Case Report

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Myoepithelioma of Right Buccal Mucosa: A Rare Case Report

Dentistry Section

VAISHNAVI SAWANT[1], SAMKIT SAKHARIYA[2], AMITA ADITYA[3], PUSHKAR WAKNIS[4], FARHA KHATOON[5]

ABSTRACT

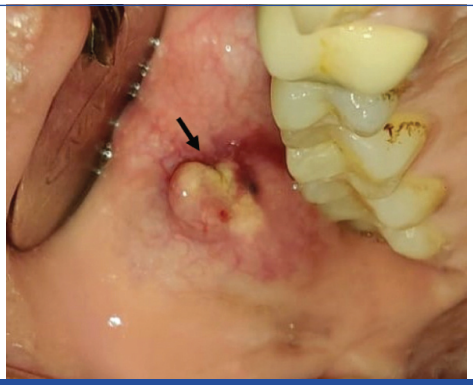
Myoepithelioma is a rare benign salivary gland tumour that has been reported sporadically. It is most often seen to be associated with the parotid gland, and the involvement of minor salivary glands remains a rarity. Amongst the minor salivary glands, myoepithelioma has been reported to occur most commonly in the palate, most commonly, followed by the tongue and upper lips. Its occurrence in the buccal mucosa is an extremely rare phenomenon and to date, only a few cases have been reported. This is a rare case of myoepithelioma of the minor salivary gland of the right buccal mucosa, adding to the list. Hereby, the authors present a case report of a 40-year-old female who visited with a complaint of tooth decay and an asymptomatic, long-standing tissue growth was noted in the right buccal mucosa. What was initially and provisionally diagnosed as irritational hyperplasia and was turned out to be rare benign tumour, i.e., myoepithelioma, on detailed investigation. It was successfully managed with surgical excision, and no recurrence has been reported to date.

Keywords: Benign, Immunohistochemistry, Minor salivary gland, Neoplasia, Tumour

CASE REPORT

A 40-year-old female reported to the Department of Oral Medicine and Radiology with the chief complaint of pain in the upper left back region of her jaw for 4-5 months, related to a reinfection of a root canal-treated tooth. The pain was gradual in onset, intermittent in nature with moderate intensity, lasted for 1-2 minutes, aggravated on eating on the left side, and relieved on its own or sometimes by taking over-the-counter painkillers. She did not have any contributory past medical or family history but had a past dental history of root canal treatment at a private clinic with three of her teeth: two in the upper left back teeth four years ago and one in the upper right back tooth three years ago. She had relatively good oral hygiene and no adverse habits. Extraoral examination did not reveal any abnormality, and lymph nodes were non palpable.

During the intraoral examination, a single soft tissue growth was noted, about which the patient was unconcerned as it was asymptomatic and had been present for 4-5 years. It was seen on the right buccal mucosa, had an ovoid shape, and measured approximately 2×1.5 cm. The growth had a broad base with a soft tissue peduncle, and its location appeared to correspond to the occlusal plane from the right first premolar to the first molar region, mesio-laterally. The colour of the growth was slightly yellowish in the peduncle region and was erythematous near the base, with an area showing petechiae. The surface of the growth superiorly showed indentation, which was due to the opposing teeth [Table/Fig-1].

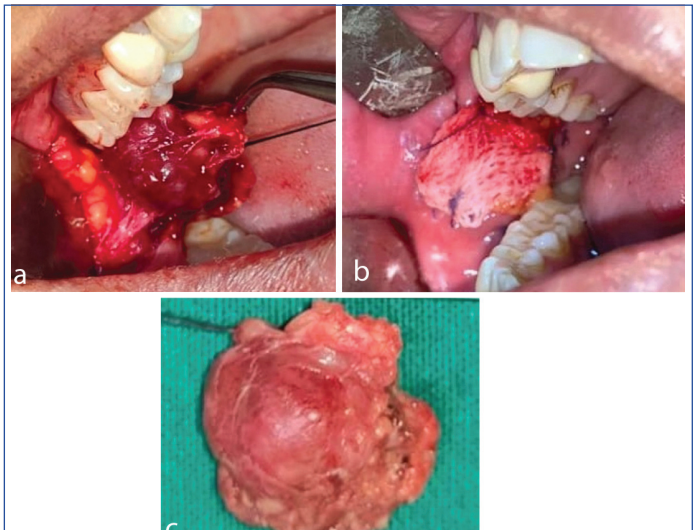


[Table/Fig-1]: A soft-tissue growth is seen on the right buccal mucosa over the occlusal plane, having ovoid shape with a broad base and peduncle along with indentation of the opposing teeth shown with the black arrow.

