

Surgical approach to pleomorphic adenomas arising in the palate: a 10-year retrospective study in a Brazilian population

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ABSTRACT:

Salivary gland tumors represent an important group of neoplasms characterized by a great diversity of types and morphological variants. Pleomorphic adenoma is the most common benign tumor occurring in the major and minor salivary glands. The majority of case series described in the literature refers to epidemiological data of major and minor salivary gland tumors, generally being of a benign or malignant nature. Studies reporting exclusively surgical treatment experience with pleomorphic adenomas of the palate are scarce. Therefore, the aim of this study was to report data on the treatment experience with surgical removal of pleomorphic adenoma of the palate together with to nearby mucosal lining with or without ostectomy. Clinical records of all patients diagnosed with pleomorphic adenoma of the palate seen at Brazilian public service dental clinic (Oral and Maxillofacial Surgery Division) and at a private clinic between March 1999 and November 2010 were reviewed. The analysis of the biopsied specimens yielded 10 cases during the 10-year period. Of all the patients 8 were women, with mean age 39 years. The most common main complaint was asymptomatic increase in volume (n = 9), with mean tumor size 2.1 cm. Most patients (n = 8) underwent excision of tumor and mucosal lining. One patient underwent bone ostectomy and in another, the tumor regressed after incisional biopsy. The mean follow-up was 26,8 months (12-42 months) with no recurrences, and the main complication was local discomfort and dysphonia (n = 2). In summary, surgical excision of the tumor and mucosal lining, with or without ostectomy, was shown to be an effective treatment modality for pleomorphic adenomas of the palate.

Keywords: epidemiology, oral pathology; salivary gland neoplasms.

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INTRODUCTION

Salivary gland tumors represent an important group of neoplasms characterized by a great diversity of types and morphological variants¹. These conditions are relatively uncommon representing approximately 3% to 10% of head and neck tumors².

Pleomorphic adenoma (PA) is the most common benign tumor of the major and minor salivary glands³. The etiology of these tumors is associated with components of the salivary intercalated duct, whose cells with pluripotency to differentiate into various morphological patterns give rise to myoepithelial cells⁴. Although PA can affect patients of both sexes and all ages, it is more prevalent in women⁴ and individuals between the 4th and 6th decades of life⁵. PA is rarely seen in children and adolescents⁶. The site mainly affected when it reaches the minor salivary glands, is the palate⁷. When it involves this region, it presents clinically as a firm, slowly and painless growing swelling, covered with mucosa of normal appearance, rarely reaching large dimensions⁸.

Most case series described in the literature refer to epidemiological data from major and minor salivary gland tumors, generally being of a benign or malignant nature. Studies reporting exclusively surgical treatment experience with pleomorphic adenomas of the palate are scarce. Therefore, the objective is to discuss the results obtained by surgical removal of these lesions and the mucosal lining, either associated with bone wall ostectomy with a drill, or not.

MATERIAL AND METHODS

In this study clinical records of all patients diagnosed with PA were included, according to standardized histopathological features, all located in the palate and treated at a Brazilian public service dental clinic (Oral and Maxillofacial Surgery) and at a private clinic from March 1999 to November 2010. The cases in other anatomical regions, or those with incomplete records, were excluded. The records of patients reporting a history of systemic diseases or drug usage that could impair the healing process, or increase the risk of infection were also removed from the sample. Figure 1 shows the algorithm for the final sample composition with inclusion/exclusion criteria.

The researchers retrieved data included in this retrospective study after reviewing all the information from the records of the two services. Three researchers were responsible for data collection from public service, whereas two researchers collected the data from the private service. The following information was collected: sex, age, location, chief complaint, size, duration, treatment modality, follow-up, recurrence rate and

