

Sebaceous Carcinoma of the Eyelid Presenting As Metastasis to the Parotid Gland: A Rare Occurrence

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Abstract

Sebaceous carcinoma of the eyelid frequently present with metastatic disease involving regional lymph nodes. However, the metastasis to parotid gland is very rare and can be mistaken for primary parotid gland tumor. We present a case of 53-year-old female who presented with right parotid swelling. MRI was suggestive of a pleomorphic adenoma. She was operated for sebaceous carcinoma of the eyelid four years back. Characteristic cytological findings of a mixture of high-grade malignant cells with vacuolated cells along with clinical correlation were helpful in the diagnosis of this tumor following which the patient underwent parotidectomy.

Keywords: Sebaceous carcinoma, Eyelid sebaceous carcinoma, parotid metastatic malignancy.

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I. Introduction

Sebaceous carcinoma is a rare and aggressive tumor showing high rate of local recurrence and metastasis.^[1] Most cases of sebaceous carcinoma involve the eyelid but it has also been reported in extraocular sites including the parotid gland. Metastatic spread of this tumor occurs by continuous growth, lymphatic and hematogenous routes. Most common sites of local metastasis are preauricular, parotid and cervical lymph nodes.^[2,3] Sebaceous carcinoma presenting as metastasis to parotid gland is a rare entity and can pose a diagnostic challenge on cytology as it has to be differentiated from primary salivary duct carcinoma^[2]. We report a rare case of metastatic sebaceous carcinoma to parotid gland presenting 4 years after primary eyelid involvement highlighting its cytological and histological features.

II. Case Report

In this study we present a rare case report of a 53-year female came with the complaints of a gradually increasing swelling in the right preauricular region for 1 year. On examination, swelling measured 5x6 cm and is firm, mobile and non-tender. **Magnetic Resonance Imaging (MRI)** face was done before and after intravenous injection of contrast. MRI revealed a well demarcated lobulated heterogeneously enhancing soft tissue lesion in the superficial lobe of the right parotid gland measuring 2.7cmx2.3cmx3.1 cm in AP x TR x CC dimension. It does not involve deep lobe of the parotid gland. It abuts mandibular condyle; however, it does not seem to be involved. Multiple subcentimetric sized lymph nodes were seen in bilateral cervical Ib, II, and III levels. An enlarged lymph node of size 1.7 X 1.4 cm in size at right level II level showed partial central necrosis within. Rest of the facial structures and soft tissue were unremarkable. A final impression of pleomorphic adenoma was given in MRI and was advised for histopathological correlation.

FNAC of the right parotid swelling showed a cellular aspirate with two population of cells comprising of cohesive clusters of epithelial cells mixed with large cells showing vacuolated bubbly cytoplasm with high nuclear/cytoplasmic ratio, anisonucleosis, pleomorphism and giant cells with foamy cytoplasm. [Figure 1]. Background showed lipid vacuoles and necrosis. A diagnosis salivary gland duct carcinoma was considered. On further enquiry patient gave history of being operated for an eyelid mass operated four years back at another hospital. Earlier biopsy was retrieved and showed features of sebaceous carcinoma. Cytological features were reviewed and a diagnosis of metastatic sebaceous carcinoma was made.

Positron Emission Topography scan showed hypermetabolic lesion in the right parotid gland with heterogenous FDG uptake with SUV_{max} of 5 with loco-regional lymphadenopathy with no eyelid involvement. FDG avid bilateral enlarged cervical lymph nodes are seen at level II, III, and IV, largest being right level III with a SUV_{max} of 5. Patient underwent right superficial right parotidectomy with right sided selective neck dissection. Gross examination showed a solid grey-white firm tumor measuring 4.5 cm x 3 cm x 3cm involving the superior part of parotid gland [Figure 2].

Histopathological examination of multiple sections studied showed normal salivary gland along with tumor cells arranged in lobules and nests which were composed of mixture of malignant epithelial cells some showing foamy vacuolated cytoplasm. Tumor cells showed high grade nuclear features and brisk mitosis [Figure 3]. Some lobules showed comedo necrosis. There was no evidence of lymphovascular and perineural invasion All lymph nodes dissected were free of tumor and showed granulomatous inflammation. Based on the clinical and histopathological features a diagnosis of metastatic sebaceous carcinoma of parotid gland was given.

Figure 1: (A) 10x : PAP stained cellular smears show two population of cells comprising of epithelial cell cluster mixed with cells with vacuolated and bubbly cytoplasm amidst giant cells in a lipid rich necrotic background. (B) Epithelial cell cluster comprising of mixture of pleomorphic cells with hyperchromatic nuclei along with (C, D : H & E stain) epithelial cells with high N/C ratio, anisonucleosis, nuclear pleomorphism and prominent nucleoli. (E, F: H&E) Sebocytes/ Cells with sebaceous differentiation. (G: PAP Stain & H: H& E) showing giant cell with foamy cytoplasm.(Contd...)

