Innovative Journal of Medical and Health Science
Received 15 Dec 2020 | Revised 30 Jan 2021 | Accepted 15 Feb 2021 | Published Online 22 Mar 2021

DOI: https://doi.org/10.15520/ijmhs.v11i03.3262
I Jour Med Health Science 11 (03), 1622–1626 (2021)
ISSN (O) 2589-9341 | (P) 2277-4939 IF:1.6

CASE REPORT

SEBACEOUS CARCINOMA OF GLUTEAL REGION – AN UNUSUAL TUMOUR IN A RARE EXTRAOCULAR SITE

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Abstract

Sebaceous carcinoma is a rare aggressive malignant tumor derived from the adnexal epithelium of sebaceous glands. Sebaceous carcinomas are generally divided as ocular or extraocular locations. Very few cases of extra ocular sebaceous carcinomas have been reported till date. Hereby, we report a 62-year-old female diagnosed with sebaceous carcinoma on the gluteal region with recurrence, and was treated surgically with wide local excision. The extra-ocular site is rare and high degree of suspicion is required and sebaceous carcinoma should be considered as one of the differential diagnosis.

Keywords: SEBACEOUS CARCINOMA, GLUTEAL REGION, EX-TRAOCULAR SITE

1 | INTRODUCTION

First described by Allaire in 1891, sebaceous gland carcinoma (SGC) is a highly malignant and potentially lethal tumor of sebaceous glands.[1,2] Sebaceous carcinoma is a rare adnexal tumor that follows an aggressive clinical course, with a risk of local recurrence and distant metastasis.[3] Three-quarters of cases occur in the periocular region, particularly on the eyelids. The most common extraocular site described in the literature is the head and neck (20%).[4] Reported risk factors for sebaceous carcinoma include advanced age, Asian or South Asian race, women, previous irradiation to the head and neck, and a genetic predisposition for Muir-Torre syndrome, or possibly familial Retinoblastoma.[5–11] The very rare sites of sebaceous carcinoma so far reported are chest wall, breast nipple, parotid gland, thigh.[12–17.] To our knowledge, this is the first case report of sebaceous carcinoma occurring in the gluteal region.

2 | CASE REPORT

A 62-year-old female patient, known diabetic presented to our out patient department with complaints of thickened scar in the right gluteal region. She had history of nodular lesion in the gluteal region for 2 months duration for which clinical diagnosis of sebaceous cyst was made and excision was done 4 months back. The post operative histopathology was suggestive of sebaceous carcinoma and the patient defaulted for further treatment. She came to the surgical oncology clinic for further management with complaints of progressively increasing in size of the swelling for the last three months duration. Clinical examination revealed a painless, single well defined nodular lesion of size 2.5 x 2.5 cm with surrounding hyperpigmentation in the right gluteal region, 7 cm lateral to the gluteal cleft.(Fig.1)A linear scar with surrounding induration extending to 0.5 cm around the nodule was present. There was no significant inguinal node enlargement.

Magnetic resonance imaging revealed 4 x 3.3 cm well defined T2 hypointense and T1 iso-intense lesion with superficial eccentric areas of T2, hyperintense fluid signal in the skin and the superficial subcutaneous plane of supero-lateral aspect of right gluteal region.(Fig.2) Biopsy was done which was suggestive of sebaceous carcinoma. Ultra-sonographic examination of the inguinal region was not suggestive of any enlarged metastatic nodes. Computed tomographic study of the chest and ultrasonographic examination of the liver were negative for lung and liver metastases.

In view of the non metastatic nature of the disease, patient underwent wide local excision with 2 cm margin clearance around the induration and deep fascia was taken as deeper margin and closure was done primarily. Post operative period was uneventful. The post operative histopathology showed well circumcised tumour composed of lobules of undifferentiated cells with scattered sebaceous cells with vacuolated foamy cytoplasm. The tumour showed large areas of geographic necrosis with focal squamous differentiation and focal infiltration into the capsule. The surface of the tumour showed ulceration with suppuration.(Fig.3) There was no pagetoid spread. Immunohistochemistry showed positivity for androgen receptor, p53, focal positivity for EMA with high Ki67 and was negative for CEA and S100 confirming the diagnosis of sebaceous carcinoma.(Fig.4) The patient is disease free and on regular follow up with clinical examination for the past five months.

Supplementary information The online version of this article (https://doi.org/10.15520/ijmhs.v11i03.3262) contains supplementary material, which is available to authorized users.

