



## CASE REPORT

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## Plasma cell granuloma in oral cavity: A case report.

**Abstract:** Plasma cell granuloma is a rare benign tumor lesion that is classified and described under the pseudoinflammatory tumor category. Its occurrence in the oral cavity is rare, making diagnosis and treatment really difficult, as it bears some clinical similarity with malignant tumor diseases. Proper diagnosis and treatment of PCG requires performing biopsy and a histopathological/immunohistochemical study to rule out possible plasma and neoplastic cell dyscrasias. Consequently, the use of these auxiliary diagnostic devices will enable us to provide the appropriate treatment for the patient. In this study, we present the case of a 63-year-old female patient with a tumor/ulcerative lesion of the left buccal mucosa of a month of evolution and a tumor/ulcerative lesion on the right buccal mucosa of 15 days of evolution after the onset of the first lesion. The patient was treated successfully for a period of one year with immunosuppressive drugs, and to date the disease is inactive. The purpose of this paper is to show one of the most unusual locations in the oral cavity affected by this pathological entity, its clinical and histological features, and establish the differential diagnosis correctly with other malignant or benign disease entities, suggesting the most suitable treatment for this type of condition.

**Keywords:** *Plasma cell granuloma, benign tumor/ulcerative lesion, oral cavity.*

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## INTRODUCTION.

Plasma cell granuloma (PCG) has received the following names: inflammatory myofibroblastic tumor, inflammatory pseudotumor, fibrocystic inflammatory proliferation, xantomatosis pseudotumor, benign miofibroblastoma and inflammatory fibrosarcoma; in this article the first term is used. PCG is a rare non-neoplastic lesion first described in 1973 by Bahadori and Liebow, consisting of the proliferation of inflammatory cells with the predominance of plasma cells. It has been classified as an inflammatory pseudotumor that can affect any organ or soft tissue being common in lungs, vagina, and larynx<sup>1-3</sup>.

Bhaskar *et al.* first discovered this pathological entity on gingival tissue, since then few case reports have been

documented. Avecco *et al.* have reported the occurrence of this lesion on gingival tissue of the oral cavity. These authors report a gingival growth with well-circumscribed appearance, reactive and asymptomatic, treated through incisional biopsy. Kim *et al.* reported about the presence of PCG on the gingival surface in patients receiving immunosuppressive drugs (cyclosporine A), showing hyperplasia in that area. Consequently the use of these drugs may be considered as an etiological factor<sup>4</sup>.

PCG is a benign inflammatory lesion that requires biopsy and histopathological/immunohistochemical study to rule out possible neoplastic and plasma cell dyscrasias, and thus establish a differential diagnosis with other similar diseases such as multiple myeloma<sup>5</sup>.

A clinical case of PCG is presented in this paper,

highlighting the clinical and pathological characteristics of this condition as well as the treatment given to the patient.

### CLINICAL CASE.

The case of a 63-year-old Hispanic female, who worked as a surgeon dentist is presented. The patient was accepted for assessment and treatment at the Department of Oral and Maxillofacial Surgery. Her condition suggested the presence of a lesion on the right buccal mucosa of a month of evolution; symptomatic, showing localized pain with variable intensity VAS 7/10, exacerbated when swallowing. The patient denied a history of chronic degenerative diseases as well as the current presence of those conditions. Transfusions, surgical procedures, allergies, and history of addiction were also denied by the patient.

The oral clinical examination revealed tumor lesion on the right buccal mucosa, with a diameter of 5cm, irregular shape, fuzzy boundaries, firm consistency and fixed, with many whitish fibers along it.

Incisional biopsy of tumor lesion on right buccal mucosa was performed extracting approximately a 2cm sample from the lesion (Fig. 1), with a presumptive diagnosis of epidermoid carcinoma.

Fifteen days after the completion of the incisional biopsy, the patient came for a checkup and after exploration the emergence of a new lesion on the left palatal area was detected. The lesion was approximately 6cm long, irregularly shaped, fuzzy boundaries and whitish edges, flat, reddish and ulcerative surface, tender to palpation (Fig. 2). A tumor lesion of approximately 3cm in diameter was observed on the right buccal mucosa. The lesion was irregularly shaped, with fuzzy boundaries, whitish color, with firm consistency, movable and with ulceration in the periphery (Fig. 3).

The patient was referred to the Unit of Dermatology. There it was decided to carry out a second sampling through incisional biopsy, with a presumptive diagnosis of pemphigus vulgaris.

