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Case Report / Приказ случаја

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Myoepithelioma originating from floor of the mouth

Миоепителиом пода уста

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SUMMARY

Introduction Myoepithelioma Primarily affects the parotid gland. Usually presents as a painless lump with slow growth.

The aim of this study to report a case of myoepithelioma in the mouth floor.

Case outline A young man presented with a painless increased volume in the left side of the mouth floor region, one year of evolution. Noticed a sessile tumor with normal colored mucosa and absence of secretion output. Computed tomography with contrast showed an image with slightly heterogeneous density, with well-defined limits. Incisional biopsy performed under local anesthesia, after pathology examination, revealed a myoepithelial neoplasm. Then, a total excision of the lesion was performed under general anesthesia, which revealed the diagnosis of salivary gland myoepithelioma. The patient did not present signs of relapse after two years of follow up.

Conclusion Despite myoepithelioma originated in salivary gland are considered rare, especially in the mouth floor, this tumor should be considered in the differential diagnosis of similar lesions. Proper treatm-ent appears to be complete surgical excision and post-operative follow-ups shows should be carried out as long as possible, despite the fact that relapses are extremely rare.

Keywords: Salivary Gland; Oral Pathology; Myoepitheliomas

САЖЕТАК

Увод Миоепитхелиоми се првенствено налазе на паротидној жлезди као безболни израштај са спорим растом.

Циљ овог рада је да прикаже болесника са миоепитхелиомпм у поду уста.

Приказ болесника Младић је приметио безболну отеклину на левој страни пода уста, са једногодишњом еволуцијом у сесилан тумор, нормално пребојен, без секреције. Компјутеризована томографија са контрастом приказала је добро ограничену промену мале хетерогене густине Биопсија је урађена у локалној анестезији, а хистопатолошки преглед указао на миоепителну неоплазму. Промена је у целости одстрањена у општој анестезији, а патохистолошки налаз потврдио дијагнозу миоепитхелиома пљувачне жлезде. Болесник је праћен две године и без рецидива болести је.

З**акључак** Миоепитхелиоми пљувачних жлезда су ретки, посебно у устима пода, али овај тумор треба размотрити у диференцијалној дијагнози сличних лезија. Неопходна је потпуна хируршка ексцизија а време постоперативног што је могуће дуже и ако су рецидиви изузетно ретки.

Кључне речи: пљувачне жлезде; Орална патологија; Миоепителиоми

INTRODUCTION

Myoepithelioma was first described in 1943 [1]. However, it was only in 1991 that was considered by the World Health Organization as a distinct pathological entity. Also known as myoepithelial adenoma [2], this tumor is composed entirely of myoepithelial cells, without ducts formation inside and is about 1.0 to 1.5% of all salivary gland tumors [3–14]. It affects both minor and major salivary glands, but is more commonly found in the parotid glands (about 50%), sublingual (33%) and submandibular (13%) [13,15]. Patients between the fourth and sixth decades of life are the most often affected [4,7,11,15], and there is no predilection for gender [11, 14]. Usually present as a painless nodule with slow growth [13–15].

Myoepithelial cells are part of the normal composition of the salivary glands and are important components of many types of salivary gland tumors, such as Pleomorphic adenoma, adenoid cystic carcinoma and terminal duct carcinoma [16,17]. These cells are located between the basal lamina and the acinar and ductal cells. They have structural characteristics similar to epithelial and smooth muscle cells [3, 18].

The myoepithelioma is rarely found with more than 200 cases reported [18]. Thus, this study aims to report what we believe is the second case of myoepithelioma in the mouth floor described in

the English literature of the past 20 years, highlighting its clinical and pathologic characteristics and appropriate treatment.

CASE REPORT

A 28-year-old black male, attended the Oral and Maxillofacial Surgery and Traumatology Clinic at Baiano's Center for Dental Studies, Salvador, Brazil, complaining of painless swelling in the left mouth floor region, which lasted one year. Regarding its previous medical history, there was nothing to consider. At physical examination, a swelling, firm to palpation and lined by a normal mucosa could be noticed. It was located in floor of the mouth, in the left anterior region (Figure 1A).

Computed tomography with contrast images showed a hiperdense lesion in a region close to the left base of the tongue, with contours well-defined and slightly heterogeneous density, measuring about 4.0x2.5x1.5 cm in its greatest diameter (Figure 1B). At the ultrasound examination, it was possible to observe epithelial, subcutaneous and muscle tissues within normal limits and the presence of fluid collection within the lesion was not detected.

