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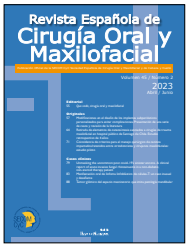

Clinical case

Glomus tumour of the masticatory space mimicking oral mandibular pathology

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ABSTRACT

Glomus tumour is a mesenchymal neoplasm from modified smooth muscle cell of the glomus apparatus. Only 39 cases have been reported in the oral cavity. A 32-year-old female presented with intense pain and hypoaestesia in the mandibular nerve area. Oral physical examination revealed a soft bulging tender to palpation. Orthopantomography showed a radiolucent, well-defined image in the mandibular ramus suggesting an osseous disorder. However, CT and angio-CT showed a hypervascular lesion in the right masticatory space. An intraoral surgical approach to the lesion was chosen. Histopathological analysis showed diffuse and strong positivity for vimentin. Smooth muscle actin, muscle specific actin, and smooth muscle myosin heavy-chain were also positive, but immunoreactivity for markers varied in extension and intensity between different tumour areas. Calponin showed focal and weak immunostaning. Proliferation index (Ki67-MIB1) was less than 1 %.

Based on these morphological and immunohistochemical findings, the tumour was diagnosed as a glomus tumour (solid type). This first description of a glomus tumour in the masticatory space makes us include it in the differential diagnosis of neoplasms in this area.

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Tumor glómico del espacio masticatorio que imita patología mandibular

Palabras clave:

Tumor glómico, neoplasias de cabeza y cuello, casos clínicos.

RESUMEN

El tumor glómico es una neoplasia mesenquimal derivada de la célula muscular lisa modificada del aparato glómico. Solo se han descrito 39 casos en la cavidad oral. Una mujer de 32 años presentó dolor intenso e hipoestesia en el área del nervio mandibular. El examen físico oral reveló un abultamiento suave sensible a la palpación. La ortopantomografía mostró una imagen radiolúcida y bien definida en la rama mandibular que sugiere un trastorno óseo. Sin embargo, el TC y el angio-TC mostraron una lesión hipervascular en el espacio masticatorio derecho. Se optó por un abordaje quirúrgico intraoral de la lesión. El análisis histopatológico mostró una positividad difusa y fuerte para la vimentina. La actina del músculo liso, la actina específica del músculo y la cadena pesada de miosina del músculo liso también fueron positivas, pero la inmunorreactividad para los marcadores varió en extensión e intensidad entre las diferentes áreas tumorales. Calponina mostró inmunotinción focal y débil. El índice de proliferación (Ki67-MIB1) fue inferior al 1 %. Basado en estos hallazgos morfológicos e inmunohistoquímicos, el tumor se diagnosticó como un tumor glómico (tipo sólido). Esta primera descripción de un tumor glómico en el espacio masticatorio nos hace incluirlo en el diagnóstico diferencial de las neoplasias en esta área.

INTRODUCTION

Glomus tumour is a mesenchymal neoplasm originated from modified smooth muscle cell of the glomus apparatus involved in the thermoregulation process[1].

This tumour can be located in any part of the body and represents less than 2 % of soft tissue tumours[2]. It preferably arises in limbs, particularly in the fingertips (subungual area) and, occasionally in the respiratory, digestive and genitourinary tracts[3].

Less than 1 % of these tumours are located in the head and neck area, and just 39 cases of oral and 3 of masticatory (oropharynx, pterygoid fossa and parotid) glomus tumours have been described so far[1,3,4]. They are generally benign in nature, although isolated descriptions of intraoral malignant glomus tumours[5] and multiple-located benign tumours have been published[6].

Different pathogenetic hypothesis have been proposed to explain the different locations of glomus tumours: either considering them as hyperplasias or hamartomas of glomus cells, as a heterotopic proliferation of glomus cells, or even as perivascular cells with glomocytic differentiation[3].

