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RESEARCH ARTICLE

Trichilemmal carcinoma: A rare cutaneous adnexal tumour

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Abstract

Trichilemmal carcinoma is a rare neoplasm arising from outer root sheath of hair follicle on sun exposed areas, the diagnosis of which is predominantly confirmed on histological features as it resembles other malignant skin tumours. Immunohistochemistry helps in identifying the cell of origin. As the tumor is of low malignant potential and has indolent clinical course, the treatment is exclusively surgical. We report a case of 62-years-old healthy woman who presented with a painless nodule on scalp growing rapidly for the past 4 months. Wide surgical excision was conducted and the patient was followed up for one year with no signs of recurrence. Therefore, we emphasize that early detection and initiation of treatment with careful follow up is the only requirement needed to manage this tumour.

INTRODUCTION

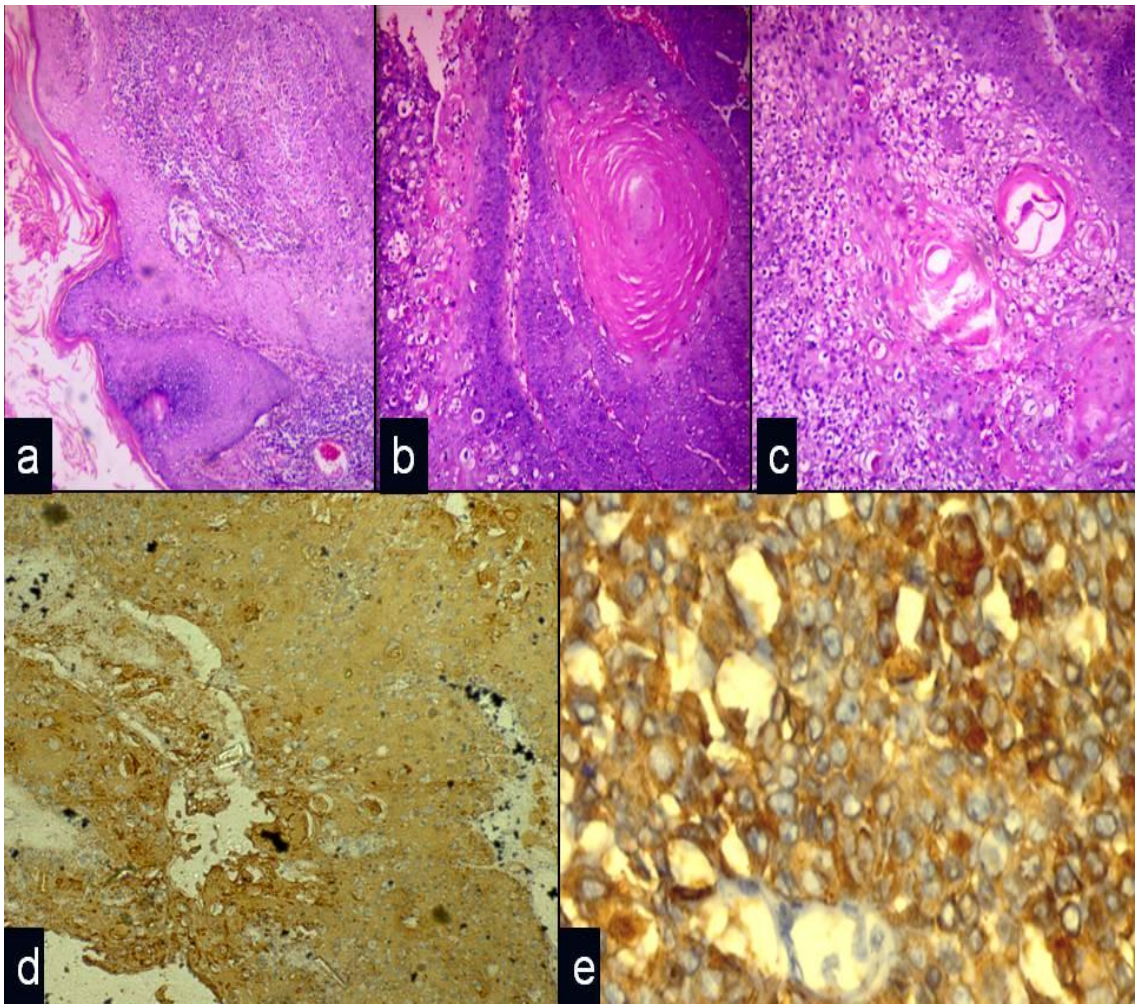
Trichilemmal carcinoma is a rare cutaneous adnexal tumour arising from the outer root sheath of the hair follicular epithelium. [1] It is often observed as a solitary lesion occurring on sun exposed areas especially head and neck, face, extremities and trunk. [2] Morphological resemblance and overlapping features with other high grade skin tumours often lead to confusion and misinterpretation. In such cases, ancillary tests like immunohistochemistry helps in identifying the lesion. Wide surgical excision is the treatment of choice.

