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Case Report

A rare convergence: Unveiling the uncommon presentation of trichoblastic carcinoma with cylindroma – A singular case report

Pratiksha Mishra[1], Meenakshi Mohapatro[1]*, Chinmayee Lenka[1], Fakir Charan Munda[1], Suryakanti Prusty[1], Lity Mohanty[1]

[1]Dept. of Pathology, SCB Medical College & Hospital Cuttack, Odisha, India

Abstract

Trichoblastic carcinoma is an infrequent and malignant neoplasm of cutaneous adnexal origin, arising subsequent to follicular differentiation. Clinically, it manifests as a well-circumscribed, solitary lesion, predominantly observed on the scalp. Cylindroma, conversely, is a benign and indolent adnexal tumor, with a predilection for the scalp, head, neck, and trunk, exhibiting a higher incidence in females.A 54-year-old female presented with a 2 cm swelling on the left anterior chest wall, which had progressively enlarged over the course of one month, accompanied by severe pain and mild pruritus. Her family and surgical histories were unremarkable, and clinical examination yielded no significant findings. The initial diagnosis of an infected sebaceous cyst was reconsidered following FNAC, which suggested a granulomatous lesion. Biopsy revealed a 2 cm globular mass containing yellowish-white material.Microscopic examination disclosed highly pleomorphic basaloid and malignant stromal cells arranged in an irregular pattern, along with a cylindromatous component exhibiting the characteristic jigsaw pattern. Immunohistochemical (IHC) analysis demonstrated strong PanCK and EMA positivity, leading to a final diagnosis of trichoblastic carcinoma with coexisting cylindroma.Trichoblastic carcinoma, a rare malignant cutaneous adnexal tumor, originates from the external root sheath of the hair follicle. Key histological features include highly pleomorphic basaloid and stromal cells with densely packed, hyperchromatic nuclei and abundant mitotic activity. Cylindromas may present as solitary or multiple lesions, though their histogenesis, whether apocrine or eccrine, remains ambiguous. Notable characteristics include a biphasic cell population arranged in a jigsaw pattern.

Keywords: Adnexal tumor, Trichoblastic carcinoma, Cylindroma, Immunohistochemistry.

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1. Introduction

Trichoblastic carcinoma, a rare malignant adnexal cutaneous tumor usually affects scalp (90% cases) with rare involvement of the trunk and extremities.[1] It usually originates from the pilar epithelium.[1] Any cutaneous tumor can develop due to the histomorphological differentiation of the primary adnexal structures present like sebaceous glands, sweat glands, apocrine and eccrine glands. So, trichoblastic carcinoma represents the malignant neoplasm of the follicular these primary germinative cells. However cylindroma is another rare, slow growing, benign cutaneous appendage tumor, constituting 0.7% of all adnexal neoplasms.[2] It usually affects the scalp, face and neck region, predominantly seen in females. Herein we present a case of trichoblastic carcinoma along with coexisting cylindroma in a 54 year female patient.

2. Case Report

A 54-year-old female presented to our surgery OPD with a gradually increasing swelling of 2 cm in diameter on left anterior chest wall since 5 years. The swelling was associated with severe pain and mild itching since 1 month. She was diagnosed with pulmonary tuberculosis and was under anti-tubercular medications since two months. There was no family history of any malignancy. Her past surgical history was also insignificant. On clinical examination, a 2 cm diameter firm non-tender nodular mass was present on the left postero-lateral back with no surrounding skin changes. Comprehensive blood examinations, laboratory tests, and

*Corresponding author: Meenakshi Mohapatro
Email: drmeenakshi1988@gmail.com

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imaging studies done were unremarkable. With the clinical diagnosis of infected sebaceous cyst, fine-needle aspiration cytology (FNAC) was done. The aspirated material was dirty. Cytosmear showed nonspecific findings with the presence of clusters of histiocytes and multinucleated giant cells on an inflammatory background. Hence, a provisional diagnosis of granulomatous lesion was given and the patient was advised for biopsy. Under all aseptic precautions, an elliptical incision was given and entire cyst sac was excised and tissue was sent for histopathological examination. Gross examination showed a whitish globular structure measuring 2cm in diameter whose cut section revealed yellowish whitish material. Microsection showed irregular nests, sheets, island, trabeculae and cords of markedly pleomorphic basaloid cells (**Figure 1**a, b) and malignant spindle cells scattered in fascicles (**Figure 2**a-c). Scattered foci of necrosis was noted (**Figure 4**b). Mitotic activity was brisk with 6 mitotic figures/10 HPF in both basaloid and malignant spindle cells were present. Intervening hyalinized collagenized stroma in between both basaloid and spindle cell islands were found (**Figure 2**c). One area showed a nodular proliferation of basaloid cells in compact nests fitted together in jigsaw pattern with intervening thick basement membrane material (**Figure 3**a,b). The nests showed a bimodal population of cells with peripherally arranged undifferentiated palisading cells and centrally located differentiated ductal cells. Few areas show clear cell and squamoid differentiation.

