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Deep lamellar keratoscleroplasty for epibulbar dermoids: a case series

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Background: Corneal and limbal dermoids are benign congenital tumors which are most commonly located in the inferotemporal quadrant. Tumor growth results in astigmatism, leading to anisometropic amblyopia. It is important to select a surgical treatment option contributing to a reduction in astigmatism and an improvement in visual acuity, and leading to good or excellent cosmetic results.

Purpose: To report a case series of epibulbar dermoids treated with deep lamellar keratoscleroplasty.

Material and Methods: We report on 4 cases (age range, 14 to 40 years) with epibulbar corneal limbal dermoids (2 eyes) and limbal dermoids (3 eyes). Dermoid excision by deep lamellar keratoscleroplasty was performed in three eyes, and by peripheral lamellar keratoscleroplasty, in one eye. In addition, no surgery was performed in one eye with a grade I dermoid. Patients underwent general eye examination, ocular photography was performed for documentation purposes, and excised dermoids were sent for histomorphological examination.

Results: In all cases, histomorphological studies confirmed the benign nature of the disease. In addition, the corneal portion of the corneoscleral graft or the corneal graft was clear. Deep lamellar keratoscleroplasty for corneal and limbal dermoids contributed to a significant reduction in corneal astigmatism, improvement in visual acuity and satisfactory cosmetic results. No corneoscleral graft rejection or neovascularization was noted and no infectious complication was observed.

Keywords:

corneal dermoid, limbal dermoid, deep lamellar keratoscleroplasty, cornea

Introduction

Epibulbar dermoids are congenital conjunctival and/or corneal neoplasms which represent slow-growing, minimally vascularized whitish or yellowish semicircular benign lesions that may contain hair follicles and eyelashes [1, 2]. Maternal viral disease in pregnancy may result in the development of epibulbar dermoids in the fetus [3]. Corneal dermoids are the most common congenital orbital lesions and account for 25% of invasive ocular growths and 75% of cystic lesions [1, 2, 4]. Corneal and/or limbal dermoids are most commonly located in the inferotemporal quadrant. A study by Nevares and colleagues [5] indicates that the majority (76%) of ocular dermoids occur at the inferotemporal bulbar location of the eye. Ocular dermoids, however, may be present entirely within the perilimbal cornea or confined to the conjunctiva or sclera. Limbal dermoids may include four pathological types: dermoids, lipodermoids, complex choristomas and epibulbar osseous choristomas [6]. Most of these tumors manifest as dermoids containing piloceleous structures, stratified squamous epithelium, and choristomatous tissue, a dense connective tissue similar to the skin corium [5, 7]. Complex dermoids contain not only the aforementioned structures

but also bony structures (Fig. 1). Most epibulbar dermoids are unilateral and are typically located close to or in the limbus in the temporal and inferior quadrants, with partial extension to the sclera [5]. They are frequently associated with other anomalies of the body. Associated systemic abnormalities include preauricular appendages and auricular



Fig. 1. Complex dermoid (conjunctival osseous choristoma)

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Table 1. Visual scoring system for limbal dermoid

Item	Score			
	0	1	2	3
Area of corneal involvement	None involved	≤ Outer ¼ quadrant, not involving the optical axis	Outer ¼ quadrant < diameter < outer ½ quadrant, not involving the optical axis	Diameter ≥ outer ½ quadrant, involving the optical axis
Surface shape	None involved	Slightly raised, cannot be observed when the eye is closed	Moderately raised, can be observed when the eye is closed	Highly raised, interferes with closing the eye
Area of conjunctival involvement	None involved	≤ 50% of the conjunctiva	> 50% of the conjunctiva	In addition to the conjunctiva, the sclera or orbital tissue are also involved

Note: A total score of 0 to 3 is categorized as grade I, a total score of 4 to 6 as grade II, and a total score of more than 6 as grade III

fistulae. In Goldenhar syndrome, conjunctival dermoids are associated with preauricular appendages, whereas the classical facial defects are hemifacial microsomia with associated ipsilateral micrognathia and macrostomia. Secondary ocular abnormalities in Goldenhar syndrome include eyelid coloboma, lacrimal abnormalities, and scleral and lid staphyloma [2, 8, 9]. It should be mentioned that, epibulbar dermoids in Goldenhar syndrome may be bilateral [10]. Mann created a classification scheme for dermoids based on the depth of corneal involvement [11]. Based on the depth of corneal involvement dermoids are classified into three grades (grade I, II and III), which facilitates the selection of optimal treatment strategy.

