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Case Report

Hidradenocarcinoma of scalp: a case report and review of literature

Sowjanya Gandla*, Satakshi Chatterjee, Ashuthosh Patil, Vishal Rao

Department of Head and Neck Surgical Oncology, HCG, Bangalore, India

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***Correspondence:**
Dr. Sowjanya Gandla,
E-mail: sowjanya233@gmail.com

ABSTRACT

Hidradenocarcinoma of the scalp is a rare skin adnexal tumour accounting for less than 0.001% of all tumors. In this study, we describe a rare presentation of the hidradenocarcinoma of the scalp in a 64 years old man. Patient presented with a right side scalp lesion in the occipital area, for which biopsy was done which showed hidradenocarcinoma. Patient underwent wide local excision of the posterior scalp lesion and posterolateral neck dissection and the patient was advised to receive adjuvant chemotherapy and radiotherapy, but the patient refused to undergo adjuvant therapy. Six months postsurgery patient had local recurrence in the posterior margin resection area and lung metastasis, which could have been prevented by adjuvant therapy. Malignant hidradenocarcinoma is a rare aggressive malignant tumour. Patients with this tumour should undergo surgical excision followed by adjuvant therapy..

Keywords: Hidradenocarcinoma, Adjuvant therapy

INTRODUCTION

Hidradenocarcinoma is a type of skin adnexal tumour arising from skin appendages.[1] Skin appendages are subdivided into three categories Pilosebaceous unit, apocrine cutaneous sweat gland and eccrine cutaneous sweat gland.[2] Hidradenocarcinoma of the scalp is a rare malignant tumour of eccrine cutaneous sweat glands.[2] Carcinomas of the eccrine sweat gland represents a rare group of tumours with potential for local destruction and metastasis.[3] Other malignant tumours of eccrine sweat glands are porocarcinoma, spiradenocarcinoma, malignant cylindroma, adenoid cystic carcinoma and microcystic adenocarcinoma.[3] Hidradenocarcinoma accounts for less than 0.001% of all tumours.[1] Head and neck is the most common site for hidradenocarcinoma.[2] Here we present a rare case of metastatic hidradenocarcinoma of the scalp and the review of literature of management of hidradenocarcinoma.

CASE REPORT

A 64 years old man presented with a swelling in the occipital area of right side scalp of 5 years duration. The swelling was insidious in onset, gradually progressive, but there was sudden increase in size of swelling for 3 months before seeking for consultation. There was no history of trauma to the area, tobacco chewing and smoking. He had consulted general practitioner for the above complaints. Punch biopsy was performed from the scalp lesion and also from enlarged right level V lymph node, which were reported as adenocarcinoma arising from nodular hidradenoma and metastatic adeno-carcinoma respectively. The patient was referred to our center for further management.

The physical examination revealed 5 cms×4 cms painless swelling in the occipital area of right side scalp region. Enlarged lymph nodes were present in the right side posterior triangle as shown in Figure 1.

Figure 1: Preoperative image of the lesion.

USG neck showed, enlarged right posterior cervical lymph nodes with distorted architecture, largest measuring 1.1×0.7cms. PET CT scan was done, which showed 3.2×2.4 cm metabolically active soft tissue cutaneous/subcutaneous thickening in the right lower occipital region, extending to muscular plane (SUV: 7.6) and multiple metabolically active right occipital and level V cervical lymph nodes (SUV: 15.8) as shown in Figure 2.

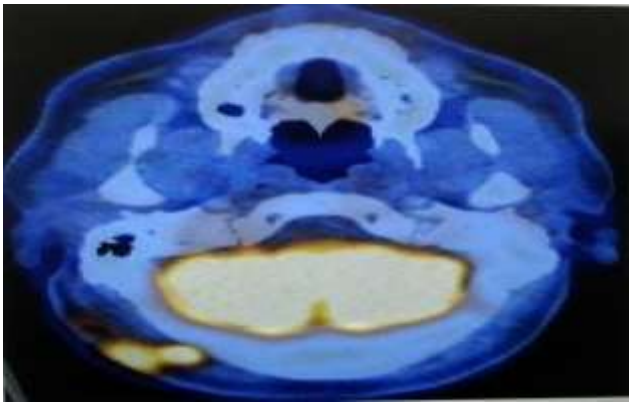

Figure 2: Preoperative PET CT image.