Most oral glomus tumours appear as a painless swelling[2] or neoplasm, and occasionally as a painful mass[1]. The case reported here is, to the best of our knowledge, the first glomus tumour described in the masticatory space, whose clinical presentation was a painful oral swelling affecting the mandibular ramus.

Therefore, the aim of this report was to describe the tumour presentation with it´s main clinical, pathological and immunohistochemical features, together with it´s surgical management.

CASE REPORT

A 32-year-old female, with no relevant medical history, was referred to the Service of Oral and Maxillofacial Surgery of Santiago de Compostela University Hospital because of intense pain and hypoaestesia in the area of the mandibular nerve. Pain increased during mouth opening, but without any mechanical limitation for mandibular movement.

The patient had been treated with steroideal and non-steroideal anti-inflammatory drugs (both local and systemic), together with myorelaxants and minor opioid drugs with poor results.

Physical examination revealed a soft bulging in the masticatory space, tender to palpation, which does not change colour with gentle pressure. Orthopantomography showed a radiolucent image in the ramus of the mandible with well-defined borders at the lingula mandibulae area. CT and angio-CT showed a hypervascular lesion in the right masticatory space with a maximum diameter of 24.7 mm. The lesion was mainly made of a ball of vascular structures, between pterygoid muscles, eroding the inner side of the right mandibular ramus (Figures 1 and 2 A-B).

An intraoral approach to the lesion was chosen with cervicotomy in order to control the external carotid artery due to proximity with the tumour and to prevent haemorrhage due to a potential intraoperative lesion (Figure 3).

Pathological analysis: The samples were fixed in 10 % neutral buffered formalin for 24 hours and embedded in paraffin routinely. Sections, 4mm thick, were stained with hematoxylin and eosin (H&E). Immunohistochemical techniques were automatically performed in an Autostainer Link 48 (Dako-Agilent, Glostrup, Denmark). The following Flex primary antibodies (Dako-

