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Giant Osteochondroma in the Coronoid Process Leading to Severe Limitation of Mouth Opening: A Case Report

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Abstract

The objective of this article is to present a case report of a 37-year-old female patient affected by an osteochondroma in the region of left coronoid process, causing severe trismus (approximately 15-mm opening) continuously evolution for about two years, without pain complaints. The patient was submitted to removal of the mass in the coronoid process by an intra-oral access under general anaesthesia. Anatomopathological examination showed evidence of an osteochondroma. The patient is under post-operative follow-up, presenting significant improvement of the mouth opening with no signs of relapse.

Keywords: Osteochondroma; coronoid process; trismus, mandible

Introduction

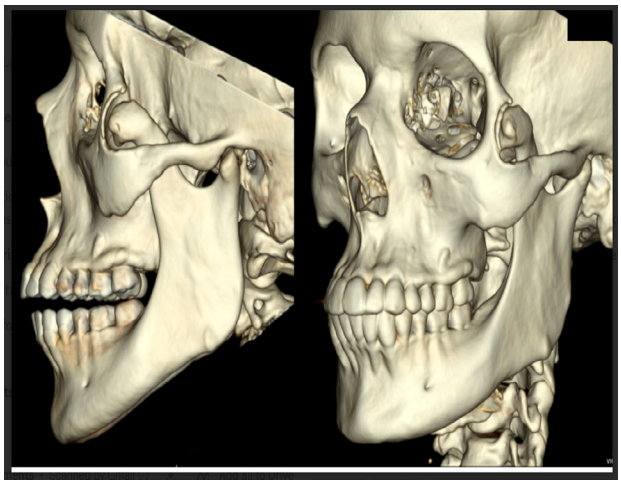
Osteochondroma is one of the most common tumours affecting the cortical bones, generally characterized by slow growth and presence of a prominent layer of cartilaginous tissue. This tumour is very commonly found in long bones, such as epiphyseal and meta-epiphyseal joints, proximal tibial meta-epiphyses, and distal femoral meta-epiphyses [1]. The condyle is the most affected site in the maxillary region, although osteochondroma has already been reported in other regions, such as skull base, maxillary sinus, zygomatic arch, and mandibular body and symphysis [2]. This type of lesion is more likely to occur in the condyle region, coronoid process and symphysis due to its ossification of embryological origin (i.e. endochondral) [3].

The origin of this type of tumour is not fully clarified, mainly due to the fact that there is no cartilaginous tissue in the region of the coronoid process. According to Lichtentein (1952) [4], the origin of osteochondromas comes from the pluripotent potential of the periosteum to undergo a process of metaplasia, thus generating chondroblasts and chondrocytes. When an osteochondroma reaches the region of the coronoid process and then develops into the region of the temporal fossa, it can lead to the formation of a pseudo-joint between the enlarged coronoid process and zygomatic arch, giving rise to the so-called Jacob disease [5]. The objective of this article is to present a case report of a patient affected by a giant osteochondroma in the region of the coronoid process characterised as Jacob disease, which was effectively treated with intra-oral excision of the lesion.

Case Report

A 37-year-old female patient presented to the Department of Oral and Maxillofacial Surgery with a chief complaint of progressively worsening limitation of mouth opening over a period of approximately two years. Initially, the maximal mouth opening was approximately 25 mm, and the patient was referred for conservative management, including the use of interocclusal splints, physical therapy, and pharmacological treatment. Despite these therapeutic interventions, the mouth opening progressively deteriorated over time. On clinical examination, before the surgical procedure, the patient had a maximum mouth opening of 15 mm, but with no complaint of pain. Nuclear magnetic resonance and computed tomography were requested for the patient, showing that the joint had not been affected. However, the presence of an extensive mass in the region of the left coronoid process, measuring approximately 40 mm in length and extending into the temporal fossa, resulted in bone resorption of the zygomatic bone region, with formation of a pseudoarthrosis between the coronoid process mass and the zygomatic arch, thereby limiting mandibular movement, a characteristic finding of Jacob's disease. (Figure 1).

Figure 1: *CT Image.*



The treatment planning involved complete excision of the coronoid process mass by an intra-oral access. Because the patient had significant trismus, intubation was performed by using a bronchoscopy and surgical access through the vestibular sulcus so that osteotomy at the base of the coronoid process could be performed with a piezo-surgical device. Displacement of the temporal muscle insertion was performed and the tumour completely removed (Figure 2). The surgical specimen was sent for histopathological analysis, being characterised as an osteochondroma with the presence of an osteocartilaginous

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lesion covered by a fibrocartilaginous cap composed predominantly of hyaline cartilage with organized endochondral ossification. The cartilaginous component showed chondrocytes arranged in clusters within a chondroid matrix, without significant atypia or mitotic activity. The underlying osseous component consisted of mature trabecular bone with fibrovascular marrow spaces. Continuity between the cortical and medullary bone and the lesion was identified, supporting the diagnosis of osteochondroma. No evidence of malignancy was observed. Condylar hyperplasia was considered unlikely due to the presence of a well-defined osteocartilaginous proliferation with a cartilage cap and tumor-like growth pattern, findings that are not characteristic of condylar hyperplasia (Figure 3).

