was initiated and the affected tooth 54 was extracted. During this intervention, loosening of the teeth 16, 55 to 53, 12 and 11 was observed. Magnetic resonance imaging (MRI) of the head (Figure 1, marked with an arrow) showed diffuse perifocal uptake of contrast media in the maxilla as a sign of pronounced inflammation with local abscesses. Furthermore, MRI showed bilateral maxillary sinusitis. No bacterial growth of pathogens was found in intraoperative swabs. Histologic examination showed infiltration of neutrophils, macrophages and T-lymphocytes, thus confirming acute osteomyelitis of the affected jaw (Figure 2-5). Despite negative bacterial cultures, antibiotic therapy was continued because of histological and radiological signs of bone inflammation. Antibiotic therapy was chosen according to the published guidelines of the German Society of Pediatric Infectiology (DGPI), which suggests IV cefuroxime (Hikma, Terrugem, Portugal) as first line empirical therapy[5]. During IV cefuroxime therapy, regression of facial swelling and inflammation markers was seen. The remaining affected teeth did not show any more signs of loosening after 10 days and were not extracted.. After 14 days of IV antibiotic treatment, administration was switched to oral amoxicillin clavulanic acid (Hexal AG, Holzkirchen, Germany) for an overall duration of four weeks. During follow-up, no loose teeth were noted; the patient demonstrated no clinical signs of maxillary osteomyelitis and was under regular clinical follow-up. Subsequent biopsies showed restoration of the affected bone with remaining actinomycosis (Figure 6).

Figure 1: MRI scan of the maxilla shows a heavily inflamed bone and soft tissue with diffuse perifocal uptake of contrast media in the maxilla around the involved teeth (indicated by the arrow).

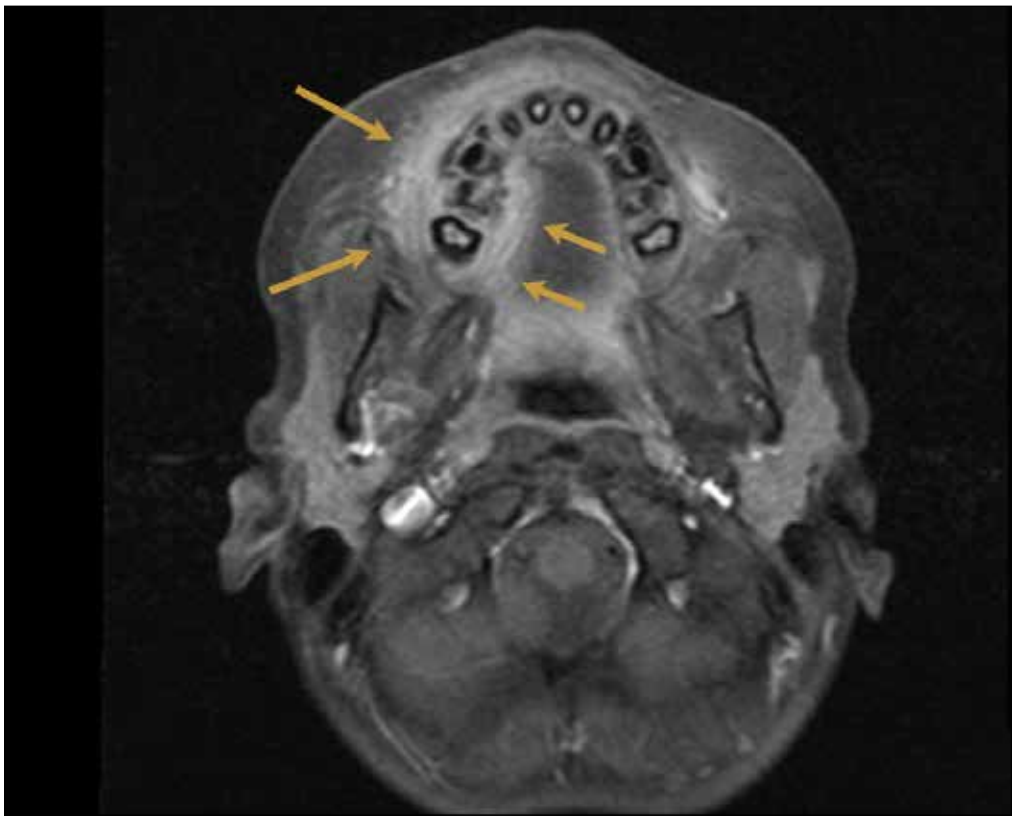


Figure 2: Hematoxylin and eosin stain (H&E stain) revealing granulation tissue (g) in bone marrow with predominantly granulocytes and destructed bone (b) (magnification 20x)

