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A rare mandibular neoplasm: case report of a Central Giant Cell Granuloma

Mandible can be affected by a great variety of neoformations, like aneurysmal bone cyst, odontogenic myxoma, CGCG (Central Giant Cell Granuloma), GCT (giant cell tumor), sarcoma, ameloblastoma, lymphoma, ossifyng fibroma, odontogenic mixoma, granuloma, arteriovenous malformations and Schwannoma. Occasionally is not possible to find clinical or radiological distinctive findings so is usefull to perform additional exams, think about rare disease and perform an explorative surgical treatment which can be adapted to the intraoperatory findings. This attitude may help to reduce overtreatment but also to be radical especially in case of rare condition like the case presented: a Central Giant Cell Granuloma of the jaws. In this case report the authors present a 19-year-old female with a slowly enlarging, painfull swelling on the left side of the lower jaw. Ortopantomography exam revealed an osteolytic bone formation confirmed by Tomographic Dental Scan, MRI and Eco-Doppler exam. No one of these procedures, however, allowed to characterize the neoformation. For that reason was planned immediately an explorative surgical treatment, instead of an agosbiopsy. Macroscopic free margins resection provided radicality on one side and saved much bone tissue as possible on the other; morever it would have permitted to be more demolitive with a further procedure if the histopathological examination of specimen didn't show complete neoformation removal.

KEY WORDS: Central Giant Cell Granuloma, Rare Mandibular Neoplasm, Explorative Surgical Treatment

Introduction

Central giant cell granuloma (CGCG) rapresents a localized aggressive benign osteolytic proliferation [1]. As regards etiology, several hypothesis have been made: Jaffe considered this tumor to be a locally reparative reaction of bone to inflammation, local trauma, or hemorrhage [2]; Hillerup and Hjørting-Hansen proposed "vascular mishap resulting from trauma, primary bone disease, or malformation, as the cause of disease [3]. It usually affects younger patients under the age of 30 years (80% of the patients being under the age of 20), with a clear female predilection (62%). It accounts for <7% of all benign tumors of the jaws; the mandible being more frequently affected than the maxilla (it occurs especially in the body region, anterior to the first molars), with a relative proportion ranging from 2:1 to 11:9. Clinically, may behave variably, exhibiting characteristics ranging from asymptomatic, indolent, and slow growth to aggressive and rapid hollowing out of bone, with cortical expansion, thinning and perforation, root resorption, displacement of adjacent structures including teeth and nerves, accompanied by pain. Actually diagnosis is based on CT scan examination demostrating a multilocular honeycomb or soap bubble-like lesion. At the histological examination CGCG shows destructive osteolytic lesion consisting of fibrous tissue with hemorrhage and hemosiderin deposits and presence of osteoclast-like giant cells with reactive bone formation [4]. Treatment options depends on the clinical characteristics and behavior and range from surgical excision or resection with continuity defect, cryotherapy, to enucleation and aggressive local curettage with or without chemical cauterization [5]. Early diagno-

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sis and treatment produce a better outcome and prognosis is good when complete removal is achieved. It is associated with a relatively high recurrence rate of 15%–20%; the more aggressive the lesion, the higher the chances of its recurrence [1].

Case Report

A 19-year-old female was brought to observation complaining a slowly enlarging, painfull swelling on the left side of the lower jaw. On intraoral examination, a small spherical swelling with no visible pulsations and normal skin overlying was noted in the left ramus of the mandible. An ortopantomography exam (Fig. 1) revealed an osteolytic bone formation which determined mandible cortical bone depletion. Tomographic Dental Scan confirmed the osteolytic erosion (Fig. 2). These clinical and radiological findings had not cleare and unique interpretation, so additional exams were perfomed: MRI showed evidence of a 7mm × 11mm well-defined lesion with cortical thinning and cortical breaches (Fig. 3), ultrasoud examination documented a hipoecogenic tumefaction and Eco-Doppler exam which showed a sclerotic lesion. Surgical excission was therefore scheduled under general anesthesia and mandible was exposed by a submandibular approach. On the subperiostal plan, the tumor mass was found under the mandibular border and appeared as a multilocular lesion solid with bone and soft tissue and in continuity with alveolar nerve without infiltrating it. (Figs. 4,5). Macroscopic free margins resection was performed including neoplasm, soft and bone tissue but saving alveolar nerve using piezosurgery and surgical specimen was sent for histopathological examination which revealed a central giant cell granuloma with negative margins. No post operative complications occurred. Follow up included postoperative MRI.

