Journal of Oral and Maxillofacial Surgery, Medicine, and Pathology xxx (2017) xxx–xxx

Contents lists available at ScienceDirect

Journal of Oral and Maxillofacial Surgery, Medicine, and Pathology

journal homepage: www.elsevier.com/locate/jomsmp

Case report

Extensive salivary myoepithelioma in pediatric patient

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ARTICLE INFO

Article history:
Received 10 May 2017
Received in revised form 15 July 2017
Accepted 18 July 2017
Available online xxx

Keywords:
Salivary gland tumors
Children
Myoepithelioma

ABSTRACT

Myoepithelioma is a benign salivary gland neoplasm, locally invasive and uncommon in children. The aim of this study is to present and discuss the clinical, morphological and immunohistochemical findings of this tumor in a pediatric patient. This is a case report of a 12-year-old female patient showed a nodular lesion in the palate with osseous resorption with one year of evolution. After incisional biopsy, the histopathological examination revealed encapsulated lesion with proliferation of myoepithelial cells, including plasmacytoid, fusiform and clear cells, arranged in sheets, nests and cords. Immunohistochemical analysis revealed immunostaining for AE1/AE3, p63, cytokeratin-7, calponin, SMA and S-100. Based on these findings, diagnosis of myoepithelioma was confirmed. The lesion was completely removed through enucleation, in follow-up 2 years; there was no recurrence of the tumor. Myoepithelioma of salivary glands although rare in children may be invasive, however it is less aggressive than other tumors, a conservative treatment avoiding facial mutilation in pediatric patients is sufficient in these cases.

1. Introduction

Myoepithelioma was first described by Sheldon in 1943. Formerly, it was considered a variant of pleomorphic adenoma with predominance of myoepithelial cells; however, in 1991 the World Health Organization recognized it as a distinct entity. Myoepithelioma is rare and represents approximately 1% of all salivary gland tumors. Most cases occur between the ages of 30 and 50, and the parotid and palate are the most common sites in major and minor salivary glands, respectively [1,2].

This tumor is composed of myoepithelial cells that exhibit variable morphology including spindle, epithelioid, plasmacytoid and/or clear cells. A limited number of ductal structures (5–10% of the tumor component) may also be observed. Overall, myoepithelioma is well-encapsulated and circumscribed, presenting a lower rate of recurrence and lower risk of malignant transformation than pleomorphic adenoma. Although more common in salivary glands, it can also appear in the skin, soft tissues, trachea, larynx and sinonasal tract [3].

Salivary gland tumors are extremely rare in children, with a similar proportion of malignant and benign tumors in some studies. Myoepithelioma accounts for 1.8–10% of all salivary neoplasms in children and adolescents [4–6]. Considering the few cases of this tumor in pediatric patients, this study reports the clinical, morphological and immunohistochemical findings of a myoepithelioma of the minor salivary gland in a child.

2. Case report

A 12-year-old female patient attended an oral diagnostic service complaining of an asymptomatic nodular lesion on the hard palate. The lesion had normal mucosa color, being slightly purple, and measured approximately 3 cm in diameter (Fig. 1). The lesion had one-year disease duration. Computed tomography (CT) showed a well-circumscribed hypodense lesion associated with bone resorption in the palatal portion of the maxilla. No rupture of the cortical bone, invasion of the nasal cavity or maxillary sinus was observed (Fig. 2).

Considering the size of the lesion, an incisional biopsy was performed to establish the definitive diagnosis and treatment. Histopathological examination revealed proliferation of myoepithelial cells, including plasmacytoid, spindle and clear cells, arranged in sheets, nests and cords. The tumor occasionally exhibited pseudocystic spaces and rare ductal structures. The stroma was composed of fibrovascular tissue with areas of hyalinization. The

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http://dx.doi.org/10.1016/j.ajoms.2017.07.007
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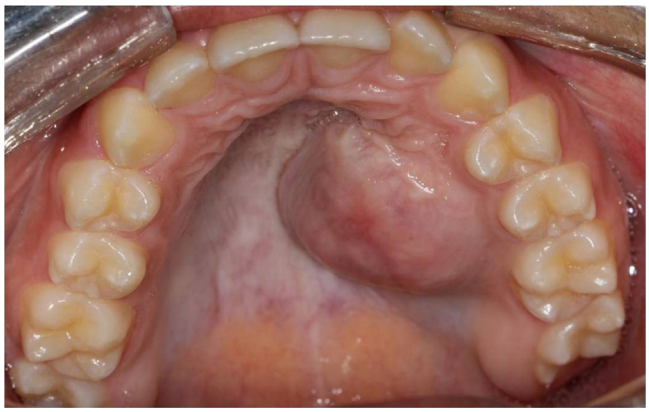

Fig. 1. Clinical presentation of myoepithelioma. The mass in palate was sessil and measured about 3 cm in its greatest diameter. Its surface was smooth with a mucosal color.

neoplasm was well delimited by a fibrous capsule (Fig. 3). Immunohistochemical analysis revealed immunostaining for p63, smooth muscle actin (SMA), calponin and S-100. Some ducts were positive for AE1/AE3 and cytokeratin-7, showed focal positivity (Fig. 4A–F). Based on these findings, the diagnosis of myoepithelioma was confirmed. The patient was submitted to conservative surgical removal of the tumor (Fig. 5) and currently, after 2 years of clinical follow-up, there is no recurrence.