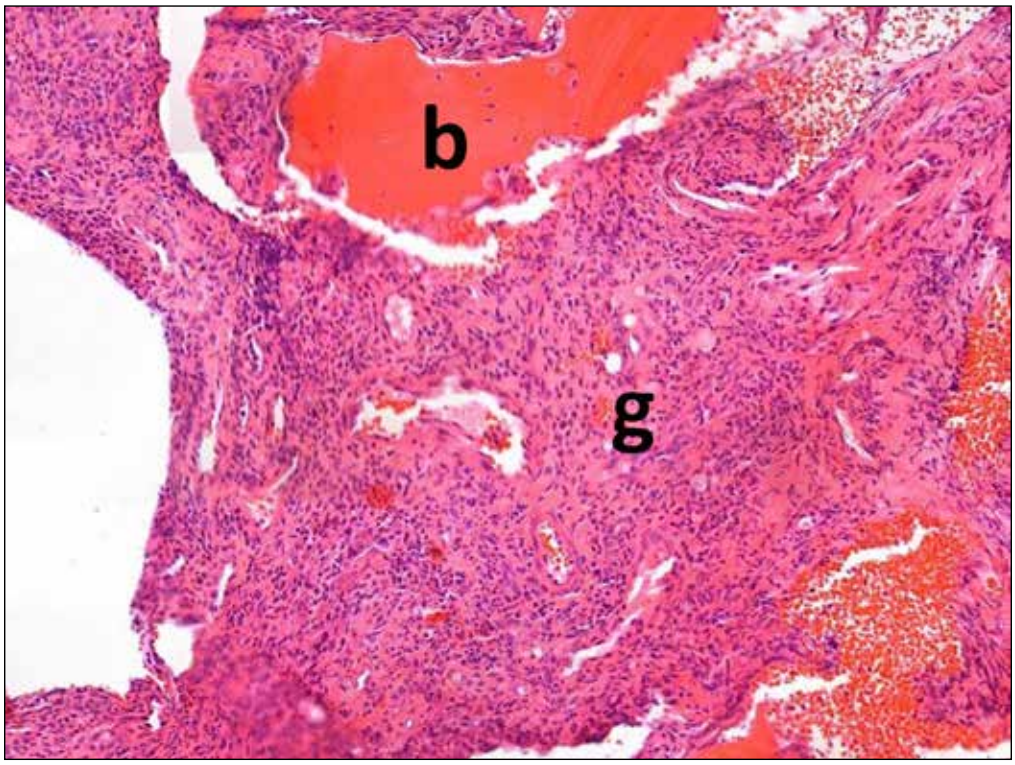


Figure 3: Immunohistochemistry revealing marked infiltrate of CD15 positive granulocytes (red structures) (ABC, alkaline phosphatase, new fuchsin stain method; magnification 40x)

