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Original Research Paper

Pathology

MUCOEPIDERMOID CARCINOMA ARISING IN SIALADENOMA PAPILLIFERUM OF THE HARD PALATE: A RARE CASE REPORT

Dr Pragati Mohan Gakhare* MBBS, Junior Resident Department Of Pathology, Government Medical College And Hospital, Nagpur 440003 *Corresponding Author

Dr Vedita Golhar MD, Associate Professor Department of Pathology, Government Medical College and Hospital, Nagpur 440003

ABSTRACT

Background: Sialadenoma papilliferum (SP) is a rare benign tumor of the minor salivary glands, most commonly affecting the hard palate. It is often clinically mistaken for squamous papilloma due to its exophytic verrucous appearance. Malignant transformation of SP is exceedingly rare, with only a few reported cases of progression to mucoepidermoid carcinoma (MEC). **Case Presentation:** We report a rare case of a 36-year-old female presenting with a rapidly enlarging lesion on the hard palate. Clinical examination revealed two adjacent swellings, one verrucous and pinkish-white, and another nodular and firm. Fine needle aspiration cytology showed mature squamous cells with inflammatory background in one lesion, and a biphasic cell population suggestive of low-grade malignant salivary gland neoplasm in the other. Wide local excision was performed. Grossly, the specimens included a well-circumscribed solid grey-white mass and a papillary lesion. Histopathological examination revealed typical features of SP in the papillary component and low-grade MEC in the solid mass, with invasion into minor salivary glands and underlying bone. The final diagnosis was MEC arising in a background of SP. **Conclusion:** This case highlights the potential for malignant transformation in SP, a tumor traditionally considered benign. Coexistence with MEC should be considered in rapidly enlarging or atypical palatal lesions. Careful histopathological evaluation is essential for accurate diagnosis and optimal management.

KEYWORDS : Sialadenoma papilliferum, Mucoepidermoid carcinoma, Hard palate tumor, Minor salivary gland neoplasm, Squamous papilloma mimic, Malignant transformation

INTRODUCTION

Sialadenoma papilliferum (SP) is an exceptionally rare, benign tumor of the minor salivary glands, constituting less than 1% of all salivary gland neoplasms[1]. First described by Abrams and Finck in 1969, SP predominantly affects the hard palate and is histologically characterized by a distinctive biphasic architecture with papillary projections of stratified squamous epithelium overlying salivary ductal elements[2]. Due to its papillary, exophytic growth, SP is often clinically mistaken for more common lesions such as squamous papilloma or verrucous carcinoma, necessitating histopathological evaluation for accurate diagnosis[3].

Despite its benign classification, SP has recently garnered attention for its potential—albeit rare—malignant transformation. Few documented cases in the literature have reported SP evolving into salivary gland malignancies, including mucoepidermoid carcinoma (MEC), epithelial-myoepithelial carcinoma, and high-grade carcinomas[4-6]. These findings challenge the traditional perception of SP as entirely indolent and underscore the importance of comprehensive histological and molecular analysis, especially in atypical presentations.

Mucoepidermoid carcinoma is the most common malignant tumor of the salivary glands and frequently affects both major and minor glands, including the palate. MEC is histologically composed of mucin-producing, intermediate, and squamoid (epidermoid) cells arranged in cystic and solid patterns, and its clinical behavior varies with grade[4]. The rare coexistence of SP and MEC in the same lesion or anatomical site raises important diagnostic and therapeutic considerations, especially when the lesion exhibits both benign and malignant histologic features.

Additionally, SP is frequently misdiagnosed due to its clinical similarity to squamous papilloma, particularly when affecting the palate—a common site for HPV-induced papillomas. However, most studies have shown that SP is not associated with HPV, unlike squamous papillomas, highlighting the importance of molecular and immunohistochemical differentiation[7].

Herein, we present a rare case of SP with coexisting low-grade MEC on the hard palate in a young female patient. The lesion clinically mimicked a squamous papilloma, underscoring the diagnostic dilemma such presentations pose. This case adds to the limited literature on malignant transformation within SP and highlights the importance of histopathologic evaluation in seemingly benign oral lesions.