The Unit of Pathology reported the condition as PCG,

**Figure 1.** Initial lesion on the right buccal mucosa, after incisional biopsy.



**Figure 2.** lesion on the hard palate of 15 days of evolution.

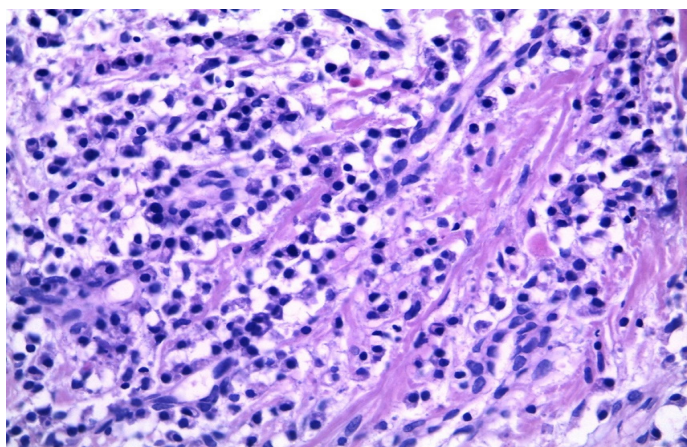


**Figure 3.** Tumor and ulcerative lesion on the left buccal mucosa of 15 days of evolution.

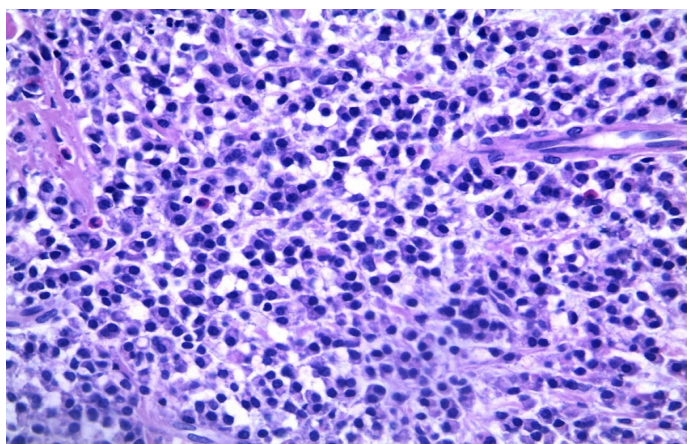




**Figure 4. HE. 40x** Note the dense fibrous stroma with dense collagen, alternating with plasma cells.



**Figure 5. HE 40x.** Note the presence of abundant plasma cells with intense golgian activity, showing an active metabolic state.



since the following characteristics were found: more than 50 plasma cells present in a high power field and presence of fibrous stroma (Fig 4 and 5). Immunohistochemistry to determine the presence of kappa and lambda chains, including clonality was performed<sup>6</sup>. The result was positive for both, indicating a polyclonal reactive process. They also reported positive IgG4, accounting for over 40% of total IgG.

Pharmacological treatment was initiated as follows: prednisone 50mg orally every 24 hours for an indefinite period of time and azathioprine 150mg a day orally.

After 1 month of treatment the patient reported a de-

creased in pain VAS 3/10, without feeling of burn in the area when performing oral hygiene, lesions decreased in size and became less reddish. Clinical examination showed a reduction in size and extent of lesions, so consolidation is scheduled for a month later. Subsequently, there was a reduction in the drug doses initially prescribed.

One year after treatment, the patient reported that the lesions had not reappeared and that she was asymptomatic at that moment. Clinical examination revealed that the disease was inactive and treatment continued with: prednisone 10mg orally every 24 hours and Azathioprine 50mg orally every 24 hours, attending regular checkups at both units.

To date, the patient is asymptomatic and her disease is inactive. With treatment suspended at 1 year and 2 weeks after onset.

## DISCUSSION

The recent WHO<sup>7</sup> classification of soft tissue tumors includes three basic variants of plasma cell granulomas:

Miofibroblast pattern loosely arranged in a myxoid edematous background, showing plasma cells, lymphocytes, eosinophils and blood vessels.

Presence of dense aggregates of spindle cells arranged in a variable myxoid stroma and collagenized background, mixing a distinctive inflammatory infiltrate, diffuse groups of plasma cells and lymph nodes.

Predominance of collagen fibers, resembling scar tissue, with the presence of plasma cells and scattered eosinophils. This variant may have cytologic atypia with nuclear pleomorphism and increased mitotic activity; these characteristics are rare and may be associated with malignant transformation.

