Case Report

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Recurrent soft palatal tumour: from pleomorphic adenoma to mucoepidermoid carcinoma

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ABSTRACT

Cases of mucoepidermoid carcinoma (MEC) is common among major salivary gland neoplasms, however infrequent cases are reported for this growth over the palatal region. We presented a case of a 50-year-old gentleman whom presented with tumour over soft palate with 3 different histopathological reports. This gentleman had history of excision of right soft palatal mass on his first presentation which was reported as palatal pleomorphic adenoma, he however had tumour recurrence over the same site after 3 years whereby biopsy was read as squamous cell carcinoma. A wide local excision of the tumour was performed and final report was disclosed as mucoepidermoid carcinoma.

Keywords: Histology, Minor salivary gland, Mucoepidermoid carcinoma, Neoplasms, Soft palate

INTRODUCTION

Mucoepidermoid carcinoma (MEC) is a variant of malignant epithelial salivary gland neoplasm. It accounts for 10% of all salivary tumours and 30% of all salivary malignancies. The incidence of palatal MEC is 28% among the minor salivary glands and is usually located at junction of hard and soft palate. 5

Massao et al first reported MEC as a distinct pathologic entity in 1942.³ MEC typically arises from ductal epithelium and known to possess both epidermoid and mucinous components.¹ The pathological diagnosis of MEC is based on the presence of squamoid, mucin-producing, and intermediate cells. The lesions are classified as low, intermediate and high grade. They can be differentiated according to their appearances. Low grade lesions are well circumscribed and in homogenous

which may resemble pleomorphic adenoma, whereby high grade lesions are ill defined, invasive with possibility of nodal spread. Intermediate grade fall between high and low grade. About two-third of these tumour arise from major salivary gland commonly from parotid, and the rest from minor salivary gland.³

CASE REPORT

A 50-year-old Chinese man presented with recurrent swelling over the soft palate for one year duration. He had a history of excision of soft palate tumour 3 years prior and histopathological examination was reported as pleomorphic adenoma. Currently the swelling recurred at the same site of previous excision, which was slow growing and painless. He had mild throat discomfort but no dysphagia or odynophagia. There was no bleeding from the tumour. Examination revealed a mass at the soft

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palate, more towards the right side between the uvula and the palatoglossal arch (Figure 1). The mass appeared lobulated with smooth surface, measured approximately 3×2 cm. It was firm and non-tender on palpation. Nasolaryngoscopy showed normal mucosal surface over the pharyngeal surface of the soft palate. There was no significant palpable cervical lymph node.



Figure 1: Picture of the oral cavity shows the soft palatal mass predominantly on the right side, involving the uvula and the palatoglossal arch.

He was initially seen in a different centre where an incisional biopsy was performed and histopathology reported as stromal glandular structures within inflammed stroma suspicious of malignancy. In view of the inconclusive histopathology result, a repeated biopsy was done in our centre. The report confirmed that the tumour is a squamous cell carcinoma. Contrast enhanced computed tomography (CECT) of the neck showed the tumour was mainly confined to the right side of the soft palate measuring 3.2×2.2 cm. The mass was abutting the uvula and the base of tongue with no obvious infiltration. was no extension into nasopharynx pterygoplataine fossa. There were subcentimetre cervical lymph nodes bilaterally with the largest node at left cervical level II, measuring 1.2×0.9 cm. The swelling however was not concurrent with the intraoral symptoms and was an incidental finding.

He underwent wide local excision of the tumour under general anaesthesia. Intraoperatively, the mass was hard, extended medially to the uvula not crossing midline, laterally to superior constrictor muscle and partially involved the palatoglossal and palatopharyngeal muscles. Histopathological report of the tumour confirmed the diagnosis of mucoepidermoid carcinoma of intermediate grade, with close margin (Figure 2). The tumor extended into uvula bounded by non-ulcerated mucosa located very close (0.5 mm) from lateral margin, less than 1mm from anterior and superior margins and 6 mm from medial margin. Lymphovascular invasion, perineural invasion was not evident with brisk mitotic activity. He later received adjuvant radiotherapy. He is tolerating food well without aspirations or regurgitations. He is disease free at 6 month follow-up with slight hypernasality.

