

Surgical Management of Palatal Pleomorphic Adenoma (PPA) Recurrence After 10 years, Treated at a Brazilian Center - A Case Report

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Abstract

Pleomorphic adenoma, considered the most frequent benign mixed neoplasm of the minor salivary glands, occurs mainly in the region of the hard palate, with slight predilection in females and peak of incidence between the third and fifth decades of life. An increase in recurrence rates has been associated with the histopathological variants of the tumor, cellular characteristics, stroma, and capsule rupture during surgical removal of the lesion. The present case report aims to describe the surgical approach performed on the patient, a 45-year-old woman with a recurrent Pleomorphic Adenoma (PA) in the region of the hard palate on the right side, 10 years after initial enucleation of the lesion; her main complaint was an increase in volume in the palatal region. After extensive local surgical excision of the tumor and 2 years of follow-up, there were no signs of recurrence. Computed tomography and a correct histopathological diagnosis are essential to enable the establishment of an appropriate surgical treatment, with the purpose of achieving complete removal of the lesion, with wide surgical margins, including the lining mucosa and the underlying periosteum, as described in the present case.

Keywords: Hard palate, local neoplasm, pleomorphic adenoma, recurrence, salivary gland neoplasm, surgical pathology

INTRODUCTION

Pleomorphic adenoma (PA) is the most common benign tumor of salivary glands of the head and neck region, accounting for about 40%–70% of all major and minor salivary gland tumors. It usually presents as a slow-growing submucosal mass in the hard palate when minor glands are affected. The palate is the most common intraoral site of PA, followed by the upper lip, buccal mucosa, floor of the mouth, tongue, tonsil, and retromolar regions.^[1-5]

The majority of minor salivary gland PAs occur in the second decade of life,^[1] but other epidemiological data^[3,6] have shown a higher prevalence in the fourth and sixth decades, with slight predilection in the female gender.^[1,3,6]

Clinically, it presents as a painless, immobile nodular lesion of firm consistency that is slow-growing, covered by healthy mucosa and of normal color.^[3,6-8] In few cases, mainly related to the parotid gland, it could grow more rapidly.^[9] Moreover, while PAs affect the palatal region, their clinical presentation is

typically characterized by a firm or rubbery submucosal mass without ulceration or surrounding inflammation.^[1]

These lesions can affect the underlying bone and may extend to the maxillary sinus.^[10] Thus, to aid the clinical examination, computed tomography (CT) is the gold standard imaginological assessment required, as it delimits the region affected by the lesion and reduces the chances of recurrence or postoperative complications.

Histologically, the lesion presents a well-delimited neoplasm with a fibrous connective tissue capsule, consisting of cells with

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ductal and myoepithelial elements.^[6,9,11] In addition, myxoid, ductiform, hyaline, plasmacytoid, osteoid, and chondroid areas are found, which is why the term benign mixed tumor has been applied to it.^[4,9,12,13]

Classically, the most reported form of treatment in the literature is wide local excision with the removal of the periosteum or bone if these tissues are involved. Simple enucleation procedures, which may or may not remove covering tissue, are associated with high local recurrence of the tumor and should be avoided.^[1,5,9]

The aim of this brief report was to describe our experience with surgical management of a recurrent palatal PA in a female patient and to compare our clinical results with those of others found in the contemporary literature review.

CASE REPORT

The patient, a 46-year-old woman, was referred to the outpatient clinic of our Division of the Oral and Maxillofacial Surgery Department at a state hospital in Sao Paulo, Brazil, to have several complaints addressed. Among these was swelling, pain in the hard palate, making it difficult to adapt her dental prosthesis correctly in the region where a surgical procedure for cyst removal had been performed 10 years previously.

An extra-oral examination showed no facial changes. Relative to the intraoral assessment, an exophytic lesion with a nodular aspect was found, which had a firm consistency, was located in the anterolateral portion of the hard palate, fixed on the right side, measuring approximately 2.2 cm × 3 cm. It was slow-growing, and the patient reported no significant pain complaints about it. CT for soft and hard tissues was performed with the purpose of delimiting the local invasiveness of tumor that showed no bone breakdown or erosions in palatal bone, as visualized in Figure 1.

Histopathological assessment was made after an incisional biopsy, which confirmed our initial diagnosis of PA. Thus, the surgical planning consisted of the complete removal of the tumor by wide surgical excision involving the periosteum and surrounding mucosa.

The surgical technique was started with regional antisepsis using chlorhexidine 0, 12%, and 2% for purposes of intra- and extra-oral decontamination, respectively. Local anesthesia was performed in conjunction with conscious sedation for better patient comfort. Moreover, a rhomboidal incision surrounding the lesion, up to the bone level, was made with an electric scalpel and the divulsion of the periosteum was performed using a molt periosteal elevator number 9. The lesion was detached and removed immediately, as shown in Figure 2.

