Scientific Investigation in Dentistry – SID

2317-2835

Caso Clínico http://periodicos.unievangelica.edu.br/index.php/scientificinvestigationindestist

PLEOMORPHIC ADENOMA OF MINOR SALIVARY GLANDS IN UNCOMMON SITES: A CASE SERIES

ADENOMA PLEOMÓRFICO DE GLÂNDULAS SALIVARES MENORES EM LOCAIS RAROS: UMA SÉRIE DE CASOS

Fernanda Zanol MATOS[1], Artur Aburad de CARVALHOSA[2], Fernanda Silva de ASSIS[3], Helder Fernandes de OLIVEIRA[4], Orlando Aguirre GUEDES[4], Andreza Maria Fábio ARANHA[3], Alessandra Nogueira PORTO[1]

[1]Doutora, Professora da Faculdade de Odontologia do Centro Universitário de Várzea Grande - UNIVAG, Várzea Grande, Mato Grosso, Brasil.
[2]Doutor, Professor da Faculdade de Medicina da Universidade de Cuiabá - UNIC, Cuiabá, Mato Grosso, Brasil.
[3]Doutora, Professora da Faculdade de Odontologia da Universidade de Cuiabá - UNIC, Cuiabá, Mato Grosso, Brasil.
[4]Doutor, Professor da Faculdade de Odontologia da Universidade Evangélica de Goiás - UniEVANGÉLICA, Anápolis, Goiás, Brasil.

Informação sobre o manuscrito
Recebido em: 15 Out 2023
Aceito em: 20 Out 2023

Autor para contato:
Orlando Aguirre Guedes
Faculdade de Odontologia, Universidade Evangélica de Goiás - UniEVANGÉLICA, Km 3,5, Cidade Universitária, 75083-515, Anápolis, Goiás, Brasil.
Tel: +55 (62) 3310-6630
E-mail: orlandoaguedes@gmail.com

ABSTRACT

Pleomorphic adenoma (PA) is the most common tumor of the salivary glands, classified as a benign mixed tumor with the involvement of epithelial, myoepithelial, and mesenchymal components. This type of tumor primarily affects the parotid glands, with a small percentage of cases involving minor salivary glands. In this study, two cases of PA with unusual locations, palate, and upper lip are presented. The clinical and histopathological characteristics of the tumors as well as the therapeutic management adopted are discussed.

KEY-WORDS: *Minor Salivary Glands; Oral Diagnosis; Pleomorphic adenoma.*

INTRODUCTION

Pleomorphic adenoma (PA) is the most common benign tumor of the salivary glands, accounting for 45-75% of all tumors affecting these structures. It is classified as a benign mixed tumor, derived from epithelial, myoepithelial, and mesenchymal elements. This type of tumor has a higher incidence in larger salivary glands compared to smaller ones, with the parotid gland being particularly predominant (85%).[1] It affects individuals of all ages and shows a predilection for the female gender. When PA occurs in the minor salivary glands, the order of prevalence is predominantly in the palate, followed by the upper lip, buccal mucosa, nasal cavity, pharynx, and larynx.[2]

In most cases, PA manifests as a firm, painless swelling with slow growth. It is palpable and has a firm consistency, is encapsulated, without ulcerations, and displays normal coloration. Additionally, it is asymptomatic in most cases. However, in rare instances,

it may undergo rapid growth, extensive ulceration, and transform into a malignant form.[3]

The histopathological characteristics of PA involve a mixed proliferation of cells. Typically, the neoplasm is well-defined by a fibrous connective tissue capsule, with parenchyma composed of ductal epithelial cells and myoepithelial cells.[1] The treatment for PA of minor salivary glands consists of conservative enucleation and surgical resections with adequate margins.[2,3] The etiology of PA is still unknown, but exposure to radiation and contact with the simian virus (SV40) may be considered important factors in the etiology of this disease.[2]

The aim of this study was to present two clinical cases of PA, one located in the palate and the other in the upper lip. The clinical and histopathological characteristics, diagnostic hypotheses, the approach to obtaining the diagnosis, and the treatment adopted are discussed.

