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Chronic nonbacterial osteomyelitis involving the mandible: A case report☆

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ABSTRACT

INTRODUCTION: Chronic nonbacterial osteomyelitis (CNO) or chronic recurrent multifocal osteomyelitis (CRMO), is a very rare condition of unknown aetiology. It is characterised by focal sterile inflammatory disease with prolonged, self-limiting and recurrent episodes.
CASE PRESENTATION: We report the discovery of this very rare disease following a mandibular abscess in a 10-year-old female. We initially focus on the difference between the preoperative orthopantomography and the maxillofacial computed tomography and magnetic resonance images obtained, and then on the improvement of strategies for correct diagnosis and treatment of this disease.
DISCUSSION: Bone pain and localised swelling can occur in a single bone or can spread to soft tissue and adjacent bone; areas commonly affected by CMRO include the metaphyseal plates of the long bones, as well as the spine, clavicle and, rarely, the maxillofacial area. The clinical presentation of CMRO includes pain, functional impairment, and swelling, similar to our case.
CONCLUSIONS: We report a very rare case of this unifocal mandibular disease in a child who presented for an abscess and was then diagnosed and treated for CNO.

1. Introduction

Chronic recurrent multifocal osteomyelitis (CMRO), or chronic nonbacterial osteomyelitis (CNO), is a very rare idiopathic non-infectious inflammatory disorder, characterised by bone lesions with pain and swelling, and periods of exacerbations and improvement in different locations over the course of several months to years [1,2]. CRMO primarily affects children, with a female-to-male ratio of 4:1, and the mean age at onset is 10 years [3,4]. The diagnosis of this disorder is based on clinical, radiological, and histological features [5].

Relatively few cases of CMRO have been reported since the first description in 1972 by Giedion, who described the disease as an unusual form of multifocal bone lesions with subacute and chronic symmetrical osteomyelitis [6]. This was followed by Bjorksten's report in 1978, which coined the term 'chronic recurrent multifocal osteomyelitis'. The term CRMO is used to described the multifocal form of CNO [7].

CNO is poorly characterised in the maxillofacial surgery literature due to the use of inconsistent terminology [8].

Many authors agree that chronic osteomyelitis involving the jawbone may be divided into two major categories: suppurative and non-suppurative. We use the Marx classification [24] to emphasise the difference between CNO and other lesions.

Mandibular lesions are found in 1.5–3% of disease foci in patients with CRMO [9] and the bone most commonly affected by unifocal disease is the mandible [10].

Here, we describe a case of this very rare disease, which was discovered during a frequently-occurring mandibular abscess in a 10-year-old female.

2. Case report

A 10-year-old girl with a recurrent mandibular abscess was referred to the Outpatient Unit of the Department of Maxillofacial Paediatric Surgery, Children's Hospital, ASST degli Spedali Civili, Brescia, Italy. Physical examination revealed only left mandibular swelling and pain with trismus (Fig. 1). The parents revealed that this was the third presentation of the abscess within the last year. Vitaly tooth tests were positive and the percussion test negative. Blood examination revealed only that the C-reactive protein (CRP)

☆ The English in this document has been checked by at least two professional editors, both native speakers of English. For a certificate, please see: http://www.textcheck.com/certificate/MEDbQ3.

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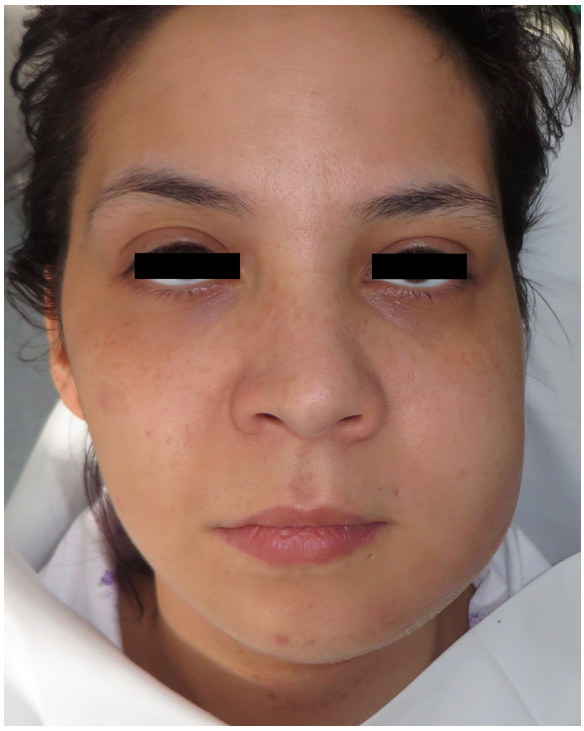

Fig. 1. Clinical view of the patient.

