

75894_Auto_Edited.docx

Rare giant corneal keloid presenting 26 years after trauma: A case report

Traumatic giant corneal keloid

Shang Li, Jiang Lei, Yinghui Wang, Xiaolin Xu, Ke Yang, Ying Jie

Abstract

BACKGROUND

Corneal keloid is a rare clinical disease with an unknown etiology, which is easily misdiagnosed. Surgery is the most effective treatment but is rarely reported in the literature. Herein, we report the clinical features, histopathology, and surgical outcome of a giant corneal keloid with trophoblastic vessels and discuss the genesis of the mass.

CASE SUMMARY

A 36-year-old young man was admitted to the hospital because of a large mass on the surface of the left cornea. The patient had suffered an injury to his left eye at the age of 6-years-old; however, as the injury did not cause cornea perforation, he did not undergo treatment. Slit lamp exam showed a large, elevated, opaque lesion that covered the entire cornea and protruded from the surface of the eyeball. Anterior segment optical coherence tomography (AS-OCT) revealed a lesion of irregular density involving the anterior stroma. We suspected a secondary corneal fibroproliferative mass based on the clinical history, and slit lamp and AS-OCT findings. The patient subsequently underwent a superficial keratectomy and keratoplasty, and the final diagnosis of corneal keloid was confirmed by intraoperative histopathological examination.

CONCLUSION

Non-penetrating corneal trauma damages corneal epithelium basement membrane, initiating stromal fibrosis and causing corneal keloids. AS-OCT and biopsy confirm diagnosis.

Key Words: Corneal keloid; Histopathology; Immunohistochemical staining; Anterior segment ocular coherence tomography; Deep anterior lamellar keratoplasty; Case report

Li S, Lei J, Wang Y, Xu X, Yang K, Jie Y. Rare giant corneal keloid presenting 26 years after trauma: A case report . *World J Clin Cases* 2022; In press

Core Tip: ³ Corneal keloid is a benign proliferation of fibrous or fibrovascular tissue in the corneal stroma, and is rarely encountered in the clinic. The onset of secondary corneal keloids can occur months to years after the surgery or trauma, and the depth of corneal infiltration varies. Histopathology is the gold standard for diagnosis, but the cause of the disease is not yet clear. Here, we report a case of a giant corneal keloid that occurred 26 years after trauma and discuss the cause in detail.

INTRODUCTION

Corneal keloid is a rare type of ophthalmic disease that presents as a white, painless, enlarged vascular or avascular solitary nodule. It can also involve the entire corneal stroma, with a smooth surface^[1]. The diagnosis of corneal keloid can be challenging. It is often referred to as corneal fibroma, myofibroma, and hypertrophic scars, and is easily misdiagnosed as corneal dermatoid tumor, ocular surface squamous neoplasia, and corneal inclusion cyst^[2-5]. Corneal keloid can appear primary or secondary to trauma and surgery; secondary keloid, in particular, can occur anytime from months to years after the corneal injury and is commonly treated with surgery^[6-8]. Since the morphology and time of onset of corneal keloid are highly variable, biopsy and histopathology are essential for a definitive diagnosis.

Herein, we report the clinical features, histopathology, ² and anterior segment optical coherence tomography (AS-OCT) features in a case of giant corneal keloid presenting approximately 26 years after corneal trauma.

CASE PRESENTATION

Chief complaints

A 36-year-old man was referred to our hospital in December 2019 to address a corneal swelling found in his left eye 4 years prior. Over the past 2 years, the mass had rapidly

increased until it protruded from the surface of the eye, preventing the eyelid from closing properly.

History of present illness

The patient reported having suffered an injury to the left eye at the age of 6-years-old; as the injury had not caused a corneal perforation, treatment was foregone. Since then, however, he had suffered from recurrent redness and a foreign body sensation, which led him to rub his eyes often.

4

History of past illness

The patient's medical history was unremarkable.

Personal and family history

Not available at this time.

Physical examination

7

On initial examination, the best-corrected visual acuity (BCVA) was light perception/30 cm for the left eye and 20/20 for the right eye. Slit lamp showed conjunctival congestion with a thick nutrient vessel in the inferior temporal quadrant extending to the corneal limbal epithelium (Figure 1A). A white, tough, round, raised mass, 7 mm high and 9 mm in diameter, with numerous tiny vessels on the surface, localized in the center of the cornea, covered almost the entire cornea surface, preventing the eyelid from closing properly (Figure 1B). Anterior chamber details could not be visualized.

Laboratory examinations

Not available at this time.

Imaging examinations

Ultrasonographic evaluation of the posterior segment yielded normal findings. AS-OCT showed ¹irregular epithelial tissue and dense subepithelial tissue with a low density of blood vessel invasion, similar to a honeycomb-like structure (Figure 1C).

FURTHER DIAGNOSTIC WORK-UP

Detailed history-taking of the patient's birth revealed that his mother had a spontaneous full-term delivery and no history of medications taken during pregnancy. No swelling was observed in any parts of the patient's body other than the cornea. The results of routine blood, urine, and immunological examinations were negative.

PRELIMINARY DIAGNOSIS

Secondary corneal fibroproliferative mass

FINAL DIAGNOSIS

Corneal keloid due to pathological examination

TREATMENT

The lesion was removed *via* superficial keratectomy. Based on the depth of infiltration of the nodule, a deep anterior lamellar keratoplasty was performed (Figure 2). The excised mass was dissected, soaked in formalin, and sent for pathological examination.

