Hyalinizing Clear Cell Carcinoma of Maxilla

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Dear Editor,

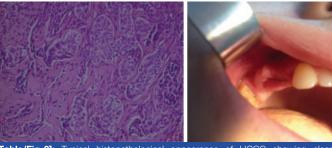
A 31-year-old female reported with a complaint of swelling and loose teeth in the right maxillary region. The patient had been aware of a progressive, painless swelling of right side and stated that she had mostly discomfort while chewing. The swelling started to grow approximately six to eight months previously. Three months later, the patient started to suffer from discomfort while chewing. The patient did not have a medical history of trauma, previous surgery or radiation therapy.

The head and neck examination revealed no evidence of submandibular and cervical lymphadenopathy, paresthesia or motor nerve deficiency. Intraorally, there was a swelling on the right site of the alveolar mucosa in maxilla between #14 and #13. Intraoral examination revealed missing teeth #16, #25, #26, #45, #46, #48 and #36, #38 teeth with grade 1 dental calculus. The swelling was tender on palpation and tooth# 14 was tender on percussion, however, there was no pus drainage from the area except a large swelling tissue in the posterior region. The panoramic radiography showed a radiolucent lesion in the maxilla with the root displacements of teeth #14 and #15 [Table/Fig-1].

An excisional biopsy was performed and the size of the specimen was 1.5 cm³. The specimen was a red and lobulated mass. The histologic examination showed trabeculae and cords of organized, wide clear cells with a vascularized and hyalinized stroma [Table/ Fig-2]. The specimen showed low activity of pleomorphism and mitotic activities, thus, the lesion indicated a low grade tumour. CK stains of the specimen demonstrated a marker status of negative CK20 but positive CK7 and on the other hand CK-19 showed mild, heterogeneous immunoreactivity in the tumour islands while vimentin was non-reactive in the tumour but there was PAS and S-100 protein positivity and vimentin was strongly positive in the fibrous stromal component only in this case. The final diagnosis was hyalinizing clear cell carcinoma in the maxilla. Since the pleomorphism and mitotic activities were low in tumour cells, the lesion was entirely excised with extraction of teeth #14 and #15. The patient was followed up at 10 days [Table/Fig-3], six month and one year and showed healing on the site.



[Table/Fig-1]: Initial panoramic imaging is showing a radiolucent lesion (arrows) in the maxilla which displaced the teeth.



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[Table/Fig-2]: Typical histopathological appearance of HCCC showing clear cytoplasm separated by hyalinised stroma (H&E, x100); [Table/Fig-3]: Post-operative intraoral Image after 10 days showing good healing.

Hyalinizing Clear Cell Carcinoma (HCCC) is an uncommon malignant salivary gland tumour with infiltrative growth pattern as the capacity for slow-growing lymphatic spread [1-3]. HCCC affects mainly the ducts of minor salivary glands without myoepithelial cell differentiation [1,4,5].

HCCC shows female predominance and older age in diagnosis, with the vast majority of cases occurring in the oral cavity [6]. In the present reported case, the lesion was seen in a 31-year-old female patient.

The classic clinical presentation of HCCC has been reported to be of a painless swelling in the mandible or maxilla but a number of patients report pain, ulceration, or alteration of speech [4]. In our case, the clinical symptoms were similar that was defined in the literature.

Several entities should be considered in the differential diagnosis of HCCC, including epithelial-myoepithelial carcinoma, myoepithelial carcinoma, clear cell mucoepidermoid carcinoma, acinic cell carcinoma, polymorphous low-grade adenocarcinoma, clear cell oncocytoma, and metastatic renal cell carcinoma can be included in the differential diagnosis [2,3].

In present reported case, histopathological report confirmed salivary gland neoplasm. The differential expression of CK7 and CK20 in carcinomas facilitates differentiation of primary salivary gland neoplasms from metastatic tumours and squamous carcinoma [7]. Thus, the expression of CK7, CK19 and CK20 was assessed by immunohistochemistry.

CONCLUSION

HCCC is difficult to diagnose, because it shares or overlaps immunohistological characteristics with other malignant tumours of the salivary glands. Maxillofacial surgeons as well as general practitioners must be aware of this rare entity especially in decision for treatment of these tumours.

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