Case Presentation

A 36-year-old female farmer presented with a rapidly enlarging swelling on the hard palate for the past two months. Clinical examination revealed two distinct lesions: one appeared as a verrucous, pinkish-white exophytic growth, adherent to a bulging submucosal mass on the hard palate (Figure 1). According to the patient, both swellings developed simultaneously. There was no significant past medical or surgical history.

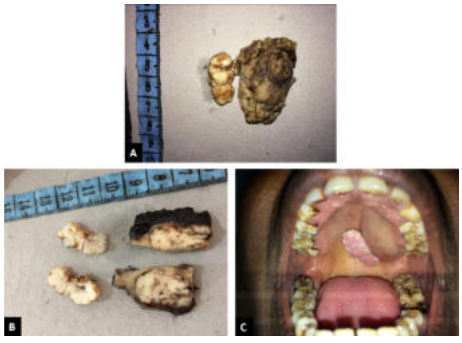

Figure 1: (A) Gross specimen showing two irregular, nodular tissue pieces. (B) Cut surface of the smaller tissue piece reveals a papillary architecture. The larger tissue piece appears well-circumscribed, partially encapsulated, and presents as a solid, grey-white tumor. (C) Intraoral clinical image demonstrating two distinct swellings on the hard palate, one of which displays a verrucous pinkish-white surface attached to a bulging lesion.

Fine needle aspiration cytology (FNAC) from the verrucous lesion revealed mature squamous cells with a few inflammatory cells, suggestive of a benign squamous

proliferation. The deeper lesion yielded smears showing a biphasic cell population consisting of mature squamous cells and small cuboidal to columnar cells with scant cytoplasm and round to oval nuclei with fine chromatin. Mucin-secreting cells and a mucinous or dirty background were appreciated (Figure 2). A provisional diagnosis of a verrucous lesion with a low-grade malignant salivary gland tumor was considered.

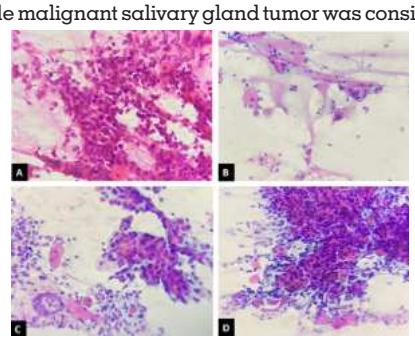

Figure 2: (A) Cellular smear showing cohesive clusters of intermediate cells with mild pleomorphism. (B) Scattered epithelial cells embedded in abundant mucoid background. (C) Occasional mature squamous cells seen among intermediate-type nuclei. (D) Dense cluster of epithelial cells with evidence of mucin production.

Wide local excision was performed, and two tissue specimens were submitted in a single container without orientation for margins. The larger specimen measured 4 × 3 × 2 cm and was covered by palatal mucosa on the inferior surface. The cut surface showed a well-circumscribed, partially encapsulated, solid gray-white tumor. The smaller specimen measured 2 × 2 × 1 cm with a gray-white, papillary cut surface (Figure 1).

Histopathological examination of the smaller specimen revealed features consistent with squamous papilloma-like architecture, while the larger specimen demonstrated mucoepidermoid carcinoma (MEC) infiltrating adjacent minor salivary glands and the underlying palatal bone (Figure 3). The MEC exhibited a mixture of mucin-producing columnar cells, intermediate cells, and epidermoid cells in varying proportions, forming both cystic and solid areas. Scattered clear cells with abundant cytoplasm and eccentric nuclei were also identified. The stroma showed a dense lymphoplasmacytic infiltrate (Figure 4).

A final diagnosis of mucoepidermoid carcinoma arising in a background of sialadenoma papilliferum was rendered.

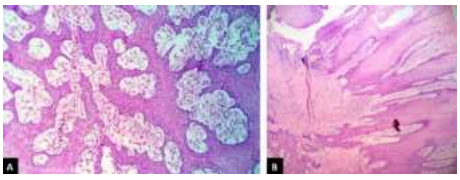

Figure 3: (A) High-power view showing multiple papillary projections lined by stratified squamous epithelium with underlying fibrovascular cores. (H&E stain, 10×) (B) Low-power view revealing the overall exophytic papillary architecture of the lesion.