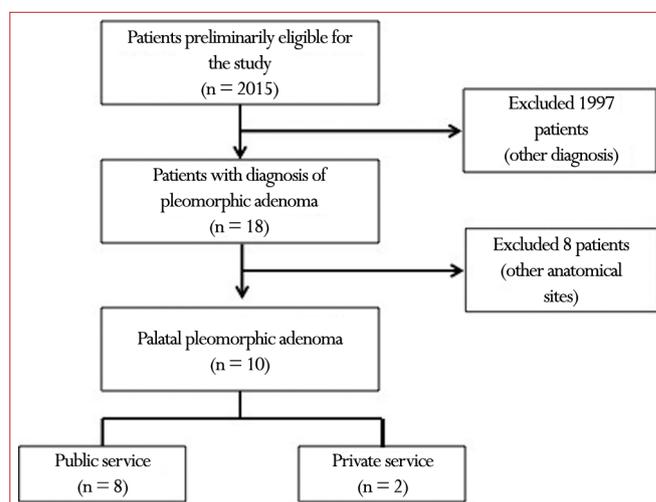


Figure 1. Algorithm for the final sample composition.

postoperative complications. As regards location, lesions in the hard or soft palate were considered, where they were present to the greatest extent.

All patients had initially undergone an incisional biopsy under local anesthesia, for histopathologic confirmation of the diagnosis of pleomorphic. With the exception of cases 6 and 7, all patients underwent the following treatment sequence: impression taking in order to fabricate an acrylic splint, which served to protect the surgical site during the postoperative period of 14 days. The technique involved the surgical excision of the lesion together with the mucosal lining, plus a small safety margin of the adjacent mucosa 5 mm in length (Figure 2). In case 6 a plus of 2 mm in length of peripheral ostectomy of the remaining bone wall was performed with a surgical drill (Figure 3). Case 7 did not require further treatment because when the patient returned later to receive the histopathology result, the lesion had involuted completely. For cases 2 and 9, the acrylic plate was not manufactured because the lesion was predominantly in the soft palate.

RESULTS

From March 1999 to November 2010, a total of 2015 biopsies were performed in the two services: 1852 in a public, and 163 in a private service. The benign and the malignant neoplasms of minor salivary glands added up 34 cases (24 benign 10 malignant), representing 1.68% of cases. Of the 24 benign lesions, 18 (75%) were diagnosed as PA, with 10 (55.5%) of them involving the palate (Table 1).

Of the ten cases in the palate, 8 (80%) occurred in women. The mean age was 39 years, in the age-range from 25 to 58 years. Most PA (80%) involved only the hard palate, with only

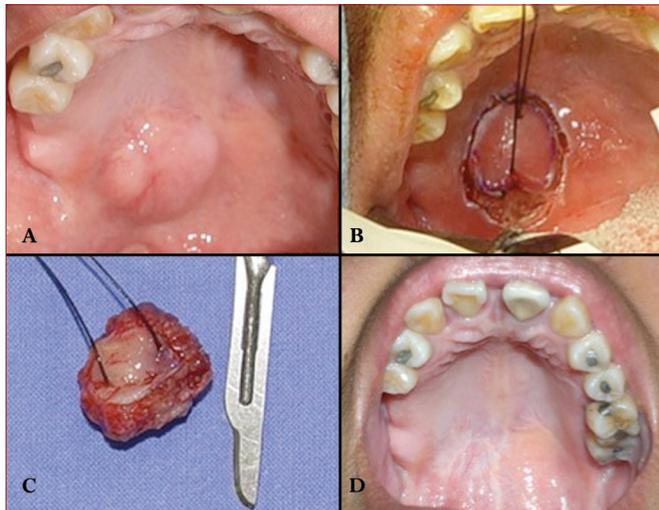


Figure 2. A: Clinical appearance; B, C: Surgical excision including the mucosal lining, in addition to a small safety margin of the adjacent mucosa; D: Follow-up period of 3 years and 6 months.