Mann's classification for epibulbar dermoids (1993)

Grade I limbal dermoids are superficial lesions measuring less than 5 mm and are localized to the limbus (one quadrant affected).

Grade II limbal dermoids are larger lesions covering most of the cornea and extending deep into the stroma down to Descemet's membrane without involving it (one to two quadrants affected).

Grade III limbal dermoids, the least common of all the presenting dermoids, are large lesions covering the whole cornea and extending through the histological structures between the anterior surface of the globe and the pigmented epithelium of the iris.

Zhong and colleagues (2018) [3] conducted a retrospective study on 261 patients with limbal dermoid to establish a new scoring system for limbal dermoid (Table 1).

Astigmatic refractive errors associated with limbal dermoids have been reported [12-14]. Therefore, although benign, dermoid may affect vision, gradually causing corneal astigmatism and leading to the development of anisometropic amblyopia. In addition, they may affect the tear film, thus leading to the development of dry eye, and the esthetics of the patient. Therefore, early and effective surgical treatment of dermoid neoplasms is important.

The purpose of the study was to report a case series of epibulbar dermoids treated with deep lamellar keratoscleroplasty.

Material and Methods

We report on 5 eyes (4 patients), including 2 eyes with corneal limbal dermoid and 3 eyes with limbal dermoid. Patients (age range, 17 to 40 years) were treated at State Institution "The Filatov Institute of Eye Diseases and Tissue Therapy of the National Academy of Medical Sciences of Ukraine". Three eyes had grade I dermoid, and two eyes, grade II dermoid as per Mann's classification. Three patients had a monolateral ocular lesion, and one patient (that with Goldenhar syndrome), a bilateral ocular lesion. All four patients received surgery for dermoids. Dermoid excision by deep lamellar keratoscleroplasty was performed in three eyes, and by peripheral lamellar keratoscleroplasty, in one eye. In addition, no surgery was performed in one eye with a grade I dermoid.

Patients underwent general eye examination. Excised dermoids were sent for histopathological examination. Ocular photography was performed for documentation purposes. The patients were followed for 12 months after surgery. Informed consent for processing of personal clinical data for publication was obtained.

Surgical technique for dermoid excision (deep lamellar keratoscleroplasty)

Surgeries were performed under neuroleptanalgesia with additional epibulbar and retrobulbar anesthesia. The eyelid skin was disinfected with chlorhexidine 0.5%. The conjunctival cavity was washed with sufacyl sodium 30%. Superior and inferior rectus muscles were secured with 6-0 silk sutures, if required. A scraper was used to dissect the conjunctiva from the scleral portion of the lesion and remove the dermoid from the corneal, limbal and scleral surfaces, while preserving the capsule of the dermoid. Dermoid size was measured with a caliper, and trephination of the superficial cornea (within the area of opacification) was performed. A disc knife and Castroviejo scissors were used to remove opacified corneal layers layer by layer. In addition, superficial scleral layers at the lesion site were removed, and the bed for the lamellar corneoscleral graft was cut. Thereafter, a scraper and disc knife were used to strip of a keratobioimplant (KBI) and trephine was used

to cut a lamellar corneoscleral graft conforming in size to the bed in the recipient's eye. The graft was placed in the prepared bed and secured to the cornea with interrupted or running 10-0 nylon sutures, to the limbus with interrupted 8-0 nylon sutures, and to the episclera with interrupted 8-0 silk sutures. The dissected conjunctiva was secured at the limbus with interrupted 8-0 sutures.

Affected eyes were injected subconjunctivally with an antibiotic and corticosteroid. A sterile monocular dressing was applied to conclude the procedure. Postoperatively, we applied topical antiseptics; antibiotics (7 days); regenerative agents, and dexpanthenol. In addition, dexamethasone was administered in tapering doses, and preservative-free tear substitutes (trehalose 3% and hyaluronic acid 0.15%) were applied.

Peripheral lamellar keratoplasty was performed after dermoid removal in an eye with a small (< 4 mm) limbal dermoid.

