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Case report

Report of a unique case of myoepithelial carcinoma of left parotid gland with metachronous bilateral cavernous sinus metastasis

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ABSTRACT

Myoepithelial carcinoma (MC) is a rare, locally aggressive malignant neoplasm of the salivary glands. Only few evidences on its metastatic behavior are available in the literature. We herein present a unique case of MC of left parotid gland which metastasized to bilateral cavernous sinuses. The patient was successfully treated with palliative radiotherapy and chemotherapy.

Introduction

Myoepithelial carcinoma (MC), also known as malignant myoepithelioma, is a rare locally aggressive primary salivary gland neoplasm. However, it has also been identified in other sites like upper aerodigestive tract, breast, skin and other soft tissues [1]. The recognition of this tumor is challenging as there is considerable morphologic heterogeneity and exact histopathologic and immunohistochemistry profiles remain to be delineated. It is composed of neoplastic cells with prominent myoepithelial differentiation and characterized by infiltrative growth and potential for distant metastasis [1]. Due to its rarity, the clinical course of this tumor including its metastatic behaviour and the treatment are not well defined yet. Few scarce reports on its metastatic potential are available in the literature. The metastatic sites included the lungs, kidneys, cervical lymph nodes, bones, thyroid and the scalp [2]. We herein present a unique case of MC of left parotid gland with metastasis to bilateral cavernous sinuses. The patient was successfully treated by palliative radiotherapy and chemotherapy. To the best of our knowledge, this is the first reported case of its kind.

Case report

A 46-year-old gentleman, known hypertensive, adequately controlled with anti-hypertensive medications initially presented to our clinic in September 2015 with left parotid swelling for five years which was rapidly increasing for last two months associated with left neck swelling for two months. Local examination showed a 10 × 8 cm left parotid swelling which was displacing ear lobule superiorly and one lymph node palpable at submandibular region with no sign of facial nerve palsy. Core biopsy from parotid mass revealed features of poorly differentiated carcinoma. Contrast enhanced computed tomography (CECT) scan of neck, chest, abdomen and pelvis was done which showed a left parotid superficial lobe mass and enlarged conglomerate necrotic left cervical lymphadenopathy (Fig. 1). He underwent left total parotidectomy along with left radical neck dissection on October 2015. Post operative histopathologic examination (HPE) revealed a tumor showing feature of myoepithelial carcinoma (Fig. 2). Tumor cells were positive for cytokeratin (CK), Epithelial membrane antigen (EMA), S100 (focally) and p53 and negative for CD10 and smooth muscle actin (SMA) (Fig. 3). Five out of sixteen lymph nodes dissected were involved by tumor, three out of which were showing soft tissue extension. He developed right cervical lymph node swelling after a gap of one month for which he underwent type-III right modified

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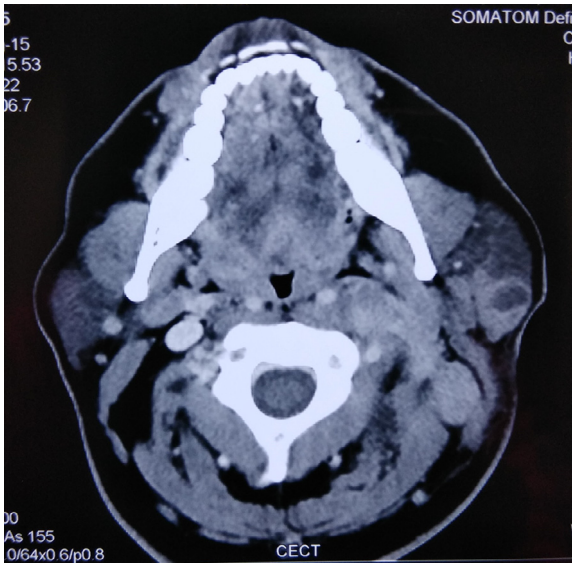

Fig. 1. CECT scan of neck showing a left parotid superficial lobe mass and enlarged necrotic left cervical lymphadenopathy.