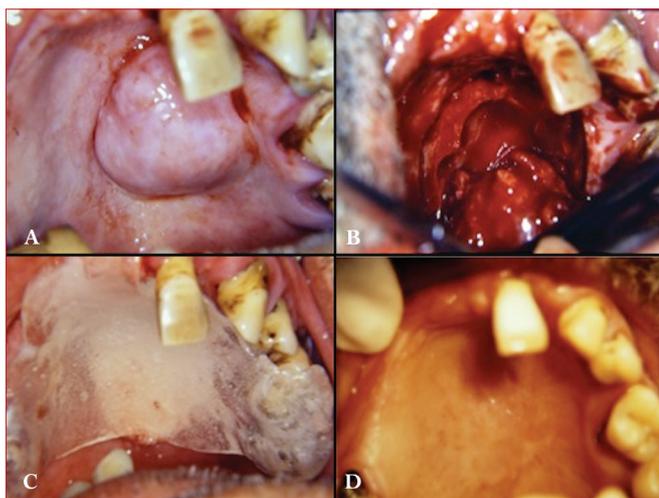


Figure 3. A: Clinical appearance; B: Peripheral osteotomy after surgical excision; C: Removable acrylic appliance on surgical site; D: Follow-up period of 12 months.

20% of cases affecting the soft palate. The main complaint was a painless swelling (n = 9); only in Case 9 the patient reported that pain was the reason that led him to seek treatment. The mean size found was 2.1 cm (1.5 to 3.5 cm). The average time of lesion development before the patient sought care was 9.6 months, ranging from 2 to 18 months (Table 1).

The mean follow-up period was 26,8 months (12-42 months) Recurrence was detected in no case up to the time this work was concluded. The most prevalent postoperative complications were local discomfort and dysphonia (Table 2).

DISCUSSION

There are mucous salivary glands distributed throughout the oral cavity in greater concentration in the palate and lips⁵. The findings are in agreement with those of several studies that have pointed a percentage of PA of benign minor salivary gland neoplasms very close to 75% found in the present study^{1,2,9-16}. In addition, of the 18 cases found in this study, 10 occurred in the palate, demonstrating its high incidence in this location, as supported by several authors^{9,11,12,15}.

The sample included 80% of PA in women and 20% in men. The male:female ratio to the order of 1:1.25¹², and 1:1.0⁸. that has been presented by other authors confirmed the gender predilection of the lesion, described in the literature, however the ratio found was high (1:4). The small number of cases in the present sample may explain that difference. It is important to note that there are articles in which men were more affected, such as that of Chau, Radden¹⁰ who retrospectively assessed 53 patients affected by PA of 98 cases of neoplasms of the salivary glands, found a higher prevalence for males (57%). Likewise, Jones et al.¹³ found a slight predilection for males.

In the present study, the age ranged from 25 to 58 years (mean 39 years), with prevalence of the fourth decade of life. The mean age for males was 55 years (52-58 years) while for females was 35 years (25-47 years). In contrast with these findings,

Table 1. Distribution of patients according to sex, age, clinical features, and course of PA.

Patient	Sex	Age (years)	Site	Clinical features			Course
				Size (cm)	Swelling	Pain	
Case 1	Female	36	Hard palate	3	Yes	No	2 months
Case 2	Female	31	Soft palate	3.5	Yes	No	8 months
Case 3	Female	47	Hard palate	1.5	Yes	No	5 months
Case 4	Female	25	Hard palate	2.0	Yes	No	15 days
Case 5	Male	58	Hard palate	2.0	Yes	No	8 months
Case 6	Female	43	Hard palate	2.0	Yes	No	9 months
Case 7	Female	34	Hard palate	2.0	Yes	No	4 months
Case 8	Female	27	Hard palate	1.5	Yes	No	Not informed
Case 9	Male	53	Soft palate	2.5	Yes	Yes	18 months
Case 10	Female	37	Hard palate	1.5	Yes	No	6 months

Table 2. Distribution of patients according to surgical approach.

Patient	Treatment			Postoperative complications	Follow-up	Recurrence
	Enucleation	Bone wall ostectomy	Provisional prosthesis			
Case 1	Yes	No	Yes	Local discomfort	3 years 6 months	No
Case 2	Yes	No	No	Local discomfort	2 years	No
Case 3	Yes	No	Yes	Local discomfort	1 year	No
Case 4	Yes	No	Yes	Local discomfort	2 years 10 months	No
Case 5	Yes	No	Yes	Local discomfort	3 years	No
Case 6	Yes	Yes	Yes	Local discomfort	1 year	No
Case 7	No*	No	Yes	Local discomfort	1 year 9 months	No
Case 8	Yes	No	Yes	Local discomfort	2 years 8 months	No
Case 9	Yes	No	No	Dysphonia	3 years 1 month	No
Case 10	Yes	No	Yes	Local discomfort	1 year 6 months	No

* Incisional biopsy

Isacson, Shear⁹ studying 140 cases of PA observed a peak age in the 3rd decade of life, with an average age for men of 37 years and for women of 41 years, which differs from the findings of the present study. Jaber¹⁵ who observed a higher prevalence of PA in the fourth decade of life, found similar results.

