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CASE REPORT

Recurrent ameloblastoma 30 years after surgical treatment

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Abstract:

Introduction: Ameloblastoma is a locally invasive, slowly growing odontogenic epithelium, which can reach large proportions. It is preferentially located in the region of mandibular molars. The most indicated treatment is resection with safety margin, since its potential for recurrence is high. **Objective:** To report a case of recurrence the ameloblastoma. **Case report:** A 60-year-old female patient came to the hospital with difficulty speaking and eating because of the mass inside the mouth and earaches. The extra-buccal physical examination had an increase in volume in the jugal and right temporal regions. At the intra-buccal physical examination there was a nodule approximately 5X6X7 cm, firm at palpation with normal overlying mucosa in color and continuity. The patient reported that 30 years ago she had undergone a surgical procedure to remove an ameloblastoma in the region of the right mandibular body. Imaging revealed a wide extension of the lesion, extending from the symphysis to the cranial base, in the middle cranial fossa region. Previous incisive biopsy revealed the diagnosis of follicular ameloblastoma. **Conclusion:** Ameloblastoma should be followed for a long period of time because it is a locally infiltrative benign neoplasm that, even when treated with surgery and safety margin, may recur late. The approach taken was the removal of the lesion associated with mandibular reconstruction.

Keywords: Ameloblastoma; Jaw Neoplasms; Mandibular Reconstruction; Odontogenic Tumors.

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INTRODUCTION

Ameloblastoma is an odontogenic tumour classified as benign, despite having an aggressive behaviour that resembles malignancies. It presents clinical characteristics of fast and invasive growth, more commonly affects the mandibular posterior region, and appears uni- or multilocular in radiographic images, which is described as soap bubble or honeycomb[1]. A recurrence rate is most reported in the literature after conservative treatment[2-8]. Adequate treatment is defined considering the clinical and imaging characteristics, which demonstrate if the lesion is more or less aggressive[9].

An article by Laborde et al.[7] reported a series of cases of patients with ameloblastoma from the oral and maxillofacial surgery department of the Lille Hospital between 1991 and 2013. It had a total of 31 cases, where 27 cases were included in the study, the age, location, type of treatment, if any Conservative or radical. They concluded that the earlier the intervention, the lower the chance of recurrence were, and that when they adopted a radical treatment, the relapses decreased significantly.

In the literature, there are reports of recurrence in the iliac bone after 20 years, showing the potential for recurrence of this pathology[10].

A literature review performed by Henriques et al.[11] reported the capacity of the ameloblastoma to present late recurrences. Diagnosis is reached histopathologically; according to the WHO classification, the basic subtypes are the follicular and plexiform types[12]. The recommended treatment is surgical removal, which can be performed conservatively or radically, depending upon the case to decrease the rates of relapse.

CASE DESCRIPTION AND RESULTS

A 60-year-old female patient, sought care with food difficulty, reported having undergone surgery 30 years ago for the removal of ameloblastoma. Clinically, the patient presented with a volumetric increase in the temporal region and the right side of the face, causing an asymmetry in the region of the base of the mandible direct side we can observe the scar due to surgery 30 years ago. Intra-buccally, a nodule of approximately 5 x 6 x 7 cm could be observed. It was firm on palpation, with fibrous and vascularized consistency (Figure 1).

In the panoramic image (Figure 2A), we noticed that the patient underwent a hemimandibulectomy and reconstruction with Kirschner's wire; in the same image,

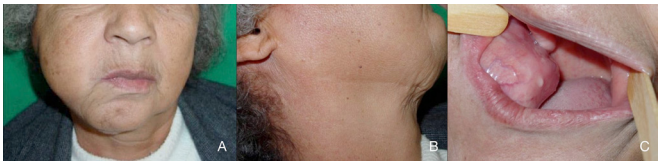

Figure 1. (A) -Facial deformity right side involving temporal region and face, (B) scar in the base region of the mandible regarding the reconstruction of 30 years ago and (C) intrabuccal lesion involving jugal mucosa right side.

we also observed a great mass was present in the region of the right condyle (Figure 2B). The patient was asked to undergo computed tomography (CT) scan with soft tissue window for better evaluation. In CT images, we noticed the presence of two major lesions, one in the temporal region and skull base (Figure 2C) on the right side and another involving the right mandible.

