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Clinical case

Salivary gland oncocytoma. Case report and literature review

Oncocitoma de glándulas salivales. Reporte de caso y revisión de la literatura

Iván André Avilés Hidalgo,*,‡ Zeyda María Oliman Hoyos,*,§ Tania Chávez Priego*,¶

ABSTRACT

Introduction: oncocytoma is a rare benign neoplasm with clinical and imaging characteristics similar to other benign glandular tumors, representing 2% of them. Histopathologically, oncocytic cells with eosinophilic granular cytoplasm, abundant mitochondria, and rounded nuclei with characteristic prominent nucleoli are observed. **Objective:** to present the diagnostic and therapeutic approach to a parotid oncocytoma with a literature review describing its differential characteristics. **Case report:** a 58-year-old female patient with a symptomatic swelling in the right preauricular region with a six-month evolution. Simple and contrast-enhanced tomography revealed a spherical iso-dense lesion associated with the superficial lobe of the parotid gland. Surgical protocol was established to perform a conservative superficial parotidectomy for lesion excision. **Results:** postoperative evolution was favorable, without evidence of facial nerve injury or recurrence of the injury at 24 months of follow-up. Conclusions: the identification of oncocytic cells by histopathological study is the key to the differential diagnosis of oncocytoma compared to other salivary gland tumors with similar clinical symptoms and

RESUMEN

Introducción: *el oncocitoma es una neoplasia benigna poco prevalente con características clínicas e imagenológicas similares con otros tumores benignos glandulares, representando 2% de los mismos. Histopatológicamente se observan células oncocíticas con citoplasma granular eosinofílico, mitocondrias abundantes y núcleos redondeados con nucleolos prominentes característicos.* ***Objetivo:*** *presentar el abordaje diagnóstico y terapéutico de un oncocitoma en glándula parótida con una revisión de la literatura describiendo sus características diferenciales.* ***Presentación del caso:*** *mujer de 58 años con un aumento de volumen sintomático en la región preauricular derecha con una evolución de seis meses. La tomografía simple y contrastada evidenció una lesión isodensa esférica asociada al lóbulo superficial de la glándula parótida. Se protocolizó quirúrgicamente para realizar la escisión de la lesión mediante una parotidectomía superficial conservadora.* ***Resultados:*** *la evolución postoperatoria fue favorable, sin evidencia de lesión del nervio facial ni recurrencia de la lesión en 24 meses de seguimiento.* ***Conclusiones:*** *la identificación de células oncocíticas mediante el estudio histopatológico es la*

* Centro de Alta Especialidad «Dr. Rafael Lucio», Veracruz, Mexico.
‡ Oral and Maxillofacial Surgery Resident. Department of Oral & Maxillofacial Surgery.
§ Oral and Maxillofacial Surgeon. Department of Oral & Maxillofacial Surgery.
¶ Oral and Maxillofacial Pathologist. Anatomical Pathology Department Chief.

Correspondence:
Iván André Avilés Hidalgo
Av. Adolfo Ruiz Cortines Núm. 2903, Unidad Magisterial, 91020 Xalapa-Enríquez, Veracruz, Mexico. Tel: +52 221 764 0039.
E-mail: livingtheglory@gmail.com

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imaging, its treatment is surgical and requires long-term surveillance due to the risk of recurrence.

Keywords: *oncocytoma, benign tumor, salivary glands, parotidectomy.*

clave para el diagnóstico diferencial del oncocitoma frente a otros tumores de glándulas salivales con clínica e imagen similares, su tratamiento es quirúrgico y requiere vigilancia a largo plazo por el riesgo de recidiva.