¹ Histopathological examination revealed irregular epithelial hyperplasia without Bowman's layer and abundant subepithelial collagen fibers with irregular proliferation and degenerative changes, which were lined with fibroblasts and vascular cavities (Figures 3A,3B). Immunohistochemical staining of the keloid showed diffuse positivity for vimentin and smooth muscle actin (SMA), highlighting the smooth muscle wall of the vasculature and myofibroblasts (Figures 3C, 3D).

OUTCOME AND FOLLOW-UP

¹ Postoperatively, the patient was prescribed topical steroids, antibiotics, and lubricants. On the first day after surgery, the BCVA was 20/100, and epithelialization of the ocular surface was completed 1 wk later (Figure 4A). There was no recurrence of swelling or post-operative complications during the 4-months follow-up, and the patient reported satisfaction with the postoperative visual acuity and appearance (Figure 4B). ¹ AS-OCT showed that the corneal graft was closely attached to the bed, and the residual stroma thickness was about 100 μ m (Figure 4C).

DISCUSSION

Corneal keloid is a rare clinical disease, classified as congenital, primary, or secondary depending on the etiology^[1]. The earliest case of corneal keloid was reported by Szokalski in 1865^[9], and since then approximately 80 cases of corneal keloid have been reported in the literature^[2]. Most of the reported cases have occurred secondary to trauma, inflammation, or surgery, and the duration of onset has varied from a few months to several years. Herein, we reported a case of giant corneal keloid presenting approximately 26 years after corneal trauma.

There is no uniformity in the pathogenesis of corneal keloid, according to the collective cases reported. Some scholars believe that keloids result from the replacement of embedded iris and inflammatory exudate by fibrovascular tissue^[9]. However, in our case, the patient had no history of penetrating corneal injury, so we believe that the keloid originated from the injury and persistent eye rubbing, which led to excessive corneal repair.

Under normal conditions, an intact corneal epithelial basement membrane (EBM) protects the corneal stroma from the presence of myofibroblasts. Once the EBM is damaged, cytokines such as transforming growth factor beta-1 (TGF β 1), TGF β 2, and platelet-derived growth factor of corneal epithelial origin enter the stroma layer, contributing to the proliferation and differentiation of myofibroblast precursors^[10]. If the EBM is repaired within 1-3 wk, the fibroblast precursors do not mature^[11]. We assumed that the EBM could not repair the damage in this patient due to frequent eye

rubbing and cytokines that were continuously secreted to stimulate myofibroblast activation. As this persisted for years, secretion of large amounts of extracellular matrix, in turn, led to the formation of a giant corneal keloid. Thus, both vimentin and SMA staining within the keloid were positive.

The enlargement of the swelling also depends on the blood vessel supply. The growth of neovascularization into the cornea is a complex process, in which multiple mediators such as inflammatory regulators, cytokines, growth factors, and matrix metalloproteinases interact. The neovascularization itself can assist dendritic cell (DC) migration into the central area^[12]. DCs promote keloid proliferation by secreting a series of cytokines that activate inflammatory pathways in the cornea^[13].

² After being diagnosed with corneal keloids, 82.6% of patients underwent surgery, these include superficial keratectomy (SK), lamellar excision/keratoplasty, penetrating keratoplasty (PK), sclerokeratoplasty, and enucleation^[6, 14-16]. SK remains the preferred choice of most physicians, with 70% of patients undergoing the procedure receiving SK. However, according to the literature, a non-dissected mass leads to recurrence in 40% of patients^[3]. Bakhtiari performed SK in 1 patient, after which the corneal opacification recurred, so he had to undergo 2 SK procedures combining PK ⁶ and mitomycin C, followed by a femtosecond laser-assisted deep anterior lamellar keratoplasty^[15]. Therefore, we recommend choosing surgery based on the depth of the infiltrated mass, with the aim of complete removal of the mass from the corneal stroma. For lesions located in the corneal stroma that do not breach the Descemet's membrane, SK combined with amniotic membrane graft or deep lamellar corneal graft may be an option. For lesions that infiltrate into the endothelium or do not reach the endothelium but cause corneal endothelial edema, PK surgery is an option. In our case, the corneal keloid and neovascularization infiltrated the deep stromal layer of the cornea without breaking through the Descemet's membrane; thus, the deep anterior lamellar keratoplasty procedure was selected.

CONCLUSION

Corneal keloids can occur up to 26 years secondary to non-penetrating corneal trauma. The genesis of such keloids may be related to EBM injury with cytokine clustering. AS-OCT plays an important role in early diagnosis of the disease, but confirmation of the diagnosis still relies on histopathological examination. Surgery is the treatment modality of choice for corneal keloid, and the operator should choose the procedure based on depth of infiltration.

ORIGINALITY REPORT

10%

SIMILARITY INDEX

PRIMARY SOURCES

1	www.researchsquare.com Internet	75 words — 4%
2	Joel R. Palko, Safa Arfeen, Asim V. Farooq, Catherine Reppa, George J. Harocopos. "Corneal keloid presenting forty years after penetrating injury: Case report and literature review", Survey of Ophthalmology, 2019 Crossref	34 words — 2%
3	www.ncbi.nlm.nih.gov Internet	16 words — 1%
4	www.wjgnet.com Internet	15 words — 1%
5	f6publishing.blob.core.windows.net Internet	14 words — 1%
6	mediskin.cn Internet	13 words — 1%
7	Www.dovepress.com Internet	12 words — 1%