An incisional biopsy under local anesthesia was performed, and histopathological examination revealed a well circumscribed neoplasm characterized by the presence of plasmocytoid myoepithelial, epithelioid and eventually cuboid cells in a fibrous or hyaline matrix (Figure 1E). The patient was thereafter submitted to excisional biopsy under general anesthesia through intraoral access in the left anterior floor of the mouth region. During surgery we could notice that the lesion had well-defined boundaries, easy identification and cleavage, with rubber-consistency and predominantly yellowish color with purplish spots (Figures 1C and 1D).

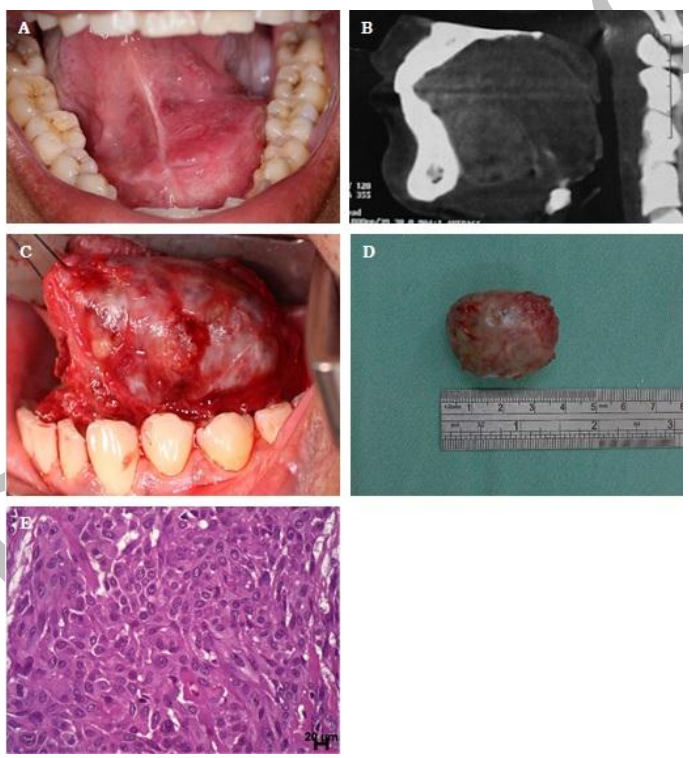

Figure 1: A) Clinical image showing a swelling located in the floor of mouth; B) Tomographic image (sagittal multiplanar reconstruction) presenting a well located lesion; C) Nodular and well delimited lesion located in the floor of mouth; D) Nodular lesion measuring 4.0 x 2.5 x 1.5 cm; E) Solid area showing proliferation of plasmocytoid, epithelioid and, eventually, spindle shaped cells.

The surgical specimen was stored in formaldehyde at 10% and sent for histopathological examination and a diagnosis of myoepithelioma was established. The patient did not have any postoperative complication and after two years of follow-up, showed no signs of recurrence.

DISCUSSION

The occurrence of myoepithelioma in the head and neck area is rare, while the involvement of the oral cavity is extremely rare [19], representing about 1.0 to 1.5% of all salivary gland tumors [5, 18, 20]. According to Table 1, only one article was published about myoepithelioma located in the floor of the mouth, and maxilla was the most frequent site. There was no predilection for gender and age group most affected were persons in the third and fifth decades of life.

Table 1. Summarization of face myoepitheliomas clinical cases reported in 20 years.