Palabras clave: *oncocitoma, tumor benigno, glándulas salivales, parotidectomía.*

Abbreviations:
CT = Computed Tomography
EMA = Epithelial Membrane Antigen

INTRODUCTION

Oncocytoma, also referred to as oxyphilic adenoma,[1,2] is a benign epithelial neoplasm initially described by Jaffé in 1932. It is classified by the 5th edition of the World Health Organization's histological classification in 2022 among the benign tumors of the salivary glands.[3,4] Cases of oncocytomas have also been described in seromucous glands of the respiratory tract, as well as in other secretory organs such as the thyroid, parathyroid, lacrimal glands, pancreas, and kidney.[5]

Regarding its epidemiology, it is worth mentioning that it represents 2% of salivary gland tumors, with an annual incidence ranging from 0.4 to 13.5 per 100,000 cases worldwide.[1,6] It occurs most frequently in old age, with a higher prevalence between the sixth and eighth decades of life, and it has a slight inclination for the female sex.[2,7] They are characteristic of the major salivary glands, with the parotid gland being the most affected with 90% of cases, while its presence in minor salivary glands is rare.

Clinically, it is identified as a firm, asymptomatic, solitary swelling, with a growth rate that can vary from a few months to several years, with an approximate diameter of 2.3 cm.[8] When located in the parotid gland, they are associated with the superficial lobe and are difficult to distinguish from other benign tumors, making their histopathological study the key to their definitive diagnosis. In some cases, it can be associated with oncocytosis with the presence of bilateral tumors.

Contrast-enhanced Computed Tomography (CT) scans are useful for the diagnosis of parotid oncocytomas, as they allow identification of their capsule and association with the gland of origin.[9] Another type of study, such as fine-needle aspiration biopsy, has a sensitivity of 70% and a specificity of 81% for salivary gland tumors; however, due to the overlap of cytological characteristics with other entities, it can lead to an erroneous diagnostic suspicion.[7,10] The differential diagnosis can be made with pleomorphic adenoma, Warthin's tumor, and malignant or metastatic tumors with similar cytological characteristics.[3,11]

The histopathological analysis shows a lesion delimited by a thin fibrous connective tissue capsule enclosing nests, sheets, or trabeculae of oncocytes, which are polyhedral cells with central hyperchromatic nuclei.[11] The cytoplasm is clear due to the high collagen content, with the presence of eosinophilic granules and abundant mitochondria, which are identified with electron microscopy, mitochondrial antibodies, or staining with hematoxylin and phosphotungstic acid. The use of immunohistochemical tests for the following markers has been mentioned: cytokeratin (CK7, CK8 and CK19) and Epithelial Membrane Antigen (EMA).[12] The treatment of choice for oncocytoma is the surgical excision of the lesion, considering the risk of recurrence if the surrounding capsule ruptures. The procedure should be carried out with the excision of the superficial lobe of the parotid gland (superficial parotidectomy) and with total enucleation in the case of the submandibular gland. When it occurs in minor salivary glands, a safety margin of healthy tissue is recommended.[4,13]

The recurrence rate is approximately 2.6%, with a favorable prognosis in most cases; however, oncocytic carcinomas, which have malignant histopathological characteristics, have been described in the literature.[13] Another described variant is oncocytomas of the nasosinusal glands, which are locally aggressive, leading to their classification as low-grade malignant neoplasms. In both cases, the prognosis is poor.[7,14,15]

This study aims to report a clinical case of salivary gland oncocytoma along with a literature review that provides a broader perspective with updated evidence to establish precise clinical, imaging, and histopathological criteria, facilitating differential diagnosis, surgical management, and prognosis of

this rare benign salivary gland neoplasm, highlighting the importance of this case.

CASE REPORT

We describe the case of a 58-year-old woman who attended the clinic due to swelling in the right pre-auricular region that had been developing over a period of six months and was associated with pain. She had previously received pharmacological treatment with non-steroidal anti-inflammatory drugs at her health center. Upon direct questioning, she reports arterial hypertension controlled with telmisartan 40 mg every 24 hours. She denies personal pathological history associated with the manifested lesion, and denies personal surgical and allergic history.