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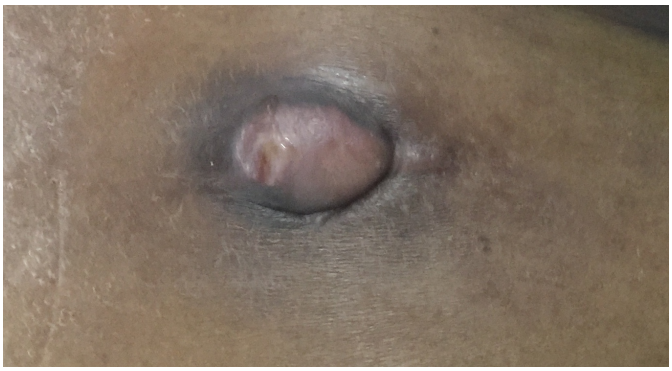

FIGURE 1: PHOTOGRAPH OF THE LESION AT THE GLUTEAL REGION WITH NODULARITY AND PREVIOUS SURGICAL SCAR CHANGES 480x261mm (72 x 72 DPI)

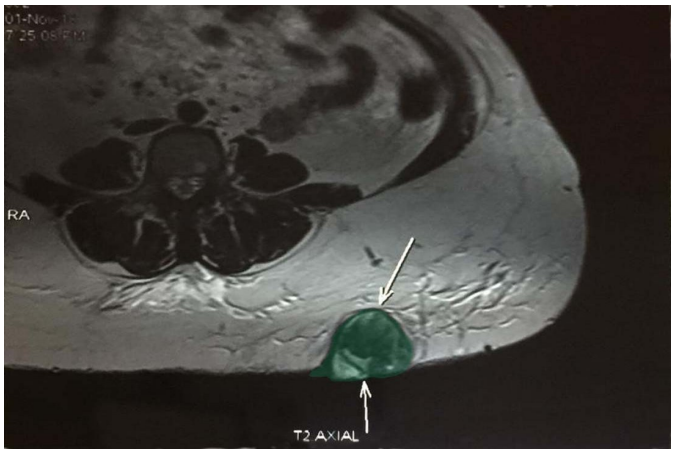

FIGURE 2: MRI OF THE GLUTEAL REGION – T2 AXIAL- SHOWING THE LESION IN THE SUBCUTANEOUS REGION. 385x278mm (72 x 72 DPI)

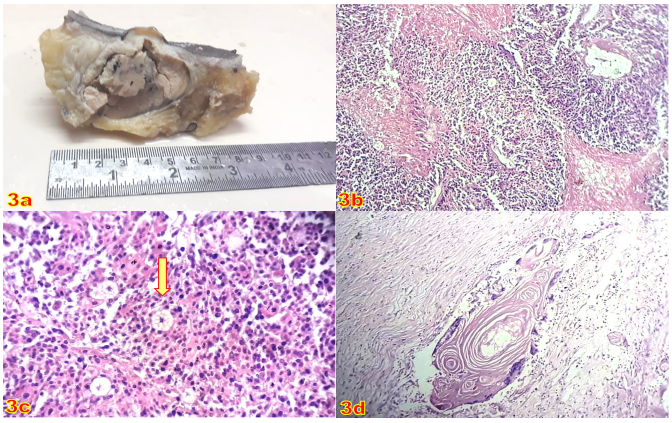

FIGURE 3: a) Gross picture showing tumor mass , b) Histopathological examination showing lobules of undifferentiated basaloid cells with areas of necrosis (H&E, 100X), c) Shows scattered sebaceous cells (arrow pointing) with foamy cytoplasm (H&E, 400X), d) shows area of squamous differentiation infiltrating the fibrocollagenous capsule (H&E, 400X). 342x228mm (300 x 300 DPI)

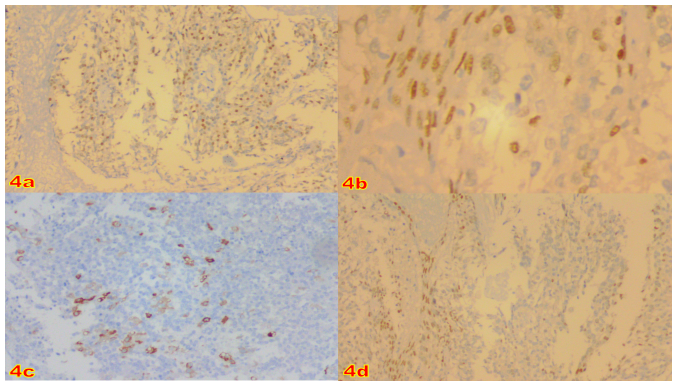

FIGURE 4: a) Immunohistochemistry showing positivity for Androgen receptor (IHC, 100X), b) p53 positivity (IHC, 400X), c) EMA showing focal positivity (IHC, 100X), d) High Ki67 (IHC, 100X) 342x228mm (300 x 300 DPI)