You may notice that the lesion at the base of the skull is causing bone resorption, leaving the base of the skull with marked resorption. The CT scan shows the presence of cystic cavities within the lesion (Figure 2D). An incisional biopsy was performed at the outpatient level, and a specimen was sent to the pathologist.

The sections stained with hematoxylin-eosin revealed epithelial islands resembling the enamel organ epithelium in a fibrous mature connective tissue stroma. These nests of epithelium had a central area constituted of loosely arranged cells. It was further noted that this central portion was surrounded by a single layer of elongated, polarized, ameloblast-like columnar cells.

With the diagnosis, the surgical removal and rehabilitation of this patient were planned.

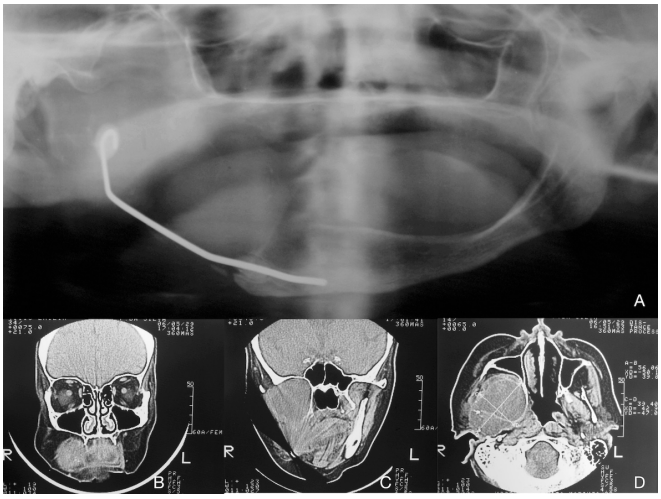

Figure 2. (A) Panoramic image showing lesion mandible right side, (B) CT image revealing intrabuccal soft tissue lesion on right side, (C) CT showing lesion causing resorption of skull base and (D) CT showing lesion in the condylar region and temporal region.

After diagnosis of ameloblastoma, the patient was admitted to the hospital and underwent general anaesthesia to remove the recurrent lesion and carry out reconstruction. Exposure of the lesion was performed

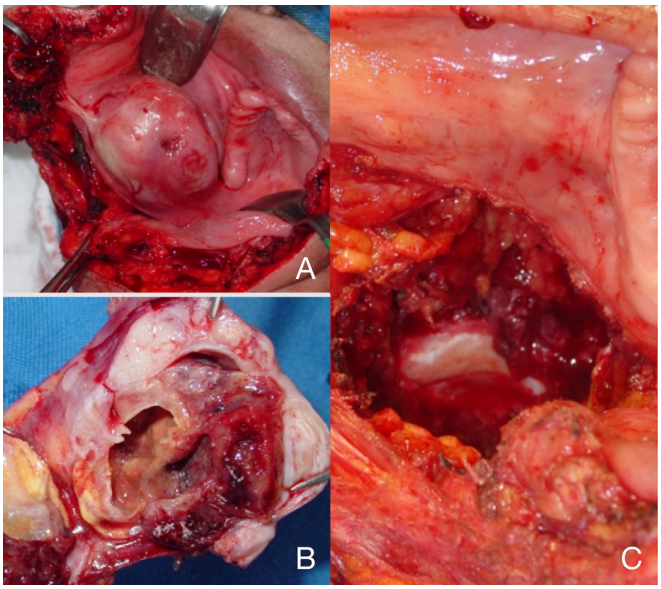

Figure 3. (A) Intra-oral Surgical Trans, (B) Surgical specimen evidencing cystic cavities and (C) temporal fossa.

through submandibular access with superior extension in the mandibular midline (Figure 3).

The region was reconstructed with a reconstruction plate of the 2.4 system, and in the region of the condyle, a resin device similar to the condylar anatomy was made (Figure 4). Suturing was performed in the anatomical planes. The patient is being followed up, and has reported no pain or infection. The followed up was programed for 1 month, 6 months and, 1 year after surgical procedure until 5 year follow up.

