

Oncocytic Carcinoma of Parotid Gland: A Case Report

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ABSTRACT

Introduction: Oncocytic carcinoma (OC) of salivary gland origin is an extremely rare proliferation of malignant oncocytes with adenocarcinomatous architectural phenotypes, including infiltrative qualities. To help clarify the clinicopathologic feature of this tumor group, herein, we report a case of oncocytic carcinoma arising from the salivary glands, together with radiological and immunohistochemical observations.

Case report: The tumor was reported in the parotid region of a 50 year old female. The tumors were unencapsulated and was seen invading into the nearby lymphatic tissues. The neoplastic cells had eosinophilic granular cytoplasm and round vesicular nuclei with prominent red nucleoli. Cellular atypia and pleomorphism was mild. Immunohistochemistry revealed DOG1 and p63 positivity. In summary, OC of salivary gland origin is a high-grade tumor, often with local recurrence, regional or distant metastasis, diagnosis of which is based on a combination of clinical and histopathological features.

Conclusion: Complete surgical excision is the treatment of choice while the role of radiotherapy or chemotherapy is controversial, and careful follow-up is necessary.

Keywords: Oncocytic Neoplasm, Parotid Gland, Carcinoma

INTRODUCTION

The diversity of epithelial salivary gland tumors as well as their rarity and varied morphological aspects often make the diagnosis of these neoplasm difficult. Salivary gland tumors are rare, occurring in 0.4-13.5 cases/100000 people. Most primary epithelial salivary gland tumors occur in the parotid gland.¹

Malignant salivary gland tumors represents 6% of head and neck cancers and 0.3% of all human malignancies. Oncocytic carcinoma is an extremely rare malignancy of salivary gland, accounting for 11% of all oncocytic salivary gland neoplasm, 0.5% of all epithelial salivary gland malignancies and 0.18% of all epithelial salivary gland tumors.

“Oncocyte” is derived from a Greek word “onkousthai”, meaning “increase in bulk”, swollen or tumor. This type of cells were originally referred to as “oncocytes” by Hamperl at 1931,² which are seen in organs like salivary gland, thyroid, parathyroid, and kidney, under various pathological conditions including neoplasia (such as oncocytomas).³

Oncocytic carcinoma of salivary gland origin is an extremely rare tumor, defined by WHO as a proliferation of cytomorphologically malignant oncocytes with adenocarcinomatous architectural phenotypes including infiltrative qualities, which may arise de novo and also can be seen in association with a pre-existing oncocytoma.⁴

A growing number of both benign and malignant salivary

gland tumors are characterized by recurrent genetic alteration particularly chromosomal translocation.⁵ Oncocytic carcinoma occurs mainly in the parotid gland and only a very few cases have been reported in the literature.^{6,7} We report a case of 50 year old female who presented with oncocytic carcinoma of parotid gland with metastasis to local lymph nodes.

CASE REPORT

A 50 year old female presented in the surgery OPD with left parotid region swelling since last 2 years with history of significant weight loss. There was no history of fever, pain, difficulty in chewing food or sudden increase in the size of swelling. Facial nerve involvement was excluded on thorough physical examination. MRI scan revealed an irregular hypointense lesion on T1/T2 predominantly involving the deep and superficial lobe of left parotid gland suggestive of a salivary gland neoplasm (Figure 1).

An FNA of the parotid swelling performed outside revealed features suggestive of an oncocytic neoplasm. A total conservative parotidectomy involving both the superficial and deep lobe of parotid gland (both enlarged) along with the resection of sentinel group of lymph nodes was performed, preserving the facial nerves and all its branches.



