International Journal of Pediatric Otorhinolaryngology 75 (2011) 811–814

Contents lists available at ScienceDirect

International Journal of Pediatric Otorhinolaryngology

journal homepage: www.elsevier.com/locate/ijporl

Mandibular osteomyelitis in children mimicking juvenile recurrent parotitis

Riitta T. Saarinen [a,*], Kaija-Leena Kolho [d], Risto Kontio [b], Riste Saat [c], Eeva Salo [d], Anne Pitkäranta [a]

[a] Department of Otorhinolaryngology, Helsinki University Central Hospital, Helsinki University, Helsinki, Finland
[b] Department of Maxillofacial Surgery, Helsinki University Central Hospital, Helsinki University, Helsinki, Finland
[c] Department of Radiology, Helsinki University Central Hospital, Helsinki University, Helsinki, Finland
[d] Hospital of Children and Adolescents, Helsinki University Central Hospital, Helsinki University, Helsinki, Finland

ARTICLE INFO

Article history:
Received 26 November 2010
Received in revised form 14 March 2011
Accepted 15 March 2011
Available online 13 April 2011

Key words:
Pediatric
Actinomycosis
Hyperbaric oxygen therapy
Tooth follicle
Glandula parotis

ABSTRACT

Objective: To describe pediatric cases with mandibular osteomyelitis initially diagnosed and treated as juvenile recurrent parotitis.
Methods: We reviewed the patient data of all our pediatric patients treated at Helsinki University Central Hospital, a tertiary care hospital, between 1998 and 2010 who had the initial diagnosis of recurrent parotitis which in fact was osteomyelitis.
Results: Over a period of 12 years, six children (aged 5–17 years, five girls) presented with mandibular osteomyelitis primarily diagnosed as recurrent parotitis. Diagnostic delay ranged from 1.5 months to 6.0 years before the final diagnosis of mandibular osteomyelitis confirmed in MRI. Of the six cases undergoing biopsies, bacterial culture showed *Actinomyces* or *Streptococcus viridans* in four cases. All patients received antimicrobial treatment. Two received hyperbaric oxygen therapy with no resolution of symptoms. Debridement was performed in these two cases as well, and in the second case persistent symptoms led to bisphosphonate treatment.
Conclusions: Juvenile parotitis is in most cases a clinical diagnosis, and treatment is symptomatic. In contrast, mandibular osteomyelitis is a severe disease requiring lengthy treatment. Because symptoms of these two entities may mimic each other, unclear cases require MRI.

1. Introduction

Mandibular osteomyelitis during childhood and adolescence has been addressed in a limited number of papers [1]. This inflammatory process involving cortical and cancellous bone is classically divided into acute, subacute and primary chronic forms [2]. Juvenile recurrent parotitis, on the other hand, is a recurring inflammation of the parotid gland of unknown etiology. The symptoms of these two entities may mimic each other: recurrent swelling and pain in the mandibular area, difficulties in mastication and occasional fever.

The diagnosis of primary parotitis in pediatric patients is based on clinical examination, and in most cases no imaging or laboratory investigations are necessary [3]. However, if the symptoms recur, other pathologies should be excluded. Ultrasound is an applicable tool for differential diagnosis [4]. The prognosis of juvenile recurrent parotitis is excellent, and the disease seldom persists into adulthood [3].

Conversely, osteomyelitis in general is a severe disease necessitating intensive treatment. It may be associated with local infection or trauma or be secondary to vascular insufficiency or be of hematogenous origin [5]. The long bones are most commonly affected, and in some cases osteomyelitis may be multifocal. The annual incidence of osteomyelitis in children and adolescents varies between 1:5000 and 1:10 000, with a male predominance [6,7]. In the pediatric population, osteomyelitis is most often of hematogenous origin traditionally treated with long-term antibiotic therapy and in many cases with debridement of the infected area. Some evidence, however, exists of the equal efficacy of shorter antibiotic treatment and a more conservative surgical approach in children [8,9]. Methods such as plain radiograph, CT, MRI, and nuclear imaging can serve in the diagnostic work-up. Of these, plain radiograph is most widely available, but in the early phase of the disease has the lowest sensitivity.

The adjuvant treatment options for osteomyelitis include hyperbaric oxygen therapy (HBO). Higher levels of available oxygen have many beneficial effects in diseased tissue: i.e. induction of capillary formation, increased activity of leucocytes and a suppressive effect on anaerobic bacteria [10,11]. Bisphosphonate treatment as an adjunct therapy for chronic osteomyelitis has recently shown promising results in cases resistant to conventional therapy [12,13].

* Corresponding author at: Department of Otorhinolaryngology, Helsinki University Central Hospital, POB 220, Haartmaninkatu 4E, 00029 HUS, Finland. Tel.: +358 50 4271 496.
E-mail address: riitta.t.saarinen@hus.fi (R.T. Saarinen).

0165-5876/$ – see front matter © 2011 Elsevier Ireland Ltd. All rights reserved.
doi:10.1016/j.ijporl.2011.03.013

Table 1
Characteristics of the six children treated at our institution due to osteomyelitis first diagnosed as recurrent juvenile parotitis.

