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Pseudoepitheliomatous Hyperplasia as a Limbal Mass Mimicking Nodular Episcleritis

Dear Editor,

Pseudoepitheliomatous hyperplasia (PEH) is a benign proliferation of the conjunctival or corneal epithelium that occurs in response to inflammatory conditions [1]. There are a few reports of occurrence of PEH presenting as a limbal mass, especially in inflammatory conditions such as vernal keratoconjunctivitis (VKC) [2-4]. Here, we describe an uncommon case of PEH with vascular tortuosity of the conjunctiva in a patient misdiagnosed with nodular episcleritis. The diagnosis of PEH was confirmed by histopathologic findings.

A 28-year-old man was referred to Farabi Eye Hospital emergency department with a complaint of local pain and redness in his right eye for 1 month. The past medical and ocular surgical history were unremarkable, with no history of atopy or contact lens wear. He had been diagnosed with nodular episcleritis in another ophthalmology clinic and was treated with topical betamethasone and diclophenac drops every 6 hours for 8 weeks. He had no improvement using these medications. Upon initial examination, his visual acuity was 10 / 10 in both eyes. Slit lamp examination was normal except for the right eye, in which there was a grayish nodular, placoid-free mobile elevated mass with white pearly hypertrophied conjunctiva, sized approximately 2 mm × 3 mm and located at the nasal side of the limbus (Fig. 1A). There was a ciliary injection at the nasal conjunctivae and around the lesion, with tortuosity and dilation of episcleral vessels resembling sentinel vessels. Other ophthalmic examinations were unremarkable in both eyes. Based on the fact that the patient had no response to topical corticosteroids and nonsteroidal anti-inflammatory drugs, and also considering the features of the lesion (nodular mass with leukoplakia), concerns were raised regarding malignancy. An excisional biopsy was performed. The lesion was excised with a 1 mm margin with amniotic membrane transplantation and the specimen was sent in formalin for histopathologic examination. The pathology report (Fig. 1B) revealed a stratified squamous

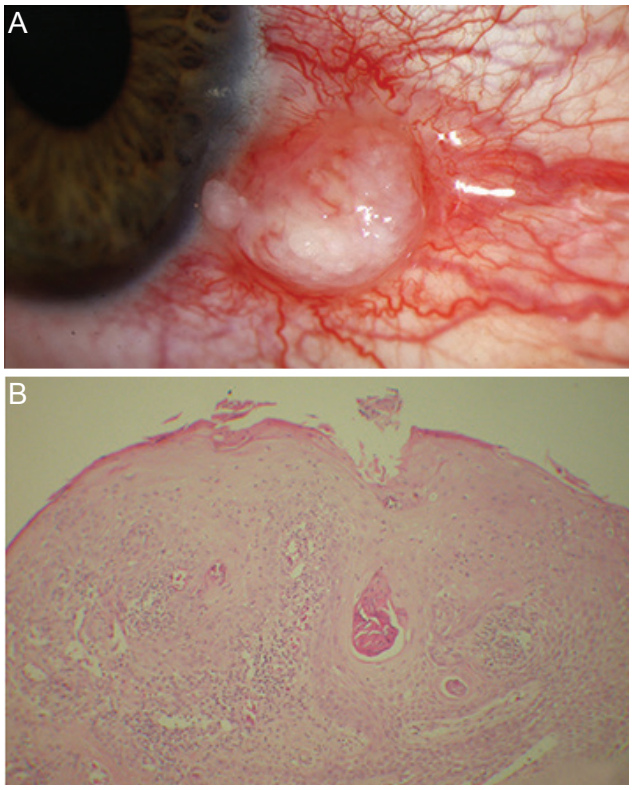

Fig. 1. (A) Limbal white nodular mass with conjunctival vascular tortuosity mimicking ocular surface squamous neoplasia. (B) Acanthosis, spongiosis, and sparse intraepithelial infiltration of leukocytes with marked downward proliferation of the surface stratified squamous epithelium are observed. There is also evidence of keratinization at the squamous cells, accompanied by the complete loss of goblet cells. Substantia propria showed edema, vascular proliferation and prominent infiltration of lymphoplasmacytic and neutrophils leukocytes. No considerable nuclear atypia or any atypical mitotic figure was noted (H&E, ×100).

epithelium showing PEH, spongiosis, and infiltration of leukocytes. There was marked infiltration of lymphoplasmaocytic leukocytes admixed with numerous neutrophilic leukocytes in an edematous substantia properia. Based on the microscopic findings, the patient was diagnosed with PEH of conjunctivae with acute and chronic inflammation. Two months following excisional biopsy, there was no recurrence of the lesion.

With the typical presentation of a white hyperkeratotic elevated mass, PEH is a benign proliferation of the conjunctival or corneal epithelium that occurs in response to inflammatory conditions. It is important to clinically differentiate PEH from ocular surface squamous neoplasia, especially when it arises at the limbus [1]. There has been a reported case of epithelial hyperplasia as a rare complication of cultivated limbal epithelium transplantation [4]. There are also two reports of occurrence of such a mass in patients with VKC [2,3]. Schwab et al. [3] reported a case of limbal VKC with nodular limbal mass associated with superior hypertrophic conjunctival thickening. Malhotra et al. [2] reported PEH in a case of palpebral VKC presenting as a limbal mass. They confirmed the diagnosis based on histopathologic findings that were consistent with PEH. The lesion had no features suggestive of malignancy. They hypothesized that chronic underlying inflammation aggravated by mechanical microtrauma to the limbal epithelium due to eye rubbing led to the hyperplasia [4]. Herein, we report a case of limbal PEH with no known history of VKC or other irritating conditions that had been misdiagnosed as nodular episcleritis. The presence of white pearly hypertrophied conjunctiva with vessel tortuosity resembling sentinel vessels, raised concerns towards malignancy as the existence of capillary fronds favored the diagnosis of ocular surface squamous neoplasia or papillomas in contrast to PEH, which usually lacks vessel tortuosity [2]. However, histopathological examination of the excision biopsy revealed PEH, spongiosis, and infiltration of leukocytes. No goblet cells were seen in the region and there was no evidence of dysplasia or abnormal mitotic activity. We hypothesized that the lesion must have been caused by an underlying inflammatory condition, e.g., a foreign body at the limbus or neglected nodular episcleritis. We concluded that PEH should be considered as a differential diagnosis of such masses, especially in cases with a history of other causes of chronic inflammation other than VKC in order to avoid the surgical excision with broad margins required for ocular surface squamous neoplasia.

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Conflict of Interest

No potential conflict of interest relevant to this article was reported.

References

1. McLean IW, editor. *Tumors of the eye and ocular adnexa*. Washington, DC: Armed Forces Institute of Pathology; 1995. p. 53-4.
2. Malhotra C, Jain AK, Thapa B. Limbal pseudoepitheliomatous hyperplasia mimicking ocular surface squamous neoplasia in palpebral vernal keratoconjunctivitis. *Case Rep Ophthalmol Med* 2013;2013:527230.
3. Schwab IR, Schwab L, Cavender JC. Limbal vernal keratoconjunctivitis with a hypertrophic limbal mass lesion. *Ann Ophthalmol* 1987;19:79-80.
4. Fatima A, Matalia HP, Vemuganti GK, et al. Pseudoepitheliomatous hyperplasia mimicking ocular surface squamous neoplasia following cultivated limbal epithelium transplantation. *Clin Experiment Ophthalmol* 2006;34:889-91.