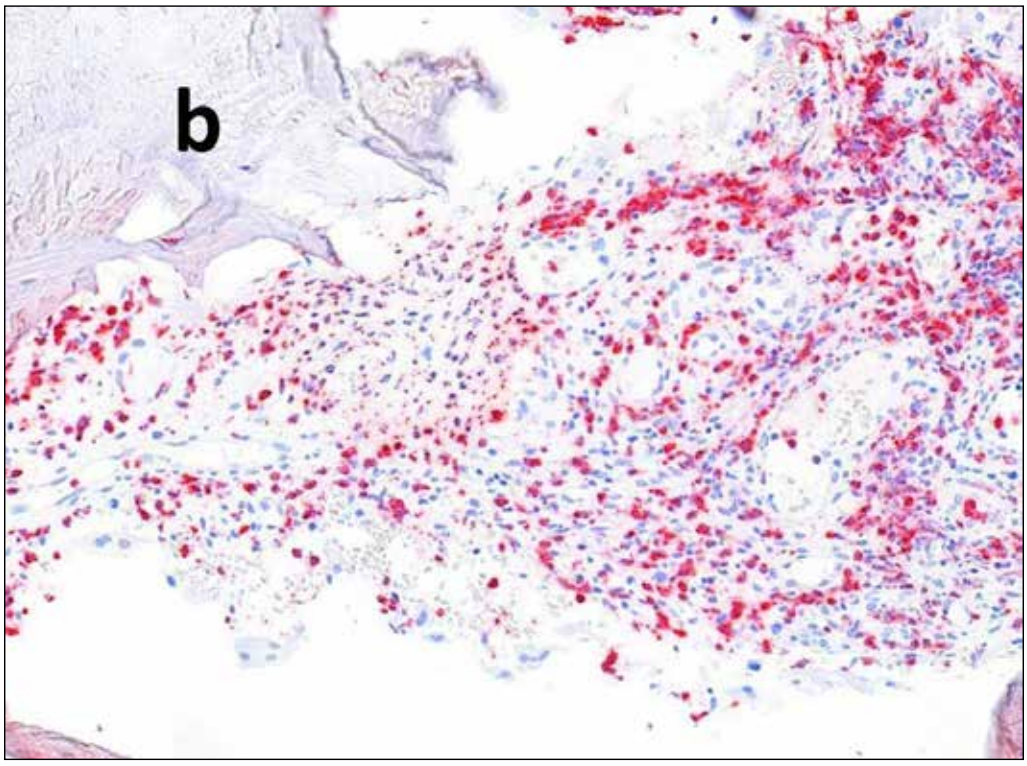


Figure 4: Immunohistochemistry showing moderate infiltrate of CD3 positive T- cells in the granulation tissue (in red) (ABC, alkaline phosphatase, new fuchsin stain method; magnification 40x)

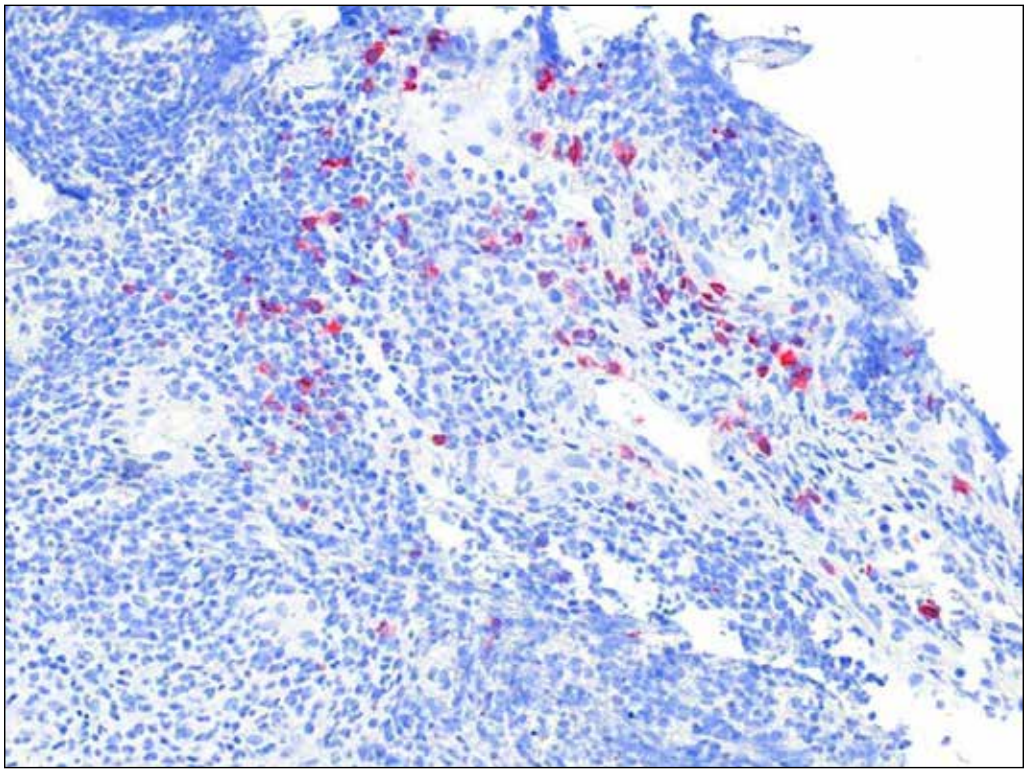


Figure 5: Immunohistochemistry indicating moderate infiltrate of CD68 positive macrophages in the granulation tissue (in red) (ABC, alkaline phosphatase, new fuchsin stain method; magnification 40x)