3 | DISCUSSION

According to the Surveillance, Epidemiology, and End Results (SEER) study from 2002 to 2012, Sebaceous carcinoma can be classified into two groups: ocular type and extraocular type.[18]Sebaceous carcinoma represents less than 0,1% of all cutaneous malignancies. The tumor typically arises in the sixth–seventh decade of life (57–72 years) but younger age incidence also have been described in the literature.[19] The etiology of the sebaceous carcinoma is not known. One hypothesis is that the tumor originates in the epithelium of sebaceous glands, but recent studies suggest that it may be derived from a pleuripotent cell able to differentiate into any cell line, including sebaceous cells.[20]In sporadic cases, Human Papilloma Virus infection, dysregulated cytokine secretion and mutations in tumour suppressor genes such as p53 might contribute to sebaceous carcinoma development. [21]

Among all the associations described in the literature, the most common associations are with the Muir- Torre syndrome , immunosuppression, lesions associated with nevus sebaceous of Jadassohn.[21] About the association with Muir- Torre Syndrome, it was suggested that expression of retinoid X receptor beta and gamma could be related to the development of SC. Muir- Torre syndrome is a phenotypic variant of hereditary non-polyposis colorectal cancer (HNPCC) or Lynch syndrome.[22]

Histopathology of Sebaceous carcionoma shows irregular lobular formations with great variations in

the size of the lobules with many undifferentiated cells; distinct sebaceous cells showing a foamy cytoplasm are present in the center of most lobules and the number of these cells may vary depending on the differentiation. It is hard to find these cells in poorly differentiated tumors and needs special stains like oil red O to highlight these cells. Sebaceous carcinomas are usually positive for EMA, Androgen receptor, p53, adipophillin, and negative for CEA and S100. Ki67 shows high proliferation index.[23]

Extraocular sebaceous carcinoma is clinically and histopathologically different from ocular sebaceous carcinoma. Reported clinicopathological differences include more frequent ulceration in extraocular tumors and more frequent pagetoid spread of tumor cells in the epidermis in cases of ocular sebaceous carcinoma.[24]Extraocular sebaceous carcinoma has been traditionally claimed to have a better prognosis than the ocular forms. However, review of the recent literature has revealed that extraocular sebaceous carcinoma has a tendency to be locally aggressive, to develop cutaneous recurrence and to metastasize widely. The most common site of metastasis is in the draining lymph node. Distant metastases have been reported in less than 10% of all ESC cases.[25,26.]The clinical differential diagnosis of extraocular sebaceous carcinoma includes various neoplastic conditions such as basal cell carcinoma,squamous cell carcinoma, amelanotic melanoma, Merkel cell carcinoma, and cutaneous lymphoma, or non-neoplastic benign sebaceous conditions such as nevus sebaceus, xanthoma, and sarcoidosis.[27]

Factors associated with poor prognosis are vascular, lymphatic, orbital metastases, poor differentiation, multicentric origin, duration of symptoms more than six months, tumor diameter > 10mm and a highly infiltrative pattern.[28] Calculation of extraocular sebaceous carcinoma survival rate is difficult because of the relatively low number of cases with sufficient follow-up. The primary modality of the treatment of non metastatic sebaceous carcinoma is excision with negative margins, whereas Moh's microsurgery can play an important role in cosmetic areas. Radiation therapy in the management of extraocular sebaceous carcinoma is considered largely palliative and for post surgical management of metastatic disease.[29]

4 | CONCLUSION

Sebaceous neoplasms, especially in extraocular regions, often mimic more benign cystic lesions leading to misdiagnosis. Because of the rarity in the region other than head and neck, extra ocular sebaceous carcinoma continues to defy clinicians and the pathologists in suspicion and diagnosis confirmation and treatment challenges. High degree of suspicion is required and sebaceous carcinoma should be considered as one of the differential diagnosis.

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How to cite this article: R.A.V.I.S.A.N.K.A.R.P.A.L.A.N. GIRIEASAN S.A.B.A.R.I., KALAIVANI S.U.B.R.A.M.A.N.I.A.N., HARIPRIYA R.A.G.A.V.A.N., DURAIRAJU P.R.I.Y.A.D.H.A.R.S.H.I.N.I., KOTHANDAPANI S.E.L.V.A.K.U.M.A.R. SEBACEOUS CARCINOMA OF GLUTEAL REGION – AN UNUSUAL TUMOUR IN A RARE EXTRAOCULAR SITE. Innovative Journal of Medical and Health Science. 2021;1622–1626. https://doi.org/10.15520/ijmhs.v11i03.3262