CASE REPORT 1

A 36-year-old female patient presented to the Stomatology Service of the University of Cuiabá (UNIC) with a complaint of a "lump in the roof of the mouth" that had been present for 5 years. During the anamnesis, the patient reported no systemic alterations. The extra-oral examination did not reveal any major clinical signs of disease. The patient presented with normal facial color and with no facial asymmetries or any deviation from normality.

The intra-oral examination revealed the absence of teeth and the presence of an upper removable partial denture. A non-ulcerated rounded lesion was observed, causing mild pain symptoms. The lesion appeared more reddish than the adjacent mucosa, had a sessile base, was slightly soft upon palpation, measured 2 cm in diameter, and was located in the region of the maxillary tubercle between the hard palate and the soft palate on the left side (Figure 1A). Computed tomography imaging showed preserved bone structure without signs of lesion infiltration (Figure 1B). The diagnostic hypotheses included benign glandular neoplasia and benign mesenchymal neoplasia.

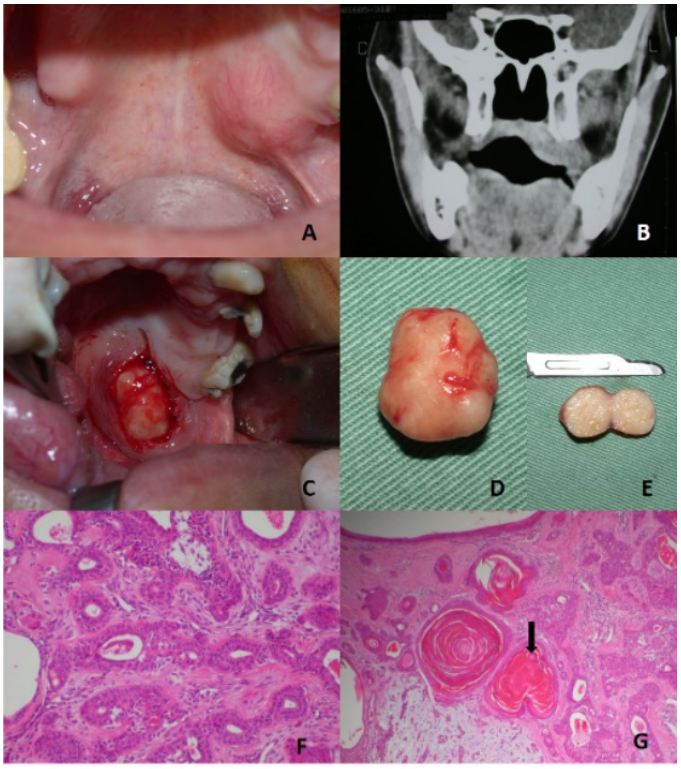

Figure 1. (A) Mass of firm consistency on the hard palate on the left side; (B) Coronal tomography showing preserved bone structure; (C) Surgical excision of the lesion adhered to the mucosa; (D) Well-defined surgical specimen with a lobulated surface; (E) The cut surface shows a solid and compact lesion with a gray-white aspect and fibroelastic consistency; (F) Histological image with epithelial and myoepithelial cells composing the glandular pattern; (G) Histological image in the lower corner with a chondroid-like mixohyaline area containing myoepithelial cells; the arrow indicates areas with metaplastic squamous epithelium without atypia, forming cysts containing concentric keratin.

An excisional biopsy was performed for diagnostic purposes, during which the entire lesion was surgically removed under local anesthesia (Mepivacaine 1: 100,000: Nova DFL, Rio de Janeiro, RJ, Brazil). In summary, an incision was made on the lesion, and a mucoperiosteal palatal flap was detached to expose and remove the neoplastic tissue (Figure 1C to D). The surgical wound was irrigated with 0.9% saline solution and sutured with absorbable thread (Vicryl 4.0, Ethycon São Paulo, SP, Brazil).