One of the pathologies that must be considered in the differential diagnosis due to its macroscopic similarity is extramedullary plasmacytoma, given the poor prognosis for this neoplasia. Its histopathological examination consists of a pure infiltrate of plasma cells arranged in relatively large sheets with a fine reticular stromal, while PCG shows a capillary network as its main feature.

Another feature to differentiate them is tissue replacement by the plasmacytoma, while PCG stick to cells at the tissues<sup>8</sup>.

Clinically, PCG takes at least two morphological types in the oral mucosa: exophytic/tumor or unilateral ulcerative. In this case it adopted both types and was bilateral. According to the limited literature reviewed there was not another case with clinical ulcerative morphology, if so, this would be the first case with this type of manifestation.

If the lesion looks like a tumor, differential diagnosis must be performed first because of its frequency with squamous cell carcinoma, with extramedullary plasmacytoma and multiple myeloma in the oral cavity<sup>9,10</sup>.

If the lesion looks like an ulcer, differential diagnosis will be performed with autoimmune processes such as pemphigus vulgaris, bullous pemphigoid, mucous membrane pemphigoid and erosive lichen planus, and systemic lupus erythematosus.

Histologically it can mimic other diseases, depending on the most prominent type of infiltration: multiple myeloma, lymphomas, sarcomas, plasma cell gingivitis and other benign and malignant growths with predominant spindle cells. During the review of the literature, we found involvement of IgG4<sup>11</sup>, with high concentrations in PCG and other immune diseases<sup>12,13</sup>.

As for the pathogenesis, the PCG case presented in this paper has as a chemical mediator (IgG4) acting as

a destructor of tissues in response to antigens of various origins and is in high concentrations in these cases<sup>14,15</sup>.

The biological behavior of PCG is variable. There are cases where it acts as a reactive inflammatory process, in others as a benign tumor, but there are also reports of cases with malignant neoplastic behavior at least locally, with obvious infiltration and destruction of the affected tissues. The treatment of cases that seem to be malignant neoplasias is surgical excision, chemotherapy and/or radiotherapy; cases of benign neoplasia require only surgery. Reactive proliferations respond to medical management with corticosteroids and other immunosuppressants such as azathioprine<sup>16</sup>. This case has so far showed a benign course, as a reactive process controlled with immunosuppressants.

## CONCLUSION

PCG poses a major diagnostic challenge given its similarity to a large number of disease entities of the oral cavity and the limited information regarding its pathogenesis.

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## Granuloma de células plasmáticas en cavidad oral: Informe de un caso.

**Resumen:** El Granuloma de Células Plasmáticas es una rara lesión tumoral benigna descrita dentro de la clasificación de tumores pseudoinflamatorios, es rara su aparición dentro de la cavidad oral, haciendo realmente complicado su diagnóstico y tratamiento, ya que presenta similitud clínica con patologías tumorales malignas. Para el adecuado diagnóstico y tratamiento del GCP, se requiere de la realización de biopsia y estudio histopatológico/inmu-

nohistoquímico, para descartar posibles discrasias de las células plasmáticas y neoplásicas, de tal forma que el uso de estos auxiliares de diagnóstico nos permitirá sustentar de manera adecuada el tratamiento otorgado al paciente. Se presenta el caso de paciente femenino de 63 años de edad, con una lesión tumoral/ulcerativa de la mucosa yugal izquierda de un mes de evolución y lesión tumoral/ulcerativa de la mucosa yugal derecha de 15 días de evolución posterior a la aparición de la primera lesión, tratada por un periodo de un año con inmunosupresores de forma

exitosa ya que a la fecha la paciente presenta inactiva la enfermedad. El propósito de este manuscrito es mostrar una de las localizaciones más extrañas de esta entidad patológica en la cavidad oral, sus características clínicas e histológicas, establecer de forma correcta el diagnóstico

diferencial con otras entidades patológicas malignas ó benignas y así emplear el tratamiento requerido para la forma en que se presente la misma.