3. Discussion

Myoepithelioma is a rare benign tumor of the salivary glands characterized by the proliferation of myoepithelial cells with varied morphology. Although it can occur in any age group, most cases are observed between 30 and 50 years, with a mean age of 36. Most myoepitheliomas affect the parotid gland and minor salivary glands of the palate. Clinically, they are similar to other benign salivary tumors, presenting as solid nodules with a diameter less than 3 cm, rarely reaching large dimensions [2,3,7].

This tumor is relatively rare in children and adolescents. As observed in Table 1, between the years 2007 and 2017 only 6 cases were reported. The age ranged from 11 to 15 years and the palate was the most common location. Only one case occurred in the parotid gland and none of the cases had recurrence.

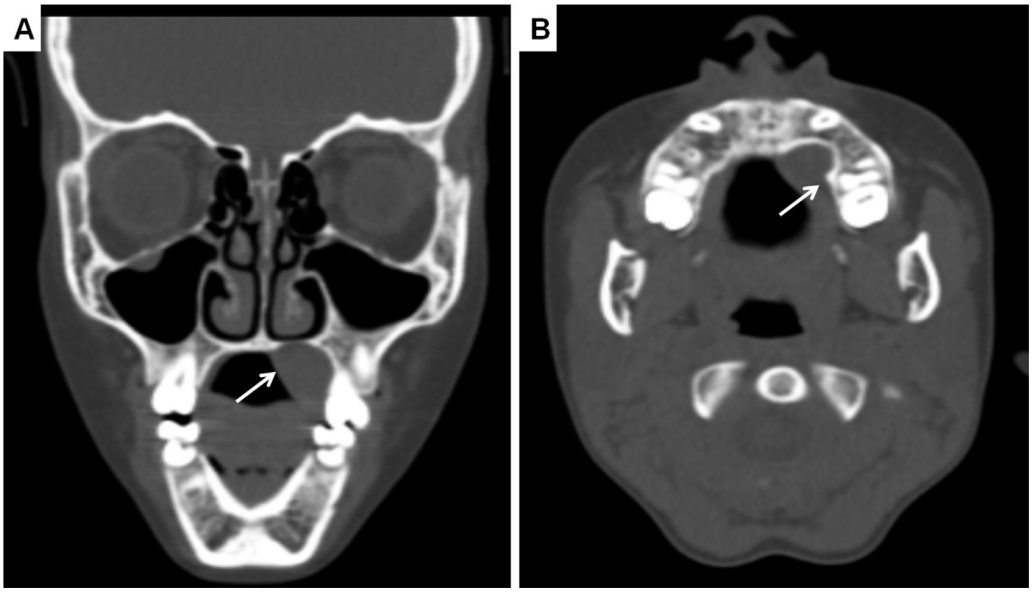

Fig. 2. (A) Frontal CT shows hypodense mass (arrow) with proximity to the nasal cavity. (B) Axial CT shows osseuos resorption in maxilla.

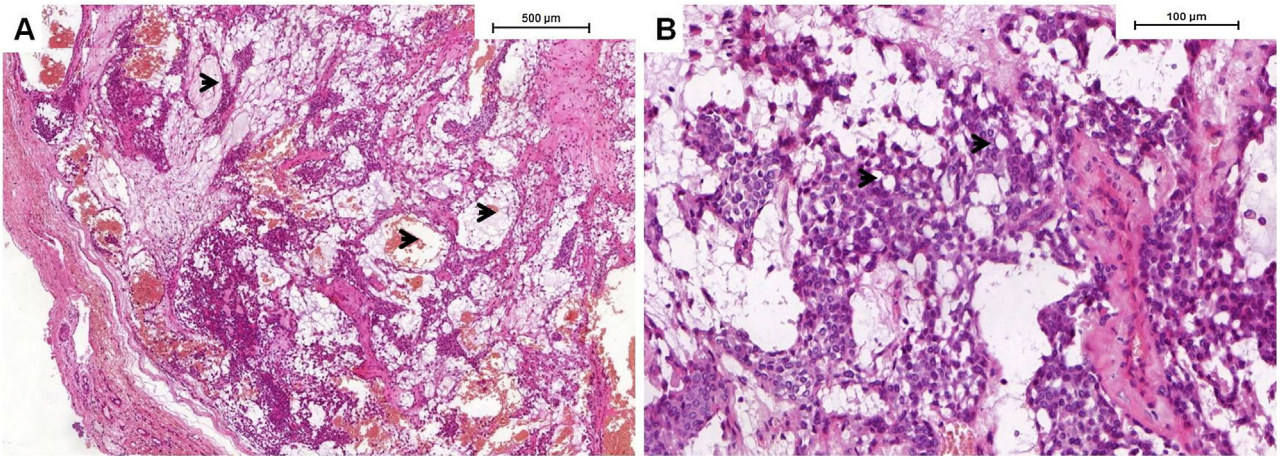

Fig. 3. (A) Histopathological findings. (A) Proliferation of myoepithelial cells in a myxoid stroma with pseudocystic spaces (arrowheads) and presence of fibrous capsule. (B) Absence of extensive ductal proliferation, but a few remnants of ductal structures were seen (arrowheads) (H&E, Bars indicate in figure).

Please cite this article in press as: da Silva LAB, et al. Extensive salivary myoepithelioma in pediatric patient. J Oral Maxillofac Surg Med Pathol (2017), http://dx.doi.org/10.1016/j.ajoms.2017.07.007