Patient underwent wide local excision of the posterior occipital region of the scalp with right posterolateral neck dissection as shown in Figure 3, 4 and 5. Histopathological examination showed scalp hidradeno-carcinoma with multiple foci of perineural invasion. All the margins were shown to be free of tumor on frozen section. Multiple Lymph nodes were Positive with extracapsular extension. The Postoperative period was uneventful. Patient was advised to receive adjuvant chemotherapy with radiotherapy, but the patient refused to undergo adjuvant therapy and wished to opt for alternative medicine treatment. Six months post-surgery, patient presented with local recurrence in the area of posterior margin resection, which could have been prevented by receiving adjuvant therapy. PET CT scan performed to reassess showed local recurrence at the region of scalp margins, lung metastasis and iliac bone metastasis. The case was discussed in our multidisciplinary clinic and the patient was advised to receive chemoradiotherapy. However patient opted for alternative medicine treatment.

Figure 3: Intraoperative image.

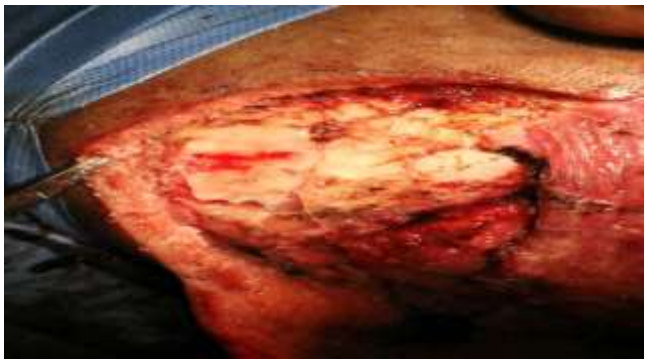

Figure 4: Intraoperative image.

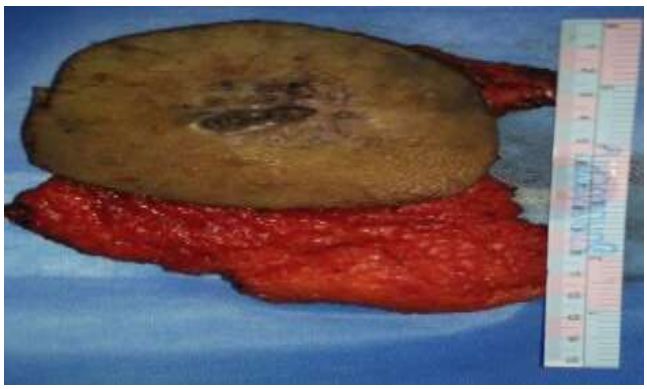

Figure 5: Excised specimen.

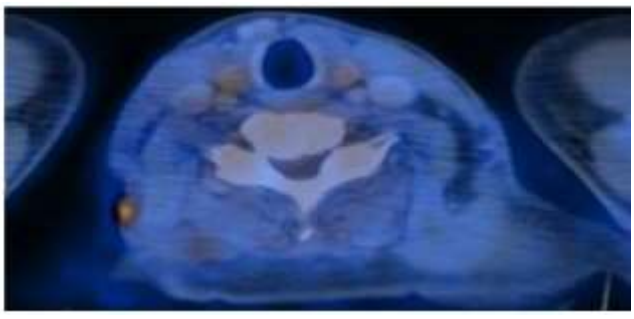

Figure 6: PET CT showing local recurrence.

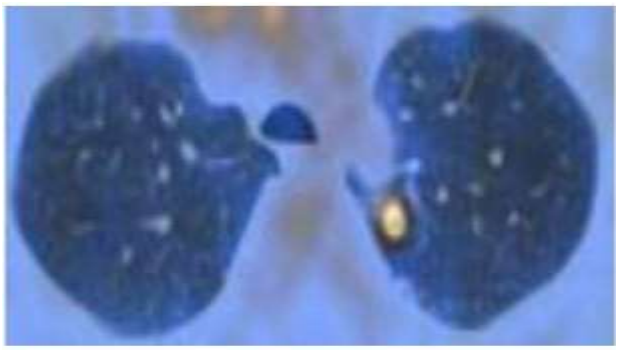

Figure 7: PET CT showing lung metastasis.