Analysis of the surgical specimen revealed a lobulated lesion with a solid, compact interior with a gray-white fibroelastic consistency (Figure 1E). The specimen was stored in a container containing 10% formalin solution and submitted for anatomopathological analysis.

Microscopic examination revealed the presence of an incomplete fibrous capsule with a proliferation of epithelial and myoepithelial elements, forming the glandular pattern (Figure 1F). In addition, a chondroid-like mixo-hyaline area containing myoepithelial cells was observed, along with areas with metaplastic squamous epithelium without atypia, forming cysts containing concentric keratin (Figure 1G). These combined features supported a conclusive diagnosis of PA.

The patient was advised about the importance of attending follow-up appointments. After three years, no signs of recurrence were observed.

CASE REPORT 2

A 34-year-old male patient presented to the Service of Stomatology of the University of Cuiabá (UNIC) complaining of "a hard lump inside the lip" that had been present for 1 year. An anamnesis was conducted to gather information about the patient's general health, revealing no systemic disease or other comorbidities. During a physical examination, a slight swelling was observed in the region described by the patient.

Upon intra-buccal inspection and palpation, a submucosal nodule was identified in the upper lip. The nodule was painless, showed no signs of ulceration, had a sessile base, and fibrous consistency, was mobile, asymptomatic, and had well-defined limits, measuring approximately 1.5 cm in diameter (Figure 2A).

An aspiration puncture was performed, and no liquid content was found in the lumen of the lesion (Figure 2B). The diagnostic hypotheses included lipoma, dermoid or epidermoid cyst, or salivary gland tumor. Subsequently, an excisional biopsy was conducted, removing a lobulated and encapsulated lesion (Figure 2C to D) that was then submitted for histopathological analysis.

In the histological section, the presence of epithelial tissue, cell proliferation, and nests with anastomoses was detected (Figure 2E). Ductiform structures and chondroid-like myoepithelioid cells containing myoepithelial cells (Figure 2F) were also observed, leading to the diagnosis of PA. The patient has been under follow-up for 2 years, and no recurrence has been detected thus far.

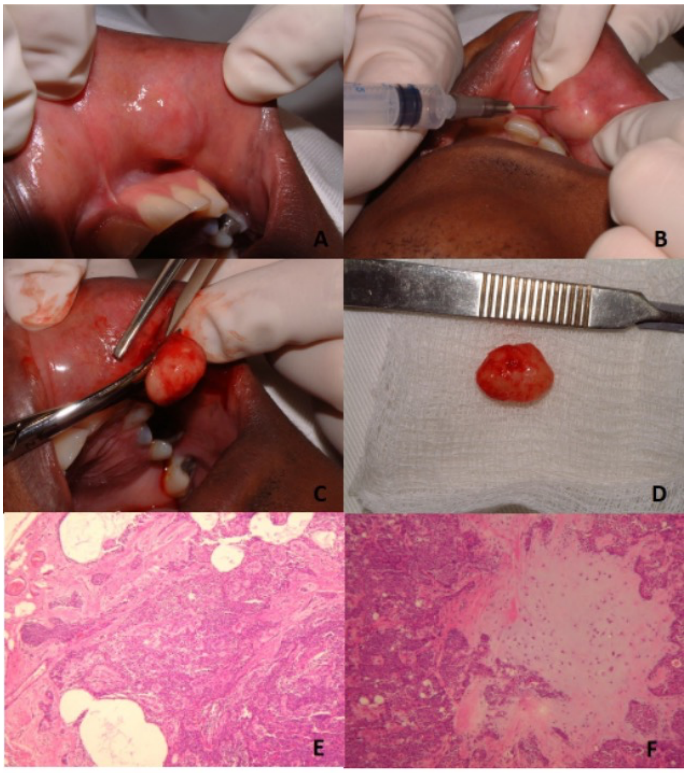

Figure 2. (A) Nodular mass of firm consistency in the upper left lip; (B) Aspiration puncture of the lumen of the lesion indicating the absence of liquid content; (C) Encapsulated lesion of firm consistency during surgical procedure; (D) Complete surgical specimen showing presence of lobes; (E) Histological image showing cord-like cell proliferation and epithelial nests with anastomoses; (F) Histological image showing ductiform structures and myoepithelioid cells with chondroid aspect containing myoepithelial cells.

