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Trichoblastic carcinoma of the penile shaft: A case report of a rare condition

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ABSTRACT

Trichoblastic carcinoma is a sporadic malignant tumor. It usually arises in the scalp, trunk, and extremities. However, its occurrence in the base of the penis is highly uncommon. To our knowledge, there were no similar cases reported in the literature. Case Presentation. A 65-year-old Tunisian man with no significant medical history consulted our department with a 10 cm ulcerative lesion on the base of the penis. The lesion biopsy was concordant with the diagnosis of basal cell carcinoma. The patient underwent a wide local excision. The tissue defect was reconstructed by an anterolateral pedicled flap of the thigh. The final histopathology report was concordant with the diagnosis of a trichoblastic carcinoma. Conclusion. There is no treatment consensus due to the few reported cases of trichoblastic carcinoma in the literature. Surgical excision with free margins remains the cornerstone of the treatment.

Category: Case Presentation

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Introduction

Trichoblastic carcinoma is a sporadic skin adnexal malignant tumor arising from the epithelium of the pilar sheath. Typically, it develops on a trichoblastoma after a long period of evolution [1,2]. However, unlike other skin malignancies, it occurs in non–sun-exposed sites [3]. This malignant tumor usually mimics basal cell carcinoma. Unlike basal cell carcinoma, trichoblastic carcinoma is not only more aggressive, but also with a much worse prognosis [4,5]. The diagnosis is usually difficult to establish. Consequently, the distinction between these types of malignancies is challenging but needs to be accurately established [6-8]. Treatment modalities have not been standardized, and various protocols have been used due to the rarity of this form of neoplasm [9,10].

We report a trichoblastic carcinoma case affecting an infrequent location, the base of the penis. It was treated by wide local excision and covered by an anterolateral skin flap of the thigh. We aim to discuss the importance of reconstructive surgery in such infrequent presentation through this case report.

Case Presentation

It is presented the case of a 65-year-old North African Caucasian man with no significant medical history, who presented to our department with a neglected slowly-enlarging ulcerative mass on the base of the penis of 10-years duration, with an ulceration emerging 5 months before the medical consultation. The mass measured 10 cm and was associated with erectile dysfunction. The patient denied any previous treatment history, sun or radiation exposure, sexually transmitted disease, or other skin tumors (Figure 1).

There was no evidence of dysuria and neither appreciable inguinal lymphadenopathy on physical examination. The pathology report of the biopsy was concordant with a basal cell carcinoma. The abdomino-pelvic US found no signs of metastases.

After evaluation in a multidisciplinary team, it was decided for wide local excision and reconstruction with a local skin flap (Figures 2, 3).

The operation lasted 240 minutes. The tumor was highly adherent to the deep anatomical structures, making

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it difficult to remove. The frozen section showed free surgical margins. The maximum dimensions of the removed specimen were 90mm/60mm/25mm (Figure 3). The recovery was uneventful. The patient was released on the third day.

