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Clival Salivary Heterotopia (Salivary Choristoma): A Rare Radiopathogical Lesión

Javier Ortiz[1*], Juan Carlos Paniagua[2], Luis Chinchilla[1], Enrique Montero[1], Elisa Muñoz[1], María Dolores Ludeña[1]

[1]Dpto. de Biología Celular y Patología (Universidad de Salamanca), Servicio de Anatomía Patológica (Hospital Universitario de Salamanca), Salamanca, Spain
[2]Servicio de Radiodiagnóstico (Hospital Universitario de Salamanca), Salamanca, Spain
[3]Dpto. de Biología Celular y Patología (Universidad de Salamanca), Salamanca, Spain
Email: *jortiz@usal.es

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Abstract

Objective: To describe the radiological and pathological presentation of a salivary heterotopia in clivus. The lesion was detected incidentally in a 57-year-old woman. **Material and Methods:** Radiologic imaging (CT scan and MRI) and histological picture obtained from a clival lesion. **Results:** Radiologically the lesion presented as a well-limited lytic lesion with a diameter of 1.5 cm and accompanied by a cerebrospinal fluid secondary fistula. Histological examination revealed non-pathological salivary gland tissue. **Conclusion:** The differential diagnosis of this rare entity mandates a histological examination of the lesion.

Keywords

Salivary Heterotopia, Clivus

1. Introduction

The increasing use of sophisticated radiological studies for neurological patients provides a thorough and ever-growing body of knowledge on lesions in the craniospinal axis.

This statement is particularly true when the diagnostic process is confronted with rare entities, imposing on both radiologists and pathologists the need to delve into deeper knowledge about the differential diagnosis of these often incidentally discovered lesions. Our case report on a salivary heterotopia in clivus adequately illustrates this problem.

2. Case Report

A previously healthy 57-year-old woman was referred to the Neurosurgery Service of our Hospital for the study of a persistent cerebrospinal fluid fistula, which was iatrogenically triggered by a nasopharyngeal swab for COVID19 diagnosis. CT scan revealed in upper clivus a well delimitated lythic lesion measuring 1.5 cm on its maximal diameter (Figure 1(A)). MRI showed an infrasellar clival process which protruded into cisterna pontis and the sphenoidal sinus, where an air-fluid level is seen (Figure 1(B)). Surgical repair of the fistula was indicated, and the lesion was removed followed by muscular dural patch plastia.

The sample submitted for pathological study was an irregularly shaped fragment of greyish tissue with petrous consistency. After decalcification with nitric acid, the histological picture showed normal salivary tissue into the intraosseous lesion (Figure 2 and Figure 3). Ki67 immunohistochemistry revealed a very low proliferative index (<1%). Therefore, a final diagnosis of salivary heterotopia (choristoma) was established.

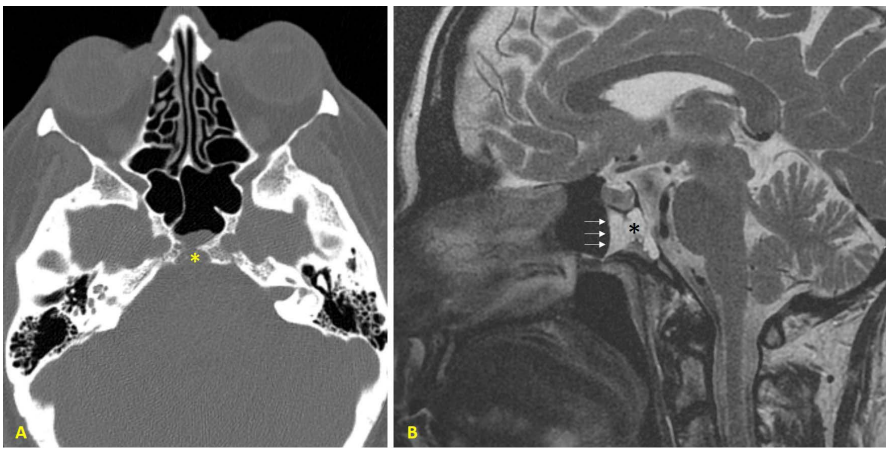

Figure 1. (A): Axial CT scan image shows a lytic lesion on the cranial aspect of clivus, with well-defined and partially sclerosed borders. (B): Sagital T2 enhanced MR image shows an hyperintense lesion in clivus protruding into the pontine cisterna and the sphenoidal sinus (asterisks). An air-fluid level (arrows) is observed in the sphenoidal sinus.

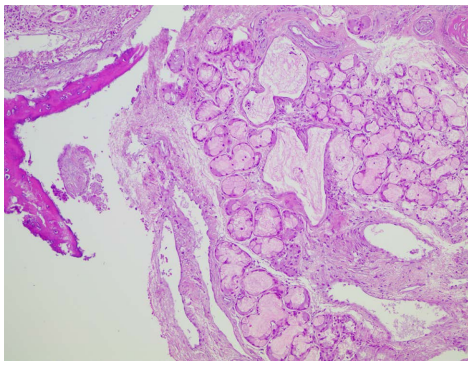

Figure 2. Pathology: Hematoxilin-eosin histologic picture (×200) showing normal salivary tissue surrounded by osseous tissue.

