Solitary Fibrofolliculoma of the Upper Eyelid in a 68-year Old Female: A Case Report

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Case report

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Abstract

Background Fibrofolliculoma is a benign, perifollicular, connective tissue tumor, which is a clinically asymptomatic, 2-4 mm, skin-colored, dome-shaped smooth papule. It usually arises in the form of multiple lesions, and very rarely presents as a solitary lesion. Case presentation A 68-year-old female presented with an asymptomatic, flesh-colored mass on the right upper eyelid. The 5×5×4 mm lesion was located near upper lid margin. It was a skin-colored, dome-shaped, smooth nodule, with eyelashes on the surface, rubbery to palpation and not painful. The lesion was excised completely, and the diagnosis of fibrofolliculoma was confirmed through a histological exam. Conclusions Solitary fibrofolliculomas is rarely arising in the eyelid. However, it should be suspected when a localized mass lesion of the eyelid is encountered.

Background

Fibrofolliculoma usually is clinically asymptomatic multiple connective tissue tumor□apperaring perifollicular, skin color, dome, smooth papules, with diameters of 2-4 mm, located on head and neck. Multiple fibrofolliculomas generally are inherited as an autosomal dominant trait and share clinically characteristic of Birt-Hogg-Dubé (BHD) syndrome, which is associated with multiple fibrofolliculomas, acrochordons, trichodiscomas, and internal neoplasms [1, 2].

Fibrofolliculoma very rarely presents as a solitary lesion, firstly reported in 1984. Solitary forms are usually unassociated with other cutaneous abnormalities with typically nonhereditary[3,4]. Only 12 cases are so far published in literature. To our knowledge, our case is the second one reported on the eyelid. Herein, we presented the clinical feature and surgical treatment of the rare solitary fibrofolliculoma located on upper eyelid in a 68-year-old female.

Case Presentation

A 68-years-old Mongoloid woman presented with an asymptomatic, flesh-colored lesion in the right upper eyelid. The lesion increased in size slowly for 5 years. No similar lesion was found in other parts of the body. Her medical and family histories were unremarkable, and she had experienced no triggering trauma.

Ophthalmologic examination, the protruding lesion was around 5×5×4 mm, located on the upper lid margin (Fig. 1). Palpation of the lesion did not elicit pain. The visual acuity of both eyes was 20/20. On the photography of anterior segment, the nodule was fresh-colored, dome-shaped, with eyelashes on the smooth surface, and the lesion located on the anterior lamella of the lid margin (Fig.1) the anterior segment was unmarkable and so as the fundus examination results. Examinations showed that her left eye was normal.

Under local anesthesia, anterior lamella of the eyelid was resected with a trigonal wedge, with the removal of 1 mm of extra tissue from the margin of the lesion, and thin layer of the lid plate. The anterior lamella defect of the upper eyelid was repaired using A-T flap. A gray line split was performed on the cut ends of

skin defects, then, the skin defects were sutured directly. After the operation, the pressure was bandaged for 48 hours.

Histologic examination of the lesion showed a well-defined tumor mass involving a hair follicle and a proliferation of multiple thin strands of basaloid cells extending from the central follicle into the surrounding fibrous stroma. The fibrous stroma showed a sharp contrast with the surrounding dermis. Hematoxylin-eosin stains showed mucin content in the stroma (Fig. 2). The histologic findings were characteristic of fibrofolliculoma.

During the 3-month follow-up, no signs of recurrence or new lesions appeared.

Discussion

The cases of solitary fibrofolliculoma were extremely rare since being first reported in 1984 [3]. Only eight previous papers involving 12 cases have been published worldwide [3-10]. We reviewed the previously published cases and found that most lesions were found on the face, and only 2 around eyes, one on the eyelid and the other on the eyebow (Table 1). Among all the cases, no sex preponderance is seen (5 men and 8 women), and the mean age at presentation was 51.4 years. The duration of symptom varying from several months to years. Solitary fibrofolliculomas share clinic apperance of multiple fibrofolliculomas, yellowish to flesh-colored and dome-shaped papules.