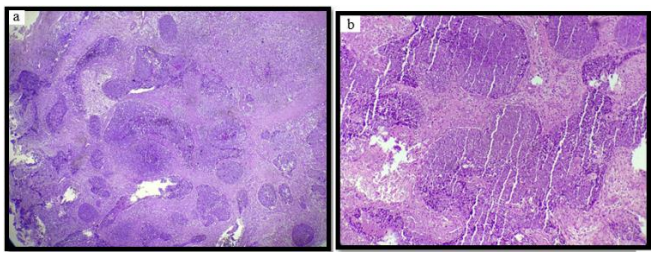

Figure 1: a,b: (100x, 400x): Microphotographs show pleomorphic basaloid cells arranged in irregular nests, cords, trabeculae and few in diffuse sheets.

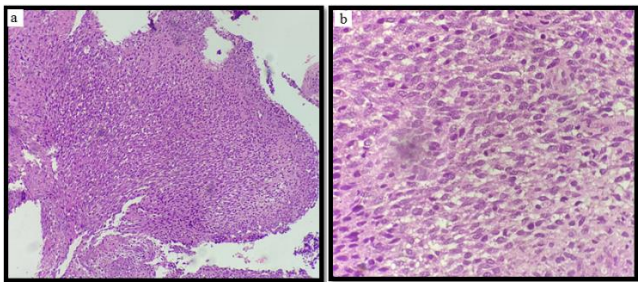

Figure 2: a,b: (100x, 400x): Microphotographs show highly pleomorphic stromal cells arranged in in diffuse sheets and in small fascicles.

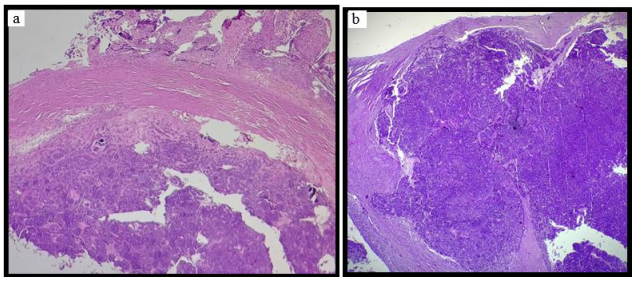

Figure 3: a,b: (100x): Microphotographs show compact nest of basaloid cells arranged in jigsaw pattern

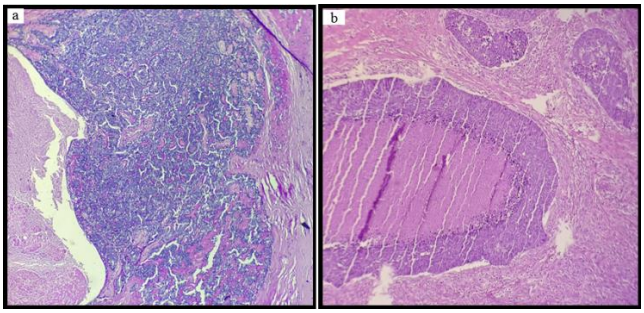

Figure 4: a: (100x): Microphotographs show PAS positivity in the intervening thickened basement membrane of cylindromatous component; **b:** (400x): Scattered foci of comedonecrosis noted.

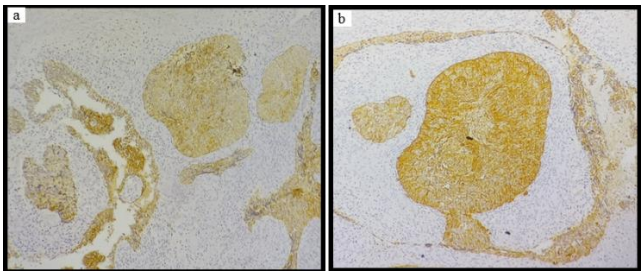

Figure 5: a,b: IHC (40x, 100x), PanCK showed strong cytoplasmic positivity in epithelial component of trichoblastic carcinoma.