Figure-1: MRI showing irregular lesion in left parotid gland (red arrow)

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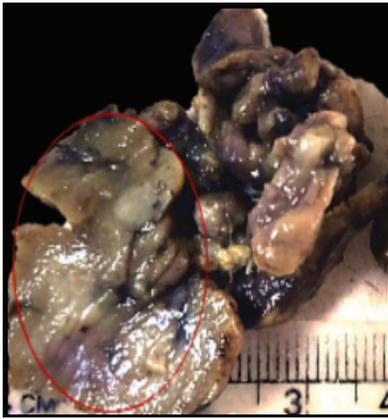


Figure-2: Gross image-Grey white tumor infiltrating the salivary gland

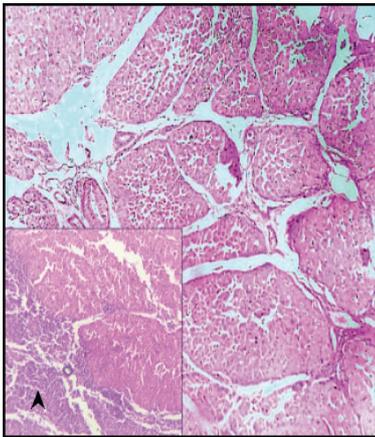


Figure-3: Tumor cells arranged in sheets and nests.(H & E; 100X). Inset shows lymph node(arrow head) with tumor deposit having similar morphology.

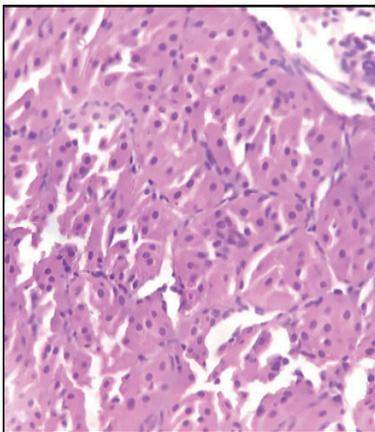


Figure-4: Tumor cells having abundant eosinophilic granular cytoplasm with small round nuclei and granular chromatin(H & E; 400X)

Gross

We received a gross specimen labelled as superficial lobe, deep lobe and sentinel lymph nodes. Superficial lobe measured 4x4x1.5 cm and was irregular on the outer surface. Cut section showed a heterogenous grey white tumor approximately measuring 1x0.8x0.3 cm (Figure 2).Similarly the deep lobe and the sentinel lymph node were received measuring 2x2x1.5 cm and 6x4x2 cm.

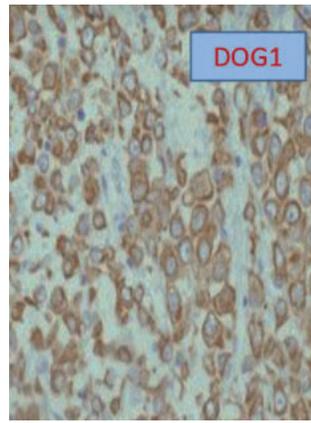


Figure-5: Tumor cells positive for DOG1 (IHC)

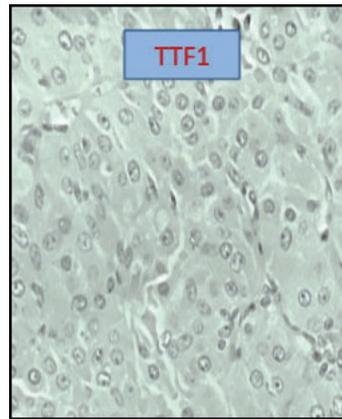


Figure-6: Tumor cells negative for TTF1.(IHC)

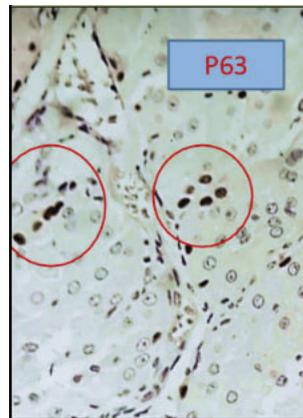


Figure-7: Tumor cells showing focal positivity for p63 in the myoepithelial cells (IHC).