III. Discussion

Sebaceous carcinoma arises predominantly in periocular area and ocular adnexa and has great propensity for metastasis. This tumor is generally seen in older individuals between 50 to 70 years of age with a female predominance.^[3] The early diagnosis of these tumors is missed as they mimic various inflammatory conditions like chalazion, blepharo-conjunctivitis and can present with metastatic disease. Our patient was also an elderly female who presented with parotid metastasis in absence of recurrence.

Secondary neoplastic lesions of parotid gland are rare and comprise 6-8% of all parotid tumors with 90% of them arising from a head and neck primary. This metastasis is through lymphatic spread as many lymph nodes are present in preauricular area and within the parotid gland which drain superficial areas of face and scalp. Most common primary tumors causing metastasis to parotid gland are squamous cell carcinoma, melanoma and basal cell carcinoma.^{[4],[5]} Metastatic sebaceous carcinoma to the parotid is very rare with only four cases reported in literature.^[2] Margarita V et al have reported a case of recurrent sebaceous carcinoma of eyelid with parotid gland and lymph node metastasis 4 year after previous surgery.^[2] However, in our case there was no recurrence at primary site and no lymph nodes were involved.

FNAC is a useful technique for preoperative assessment of metastatic parotid tumors and they show 71.9% sensitivity when correlated along with history, clinical examination and imaging.^[4] Cytological features of sebaceous carcinoma described are single or loosely cohesive cell clusters with large round to oval vesicular nuclei and prominent nucleoli, varying degrees of cytoplasmic vacuolization, multinucleate giant cells and lipidic or necrotic background.^{[1],[6],[7]} The presence of vacuolated cells with bubbly cytoplasm and central nuclei with a lipid rich background suggest sebaceous differentiation and can aid in the diagnosis as in the present case.

Histopathology of sebaceous carcinoma shows lobules of undifferentiated cells mixed with cells showing sebaceous differentiation with foamy vacuolated cytoplasm.^{[7],[8]} In our case all the above features were observed in both primary eyelid tumor as well as in sections from the parotid gland metastasis. Our cytological findings were also in concordance with histological findings.