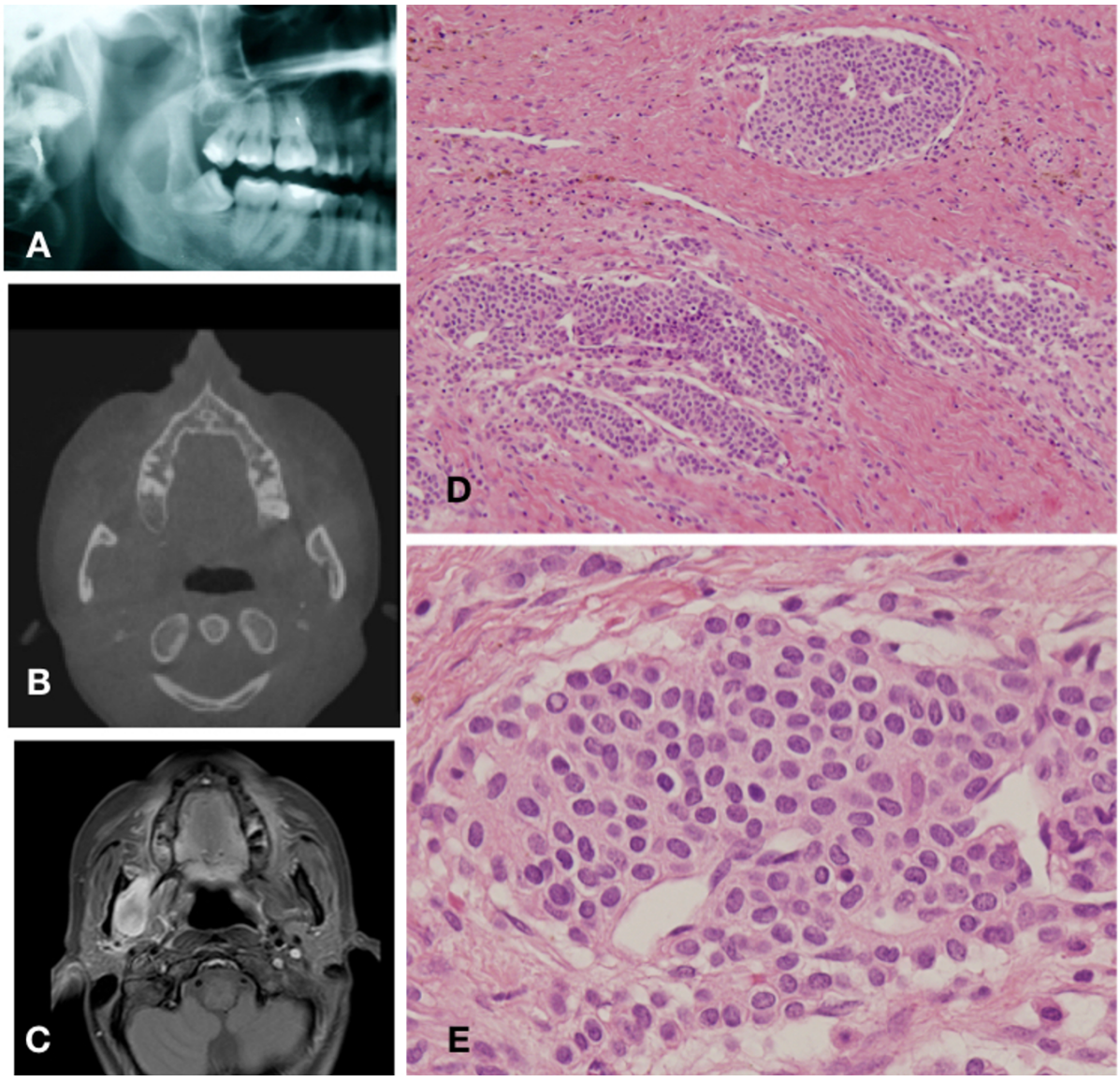

Figure 1. A: radiolucent image in mandibular ramus. B, C: mass within the masticatory space compressing the inner side of the mandibular ramus. D: histopathology: The tumour was composed by nests or sheets of cells with small vascular elements. The stromal tissue was dense (HE, x10). E: tumour cells showed round or polygonal shape, eosinophilic cytoplasms and round or ovoid nuclei. No atypia or mitosis was observed (HE, x40).

Agilent) were employed: vimentin, smooth muscle actin (SMA), muscle specific actin (HHF35), smooth muscle myosin-heavy chain (SMM-HC), calponin, desmin, S100 protein, CK AE1/AE3, CD31, CD34, Factor VIII, D2-40, CD56, CD68, chomogranin, synaptophysin and Ki67 (MIB1). EnVision FLEX/HRP (Dako-Agilent), a dextran polymer horseradish peroxidase, was used as detection system, and 3,3'-diaminobenzidine as chromogen.

Histologically, the tumour showed an organoid pattern composed of solid masses and islands of tumour cells with thin vascular spaces surrounded by dense connective tissue. Tumour cells were small and uniform. They showed round or polyhedral morphology and eosinophilic cytoplasms. Nuclei were round or ovoid, small and regular. No atypia was found, and mitotic figures were inconspicuous.

Immunohistochemical techniques showed diffuse and strong positivity for vimentin (Figure 4). Smooth muscle actin (SMA), muscle specific actin (HHF35) and smooth muscle myosin heavy-chain (SMM-HC) were also positive, but immunoreactivity for these markers varied both in extension and intensity between different tumour areas. Calponin showed focal and weak immunostaining. Negativity was observed for desmin, S100 protein, CK AE1/AE3, CD31, CD34, Factor VIII, D2-40, CD56, CD68, chromogranin and synaptophysin (not shown). Proliferation index (Ki67-MIB1) was less than 1 %.