Results

Case 1

A 17-year-old male presented with complaints of a bilateral ocular lesion, cosmetic defect, an increase in size of the lesion, and reduced vision in the left eye. At presentation, visual acuity was 1.0 OD and 0.3 OS, improving to 0.6 using +9.25 D sphere/-1.75 D cylinder at 35 degrees.

Slit-lamp biomicroscopy of the right eye revealed a whitish round limbal tumor measuring ≤ 4.0 mm in the largest dimension and located temporally at 7 o'clock. The clinical diagnosis was grade I corneal limbal dermoid OD (Fig. 2).

Slit-lamp biomicroscopy of the left eye revealed a white round corneal mass (with partial involvement of the limbus and sclera) measuring ≤ 7.0 mm in the largest dimension and located temporally between 2 and 5 o'clock. The rest of the corneal surface was clear. The clinical diagnosis was grade II corneal limbal dermoid OS (Fig. 3).

The left eye underwent peripheral lamellar keratoscleroplasty. The graft surface epithelialized within 7 days. The corneal portion of the lamellar graft remained clear within a one-year follow-up period.

Postoperatively, UCVA OS was 0.5, improving to 0.85 with -1.0 D cylinder at 110°.

On histopathology, the tumor surface appeared as stratified squamous epithelium, and subepithelial tissue showed degenerative hyaline changes with localized fat tissue.

Case 2

A 24-year-old male presented with complaints of an increase in size of a lesion, reduction in vision and cosmetic defect in the left eye over the most recent two years. At presentation, visual acuity was 1.0 OD and 0.4 OS, improving to 0.5 using -9.25 D sphere/+1.75 D cylinder at 20 degrees.

Slit-lamp biomicroscopy of the left eye revealed a whitish round corneal mass with partial limbal involvement measuring ≤ 6.0 mm in the largest dimension and

located temporally between 2 and 6 o'clock. The rest of the corneal surface was clear.

The clinical diagnosis was grade II corneal limbal dermoid OS (Fig. 4). The left eye underwent deep lamellar keratoscleroplasty. The graft surface epithelialized within 6 days. The corneal portion of the graft was clear at 1 year.

Postoperatively, visual acuity was 0.7 OS, improving to 1.0 using -1.0 D sphere (Fig. 5).

Case 3

A 40-year-old male presented with complaints of a lesion and cosmetic defect in the left eye. He also complained of a feeling of discomfort and foreign body sensation which he believed to be due to dusty conditions causing eye irritation.

At presentation, visual acuity was 1.0 OD and 0.6 OS. Slit-lamp biomicroscopy of the left eye revealed a moderately vascularized limbal conjunctival mass measuring 5 x 6 mm, located between 4 and 5 o'clock and containing hair. There was local degenerative opacification of the cornea, but the rest of the cornea was clear. The clinical diagnosis was grade I limbal dermoid OS (Fig. 6).

The patient underwent surgical excision of the dermoid with peripheral lamellar keratoscleroplasty. The graft surface epithelialized within 6 days. Postoperative UCVA was 0.85 OS (Fig. 7). Histopathology staining revealed a fragment of corneal tissue with stromal accumulation of amyloid.

Case 4

A 33-year-old female presented with complaints of an ocular lesion and cosmetic defect in the right eye. Over the recent year, she had a feeling of discomfort and foreign body sensation in the eye.

At presentation, patient's UCVA was 1.0 OD and 1.0 OS. Slit-lamp biomicroscopy of the right eye revealed a whitish conjunctival tumor extending to the limbus, measuring 5 x 6 mm, located between 8 and 9 o'clock, and containing hair fragments. There was degenerative opacification of the peripheral cornea, but the rest of the cornea was clear. The clinical diagnosis was grade I limbal dermoid with peripheral corneal degeneration OD (Fig. 8).

The patient underwent surgical excision of the dermoid with deep lamellar keratoscleroplasty. The corneal graft surface epithelialized within 6 days. The corneal portion of the graft was clear. Postoperative visual acuity was 1.0 OD (Fig. 9).

Histopathology staining revealed stratified squamous epithelium with sites of acantholytic squamous epithelial cells. In addition, amorphous cotton-like deposits were seen in the stroma underlying and adhering to the basal corneal epithelium. Moreover, numerous capillary type vessels were seen in the surrounding stroma.