Indeed, the surgical specimen was fixed in a 10% formaldehyde solution and sent for definitive histopathological examination by the oral pathology laboratory. Finally, the wound was allowed to granulate and heal by itself, and the patient's

obturator prosthesis was repackaged with silicone material to avoid further contamination of the surgical site and diminish painful symptoms. Corticosteroid and analgesic medications were recommended for use in the following 5 days.

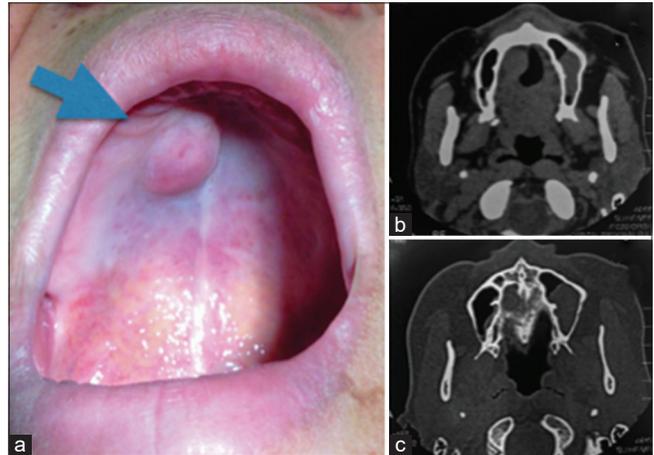


Figure 1: (a) Intraoral examination showed well-delimited swelling in the anterior region of the hard palate. (b) In computed tomography scan, generating images in axial section, size, and extension of lesions associated with surrounding tissues may be visualized. (c) Relative to hard tissues, intact hard palate bone may be observed in the axial section of computed tomography

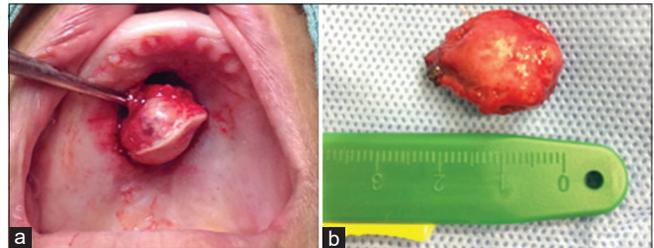


Figure 2: (a) Wide local excision for removal of the tumor in the hard palate, which included underlying mucosal-periosteal tissue, (b) Macroscopically, well-delineated submucosal mass covered with normal mucous membrane may be visualized, corresponding to the surgical specimen

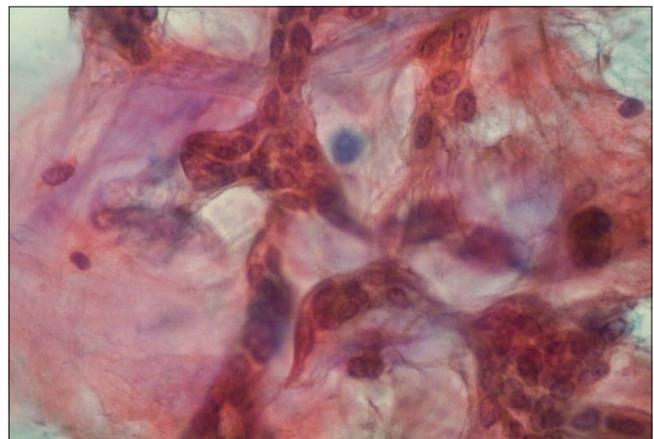


Figure 3: Epithelial cells arranged in duct-form or cord-like cell patterns, along with areas of epidermoid metaplasia

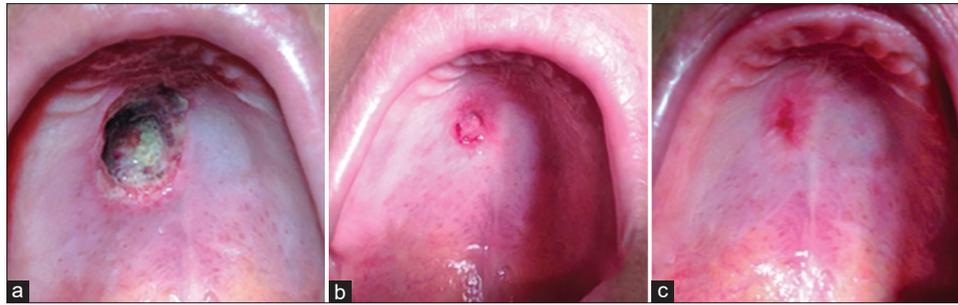


Figure 4: Follow-up after (a) 7 days, (b) 4 weeks, and (c) Almost 2 months after surgical procedure

Histopathology with hematoxylin and eosin staining [Figure 3] showed the presence of proliferation of cells such as fusiform/plasmacytoid myoepithelial tumor-type cells. The neoplastic parenchyma was made up of epithelial cells arranged in cord-like and duct-like cell patterns. These were distributed in small stroma mixtures that confirmed the characteristics of a benign PA.

