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Case Report

Intraoral exophytic lesion in an adolescent: A case report of myoepithelioma with unique clinical presentation

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ABSTRACT

Myoepithelioma is an infrequent benign tumor of the salivary glands, characterized by its composition of myoepithelial cells which can show different shapes and be arranged in various patterns with a well-circumscribed or encapsulated growth. This tumor commonly presents in adults as an asymptomatic swelling of the parotid gland, very rarely in minor salivary glands of children or adolescents, and even rarer in the buccal mucosa, with only six cases reported to date and only one of them presented in an adolescent. We present an additional case of myoepithelioma in the buccal mucosa of a 16-year-old male, with a novel clinical presentation as a non-submucosal exophytic mass. Immunohistochemically, neoplastic cells were positive for CK, S100, p63, and GFAP. The tumour was treated surgically, and the patient showed satisfactory evolution at 1 year of follow-up. The clinical and histopathological characteristics of the reported cases are discussed.

1. Introduction

Myoepithelioma is a benign salivary gland tumor, constituting only 1% to 1.5% of all salivary gland tumors. This tumor consists entirely of myoepithelial cells with variable cellular characteristics, including spindle, epithelioid, plasmacytoid and clear cells. A myoepithelioma may be composed of one or a mixture of these cell types, with a variable stromal component [1]. The diagnostic term myoepithelioma was first used by Sheldon in 1943 [2], but the tumor was reclassified as an independent entity in 1991 in the International Classification of Salivary Gland Tumors of the World Health Organization [3].

Myoepitheliomas have clinical and biological behavior similar to that of other benign tumors of the salivary gland, they present as asymptomatic swellings, either encapsulated or well circumscribed, slow growing, with a peak incidence in the third decade of life, without predilection by gender [4]. The most common site of presentation is the parotid, followed by the minor salivary glands of the hard and soft palate [5]. However, myoepithelioma of the buccal mucosa is extremely rare, to the best of our knowledge, only 6 cases have been previously reported in the English-language literature [4,6–10].

A case of plasmacytoid myoepithelioma is reported due to the rarity of this tumor, especially in adolescents and in the buccal mucosa, furthermore with novel clinical presentation, which was successfully treated by surgical excision. In addition, the clinical and histopathological characteristics of the reported cases are reviewed and discussed.

2. Case presentation

A 16-year-old man with no pathological history relevant to the current condition, no obvious facial asymmetry, presents an asymptomatic exophytic lesion with a nodular appearance on the buccal mucosa on the left side, of slow growth with three years of evolution approximately, multi-treated for infection. The lesion was sessile, with intact surface and telangiectasias, firm in consistency (Fig. 1 a and b). No other head and neck injuries were identified, and motor and sensory functions of the oral and maxillofacial area were normal. With the presumptive diagnosis of a benign lesion compatible with a lipoma, an excisional biopsy was performed.

Macroscopically, the well circumscribed specimen measured 2.2 × 1.9 × 1.6 cm, ovoid in shape, with a smooth surface. On cut surface, a homogeneous white solid, well delimited, with semi-translucent areas of mucoid appearance as well as discrete areas of hemorrhagic appearance were observed (Fig. 2 a and b). Microscopically, a well-circumscribed proliferation of round to polygonal cells was observed, mostly with a plasmacytoid appearance, with round and eccentric nuclei and abundant eosinophilic cytoplasm, and in focal areas with a fusiform appearance, arranged in trabeculae and

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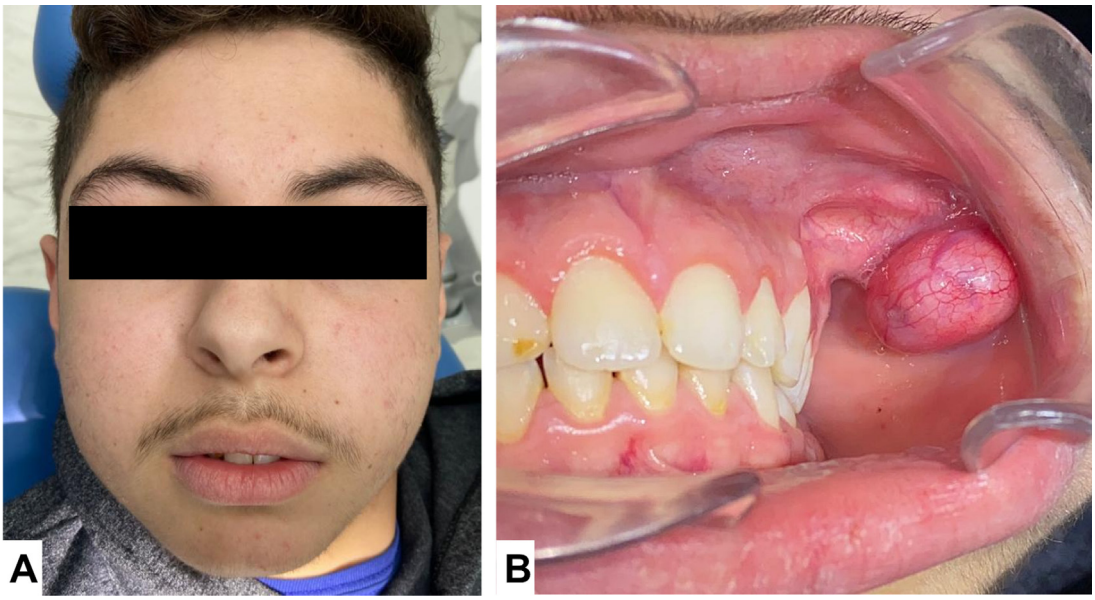

