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Intrastromal keratopigmentation for photophobia secondary to traumatic aniridia

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ABSTRACT

Purpose: To present a case of therapeutic intrastromal keratopigmentation to resolve intractable photophobia secondary to traumatic aniridia in a hypotonus eye.

Observations: A 66-year-old male presented with intractable photophobia for several years in the left eye following a ruptured globe and multiple subsequent retinal surgeries for retinal detachments complicated by proliferative vitreoretinopathy. The patient underwent intrastromal keratopigmentation given surgical limitations due to the presence of hypotony and silicone oil dependence. The patient's symptoms were fully resolved, and the pigmentation remained stable at 18 months.

Conclusions/Importance: Keratopigmentation can be an effective surgical approach to managing patients with symptomatic photophobia in eyes where intraocular surgery is not an amenable option.

1. Introduction

Keratopigmentation, or corneal tattooing, has been historically used for cosmetic purposes by using various mixtures of chemicals and pigments; however, keratopigmentation also serves as a therapeutic option to address photophobia or monocular diplopia from peripheral iridotomies, aniridia, and other iris defects.[1,2] Keratopigmentation has demonstrated good cosmetic outcomes for disfigured eyes and stable pigment longevity.[3] We describe a case of a patient with intractable photophobia secondary to traumatic aniridia in an eye with silicone oil and hypotony who had resolution of his visual symptoms with manual intrastromal keratopigmentation.

2. Case report

The patient is a 66-year-old male with a history of ruptured globe in the left eye (OS) who presented with several years of intractable photophobia. His ruptured globe was complicated by uveal, iris and ciliary body loss, as well as a subsequent total retinal detachment with proliferative vitreoretinopathy status post multiple pars plana vitrectomies with silicone oil. Upon presentation, his visual acuity was hand motion OS. His intraocular pressure was 3 mmHg and there was an afferent pupillary defect OS. On slit lamp exam, he had early band keratopathy and no visible iris tissue or stump (Fig. 1A). Additionally, his intraocular lens (IOL) was dislocated and encased in fibrosis within the ciliary body. There was pallor to his optic nerve and his retina was flat posteriorly with silicone oil fill. The right eye was unremarkable. He was intolerant to contact lens. Given his hypotony, presence of silicone oil, inadequate capsular support in addition to the dislocated IOL, he was not a good surgical candidate for insertion of an intraocular prosthetic iris.

He subsequently underwent manual intrastromal keratopigmentation with stromal punctures to match the color and appearance of his fellow iris (Fig. 1C). A vacuum corneal trephine (Hessburg-Barron vacuum trephine, Jedmed Instrument Co., Oakville, MO) was used to create a 35–40% partial thickness trephination based on pachymetric data obtained by Scheimpflug tomography. A keratome was then used to dissect a lamellar pocket from the trephine and extended circumferentially towards the limbus. Various micropigmentation (Permark micropigmentation, PMT Corporation, Chanhassen, MN) were manually mixed in the operating room to match the iris stroma color to the contralateral eye (White: P200485, Navy Blue: P200301, Slate Blue: P200303, Charcoal Green: P200294, Green: P200296). A 30-gauge needle coated with white ink was then used to perform paracentral stromal punctures to create the appearance of Wofflin nodules. Artificial pupil sizing was carefully planned to allow vitreoretinal specialists to

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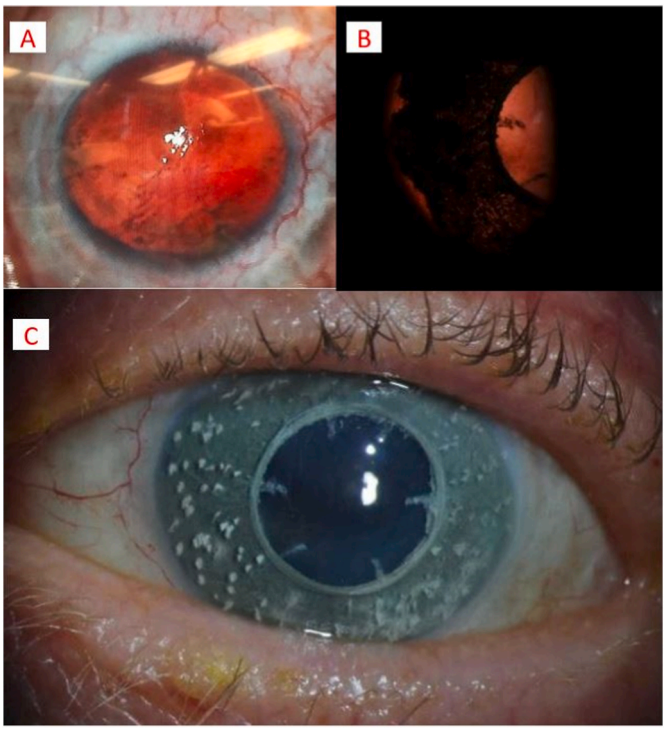