Based on these morphological and immunohistochemical findings, the tumour was diagnosed as a glomus tumour (solid type of the WHO classification). No relevant incidences were observed in the intra-operative and post-operative period. The

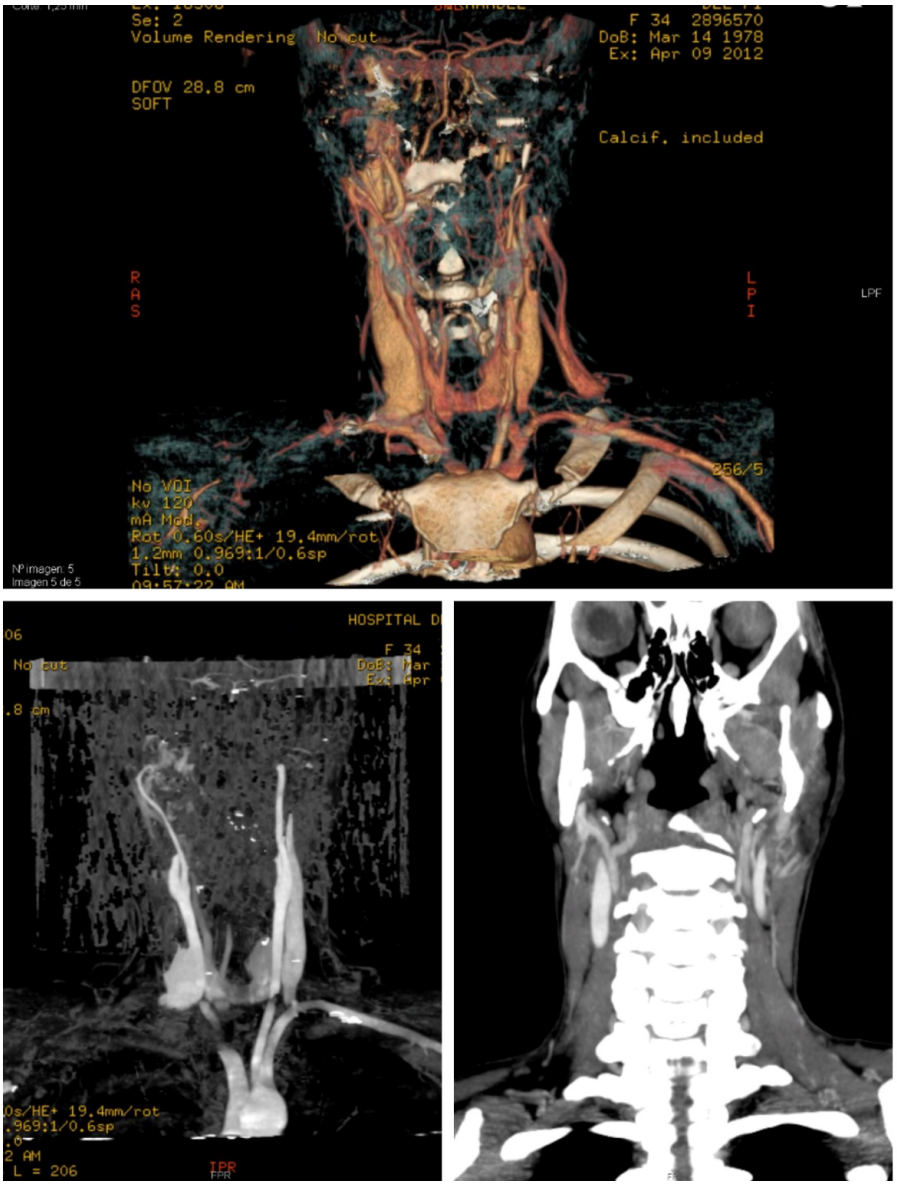

Figure 2. A: preoperative angiography in colour contrast in which lesion is clearly perceived. B: preoperative angiographiy in black and white contrast in which lesion is clearly perceived. C: angio-CT of control after more than 10 years showing no pathology.

patient remains asymptomatic after more than 10 years of follow-up with a hypoaesthesia in the mandibular nerve area as the only sequel. Angio-CT did not show signs of recurrence (Figure 2C).