Fig. 1. (a) Extraoral features, without facial asymmetry; (b) Exophytic intraoral lesion, with intact mucosal surface.

nests, with areas of solid pattern. The stroma was myxomatous-mucoid with erythrocyte extravasation. Although the neoplasm was well circumscribed, no capsule was seen, and there was no involvement of the resection margins (Fig. 3). On immunohistochemical examination, the lesion was positive for CK7, GFAP, p63, S-100, and negative for CK5/6 (Fig. 4).

The lesion was diagnosed as plasmacytoid myoepithelioma arising from a minor salivary gland of the buccal mucosa. Postoperative follow-up at 1 year reported no recurrence of the lesion (Fig. 5).

3. Discussion

Myoepithelioma is a benign salivary gland tumor that is very rare in children and adolescents, Perez et al. [11] reported a case of plasmacytoid myoepithelioma in the palate of a child, and in their review, they reported that all cases of myoepithelioma in children and adolescents were of the plasmacytoid variant and in the palate. Interestingly, myoepitheliomas in pediatric patients exhibited cellular pleomorphism and the absence of a well-defined capsule (which is often present in myoepitheliomas in general), although the tumors were well circumscribed. Our adolescent patient who presented a plasmacytoid-type myoepithelioma, not encapsulated but well circumscribed, is distinguished by its buccal mucosa location.

Myoepithelioma can exhibit a solid, myxoid, reticular growth pattern, or a combination of these three growth patterns. Regarding architectural variations, the solid growth pattern is the most common, with the myxoid and reticular patterns being less common [12].

Myoepitheliomas are rare and could be misdiagnosed with the most common pleomorphic adenoma, which is considered the main differential diagnosis. Some authors have considered that myoepithelioma represents one end of the morphological spectrum of pleomorphic adenomas [4,8]. However, myoepithelioma must be distinguished from pleomorphic adenoma considering the following diagnostic criteria: the lack of ductal structures (although occasional ducts may be present), and the absence of chondroid or chondromyxoid matrix typically seen in pleomorphic adenoma [12].

Among the histological types of myoepithelioma, the plasmacytoid appears to have a predilection for the oral cavity, especially the palate [13], in slightly younger subjects [14]. This was the type seen in our adolescent patient, although localized on the buccal mucosa.

A myoepithelioma arising in the buccal area of the cheek is considered a rare entity, Table 1 shows the 6 cases reported to date, including the present case.

Recently, de Araujo Gomes et al. [10] informed a case of myoepithelioma that presented as a submucosal mass in the buccal mucosa of a 13-year-old adolescent. The authors described the case as the first presenting these characteristics. The present case is consistent with this reported case and differs from the other five reported cases in terms of age group.

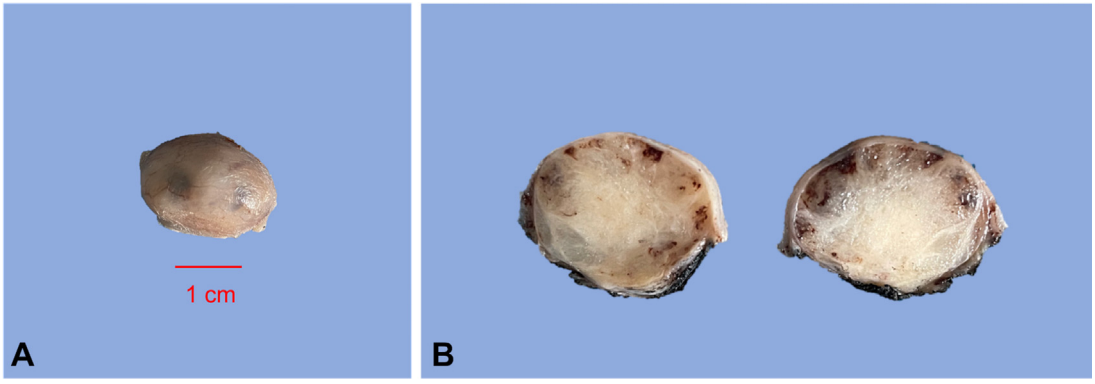

Fig. 2. (a) Well-circumscribed specimen fixed in 10% formalin; (b) Solid mucoid cut surface, with hemorrhagic areas.

