

Case Report

Oncocytic carcinoma of the right nasal side lacrimal apparatus with androgen receptor positive

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Abstract: Oncocytic carcinoma (OC) is a rare salivary gland malignant tumor without effective treatment method till now. We reported a 53-year-old man with tumor in the right eye with visual acuity 0.3. The serum of prostatic-specific antigen (PSA) was 13.2 ug/L. The tumor was resection. Microscopically, the lesion consisted of large atypical and pleomorphism epithelial tumor cells with fine, granular, eosinophilic cytoplasm composed of sheets, islands and nests with abortive glandular structures. Some cells were obviously mimicked prostate carcinoma. Immunohistochemically, the tumor cells were positive for AR, cytokeratin (AE1/AE3), CK18, CK19, alpha-1-antitripsin and negative for P504s, PSA, synaptophysin, chromogranin, and HMB-45. The pathological diagnosis was oncocytoma carcinoma of the right orbit. The prostatic was had no carcinoma with fine needle aspiration biopsy. The patient had radiotherapy and recurred in the 5 months after the first surgery. The patient died after the second surgery in the 6th month. There were some tumors with AR positive and androgen deprivation therapy were effect for them. We do not know that if all the OC were positive for AR and the androgen deprivation therapy was effective.

Keywords: Oncocytic carcinoma, androgen receptor, therapy

Introduction

Oncocytic carcinoma (OC) is a rare and aggressive subtype of salivary gland tumor with a few cases reports in the literature till now [1, 2]. It was a malignant tumor with poor prognosis. Till now there was no effective treatment for it except surgery.

Case report

Clinical summary

We described a 53-year-old man presented with a 15 months history of progressive swelling of the right eye without inducement. The patient had the eye pain and lacrimation. Ophthalmic examination showed visual acuity was unusual. The right eye visual acuity was 0.3 and the left one was 1.2. There was a firm mass without boundary in the right lower eye lip caused 2 mm superior of the right eye globe. Ocular motility testing showed limited to the upward and downward and adduction. Laboratory examination showed that serum was PSA13.2 ug/L. The other tumor markers

include AFP and CEA were normal. CT scan revealed a mass located in the right inside orbit infiltrated the periphery muscles of the right eye (**Figure 1**). Others were normal. The tumor was resection including the involved muscles around the tumor.

Histopathological findings

Microscopically, the lesion consisted of large atypical and pleomorphism epithelial tumor cells with fine, granular, eosinophilic cytoplasm composed of sheets, islands and nests with abortive glandular structures (**Figure 2**). Some cells lacked prominent eosinophilic cytoplasm and glandular structure was obviously mimicked prostate carcinoma (**Figure 3**). There were multinucleated cells. Mitotic figures were rare. Necrosis was not been seen. The mass was unencapsulated and invaded the muscle and nerves. Vascular invasion was not found. Immunohistochemically, the tumor cells were positive for AR (**Figure 4**), cytokeratin (AE1/AE3), CK18, CK19, alpha-1-antitripsin and negative for P504s, prostatic-specific antigen (PSA), Myoglobin, Myogenin, Desmin, GFAP,

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Figure 1. CT scan revealed a homogenous, irregularly shaped hyper dense lesion located in the right inside orbit with indistinct borders. It infiltrated the medial and inferior rectus muscles of the right eye. The nasal side orbit including the lacrimal sac and duct was involved. There was no bone invaded.

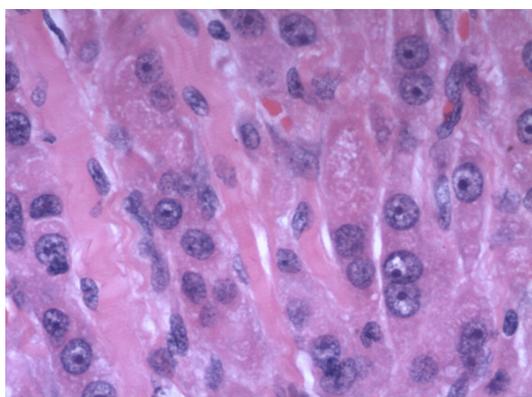


Figure 2. Microscopically, the lesion consisted of large epithelial cells composed of sheets, islands and nests with abortive glandular structures. The atypical and pleomorphism tumor cells were round to polyhedral with fine, granular, eosinophilic cytoplasm and central or peripheral located, round vesicular nuclei with prominent nucleoli (hematoxylin and eosin; original magnification $\times 40$).

S-100 protein, thyroid transcription factor-1, synaptophysin, chromogranin, and HMB-45. The pathological diagnosis was oncocytoma carcinoma of the right orbit while suggested to check the prostate.