Autors	Year	Age	Gender	Color	Local	Treatment	Follow-up	Recurrence
Kanazawa et al.	1999	42	Female	Yellow	Hard Palate	Local excision	2 years	No
Piatelli et al.	1999	47	-	-	Jugal Mucosa	Excisional biopsy	3 years	No
Carinci et al.	2001	30	Male	-	Tongue Base	Local resection + Chemotherapy	4 years and 4 months	No
Isogai et al.	2003	47	Female	Yellow	Bucal Mucosa	-	6 months	No
Nair et al.	2004	58	Male	Brown	Hard Palate	Local excision	6 months	No
Onbas et al.	2005	65	Female	-	Hemiface	-	-	-
Woo et al.	2005	22	Female	-	Dorsal Tongue	Excisional biopsy	1 year and 2 months	No
Cuesta Gil et al.	2008	54	Female	White	Maxilla	Hemimaxillectomy	3 years	No
Patrocinio et al.	2009	38	Male	-	Maxilla	Local resection	9 years	No
Nikitakis et al.	2010	45	Male	White	Dorsal Tongue	Excisional biopsy	2 years	No
Hunt et al.	2011	21	Male	White	Mouth Floor	Excision of the submandibular gland	-	-
Park and Seo	2011	23	Male	Yellow	Bucal Mucosa	Local Excision	2 years	No
Rishabh et al.	2011	22	Male	Brown	Orbit	Local Excision	5 months	No
Sperandio et al.	2011	42	Female	Black	Soft Palate	Local Excision	1 year	No
Badal S et al.	2013	55	Male	-	Maxilla	Hemimaxillectomy	-	-
Gore et al.	2013	70	Female	-	Maxilla	-	-	-
Gore et al.	2013	62	Female	-	Maxilla	-	-	-
Gore et al.	2013	30	Female	-	Maxilla	-	-	-
Mochizuki et al.	2013	40	Female	White	Parotid Gland	Enucletion	1 year	No
Yadav et al.	2013	40	Male	-	Soft Palate	Local Excision	6 months	No
Present case	**2016**	**28**	**Male**	**Black**	**Mouth Floor**	**Local Excision**	**2 years**	**No**

Clinically, myoepithelioma presents itself as a slow-growing, circumscribed and painless swelling [4,10,12,13,18,19,21]. The presented case showed an evolution period of two years, without painful symptoms associated, and imaging studies revealed a circumscribed lesion in floor of the mouth in the left anterior region. Myoepithelioma shows no predilection for gender and affects a wide age range [14,19,21], but some authors claim that the fifth decade of life is the most affected age group [13,18,20].

Most myoepitheliomas of salivary glands occur in parotid glands (50%), sublingual (33%) and submandibular gland (13%) [2,6,13,15]. Rarely, it affects locations such as the maxillary sinus, lacrimal gland, nasal cavity, larynx or dermis [6]. The origin of the tumor described in this study appears to be a minor salivary gland and its location is extremely rare, with only one case documented in English literature researched [20].

The reported case fills in the criteria for myoepithelioma. Myoepithelial cells are similar to smooth muscle cells, probably of ectodermal origin, but performing functions of mesodermal cells [22]. Usually, myoepithelioma presents multiple cellular patterns as fusiform, plasmacytoid,

epithelioid, clear cell, mixed pattern and abundant presence of mucoid acellular stroma [3,4,6,9,12, 18–20]. The plasmacytoid type tends to occur more often in the oral cavity, especially palate, when compared with other types of myoepithelioma, although the fusiform pattern is the most common and often primarily affects the parotid gland [3,4,18]. Patterns containing epithelioid cells and clear cells develop in the parotid glands and often suffer malignant transformation [6]. Histological pattern does not influence the biological behavior of the lesion [18].

The myoepithelioma is often confused with pleomorphic adenomas due to the large amount of myoepithelial cells present in these two tumors [3, 9, 19, 20]. Myoepitheliomas were once considered as a variant of pleomorphic adenoma [13]; however, since 1991, the World Health Organization clearly differentiated myoepithelioma from pleomorphic adenoma, showing that myoepithelioma presents epithelial cells, but it has no duct differentiation or presence of chondroid or mixochondroid matrix.

The differential diagnosis includes pleomorphic adenoma and other salivary gland tumors, including cancer. The first suspect in the presented case was pleomorphic adenoma, followed by plunging ranula; however, in floor of the mouth, other tumors such as lipomas and neurofibromas can also be found.

Myoepitheliomas are less likely to recur than pleomorphic adenoma; however, they can undergo malignant transformation, especially when there are recurrent relapses or tumor existence for a long time without treatment [9]. The prognosis is based on the histopathology, being favorable for the benign form, which does not eliminate the need for regular monitoring to detect local recurrence, though it is rare when the lesion is completely removed [9,18,20].

According to the Table 1 and the current literature [2,9,12,13,18], the treatment usually consists of complete removal of the lesion, with no reports of recurrence after an average time of 25 months following surgery. Recurrence rates of 10% and 18% are reported, probably due to incomplete removal of the lesion. The prognosis is favorable [12, 14]. In the present case, after two years of postoperative follow-up, there were no signs of recurrence. However, it is wise to carry out follow-ups as long as possible, despite the relapses are extremely rare.