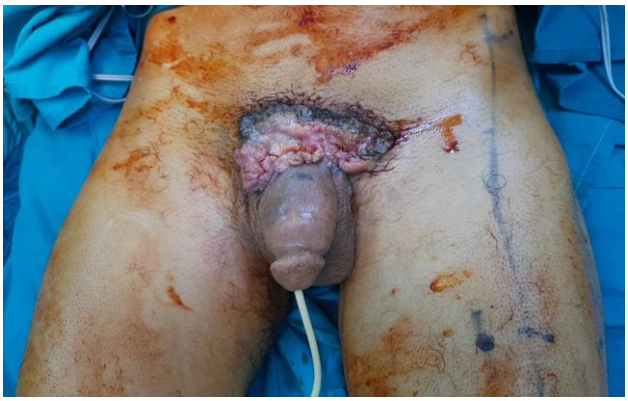

Figure 1. Trichoblastic carcinoma of the base of the penis

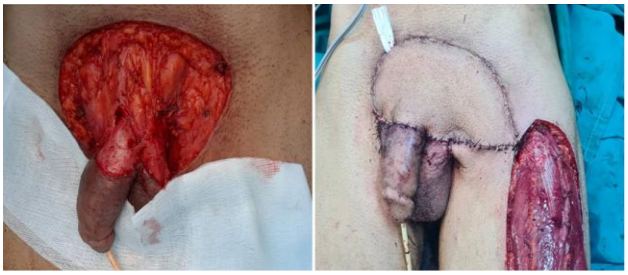

Figure 2. Tissue defect reconstructed by an ALT flap

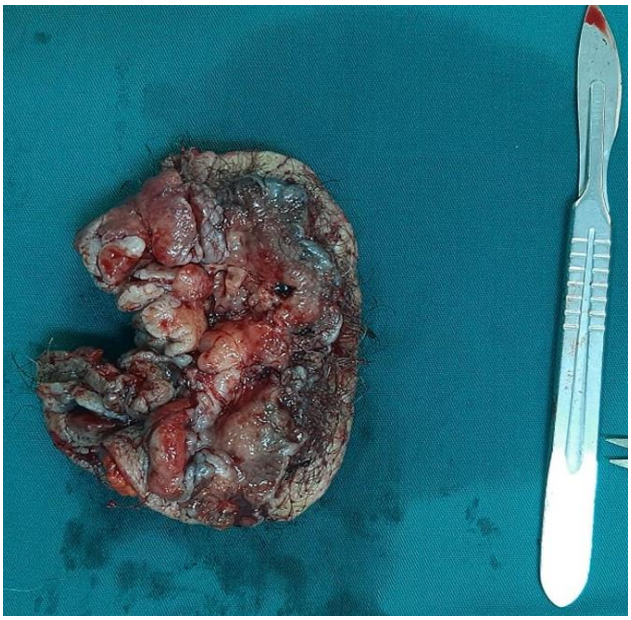

Figure 3. The excised specimen

Histopathological definitive examination revealed a large, ulcerated epithelial proliferation infiltrating dermis and hypodermis and focally connected to the epidermis (Figure 4A). The tumor consisted of trabeculae, islands, adenoid structures, and sheets of basaloid cells with inconstant peripheral palisading and clefting between tumor and stroma (Figure 4B). The stroma was fibrous and cellular (Figure 4C). Tumor cells showed enlarged atypical nuclei (Figure 4D). There was no perineural nor vaso-invasive growth.

The multidisciplinary meeting decision was to start with a close follow-up. Every four months the first two years, then every six months until five years of follow-up.

The follow-up will be based on clinical examination. The other exams will be performed according to the clinical signs. After 8 months of follow-up, the patient shows no sign of relapse.

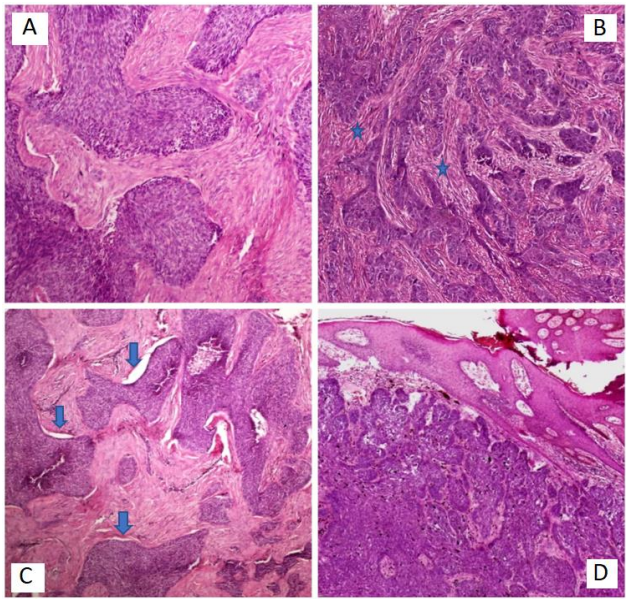

Figure 4. Pathological findings

A - (HE X 400): Tumor cell showing enlarged atypical nuclei
B - (HE X 200): Cellular fibrous stroma (★)
C - (HE X 200): Trabeculae and sheets of basaloid cells with inconstant peripheral palisading and clefting (⬇)
D - (HE X 200): Tumor infiltrating the dermis and focally connected to the epidermis

Discussion

Trichoblastic carcinoma is often a sporadic adnexal malignant tumor [11,12]. It arises from the epithelium of the pilar sheath, still having an insufficiently known pathogenesis [13,14]. It usually develops de NOVO, but it can also develop after a long evolution of a trichoblastoma [15,16]. Generally, it occurs in non-sun-exposed sites, unlike other skin malignancies [2]. The most common locations are the scalp, trunk, and extremities [17,18]. Its occurrence on the base of the penis is infrequent. It usually affects aged people around the seventh decade of life [19,20].