Historically, fibrofolliculoma has both an epithelial and a mesenchymal origin, showing distinctive and characteristic features with minor variation [3]. The center of the lesion shows a hair follicle and consists of an expansion of the fibrous root sheath, which typically surrounds the hair follicle along with proliferating bands or ribbons of perifollicular connective tissue. Cesinaro and coauthors found immunohistochemical expression of factor XIIIa in the bizarre perifollicular cells in a background of CD34-positive spindle cells, which aids for better characterize the nature of the lesion [9].

Since solitary fibrofolliculoma are extremely infrequently and definitively diagnosed only by historical results, it can be easily overlooked or clinically misdiagnose. The 37-year female patient reported by Chang and coauthors had previously misdiagnosis as chalazion and received incision and curettage only. For years, her condition had not improved and recurred several times [5]. Fortunately, fibrofolliculoma rarely develop to malignant ones. Once this type of a lesion in the eyelid is observed, a diagnosis of fibrofolliculoma should be considered. As the lesion of the present case located on the eyelid, it also should be included as a differential diagnosis from malignant conditions such as basal cell carcinoma and squamous cell carcinomas, which are most common malignant eyelid tumors.

As sharing the characteristic of BHD syndrome, fibrofolliculoma are considered to be hamartomas composed of both connective tissue and follicular epithelial component [11]. Most fibrofolliculoma may have some common histogenesis such as abnormal function of hair follicle bulge cells, and differential diagnosis should be considered in the histopathological exam.

Surgical excision is usually chosen for the skin fibrofolliculomas at first operation for pathological dignosis. CO2 laser or erbium-doped YAG laser might be a better choice for multiple fibrofolliculomas or recurrent lesions[12-13]. Currently, there is no uniform standard for the eyelid lesion. Surgical treatment for the eyelid lesion should be individualized based on the size, growth rate, invasion, and interference with eyelid function and esthetics[14-15]. For clinically supposed eyelid benign tumor, surgery is preferred with an excisional biopsy under frozen section or clinical margins of at least 1-mm and with reconstruction of the eyelid at the same time. Also, regular postoperative follow-up is necessary to observe the recurrence, and to improve facial appearance as required.

Conclusions

Although rare, solitary fibrofolliculomas should be suspected when a localized mass lesion of the eyelid is encountered. We present a rare case of the lesion arising on the unusual location of the upper eyelid. This report highlights both the clinical and the histopathologic features. The findings may extend the knowledge on experiences of solitary fibrofolliculoma on eyelids.

Declarations Section

Ethics approval and consent to participate

All procedures performed were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki Declaration and its later amendments or comparable ethical standards. This is a case report, retrospectively describing the course of the diagnostics and therapy, thus does not require the local Bioethical Committee approval.

Consent for publication

Written informed consent was obtained from the patient for the publication of this report and any accompanying images.

Availability of data and materials

More data if necessary are available from the corresponding author on reasonable request.

Competing interests

The authors declare no competing financial interests

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Authors' contributions

W.W and J.C.were responsible for the design of the study, surgery for the patient, collected the data, wrote the manuscript, and revised the manuscript and all authors have read and approved the manuscript.

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Abbreviations

BHD syndrome : Birt-Hogg-Dubé syndrome

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Table

Due to technical limitations, Table 1 is only available as a download in the supplemental files section