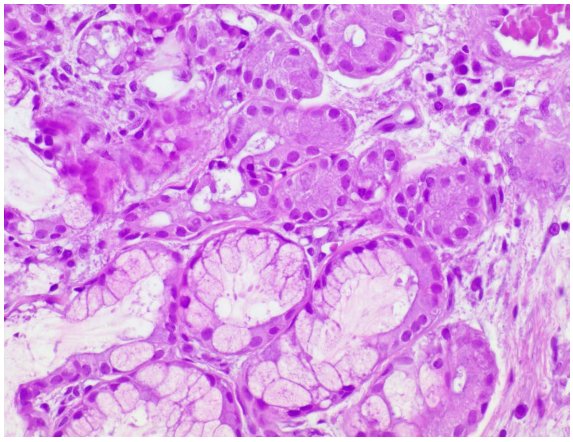

Figure 3. Pathology: Hematoxilin-eosin histologic picture (×400) confirms no abnormality in salivary tissue.

3. Discussion

The presence of normal tissue in ectopic localization is the fundamental criterium for the diagnosis of heterotopic lesions, also called choristomas [1] [2] [3]. These are very rare benign entities usually appearing as a mass lesion detected clinically or radiologically. They have been described for different tissues, such as pancreatic choristoma in stomach [4], nasally located neural tissue (glial nasal heterotopia, nasal glioma) [5], thyroid tissue in tongue (lingual thyroid heterotopia) [6] or endometriosis [7].

Salivary heterotopia can present in two forms [8]. The most common one is intranodal, affecting peri-parotideal lymphatic nodules in babies and toddlers. Extranodal presentation can be subdivided into two forms. Lower extranodal lesions are related to pharingeal pouches and localize on the inferomedial aspect of the sternocleidomastoid muscle [9] [10] [11]. Cranial extranodal presentation embraces cases described in mandibles, ear, inner ear, palatine tonsil, mylohyoid muscle, pituitary gland and pontocerebellar angle [12] [13] [14]. It has to be stressed that ectopic salivary tissue can harbor abnormalities similar to those in its orthotopic counterpart, such as metaplasia, cystic transformation or neoplasia [15] [16].

Our case is a hitherto undescribed localization of salivary heterotopy. Sphenoidal involvement cases are localized beneath sella turcica and affect pituitary gland. To the best of our knowledge, no cases localized in clivus have been reported.

An incidentally discovered lesion in clivus must be differentiated from other rare entities encountered on this localization. Chondrosarcoma, malignant neoplasia of cartilaginous lineage [17], chordoma, a locally aggressive tumor of notochordal origin [18], ectopic hypophysiary adenoma [19], salivary carcinoma metastases or the very rare ecchordosis physaliphora, a benign congenital lesion due to notochordal tissue remnants [20], should be borne in mind.

In our case, the benign clinical and radiological presentation and, conclusively,

the histological picture of a normal salivary gland discard the above-mentioned primary lesions.

A year after surgery the patient remains asymptomatic and without recurrence of the lesion.

Finally, we would like to emphasize the relevance of a close collaboration between clinicians, radiologists, and pathologists to optimize our healthcare endeavor.

Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

References

[1] Lee, K.H. and Roland, P.S. (2013) Heterotopias, Teratoma, and Choristoma. In: Kountakis, S.E., Ed., *Encyclopedia of Otolaryngology, Head and Neck Surgery*. Springer, Berlin, 1179-1183. https://doi.org/10.1007/978-3-642-23499-6_642

[2] Ozolek, J.A. and Tekkesin, M.S. (2021) The "-OMAS" and "-OPIAS": Targeted and Philosophical Considerations Regarding Hamartomas, Choristomas, Teratomas, Ectopias, and Heterotopias in Pediatric Otorhinolaryngologic Pathology. *Head Neck and Pathology*, **15**, 25-40. https://doi.org/10.1007/s12105-020-01251-y

[3] Batsakis, J.G., El-Naggar, A.K. and Hicks, M.J. (1993) Epithelial Choristomas and Teratomas of the Tongue. *Annals of Otology, Rhinology & Laryngology*, **102**, 567-569. https://doi.org/10.1177/000348949310200715

[4] LeCompte, M.T., Mason, B., Robbins, K.J., *et al*. (2022) Clinical Classification of Symptomatic Heterotopic Pancreas of the Stomach and Duodenum: A Case Series and Systematic Literature Review. *World Journal of Gastroenterology*, **28**, 1455-1478. https://doi.org/10.3748/wjg.v28.i14.1455