The patient was followed up weekly for the first 4 weeks. During this period, complaints of painful symptoms were recorded in the initial 15 days [Figure 4]. Moreover, the patient has been followed up clinically and radiographically for 1 year and 6 months (October 2016 to April 2018) without signs of recurrence.

DISCUSSION

PA is one of the common benign salivary neoplasms, affecting major^[13] and minor salivary glands.^[9,12] According to Waldron *et al.*, 1988,^[10] relative to their study with 466 cases, 174 cases of these neoplasms were located in minor salivary glands. In the majority of cases, the hard palate was affected in almost 54%, as found in other studies.^[8,9] In recent studies, authors found high injury rates in the hard palate region, ranging from 40% to 70% of the total number of cases.^[1,12]

The diagnosis of PPA is based on the medical history reported by the patient, intra- and extra-oral clinical examination, with the main clinical sign being an increase in volume in the region of the hard palate, as shown in this case report. CT has become an essential tool for detecting the exact location, size, and extent of the lesion, and in the contemporary literature,^[1,14] it is considered an image examination superior to MRI with reference to the tumor characteristics mentioned above. The definitive diagnosis will be determined by histopathological analysis.^[14]

A retrospective study^[2] evaluated the clinicopathological characteristics of 74 patients with PPA and determined that of this group, 40 (84%) of the adenomas were partially encapsulated and that they had a larger amount of plasmacytoid myoepithelial tumor cells (50 cases). These histopathological characteristics were found in this case report after histological examination and could probably be associated with the recurrence shown in our patient 10 years after the initial surgical removal of the tumor.

PA, the most common benign neoplasm in minor salivary glands, has a predilection for female patients as has been shown by recent studies.^[14,15] Patigaroo *et al.*,^[1] however, reported predominance of palatal PA in males over females, and the most common age group was 16–30 years. This present case report showed a single case of a 45-year-old female patient with recurrence of a PPA that had been submitted to surgical removal 10 years previously. This was consistent with the relevant literature that showed a peak of incidence between the third and fifth decades of life.^[14,15]

The surgical technique is a relevant factor in order to avoid the recurrence of tumors. Indeed, simple enucleation is associated with clinical failure and local recurrence. For this reason, it should be avoided,^[1,14,16] whereas a conservative wide local surgical excision, instead of a resective surgical approach, would be the treatment of choice that involves complete removal of mucosal-periosteal tissue surrounding the lesion, or the curettage of bone affected, as described in the retrospective study.^[14] Bone removal is usually not necessary if the PA has not invaded any bone tissue.^[1,6,9,12]

In cases of erosions detected in the hard palate by means of CT that indicated a wide destruction and/or rupture of the floor of the nasal cavity and/or paranasal sinuses, the best alternative would be the removal of tumor in conjunction with the compromised bone, with enlarged margins, as described in the literature.^[1,14,16] For more advanced cases, partial maxillectomy has been described as the main approach, followed by rehabilitation with obturator prostheses.^[17]

Recurrences usually appear due to an inadequate surgical technique, as previously described, which could happen in a long-term period of follow-up, after a period of 2 years onward after the surgical procedure. However, other previous studies^[9,14] have detected recurrences between 7 and 10 years after initial surgery, as shown in this case report, as already mentioned.

Multiple recurrences of PPA, which take a longer time to develop, are associated with a higher probability of these becoming a carcinoma ex-PA (ca ex PA), which is an aggressive malignancy that can lead to death in most cases.^[9,10,12-14] Histopathological findings and other stroma alterations have been associated with malignant transformation such as focal necrosis, extensive hyalinization, vascular and capsular invasion, and atypical mitosis; however, these features are

rarely seen in these tumors as shown in this case report and by other recent retrospective studies.^[15]

Recurrence rates vary from 0% to 14%, and the majority were shown in cases in which the surgeon opted for an extremely conservative excision (simple enucleation) only, without removing a safety margin, and often with rupture of the lesion capsule, so that the tumor achieved greater capacity for spreading into the adjacent tissues.^[1,13]

CONCLUSION

PPA is the most frequent benign neoplasm in the minor salivary glands, and its recurrence may occur after a prolonged period of follow-up of the lesion, due to its wide spectrum of cellular and histopathological variants or an inappropriate surgical approach. A correct surgical technique that involves the complete removal of the lesion, margins involved, including the lining mucosa and the underlying periosteum has shown a reduction in the rates of local tumor recurrence. Nevertheless, an accurate clinical, tomographic, and histopathological evaluation will be essential for the establishment of an adequate surgical treatment plan. A secondary healing process with obturator prostheses showed positive results in the present case after a considerable period of clinical follow-up.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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