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SEBACEOUS CARCINOMA OCCURRING SIMULTANEOUSLY IN PAROTID AND SUBMANDIBULAR SALIVARY GLAND WITH CERVICAL LYMPHNODE METASTASIS - A CASE REPORT

Pathology

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ABSTRACT

Sebaceous carcinoma is a rare malignancy occurring predominantly in the eyelids and very rarely in salivary glands with 11% to 28% in parotid gland and 6% in submandibular gland. Till date only 33 cases of sebaceous carcinoma in parotid gland and 3 cases of sebaceous carcinoma in submandibular salivary gland have been reported. Upto our knowledge there is no case report of sebaceous carcinoma occurring simultaneously in parotid and submandibular salivary gland in world literature.

Diagnosis may be difficult because of the unusual location and morphological resemblance to other salivary gland neoplasms. IHC is essential for confirming the diagnosis.

KEYWORDS

Sebaceous carcinoma, parotid and submandibular salivary gland.

INTRODUCTION:

Sebaceous Carcinoma is a rare malignancy occurring predominantly in the eyelids and very rarely in salivary glands. Histologically, this tumour is characterized by cells with clear cytoplasm. This tumour has to be differentiated from other malignant conditions having similar morphological features. This case is presented because of its rarity of presentation. Initially, the patient presented with a lymphnode enlargement, which was diagnosed to be sebaceous carcinoma and later further investigations were done to find out the primary lesion.

CASE HISTORY

61 year old male presented with left sided cervical lymphadenopathy for 6 months. There was no significant personal or family history. No history of smoking, alcohol, irradiation or associated with pain, fever or weight loss.

O/E: Enlarged left cervical lymphnodes, 2x2cm, firm in consistency.

Patient was taken up for cervical lymphnode biopsy and same was sent for HPE, which showed a poorly differentiated carcinomatous deposits (Fig 1-3). Tissue was taken up for IHC and diagnosis of sebaceous carcinoma metastasis was made.

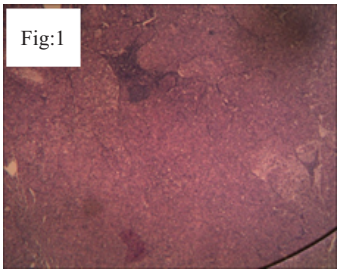

Fig-1: Effaced architecture of lymphnode with metastatic deposits (4x)

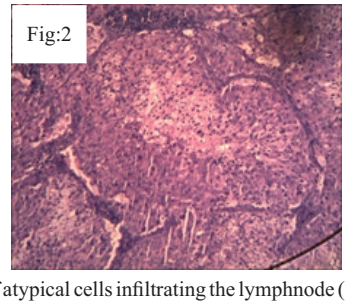

Fig-2: Nests of atypical cells infiltrating the lymphnode (10x)

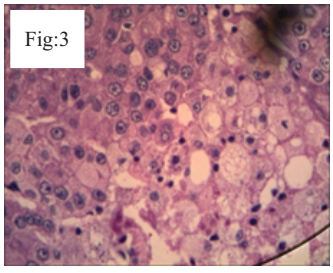

Fig-3: Nests of cells with epitheloid morphology admixed with foamy macrophages (400x)

IHC showed CK 7 & Androgen Receptor Positivity (Fig4&5) and TTF - 1, Synaptophysin, Napsin, CK 20, Calretinin, P 40, CD 68, P63, CK 5/6 negativity.

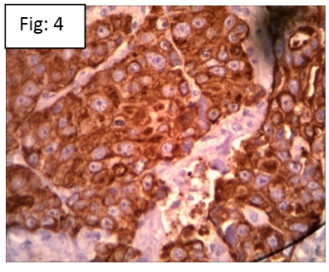

Fig:4- Cells showing positivity for Cytokeratin (400x)

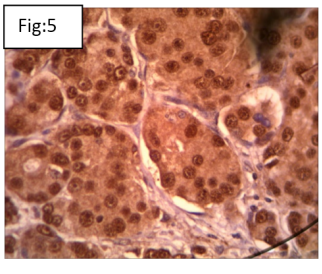

Fig:5- Cells showing positivity for Androgen receptor (400x)

Following histopathological diagnosis of metastasis of sebaceous carcinoma, the patient was worked up to find out the primary lesion. There was no primary in the ocular region or in the genitalia. The patient was planned and posted for radical neck dissection.

Grossly, submandibular gland and parotid gland showed a firm tan greyish lobulated mass with multiple enlarged lymphnodes largest measuring 2x1cm (Fig:6). The surgical margins of submandibular gland was involved by tumour where as the surgical margins of parotid gland was free of tumour. The adjacent lymphnodes did not show any adherence to the gland that ruled out direct extension of the lymphnodal mass to the salivary gland.