The patient had radiotherapy after the operation. He had prostate eight sports fine needle

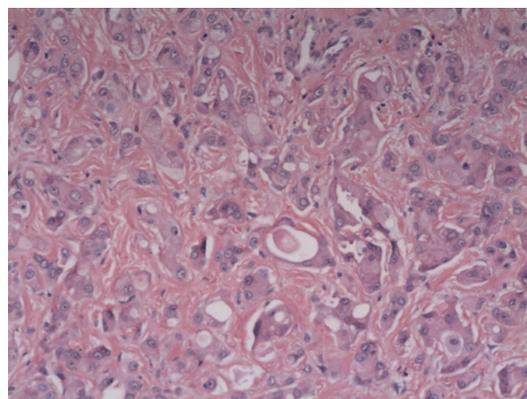


Figure 3. Some cells lacked prominent eosinophilic cytoplasm and glandular structure was obviously mimicked prostate carcinoma (hematoxylin and eosin; original magnification $\times 40$).

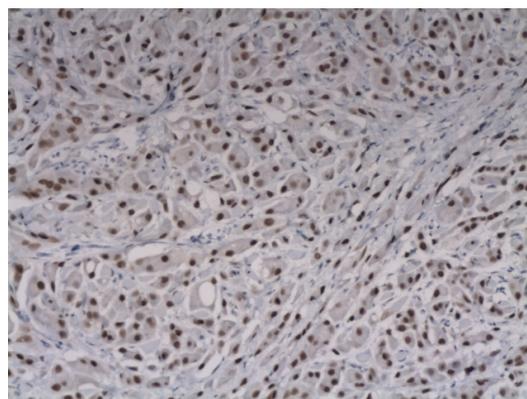


Figure 4. The tumor cells were positive for AR (original magnification $\times 40$).

aspiration biopsy and showed prostate hyperplasia without prostate carcinoma. 5 months later, the tumor recurred. The clinical symptom was the same as the first one. CT showed it involved almost all the periphery tissue of the right orbit included the osseous nasolacrimal duct. The ophthalmologists had the right eye-ball resection.

Macroscopically, there was a whole eyeball with gray tumor tissue major located in the nasal side. The lacrimal apparatus was not been identified. Microscopically, it was the same as the first one. The conjunctiva, the eye lip was all infiltrated. Immunohistochemistry was the same as the first one. That was to say AR was positive in the recurred tumor cells. The pathology diagnosis was oncocytoma carcinoma. After the second surgery, the patient had radiotherapy.

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He died in the 6th month after the second surgery.

Discussion

OCs mainly presented in the organs such as parotid gland, thyroid, breast, nasal cavity and other minor salivary glands [1, 2]. There were few reports of the periocular region OC. The etiology is unclear. In our case, the neoplasm located in the right lower nasal side orbit. It was too large to be identified the primary region such as lacrimal sac, lacrimal caruncle or nasolacrimal duct.

In microscopically, we first diagnosed OC but need differential diagnosed with the rhabdomyosarcoma because of the histology and prostate carcinoma because of the higher of serum PSA. The rhabdomyosarcoma cells also have abundant, eosinophilic granular in the cytoplasm with well defined borders and small, round, centrally or peripherally located nuclei and prominent nucleoli. In immunohistochemistry, myoglobin and desmin are positive while OC negative. The serum PSA of the patient was higher. The AR of the tumor cells was positive. We had never found a report about AR-positive OC. So prostate carcinoma must be differential diagnosis. However, other prostate carcinoma markers of the tumor cells such as P504s and PSA were negative. There was no prostate carcinoma in fine biopsies.

Surgical especially radical resection is the widely accepted treatment for OC [2]. The adjuvant radiotherapy was also necessary after surgery. But the prominent character of OC was multiple local recurrences and regional or distant metastases even had radiotherapy therapy [2]. Our case recurred in the 5th month after the first surgery even he had radiotherapy after the tumor resection. The recurred neoplasm was more progressive. So he had the radical resection. That is to say, till now there was no effective treatment for the high grade disease.

We all know that there were some tumors with AR positive not only in prostate carcinoma and androgen deprivation therapy were effect for them [3-5]. In our case, the primary and recurred tumor cells were positive for AR. To our knowledge, this is the first time that the OC patient with AR-positive has been described. We couldn't certify that all OCs were positive for

AR because of rare cases. So the authors hope that this report will encourage others to report further OC cases with AR positive. Thus, androgen deprivation therapy in patients especially with recurrent or disseminated OC patients may be beneficial.

Disclosure of conflict of interest

None.

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