Upon physical examination, a swelling on the right pre-auricular region, of indurated consistency, well-defined, mobile, not adhered to deep planes, measuring approximately 2 × 2 cm was observed. The ipsilateral Stensen's duct was patent, with no apparent adenopathies. Oral opening was preserved and mucous membranes with adequate hydration and color.

A simple and contrast-enhanced CT scan of the head and neck was requested, which showed an isodense tumor mass dependent on the superficial lobe of the right parotid gland, well-defined, unilocular, measuring 2.0 × 2.4 × 2.5 cm *(Figure 1)*. Taking into account the clinical presentation and the tomographic findings, the decision was made to schedule a surgical protocol, beginning with her preoperative evaluation.

An approach was performed using a modified Blair incision *(Figure 2A)*, followed by flap elevation and blunt dissection by planes, identifying the facial nerve using the Borle's triangle, the main trunk of the facial nerve was dissected very carefully, also identifying its terminal branches *(Figure 3A)*. Then the excision of the superficial lobe of the parotid gland along with the associated tumor was performed *(Figure 3B and Figure 4)*. Hemostasis was achieved and an active Jackson-Pratt drain was placed. The incision was closed in layers, and finally, a compressive dressing was applied *(Figure 2B)*.

The patient showed a favorable evolution in the immediate postoperative phase with preserved facial mobility, surgical wound without dehiscence and drainage of 10 mL in 24 hours, being removed 48 hours postoperatively. Therefore, following the recommendations of the Enhanced Recovery After Surgery (ERAS) protocol, it was decided to discharge him from hospital.

The histopathological report described a benign neoplasm of epithelial origin characterized by the proliferation of polyhedral cells arranged in anastomosing cords with eosinophilic granular cytoplasm, with scarce fibrovascular stroma, and the absence of pleomorphism and nuclear and cellular atypia *(Figure 5)*.

The lesion is delimited by a capsule of fibrous connective tissue, towards the periphery, serous-predominant salivary gland acini are appreciated with abundant unilocular adipose tissue in the stroma, and no suspicious signs of malignancy were identified *(Figure 6)*. Immunohistochemical studies

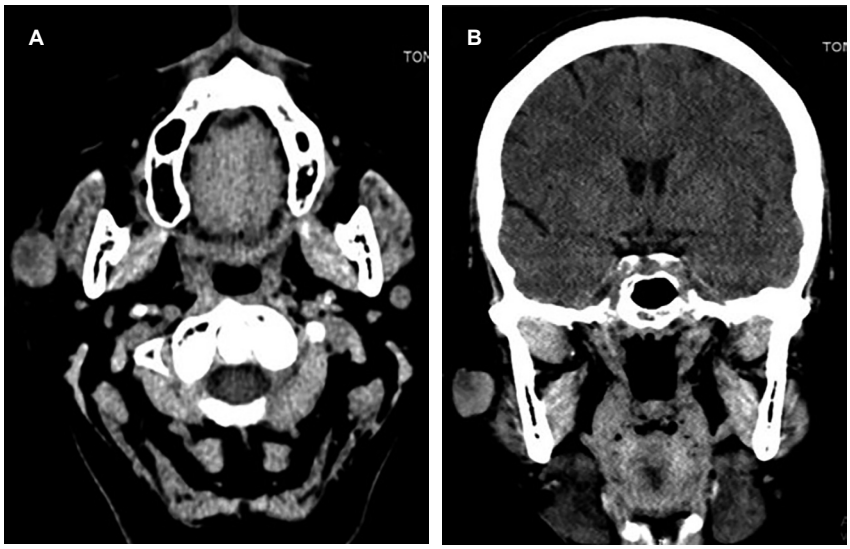

Figure 1:

Isodense mass associated with the superficial lobe of the right parotid gland. **A)** Axial view. **B)** Coronal view.

