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A Rare Pleomorphic Adenoma in an Uncommon Area: A Case Report

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ABSTRACT

The Pleomorphic Adenoma is the most frequent benign tumor of the salivary glands. The cheeks, lips and gingiva are rare sites of appearance. The objective of this study is to report a case of a Pleomorphic Adenoma arising from the buccal mucosa. A 57-year-old female patient who presented with an increase in volume in the right facial region with one year of evolution, asymptomatic and without treatment. Physical examination revealed a nodular lesion of approximately 3 cm in diameter, located in the right cheek. MRI revealed a hyperintense image in STIR sequence. Excisional biopsy of the lesion was made under balanced general anesthesia, where extracapsular dissection of the tumor was performed, using a transoral approach. The histopathological study resulted in pleomorphic adenoma. Despite the low rate of recurrence of this neoplasm, short, medium and long term follow-ups are recommended.

1. Introduction

The pleomorphic adenoma (PA) is the most frequent benign tumor of the salivary glands.[1] About 60% - 90% of PA occur in the parotid gland, 40% - 60% in the submandibular gland[1, 2] and 7% - 10% in minor salivary glands.[3] Intraorally, the minor glands of the palate are the most affected, with a prevalence of 43% to 70%. The cheeks, lips and gingiva are rare sites of appearance.[3, 4] PA is a neoplastic proliferation of glandular parenchymal cells with an associated myoepithelial component[5] that usually appears as a solitary, painless mass on the oral mucosa.[3] It varies in consistency from soft and fluctuant to firm and rubbery, depending on the presence of cystic or mucoid degeneration or the formation of chondroid or osteoid tissues.[6] The diagnosis of PA is established based on a complete history, physical examination and histopathological analysis.[7, 8] Other advanced imaging techniques like computed tomography (CT), magnetic resonance (MRI) and ultrasound (USG) provide information regarding location, size and extension of the tumor to surrounding structures.[8] The objective of this study is to report a case of pleomorphic adenoma from the buccal mucosa.

2. Case presentation

A 57-year-old female patient who attended to the Oral and Maxillofacial Surgery Unit of the University Hospital of Maracaibo, Venezuela, presenting an asymptomatic mass in the right facial region with approximately one year of evolution, without any signs of facial asymmetry. Her medical history showed arterial hypertension and nephrolithiasis. Physical examination revealed a nodular lesion of roughly 3 cm in diameter, firm, asymptomatic, mobile and located in the right cheek, without involving the parotid duct. Sensory and motor functions were unchanged. The overlying skin and deep layer of mucosa were unaffected, and no cervical adenopathies were palpated. The imaging study using a contrasted magnetic resonance imaging (MRI) revealed a hyperintense image in short-tau inversion recovery (STIR) sequence with fat suppression, located in the right buccal region, anterior to the ipsilateral masseter muscle, without the involvement of the Stensen's duct (Figure. 1).

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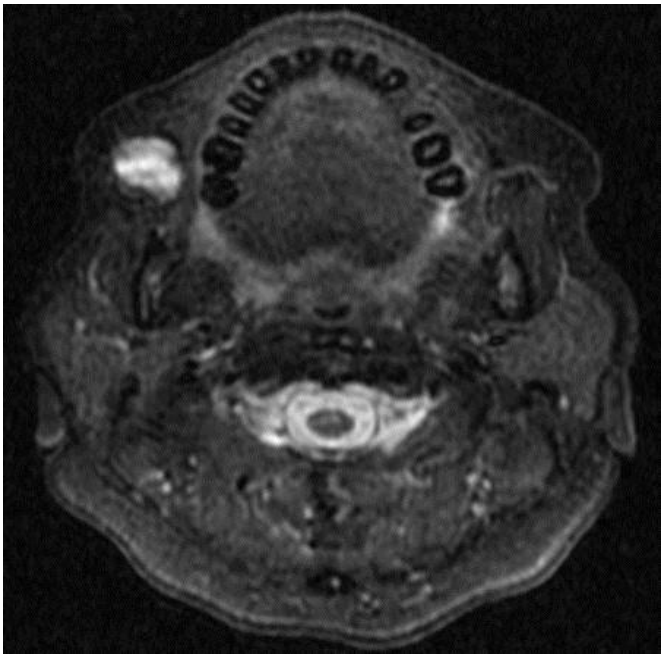

Figure 1: Contrasted Magnetic Resonance in STIR sequence showing a well-defined homogeneous lesion in the right buccal space region.

Excisional biopsy of the lesion was made under general anesthesia, where extracapsular dissection of the tumor was performed, using a transoral approach. During surgery, it was evident that the tumor was not associated with the Stensen's duct and, thus, an accessory parotid gland tumor was ruled out (Figure. 2).