Case report:

A 62-years-old healthy woman with low socio-economic background presented to the department of dermatology with a nodule on the scalp which was painless but rapidly growing for the past 4 months. There was no history of any chronic disease in the patient. Personal and family history was not significant. On local examination, the nodule was non tender with no pigmentation, ulceration or fungation and measured 2x2 cms in size. A clinical diagnosis of proliferating trichilemmal tumour was made. It was excised and sent for histopathological examination.

Grossly, a greyish white, multinodular mass was received which was 2.5 X 2 X 1 cms in size. Cut surface revealed greyish white solid areas with multiloculated cystic spaces filled with yellow-tan fluid. Microscopically, a well circumscribed cystic tumour was seen that was mostly confined to dermis but seen in continuity with the epidermis. (Figure 1(a)) The epidermal layer revealed palisading of small basaloid cells at the periphery with differentiation towards large keratinocytes with ample amount of eosinophilic cytoplasm. At places shadow cells and abrupt keratinisation without a granular layer was observed. (Figure 1(b)) The neoplastic cells were pleomorphic with large vesicular nuclei and clear cytoplasm. (Figure 1(c)) Occasional mitotic figures, focal areas of calcification and necrosis were also present. Adjoining area showed chronic inflammatory cells comprising of lymphocytes and plasma cells. Immunohistochemistry was performed using CK7, Pan Keratin (AE1/3) and CD34. The tumour cells showed intense staining with Pan Keratin (AE 1/3) (Figure 1(d)), CK7 (Figure 1(e)) and CD34. Thus, a diagnosis of trichilemmal carcinoma was confirmed based on the above findings.

Figure 1: Trichilemmal carcinoma (a) tumour seen in continuity with the epidermis (H & E, X50) (b) tumour showing abrupt keratinisation (H & E, X100) (c) neoplastic cells showing marked cytological atypia with clear cytoplasm (H & E, X100) (d) tumor cells were positive for AE1/3 (Immunoperoxidase, X100) (e) tumor cells showing CK7 positivity (Immunoperoxidase, X400)



Discussion:

Trichilemmal carcinoma was initially described by Headington and French in 1962 as a histologically invasive and cytologically atypical clear cell neoplasm that arises from outer root sheath of hair follicle [1]. It is usually seen in elderly age group with male to female ratio of 1:1. The commonest sites are head and neck, face, extremities and trunk but occasionally it has been reported in eyelid and upper lip. [2],[3] The pathogenesis for this carcinoma is still not clear, but sun-exposure, post-surgical radiation for other lesions (basal cell carcinoma for example), immuno-suppression after renal transplantation, or a pre-existing burn scar, predispose to the development of trichilemmal carcinoma. Transformation from benign trichilemmoma to trichilemmal carcinoma has also been postulated. [4],[5]

Clinically, it presents as a white or pale-tan papule, plaque or nodule on sun exposed areas measuring less than 3 cms in size. It may be associated with ulceration, hyperkeratosis or scabs. [6] It is often seen as a solitary lesion but reports on multiple presentations have been described. [7] One common syndrome associated with trichilemmal carcinoma is Cowden's syndrome. [8]

Microscopically, it is an invasive tumour that is seen in continuity with the epidermis or follicular epithelium. The tumour cells are arranged in lobules with foci of trichilemmal keratinisation. The neoplastic cells show marked cytological atypia with clear cytoplasm and hyperchromatic, pleomorphic & large nuclei. Accumulation of glycogen in the cytoplasm makes it PAS positive. Areas of haemorrhage and necrosis or cells with subnuclear basal

vacuolization can be seen. [9] Immunohistochemistry shows positivity for cytokeratins, like CK1, CK10, CK17, Pankeratin AE 2/3 and CD34 but negativity for carcinoembryonic antigen (CEA) and epithelial membrane antigen (EMA). [10]

The differentials considered are squamous cell carcinoma, basal cell carcinoma, nodular melanoma and proliferating trichilemmal tumors. [11] Trichilemmal carcinoma has a low malignant potential with indolent clinical course and the treatment includes wide surgical excision. It has a good prognosis and reports of deep invasion and local recurrence are unlikely.

Conclusion:

Trichilemmal carcinoma is a rare cutaneous adnexal tumour and the presence of clear cells often poses a great difficulty in distinguishing it from other tumours with clear cell morphology. Thus, a careful morphological interpretation along with the findings of immunohistochemistry is required to confirm the diagnosis. Complete wide excision and careful follow-up of the patient is sufficient to manage this tumour.

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