On palpation, all the inspection findings were confirmed. The growth was non tender, firm in consistency, non pulsatile, non fluctuant, non reducible, and non compressible. Sharp cusps were noted with the maxillary right second premolar and molars. Considering the clinical presentation, a provisional diagnosis of irritational fibroma was given, and benign salivary gland tumour, lipoma, and mucocele were considered as differential diagnosis.

Regarding her chief complaint, a radiologic evaluation was carried out, followed by root canal treatment for the concerned teeth. Also, enameloplasty was done for the sharp cusps of the affected teeth.

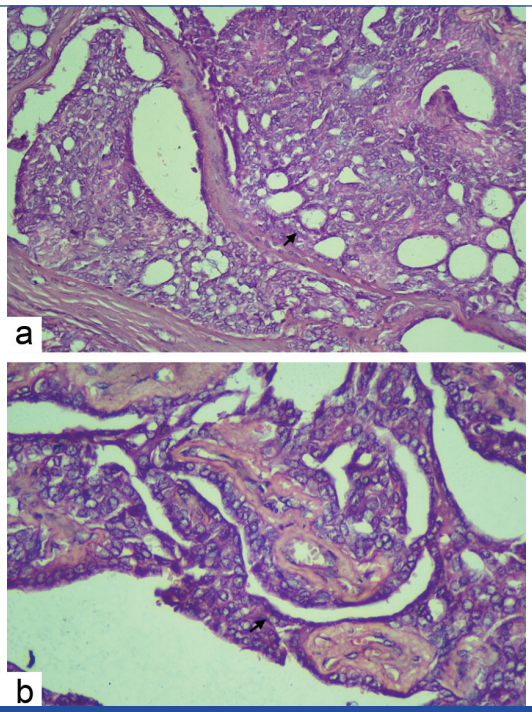
An excisional biopsy of the lesion was planned. Haematological investigations performed prior to the procedure did not reveal abnormal findings. The biopsy was performed under local anesthesia, and while excision, the lesion was surprisingly found to be invading deep into the submucosa [Table/Fig-2a]. The excision left a defect, which was filled by bringing in a vascularised buccal fat pad that was sutured in place along with the dressing [Table/Fig-2b]. The excised specimen was sent for histopathological evaluation [Table/Fig-2c]. Considering the unusual features of the



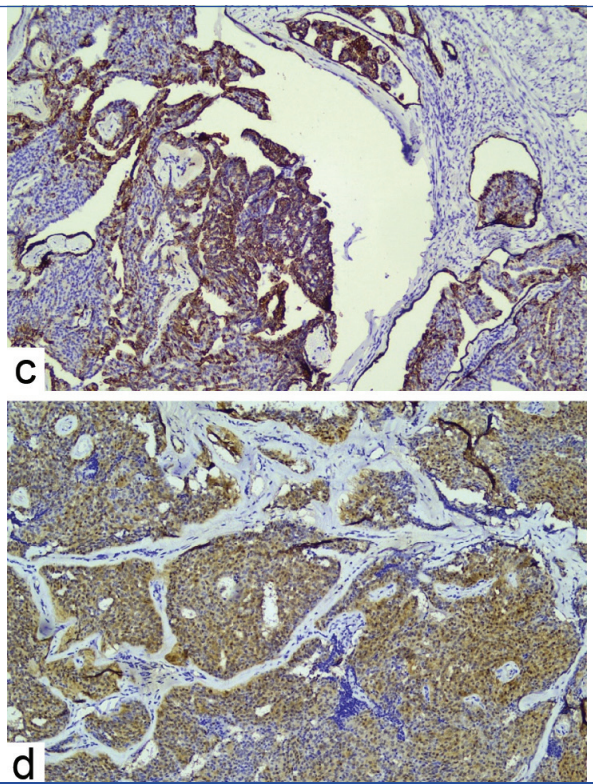
[Table/Fig-2]: a) Lesion while excision was seen to be invaded deep into the mucosa; b) Image shows surgical dressing of the buccal fat pad that was used for filling the post-excision defect; c) Image of the specimen post-excision showing an irregular shape and surface.

lesion during excision, Immunohistochemical (IHC) evaluation was also considered.

