

Original Article

Trichilemmal carcinoma of cheek mimicking basal cell carcinoma : review of literature

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ABSTRACT:

Trichilemmal carcinoma (TC) is a rare, cutaneous adnexal neoplasm that typically occurs in elderly patients on sun-exposed areas of the body. It is conventionally considered to be a neoplasm derived from adnexal keratinocytes with glycogenated clear cells and evidence of outer root sheath (ORS) or trichilemmal differentiation, and is the malignant counterpart of trichilemmoma. Trichilemmal carcinoma has a benign clinical course, but exhibits a malignant and invasive histological appearance.¹ Since it is a rarely encountered tumour, it is very challenging even for an experienced histopathologist to make a definitive diagnosis. We report here a rare case of trichilemmal carcinoma which was clinically diagnosed as basal cell carcinoma.

INTRODUCTION:

The term trichilemmal carcinoma (TLCA) was originally described as a clinical entity by Headington in 1976, although it was not adopted by pathologists for many years. Recently, the publication of some series has contributed to distinguish the TLCA from other groups of follicular tumours.² It is considered a carcinoma of low malignant potential with rare metastases. As a rule, it appears as a solitary lesion after the fifth decade of life.³ The number of reported cases has been increasing lately. The tumor originates on the outer root sheath and is a malignant form of trichilemmoma. Most of the time, lesions present as papule or nodule with possible keratosis or ulceration on the top of the lesion. The diagnosis is established by means of histopathological examination using hematoxylin-eosin staining which, when necessary may be complemented by immunohistochemistry of the lesion. The treatment may be Mohs' technique or simple lesion excision.² Prognosis is generally good. Rate of local recurrence is low and it rarely metastasizes.⁴

CASE REPORT:

A 75 years postmenopausal lady presented to our hospital with ulcer over left cheek since 2 weeks. It was associated with pain since 5 days.

Local examination of left cheek showed a well circumscribed exophytic ulceroproliferative growth measuring 0.5X0.5 cms. Tenderness was present. Clinically, differential diagnosis of pyogenic granuloma or basal cell carcinoma of left cheek was made.

Wide excision biopsy was sent to our histopathology lab. On gross examination tumour was well circumscribed with exophytic ulceroproliferative growth measuring 0.5X0.5 cms. Cut surface showed uniform pale white areas. Surgical margins were not involved grossly.

Microscopically, lesional tissue showed ulcerated epidermis with an exophytic tumour composed of cells arranged in nodular pattern separated by thin fibrous septa.(Fig 1A) Tumour cells were round to oval with nuclear atypia, peripheral palisading, moderate amount of eosinophilic cytoplasm, vesicular nuclei and prominent nucleoli (Fig 1B). Atypical mitosis (2/HPF) along with pilar keratinization was noted (Fig 1C). Dysplastic cells in lobules surrounded by plasma cells and lymphocytes are seen infiltrating the dermis. Surgical margins were free from tumour. Periodic Acid Schiff stain was contributory with tumour cells staining positive with and without diastase. Histopathological diagnosis was Trichilemmal Carcinoma.

DISCUSSION:

The term Trichilemmal Carcinoma (TLC) is described as a skin tumour which is a locally invasive, histologically malignant neoplasm of clear cells derived from the external root sheath epithelium of a hair follicle. The pathogenesis of TLC remains unclear. It is the malignant counterpart of trichilemmoma and has also been reported to be associated with the burn scar. Other factors like exposure to sunlight, repeated as well as long term doses of radiation have been implicated in the pathogenesis of trichilemmal carcinoma.⁵

TLC must be distinguished histologically from other clear cell skin tumours, particularly the clear cell variants of BCC, SCC and proliferating trichilemmal tumour. Lesions of SCC do not show lobular proliferation and trichilemmal keratinisation. Keratinisation in SCC, when present, is infundibular. Presence of peripheral palisading alone does not confirm the diagnosis of basal cell carcinoma. The clear cells in BCC occupy only a small portion of the tumour aggregate.⁶ According to Barr et al, electron microscopic studies have demonstrated that the clear cell change is the result of a degeneration phenomenon involving lysosomes. Proliferating trichilemmal tumours also have a similar histological picture but they usually arise in a pre existing cyst and have a predilection to occur in scalp. They also show high mitotic activity and sometimes keratinization with frequent metastasis. Establishing a diagnosis of malignancy is very important for therapeutic and prognostic purposes. Moreover some of these tumours may be associated with syndromes like multiple trichilemmoma seen in Cowden syndrome.⁷ Since it is a rare malignant skin tumour, the diagnosis mainly relies on the histological evaluation.

CONCLUSION:

Trichilemmal Carcinoma is a locally invasive and cytologically atypical tumour with very few case reports of distant metastasis. Its treatment is less aggressive compared to the more common high grade malignant skin tumours like SCC, BCC and other clear cell tumours. Because of its clinical as well as histological similarity to these tumours it is essential to consider the diagnosis of TLC, so that appropriate treatment can be rendered to the patient with minimum morbidity. Further, as the occurrence of TLC is rare and the literature on the same is insufficient, the addition of this case to the literature may prove helpful for reference in future.

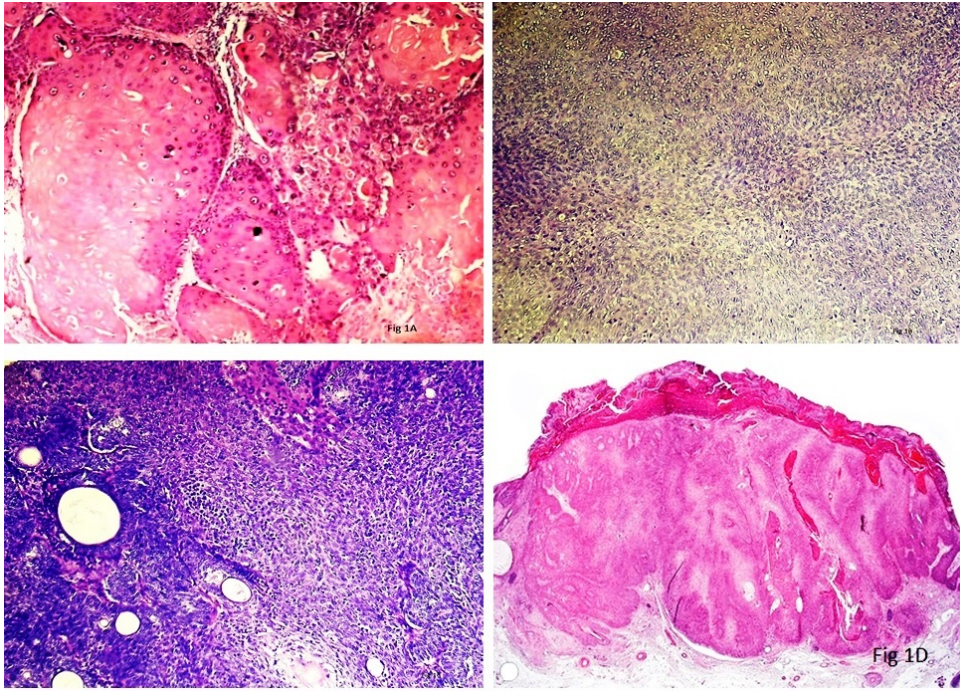


Fig 1A: Exophytic tumour composed of cells arranged in nodular pattern separated by thin fibrous septa.

Fig 1B: Tumour cells with vesicular nuclei.

Fig 1C: Tumour cells with pilar keratinization.

Fig 1D: Periodic Acid Schiff stain was contributory with tumour cells staining positive.

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