Fig. 1. External photograph montage of the left eye. (A) Fundus retroillumination revealing the absence of iris tissue and stump, and aphakia with moderate corneal edema. (B) Fundus retroillumination following intrastromal keratopigmentation demonstrating blocking of the peripheral to mid-peripheral cornea from pigmentation. (C) Direct, diffuse illumination showing color pigmentation to match the fellow eye, with white pigment from stromal punctures to mimic the iris texture. The artificial pupil was left a 6 mm to allow for visualization of the posterior segment pathology. (For interpretation of the references to color in this figure legend, the reader is referred to the Web version of this article.)

continue monitoring his posterior segment, as he required multi-specialty ophthalmic care. He endorsed significant improvement in his photophobia following the procedure. Fundus retroillumination demonstrated blocking of peripheral light consistent with his successful keratopigmentation (Fig. 1B). His visual acuity was stable at 18 months and there was no fading in his pigmentation.

3. Discussion

Keratopigmentation has a wide variety of uses for both cosmetic and functional repair in eyes with significant anterior segment defects.[1] Even in patients who have anatomically successful vitreoretinal surgery, they may continue to experience debilitating visual symptoms such as diplopia, dysphotopsia, or photophobia. Color contact lenses can be considered for initial management, but they carry a risk of infectious keratitis associated with contact lens wear.[4] Additionally, contact lens fitting may be difficult in patients with an irregular corneal surface secondary to scarring, especially in eyes with a history of trauma.

Intraocular iris prosthetics have been reported in management of aniridia and other iris defects.[5] Various models have been used with some success. Serious complications include uveitis-glaucoma-hyphema syndrome with early anterior chamber implants, with some studies reporting up to 52.3% of eyes diagnosed with secondary glaucoma with evidence of structural and functional impairment.[6,7] In this case, a compromised capsular bag precludes the possibility of implanting an endocapsular iris or capsular tension ring-based prosthetic iris. Prosthetic iris implantation also carry a risk of persistent ocular inflammation, hypotony, corneal decompensation, and retinal detachment.[8]

More invasive reconstructive procedures such as evisceration and enucleation can be considered alternatives, but carry higher prosthetic-related complications.[9] Our patient was not considered a good surgical candidate given his history of a fibrosed IOL within the ciliary body, presence of silicone oil, and persistent hypotony. It was felt that keratopigmentation would be the best option for this patient.

A five-year study by Kim et al. evaluated 147 eyes with corneal opacities and blind eyes and reported good cosmetic results, and had 17 eyes (12%) requiring retattooing, calcium plaque removal, or epithelial keratectomy.[3] Another study of 136 demonstrated that most patients are satisfied with the cosmetic outcome (98.5%), although 44.7% of the eyes with superficial keratopigmentation as a first-stage procedure required a subsequent operation to retouch the pigmentation.[10] Most complications include foreign body sensation, fading of pigmentation requiring a second operation, and epithelial ingrowth. There were no serious complications reported in either study.

Alio and authors have reported good success using a variety of keratopigmentation techniques, such an automated, manual, and using femtosecond laser.[11,12] A manual approach was preferred as docking a hypotonus eye would be challenging for a femtosecond laser approach. The presence of band keratopathy was limited to the limbus, so intrastromal dissection was carried out without complication from the trephine. Had the band keratopathy been more extensive or denser, it would be reasonable to consider pre-treatment with disodium ethylenediaminetetraacedic acid (EDTA) to allow for better visualization of the corneal pigment.

This case highlights that keratopigmentation is a good and effective alternative for therapeutic management in patients with photophobia secondary to iris defects related to trauma. While newer techniques such as superficial automated keratopigmentation and femtosecond-assisted keratopigmentation have successful outcomes, eyes that have suffered trauma and undergone extensive surgeries are often complex in presentation and require individualized surgical approaches. This case demonstrates that manual intrastromal keratopigmentation can still have excellent outcomes. Our patient's visual acuity remained stable and had no fading his pigmentation at 18 months. A subjective questionnaire at his follow up appointment revealed that he was satisfied with the outcome and had a significant improvement in his visual symptoms.

Consent

Written consent to publish this case has not been obtained. This report does not contain any personal identifying information.

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Research ethics

We further confirm that any aspect of the work covered in this manuscript that has involved human patients has been conducted with

the ethical approval of all relevant bodies and that such approvals are acknowledged within the manuscript.

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