



Basaloid follicular hamartoma of the eyelid in a pediatric patient

Nicole Jedrzynski, William Plum, Ann Q. Tran, Andrea A. Tooley, Mitra Dowlatshahi, Michael Kazim*

Edward S. Harkness Eye Institute Columbia University College of Physicians and Surgeons, New York, NY, USA

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ABSTRACT

Purpose: We report a case a pediatric patient with an eyelid lesion found to be a basaloid follicular hamartoma. **Observations:** A six-year-old female with juvenile diabetes who presented with a benign eyelid lesion harboring an aberrant eyelash. **Conclusions and Importance:** Basaloid follicular hamartoma is a rare benign neoplasm arising from hair follicles. These lesions can resemble basal cell carcinomas and require complete excision.

1. Introduction

Basaloid follicular hamartoma (BFH) is a rare benign neoplasm arising from hair follicles. Clinically and histopathologically, BFH can resemble basal cell carcinomas and are associated with autoimmune disease. There are two reports of eyelid BFH, however, none in the pediatric population. We report a six-year-old girl with juvenile diabetes who presents with a flesh colored papule of the eyelid harboring an aberrant eyelash found to be a basaloid follicular hamartoma.

2. Case report

A six-year-old Caucasian girl was referred for evaluation of a slowly progressive right upper eyelid lesion, which had developed over the prior two years. The lesion was neither painful nor pruritic. The patient denied a history of trauma, or discharge. The patient's medical history included juvenile diabetes mellitus. Present on the right upper eyelid was a non-tender, flesh colored papule measuring 1.5×1.0 mm with single white eyelash, approximately three times the length of the surrounding, normal lashes (Fig. 1). There was no surrounding madarosis or eyelid distortion.

A gross total surgical excision of the lesion was performed. Histopathological analysis demonstrated proliferating anastomosing cords of basaloid epithelium with variable peripheral palisading surrounded by a scant fibroblastic stroma consistent with basaloid follicular hamartoma (BFH) (Fig. 2). Four months post-operatively, there has been no recurrence.

3. Discussion

Basaloid follicular hamartomas (BFH) are benign tumors of the hair follicles, which typically occur as a single lesion or in linear arrangements or clusters along the head, neck, and trunk.¹ There are two prior reported cases of upper eyelid BFH in adults.^{8,9} While they are considered benign, BFH may resemble basal cell carcinomas and rare cases of malignant transformation have been documented.² Additionally, they are often associated with congenital syndromes, myasthenia gravis, systemic lupus erythematosus, alopecia, and Graves' Disease.³⁻⁷

Clinically, they often present as a flesh-colored, smooth papules and histopathologically demonstrate lattice patterns of interlaced basaloid strands and may be adjacent to the infundibulum of a hair follicle. In our case, the pathology was distinguished from basal cell carcinoma by the absence of nuclear atypia, mitotic activity, and artifactual retraction spaces between the stroma and the epithelium. Immunohistochemical staining has been advocated to distinguish BFH as they stain positively for the BCL-2 gene only around the perimeter, exhibit CD34 and CD10 positivity throughout the stroma and vasculature, and are Ki-67 negative.^{10,11} In contrast, basal cell carcinomas stain positively for BCL-2 gene throughout the lesion and are typically Ki-67 positive.

The differential diagnosis for basaloid follicular hamartoma differs depending on the age, location, and distribution of the lesion. The more common benign hair follicle tumors described in the ophthalmic literature include trichoepithelioma, trichilemmoma, and tricolliculoma. Absent Gorlin syndrome, the suspicion for basal cell carcinoma in a pediatric patient would be low however must be considered in an adult.

* Corresponding author. Edward S. Harkness Eye Institute, Columbia University College of Physicians and Surgeons, 635 West 165th Street, New York, NY, 10032, USA.

E-mail address: Mk5477@nyp.org (M. Kazim).

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Fig. 1. Basaloid follicular hamartoma on the eyelid. Raised, flesh colored lesion found to be a basaloid follicular hamartoma of the right upper eyelid.

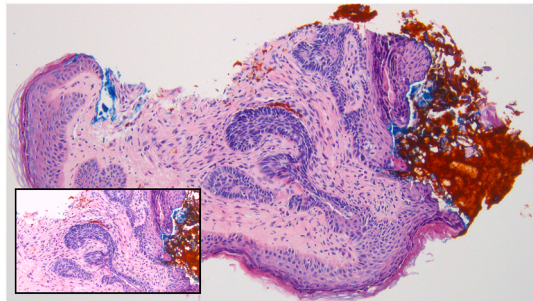


Fig. 2. Histologic staining of basaloid follicular hamartoma. Cords of basaloid epithelium with peripheral palisading within fibrous stroma of the dermis, detailed inset of basaloid follicular hamartoma at higher magnification (H&E 10x, inset 20x).

Therefore, complete surgical excision, where possible, is recommended.

4. Conclusion

We present the first case of basaloid follicular hamartoma (BFH) of the eyelid in a pediatric patient with juvenile diabetes mellitus. Given the rare potential of malignant transformation, complete surgical excision is recommended. As well, further monitoring for the development of autoimmune conditions should be considered as the child ages. Basaloid follicular hamartoma should be considered among the differential diagnosis of an eyelid lesion along the eyelash margin.

Patient consent

The patient's legal guardian consented to publication of the case in

writing.

Authorship

All authors attest that they meet the current ICMJE criteria for Authorship.

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Declaration of competing interest

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