neck dissection. Postoperative HPE of specimen revealed two out of fifteen lymph nodes involved by tumor. Then he received adjuvant radiotherapy of dose 60 Gray in 30 fractions over six weeks to parotid bed and bilateral neck till February 2016. He was disease-free for one year. Then he presented with bilateral diminished vision and ptosis in January 2017. Magnetic resonance imaging (MRI) of brain showed soft tissue density mass lesion in both cavernous sinus extending in bilateral orbital apices suggesting bilateral cavernous sinus thrombosis (Fig. 4a). He was started on anticoagulant therapy immediately but his symptoms were worsening. As it was too risky to perform biopsy from this site, patient was administered palliative radiotherapy of dose 30 Gray in10 fractions over two weeks to cavernous site by three dimensional conformal radiotherapy after obtaining proper informed consent from the patient. His vision started improving and ptosis recovered post treatment. Then he was also put on systemic chemotherapy and was planned for six cycles of three weekly Cisplatin, Doxorubicin and Cyclophosphamide. Follow up MRI brain was done five months post radiotherapy which showed near complete resolution of thrombosis (Fig. 4b).

Discussion

Myoepithelial carcinoma (MC) of the parotid gland represents less than 2% of all salivary gland tumors [3]. Owing to its rarity, natural history of this malignancy and its management are not clearly defined. MC usually present as a painless swelling but they are more frequently symptomatic than benign lesions: pain, facial nerve palsy and fixation of the mass to the underlying structures are the most presented symptoms [4]. Similarly in our presented case, the gentleman presented with parotid and neck node swellings; however it was not associated with pain or facial nerve paresis. MRI is the preferred imaging study to evaluate salivary gland masses as it delineates the tumor more sharply than does the CT scan and it is helpful in detecting facial nerve involvement [5]. Earlier classified as mixed tumors, the majority of MCs develop in a pleomorphic adenoma; in most cases they are low-grade malignancies. When they appear de novo, as in our case, the malignancy is often high grade. IHC of MCs show that these tumors usually express CK and EMA, calponin (75%) and SMA (50%). Other markers are expressed in varying degrees: S-100 protein (100%), Vimentin (100%),and gliofibrillary protein acid (31%) [3]. In our case, the tumor cells are immunopositive for Pancytokeratin, S100 (Focal positivity), EMA (Focal) and P53. However, the tumor cells are immunonegative for CD10 and SMA.

Although there are no clear guidelines for the management of MC, wide surgical excision for localized tumors with or without cervical lymph node dissection is the mainstay of therapy and adjuvant radiation therapy is commonly considered [3]. Compared to its benign counterpart (myoepithelioma), MCs are associated with increased frequency of local recurrences and metastases, which warrants close clinical follow-up. Adjuvant chemotherapy or radiotherapy may help to prevent metastasis and recurrence [6]. Guyan et al. followed up 18 cases of myoepithelial carcinoma over 17 years and found that this tumor metastasized to the regional lymph nodes, lungs, bones and liver [7]. Our case is unique because, to our knowledge, no cavernous sinus metastases from a myoepithelial carcinoma has been reported yet. The presented case was managed by local conformal radiotherapy and systemic chemotherapy to which he responded significantly, both clinico-radiologically.

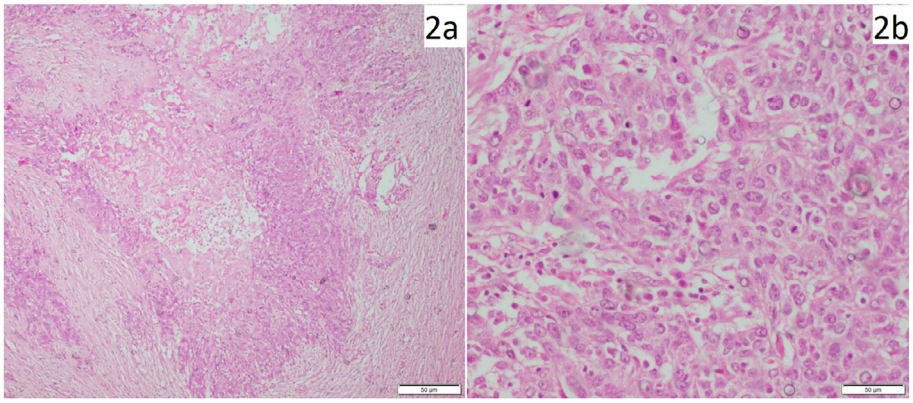

Fig. 2. (a) Tumor cells are arranged in irregular nodules with central necrosis which are separated by desmoplastic stroma (Hematoxylin and eosin stain, 40x); (b) The tumor cells are polygonal to spindle with moderate amount of pale eosinophilic to vacuolated cytoplasm. The nuclei are round to oval with moderate nuclear atypia, vesicular chromatin and conspicuous to prominent nucleoli. There are frequent mitotic figures.

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