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A CASE REPORT AND LITERATURE REVIEW: PRIMARY SEBACEOUS CARCINOMA OF LACRIMAL GLAND TREATED WITH ADJUVANT CHEMO-RADIATION.

Oncology/Radiotherapy

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ABSTRACT

Primary Sebaceous Carcinoma of Lacrimal Gland, possibly arising from heterotopic sebaceous tissue, is extremely rare and must be differentiated from secondary invasion of the Orbit by a primary Eyelid tumor/metastatic spread from other areas of the body. It is known to be aggressive So, delay in diagnosis can increase the chance of local recurrence and metastasis. In this article we report a case of Primary Sebaceous Carcinoma of the lacrimal gland, who underwent surgery with positive margin received Adjuvant radiotherapy in our Institute. **CASE REPORT:** A 55-year-Old Lady presented with Left eyelid swelling. MRI ORBITS & PNS reveals malignant mass of approx. 4.3 X 3.8 cm size in superior & temporal aspect of the left Orbit arising from the left upper eyelid compressing left globe and inseparable from the left lacrimal gland with approx. 2.3X 1.5 cm sized enlarged left Pre- auricular Node. No optic nerve encasement, Orbital wall Destruction & Extension to adjacent paranasal sinuses is seen. CECT Thorax & Whole abdomen – Normal. FNAC from eyelid swelling & Pre- Auricular Node S/o Sebaceous Carcinoma with metastasis. Patient underwent left Orbital Exenteration + left superficial parotid split skin graft. HPE show the tumor mass of size 4.5 X 4X 3 cm, completely replacing tarsal plate of eyelid & tumor mass show a highly infiltrative malignant tumor with morphology consistent with sebaceous carcinoma with positive margin and tissue from left parotid show infiltration by the sebaceous carcinoma (Possibly metastasis to the Intra-parotid node with extra nodal extension). Patient received Adjuvant/post op Chemoradiation 60 Gy/30#/ 6weeks/Rapid Arc technique with weekly 6 cycles chemotherapy Inj. Cisplatin. Patient requires close monitoring for any sign of recurrence / Distant metastasis. **CONCLUSION:** A Rare case report of sebaceous carcinoma of the Lacrimal Gland which requires appropriate evaluation & management for better prognosis.

KEYWORDS

INTRODUCTION:

Tumors of the Lacrimal gland are rare and present in less than 1/100000 individual per year[1]. Among tumors of the lacrimal gland, sebaceous carcinoma of the lacrimal gland is extremely rare. In this article, we report a case of Primary sebaceous carcinoma of lacrimal gland who underwent surgery with positive margin receive Adjuvant radiation in our institute.

CASE REPORT:

A 55-year-old lady presented with left eyelid swelling for 1 month. She was evaluated with MRI Orbit & PNS s/o lobulated mass of approx. 4.3 x 3.8 cm size noted in the superior and temporal aspect of left Orbit arising from left upper eyelid. Mass is Isointense to Hypointense on T2W images shows Intermediate signal on T1W images along with dense heterogenous enhancement, Diffusion restriction & low ADL. It is compressing the left Globe with flattening of the Globe wall. It is inseparable from left lacrimal gland & Anterior aspect of left LR muscle. There is approx. 2cm Retrobulbar extension into lateral extra-Conal space. No optic nerve encasement/ extension to orbital apex / SOF is seen. It is abutting the orbital roof & Lateral Orbital wall, however no bone erosion, Intrasinus/ Intra cranial Extension is noted. Left Optic nerve and rest of Extra-ocular muscles are normal. There is enlarged node of approx. 2.3 x 1.5 cm sized in the left pre- Auricular region noted underneath skin and subcutaneous tissues. The enlarged node is closely abutting the superficial lobe of left parotid gland with loss of fat planes. FNAC from left eyelid and parotid swelling s/o sebaceous carcinoma. CT chest & abdomen– Normal. She was diagnosed as sebaceous carcinoma of lacrimal Gland.