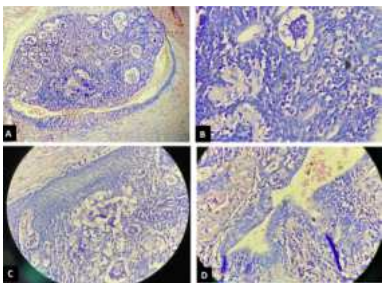

Figure 4: (A) Low-power view showing cystic and solid islands of tumor cells in sclerotic stroma. (B) High-power view highlighting mucous and intermediate-type cells. (C) Epidermoid (squamous) and mucous cells are distinctly visible within the neoplastic proliferation. (D) Cystic spaces with mucous cells are evident.

DISCUSSION

The current case presents a highly uncommon coexistence of SP and MEC in a 36-year-old female with a lesion located on the hard palate. Such dual pathology is exceedingly rare and contributes to the limited but growing body of evidence documenting malignant transformation occurring within or adjacent to SP. Prior case reports have noted similar transformations, frequently characterized by the preservation of benign morphology in the exophytic SP component, while malignant changes—such as those characteristic of MEC—predominate in the deeper, endophytic regions[4-6].

Initial cytologic assessment in our case, via fine-needle aspiration, suggested a dual epithelial population indicative of low-grade malignancy, though lacked definitive mucinous differentiation. Subsequent surgical excision and histopathological analysis confirmed the presence of a biphasic neoplasm, comprising mucin-secreting, intermediate, and epidermoid cells consistent with MEC, juxtaposed with a papillary epithelial proliferation characteristic of SP. The invasive nature of MEC, extending into the minor salivary glands and underlying bony palate, underscored its malignant potential and highlighted the clinical significance of timely diagnosis and intervention.

Notably, the literature corroborates the possibility of such malignant transformations within SP. Liu et al. (2009) documented a high-grade MEC arising from an SP at the tongue base, while Shimoda et al. (2004) described transitions to both epithelial–myoepithelial carcinoma and high-grade carcinoma within SP, thereby illustrating the broad histological spectrum of its malignant evolution[5,6]. These findings suggest that SP, although typically benign, may in rare instances serve as a precursor or nidus for malignant salivary gland neoplasms.

Emerging molecular data have further substantiated the neoplastic nature of SP. The identification of BRAF V600E mutations in SP, as recently reported by Alsugair et al. (2024), supports its classification as a true neoplasm rather than a reactive hyperplasia[8]. These molecular markers may provide valuable diagnostic adjuncts, particularly in differentiating SP from morphologically similar lesions or confirming early malignant transformation.

A key diagnostic dilemma lies in distinguishing SP from squamous papilloma, particularly those induced by human papillomavirus (HPV). Clinically, both lesions can manifest as exophytic, verrucous proliferations; however, SP typically lacks HPV association and demonstrates a more complex glandular architecture[7]. These overlapping clinical features reinforce the necessity of biopsy and histopathological evaluation for any persistent or atypical oral lesion.

CONCLUSION

Sialadenoma papilliferum (SP) is a rare, benign tumor of minor salivary glands that can closely mimic more common lesions like squamous papilloma. Although traditionally considered indolent, this case highlights the potential for malignant transformation, specifically to mucoepidermoid carcinoma (MEC), an association that has been reported only rarely in the literature. The dual presentation of SP and MEC within the same lesion emphasizes the need for thorough histopathological evaluation, especially in rapidly growing or clinically atypical palatal masses. Given the diagnostic challenges posed by overlapping clinical and cytological features, surgical excision with careful histologic assessment

remains the gold standard for accurate diagnosis. Early recognition of malignant change within SP is critical for appropriate treatment planning and to reduce the risk of recurrence or metastasis. This case reinforces the importance of considering hybrid or transforming lesions in differential diagnoses of salivary gland tumors and contributes to the evolving understanding of SP's biological behavior.

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