In all cases reported here, the corneal portion of the corneoscleral graft or the corneal graft remained clear at 12-month follow-up, and high visual acuity was achieved and maintained. No corneoscleral graft rejection or neovascularization was noted and no infectious complication was observed.

Discussion

Dermoids are benign tumors located both over the cornea and sclera and possibly extending deep into the corneal stroma and anterior chamber of the eye [11]. Some authors believe that dermoids are choristomas denoting masses of normal fetal tissue found in an abnormal location. Dermoids are treated surgically (dermoid is excised, and the defect is closed, with the method of closure depending on the size, location depth and involvement of other ocular structures) [15]. Various techniques for surgical removal of epibulbar dermoid have been reported in recent decades. Simple dermoid excision from the limbus may result in corneal vascularization, persistent epithelial defects, scarring, and development of pseudopterygium and conjunctival symblepharon [16]. Hong and colleagues [17, 18] used autologous limbal cell transplantation in two patients with good results, with none of the aforementioned complications.

Others performed dermoid excision with peripheral amniotic membrane graft [19-21]; peripheral keratoplasty using donor cornea [13, 22] or sclerokeratoplasty. A technique of small-incision lenticule extraction (SMILE)-assisted sutureless resurfacing with interface tattooing for superficial limbal dermoids have been reported [14, 23]. After excision at a plane minimally below surrounding normal cornea and sclera, a corneal tattoo powder is carefully applied within an inked circular outline of an intended corneal margin. Fibrin-glue assisted corneal resurfacing is performed with the lenticule [23, 24].

In all cases reported here, the clinical diagnosis of corneal limbal dermoid or limbal dermoid was made based on biomicroscopy, intracapsular excision of the dermoid was followed by keratoscleroplasty, and histomorphological studies confirmed the benign nature of the disease. No intraoperative complications were seen. Of note that, although the clinical picture conformed to that in grade I or grade II dermoids, there was evidence of dermoid extending deep into the cornea to Descemet's membrane, which required the use of deep or peripheral lamellar keratoscleroplasty. In all cases reported here, the corneal surface epithelialized within 7 days, and there was improvement in astigmatism and visual acuity over a one-year follow-up.

Conclusion

Deep lamellar keratoscleroplasty for corneal and limbal dermoids contributed to a significant reduction in corneal astigmatism, improvement in visual acuity and satisfactory cosmetic results.

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Disclosures

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Conflict of Interest: The authors declare no conflict of interest that could influence their opinion regarding the subject matter or material described and discussed in this manuscript.

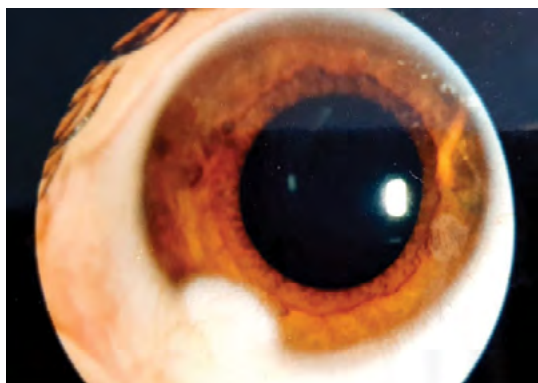


Fig. 2. Grade I limbal dermoid in the right eye of a 17-year-old male patient

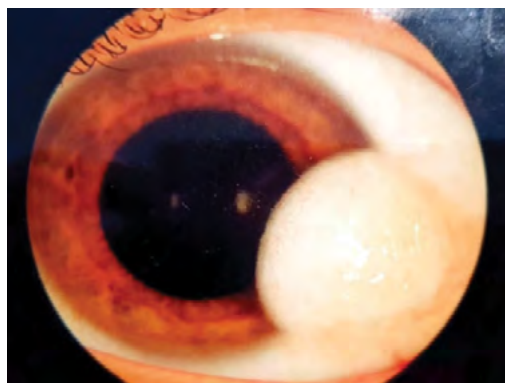


Fig. 3. Grade II corneal limbal dermoid in the left eye of the aforementioned 17-year-old male patient