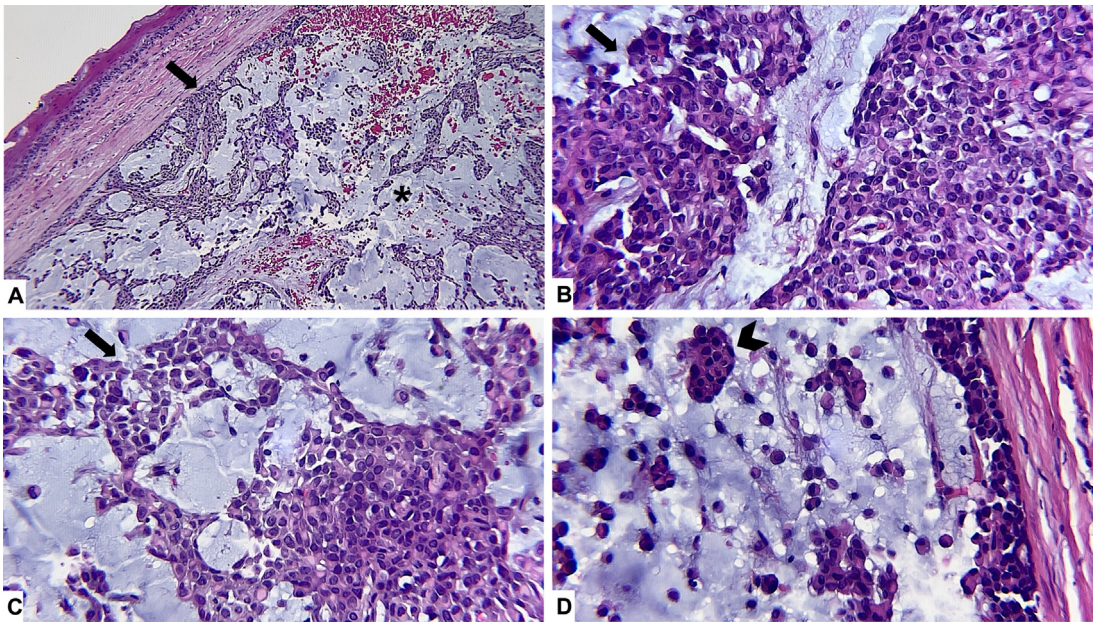

Fig. 3. (a) Well-defined neoplastic proliferation of plasmacytoid cells (arrow) on a muco-myxoid stroma (asterisk), H&E 40X; (b, c, d) Plasmacytoid cells arranged in trabeculae (arrow) and nests (arrowhead) of variable size, H&E 100X.

Ferri et al. [8] reported a case of myoepithelioma in the right buccal mucosa, in an 81-year-old woman. In addition, Sugiura et al. [7] reported a case of myoepithelioma in the left buccal mucosa, in a 54-year-old woman, and Tajima et al. [6] informed a case of myoepithelioma from buccal mucosa of a 56-year-old female. Interestingly, Argyris et al. [9] reported a case of polymorphous low-grade adenocarcinoma in the upper lip with a metachronous myoepithelioma in the buccal mucosa. The myoepithelioma reported by these authors shares the plasmacytoid characteristics of the neoplastic myoepithelial cells and the mucinous stroma of our case, although their patient was a 91-year-old female, which differed with the age of our adolescent patient.