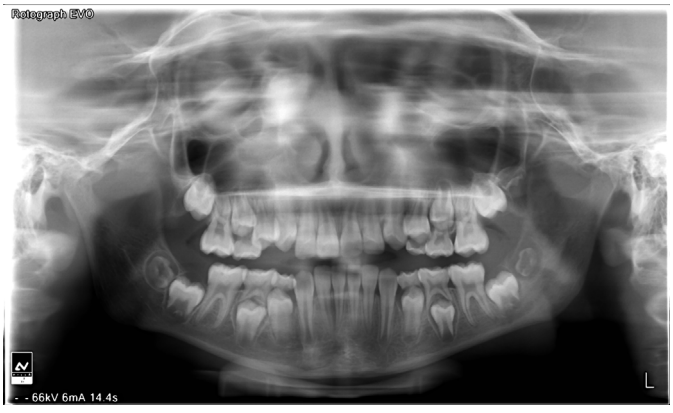

Fig. 2. The preoperative orthopantomogram.

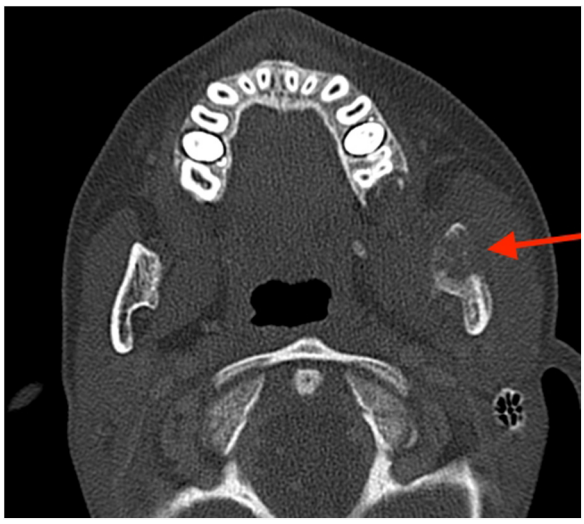

Fig. 3. The CT axial view of the mandibular lesion.

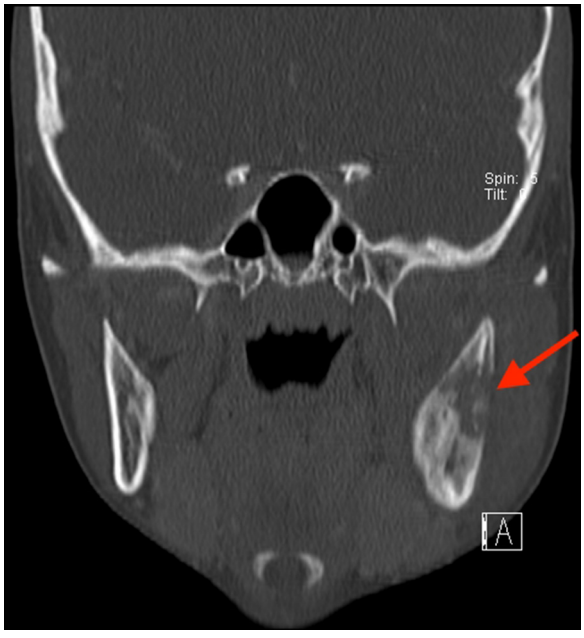

Fig. 4. The CT coronal view of the mandibular lesion.

level was elevated to >30 g/L; the blood count was normal and she was in good general health.

Orthopantomography revealed mixed dentition with nothing of note in the mandible or maxilla (Fig. 2). The patient had no history of trauma, previous medication, gingival bleeding, or caries.

To treat the abscess, the patient was immediately started on intravenous antibiotic treatment with rocefin 1 g per day and metronidazole 500 mg every 6 h.

As, clinically, abscess neoformation was evident, we decided to commence broad-spectrum antibiotic therapy [25].

One day later, it was decided to perform a contrast-enhanced computed tomography (CT) maxillofacial scan to better understand the causes of this recurrent problem.

The CT showed remarkable results. The left masseter muscle was diffusely swollen by oedematous imbibition and was also associated with nuanced imbibition of the surrounding subcutaneous soft tissue, down to the muscle itself and in contact with the mandibular angle bone surface. There was a significantly thin, soft, hypodense component, with a thickness of about 5–6 mm and an anteroposterior extension of approximately 2 cm. The seat of the cortical bone of the left mandibular branch was interrupted (maximum 1 cm extension) and the structure of the underlying cancellous bone appeared to be particularly inhomogeneous (2 cm craniocaudal extension) and showed the presence of multiple areas of osteolysis (Figs. 3–5).

Two days later, on the suspicion of mandibular osteomyelitis, the patient underwent contrast-enhanced maxillofacial magnetic resonance imaging (MRI), which revealed diffuse structural alteration of the left vertical mandibular branch and horizontal branch in the molar region (Figs. 6–8). The alteration consisted of:

- an irregularity of the cortical focal profile with continuation into the cortical branch of the vertical branch on both sides;
- widespread alteration of the cancellous bone signal, characterised by an intermediate T2 signal with moderate impregnation (the alteration also extended to the coronoid);