

Pleomorphic adenoma of the hard palate with calcifications: an unusual presentation.

Adenoma pleomórfico del paladar duro con calcificaciones:
una presentación inusual.

Jorge Obando.^{1,2}
Noelia Coronado.^{1,3}
Ana Trevejo-Bocanegra.¹

Affiliations:

¹Universidad Peruana Cayetano Heredia.
Lima, Perú.

²Universidad de Antioquia. Medellín,
Colombia.

³Universidad Mayor Nacional de San
Marcos. Lima, Perú.

Corresponding author: Jorge Obando. Av.
Honorio Delgado, San Martín de Porres
15102. Lima, Perú. **Phone:** (57) 3166841690.
E-mail: jorge.obando@upch.pe

Receipt : 08/12/2019 **Revised:** 02/23/2019
Acceptance: 03/30/2020

Abstract: Pleomorphic Adenoma (PA) is the most common benign salivary gland tumor. The most common sites for minor salivary gland from which PA arises are the palate followed by the lips and buccal mucosa. Calcifications are a common finding in major salivary glands with chronic inflammatory disorders. Major salivary gland tumors rarely show calcifications and it is less common to find them in minor salivary gland tumors. We report a case of pleomorphic adenoma of the hard palate in a 67-year-old female patient with intra-tumoral, irregular and scattered calcifications visible on computed tomography (CT). The treatment was complete surgical excision of the lesion. The diagnosis was confirmed with the histopathological study.

Keywords: *Palate, hard; salivary glands, minor; salivary gland neoplasms; adenoma, pleomorphic; tomography, x-ray computed; calcinosis.*

Cite as:

Obando J, Coronado N & Trevejo-
Bocanegra A.

Pleomorphic adenoma of the hard
palate with calcifications: an unusual
presentation.

J Oral Res 2020; 9(2):150-154.

Doi:10.17126/joralres.2020.022

Resumen: El adenoma pleomórfico (AP) es el tumor benigno de las glándulas salivales más común. Los sitios de mayor frecuencia donde surge el AP en glándulas salivales menores es el paladar seguido de los labios y la mucosa bucal. Las calcificaciones son un hallazgo común en las glándulas salivales mayores con trastornos inflamatorios crónicos, pero en el caso de tumores rara vez muestran calcificaciones y es menos común encontrarlos en tumores de las glándulas salivales menores. Presentamos un caso de adenoma pleomórfico del paladar duro en una paciente de 67 años con calcificaciones intratumorales, irregulares y dispersas visibles en la tomografía computarizada. El tratamiento fue la extirpación quirúrgica completa de la lesión. El diagnóstico se confirmó con el estudio histopatológico.

Palabra Clave: *Paladar duro; glándulas salivales menores; neoplasias de las glándulas salivales; adenoma pleomórfico; tomografía computarizada por rayos x; calcinosis.*

INTRODUCTION.

Salivary gland tumors represent less than 3% of head and neck tumors. Pleomorphic adenoma (PA) is considered the most common benign neoplasm and accounts for 45%-74% of all major and minor salivary gland tumors.^{1,2} PA occurs in individuals of all ages but it is most common between the third and sixth decades of life; the average patient age at presentation is approximately 45 years. The female to male ratio is 2:1.³⁻⁵ In the oral cavity, PAs grow slowly as a painless and dome-shaped mass, the symptoms are mainly due to mass effect of the swelling and include difficulty in mastication, swallowing, and speech.^{2,6} On computed tomography (CT) and magnetic resonance (MR) imaging, PAs show as well-defined lesions, occasionally accompanied by lobulated contours. Histologically, PA

exhibit epithelial and mesenchymal components and it is also called as a benign mixed tumor.

The terms pleomorphic adenoma and mixed tumor both represent attempts to describe the unusual histopathological features of this tumor.⁵ Imaging findings of PAs depend on the cellular density, proportion of epithelial and stromal components, and second histological changes (fibrosis, lipometaplasia, ossification, cystic degeneration and infarction).^{1,7}

Although calcifications are a common finding in inflammatory salivary gland disorders, salivary gland tumors rarely shows calcifications;⁸ however, calcifications have been found in both benign and malignant salivary gland tumors.⁹ We report a case of pleomorphic adenoma on the hard palate with intratumoral, irregular and scattered calcifications visible on CT.

Figure 1. Intra oral view of swelling in the left side of the palate, with crossing of the midline.



Figure 2. Bone windows CT.