Furthermore, a male-to-female ratio of 3:4 is observed among reported cases [4,6–10] including the present case. Macroscopically,

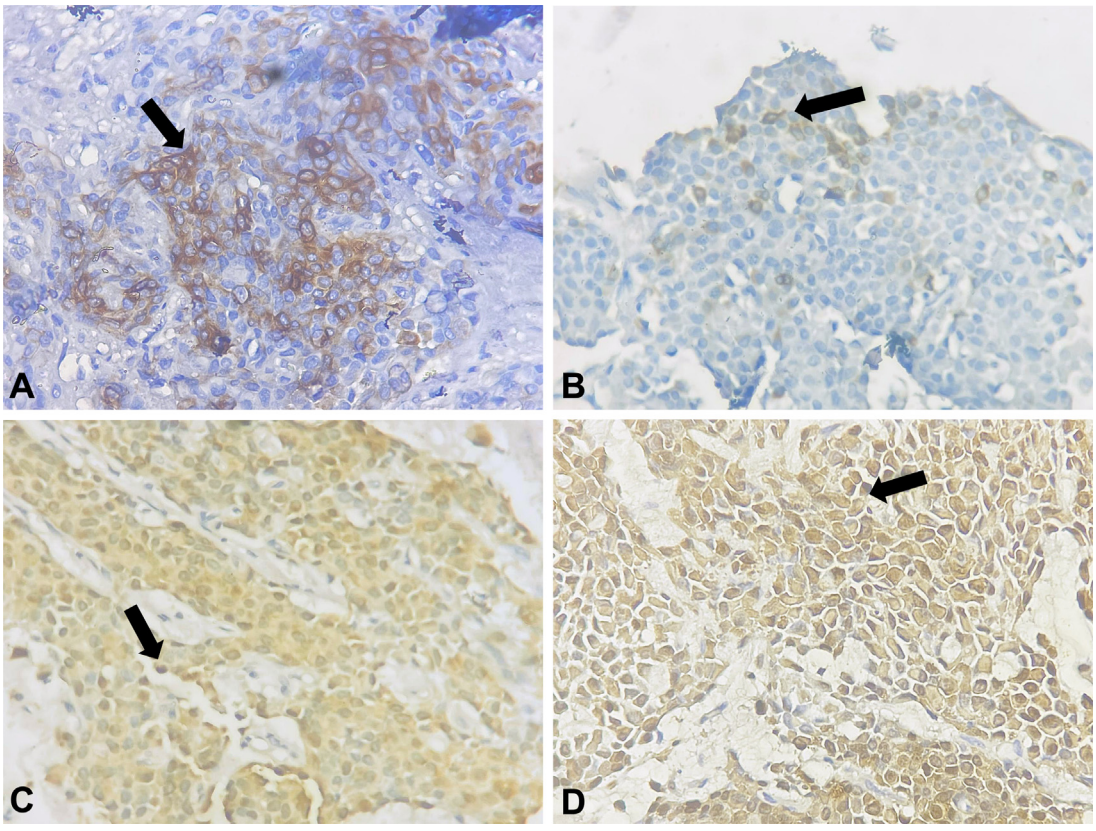

Fig. 4. Neoplastic cells positive (arrows) for (a) CK7; (b) GFAP; (c) p63; (d) S100.

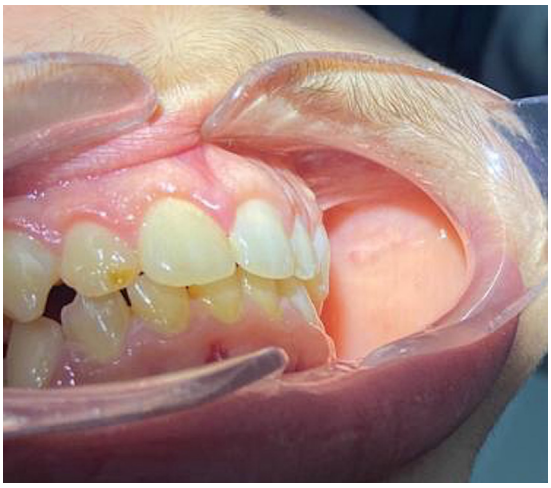

Fig. 5. Complete healing and no evidence of recurrence at 12-month follow-up.

Sugiura et al. [7], Ferri et al. [8] and Park et al. [4] reported an encapsulated specimen, in our case it was well delimited but not encapsulated, with a solid cut surface of muco-myxoid appearance. Histologically, the plasmacytoid cell type was identified in 5 of 7 cases [4,6,7,9], including the present case. Park et al. [4] reported a case of myoepithelioma in a 23-year-old man, a case histopathologically very similar to that of our 16-year-old patient.

Remarkably, all reported cases of myoepithelioma on the buccal mucosa presented as an asymptomatic mass, generally with a long evolution time, with a submucosal presentation [4,7–10], which differs from the unique clinical presentation as a non-submucosal exophytic mass in our patient.

