Hyalinizing Clear Cell Carcinoma of Buccal Vestibule: A Rare Case

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Abstract

Hyalinizying clear cell carcinoma is a rare tumor which affects women more than men and is more common in minor salivary glands. The purpose of this paper was to describe a case of hyalinizing clear cell carcinoma of buccal vestibule in a 55-year-old woman. It is important to differentiate this neoplasm from other tumors with clear cell features because of their differences in treatment and clinical outcome.

Key words: Hyalinizying clear cell carcinoma, minor salivary gland, mucicarmine, salivary gland neoplasm.

Introduction

Hyalinizing clear cell carcinoma (HCCC) was first described as a distinct entity by Milchgrub et al. in 1994 (1,2). It almost exclusively affects the minor salivary glands of the oral cavity with a predominance of palate and tongue in adult females (3). It presents as a small, painless submucosal mass that is seldom ulcerated (1). The presence of clear cells in many types of tumors makes diagnosis difficult. In most cases; however, they constitute only a minor component and appropriate classification of the tumor is easily established on the basis of the typical features (1). When these cells make up the major cellular component, the diagnostic challenge is the greatest (3). Accurate diagnosis is vital as the treatment strategies may vary accordingly (4). The purpose of this paper was to describe a case of hyalinizing clear cell carcinoma of buccal vestibule in a 55-year-old female. We found that there were 68 cases of HCCC reported from 1980 to 2013 in the English language literatures of the PubMed.

Case Report

A 55-year-old female referred to the dental clinic with the chief complaint of a painless slow-growing submucosal mass on the right buccal vestibule of the mandible for16 months. The overlying mucosa was intact and pink in color and the lesion had a firm consistency.No significant enlargement of the lymph nodes in the neck was noted in extraoral examination. With provisional diagnosis of soft tissue tumors the excisional biopsy was performed. The gross appearance of the tumor was well-circumscribed and circular with a diameter of 2 cm (Fig. 1). The cut surface was creamy white and firm (Fig. 2). Histopathologic sections showed an epithelial neoplasm composed of bland cells, predominantly with clear cytoplasm. The tumor cells arranged in solid nests, trabeculae, islands and cords

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surrounded by a prominent hyaline stroma. The other population of cells had eosinophilic cytoplasm (Fig.3). Both populations of tumor cells lacked nuclear pleomorphism. Perineural invasion was also evident (Fig. 4).Based on the microscopic feature; the diagnosis of hyalinizing clear cell carcinoma was made. Mucicarmine staining and immunohistochemistry for S100 (Fig.5), SMA and Calponin (myoepithelial markers) were negative whereas Cytokeratin (Fig. 6) was positive. These findings confirmed the diagnosis. General evaluation including a chest X-ray and abdominal sonography showed no evidence of distant metastasis. As the tumor is low-grade, additional chemotherapy or radiotherapy was not indicated.There has been no evidence of recurrence in annual follow- up examinations since 2010.

Figure 1. Grossly, the tumor was well circumscribed

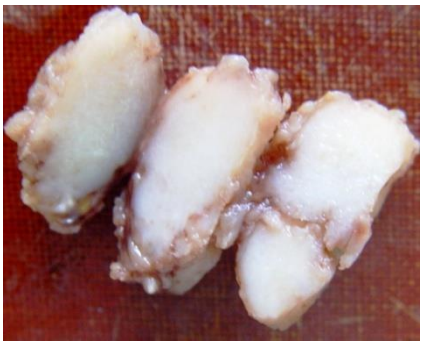

Figure 2. The cut surface was creamy white and solid

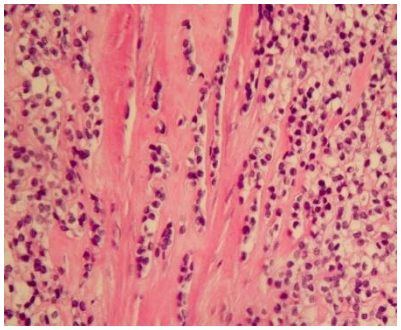

Figure 3. Histopathologic sections showed an epithelial neoplasm composed of bland cells, predominantly with clear cytoplasm (*200)

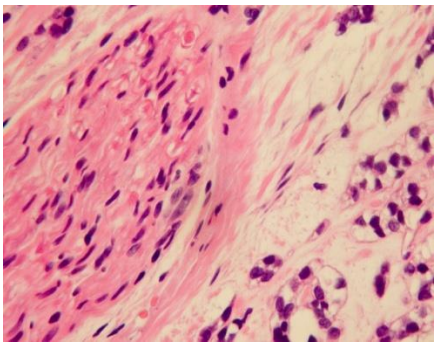

Figure 4. Perineural invasion (*400)

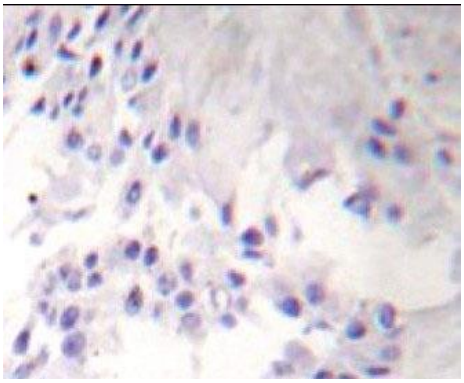

Figure 5. The tumoral cells were negative for S100 protein (*400)

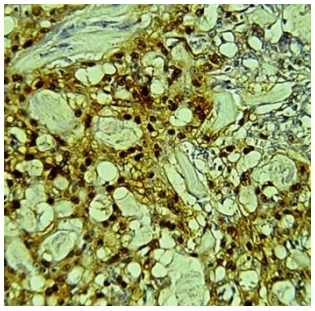

Figure 6. The tumoral cells were positive for Cytokeratin (*400)