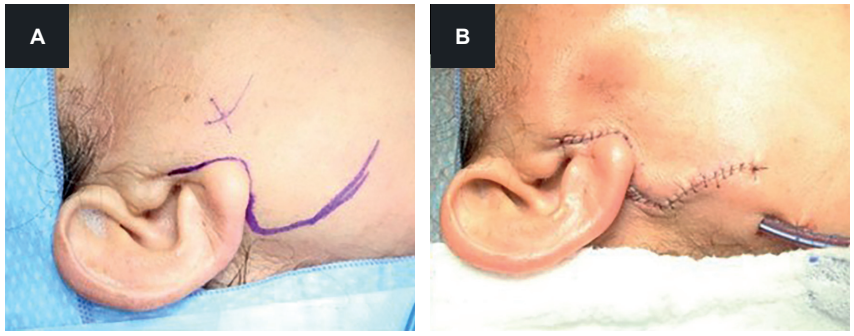

Figure 2:

Modified Blair incision. **A)** Pre-surgical. **B)** Post-surgical.

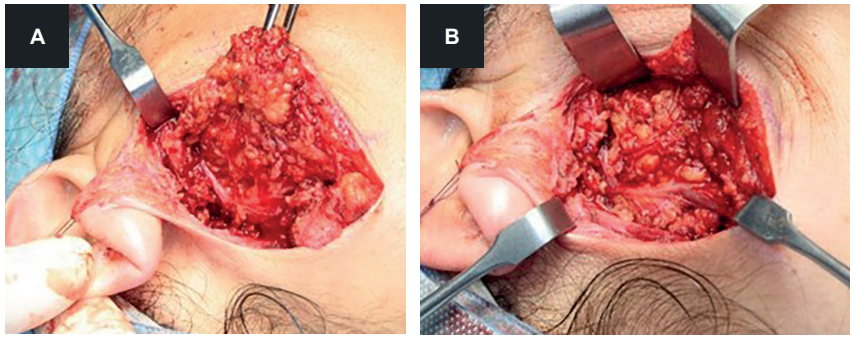

Figure 3:

Surgical approach. **A)** Pre-surgical. **B)** Preserved facial nerve.

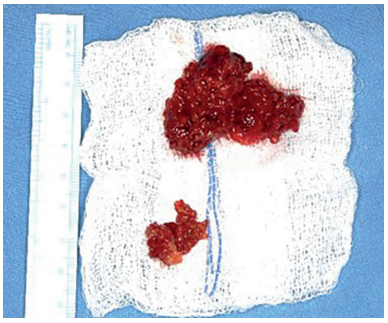

Figure 4:

Surgical specimen.

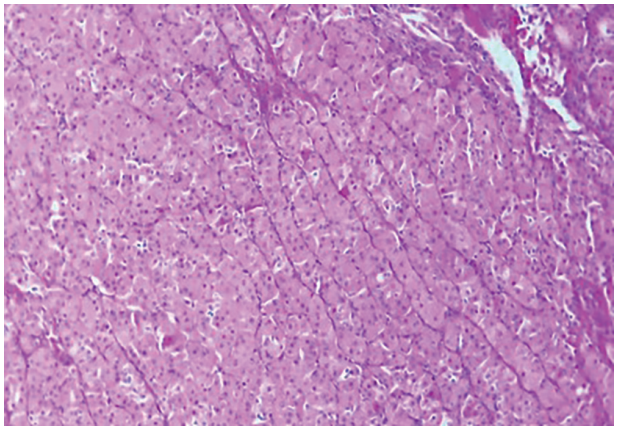

Figure 5: Typical oncocytes with eosinophilic granular cytoplasm and clear cells.