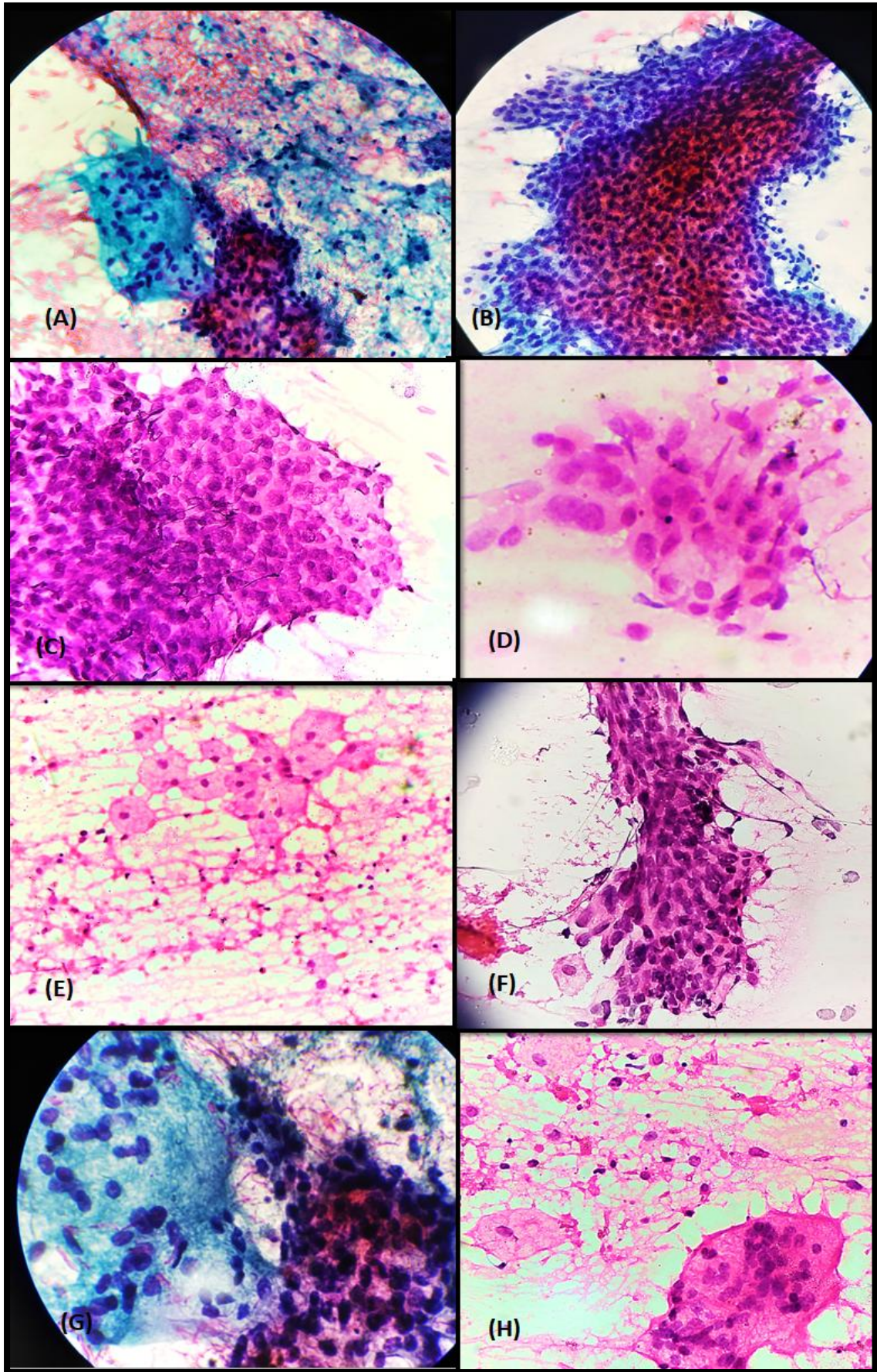


FIGURE 1

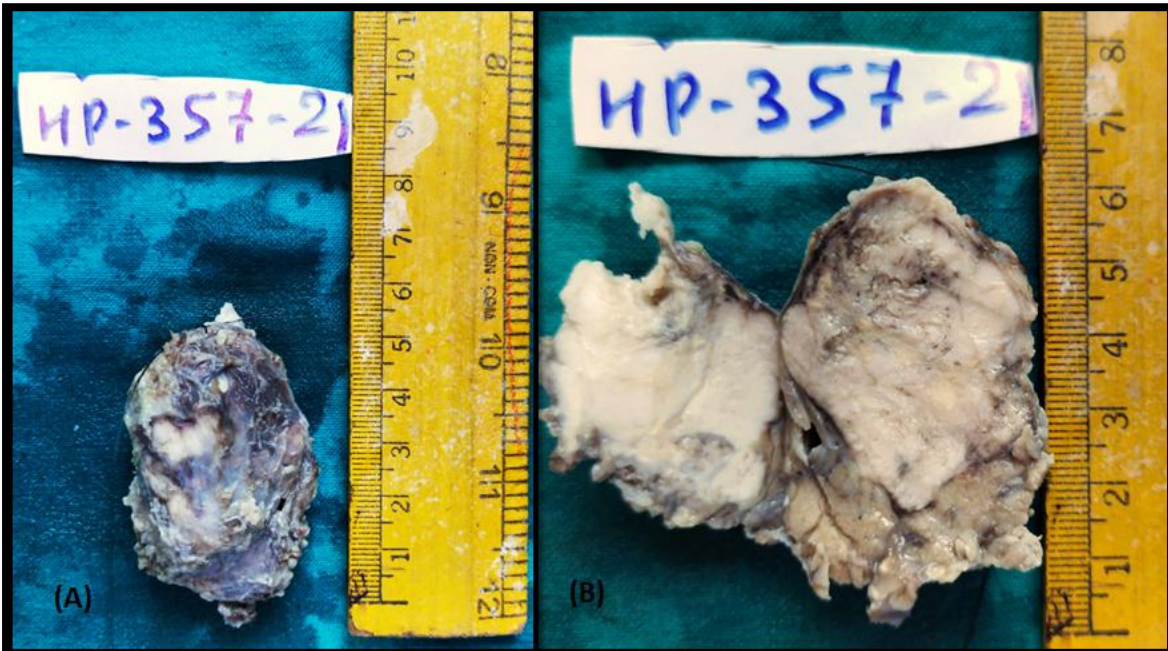


Figure 2: Gross examination of parotid mass (A & B) : A solid greyish white tumor nodule in the parotid gland.