[5] Zhang, W., Tang, L.X., Wang, P.P., Ge, W.T. and Ni, X. (2020) Intranasal glial Heterotopia in a Male Infant: A Case Report. *Medicine* (*Baltimore*), **99**, e21200. https://doi.org/10.1097/MD.0000000000021200

[6] Amr, B. and Monib, S. (2011) Lingual Thyroid: A Case Report. *International Journal of Surgery Case Reports*, **2**, 313-315. https://doi.org/10.1016/j.ijscr.2011.10.004

[7] Laganà, A.S., Garzon, S., Götte, M., *et al*. (2019) The Pathogenesis of Endometriosis: Molecular and Cell Biology Insights. *International Journal of Molecular Sciences*, **20**, Article 5615. https://doi.org/10.3390/ijms20225615

[8] Martínez-Madrigal, F., Bosq, J. and Casiraghi, O. (2007) Major Salivary Glands. In: Mills S.E., ed., *Histology for Pathologists*. 3rd Edition, Lipincott Williams & Wilkins, Philadelphia, 445-469.

[9] Adams, W.P. and Donahoe, P.K. (1979) Salivary Gland Heterotopia in the Lower Part of the Neck. The Archives of Surgery, **114**, 79-81. https://doi.org/10.1001/archsurg.1979.01370250081017

[10] Jain, S., Aggarwal, A., Deshmukh, P., Singhvi, P. and Sudarshan, K. (2011) Heterotopic Salivary Gland Presenting as a Discharging Sinus in the Base of the Neck. *International Journal of Clinical Practice*, **1**, e131. https://doi.org/10.4081/cp.2011.e131

[11] Haemel, A., Gnepp, D.R., Carlsten, J. and Robinson-Bostom, L. (1996) Heterotopic Salivary Gland Tissue in the Lower Neck. *The Journal of Dermatology*, **23**, 287-289.

https://doi.org/10.1111/j.1346-8138.1996.tb04015.x

[12] Wise, J.B., Sehgal, K., Guttenberg, M. and Shah, U.K. (2005) Ectopic Salivary Tissue of the Tonsil: A Case Report. *International Journal of Pediatric Otorhinolaryngology*, **69**, 567-571. https://doi.org/10.1016/j.ijporl.2004.11.019

[13] Tanaka, Y., Kubo, A., Ayabe, J., Watanabe, M., Maeda, M., Tsuura, Y. and Tanaka, Y. (2015) Intrasellar Symptomatic Salivary Gland Rest with Inflammations. *World Neurosurgery*, **84**, 189.e13-189.e18. https://doi.org/10.1016/j.wneu.2015.02.018

[14] Rodriguez, F., Scheithauer, B.W., Ockner, D.M. and Giannini, C. (2004) Solitary Fibrous Tumor of the Cerebellopontine Angle with Salivary Gland Heterotopia a Unique Presentation. *The American Journal of Surgical Pathology*, **28**, 139-142. https://doi.org/10.1097/00000478-200401000-00017

[15] Oh, S.J., Lee, D., Suh, Y.L. and Lee, J.I. (2013) Heterotopic Glioneuronal Mass of the Cerebellomedullary Cistern with Duct Cyst Formation of Ectopic Salivary Gland Tissue. *Neuropathology*, **33**, 179-184. https://doi.org/10.1111/j.1440-1789.2012.01334.x

[16] Daniel, E. and McGuirt, W.F (2005) Neck Masses Secondary to Heterotopic Salivary Gland Tissue: A 25-Year Experience. *American Journal of Otolaryngology*, **26**, 96-100. https://doi.org/10.1016/j.amjoto.2004.08.009

[17] Ding, C., Chen, W., Liu, F., Xiong, M. and Chen, J. (2019) Skull Base Chondrosarcoma Caused by Ollier Disease: A Case Report and Literature Review. *World Neurosurgery*, **127**, 103-108. https://doi.org/10.1016/j.wneu.2019.03.037

[18] Kremenevski, N., Schlaffer, S.M., Coras, R., Kinfe, T.M., Graillon, T. and Buchfelder, M. (2020) Skull Base Chordomas and Chondrosarcomas. *Neuroendocrinology*, **110**, 836-847. https://doi.org/10.1159/000509386

[19] Campana, C., Nista, F., Castelletti, L., Caputo, M., Lavezzi, E., Marzullo, P., Ferrero, A., Gaggero, G., Canevari, F.R., Rossi, D.C., Zona, G., Lania, A., Ferone, D. and Gatto, F. (2022) Clinical and Radiological Presentation of Parasellar Ectopic Pituitary Adenomas: Case Series and Systematic Review of the Literatura. *Journal of Endocrinological Investigation*, **45**, 1465-1481. https://doi.org/10.1007/s40618-022-01758-x

[20] Park, H.H., Lee, K.S., Ahn, S.J., Suh, S.H. and Hong, C.K. (2017) Ecchordosis Physaliphora: Typical and Atypical Radiologic Features. *Neurosurgical Review*, **40**, 87-94. https://doi.org/10.1007/s10143-016-0753-4