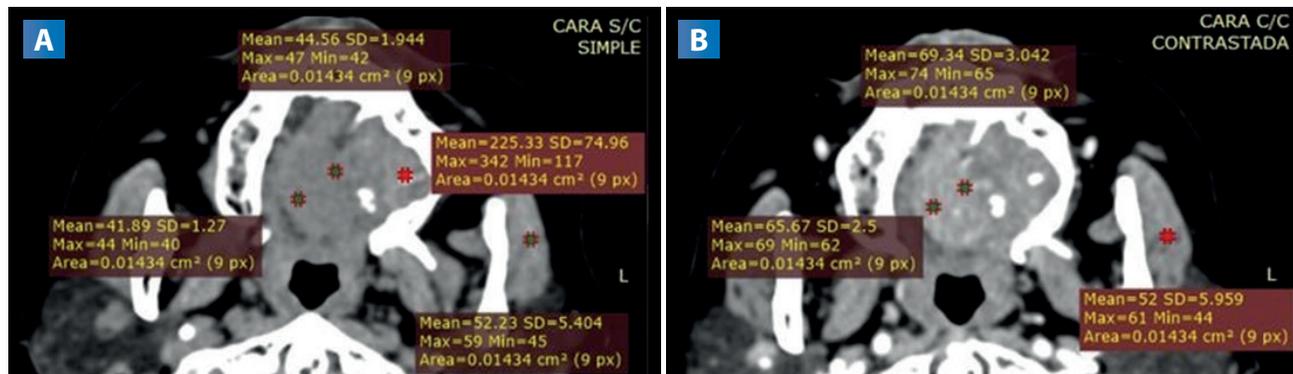


(A-B): Axial images show bone resorption in the left alveolar bone and hard palate with corticated borders in the underlying bone. Intra-tumoral, irregular and scattered calcifications are seen.

(C): Coronal image shows bone resorption of the left nasal floor and the floor of the left maxillary antrum.

(D): Posterior coronal Image shows no changes of the greater palatine canals.

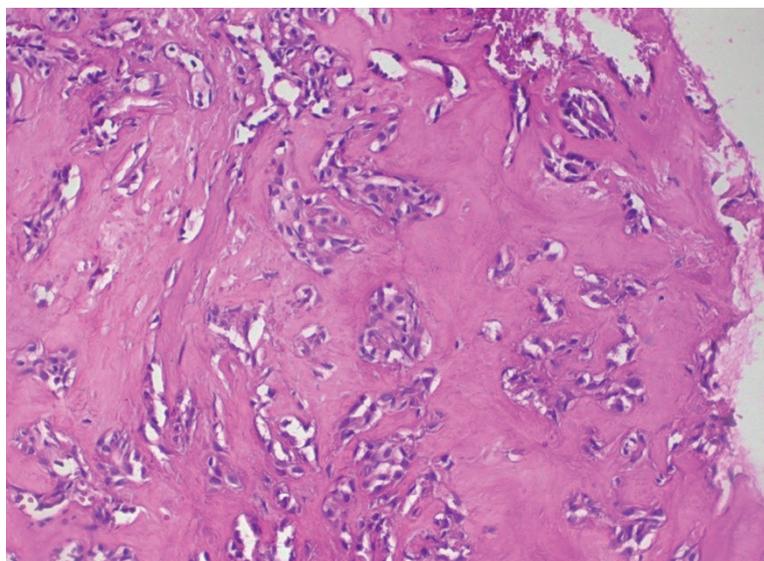
Figure 3. Axial CT images in soft-tissue window.



A: Before contrast medium administration: a mass in the palatal area can be seen. The mean Hounsfield Units (HU) values of the mass are 41.89 in the periphery and 44.56 in the center. The mean HU value of a calcification is 225.33.

B: After medium contrast administration, an intermediate enhancing mass with heterogeneous pattern representing the solid component of the tumor can be observed; the mean HU values are 65.67 in the periphery and 69.34 in the center, higher values than those found in muscle tissue (52 HU).

Figure 4. Hematoxylin and eosin stained microscopic image of the tumor, showing myoepithelial cells with ovoid nuclei submerged in myxoid and chondromyxoid stroma.



CASE REPORT.

A 67-year-old female patient reported to the dental clinic of the Universidad Peruana Cayetano Heredia in Lima, Peru. The patient was derived from a private dentist, with an approximately 30-year history of painless swelling in her mouth, which was small initially and gradually increased in size. The patient complained that the lesion causes her difficulty in mastication and speech, being asymptomatic at all times of its growth. The patient had no relevant medical or family history.