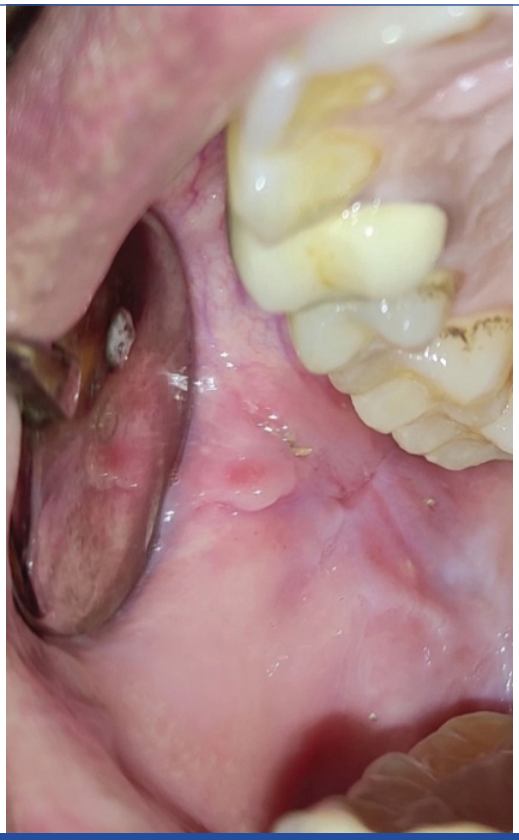
The specimen measured approximately 1.7×1.6 cm in dimensions. Histologically, it showed a varied picture with few areas of inflammatory cell infiltration, neoplastic cells, areas showing a ductal pattern of cells, and monomorphic cells. Epithelioid cells were observed displaying a plasmacytoid appearance with a scanty and focally myxoid stroma. It underwent further Immunohistochemistry (IHC) examination, which showed that neoplastic cells tested positive for Cytokeratin, S-100 protein, and C-Kit (Focal) [Table/Fig-3a-d]. These features were suggestive of myoepithelioma of the minor salivary gland with no definite features of malignancy noted. An adequate healing of the excised area was noted in the 14-day follow-up visit [Table/Fig-4]. The patient is currently under follow-up, and the lesion has not shown any recurrence in the last seven months.



[Table/Fig-3a,b]: Plasmacytoid appearance of epithelioid cells; scanty and focally myxoid stroma noted and the cells are arranged in small nests, lobules and papillae shown with black arrows {Haematoxylin and Eosin (H&E 40x)}.



[Table/Fig-3 c,d]: c) IHC stained section positive for CK; d) IHC stained section positive for S-100 protein (10x).



[Table/Fig-4]: Image taken at the time of follow-up which showed adequate healing of the surgical site.

DISCUSSION

The term "myoepithelioma" was first introduced by Sheldon in 1943 [1]. It accounts for 1-1.5% of all salivary gland neoplasms. It generally affects the parotid gland, with minor salivary glands are involved infrequently involved [2,3]. Among the minor salivary glands, it is generally seen affecting the palatal region, tongue, and upper lips. Till date, there have been few reported cases of myoepithelioma involving the buccal mucosa of the oral cavity, making the present case an extremely rare one. A comparative summary of some of the cases reported has been presented in [Table/Fig-5] [4-9].

Author's name/ Location	Year	Age/ gender	Site	Clinical features/ macroscopy	Treatment
Sugiura R et al., (Japan) [5]	2000	54/F	Left buccal mucosa	Painless swelling; for 5 years; 1.0×0.7×0.6 cm	Tumour was resected
Ferri E et al., (Italy) [6]	2006	81/F	Right cheek	Non painful submucosa mass for 2 years; 3.5×3×2.5 cm	Wide local tumour-resection
Park TH, (Korea) [4]	2011	23/M	Right buccal mucosa	Firm, non tender mass; for several years; 2 cm in diameter	Locally excised
Argyris PP et al., (USA) [7]	2014	91/F	Left buccal vestibule	Asymptomatic, firm, 1.5-cm mass; within 2 years	An excisional biopsy was performed
de Araujo Gomes B et al., (Brazil) [8]	2023	13/M	Left lower buccal mucosa	Painless nodular lesion, with a sessile base, for 7 years; 3 cm in diameter	Surgical excision
Urias Barreras CM et al., (Mexico) [9]	2023	16/M	Left buccal mucosa	Asymptomatic exophytic lesion, nodular appearance; for 3 years; size 2.2×1.9×1.6 cm	Excisional biopsy

[Table/Fig-5]: Summary of reported cases of myoepithelioma in buccal region [4-9].