Microscopy

Microscopic findings revealed an unencapsulated tumor composed of lobules, sheets and nests of tumor cells (Figure 3).The tumor cells had abundant eosinophilic granular cytoplasm with small round to oval nuclei with granular chromatin and small nucleoli in some of the tumor cells, with mild anisonucleosis(Figure 4). Tumor was seen infiltrating into the surrounding normal salivary gland tissue. Deep lobe also showed a tumor with similar morphology. Sentinel nodes showed tumor deposits(Figure 3 inset). Immunohistochemistry was performed and the tumor cells showed positivity for DOG1(Figure 5) and were negative for TTF 1(Figure 6), CK, Vimentin and Actin. Interspersed

myoepithelial cells in the tumor were positive for p63 (Figure 7). Ki67 proliferation index was low.

Therefore, based on radiological and morphological findings, a diagnosis of Oncocytic carcinoma with metastasis to sentinel lymph nodes was made.

DISCUSSION

Oncocytic carcinoma is a rare tumor arising in a major salivary gland presenting as 0.5% of all epithelial salivary gland malignancies and 0.18% of all epithelial salivary gland tumors.^{1,5,6} Most common age of presentation is 50-60 years with slight preponderance in males in two third of cases. Clinically presenting as a swelling over the pre auricular area with or without pain and sometimes involving the facial nerve. However, our case showed similar age of presentation with a painless swelling, uninvolving the facial nerve and presenting in a female.

Oncocytic carcinoma can arise from benign variant or can arise de novo. Its infiltrative growth pattern is represented by perineural, perivascular or lymphatic invasion, local lymph node metastasis and its morphological features in the form of cellular pleomorphism, necrosis, frequent mitosis which helps to identify its malignant nature. In our case, there was mild cellular pleomorphism and mitotic activity was low, there was no perineural, vascular or lymphatic invasion. However the tumor was seen infiltrating into the adjacent salivary gland tissue and metastatic deposits were seen in the sentinel lymph nodes. There was no evidence suggestive of distant metastasis.

The tumor need to be differentiated from other primary and secondary salivary gland malignancy having similar morphological features. These include primary salivary gland tumors like acinic cell carcinoma and metastasis from oncocytic cell carcinoma thyroid.

Acinic cell carcinoma has a characteristic microcystic solid and a papillary growth pattern with tumor cells showing amphophilic to basophilic cytoplasmic granules. On IHC, the tumor cells stain positive for DOG1 and are negative for p63. In our case, the tumor was composed of lobules, sheets and nests, with abundant eosinophilic cytoplasm. Tumor cells were positive for DOG1 on IHC. Interspersed myoepithelial cells were positive for p63. Metastatic oncocytic carcinoma originating from thyroid and metastasizing to salivary gland also shows tumor cells with similar morphology. However there should be a history of thyroid swelling and on IHC, the tumor cells stain positive for TTF1. In our case, thyroid examination was normal and the tumor cells were negative for TTF1, thus excluding oncocytic carcinoma originating from thyroid.

Despite being described in the early decades, very little is known about these rare tumors. The biological behaviour cannot be evaluated fully because of the paucity of cases reported and the lack of follow up information.⁸

However, the main treatment modality is surgery with or without adjuvant radiotherapy. Goode and corio have reported that tumors smaller than 2 cm in diameter have a better prognosis than the larger size tumors which was in our

case and hence a bad prognosis was assumed.⁹ Prophylactic neck dissection may be indicated for tumors larger than 2 cm in diameter which was performed in our patient. Follow up of some of the cases revealed that these tumors have the potential risk of developing distant metastasis and demand a long term follow up therapy.¹⁰ In oncocytic carcinoma of the head and neck, the presence of distant rather than local lymph node metastasis is the most important prognostic indicator.

CONCLUSION

Oncocytic carcinoma is an extremely rare salivary gland malignancy. The biological behaviour of this tumor has not been evaluated fully due to its low incidence and lack of follow up information. Thus, these patients require a careful long term follow up as distant metastasis appears to be the most important prognostic feature.

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