REFERENCES

1. Sheldon W. So-called mixed tumors of the salivary glands. Arch Pathol. 1943; 35: 1–20.
2. Sperandio FF, Giudice FS, Pinto-Junior DS, de Sousa SCOM. Myoepithelioma of the soft palate: a case report giving special attention to the differential diagnosis. J Oral Maxillofac Res. 2011; 2: 4.
3. Kanazawa H, Furuya, T, Watanabe T, Kato J. Plasmacytoid myoepithelioma of the palate. J Oral Maxillofac Surg. 1999; 57: 857–60.
4. Piatelli A, Fioroni M, Rubini C. Myoepithelioma of the gingiva. Report of a case. J Periodontol. 1999; 70(6): 683–7.
5. Carinci F, Grasso DL, Grandi E, Pelucchi S, Pastore A. Malignant myoepithelioma of the tongue base: Case report and literature review. J Craniofac Surg. 2001; 12(6): 544–6.
6. Isogai R, Kawada A, Ueno K, Aragane Y, Tezuka T. Myoepithelioma possibly originating from the acessory parotid gland. Dermatology. 2004; 208(1): 74–8.

7. Onbas O, Karasen RM, Gursan N, Kantarci M, Alper F, Okur A. Giant myoepithelioma of the face: MDCT with 2D AND 3D images. AJR Am J Roentgenol. 2006; 187(4): W418–9.
8. Patrocinio LG, Damasceno PG, Patrocinio JA. Malignant myoepithelioma of the hard palate: 9-year follow-up. Braz J Otorhinolaryngol. 2009; 75(4): 620.
9. Park TH, Seo SW. Diagnostic challenges of myoepithelioma arising from a minor salivary gland. J Oral Maxillofac Surg. 2011; 69(11): 2830–2.
10. Rishabh K, Ashwarya T, R S. A Rare Case of Myoepithelioma around the Left Orbit. J Dent Res Dent Clin Dent Prospects. 2011; 5(4): 141–3.
11. Badal S, Ahmed S, Patil PS, Badal A. Malignant myoepithelioma of the maxilla posing a diagnostic dilemma. Natl J Maxillofac Surg. 2013; 4(2): 235–8.
12. Gore CR, Panicker N, Chandanwale S, Singh BK. Myoepithelioma of minor salivary glands - A diagnostic challenge: Report of three cases with varied histomorphology. J Oral Maxillofac Pathol. 2013; 17(2): 257–60.
13. Mochizuki Y, Omura K, Tanaka K, Sakamoto K, Yamaguchi A. Myoepithelioma of the Parotid Gland Presenting as a Retroauricular Cutaneous nodule: A Case Report. J Clin Diagn Res. 2013; 7(6): 1165–8.
14. Nair BJ, Vivek V, Sivakumar TT, Joseph AP, Varun BR, Mony V. Clear cell myoepithelioma of palate with emphasis on clinical and histological differential diagnosis. Clin Pract. 2014; 4(1): 628.
15. Rosai J. Ackerman`s Surgical Pathology. 8th ed. St. Louis: CV Mosby; 1996. p. 833.
16. Takeda Y. Malignant myoepithelioma of minor salivary gland origin. Acta Pathol Jpn. 1992; 42(7): 518–22.
17. Yadav AK, Nadarajah J, Chandrashekhara SH, Tambade VD, Acharya S. Myoepithelioma of the soft palate: a case report. Case Rep Otolaryngol. 2013; 2013: 642806.
18. Hunt KT, Stevens MR, Abdelsayed RA, Nguyen CT. Benign myoepithelioma of floor of mouth with mandibular involvement: a case report and literature review. J Oral Maxillofac Surg. 2011; 69(12): 3001–5.
19. Nikitakis NG, Argyris P, Sklavounou A, Papadimitriou JC. Oral myoepithelioma of soft tissue origin: report of a new case and literature review. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 2010; 110(5): e48–51.
20. Cuesta Gil M, Bucci T, Navarro Cuellar C, Duarte Ruiz B, Pannone G, Bufo P, et al. Intraosseous myoepithelioma of the maxilla: clinicopathologic features and therapeutic considerations. J Oral Maxillofac Surg. 2008; 66(4): 800–3.
21. Peel Rl, Gnepp DR. Diseases of the salivary glands. In Barnes L. Surgical Pathology of the head and neck, 2th ed. Vol. 1. New York: Marcel Dekker; 1985. p. 534
22. Woo VL, Angiero F, Fantasia JE. Myoepithelioma of the tongue. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 2005; 99(5): 581–9.

