Periocular Trichoblastoma: A Diagnostic Dilemma

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Abstract
A 41-year-old woman presented with a large medial canthal lesion that extended over the midline. Excisional biopsy revealed a multinodular basaloid tumor, located within the dermis and subcutaneous fat that extended into skeletal muscle at the deep and peripheral margin. Histology was consistent with trichoblastoma. A review of the literature reveals a lack of specific and sensitive immunohistochemistry markers to establish the diagnosis of trichoblastoma. Following reconstruction, the patient has shown no signs of recurrence of the lesion at 24 months.

Introduction
Originally described by Headington in the 1970s, trichoblastoma (TBL) is now appreciated as a rare benign tumor that originates from the follicular germ cell line \cite{1,2}. Typical presentation involves papules or nodules around 1–2 cm in diameter in adults 40–50 years of age in the region of the head and neck. TBL is classified in the latest skin tumor classification by the World Health Organization as one with a favorable outcome. Diagnosis of TBL is challenging given its resemblance to basal cell carcinoma (BCC) but is important given possible malignant transformation and potential metastasis. There are only six previously
reported cases of TBL in the periocular region with varying methods used for diagnosis. The study adhered to the principles of the Declaration of Helsinki. The patient provided written consent to publication of her case and clinical photograph.

**Case Report/Case Presentation**

A 41-year-old Caucasian female presented with an 8-month history of an enlarging right medial canthal lesion, which had extended across the midline. She believed it had commenced as a "black head," from which she had expressed cheesy material upon squeezing. The lesion measured $30 \times 15$ mm in size, was fluctuant in nature, and appeared to be tethered to the bridge of the nose as shown in Figure 1. Skin changes overlying the lesion appeared to be consistent with the patient's history of chronic picking and rubbing of a (previously ruptured) cystic lesion. The patient smoked tobacco, and her medical history was significant for depression for which she took mirtazapine. She underwent an excisional biopsy and reconstruction using a supraclavicular skin graft.

Histology showed a large, multinodular basaloid tumor with background solar elastosis. It was located in the dermis and subcutaneous fat, extended into skeletal muscle, and was focally attached to the epidermis as shown in Figure 2. A few separate tumor islands were present in the dermis. The tumor cells had high nuclear:cytoplasmic ratios and inconspicuous nucleoli. Mitotic activity was present, but no abnormal forms were seen. There were some apoptotic cells, but no necrosis. No perineural or vascular invasion was identified. A prominent finding was a ribbon-like arrangement of the cells, in which the nuclei were elongated and aligned in palisades as shown in Figure 3.

**Fig. 1.** Clinical photography demonstrating a medial canthus tumor extending over the midline.

**Fig. 2.** Multinodular basaloid tumor with background solar elastosis.
The tumor focally extended to the deep and peripheral margins. Immunocytochemistry confirmed the epithelial nature of the tumor (MNF116, AE1/3, BerEP4 all positive). Neuroendocrine markers (synaptophysin, chromogranin, CD56), melanocytic markers (Melan-A, S-100, HMB45), TTF1, and CK20 were all negative.

At 1-month post-excision, there was no sign of any recurrence and the patient declined further excision following full discussion of the histology results. There were no signs of recurrence at 24-month follow-up.

**Discussion/Conclusion**

TBL, trichoepithelioma, and BCC are all dermatological tumors of the follicular germ cell line. The term TBL was originally reserved for one of four hair tumors that had primitive cellular appearances to an embryonic hair [1]. However, the term has undergone several alterations since and the collective term of “trichogenic tumors” has been proposed for benign neoplasms of hair follicles, including TBL and trichoepithelioma [3, 4]. A diagnostic dilemma presents itself when trichogenic tumors – rare, benign epithelial tumors – and BCC – the most common malignant skin cancer – share several overlapping clinical and histological characteristics [2, 5–7]. Establishing the correct diagnosis is vital in these instances as surgical treatment for each differs: BCCs require surgical resection with free margins, while TBL requires simple surgical resection [8]. Histologically, our case had considerable overlap with BCC with a slight irregular border, sun damage, and focal epidermal attachment. Lack of retraction artifact, tumor necrosis, and the ribbon-like arrangement of cells combined with the clinical findings favored the diagnosis of TBL.

Establishing a differential diagnosis between benign dermatological tumors and their counterpart malignancies is mainly achieved through histology with hematoxylin and eosin staining. Microscopically, the hallmark features of BCCs are a uniform proliferation of cells that have elongated oval hyperchromatic nuclei, minimal pale cytoplasm, and prominent palisading of cells near the periphery of the tumor [3]. TBL shares significant histological overlap with BCC, including fissures between the epithelium and stroma and peripheral palisades. However, TBL rarely presents with necrosis, mitotic activity, or lymphocytic...
infiltration [8]. A highly specific, but not always present – such as in our case, feature of TBL is the presence of papillary mesenchymal bodies [2]. Dermoscopic differences between BCC and TBL include the presence of blue-gray ovoid nests and globules that are more frequently found in BCC than TBL. However, the use of a dermascope alone to establish a diagnosis is not advised [9]. Clinically, the distribution of the dermatological tumors differs. Both BCC and TBL classically present in middle-age and elderly individuals; however, BCC can affect numerous sun-exposed sites: face, scalp, neck, chest, and back. TBLs on the other hand are less likely to affect peripheral sites beyond the head and neck, although this is not a strict rule [3].

In certain cases, the use of immunohistochemistry may provide ancillary data. Three of the six publications of periocular TBL have documented positivity for CD34 and CD10 in diagnosed cases [10–12]. In the only other publication to state the use of immunohistochemistry, the specimen was positive for chromogranin A and neuron-specific enolase [13]. The diagnosis of TBL in our case was questioned given the clinical findings of a pearly, ulcerated nodule with the presence of stromal clefting histologically closely fitting the description of an infundibulocystic BCC; however, immunocytochemistry confirmed the epithelial nature of TBL [14]. A specific marker has not been identified to differentiate TBL from BCC, and it may be that both of these entities exist on a spectrum with considerable overlap of features [2].

Management of previously documented TBL cases has included wide local excision as well as Mohs’ micrographic surgery [10–15]. Recurrences requiring multiple re-excisions have been reported following positive margin excisions [10, 14]. Although our case demonstrated positive margins, the patient declined further treatment and there were no signs of recurrence at 24 months.

**Statement of Ethics**

Ethics approval for this study was not required as per the Human Research Authority (HRA) online decision-making tool. Written informed consent was obtained from the patient for the publication of her case and clinical photograph.

**Conflict of Interest Statement**

The authors have no conflicts of interest to declare.

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**Author Contributions**

Pav Gounder, Carole Caldwell, Paul Craig, Daisy Bassey-Duke, and Simon N Madge – substantial contribution to conception and design of the case report and drafting the manuscript and gave final approval to be published version. Yarrow Scantling-Birch – substantial contribution to revision of the manuscript following first review and gave final approval to be published version.
Data Availability Statement

All data generated or analyzed during this study are included in this article. Further inquiries can be directed to the corresponding author.

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