Image-1: Clinical Image prior to surgery

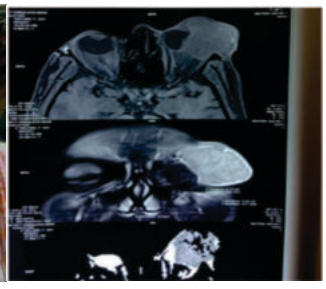
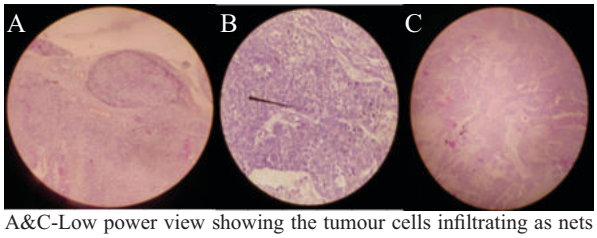


Image-2: Axial and coronal MRI scans: Before Surgery

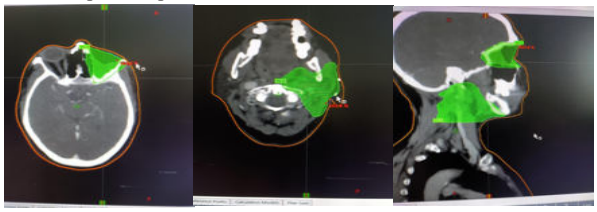
Surgery Patient underwent left Orbital Exenteration + left superficial parotidectomy + split skin graft. HPE s/o tumor mass shows a highly Infiltrative malignant tumor with morphology, Consistent with sebaceous carcinoma. The tumor is seen infiltrating the adjacent fibromuscular and fatty tissue. It does not infiltrate the eyeball. Tumor infiltrates the superior margin, inferior margin, posterior margin and lateA&C-Low power view showing the tumour cells infiltrating as nets and sheets surrounded by fibrous storms.

B-High power microscopic view of resected specimen showing sheets of multi vacuolated cells with mild pleomorphism and mitosis.

Axial and sagittal sections shows Dose colour wash:- 95% PTV coverage.ral margins, LVSI (+)/Perineural invasion not seen. Tissue from left parotid show infiltration by the sebaceous carcinoma (Possibly metastasis to the Intraparotid node with Extra nodal extension). Patient received Adjuvant/post-op radiation 60Gy/30#/6 weeks / Rapid arc technique with weekly 6 cycles of chemotherapy Inj.Cisplatin. She needs to be followed up closely to see any sign of recurrence.



A&C-Low power view showing the tumour cells infiltrating as nets and sheets surrounded by fibrous storms.B-High power microscopic view of resected specimen showing sheets of multi vacuolated cells with mild pleomorphism and mitosis.



Axial and sagittal sections shows Dose colour wash:- 95% PTV coverage.

DISCUSSION:

Tumors of lacrimal gland are relatively rare. It accounts for approx. 22-23% of all orbital space occupying lesions and 20-50 % of these tumors are malignant[7-9].

Primary sebaceous carcinoma of lacrimal gland is extremely rare, and possibly arising from heterotropic sebaceous tissue. It must be differentiated from secondary invasions of the orbit by a primary eyelid tumor / metastatic spread from other areas of the body. Tumor is highly malignant and can metastasize to preauricular and deep cervical lymph nodes occur early in the disease. There are various possible processes by which sebaceous carcinoma might be present in the lacrimal gland. Posterior extension of a Meibomian/Zeis gland sebaceous carcinoma into the orbit, mimicking a lacrimal tumor, has been described by Shields and Front[12] . In their patient, however, while the upper eyelid was extremely normal, on lifting and everting of the lid, the mass was clearly visible in the upper outer quadrant. Additionally, the conjunctives showed chemosis and injection there was early corneal pannus and the anterior segment was involved with cells and flare.