DISCUSSION

Computed tomography, magnetic resonance imaging and PET are the imaging modalities for skin adnexal tumors. In MRI, the lesion shows hypointense signal on T1 weighted images and hyperintense signal on T2 weighted images.[2] Hidradenocarcinoma was first described by keasbay and Hadley in 1954.[4] Hidradenocarcinoma is a rare aggressive malignant tumour of the eccrine cutaneous sweat glands.[2] Hidradenocarcinoma accounts for approximately 6% of malignant eccrine tumours.[2] The most common age group of presentation is fifth to seventh decade with the mean age of onset being 50 years.[5] Hidradenocarcinoma presents with a slight female preponderance.[3]

The clinical history of hidradenocarcinoma patients with which they present to the hospital is very non-specific. Most of the patients with this tumour are asymptomatic. Usually the patients present with prolonged history of non-tender cutaneous or subcutaneous nodular swelling commonly in the head and neck region but can also occur rarely in extremities.[6] Usually the skin over the swelling is normal in most of the cases, but in patients with prolonged history and aggressive tumors, the skin may be ulcerated. According to the literature, there is a delay in diagnosis in most of the cases of hidradenocarcinoma.[7] The factors attributing to the delayed diagnosis could be slow growing asymptomatic nodule and most of these nodules can be mistaken clinically with more common lesions like lipoma, epidermoid cyst and sebaceous cyst. The differential diagnosis include lipoma, metastatic tumors to the skin, nodular basal cell carcinoma, hemangioma, lymphangioma, nodular squamous cell carcinoma and epidermoid cyst. The tumour usually arises conventionally de novo or rarely hidradeno-carcinoma may arise from pre-existing hidradenoma.[8] If the diagnosis of hidradenocarcinoma is made, then it is advisable to undergo a whole body scan to exclude distant metastasis, as the distant metastasis is more common in these tumors.

The diagnosis is based on histological and immune-histochemical analysis. On gross examination, the tumour appears like well circumscribed nodule over the skin. On histopathological examination, the tumour cells exhibit characteristic vacuolated cytoplasm (due to the presence of glycogen), another characteristic feature of hidradenocarcinoma is the presence of ductal lumina lined by epithelial cells.[9] The tumour contains two different cell types, clear cells and fusiform cells. The histology of benign hidradenoma and malignant hidradenocarcinoma is similar but malignant hidradeno-carcinoma exhibits infiltrative or invasive pattern, atypical mitosis, lymphovascular invasion and perineural invasion.[3] On immunohistochemistry, hidradeno-carcinomas are variably positive for epidermal growth factor receptor (EGFR) in 80-85% of cases and are also positive for androgen receptor, oestrogen receptor, progesterone receptor.[10] Shu et al did immune-histochemistry for both of their hidradenocarcinoma patients and found that both cases were diffusely positive for EMA (epithelial membrane antigen) and Vimentin.[11]

Surgery is the mainstay of treatment modality and postsurgical 5 year survival rate is found be less than 30%.[2] Wide local excision of the tumour with adequate margins along with neck dissection is the treatment modality of choice. Even with aggressive surgical resection the rate of local recurrence ranges from 20%-50%.[3] Regional lymph nodal spread is the commonest route of metastasis. Hematogenous spread to bones, vertebrae, lungs have also been reported.[3]

A case report of hidradenocarcinoma by ohta et al and Park et al, have concluded that early wide local excision of the tumour is the treatment of choice.[7,12] In their case reports, patients were only treated with wide local excision and there was no recurrence observed in 24 months and 16 months of follow up respectively. In a study by Stanislav et al, the authors performed a retrospective review of hidradenocarcinoma patients treated with Mohs micrographic surgery at Mayo clinic from 1993 to 2013 and they found that Mohs micrographic surgery seems to be a useful treatment modality and there was no recurrence, metastasis and disease related mortality in patients treated with Mohs micrographic surgery.[13]

In contrary a case report by Lalya et al, they have concluded that adjuvant therapy should be considered postoperatively for improved loco-regional control and should be mandatory in certain histopathological features like dermal lymphatic invasion, perineural invasion, high anaplastic morphology, extracapsular lymph nodal extension, infiltration of deep structures and positive surgical margins.[8] Their patient was initially treated with only surgery and had recurrence within 5 months of surgery and this recurrence was managed successfully by high dose radiotherapy. Harari and colleagues reported complete remissions with external beam radiotherapy of sweat gland tumours with positive margins after surgery.[14] Lerner et al have mentioned that their patient with unresectable metastatic hidradenocarcinoma had complete clinical and radiological response with 3 months of oral capecitabine.[15]

Six months post-surgery, our patient had local recurrence in the posterior margin resection area, despite complete surgical excision with adequate clear margins. This local recurrence in this case could have been prevented by adjuvant chemotherapy and radiotherapy. This indicates that malignant hidradenocarcinoma is a very aggressive tumour and patients with this tumour should undergo complete surgical excision with clear margins followed by adjuvant therapy.

CONCLUSION

Hidradenocarcinoma is a rare aggressive tumor with high rates of local recurrence and distant metastasis when treated with surgical modality alone. It is desirable that these patients be treated aggressively with surgical excision followed by adjuvant therapy.

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