DISCUSSION

Pleomorphic adenoma is a prevalent benign mixed tumor typically found in major salivary glands, with the parotids being particularly affected. However, it can

also affect minor salivary glands, albeit less commonly.[1] A study by Rahnama et al. (2013)[4] analyzed 174 cases of PA in the oral cavity, noting that the palate was the most affected region (54%), followed by the upper lip (18%), buccal mucosa (11%) and maxillary vestibule (10%).[4] Interestingly, in the present study, both cases were diagnosed in rare regions of the oral cavity, specifically the palate and the upper lip.

There is a slightly higher occurrence of PA among females,[5] as well as among individuals aged from 40 to 60 years.[6] However, the cases reported in this study involve individuals of both sexes in their third decade of life. This aligns with the findings of Chidzonga et al. (1995)[7] who observed a higher incidence of cases among patients in their third and fourth decades of life in their study with 206 patients.

Clinically, PA presents as a well-delimited swelling with slow growth, and firm consistency, and is often asymptomatic. There is usually a significant time lapse between the onset of clinical symptoms and the diagnosis.[8] In the cases described in this study, the patients sought specialized treatment 1 and 5 years after the initial appearance of the lesions, respectively. The delayed diagnosis in both cases can be attributed to limited access to healthcare facilities for these patients.

PA derives its name from the diverse histological features presented within the tumor. There are three primary components: epithelial cells, myoepithelial cells, and the stroma (mesenchymal) components.[9] However, the quantities of these cell types can vary depending on the type of tumor. The epithelial component has the potential to form various tubular and ductiform structures.[10] The stromal element typically exhibits a slightly fibrous characteristic with varying degrees of myoxoid, hyaline, cartilaginous, or bone differentiation. This tumor is encapsulated by a fibrous capsule, displaying well-defined borders, and may manifest as single or multiple nodules.[11]

The differential diagnosis of salivary gland lesions is crucial for guiding clinical decision-making. Benign and malignant salivary gland lesions often present similar clinical characteristics, especially in their early stages. Therefore, distinguishing between various lesions is essential. In the first clinical case, the differential diagnosis of the reported lesion included mucoepidermoid carcinoma, mesenchymal neoplasms, and adenocarcinoma. In the second case, potential differential diagnoses encompassed lipoma, mucocele, dermoid or epidermoid cyst, or salivary gland tumor. The consideration of differential diagnosis significantly influences patient management, aiding in determining the appropriate diagnostic exams and approaches leading to a precise final diagnosis.[12]

Various diagnostic tools and methods can be used to evaluate salivary gland lesions, including imaging tests like computed tomography, ultrasonography, magnetic resonance imaging, and X-rays. However, radiographs of lesions located on the palate may offer limited contribution due to overlapping images, which can hinder visualization. Therefore, histopathological examination remains the primary and most appropriate therapeutic management for salivary gland lesions.[13]

The treatment for PA involves the surgical removal of the lesion.[14] Adopting appropriate safety margins during the surgical procedure enhances the reliability of the diagnosis, reduces the risk of potential malignancy, and minimizes the possibility of recurrence.[15] Although cases of recurrence are rare, they can occur due to inadequate surgical removal of the lesion or enucleation during the surgery.[15] Recurrence rates are estimated to range from 2 to 44%.[16] In both cases reported here, the patients underwent surgical removal of the lesion with safety margins, and during the follow-up period, no recurrence was observed.