**Palabras clave:** *Granuloma de Células Plasmáticas, Lesión tumoral benigna, Ulcerativa, Cavidad oral.*

## REFERENCES.

1. Small O, Small M. Granuloma de Células Plasmáticas Labial. Presentación de un caso. *Dermatol Perú* 2008;18(1):51-54.
2. Gleason BC, Hornick JL. Inflammatory myofibroblastic tumours: where are we now? *J Clin Pathol.* 2008;61(4):428-37.
3. Urschel JD, Horan TA, Unruh HW. Plasma cell granuloma of the lung. *J Thorac Cardiovasc Surg.* 1992;104(4):870-5.
4. Yao X, Alvarado Y, Brackeen J, Juniper C, Cobos E. Plasma cell granuloma: a case report of multiple lesions in the lung and review of the literature. *Am J Med Sci.* 2007;334(5):402-6.
5. Fumio I, Tetsuo S, Norio H. Plasmacell granuloma of the oral mucosa with angiokeratomatous features. A possible analogue of cutaneous angioplasmocellular hyperplasia. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod.* 2000;89:204-207.
6. Coffin CM, Watterson J, Priest JR, Dehner LP. Extrapulmonary inflammatory myofibroblastic tumor (inflammatory pseudotumor). A clinicopathologic and immunohistochemical study of 84 cases. *Am J Surg Pathol.* 1995;19(8):859-72.
7. Leon B, Eveson J, Reichart P, Sidransky D. World Health Organization Classification of Tumors. Pathology and Genetics Head and Neck Tumors IARC Press Lyon. 2005.
8. Ono K, Shiiba M, Yoshizaki M, Ogawara K, Ishihara T, Yonemori Y, Oide T, Uzawa K, Nakatani Y, Tanzawa H. Immunoglobulin G4-related sclerosing inflammatory pseudotumors presenting in the oral cavity. *J Oral Maxillofac Surg.* 2012;70(7):1593-8.
9. Ide F, Shimoyama T, Horie N. Inflammatory pseudotumor in the mandibular retromolar region. *J Oral Pathol Med.* 1998;27(10):508-10.
10. Seoane J, Aguirre-Urizar JM, Esparza-Gómez G, Suárez-Cunqueiro M, Campos-Trapero J, Pomareda M. The spectrum of plasma cell neoplasia in oral pathology. *Med Oral.* 2003;8(4):269-80.
11. Deshpande V, Zen Y, Chan JK, Yi EE, Sato Y, Yoshino T, Klöppel G, Heathcote JG, Khosroshahi A, Ferry JA, Aalberse RC, Bloch DB, Brugge WR, Bateman AC, Carruthers MN, Chari ST, Cheuk W, Corneli LD, Fernandez-Del Castillo C, Forcione DG, Hamilos DL, Kamisawa T, Kasashima S, Kawa S, Kawano M, Lauwers GY, Masaki Y, Nakanuma Y, Notohara K, Okazaki K, Ryu JK, Saeki T, Sahani DV, Smyrk TC, Stone JR, Takahira M, Webster GJ, Yamamoto M, Zamboni G, Umehara H, Stone JH. Consensus statement on the pathology of IgG4-related disease. *Mod Pathol.* 2012;25(9):1181-92.
12. Kamisawa T, Shimosegawa T, Okazaki K, Nishino T, Watanabe H, Kanno A, Okumura F, Nishikawa T, Kobayashi K, Ichiya T, Takatori H, Yamakita K, Kubota K, Hamano H, Okamura K, Hirano K, Ito T, Ko SB, Omata M. Standard steroid treatment for autoimmune pancreatitis. *Gut.* 2009;58(11):1504-7.
13. Kamisawa T, Okamoto A. IgG4-related sclerosing disease. *World J Gastroenterol.* 2008;14(24):3948-55.
14. Umehara H, Okazaki K, Masaki Y, Kawano M, Yamamoto M, Saeki T, Matsui S, Sumida T, Mimori T, Tanaka Y, Tsubota K, Yoshino T, Kawa S, Suzuki R, Takegami T, Tomosugi N, Kurose N, Ishigaki Y, Azumi A, Kojima M, Nakamura S, Inoue D, Research Program for Intractable Disease by Ministry of Health, Labor and Welfare (MHLW) Japan G4 team. A novel clinical entity, IgG4-related disease (IgG4RD): general concept and details. *Mod Rheumatol.* 2012;22(1):1-14.
15. Okazaki K, Uchida K, Koyabu M, Miyoshi H, Takaoka M. Recent advances in the concept and diagnosis of autoimmune pancreatitis and IgG4-related disease. *J Gastroenterol.* 2011;46(3):277-88.
16. Sato Y, Yoshino T. IgG4-Related Lymphadenopathy. *Int J Rheumatol.* 2012;2012:572539.