DISCUSSION

Ameloblastoma is a pathology with infiltrative potential and the location in this case leaves us cautious because it affects a region that can lead to death or severe sequelae for the patient, as it infiltrates the base of the skull, and it is not possible to remove the pathology with a safety margin. This case is very similar to some cases reported in the literature[4,5,13] that reported the recurrence of ameloblastoma causing facial deformity in the temporal region. In the case reported by Al-Bayaty et al.[13], the authors present a schematic showing the possible path covered by the tumour to reach the temporal region, following the muscular insertions, serving as a basis

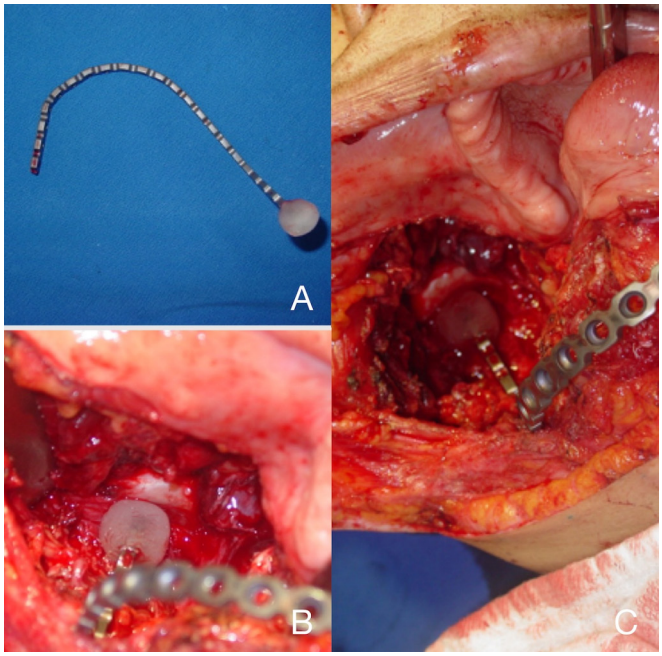

Figure 4. (A) Titanium plate with condylar process reconstructed in resin, (B) condyle in resin positioned in the temporal fossa and (C) condyle in resin positioned in the temporal fossa.

to explain the pathway of the recurrence reported in this article. It is suggested that the pathway covered by ameloblastoma in soft tissue in this case follows the direction of the masticatory muscles.

We can observe the same growth pattern in another article[5] that reports two cases of recurrences of ameloblastomas in soft tissue, where the recurrences were observed. In one of the cases, recurrence occurred after 6 months of resection (original tumour in the left posterior jaw region involving soft tissue), and the recurrence of this case was observed in the temporal region. In the other reported case, the recurrence was in the posterior mandibular region on the right side, and reconstruction was performed with iliac graft. The relapse was after 25 years, evolving to the skull base and temporal region.

Another characteristic that was observed similar to the case reported by Al-Bayaty et al.[13] was that of a specimen with several cystic cavities containing liquid inside, as observed in this case (Figure 3B), and can be explained by the similarities in the histopathological descriptions. With these reports of similar cases, we can observe the growth pattern of the tumour. In these reports, it is suggested that conservative treatment should be evaluated with caution since the relapse in soft tissue can occur in vital and complex regions, where it is difficult to carry out removal with a safety

margin[5,7,13,14]. As was observed in the case presented, since the base of the skull underwent resorption due to the ameloblastoma, it became even tougher to leave a safety margin in this region.

The study by Feinberg & Steinberg[14] suggests that lesions located in the posterior region of the mandible are treated conservatively. The case presented in his first intervention 30 years ago reported a lesion in the posterior region of mandible. Radical treatment with hemimandibulectomy was performed and the patient still presented with recurrence. Thus, there is no established protocol for treatment. Each case should be approached in a unique way, depending on the histopathological, location, size, age, and other characteristics that may influence the relapse.

Follow-up should be performed by treating a lesion with tumor characteristics that, although benign, may present aggressive characteristics. We can emphasize the importance of monitoring the patients and guiding them to return for follow-up visits. The location and the path travelled by the lesion, positioning itself in the temporal region can suggest the soft tissue injury it caused in the muscular insertions, using them as way to reach the temporal region.

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