The main complaints reported were swelling, slow growing and painless lesions (8 cases) and pain as a result of trauma (2 cases). In addition to these, the literature presents other possible signs and symptoms such as radiographic changes, ulcers, difficulty with normal oral functions and misfitting dentures^{5,10,14,15}.

The fact that the lesion had developed during an average period of 9.6 months before being recognized, and reached an average size of 2.1 cm, can be explained by its the silent nature of its growth^{2,15}. Ansari² and Jaber¹⁵ found that most patients seek professional help in the first 12 months, as observed in this study. Lomeu, Finneman⁶ reported the case of a young patient in whom its clinical course lasted five years.

All cases were first submitted to an incisional biopsy. Pires et al.¹⁶ through a clinicopathological study, reported that 143 of the 181 cases of PA of minor salivary glands were diagnosed by excisional biopsy. Although these authors did not specify in which locations this treatment had been established, it is a questionable procedure because the clinical appearance of benign and malignant tumors is very similar at this anatomical site, as described by Loyola et al.¹² and Pogrel⁵. They consider it essential to perform the biopsy prior to definitive treatment of palatal lesions, because surgical treatment in this location shows considerable variation depending on the diagnosis.

Enucleation has been considered an appropriate form of treatment for PA, although Pogrel⁵ considers the palatal region the site of the most aggressive type of pleomorphic adenoma. This author points out that while considered a benign lesion, it is poorly encapsulated, which leads to a tendency to recur after inadequate resection.

Extracapsular excision, including the mucosal lining and palatal periosteum was performed in eight of the 10 cases. In case 6, this treatment was extended by peripheral ostectomy, by virtue of an unencapsulated lobular growth in contact with the bone wall having been detected intraoperatively. This aggression in the initial therapy is consistent with the principles recommended by Sacks et al.¹⁷ and Pogrel⁵. The latter author justifies such rigor in order to prevent the recurrent tumor from reaching the palatine foramen and from there reaching the base of the skull. Lim et al.¹⁴ reported a case of recurrent PA that caused the patient's death because of extensive local involvement.

The treatment of the patient in Case 7 consisted of incisional biopsy only, because the lesion resolved itself completely after completion of the biopsy and it was not perceived in the reassessment after 21 months. Although rare, spontaneous remission after incisional biopsy has been published. Chen et al.¹⁸ reported a case of a 28-year-old man with palatal PA that experienced an extensive necrosis three weeks after incisional biopsy. According to these authors, the etiopathogenesis of this phenomenon is still uncertain. It has been speculated that large tumors may compress the palatine arteries, producing tumor necrosis¹⁹. However, in the present study, the case presented as a small mass. Therefore, a precise etiology was not established in this case.

The most frequently reported postoperative complications were local discomfort and dysphonia. The discomfort was relieved by the prefabricated acrylic splint, which was made for 80% of the patients. Dysphonia occurred only in Case 9 due to the greater extent of the lesion in the soft palate. Both complications were transient and involuted completely in a period of 15 to 60 days. During a mean follow-up period of 26,8 months (12-42 months), in no case was recurrence detected during the study period. The monitoring of all cases will continue in the long term especially in Case 7, in which involution occurred after the incisional biopsy.

CONCLUSION

In summary, the clinical characteristics of cases of PAs included in the present study suggest the imposition of definitive treatment of lesions located mainly in the proximity of the palatine foramina. Under the conditions of this study, surgical excision of the tumor and the mucosal lining, either in addition to ostectomy, or not, was shown to be an effective treatment modality for pleomorphic adenomas of the palate. In addition, the use of a removable acrylic appliance in the initial healing period proved to be a reasonably good treatment option with regard to the removal of pathological tissue and postoperative complications.

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