DISCUSSION

The most frequently described oral locations for glomus tumours are lip (n = 14), hard palate (n = 7), buccal mucosa (n = 7), tongue (n = 5), gum and periodontal maxilla (n = 4)[1,3,4,7,8]. Multiple-located glomus tumours are rare (face, lip, and buccal mucosa)[6], as are intra-osseous presentations with root resorption[4]. The mentioned case we report was located within the masticatory space, which can host a wide range of disorders, such as pseudolesions, developmental and inflammatory lesions, as well as benign and malign tumours or secondary extensions of tumours to the masticatory space; these pathologies can sometimes have characteristics in common as being radiolucent, rounded, with limited shape, and can also be in relation with teeth, making it difficult to diagnose[9]. This case shows the glomus tumour causing erosive changes in the mandibular ramus, seen as a radiolucent image in the orthopantomography, suggesting a primary impression of bone lesion of cystic origin or induced by an odontogenic tumour (odontogenic keratocyst, ameloblastoma, etc.), or seeming a fibro-osseous lesion such as an ossifying fibroma, or even resembling the image produced by a neurogenic tumour. However,

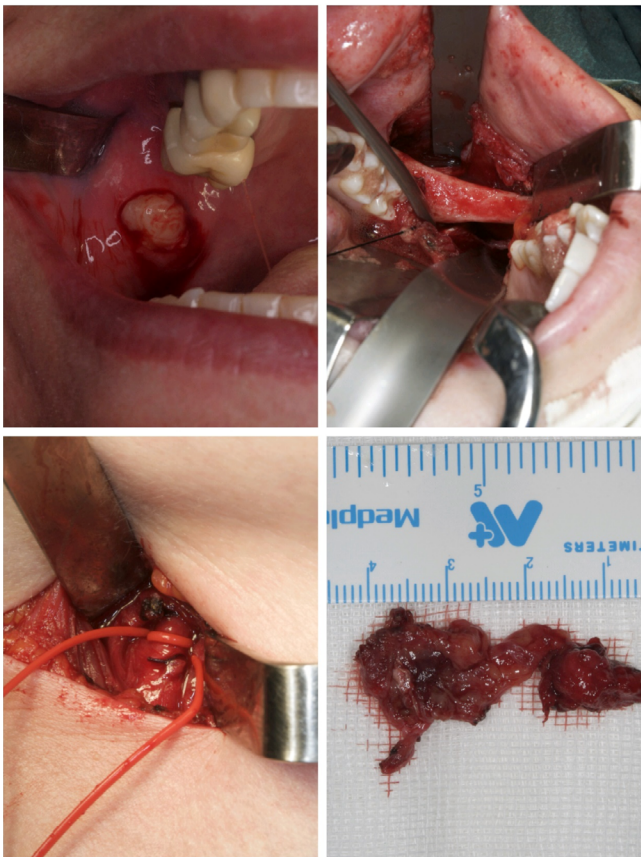

Figure 3. A: intraoperative approach showing the tumour. B: intraoperative approach showing the masticatory space localization. C: cervical approach showing the cervicotomy. D: tumour removed and measured.