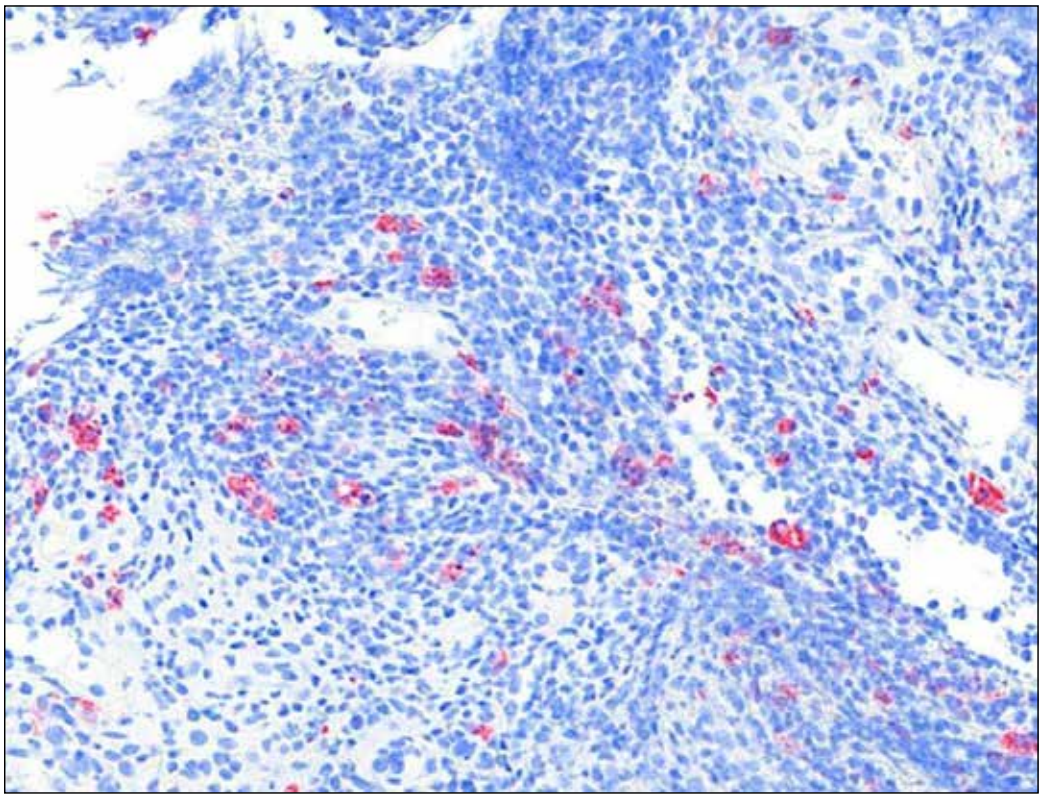
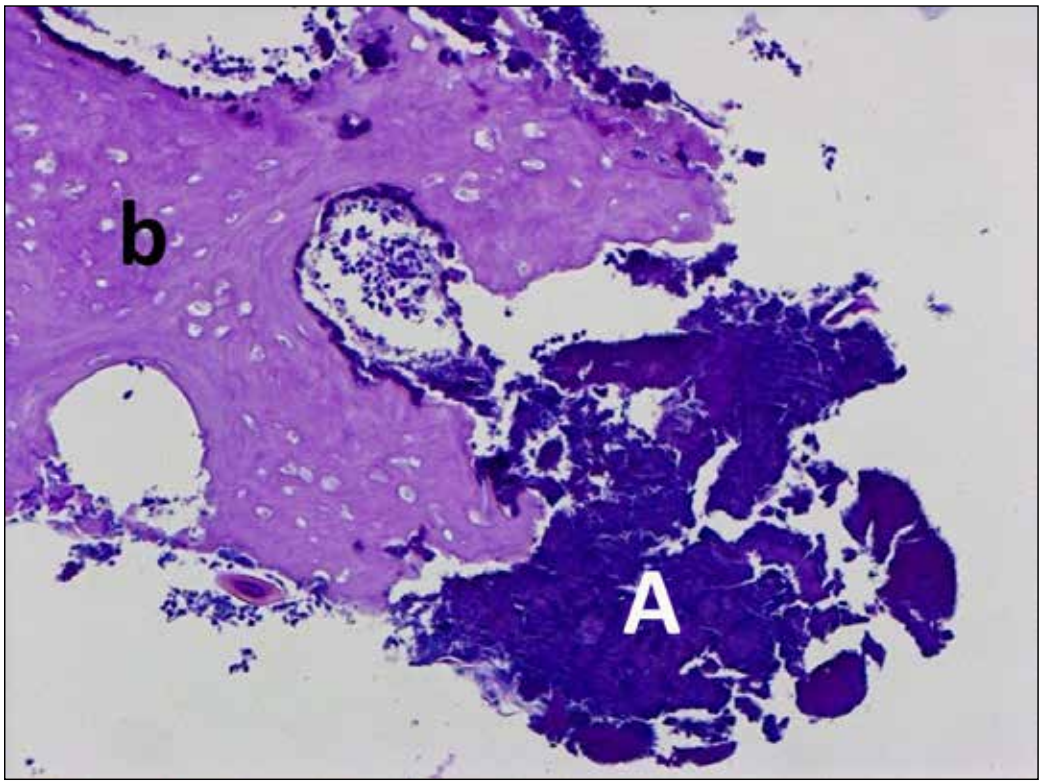