Immunohistochemical analysis can help in the diagnosis of myoepithelioma, with positivity for CK7, CK14, AE1/AE3, S-100 and p63, among other markers such as GFAP and calponin. Positivity to smooth muscle actin and calponin can vary and often occurs in the spindle cell component [12]. Usually, a combination of a keratin in conjunction with S-100 detection, vimentin, and/or a myogenic marker is required for confirmation of the diagnosis of myoepithelioma [1]. Present case showed positivity for CK7, S100, p63 y GFAP confirming the diagnosis. Regarding the immunohistochemical profile, our case coincides with those reported by Park et al. [4], Argyris et al. [9], de Araujo Gomes et al. [10] and Sugiura et al. [7], and differs with Ferri et al. [8] regarding the positivity for CK and GFAP.

Surgical removal of the myoepithelioma with a margin of uninvolved normal tissue is the first treatment option for myoepithelioma, and the prognosis is very favorable. Patients should have regular follow-up reviews to rule out local recurrence, which is associated with incomplete surgical resection [4,15]. Possibly due to its slow growth, postoperative local recurrence can occur even 10 years later, its rare recurrence can become malignant [10], thus careful initial diagnosis and long-term clinical follow-up are merited. At one year of follow-up, our patient is free of lesion, and will continue to be clinically monitored. Reported cases responded satisfactorily to surgical excision, with no recurrences in a range of 8 to 36 months follow-up [4,7–10].

Finally, our case is interesting not only for presenting on the buccal mucosa of an adolescent, which contributes to reporting the unusual location and age group of this rare salivary gland neoplasm, but also for its novel clinical presentation as a non-submucosal exophytic mass that can mimic common reactive or benign neoplastic lesions of the oral mucosa, which can lead to incorrect diagnosis and delays in proper management.

Table 1
Reported cases of myoepithelioma of the buccal mucosa.

Author	Sex/age	Clinical features	Macroscopy	Cellular type	Immunohistochemistry	Treatment/Follow-up
Tajima et al. [6]	F/56	Not mentioned	Not mentioned	Plasmacytoid and spindle-shaped	Not mentioned	Not mentioned
Sugiura et al. [7]	F/54	Painless swelling in the left buccal mucosa, for five years	Well encapsulated tumor specimen measured 1.0 × 0.7 × 0.6 cm	Plasmacytoid and spindle-shaped	Moderately positive for S100, Vimentin, GFAP Weakly positive for CK Negative for actin	Surgical excision, no recurrence at 8-month follow-up
Ferri et al. [8]	F/81	Non-painful submucosa mass in the right cheek, for two years	Well-circumscribed capsulated mass measuring 3.5 × 3 × 2.5 cm	Spindle-shaped	Strongly positive for vimentin and focally for S100 and CD100 Negative for CK, αSMA, MNF116, HMB-45 and GFAP Ki-67 1%	Surgical excision, no recurrence at 12-month follow-up
Park et al. [4]	M/23	Nontender mass in the buccal mucosa, for several years	Well-demarcated capsulated ovoid mass measuring 2.5 × 1.8 × 1.5 cm	Plasmacytoid	Positivity for CK, S100, Vimentin, Locally positive for p63 Negative for αSMA and EMA Ki-67 0%	Surgical excision, no recurrence at 24-month follow-up
Argyris et al. [9]	F/91	Asymptomatic, firm, 1.5-cm mass of the left buccal vestibule	Not mentioned	Epithelioid and plasmacytoid	Positivity for AE1/AE3, CK7 and S100 Sporadic positivity for CK5/6, CK8/18, GFAP, and calponin Rare positivity for p63 Negative for CEA, CD10, EMA and αSMA Ki-67 0%	Surgical excision, no recurrence at 36-month follow-up
de Araujo Gomes et al. [10]	M/13	Painless nodular lesion, with a sessile base, in the left lower buccal mucosa, for seven years	Not mentioned	Epithelioid	Positive for AE1/AE3, GFAP Partially positive for CK7, calponin, p63 and S100	Surgical excision, no recurrence at 24-month follow-up
Present case	M/16	Asymptomatic exophytic lesion with a nodular appearance on the buccal mucosa on the left side, for three years	Well circumscribed specimen measuring 2.2 × 1.9 × 1.6 cm	Plasmacytoid	Positivity for CK7, S100, p63, GFAP Negativity for CK5/6	Surgical excision, no recurrence at 12-month follow-up

M: male, F: female, CK: cytokeratin, GFAP: Glial fibrillar acidic protein, αSMA: alpha smooth muscle actin, EMA: epithelial membrane antigen.

4. Conclusion

Dental and pediatric health professionals must recognize this rare entity as part of the differential diagnosis of non-submucosal exophytic lesions of the oral mucosa, for its timely and correct treatment.

Declaration of Competing Interest

None.

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