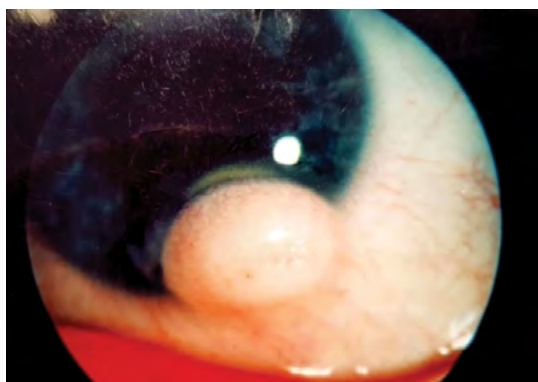


Fig. 4. Grade II corneal limbal dermoid in the left eye of a 24-year-old male patient

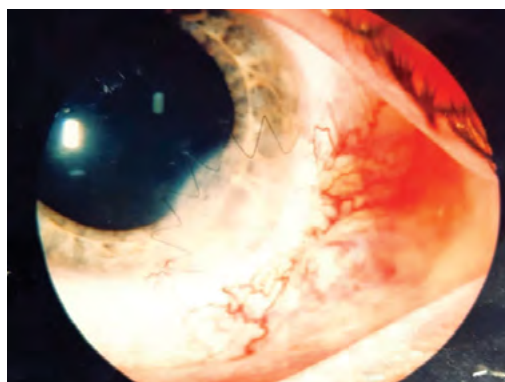


Fig. 5. The left eye of the aforementioned 24-year-old male patient two weeks after excision of the dermoid. The corneal portion of the corneoscleral graft is clear

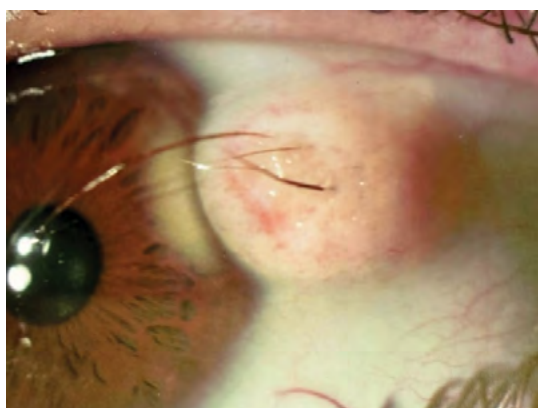


Fig. 6. Grade I limbal dermoid in the left eye of a 40-year-old male patient. Degenerative opacification of the cornea is seen



Fig. 7. The left eye of the aforementioned 40-year-old male patient two weeks after excision of the dermoid. The corneal graft is semitransparent

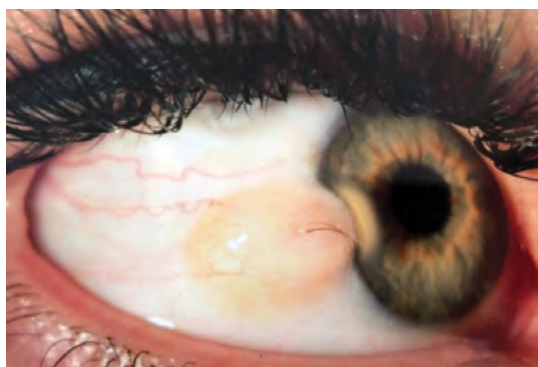


Fig. 8. Grade I limbal dermoid with a degenerative opacification of the peripheral cornea in the right eye of a 33-year-old female patient

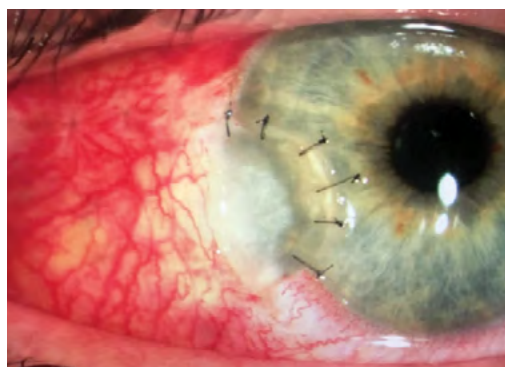


Fig. 9. The right eye of the aforementioned 33-year-old female patient 2 weeks after surgery. The corneal portion of the corneoscleral graft is clear