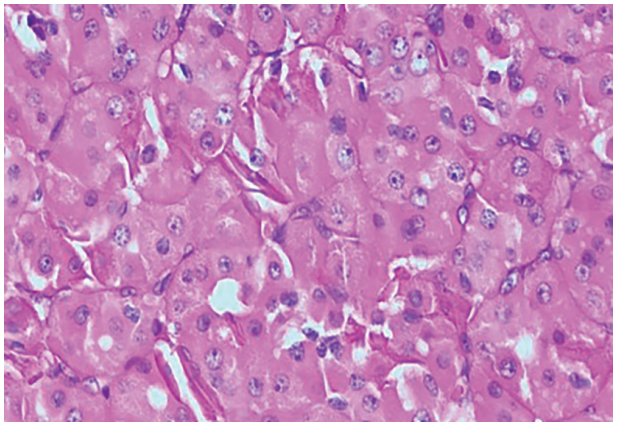

Figure 6: Polygonal cells with clear and eosinophilic cytoplasm, and large central nuclei.

were not performed because the hospital's pathology laboratory does not provide that type of analysis.

Postoperative follow-up was carried out at seven, 30, 90, and 180 days after hospital discharge, evidencing preservation of the terminal branches of the facial nerve, adequate healing of the surgical wound, and no symptomatology in the surgically intervened area. Currently, an annual 24-month control is maintained in surveillance without presenting recurrence of the tumor lesion.

DISCUSSION

Oncocytoma is a rare and underestimated entity within the presumptive diagnoses of salivary gland pathologies. Authors such as Imran et al. mention a higher prevalence between the sixth and eighth decades of life, a preference for the female sex, and association with major salivary glands in 90% of cases, consistent with our case where we present an oncocytoma in the right parotid gland in a 58-year-old woman.[5,13]

For diagnosis, data obtained from the patient's medical history and physical examination were considered. Bhushan et al. mention that they generally present as painless masses of several years' evolution; however, in our case, we found a well-defined inflammation of six months' evolution, approximately 2 cm in diameter, highlighting the particularity of presenting painful symptoms.[8]

Prabhakar et al. points to plain CT with contrast as a study that allows locating the lesion, determining its association with the superficial lobe of the gland, and delimiting its capsule, so it was requested to confirm its parotid association and determine the size of the tumor lesion.[7,11] Incisional biopsies are contraindicated because they can rupture the fibrous membrane surrounding the tumor, which can lead to recurrences after treatment.[9,12]

The treatment was performed via superficial parotidectomy where an encapsulated tumor adhered to the superficial lobe of the gland was identified. Singh et al. comment on the risk of recurrence with capsule rupture when only the lesion is enucleated, so the decision was made to perform excision it along with the superficial parotid lobe. Radical parotidectomy is reserved for cases of malignancy with facial nerve infiltration.[4,13]

The strengths of this case include the comprehensive clinical, imaging, and histopathological evaluation and the scarce documentation of this lesion in the literature. Among the limitations are the lack of complementary immunohistochemical studies that complement the histological diagnosis and documented postoperative follow-up 24 months postoperatively. In view of the above, it is evident that there is a need to document more cases of neoplasms associated with salivary glands that allow the clinician and surgeon to have weapons that facilitate the approach to this type of benign pathologies.

CONCLUSIONS

Oncocytoma is a benign neoplasm with a prevalence of 2% in salivary glands, usually associated with the parotid gland in its superficial lobe. Clinically and in imaging studies it is indistinguishable from other benign glandular tumors. Therefore, the histopathological study identifying polyhedral cells with hyperchromatic nuclei, eosinophilic cytoplasm and abundant mitochondria (oncocytes) becomes relevant.

Although they were not used in this study, immunohistochemical tests have been described for the following markers: cytokeratin (CK7, CK8 and CK19), EMA and mitochondrial antibodies. The abundance of mitochondria can be confirmed with electron microscopy or staining with hematoxylin and phosphotungstic acid.

Treatment is performed by excisional biopsy or superficial parotidectomy, which protects the integrity of the facial nerve and its surrounding structures. Management with a refined surgical technique is key, seeking to dissect and completely enucleate the lesion that is usually encapsulated, reducing the risk of recurrence which, although it is 2.6%, is always latent so it is necessary to carry out periodic postoperative follow-up.

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Conflict of interests: the authors declare that they have no conflict of interests.