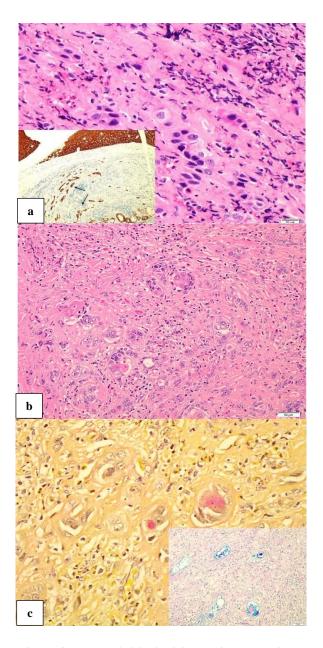


Figure 2 (a): The initial incisional biopsy specimen shows the tumor is predominantly comprised of epidermoid (squamoid) cells leading to the diagnosis of squamous cell carcinoma. Mucin secreting cells are not prominently seen here. (H&E 40×); these cells are highlighted by cytokeratin 5/6 immunostain (inset; $20\times$); (b) the repeated biopsy specimen revealed that the tumor is predominantly comprised two types of cells: epidermoid (squamoid) and intermediate cells. The predominantly seen epidermoid (squamoid) cells are arranged in clusters, nests, and trabeculae with large vesicular nuclei, single macro-nucleoli, and eosinophilic cytoplasm while the intermediate cells (arrows) are rather small with darker hyperchromatic nuclei and basophilic cytoplasm. Mucin secreting cells are not prominently seen here. (H&E 20x); (c). mucicarmine and alcian blue/periodic acid Schiff (inset) special immunohistochemical stains highlights the scattered mucin of mucin-producing cells.

DISCUSSION

Mucoepidermoid carcinoma is a common salivary gland malignancy.⁶ Following the parotid gland, hard palate minor salivary gland is the second most frequent location for MEC.⁶ It predominantly occurs at the fifth or sixth decade of life.³ MEC are histologically graded as low, intermediate, and high-grade based on the basis of architecture (cystic vs. solid), prevalence of mucus cells, and overall circumference of the tumor. Low-grade MECs are cystic, mucous cell-rich, and well-circumscribed; intermediate grade MECs are generally more solid and less circumscribed while high grade MECs displays one or more of the following features: nuclear anaplasia, necrosis, increased mitotic rate, and perineural, lymphovascular, or bony invasion.²

The diagnosis of MEC can be confusing due to its mixed cellular types. The potential differential diagnoses in a biopsy specimen include both non-neoplastic and neoplastic entities, such as necrotizing sialometaplasia, pleomorphic adenoma with squamous metaplasia, sclerosing polycystic adenosis, and squamous cell carcinoma. The lack of mucus cell in the initial biopsied specimen in this case leads to the misinterpretation as squamous cell carcinoma. Special immunohistochemical stains are needed to demonstrate the variable presence of mucin cells to establish the diagnosis and grading of MEC.

For definitive diagnosis of MEC, identification of mucus cells, squamous cells and intermediate cells are important. Unfortunately, all these smears are not present conspicuously in all cases, which leads to misdiagnosis of other malignant or benign entities. The histological grading of MEC is most predictive of prognosis and used to formulate a therapeutic plan. The treatment of choice for MEC is wide local excision with negative margins whereby high grade MEC usually requires excision radically accompanied with neck dissection.

Most of the cancer diagnosis are made on the basis of either histologic or cytologic evaluation. Inacurracy of diagnosis will have implications on patient's treatment and outcome. It was reported that anatomic pathologic errors accounts in the range of 1% to 43% of all specimens regardless of disease and origin, and 1% to 5% for oncology. These reported errors were however poorly characterized as there is no standardized measurement process.

CONCLUSION

The decision making of treatment holds the utmost importance of a disease and clinical correlation has

always been reminded throughout decades of medical practice. In this particular case, we opted for wide local excision without neck dissection when commonly a selective neck dissection would be considered for squamous cell carcinoma. The decision was made based on the clinical appearance of the tumour and patient's symptoms were not indicative of a high grade malignancy suspicion. The final intraoperative sample revealed an intermediate grade of mucoepidermoid carcinoma. Intermediate grade of intraoral minor salivary gland MEC has a very low recurrence (<10%) and rare incidence of metastases. Moraes et al suggested that wide local excision is sufficient to ensure tumor free surgical margin for low to intermediates intraoral minor salivary gland MECs. The morbidity risk was reduced by not opting for an aggressive surgical intervention.

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