CONCLUSION

Minor salivary gland tumors are relatively rare, and among them, PA is the most prevalent. The development of PA in the lip and palate, although uncommon, does occur. However, it is essential to consider this neoplasm in the differential diagnosis of head and neck lesions, even in these less common locations.

RESUMO

O adenoma pleomórfico (AP) é o tumor mais comum das glândulas salivares, sendo classificado como um tumor benigno de natureza mista, com participação de componentes epiteliais, mioepiteliais e mesenquimais. Este tipo de tumor afeta primariamente as glândulas parótidas, sendo que apenas uma pequena parcela dos casos apresenta envolvimento das glândulas salivares menores. Neste estudo, são apresentados dois casos de AP com localizações atípicas: palato e lábio superior. As características clínicas e histopatológicas dos tumores, bem como os manejos terapêuticos adotados são discutidas.

PALAVRAS-CHAVE: Glândulas salivares menores; Diagnóstico bucal; Adenoma pleomórfico.

REFERÊNCIAS

1. Jain S, Hasan S, Vyas N, Shah N, Dalal S. Pleomorphic adenoma of the parotid gland: Report of a case with review of literature. Ethiop J Health Sci. 2015;25(2):189-94.
2. Geetha NT, Deepa BV, Umashankara KV, Rai Kithikumar. Pleomorphic adenoma of minor salivary gland in the cheek. International Journal of Oral Health Sciences. 2015;5(2):117-20.
3. Khan MN, Raza SS, Hussain Zaidi SA, Haq IU, Hussain AK, Nadeem MD, et al. Pleomorphic adenoma of minor salivary glands. J Ayub Med Coll Abbottabad. 2016;28(3):620-622.
4. Rahnama M, Orzędała-Koszel U, Czupkałło L, Lobacz M. Pleomorphic adenoma of the palate: a case report and review of the literature. Contemp Oncol (Pozn). 2013;17(1):103-6.
5. Waldron,S.K. El-Mofty,D.R. Gnepp. Tumors of the intraoral minor salivary glands: A demographic and histologic study of 426 cases. Oral Surg Oral Med Oral Pathol Oral Surg Oral Med Oral Pathol. 1988;66(3):323-33.
6. Laccourreye H, Laccourreye O, Cauchois R, Jouffre V, Ménard M, Brasnu D. Total conservative parotidectomy for primary benign pleomorphic adenoma of the parotid gland: a 25-year experience with 229 patients. Laryngoscope. 1994;104(12):1487-94.
7. Chidzonga MM, Lopez Perez VM, Portilla Alvarez AL. Pleomorphic adenoma of the salivary glands. Clinicopathologic study of 206 cases in Zimbabwe. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 1995;79(6):747-9.
8. Al-Khtoum N, Qubilat AR, Al-Zaidaneen S, Al Mefleh S, Al-Qudah A. Clinical characteristics of pleomorphic adenoma of salivary glands among Jordanian patients. J Pak Med Assoc. 2013;63(3):358-60.
9. Bobati SS, Patil BV, Dombale VD. Histopathological study of salivary gland tumors. J Oral Maxillofac Pathol. 2017;21(1):46-50.
10. Bijai LK, Mathew P, Rajathi M, Austin RD. Recurrent pleomorphic adenoma: A case report. International Journal of Dental Sciences and Research. 2014;2:92-94.
11. Lopes ML, Barroso KM, Henriques ÁC, Dos Santos JN, Martins MD, de Souza LB. Pleomorphic adenomas of the salivary glands: retrospective multicentric study of 130 cases with emphasis on histopathological features. Eur Arch Otorhinolaryngol. 2017;274(1):543-551.
12. Cerulli G, Renzi G, Perugini M, Becelli R. Differential diagnosis between adenoid cystic carcinoma and pleomorphic adenoma of the minor salivary glands of palate. J Craniofac Surg. 2004;15(6):1056-60.