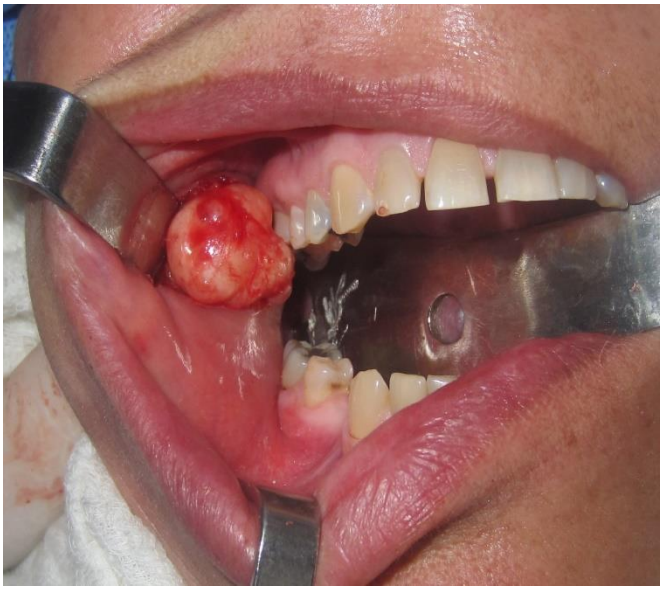

Figure 2: Intraoperative image of mass in the right cheek.

Macroscopically, a nodular light brown structure of soft tissue measuring 2.5 x 2.1 cm, was observed. When cut, the nodule was solid, firm and White (Figure. 3).

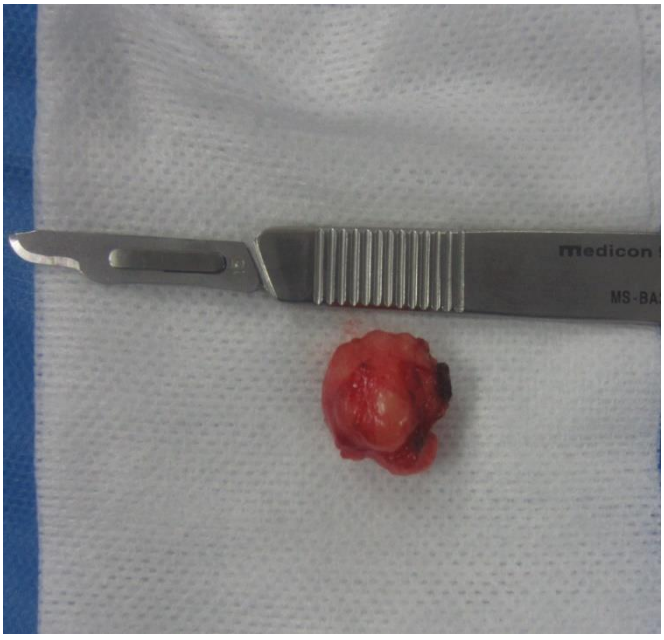

Figure 3: Gross specimen with intact soft tissue encapsulation.

The histological sections showed neoplasia of epithelial origin, surrounded by a dense fibrous connective tissue capsule, with stroma containing hyper and hypocellular areas, hyalinized in some areas, myxoid in others. Small and rounded epithelial cells and myoepithelial cells were identified. The cellular component was organized in structures that resemble glandular ducts. The histopathological study resulted in PA (Figure. 4).

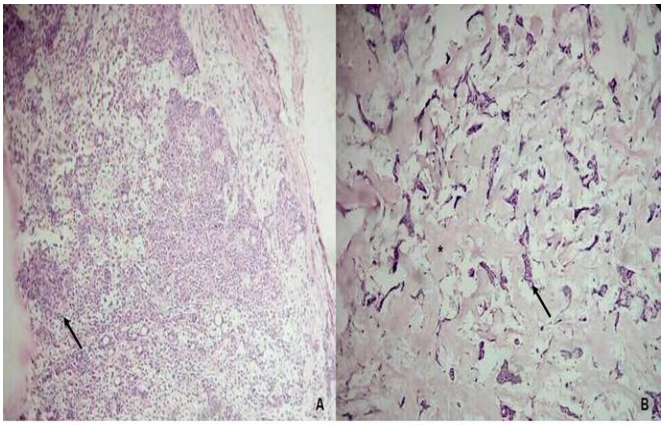

Figure 4: Histopathological examination on hematoxylin and eosin (H&E) staining. Microscope magnification A. 40x B. 100x. Neoplasia of epithelial origin (black arrows), surrounded by a dense fibrous connective tissue capsule with stroma containing hyalinized areas (asterisk).