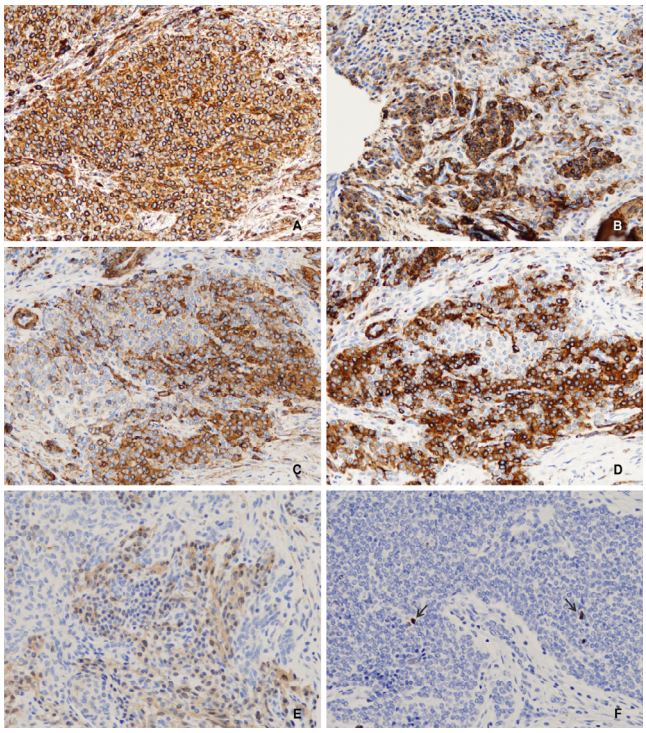

Figure 4. Immunohistochemistry. A: virtually all tumour cells presented intense immunoreactivity for vimentin (x20). B: smooth muscle actin (SMA, x20). C: muscle specific actin (HHF35, x20). D: smooth muscle myosin heavy-chain (SMM-HC, x20) showed irregular immunoreactivity, with intense positive cells intermingled with weak or negative ones. E: immunostaining for calponin was focal and weak (x20). F: proliferation index was very low, with sparse nuclei immunostained for Ki67 (MIB1) (IHC, x20).

CT-scan, angio-CT and MRI are mandatory tools of diagnose that permitted identifying clearly the lesion as extra-osseous with a vascular origin, which oriented both the diagnosis and the therapeutic approach.

Benign glomus tumours have proved able to occupy the bone marrow area and to resorb roots and cortical bone[4].

Regarding symptoms, oral tumours are usually asymptomatic while extraoral ones are commonly painful[2]. The pain experienced varies widely in intensity[3], and patients usually link it to variations in pressure[10], which may well explain the changes in pain intensity with masticatory movements reported by our patient.

Glomus tumours, particularly those not superficial, are a real challenge for pre-surgical diagnosis. Both CT and MRI are paramount in the diagnosis of masticatory space masses for distinguishing between intrinsic and extrinsic lesions to this space; CT gives precise data about hard tissues and MRI provides accurate data in terms of soft tissue, making the correct diagnose based in the lesion component and relation with the adjacent structures[7]. CT-angiography or MRI angiography are also useful for vascular mapping of the lesion.

Treatment of glomus tumours is surgical excision, with different possible approaches. In the case we present, an intraoral approach may be useful for selected small, benign tumours. Generally speaking, relapse of glomus tumours is relatively infrequent (5-17 %)[4], with 3 recurrences out of 39 (7.6 %)[4,7,9] oral tumours reported after follow-up periods ranging from 2 months to 8 years.

Three different histological patterns of glomus tumours have been reported: the most frequent are solid glomus tumour (solid masses of glomus cells), followed by the angiomatous type (glomangioma) and the glomangiomyoma (smooth muscle)[1].

The solid type was predominant in the case we present, and the immunohistochemical profile can bring light about the histogenesis and filiation of the subtypes of glomus tumours in this rare, not previously described, site.

In this vein, glomus tumours should be included in the differential diagnosis of neoplasms in the masticatory space as it can appears even being an infrequent tumour in an atypical localization.

CONFLICT OF INTEREST STATEMENT

We declare no conflict of interest.

ETHICAL APPROVAL

No authorization was required but the article is in law with Helsinki Declaration and the patient signed an informed consent about their participation and publication of the data obtained.

COMPETING INTERESTS

Not applicable.

FUNDING

We have not sources of funding.

AVAILABILITY OF DATA AND MATERIALS

The data sets used and/or analysed during the current study are available from the corresponding author on reasonable request.

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