

Limbal Mass as a Presentation of Parotid Gland Undifferentiated Carcinoma: A Case Report

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Abstract- Metastatic neoplasms to the ocular surface are extremely rare. Here, we describe a case of A 56-year-old man developed simultaneously a limbal and parotid gland masses in his left side. He underwent excisional biopsy of limbal mass, parotidectomy, and systemic evaluation. Histopathologically, multislice sections of both limbal and parotid masses disclosed an undifferentiated carcinoma of both sites. Further evaluation revealed no other site of involvement and metastasis. The patient underwent systemic chemotherapy and local radiotherapy for parotid gland tumor. Distal metastasis from undifferentiated carcinoma of the parotid gland to ocular surface is very rare and to the best of our knowledge has not been previously reported. This is the first report of the manifestation of metastasis from undifferentiated carcinoma of parotid gland origin to the limbus. The limbal mass may be the initial manifestation of metastasis from this origin and should be considered in the differential diagnosis of a metastatic limbal tumor.

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Introduction

Metastatic tumors to the ocular surface are a rare event and often reflect an advanced stage of disseminated malignancy (1). Conjunctival metastasis usually occurs as a part of the widespread metastatic disease. Conjunctival metastasis has arisen from cancer of breast, lung, and cutaneous melanoma reported (2-3). Rarely, metastasis can be the presenting feature with no evidence of any systemic malignancy (4).

Undifferentiated carcinoma of the salivary gland is a rare malignant tumor of the epithelial structure that is too poorly differentiated to be placed in any of other groups of carcinoma (5). Sinonasal undifferentiated carcinoma (SNUC) is a rare, highly aggressive and clinicopathologically distinctive carcinoma of uncertain histogenesis. Also, undifferentiated carcinoma of the liver is very rare, accounting for less than 2% of all epithelial liver tumors (6).

The design and objectives of the study were explained to our patient and written informed consent was obtained. The consent for publishing this case report was taken from the institute's ethics committee; Amir-Almomenin ophthalmology research center institute of

medical sciences ethics committee.

We report here an extremely rare case of isolated distal metastasis of undifferentiated carcinoma of parotid gland to the limbal area of the ipsilateral eye, and according to our knowledge, this is the first report of ocular surface metastasis from undifferentiated carcinoma of the parotid gland.

Case Report

Here we describe a case of 56-year-old man referred with a 3-weeks history of enlarging mass lesions on his left parotid gland (Figure 1) and left eye. His medical history was not significant. His vision was 20/20 in each eye, and a complete ocular examination revealed normal findings, except for a mass lesion involving the superior limbal area of the left eye. The mass was hard, immobile, and significantly vascularized with feeding vessels, the size of the lesion was 6-7 mm with engorged vessels and reddish color. The lesion extended to the cornea and bulbar conjunctiva (Figure 2). Also, the parotid gland mass was large, hard, and immobile. The diagnostic impression on clinical findings was probable metastatic neoplasm from parotid gland tumor. He

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Limbal mass

underwent excision of both parotid gland and limbal tumors with parotidectomy of the left side and excisional biopsy of limbo-conjunctival mass (Figure 3). The systemic evaluation revealed no other site of involvement and metastasis.



Figure 1. Tumor of the left parotid gland

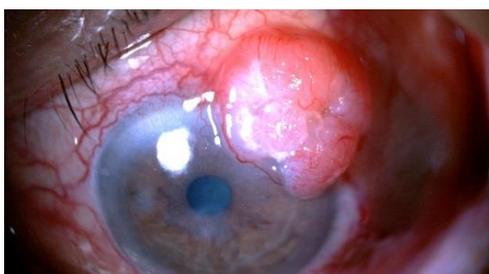


Figure 2. Metastatic tumor of the limbo-conjunctival region of the left eye



Figure 3. Post-operative site of excisional biopsy from the limbo-conjunctival region

Histopathologic evaluation (in multislice sections) of the limbo-conjunctival tumor and parotid gland tumor disclosed a neoplasm that was located in the limbo-conjunctival stroma and parotid gland stroma. The highly cellular tumor was composed of malignant cells and displayed very poorly differentiated epithelial

structures with the high mitotic count. The tumor cells were small or intermediate hyperchromatic cells with a high nuclear to cytoplasmic ratio and frequent mitosis. No evidence of differentiation was observed. Necrosis and bleeding were prominent features. An immunohistochemical (IHC) study using several epithelial, mesenchymal, and neuroendocrine markers was performed on both samples. A panel of immunohistochemical studies were used with antibodies against keratin, cytokeratin, epithelial membrane antibody (EMA) for membrane of epithelial cells, vimentin for mesenchymal cells, smooth muscle actin (a-SMA) and desmin for differentiation of muscle cells, S-100 protein for neurogenic cells and HMB-45 for malignant melanoma were negative for this samples.

The patient underwent systemic chemotherapy and local radiotherapy for parotid gland region by an oncologist. The patient had not recurrence of the limbo-conjunctival metastatic tumor in more than 11 months' follow up.

Discussion

The ocular surface area is rarely the site of metastatic neoplasms. Metastasis to the conjunctiva from primary lung cancer, mesothelioma, breast cancer, cutaneous melanoma, laryngeal carcinoma and unknown origin were reported (1-4). A rare case of distant metastasis from high-grade mucoepidermoid carcinoma of the parotid gland to the ipsilateral bulbar conjunctiva of the eye was reported (7).

Our case demonstrated limbo-conjunctival metastasis from primary parotid gland cancer that was the first sign of undifferentiated carcinoma of this region. Even though the patient had no liver and bone and other site metastasis.

Undifferentiated carcinoma of the salivary glands is classified as a malignant tumor of epithelial structure that is too poorly differentiated, devoid of any phenotypic expression under light microscopy, to be placed in any of the other groups of carcinoma. The incidence of undifferentiated carcinoma reportedly ranges from 1 to 5.5% of all malignant neoplasms of parotid gland origin. There is a male preponderance, but no data on its geographic distribution is available. In general, the prognosis for undifferentiated carcinoma of the salivary gland is poor. Some authors have indicated that tumors exceeding 4 cm diameter or at an advanced stage of disease indicate poor prognosis (5).

To differentiate undifferentiated carcinoma from adenocarcinoma, malignant melanoma or mesenchymal

tumors, mucin stains such as Alcian blue or PAS stain and immunohistochemical studies are useful. Using Alcian blue or PAS staining, undifferentiated carcinoma can be differentiated from adenocarcinoma. Malignant melanoma can be differentiated from undifferentiated carcinoma by immunohistochemical studies using HMB-45. Mesenchymal tumors can be differentiated from undifferentiated carcinoma using immunohistochemical studies such as vimentin, α -SMA, desmin and S-100 protein (8). All of these studies were negative in our patient. Undifferentiated carcinoma should be distinguished from too poorly differentiated carcinoma with small differentiated portions, the prognosis for both tumor types remain poor despite radical surgery (9).

Here, we emphasize that there was no recurrence in the site of limbal metastatic tumor excision at least 11 months after surgery. Thus the more aggressive procedures such as enucleation and orbital exenteration should not be considered if there is no intraocular tumor invasion.

Distal metastasis from undifferentiated carcinoma of parotid gland to ocular surface is very rare and to the best of our knowledge has not been previously reported. It was the first report from ocular surface metastasis originating from undifferentiated carcinoma of the parotid gland. The limbal mass may be the initial manifestation of metastasis from undifferentiated carcinoma of parotid gland origin and should be considered in the differential diagnosis of a metastatic limbal mass.

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