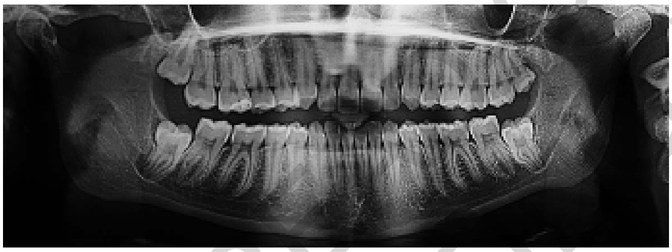

Fig 1: Ortopantomography exam.

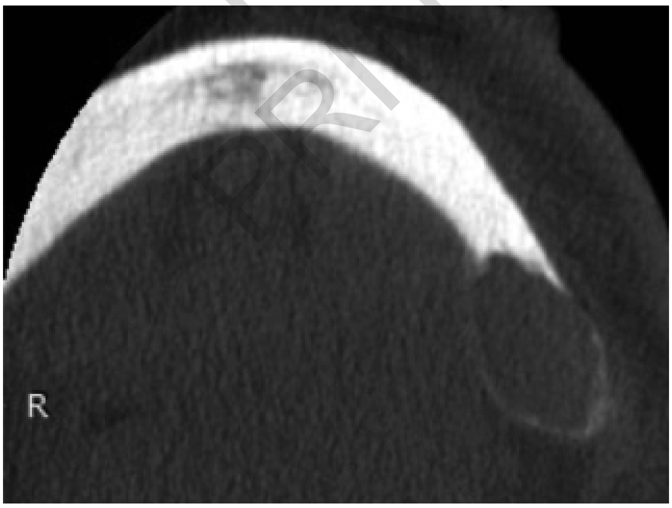

Fig 2: CT Dental Scan.

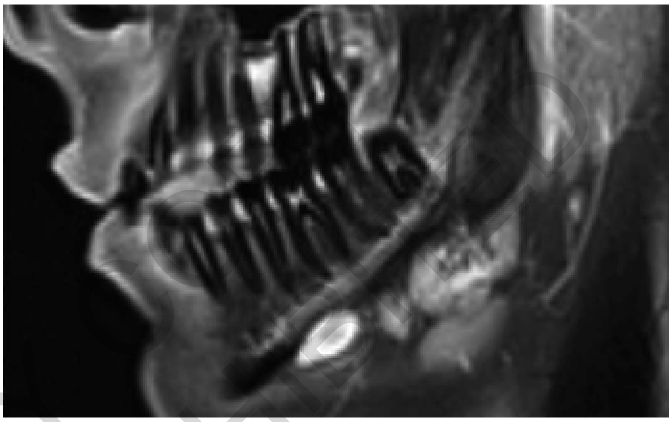

Fig 3: Magnetic Resonance.

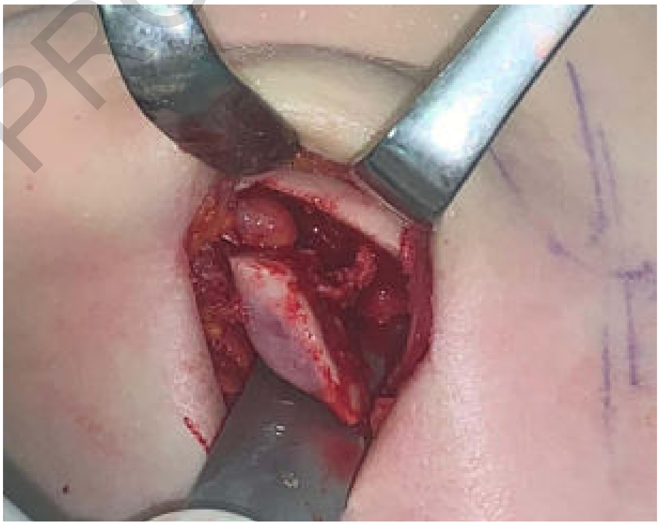

Fig 4: intraoperative picture showing inferior alveolar nerve inside the lesion.