Figure 6: Follow up biopsy revealing actinomycosis (A) in vicinity of necrotic bone (b), (PAS stain; magnification 40x)



DISCUSSION

Osteomyelitis of the jaw and especially of the maxilla is a rarely–encountered entity in the post antibiotic era[6]. It is often accompanied by chronic conditions like diabetes mellitus or immunosuppression[7]. Interestingly, no underlying medical conditions were known in our patient. Further potential predisposing factors are dental infection, foreign materials and sinusitis[7]. In the present case, deep caries of tooth 54 was treated with dental filling several days before first clinical signs on the one hand, whereas on the other hand concomitant bilateral maxillary sinusitis as a probable cause was diagnosed on MRI. Antibiotic treatment is the mainstay of therapy. Further treatment modalities comprise removal of loose otherwise healthy teeth and sequestra, debridement and reconstruction[8, 9], which were not necessary in our case. Tooth extraction is a well-known risk factor for development of osteomyelitis of the jaw[10]. Actinomycosis of the bone, which was seen histologically in the subsequent biopsy was not treated separately with antibiotics, as the histologic finding was not associated with external draining ulcers, sinuses, fistulae and the patient remained asymptomatic. As previously published treatment of actinomycosis consists of administration of oral penicillin[11]. Our patient was treated with a second generation cephalosporine (cefuroxime) therefore no additional actinomycosis-directed antibiotic therapy was added[12]. Our case describes development of maxillary osteomyelitis without prior tooth extraction, but with pre-existing caries of a deciduous tooth, serving as the possible focus of the infection.

As described in the literature, even highly loosened (and otherwise healthy teeth) should be left to maintain space due to the high probability of stabilization after successful therapy[11]. We therefore decided against removal of the six loosened teeth. The empirical antibiotic regimen was chosen based on the expected spectrum of bacteria involved in the pathogenesis of maxillary osteomyelitis[8]. Duration of antibiotic therapy remains unclear. Only recently, length of antibiotic therapy from three to six weeks and early switch to oral therapy has been proposed[12]. Given the rarity of maxillary osteomyelitis in the pediatric population, no recommendations concerning optimal duration of antibiotic therapy can be given. In our patient, overall duration of therapy was four weeks with favorable outcome. In summary, maxillary osteomyelitis is rare in the pediatric population but should be kept in mind. Loosening of teeth is a clinically important clue to diagnosis. Finally, loose but otherwise healthy primary and permanent teeth should not be extracted, during and after conservative therapy of the underlying disease.

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