Epithelial tumors are most common lesions of the lacrimal gland[8]. Epithelial lesions of the lacrimal gland include both benign and malignant tumors[10]. Most common benign tumor of the lacrimal gland is the pleomorphic adenoma, while adenoid cystic carcinoma is the most common malignant tumor[7,9,20]. Therefore, second possibility is malignant transformation and sebaceous differentiation with a pleomorphic adenoma/ other lacrimal neoplasm of epithelial origin. Mesenchymal elements in benign mixed tumors of the lacrimal gland are thought to represent ectodermal products that have undergone metaplasia[13], and epithelial (or) Myoepithelial cells can differentiate in various directions.

Primary sebaceous carcinoma of lacrimal gland has so far been treated almost exclusively by surgery. In contrast, radiotherapy is practically the only treatment for primary sebaceous carcinoma of eyelid [14-17.] The problem, however, is that sebaceous carcinoma has high incidence of metastasis/ local recurrence after surgical excision is approximately 9-36%, post-op radiotherapy is usually recommended[6]. For patients with difficulties in achieving a tumour free margin or a cosmetically acceptable reconstruction, or when the patient refuses further surgical excision in the absence of orbital extension or distant metastasis, surgical excision with adjuvant radiation or radiotherapy alone could be an alternative treatment[6]. Another issue in radiotherapy of sebaceous carcinoma of lacrimal gland is target volume. As tumor can metastasize to pre auricular and deep cervical lymph nodes early in the course of disease, parotidectomy and cervical lymphadenectomy could be considered. In our study post surgery we irradiated both surgical tumor bed including neck node. Until a consensus is reached, radiation oncologist should modify levels of neck irradiation according to physical examination, images and post-op neck dissection site.

CONCLUSION:

We report a rare case of sebaceous carcinoma of the lacrimal gland post surgery who received Adjuvant/post-op chemo-radiation. Although recent studies have reported a favourable prognosis, sebaceous carcinoma of lacrimal gland has high morbidity and mortality rate; therefore appropriate treatment, accurate and early diagnosis is important. Due to its rarity, optimal treatment is not established. Although surgery is the main treatment, radiation can be delivered post-operatively or as the only modality of treatment.

REFERENCES:

1. von Holstein SL, Coupland SE, Briscoe D, Le Tourneau C, Heegaard S. Epithelial tumours of the lacrimal gland: a clinical, histopathological, surgical and oncological survey. Acta Ophthalmol 2013; 91: 195-206 [PMID: 22471335 DOI: 10.1111/ j.1755-3768.2012.02402.x]
2. Kiratli H, Tarlan B, Fırat P. Primary sebaceous carcinoma of the lacrimal gland. Orbit 2012; 31: 352-354 [PMID: 22906082 DOI: 10.3109/01676830.2012.678921]
3. Yamamoto N, Mizoe JE, Hasegawa A, Ohshima K, Tsujii H. Primary sebaceous carcinoma of the lacrimal gland treated by carbon ion radiotherapy. Int J Clin Oncol 2003; 8: 386-390 [PMID: 14663642 DOI: 10.1007/s10147-003-0348-2]
4. Harvey PA, Parsons MA, Rennie IG. Primary sebaceous carcinoma of lacrimal gland: a previously unreported primary neoplasm. Eye (Lond) 1994; 8 (Pt 5): 592-595 [PMID: 7835459 DOI: 10.1038/ eye.1994.143]
5. Briscoe D, Mahmood S, Bonshek R, Jackson A, Leatherbarrow B. Primary sebaceous carcinoma of the lacrimal gland. Br J Ophthalmol 2001; 85: 625-626 [PMID: 11351970 DOI: 10.1136/ bjo.85.5.625]
6. Yoon JS, Kim SH, Lee CS, Lew H, Lee SY. Clinicopathological analysis of periocular sebaceous gland carcinoma. Ophthalmologica 2007; 221: 331-339 [PMID: 17728556 DOI: 10.1159/000104764]
7. Snaathorst J, Sewnaik A, Paridaens D, de Krijger RR, van der Meij EH. Primary epithelial tumors of the lacrimal gland; a retrospective analysis of 22 patients. Int J Oral Maxillofac Surg 2009; 38: 751-757 [PMID: 19369032 DOI: 10.1016 /j.ijom. 2009.02.022]
8. von Holstein SL, Rasmussen PK, Heegaard S. Tumors of the lacrimal gland. Semin Diagn Pathol 2016; 33: 156-163 [PMID: 26849904 DOI: 10.1053 /j.semdp. 2015.10.002]
9. Weis E, Rootman J, Joly TJ, Berean KW, Al-Katan HM, Pasternak S, Bonavolontà G, Strianese D, Saeed P, Feldman KA, Vangveeravong S, Lapointe JS, White VA. Epithelial lacrimal gland tumors: pathologic classification and current understanding. Arch Ophthalmol 2009; 127: 1016-1028 [PMID: 19667339 DOI: 10.1001 /archophthalmol. 2009.209]
10. Chawla B, Kashyap S, Sen S, Bajaj MS, Pushker N, Gupta K, Chandra M, Ghose S. Clinicopathologic review of epithelial tumors of the lacrimal gland. Ophthalmic Plast Reconstr Surg 2013; 29: 440-445 [PMID: 24145905 DOI: 10.1097/ IOP. 0b013e31829f3a0c]
11. Dasgupta T, Wilson LD, Yu JB. A retrospective review of 1349 cases of sebaceous carcinoma. Cancer 2009; 115: 158-165 [PMID:18988294 DOI: 10.1002/cncr.23952]
12. Shields J, Font RL (1974) Meibomian gland carcinoma presenting as a lacrimal tumor. Arch Ophthalmol 92:304–306
13. Font RL, Gamel JW (1978) Epithelial tumors of the lacrimal gland: an analysis of 265 cases. In:Jakobiec FA (ed) Ocular and adnexal tumors. Aesculapis, Birmingham, pp 787–805
14. Pardo FS, Wang CC, Albert D, et al. (1989) Sebaceous carcinoma of the ocular adnexa: radiotherapeutic management. Int J Radiat Oncol Biol Phys 17:643–647
15. Matsumoto CS, Nakatuka K, Matsuo K, et al. (1995) Sebaceous carcinoma responds to radiation therapy. Ophthalmologica 209:280–283
16. Yen MT, Tse DT, Wu X, et al. (2000) Radiation therapy for local control of eyelid sebaceous cell carcinoma: report of two cases and review of the literature. Ophthal Plast Reconstr Surg 16:211– 215
17. Nunery WR, Welsh MG, McCord CD Jr (1983) Recurrence of sebaceous carcinoma of the eyelid after radiation therapy. Am J Ophthalmol 96:10–15
18. von Holstein SL, Therkildsen MH, Prause JU, Stenman G, Siersma VD, Heegaard S. Lacrimal gland lesions in Denmark between 1974 and 2007. Acta Ophthalmol 2013; 91: 349-354 [PMID: 22471375 DOI: 10.1111/j.1755-3768.2012.02403.x]
19. Tsujii H, Kamada T. A review of update clinical results of carbon ion radiotherapy. Jpn J Clin Oncol 2012; 42: 670-685 [PMID: 22798685 DOI: 10.1093/jjco/hys104].
20. Andreasen S, Esmaeli B, Holstein SL, Mikkelsen LH, Rasmussen PK, Heegaard S. An Update on Tumors of the Lacrimal Gland. Asia Pac J Ophthalmol (Phila) 2017; 6: 159-172 [PMID: 28399336 DOI: 10.22608/APO.201707].