Discussion

Clear cells can be found in numerous salivary and non-salivary tumors in the head and neck region (5). HCCC is composed of trabeculae, cords, islands and nests of glycogen-rich clear cells and eosinophilic cells circumscribed by a hyalinizing stroma (3). The abundant hyalinizing stroma is a distinctive morphological feature in distinguishingHCCC from the other salivary gland neoplasms with clear cell phenotype (6,7). The special staining and immunohistochemistry can help to distinguish these neoplasms (1,3,8,9). The clear cells are usually PAS positive and Mucicarmine negative (3). Both clear cells and eosinophlic cells were immunoreactive for Cytokeratin and negative for myoepithelial markers (1,3). The hyalinized stroma is mostly composed of collagen I and fibronectin (9). Barsky et al. (10) showed that tumor cells with myoepithelial differentiation produce laminin and collagen IV. Some studies suggested an intercalated duct origination for the tumoral cells (3).

Clear cell variant of mucoepidermoid carcinoma was Mucicarmine stain positive that can be very helpful for highlighting the mucous cells and distinguishing them from the other clear cell neoplasms (8). The epithelial-myoepithelial carcinoma (EMC) tends to show duct-like structures with a biphasic appearance. Stroma varies from loose fibrous to hyalinized tissue and the epithelial cells are positive for Cytokeratin and epithelial membrane antigen (EMA), whereas clear cells stain positively for myoepithelial markers such as S100 (11). Acinic cell carcinoma (ACC) shows different growth patterns including solid, papillary, microcystic and follicular and usually more than one cell type (1). The location of tumor is also helpful because EMC and ACC usually affecting the parotid glands (12). Metastatic tumors like renal cell carcinoma are associated with hemorrhage and necrosis and are positive for renal cell carcinoma antigen, CD10 and cytokeratin (9).

Local wide excision was the recommended treatment method (3). This tumor often follows an indolent course with a limited metastatic potential. It is therefore important to differentiate this entity from other more aggressive clear cell tumors including metastatic tumors (5).

Prognosis is excellent with few cases metastasizing to the lymph nodes and lung. However, recurrent cases with high-grade transformation were reported (12-15).

Conclusion

Oral pathologists should be accurate in differential diagnosis of the clear cell carcinomas because the treatment and prognosis of them are varied. Immunohistochemistry and special staining may be useful methods to distinguish these neoplasms. Because recurrent and metastatic cases have been reported, long term follow-up should be done.

References

1. Baghirath PV, Kumar JV, Vinay BH. Hyalinizing clear cell carcinoma: A rare entity. J Oral Maxillofac Pathol 2011;15:335-9.
2. Dardick I, Leong I. Clear cell carcinoma: review of its histomorphogenesis and classification as a squamous cell lesion. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2009;108:399–405.
3. Sun ZJ, Zhao YF, Zhang L, Zhang WF, Chen XM, He SG. Hyalinizing clear cell carcinoma in minorsalivary glands of maxillary tuberosity. Oral Oncol Extra 2005;41:306-10.
4. Ellis GL. Clear cell neoplasm in salivary glands: clearly a diagnostic challenge. Ann Diagn Pathol 1998;2:61–78.
5. Saleh KA, Nurishmah MI, Firouzeh GN, Goh BS. Primary clear cell carcinoma of minor salivary gland of the soft palate: a case report. Med J Malaysia 2012;67:335-6.
6. Milchgrub S, Gnepp DR, Vuitch F, Delgado R, Albores-Saavedra J. Hyalinizing clear cell carcinoma of salivary gland. Am J Surg Pathol 1994;18:74–82.
7. Batsakis JG, el-Naggar AK, Luna MA. Hyalinizing clear cell carcinoma of salivary origin. Ann Otol Rhinol Laryngol 1994;103:746–8.
8. Lai G, Nemolato S, Lecca S, Parodo G, Medda C, Faa G. The role of immunohistochemistry in the diagnosis of hyalinizing clear cell carcinoma of the minor salivary gland: a case report. Eur J Histochem 2008;52:251–4.
9. Félix A, Rosa JC, Nunes JF, Fonseca I, Cidadão A, Soares J. Hyalinizing clear cell carcinoma of salivary glands: a study of extracellular matrix. Oral Oncol 2002;38:364–8.
10. Barsky SH, Layfield L, Varki N, Bhuta S. Two human tumors with high basement-membrane-producing potential. Cancer 1988;61:1798-806.

11. Solar AA, Schmidt BL, Jordan RC. Hyalinizing clear cell carcinoma case series and comprehensive review of the literature. Cancer 2009;115:75–83.
12. O'Regan E, Shandilya M, Gnepp DR, Timon C, Toner M. Hyalinizing clear cell carcinoma of salivary gland: an aggressive variant. Oral Oncol 2004;40:348–52.
13. Jin R, Craddock KJ, Irish JC, Perez-Ordonez B, Weinreb I. Recurrent hyalinizing clear cell carcinoma of the base of tongue with high-grade transformation and EWSR1 gene rearrangement by FISH. Head Neck Pathol 2012;6:389-94.
14. Hijjawi SB, Abdullah SE, Abdelhadi K, Eyzaguirre E, Willis M, Abdulla NE.Hyalinizing clear cell carcinoma of the tonsil and its treatment. OralSurg Oral Med Oral Pathol Oral Radiol 2012;114:e32-6.
15. Milchgrub S, Vuitch F, Saboorian MH, Hameed A, Wu H, Albores-Saavedra J. Hyalinizing clear-cell carcinoma ofsalivary glands in fine-needle aspiration. Diagn Cytopathol 2000;23:333–7.

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