Myoepithelioma originates from epithelial origin and phenotypically resembles smooth muscle. Previously, myoepithelioma was considered an extreme variant of pleomorphic adenoma; however, it has been considered as a separate entity by the World Health Organisation (WHO) since 1991. They can appear any time during the 3rd and 9th decades of life, with a median age of 53 years. Clinically, myoepithelioma typically presents as circumscribed, painless masses. However, in present case, its appearance was unconventional, giving more of a fibrosed look [4,10]

Given the location of the lesion in the buccal mucosa, it is important to consider other potential lesions in the differential diagnosis. Most commonly these may include fibrous hyperplasia, mucocele, and lipoma. Radiologic imaging modalities, such as Computed Tomography (CT) scans and Magnetic Resonance Imaging (MRI), can be used for investigation purposes to know about the extent, invasion, and spread of lesions if malignancy is suspected. MRI is considered the best imaging modality for evaluating salivary gland tumours, particularly for malignant palatal tumours [1]. However, a definitive diagnosis requires tissue histology.

Microscopically, myoepithelioma is composed of neoplastic myoepithelial cells, which under normal conditions are seen surrounding the ducts and acini of the exocrine glands. The cytoarchitecture of myoepithelial plasmacytoid neoplastic cells includes round to ovoid cells with eccentric nuclei and eosinophilic cytoplasm. Various growth patterns and appearances characterise it. Histologic growth patterns may be solid, myxoid (resembling pleomorphic adenoma), reticular, or a combination of these. In addition, neoplastic myoepithelial cells may have different histologic appearances (spindle, plasmacytoid, clear, and epithelioid cells). Epithelioid or clear cells may also be present. Generally no cellular atypia, necrosis, or increased mitotic figures. While a lack of ductal structures is evident, occasionally ductal cells may be present. Architecturally, myoepithelioma displays non myxoid (solid), myxoid (pleomorphic adenoma-like), reticular (canalicular-like), or mixed growth patterns. The tumour is often difficult to diagnose definitively diagnose at the light microscopic level due to its resemblance to pleomorphic adenoma [2,5,10]. Furthermore, as stated previously, there are varied histological appearances, adding to the diagnostic dilemma [10].

Immunohistochemistry (IHC) staining can aid in reducing the diagnostic challenge [4]. Positive immunoreactivity against S-100 protein, calponin, glial fibrillary acidic protein, actin, Cytokeratin (CK) 14, and various cytokeratins (AE/AE3, CAM5.2, and CK7) is a focally sensitive diagnostic tool that helps in diagnosing the exact cell of origin in cases of myoepithelioma. With the cytomorphologic features of tumour cells and IHC characteristics, a definitive diagnosis of benign myoepithelioma can be rendered with much conviction, as done in the present case [2,4,11].

Cases are generally reported to be managed effectively by conservatively excision lesions arising in minor salivary glands along with a thin rim of surrounding normal tissue. The prognosis of benign myoepithelioma is good, provided complete surgical excision is complete. Radiation therapy may be considered in cases where surgery is not feasible. Regular follow-up is indicated in cases of myoepithelioma as local recurrence can occur even after many years, and in rare cases it can turn malignant [4,10].

CONCLUSION(S)

Myoepithelioma is a rare lesion that poses a diagnostic challenge to clinicians. Like any other rare lesions of the oral cavity, meticulous work-up is required for its efficient diagnosis as well as management, and oral physicians must be cognizant of the same.

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PARTICULARS OF CONTRIBUTORS:
1. Postgraduate Student, Department of Oral Medicine and Radiology, Dr. D.Y. Patil Dental College and Hospital, Dr. D. Y. Patil Vidyapeeth, Pimpri, Pune, Maharashtra, India.
2. Postgraduate Student, Department of Oral and Maxillofacial Surgery, Dr. D.Y. Patil Dental College and Hospital, Dr. D. Y. Patil Vidyapeeth, Pimpri, Pune, Maharashtra, India.
3. Professor, Department of Oral Medicine and Radiology, Dr. D.Y. Patil Dental College and Hospital, Dr. D. Y. Patil Vidyapeeth, Pimpri, Pune, Maharashtra, India.
4. Professor, Department of Oral and Maxillofacial Surgery, Dr. D.Y. Patil Dental College and Hospital, Dr. D. Y. Patil Vidyapeeth, Pimpri, Pune, Maharashtra, India.
5. Postgraduate Student, Department of Oral Medicine and Radiology, Dr. D.Y. Patil Dental College and Hospital, Dr. D. Y. Patil Vidyapeeth, Pimpri, Pune, Maharashtra, India.

NAME, ADDRESS, E-MAIL ID OF THE CORRESPONDING AUTHOR:
Amita Aditya,
Department of Oral Medicine and Radiology, Dr. D.Y. Patil Dental College and Hospital, Sant Tukaram Nagar, Pimpri, Pune-411018, Maharashtra, India.
E-mail: dr.amitaaditya@gmail.com

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