Clinical examination revealed well-circumscribed, non-ulcerated, dome-shaped, palatal swelling

on the hard palate (Figure 1). The mass was smooth, firm and did not involve any teeth. Bone windows CT images showed a well-defined and saucer-like excavation in the hard palate with corticated borders in the underlying bone; intra-tumoral calcifications were also found (Figure 2).

Soft Tissue Windows CT images after contrast medium administration, showed a tumor with intermediate enhancement, well defined margins and a heterogeneous pattern (Figure 3). Incisional biopsy revealed a pleomorphic adenoma; due to the clinical examination, tomographic evaluation, history of the

lesion and incisional biopsy, a surgical excision and resection with margin of grossly normal surrounding tissue was performed.

The histopathological examination of the mass revealed a tumor covered by a fibrous capsule, consisting of proliferation of mantles and islands of myoepithelial cells with ovoid nuclei, the cells forming ductal structures submerged in myxoid and chondromyxoid stroma (Figure 4). This confirmed the diagnosis of PA. The patient is under regular follow-up with no signs of recurrence.

DISCUSSION.

Seventy percent of the tumors in minor salivary glands are PAs, about 80% - 90% of these tumors occur in the major salivary glands and 10% occur in the minor salivary glands. The most common intra oral site is the palate, followed by the upper lip and buccal mucosa. The unusual sites are the floor of the mouth, tongue, tonsil, pharynx, and retromolar area.

PAs most commonly occur between the third and the sixth decade of life, just as the case described here, and it is rare in children.^{3,4} Our case is clinically similar to others cases of PAs of the palate reported in the literature, as they usually present as a slow-growing, painless, unilateral, dome-shaped and firm mass, that rarely ulcerates the overlying skin or mucosa. Most common symptoms due to tumors at this location are dysphagia, difficulty in mastication and speech similar to our case; other less common symptoms included dyspnoea, acute airway obstruction and obstructive sleep apnea.²⁻⁴

Imaging benign tumors of the minor salivary glands may produce a well-defined saucer-like depression in the underlying bone; in contrast, malignant tumors may invade the bone and have poorly defined borders.^{10,11} In the current case, bone CT images show a well-defined and saucer-like excavation of the hard palate with corticated borders. Contrast-enhanced CT images of PAs usually show intermediate to high signal intensity.^{1,10,12}

Our case is not far from the tomographic features of PAs in the study conducted by Kim *et al.*,¹² who found that the majority of PAs showed high or intermediate signal intensity on contrast-enhanced CT with a median ROI of 69.7 HU (ranged -13 to 187 UH). Those tumors had smooth borders and heterogeneous patterns of contrast enhancement; the minority showed ill-defined borders, patterns of homogenous contrast enhancement

and peripheral enhancement.¹² Some authors reported that the tumoral mean density value of pleomorphic adenomas in delayed phase on contrast-enhanced CT was highest than for malignant tumors or other benign salivary gland tumors.¹³

Calcifications within the salivary gland parenchyma usually indicate the presence of chronic sialadenitis, but if calcifications occur in a discrete mass inside the gland, some authors believe the most likely lesion is a PA.^{10,14} Other authors consider that intratumoral calcifications in salivary glands could be considered as a criterion for malignancy.⁸

Farid *et al.*,¹⁴ demonstrate in a systematic review that intra-tumoral salivary gland calcifications have been found in both benign and malignant tumors, although the frequency was found to be higher in malignant (47.5%) than in benign tumors (25%); although benign salivary gland tumors are generally more prevalent than malignant ones, intratumoral calcifications could not be considered as a criterion for malignancy. Bone formation is relatively rare, the mechanism of osseous formation in PA remains controversial, but there are two traditional hypotheses regarding this: endochondral ossification and deposition of osteoid tissue by modified myoepithelial cells.¹

Histologically, morphological diversity is the most characteristic feature of PAs, which are characterized by a mixture of both epithelial and stromal (mesenchymal) components. Stromal components can be myxomatous, chondromatous, lipomatous, hyalinized, fibrous, or osseous.^{7,15} Conventionally, the treatment of PAs is essentially surgical excision. Though these benign tumors are well encapsulated, resection of the tumor with an adequate margin of grossly normal surrounding tissue is necessary to prevent local recurrence. Although tumor recurrence rates are low, long-term follow-up is required.^{16,17}

CONCLUSION.