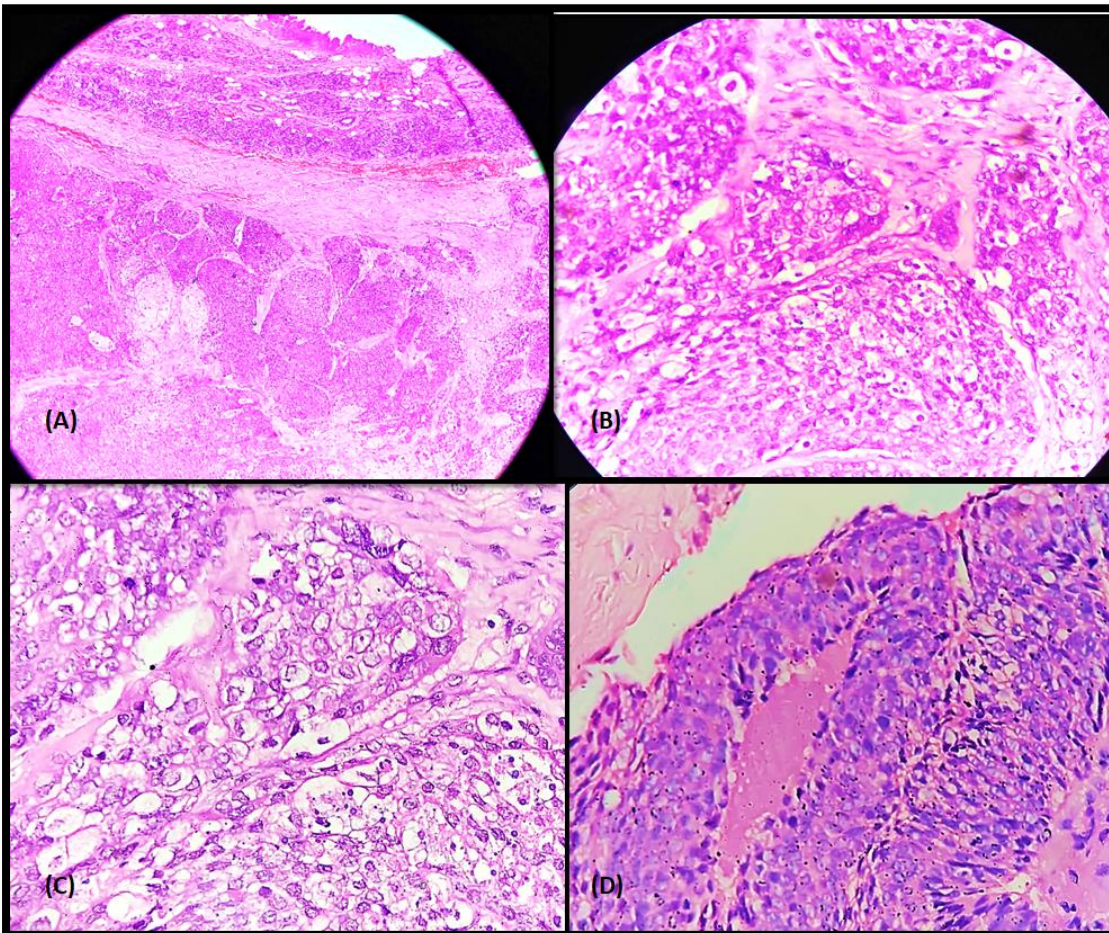


Figure 3: H & E stain: Histopathological examination revealed: (A) 10x: Tumor cells arranged in nodular pattern with normal lying parotid tissue. (B & C) 400x: Malignant cells showing high Nuclear cytoplasmic ratio, cellular and nuclear pleomorphism, hyperchromatic nuclei, mitosis and vacuolated cytoplasm with sebaceous differentiation. (H and E stain -400x) Tumor lobules with comedo necrosis.

It is important to distinguish sebaceous carcinoma from other tumors like mucoepidermoid carcinoma with mucous cells, squamous cell carcinoma with sebaceous differentiation and salivary duct carcinoma. Sebaceous carcinoma shows immunopositivity for AE1/AE3 cytokeratin, EMA, Androgen Receptor protein and is negative for CEA, S100 and Her-2/neu. Adipophilin is a monoclonal antibody with high sensitivity to identify intracellular lipids in normal sebocytes and in neoplastic cells of sebaceous lesion and is a useful marker for diagnosis of sebaceous carcinoma.^[2] Absence of mucin on special stain and staining of tumor cells with oil red O is characteristic of sebaceous carcinoma.^[7]

Management of sebaceous carcinoma metastasis to parotid gland is by superficial parotidectomy as lymph nodes of parotid glands are always confined to the superficial lobe along with neck dissection. Neck dissection is recommended along with parotidectomy even if they do not show metastasis as they are likely to develop metastasis if left behind or they might have occult metastasis at presentation.^{[5],[10]} Similar protocol was followed in our patient.

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