Localized osteodysplasia of the mandible in a seven year old child: a case report

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ABSTRACT: Aim This report concerns the most recent knowledge on osteodysplasies, anomalies in the development of the bone tissue, that may concern one or more skeleton segments affecting children. Case report The case of a 7 year old patient is reported, whose left hemimandible showed a radio transparency with a size of 10x7 mm. After the usual preliminary diagnostic investigations, it was decided to carry out an ablation. The neoplastic formation was completely enucleated using a conservative operation technique.

Keywords: Osteodysplasia, Mandible, Child

Introduction
Osteodysplasies are anomalies in the development of the bone tissue concerning one or more skeletal segments. They depend on the alteration of the osteogenic mesenchyma (connective tissue responsible for the building of the osseous tissue) [Valletta, 1997]. They include different kinds of osteopathies, each one characterised by an alteration of the bone, that turns into osteofibrous tissue, that is a fibrous component, the same as in each osteodysplastic form, that replaces normal bone [Valletta, 1997]. Each one of these forms can be recognized through a typical pathological appearance, while the fibrous component is not a typical element, because it must be considered a foreign reaction of the bone tissue to different, partly non-identified, aetiological agents [Valletta, 1997]. The most frequently observed forms are: Paget’s disease, Recklinghausen’s disease, Jaffè-Lichtenstein osteofibrous dysplasia, Albright syndrome, Cherubism and osteofibrous cement lesions.

Case report
In July 1998 a 7 year old male patient, A.R., attended the Department of Paedodontics (Department of Head and Neck, II University of Naples). Until that time the patient’s dentist had been treating him for dental caries and chronic gingivitis. The referral was because an examination of an orthopantomogram indicated a bean shaped radiotransparency. The largest diameter was 10 mm, while the smallest was about 7 mm, situated in the left hemimandible (Fig. 1). The lesion was almost completely asymptomatic. An oral examination showed caries present only in tooth 75. The medical history of the patient indicated that, beyond the usual childhood disease, he had not suffered from serious diseases or other injuries to the region under investigation.

The family history was non-contributory. Further questioning of the patient recorded no disturbances commented on by the boy, except for a sensation of trouble to the teeth in the left hemimandible. Local examination did not show any deformation of the mandibular bone structure or a mobility of the dental elements contiguous to the lesion. There was no history of any toothache either spontaneous or provoked.

Further diagnostic procedures. After the examination of the orthopantomogram it was decided to take a dental scan. This enabled an identification of the relationship of the lesion to the nearest anatomical structures (Fig. 2). After the evaluation of all radiographic examinations, laboratory analyses as well as other tests were completed (ECG, urine analysis, chest film etc.). As the patient was in good health a surgical approach to treatment was decided upon.

Treatment. Tooth 75 was removed, as its roots were involved in the lesion. The operation consisted of a mucoperiosteal incision starting from the third left inferior primary tooth to homolateral permanent tooth. After the elevation of the flap, a bone incision was
made with bone nippers to reach the lesion, that had partly eroded the mandibular cortex. After exposure, the lesion was entirely enucleated leaving a large cavity (Fig. 3). The excised lesion was sent for the histological examination.

Histology. The histological report showed fragments of fibrovascular connective tissue with small areas of chronic inflammation and bone spikules with an adherent flap of osseous tissue having the characteristics of fibrous dysplasia (Fig. 4).

Follow-up care. Four months postoperatively full skeleton radiography was carried out in agreement with the patient’s paediatrician, who had advised against carrying out a total body scintography. This examination did not show any bone alteration in the examined segments. The patient is, however, monitored every 6-8 months and this will be continued in the future. At the 5 months postoperative visit an orthopantomogram showed bone healing (Fig. 5a). A 5 years postoperative follow-up indicated that healing was complete and eruption of the permanent teeth had occurred (Fig. 5b).

Discussion

The varied forms of mandible located osteodysplasia may cause different patterns according to their localisation and their extension. In the initial phase...

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**Fig. 1** - Initial orthopantomogram showing a bean shaped radio transparent area with well defined margins, at level of tooth 36.

**Fig. 2** - a) CT scan showing an ellipsoid shaped lesion, with the largest diameter being about 1 cm. b) Interruption of the external cortex associated with an implication of the spongy bone around the roots of teeth 36 and 75.

**Fig. 3** - Intraoral picture showing cavity in bone after enucleation of the lesion.

**Fig. 4** - Histological picture showing fragments of fibrovascular connective tissue with small areas of chronic inflammation and bone spikules with an adherent flap of osseous tissue having the characteristics of fibrous dysplasia.
they often run silently. At other times, by contrast, they appear with specific pathological symptoms ranging from asthenia, anorexia, slimming to bradycardia, vomiting and abdominal pains [Guarini et al., 2000]. Each osteodysplastic form shows the same pathological symptoms characterised by local pain, that is often joined by sensory problems (such as paraesthesia, hypoaesthesia etc.) according to their localisation [Malandrini et al., 1996]. Deformations of involved bone segments may occur later on, until fractures occur that may be spontaneous but minor, depending on the extent of any injuries [Guarini et al., 2000].

The differential diagnosis involved must consider a number of alternative conditions. Von Recklinghausen’s disease is usually treated with an operation, which solves the primary cause, depending on the parathyroid glands or on kidneys [Baluta et al., 1996]. Medical treatment is only prescribed to patients presenting heavy contraindications to an operation and the most effective therapy is the use of oral monocacid phosphates. Once the dose has been established, the intake must continue uninterrupted. In the event that calcemia is above 14-15 mg/dl, an emergency therapy with use of isotonic NaCl solution at 0.9% is needed to rebalance the dehydration. Phosphates, calcitonin or diphosphate may be used as well in order to reduce the hypercalcemia. The prognosis of Jaffè-Lichtenstein disease is favourable, because its development stops with the end of growth. Treatment is usually surgical, while radiation therapy is not advisable because it could induce a sarcomatous transformation [Rabukhina et al., 1996]. Treatment of osteofibrous cement lesions varies according to the different forms; the cement osseoperiapical form does not require any treatment, while focal and florid osseocement forms require surgical treatment consisting of curettage in small fragments [Dellestable et al., 1996].

On the basis of this report it is possible to conclude that when osteodysplastic bone segments are present, the right therapy depends on careful diagnostic screening. In this present case the first examination was radiographic, but at the time of the examination it was thought that a dental scan would be very helpful. This allowed a conservative surgical technique. This approach was born out as, after only 2 weeks, the mucous injury had already healed up. The patient enjoys the best of health at present and follow-up radiographic assessment shows physiological bone healing as well as a normal development of secondary dentition.

**Conclusion**

The treatment of this described case of localised osteodysplasia of the mandible in a child was successful, due to a conservative surgical approach after full radiographic and diagnostic tests assessment.

**References**