Patient	Age/ gender	Duration of symptoms before/after diagnosis m = month, y = year	Bacteriology	Tooth removal/ depridement	Hyperbaric oxygen therapy Imaging	Antibiotic therapy after diagnosis of osteomelitis m = month	Follow-up/outcome
1	10/F	12 m/24 m	Negative cultures	No/no	No	Ciprofloxacin + rifampicin 2 m, ciprofloxacin 3 m	6 years/cured
2	11/F	10 m/25 m	Negative cultures	d38/yes	20 treatments	Clindamycin + cephalexin 3 m, rifampicin + levofloxacin 2 m	5 years/cured
3	13/M	1.5 m/4 y	*Actinomyces*, *Enterococcus faecalis*, *Candida albicans*	d38/yes	15 treatments, 10 treatments	Penicillin 3 m, amoxicillin + flukonazole 4 m, meropenem 1 m	4 years/symptoms persist
4	11/F	18 m/3 m	*Actinomyces turicencis*, *Fuco-bacterium*, *Streptococcus viridans*	d38/no	No	Penicillin 3 m	10 months/symptom-free, opening of the mouth mildly reduced
5	5/F	2.5 y/6 m	*Actinomyces*, *Fusobacterium*, *Streptococcus viridans*	No/no	No	Penicillin 3 m	6 months/symptom-free
6	17/F	6 y/5 m	*Actinomyces* type of bacteria, *Propionibacterium acnes*, *Streptococcus viridans* and *anginosus*	d38/no	No	Amoxicillin 40 days	24 months/cured

We reviewed patient data during the last decade at our tertiary care hospital and traced six children with mandibular osteomyelitis which was initially diagnosed as juvenile recurrent parotitis. These findings emphasize the importance of differential diagnosis between these two entities, and the importance of MRI in cases of persistent and severe symptoms in the parotid gland region.

2. Methods

The data base of Helsinki University Central Hospital Department of Otorhinolaryngology, according to the classification of Diseases 10th Revision (ICD-10) provided data on otorhinolaryngological patients aged 17 or under, to find all cases of regional osteomyelitis. Diagnostic codes for osteomyelitis and parotid illnesses were both included. Our hospital is a tertiary care hospital with a source population representing 28% of the total child population in Finland. These patient data were reviewed. In addition to the six children included in the study, one similar patient had been referred for consultation. However, since his full patient history was unavailable, he was excluded.

The study was approved by our institutional local ethics committee.

3. Results

Over a period of 12 years (1998–2009), six children (aged 5–17 years, five girls) presenting with mandibular osteomyelitis initially diagnosed as recurrent parotitis were observed in the Helsinki University Central Hospital Department of Otorhinolaryngology. The cases are presented in chronological order in Table 1. The data base surge with osteomyelitis as a primary diagnosis provided negative results.

All children presented with acute onset of symptoms, after which they had recurrent swellings and pain in the parotid region. An ultrasound investigation in all cases during the primary phase produced negative findings. All children had received short courses of antibiotics and NSAIDs for their recurrent symptoms. All children used good dental hygiene, and their general health was excellent. Due to persistent symptoms, all children underwent MRI that led to the correct diagnosis. The MRI revealed edema and chronic changes in the bone structure typical for osteomyelitis. In patient number 2 an MRI in the early phase was misinterpreted. In one case (patient 6) a CT was also performed but provided no additional information. In addition, ortopantomography was performed in every case. Typical radiological findings in MRI and ortopantomography are shown in Figs. 1 and 2 respectively.

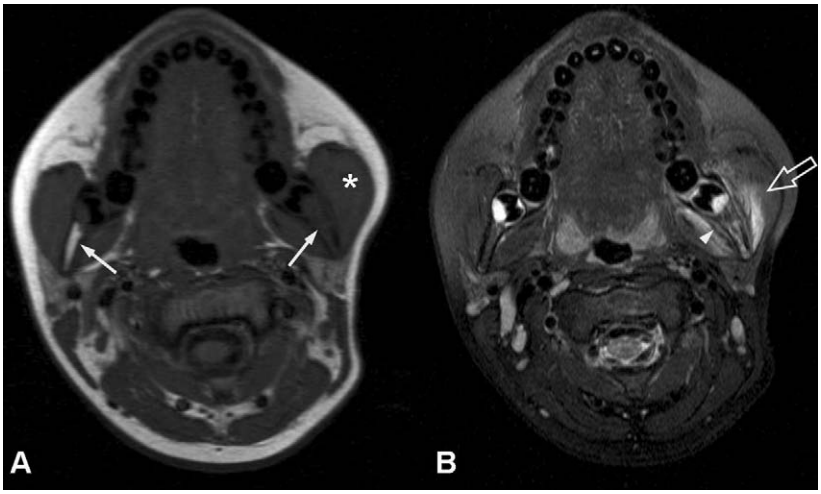

Fig. 1. Axial T1 (A) and axial T2 fs (B) MRI images showing osteomyelitis of the mandible on the left side. Marked edema and replacement of fatty bone marrow are visible as signal loss on the T1 image (A: right arrow), and a signal increase on the T2 fs image compared to the normal right side of the mandible (A: left arrow). The periosteum of mandibular angle is thickened (B: arrowhead). The masseter muscle on the diseased side (A: asterisk) is enlarged, and with inflammatory edema in the masticator muscles visible as a diffuse signal gain in a T2 fs image centered on the mandible (B: open arrow).