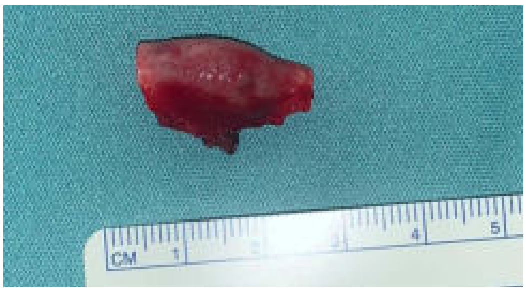

Fig 5: surgical specimen.

Discussion

Mandible can be affected by a great variety of neoformations like aneurysmal bone cyst, odontogenic myxoma, CGCG, GCT (giant cell tumor), sarcoma, ameloblastoma, lymphoma, ossifyng fibroma, odontogenic mixoma, granuloma, arteriovenous malformations and Schwannoma mostly with typical clinical and radiological presentation and when that happens, both diagnosis and treatment are based on CT scan. In the situation presented both intra/extra-oral exam and CT Dental Scan didn't allow to characterize the neoformation (it was impossible to descriminate if the mass was depending by bone, nerve or soft tissue, allowing to make diagosis of ameloblastoma, Schwannoma or sarcoma for example). In particular, CT Dental Scan scan showed deep relation between tumor and the inferior alveolar nerve canal so Schwannoma was considered as a possible diagnosis [6,7] and, moreover, addressed the problem to save the nerve [8,9]. In cases like this, when is not possible to find clinical or radiological distinctive findings, the authors suggest to perform additional exams (as ultrasoud examination, MRI or agobiopsy) and think about rare disease. Moreover, in consideration of dimension, consistency and radiological features, it's usefull to plan immediately an explorative surgical treatment, instead of an agosbiopsy. The finaly definition of the lesion nature is possible only by histopathological examination which showed, in the case presented, an unexpected diagnosis: a Central Giant Cell Granuloma with free margins of resection. Considering CGCG high recurrence rate and the patient age, it has been important, then, to perform an explorative surgical treatment which has been adapted to the intraoperatory findings. In this situation, macroscopic free margins resection provided radicality on one side and saved much bone tissue as possible on the other; morever it would have permitted to be more demolitive with a further procedure if the histopathological specimen examination didn't show complete neoformation removal. This attitude may help to reduce overtreatment but also to be radical.

Riassunto

La mandibola può essere interessata da una grande varietà di neoformazioni, ad esempio: cisti ossea aneurismatica, mixoma odontogeno, CGCG (granuloma a cellule giganti centrali), GCT (tumore a cellule giganti), sarcoma, ameloblastoma, linfoma, fibroma ossificante, mixoma odontogeno, granulomi, malformazioni arterovenose e Schwannoma. Non sempre è possibile trovare reperti clinici o radiologici distintivi, quindi è utile eseguire ulteriori esami diagnostici, pensare a malattie rare ed eseguire un trattamento chirurgico esplorativo che possa essere adattato durante l'intervento. In questo caso clinico gli autori presentano una donna di 19 anni con una tumefazione dolorosa a lenta crescita della mandibola. L'esame ortopantomografico ha rivelato, successivamente, una lesione osteolitica confermata dalla TC Dental Scan, dalla risonanza magnetica e dall'esame Eco-Doppler. Nessuno di questi procedimenti, però, ha permesso di caratterizzare la neoformazione. Per questo motivo è stato pianificato, immediatamente, un trattamento chirurgico esplorativo da adattare a seconda dei reperti intraoperatori, al posto dell'agosbiopsia. Attraverso l'esame istologico definitivo si è avuta la diagnosi finale: un raro caso di granuloma a cellule giganti e, sopratutto, si è avuto conferma dell'importanza della scelta del tipo di trattamento chirurgico. Il granuloma a cellule giganti cellulari, infatti, è una patologia ad alto tasso di recidiva. Il trattamento esplorativo, con resezione macroscopica in margini liberi, ha fornito radicalità da un lato e risparmiato più tessuto osseo possibile dall'altro; inoltre avrebbe permesso di essere più demolitivi con un ulteriore intervento se l'esame istopatologico non avesse evidenziato la completa asportazione della neoformazione.

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