PAs present multiple histological characteristics and therefore variability of imaging presentations, including the presence of calcifications, which is relatively rare and can make it difficult to establish a diagnosis.

Previously it was thought the presence of calcifications visible on CT could represent a criterion for malignancy; However, these have been reported in both benign and malignant cases, and we reported a benign case to corroborate this fact.

Conflict of interests: The authors declare that there is no conflict of interests.

Ethics approval: This research was approved by the Universidad Peruana Cayetano Heredia in Lima, Peru.

Funding: None.

Authors' contributions: All authors contributed to this manuscript.

Acknowledgements: None.

REFERENCES.

1. Kato H, Kawaguchi M, Ando T, Mizuta K, Aoki M, Matsuo M. Pleomorphic adenoma of salivary glands: common and uncommon CT and MR imaging features. *Jpn J Radiol.* 2018;36(8):463–71.
2. Chaturvedi M, Jaidev A, Thaddanee R, Khilnani A. Large Pleomorphic Adenoma of hard palate. *Ann Maxillofac Surg.* 2018;8(1):124–6.
3. El-Naggar A, Grandis J, Takata T, Slootweg P. WHO Classification of Head and Neck. 4th Ed. IARC, Lyon; 2017.
4. Passi D, Ram H, Ranjan S, Laxman D, Malkunje R. Pleomorphic Adenoma of Soft Palate: Unusual Occurrence of the Major Tumor in Minor Salivary Gland – A Case Report and Literature Review. *J Maxillofac Oral Surg.* 2017;16(4):500–5.
5. Figueiredo NR, Dinkar AD, Meena M. Pleomorphic adenoma of the hard palate: Report of a case Pleomorphic adenoma of the hard palate: Report of a case. *Int J Med Dent.* 2015;1–3.
6. Alves V, Pérez-de-Oliveira M, de Castro J, Vieira L, Leão J, Perez D. Intraoral Pleomorphic Adenoma in Young Patients. *J Craniofac Surg.* 2018; 29 (2): 209-211.
7. Pereira T, Shetty S. Pleomorphic adenoma of palate with predominant chondroid tissue: A rarity. *J Cancer Res Ther.* 2018;14(2):476–7.
8. Yoon JH, Ahn SG, Kim SG, Calcifications JK. Calcifications in a clear cell mucoepidermoid carcinoma of the hard palate. 2005;927–9.
9. Ananthaneni A, Ponnappalli H, Kiresur M, Krishna Chaitanya S. Mucoepidermoid carcinoma involving the palate with lamellated calcifications: A notable finding. *Eur J Gen Dent.* 2016;5(1):35.
10. Kakimoto N, Gamoh S, Tamaki J, Kishino M, Murakami S, Furukawa S. CT and MR images of pleomorphic adenoma in major and minor salivary glands. 2009;69:464–72.
11. Kaneda T, Minami M, Ozawa K, Akimoto Y, Okada M, Yamamoto H, et al. Imaging tumors of the minor salivary glands. *Oral Surgery, Oral Med Oral Pathol.* 1994;78(3):385–90.
12. Kim H, Kim SY, Kim YJ, Ko J mun, Park MJ, Kim JH, Hah JH, Kwon TK, Kim KH, Sung MW. Correlation between computed tomography imaging and histopathology in pleomorphic adenoma of parotid gland. *Auris Nasus Larynx.* 2018;45(4):783–90.
13. El-Atta M, Amer T, Gaballa G, El-Sayed N. Multi-phasic CT versus dynamic contrast enhanced MRI in characterization of parotid gland tumors. *Egypt J Radiol Nucl Med.* 2016;47(4):1361–72.
14. Farid MM, Mohamed W. Intra-tumoral salivary gland calcification: a systematic review. *Egypt Dent J.* 2016;61(4):4936–5204.
15. Rousseau A, Badoual C. Head and neck: Salivary gland tumors: an overview. *Atlas Genet Cytogenet Oncol Haematol.* 2011;15(6):533–41.
16. Rawson K, Kallalli B, Gokul K, Singh A. Pleomorphic adenoma of the palate: A case report and review of a rare entity. *J Indian Acad Oral Med Radiol.* 2016;28(3):329.
17. Arumugam P, Christopher P, Kumar S, et al. Pleomorphic Adenoma of the Palate: A Case Report. *Cureus.* 2019; 11 (3): e4308.