Primary Mucinous Carcinoma of Buccal Mucosa: A Rare Case Report

Shubha P. Bhat · Kishan H. L. Prasad ·
Vadisha Srinivasa Bhat · Rajeshwary Aroor

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Abstract Minor salivary gland tumors of oral cavity are very rarely encountered in day-to-day practice. Mucinous adenocarcinoma is very rare, and is commonly seen in elderly males. Palate and buccal mucosa is the commonest site. Wide local excision is the treatment. Histology shows tumor cells floating in pools of mucin, compartmentalized by fibrous tissue. Metastasis is a close mimicker, however with an evidence of negative scan and minor salivary gland proliferation in adjacent tissue diagnosis can be made. Mucin stains and immunohistochemistry can be used to support the diagnosis in problematic cases. We are presenting a rare case of mucinous adenocarcinoma-cheek in a 54-year-old male who came with gradually progressive swelling in the left buccal mucosa since 3 months and after wide excision, no evidence of recurrence was noted in 1 year and 3 months of follow up period.

Keywords Minor salivary gland · Mucinous adenocarcinoma · Buccal mucosa · PAS

Introduction

Minor salivary gland tumors are uncommon lesions, representing 9–23 % of all salivary gland tumors [1]. Mucinous adenocarcinoma (MAC) of minor salivary gland is a rare high-grade malignant tumor, comprising less than 0.1 % of epithelial salivary gland tumors [2]. Only few cases of mucinous adenocarcinoma-cheek are reported till date in the

literature. Males are frequently affected than females in the ratio of 3:1 with peak age of 5th–6th decade of life [1, 3, 4]. These patients usually present with painless exophytic growth, which clinically mimics adenoma, fibroma or pyogenic granuloma. Palate is the most common site [3]. Treatment is wide excision of the growth. The role of chemotherapy and radiotherapy is controversial. Prognosis depends on the stage of the disease [2]. We report a case of mucinous adenocarcinoma of minor salivary gland in a 54-year-old male who came with complains of growth in the left buccal mucosa. After wide excision, no evidence recurrence or nodal metastasis is noted in 1 year 3 months of follow up.

Case History

A 54-year-old male came with complains of gradually progressive painless swelling on the left cheek of 3 months duration. He was a chronic smoker. General and systemic examination was normal. Oral cavity showed an ulceronodular growth measuring 2.5 cm×2 cm was seen on the left buccal mucosa opposite to 2nd premolar and 1st molar (Fig. 1). On palpation, it was not tender, soft to firm in consistency, mobile and did not bleed on touch. There were no enlarged lymph nodes in the neck. Wide excision of the lesion was done with primary closure of the defect. Gross specimen was a single globular mucosa covered grey brown tissue mass measuring 2.5 cm×2 cm×1.5 cm. Cut surface was grey white with areas of hemorrhage. The lesional tissue was lined by oral mucosa with focus of ulceration covered by granulation tissue. Subepithelial stroma showed tumor cells in papillary fragments and tubules, floating in cystic spaces containing pools of mucin (Fig. 2). The tumor cells had round to oval moderately pleomorphic nucleus, vesicular chromatin and inconspicuous nucleoli (Fig. 3). Fibrous connective tissue was separating the mucin filled cystic spaces. Adjacent focus

S. P. Bhat · K. H. L. Prasad (✉)
Department of Pathology, K S Hegde Medical Academy of Nitte
University, Deralakatte, Mangalore, Karnataka, India
e-mail: dr_kishanpath@rediff.com

V. S. Bhat · R. Aroor
Department of Otorhinolaryngology, K S Hegde Medical Academy
of Nitte University, Mangalore, Karnataka, India 575018

Fig. 1 Clinical picture showing ulceronodular growth in the left buccal mucosa

showed minor salivary gland proliferation (Fig. 4). Periodic acid Schiff (PAS) stain was positive (Fig. 5). Features were suggestive of Mucinous adenocarcinoma. Resected margins were free of tumor. To confirm the presence of primary MAC comprehensive radiological examination using a computerized tomography (CT) of chest, abdomen, and pelvis was requested and was normal. Upper and lower gastrointestinal scopy was unremarkable. Patient is disease free since 1 year and 3 months with regular follow up.

Discussion

Mucinous adenocarcinoma is commonly seen in colon, pancreas, ovary, lung, prostate and breast [3–5]. Other atypical locations include nasal cavity, paranasal sinuses and mandibular ramus [5, 6]. Minor salivary gland mucinous adenocarcinoma is extremely rare neoplasm.