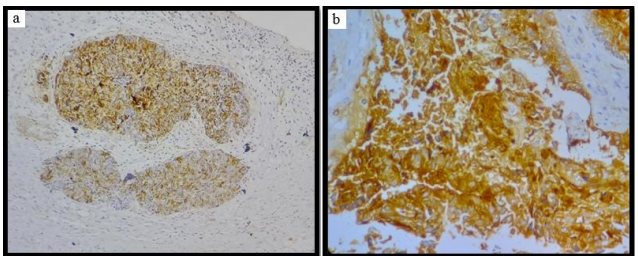

Figure 6: a b: IHC (100x, 400x): EMA) showed strong cytoplasmic positivity in epithelial component of both trichoblastic carcinoma and in ductal component of cylindroma

With a differential diagnosis of trichoblastic carcinoma with cylindroma and trichoblastic carcinoma with basal cell adenocarcinoma, immunohistochemistry was done. On immunohistochemistry (IHC), pancytokeratin (PanCK, **Figure 5**a,b) showed strong cytoplasmic positivity in

epithelial component of trichoblastic carcinoma and epithelial membrane antigen (EMA, **Figure 6**a,b) showed strong cytoplasmic positivity in epithelial component of both trichoblastic carcinoma and in ductal component of cylindroma, however vimentin, CK20 and desmin were negative for tumor cells. PAS showed strong positivity in basement membrane of cylindroma component (**Figure 4**a). Hence a final diagnosis of trichoblastic carcinoma along with coexisting cylindroma was made.

3. Discussion

Trichoblastic carcinoma is a rare malignant epithelial cutaneous adnexal tumor developing from the external root sheath of the hair follicle. Microscopically it shows irregular nests, sheets, cords, trabeculae of highly pleomorphic basaloid cells in lobules with no evidence of peripheral palisading. There could be features of squamous keratinization without intercellular bridges. Abundant mitoses is usually seen. There could be subcutaneous tissue infiltration by the tumor cells.[1] Trichoblastic carcinoma may show superficial ulceration of the epidermis similar to basal cell carcinoma (BCC), however BCC shows similar basaloid cells with features of prominent peripheral palisading, retraction artifact, necrosis and mitosis which are not present in our case. The malignant transformation of trichoblastic carcinoma is however rare and requires proper histopathological examination to distinguish between the two entities.[3] The presence of highly pleomorphic cells, crowded, hyperchromatic nucleus, and abundant mitotic figures favors malignant trichoblastic carcinoma. The presence of malignant pleomorphic stromal cells arranged in short fascicles differentiates it from other basaloid lesions.

The term cylindroma was first given by Billroth in 1959, due to its close resemblance to cylindrical shape when viewed in cross section.[4,5] They are most commonly found in the scalp and face with rare involvement of the trunk and extremities.[4] Cylindromas can be either solitary indicating a sporadic origin or can be multiple as seen in inherited familial cases which can be autosomal dominant.[6] The exact histogenesis still remains unknown whether it is of apocrine origin or eccrine origin despite extensive ultrastructural studies.[7,8] Microscopically, tumor cells are arranged in irregular nests, typical of jigsaw pattern surrounded by an eosinophilic pale pink material. The tumor cells are arranged in biphasic pattern. The central portion comprises of the well differentiated medium to large sized cells, having pale eosinophilic cytoplasm, centrally placed round nucleus having pale chromatin representing the ductal or secretory cells.[9] The peripheral portion of the nests represents the undifferentiated cells having palisaded arrangement of small to medium cells with round central nucleus and coarse clumped chromatin. Multiple cylindromas shows a higher rate of transformation to malignancy than the solitary counterparts.[10]

The coexistence of trichoblastic carcinoma along with cylindroma has not been reported yet, as seen in our case. Due to its rarity, proper identification and adequate histopathological examination along with clinical history forms the core for definite diagnosis of these two entities.

4. Conclusion

The diagnosis of skin adnexal tumors, including rare combinations such as trichoblastic carcinoma and cylindroma, is predominantly reliant on meticulous morphological evaluation. Immunohistochemistry (IHC) plays a supportive role, aiding in distinguishing between histologically overlapping entities and confirming lineage-specific markers. However, it is important to emphasize that IHC findings alone are not definitive.

In this case, the morphological assessment formed the basis of the diagnosis, with IHC used to corroborate and supplement the histopathological findings. This approach underscores the necessity of integrating IHC results with detailed microscopic examination to achieve an accurate and comprehensive diagnosis. The coexistence of trichoblastic carcinoma with cylindroma is very rare. Currently, there is no standard protocol for the management of both the entities and requires surgical excision with adequate follow ups to monitor for recurrence.

5. Conflict of Interest

None.

6. Source of Funding

None.

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