It manifests as a solitary, poorly defined, asymmetric dermal or subcutaneous mass [21,22]. Trichoblastic carcinoma is difficult to diagnose, even for professional pathologists, due to its rarity [23]. Generally, it is a basaloid poorly circumscribed tumor, having few epidermal connections [24,25].

Trichoblastic carcinoma is a biphasic malignant neoplasm with dual differentiation towards the specialized

follicular stroma (like that found in trichoblastoma) and germinative follicular cells which can be strikingly atypical [26].

The epithelial component is composed of basaloid cells resembling those seen in conventional basal cell carcinoma but showing nuclear crowding, abnormal mitotic figures, and necrosis [27,28].

The epithelial component is intimately associated with the stroma, which recapitulates specific follicular mesenchyme. Perineural infiltrations indicate a poor prognosis for local management. Occasionally, vascular emboli are seen [26,27].

Trichoblastic carcinoma is distinguished from trichoblastoma by atypia in the epithelial component.

Accurate diagnosis is challenging and is entirely dependent on adequate sampling, as both the benign and the malignant aspects of the tumor need to be represented in the biopsy. The tumour may also be mistaken for basal cell carcinoma [10], which is characterized by peripheral palisades, epithelial-stromal cleft artifact, and stromal mucin, which are not typically associated with high-grade trichoblastic carcinoma. Other basaloid carcinomas that enter the differential diagnosis are sebaceous and Merkel cell carcinoma [9].

It is crucial to make the correct diagnosis because trichoblastic carcinoma, unlike basal cell carcinoma, is generally more aggressive and has a high metastatic potential [29,30].

Due to the rarity of reported cases, there is no therapeutic consensus for this condition [31,32]. Surgical resection with free margins is the treatment of choice, especially with the lack of effectiveness of radiation and chemotherapy [33,34].

Radiotherapy is an alternative option when the tumor is unresectable or in the presence of residual disease. Seventy Gy dose should be delivered for macroscopic disease. Adjuvant radiotherapy at a dose of at least 50 Gy is recommended for microscopic residual disease [9].

Conventional chemotherapy shows disappointing results for metastatic disease [10]. Sunitinib (a tyrosine kinase inhibitor with antiangiogenic action) and vismodegib (inhibitor of the hedgehog pathway) are two novel medicines that appear to be promising [12,32].

Our patient was initially diagnosed with basal cell carcinoma which is the principal differential diagnosis of trichoblastic carcinoma. He underwent a wide local excision, and the defect was covered by a local pedicled anterolateral thigh skin flap.

The imperative of surgery with wide margins might lead to not satisfying cosmetic outcomes. Reconstructive surgery could be necessary to cover tissue defects with several techniques using skin graft, local skin flap, or myocutaneous flap. The pedicled anterolateral thigh flap is a flexible flap with several benefits. These advantages include a long and dependable pedicle that enables a wide arch of rotation, the ability to harvest a big skin area, elevating the flap with underlying fascia and muscle, as well as minor donor site morbidity [35,36].

The follow-up is imperative because of the local recurrence rate and the metastatic risk. It will be carried out at least every six months. It includes a clinical examination associated with additional examinations in case of warning signs [37,38].

Conclusions

Trichoblastic carcinoma is a very rare malignancy with a poor prognosis. The genital localization is generally rare. Chemotherapy, radiation and immunotherapy have not demonstrated remarkable effectiveness so far. Consequently, surgery remains the main therapeutic method of condition, especially in the case of voluminous formations which are unaesthetic and often associate functional deficits.

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Conflict of interest disclosure

There are no known conflicts of interest in the publication of this article. The manuscript was read and approved by all authors.

Compliance with ethical standards

Any aspect of the work covered in this manuscript has been conducted with the ethical approval of all relevant bodies and that such approvals are acknowledged within the manuscript.

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