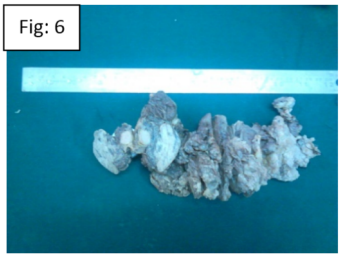

Fig:6: Specimen of Radical neck dissection showing lymphnodes and salivary gland with firm grey white cut surface

HPE of the radical neck dissection showed sebaceous carcinoma involving parotid and submandibular gland (Fig:7&8) with multiple lymphnodes metastasis - 5/30 lymphnodes positive for metastasis (Fig:9) with perinodal infiltration and lymphovascular invasion.

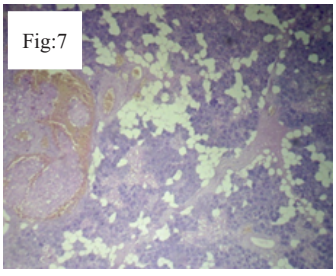

Fig:7- Salivary gland (parotid gland) with parts of a tumour (10x)

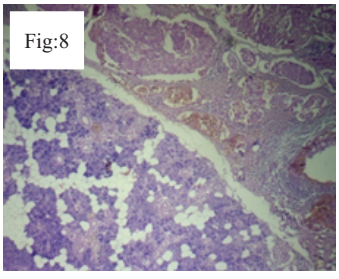

Fig:8- Salivary gland (submandibular gland) with parts of a tumour (10x)

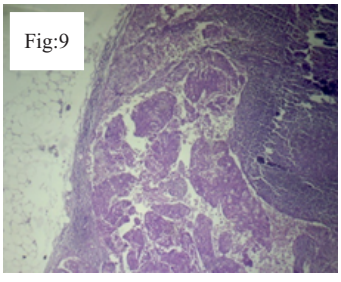

Fig:9- Metastatic deposits in cervical lymphnode (10x)

HPE showed lobulated tumour with pleomorphic cells containing eosinophilic to basophilic cytoplasm admixed with few foamy cells and centrally located nuclei. Areas of increased mitosis were seen. PAS-D was negative in foamy cells. Though differential diagnosis includes Mucoepidermoid Carcinoma, Squamous Cell Carcinoma and Epithelial Myoepithelial Carcinoma, they were excluded as the IHC showed CK 5/6 & P63 negativity and PAS - D negativity.

Though Androgen receptor positivity confirms sebaceous carcinoma in skin, there is no reference regarding AR positivity in salivary glands. The patient was started on radiotherapy.

DISCUSSION:

Sebaceous carcinoma was described in salivary glands by Rauch and Marshoff[1] and is a rare and aggressive neoplasm occurring in head and neck region[2]. 75% occurs in periocular region and is very rare in parotid and submandibular salivary gland[3].

Sebaceous glands are holocrine adnexal components of skin found in close association with hair follicles. Sebaceous carcinoma is defined by WHO as a malignant tumour composed of sebaceous cells of varying maturity that are arranged in sheets and nests with atypia and invasiveness with few foamy cells.

Diagnosis may be difficult because of the unusual location and morphological resemblance to other salivary gland neoplasms. IHC is essential for typing and confirming the diagnosis.

Histogenesis of sebaceous carcinoma in the salivary gland is unclear. Though sebaceous differentiation of salivary duct is seen in both normal and chronic sialadenitis, carcinoma arising from pluripotent stem cells differentiating into sebaceous cells cannot be excluded[3].

Sebaceous carcinoma can be associated with Muir Torre syndrome which is a variant of Lynch Syndrome (Hereditary non polyposis colorectal cancer) HNPCC with germ line mutation of hMLH-1 and hMSH[4]. Our patient was not tested for this mutation.

Sebaceous carcinoma has bimodal age distribution, peak in 2nd decade and another peak in 7th decade of life[5].

Sebaceous carcinoma is treated with complete resection and post operative radiotherapy with chemotherapy for aggressive lesions with metastasis[6].

Metastasis may occur in lung, brain and regional lymphnodes. Our patient had metastasis in the cervical lymphnodes.

Overall survival rate for sebaceous carcinoma in salivary glands is 62% which is lower than that of similar lesion in skin and orbit which is 84.5%[7].

CONCLUSION:

In summary, primary sebaceous carcinoma of salivary gland is extremely rare and primary sebaceous carcinoma involving parotid and submandibular gland has not been reported. Only 33 cases of sebaceous carcinoma in parotid and 3 cases of sebaceous carcinoma in submandibular salivary gland have been reported.

This case is presented for its extreme rarity and upto our knowledge it is first case reported in world literature.

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