Fig. 2 Histopathology showing tumor cells in papillary fragments and tubules [H&E, $\times 100$]

Fig. 3 Tumor cells in glandular pattern having moderately pleomorphic nucleus with vesicular chromatin and nucleoli [H&E, $\times 400$]

Accumulation of genomic aberrations in protooncogenes, and tumor suppressor genes play an important role in carcinogenesis [5]. MAC of minor salivary gland is a tumor of older adults and has a male preponderance. In the available literature, we have found 22 published cases of intraoral minor salivary gland MAC, located in the palate (9 cases), buccal mucosa (5 cases), and floor of the mouth (3 cases), lips (3 cases), and tongue (2 cases). Some patients complain of a dull aching pain associated with the swelling. In our case, patient was a 54-year-old male who came with complains of gradually progressing painless swelling in the left buccal mucosa. Typical feature of MAC includes well-defined speckled pattern on surface of the lesion, which are related to mucus microcysts. These lesions pose diagnostic difficulties and are often misdiagnosed as pyogenic granuloma, mucus retention cyst, adenoma, or fibroma. Complete surgical excision is the primary therapy for an MAC [3, 4]. Wide surgical excision of the growth was done in our case.

Fig. 4 Histopathology showing minor salivary gland proliferation adjacent to tumor area [H&E, $\times 100$]

Fig. 5 Tumour cells showing Periodic acid Schiff stain positivity [$\times 400$]

Mucinous adenocarcinoma on histology shows the tumor cell nests float in large extracellular mucin pools. The tumor cells will have clear cytoplasm and small, round, darkly stained nuclei. Adjacent focus showed minor salivary gland proliferation. This finding suggest MAC of minor salivary gland origin [2–4].

Differential diagnosis includes metastatic mucinous adenocarcinoma of breast, mucinous eccrine carcinoma, and malignant salivary gland tumors like mucoepidermoid carcinoma and mucin rich salivary duct carcinoma [2, 3]. In our case, radiological examination using a CT of chest, abdomen, and pelvis was done and there was no evidence of primary elsewhere. There was evidence of minor salivary gland proliferation adjacent to the tumor, with skin and major salivary glands appearing normal, indicating its origin.

Diagnosis is also aided by staining with Periodic acid Schiff, mucicarmine or alcian blue to demonstrate extracellular and intracytoplasmic mucin. In cases where histology has no evidence of minor salivary gland origin, imaging showing mass lesion elsewhere or mucin stains are inconclusive; immunohistochemistry should be done for confirmation [2, 3]. Markers like cytokeratins, SMA, vimentin, and S-100 protein are positive in mucinous adenocarcinoma. CK 7(+)/CK 20(–) phenotype is considered as salivary primary, whereas CK 7(–)/CK 20(+) is considered as intestinal primary [3]. In our case, PAS was positive and immunohistochemistry was not done as many finding suggested minor salivary gland origin.

Mucinous adenocarcinomas are high-grade tumors with local recurrences and cervical lymph node metastasis. The role of adjuvant chemotherapy and radiotherapy is not well known [3, 5, 6]. Our patient is on regular follow up since 1 year 3 months with wide excision and there is no evidence of nodal metastasis or local recurrence.

Conclusion

Mucinous adenocarcinoma of minor salivary glands though rare, does exist. Clinicians should have a second thought whenever they come across mucus retention cysts or pyogenic granuloma especially in sites like palate and buccal mucosa. Wide surgical excision is the treatment of choice. As the histology mimics metastatic MAC from other sites, one should evaluate for the evidence primary of minor salivary gland origin. In cases of diagnostic dilemma, special stains and immunohistochemistry can be used to substantiate the diagnosis.

Conflict of interest None

References

1. Pires FR, Pringle GA, Almeida OP, Chen SY (2007) Intraoral minor salivary gland tumors: a clinicopathological study of 546 cases. *Oral Oncol* 43:463–470
2. Guimaraes KB, Santos BRM, Gordon MA et al (2012) Mucinous adenocarcinoma of minor salivary gland: case report and literature review. *J Clinic Case Reports* 2(7):1–4
3. Wyszynska-Pawelec G, Koryczan P, Zapala J et al (2013) Minor salivary gland mucinous adenocarcinoma of buccal mucosa- case report and review of literature. *Pol J Pathol* 64(4):312–316
4. Seoane J, Varela-Centelles, Lopez-Nino, Vazquez I, Abdulkader I, Garcia-Caballero (2010) Gingival mucinous adenocarcinoma of a minor salivary gland. *J Periodontol* 81(4):626–631
5. Uchida K, Oga A, Mano T, Nagatsuka H, Ueyama Y, Sasaki K (2010) Screening for DNA copy number aberrations in mucinous adenocarcinoma arising from the minor salivary gland: two case reports. *Cancer Genet and Cytogenet* 203:324–327
6. Hashitani S, Sakurai K, Naguchi K et al (2004) Mucinous adenocarcinoma with neuroendocrine differentiation of the mandibular ramus: report of a case. *J Oral Pathol Med* 33:59–63