3. Discussion

Tumors that appear in the minor salivary glands represent 22% of all salivary gland neoplasms. Most are malignant, and only 18% are benign.[9] Of the benign tumors, the PA is the most common lesion. The site with the highest prevalence within the oral cavity is the palate, followed by the lips, cheek mucosa, the floor of the mouth, tongue, amygdala, pharynx, retromolar area and nasal cavity.[4] PA frequently occurs in people of middle age; between the 4th and 6th decades of life; and more commonly in the female gender,[4, 10] as in our case.

The evaluation of a mass in the cheek can be challenging, in particular, the anatomical region that is bound posteriorly by the anterior edge of the masseter, cephalad by the zygoma, anteriorly by the modiolus and caudal by the edge of the mandible. The diversity of progenitor cells found in the region make the differential diagnoses very wide, given that, from the intraoral mucosa to the dermis, different elements can be found, such as mucosa, submucosa, minor salivary glands, the buccinator muscle, neurovascular components, the buccal fat pad, the superficial musculoaponeurotic system, subcutaneous fat and dermis.[11] Differential diagnoses for a mass in the cheek include major and minor salivary gland tumors (MSGT), tumors of an accessory parotid gland, lipomas, myofibromas, neurofibromas, sebaceous cysts, epidermoid cysts, dermoid cysts, mucoepidermoid carcinoma, and adenoid cystic carcinoma.[12]

In the evaluation of salivary gland neoplasms, imaging methods such as ultrasound, CT, or MRI may be used depending on the site and size of the tumor. On ultrasound, PA appears typically as a hypoechoic, homogenous, well-circumscribed mass with posterior acoustic enhancement. On CT, it usually appears as a well-circumscribed mass of soft-tissue density that shows either heterogeneous or homogeneous contrast enhancement. On MRI, it has an intermediate or low T1 signal and a more variable T2-weighted signal of which the intensity varies from high in cellular areas to very high (higher than cerebrospinal fluid) in myxoid areas.[13] MRI is the imaging modality of choice for neoplasms in this region, due to its accuracy to evaluate soft tissues, for this reason, it is the most commonly used imaging study for neoplasms suspected in minor salivary glands,[11] as it was in our case.

Several studies have depicted varied frequency of occurrence of PA of minor salivary glands of the cheek.[4, 6, 14, 15, 16, 17, 18 – 25] (Table 1) In a review of 24 cases of PA in minor salivary glands, 75% occurred in the palate and 17% in the cheek[4] In another study of 149 cases reported in minor salivary glands, 19 were present in the cheek, representing only 13%.[18] As it was observed, the frequency is very low to these neoplasms in minor salivary glands; however, these results are not comparable with those of our study, because only one case is being reported.

Table 1: A literature review of the frequency of pleomorphic adenoma of minor salivary glands of the cheek.

Studies	Country	Total number of pleomorphic adenoma cases	Number of cases in cheek	Percentage (%) of cases in cheek
Isacsson and Shear[14]	South Africa	140	7	5
Fine et al.[15]	United State	25	4	16
Chaudhry et al.[6]	United State	476	38	8
Main et al.[16]	Scotland and Canada	31	5	16
Cohen.[17]	South Africa	144	10	7
Bablani et al.[4]	India	24	4	17
Buchner et al.[18]	United State	149	19	13
Lopes et al.[19]	Brazil	65	0	0
Yih et al.[20]	United State	93	16	17
Hayashi et al.[21]	Japan	54	6	11
Pérez de Oliveira et al.[22]	Brazil	20	6	30
Toida et al.[23]	Japan	54	6	11
Rivera et al.[24]	Venezuela	24	2	8

PA of the cheek usually presents as a mobile, slowly growing, the painless, firm, lobulated submucosal mass that does not cause ulceration of the overlying mucosa[6] as presented in our case. The size of these tumors varies within a range of 1 to 4 cm, which coincides with the case reported, and they have a recurrence rate of 6%.[4] The typical characteristics of the lesion and its location were key signs of a benign tumor of minor salivary glands, and thus, it was resected intact with its capsule.

4. Conclusion

The varied presentation of tumors of the minor salivary glands makes the diagnosis challenging even for an experienced surgeon and pathologist. PA of the cheek is a rare neoplasm and should be considered in the differential diagnosis of intraoral swellings of the buccal mucosa. Complete wide local surgical excision is the treatment of choice. Despite the low rate of recurrence of this neoplasm, short, medium and long term follow-up is recommended.

Conflict of Interest

The authors declared that there is no conflict of interest.

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