Figure 2: Surgical specimen.

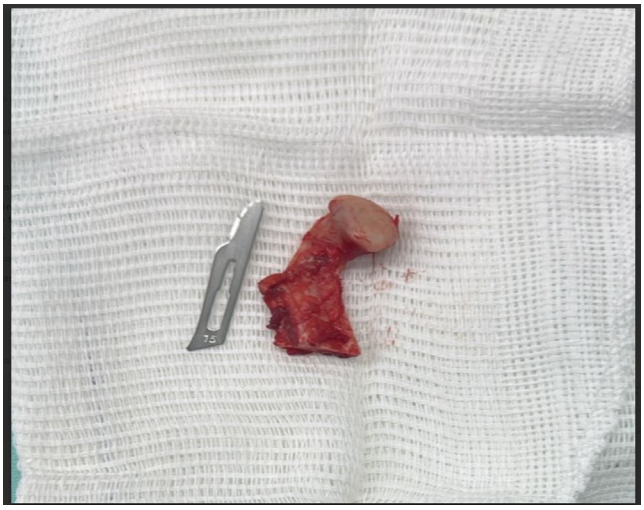
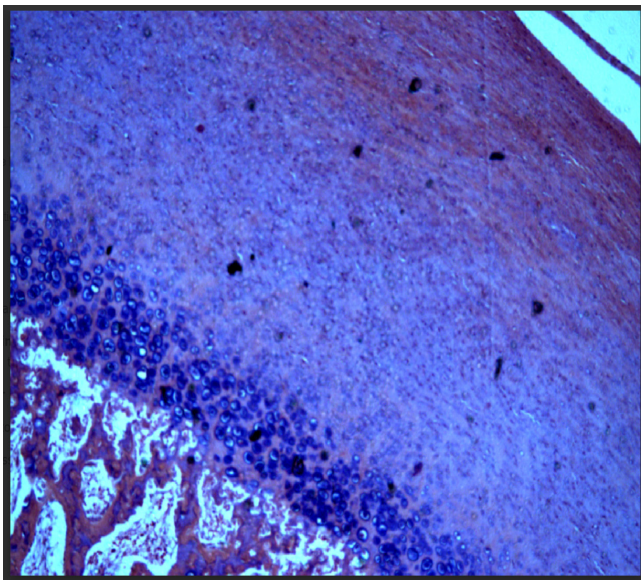


Figure 3: Histological section stained with H&E, magnification 100x.



Postoperatively, the patient underwent physiotherapeutic follow-up consisting of active exercises for mouth opening, protrusion, and lateral excursion, combined with manual temporomandibular joint (TMJ) mobilization and myofascial release of the masticatory muscles, with two sessions per week over a two-month period. At the 10-month postoperative follow-up, the patient demonstrated satisfactory mouth opening (approximately 30 mm), with no clinical signs of relapse.

Discussion

The development of an osteochondroma in the region of the coronoid process is something rare. The most recent literature review showed only 39 cases with histological evidence compared to the 50 cases reported elsewhere [6]. Computed tomography is the gold standard examination for diagnosis of this type of pathological alteration, as magnetic resonance normally cannot cover this region of the coronoid process in the mandible. The possibility of diagnosis by panoramic radiography has been cited by some studies, but because of image overlap in this region resulting in local osseous duplication, some diagnostic difficulty may be encountered [7].

This type of tumour requires surgical treatment, with coronoidectomy being recommend due to its low rate of relapse (i.e. 2%) [8]. The access for this procedure can be obtained either intra or extra-orally. The intraoral approach eliminates the risk of extraoral scarring and facial nerve injury. Nonetheless, herniation of the buccal fat pad may occur, and depending on the size of the lesion, its removal may not be feasible via the temporal fossa [9]. Extra-oral approaches are the other forms of access described in the literature, such as Akayat-Bramly [10], bi-coronal [11] and combinations of intra- and extra-oral accesses [9] all having disadvantages as described earlier.

This case differs from those previously reported in the literature due to the unusually large size of the osteochondroma and its successful removal through an intraoral approach, thereby avoiding potential facial scarring and injury to adjacent facial structures that could occur with an extraoral surgical access. In conclusion, patients presenting progressive limitation of the mouth opening should be always evaluated for the possible presence of an osteochondroma in the region of coronoid process. Exams such as computed tomography, mainly in patients with important trismus, should always be required for evaluation of temporomandibular disorder, to not delaying the diagnosis and lead to ineffective treatments due to a possible misunderstanding about intra-articular problems.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest. None of the authors received any funding for this research.

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Informed consent statement/ Ethics committee approval

According to Brazilian National Health Council Resolution No. 510/2016, approval by a Research Ethics Committee was not required for case reports. Nevertheless, the patient provided written informed consent authorizing the publication and disclosure of the case for educational and research purposes.

Credit author statement: Fábio Ricardo Loureiro Sato: Conceptualization, Writing- Original draft preparation; **José Simão de Oliveira:** Conceptualization, Resources; **Celso Luiz Ferraz:** Resources; **Fernando Vagner Araldi:** Writing - Original Draft; **Michelle Bianchi de Moraes:** Writing - Original Draft; **Gabriel Cortez da Silva Toledo**: Histopathological analysis.

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