Figures

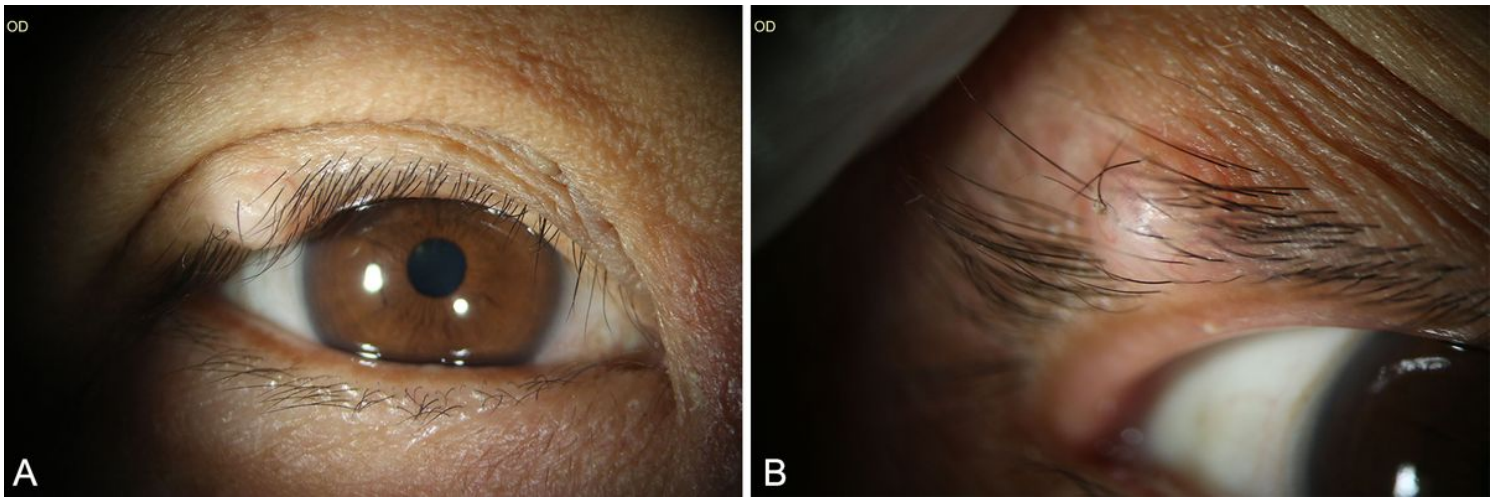

Figure 1

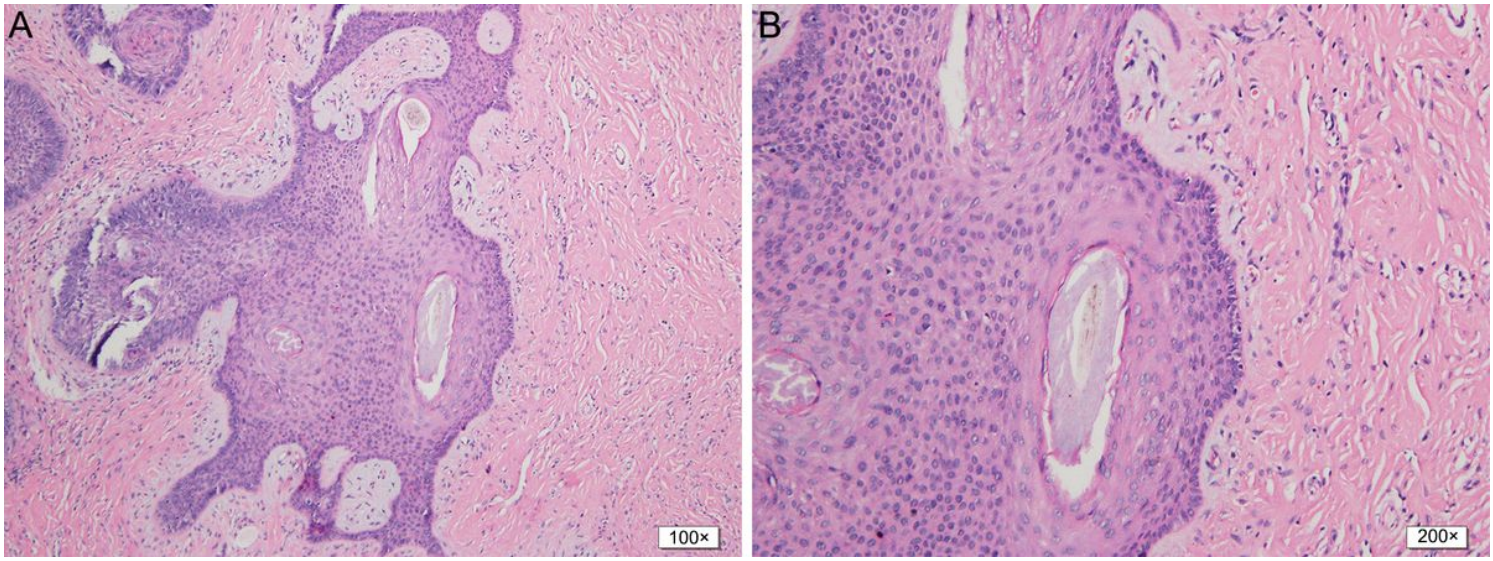

Figure 2

Supplementary Files

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