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Keratopigmentation and retrobulbar filling with hyaluronic acid for the aesthetic treatment of an enophthalmic blind eye

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Abstract
Keratopigmentation (KTP) is a technique that can be used to change the color and cosmetically enhance corneal lesions. Ocular changes such as *leukoma*, phthisis bulbi, aniridia, heterochromia, and enophthalmos, among others, generate negative social stigma for patients. We report the case of a 25-year-old female patient with unilateral enophthalmic blind eye who underwent KTP associated with retrobulbar injection of hyaluronic acid. The chosen technique was Automated Superficial Keratopigmentation using an electronic device (Ultra Pen Electric Ink). Subsequently, retrobulbar injection of hyaluronic acid Saypha® Volume Plus was performed. Keratopigmentation presented the expected outcome, and the procedure proved safe without intraoperative and postoperative complications. Retrobulbar filling improved symmetry between the eyes and reduced the prominent eyelid sulcus. The patient was highly content with the final aesthetic result. Although not widespread, both techniques have proven to be relatively safe, less painful, and with satisfactory aesthetic results.

Keywords: Keratopigmentation, tattoing, cornea, hyaluronic acid, enophthalmos

Introduction
Ocular alterations, including *leukoma*, phthisis bulbi, aniridia, and heterochromia, generate negative social stigma in patients who frequently seek alternatives to hide these changes and reduce possible secondary psychological effects. In addition, when associated with facial asymmetries, as in the case of monocular patients with dysmorphic aesthetic involvement or enophthalmos, the associated negative psychosocial effects may be even more pronounced [1]. Keratopigmentation (KTP) is a method used to change the color and cosmetically improve corneal lesions. Among its various indications, the most common one found in the literature continues to be corneal opacity in eyes without visual prognoses, the main cause of which is ocular trauma [1-2].

Patients who experienced perforating eye trauma or those with phthisis bulbi frequently show decreased eyeball volume and enophthalmos, a condition involving anteroposterior displacement of the eyeball into the orbit [3]. This condition produces the appearance of a smaller, sunken eye and very flat or irregular corneal surface, making it difficult to fit cosmetic contact lenses and eye prostheses.

The objective of this study was to disseminate the techniques of keratopigmentation and retrobulbar hyaluronic acid gel filler injection, which are still little explored in the literature, as promising alternatives for the aesthetic improvement of monocular patients with enophthalmos and dysfunctional corneal aesthetics.

Case Report
A 25-year-old female patient sought a specialized ophthalmology service desiring aesthetic improvement in the right eye (OD). The patient showed interocular differences in terms of size and color; this difference appeared after an episode of uveitis four years ago that evolved with blindness. At the time, the possible causes of uveitis were investigated, albeit without etiological elucidation. The patient denied trauma or any external pathology and reported only ocular hyperemia and progressively reduced visual acuity, followed by darkening of the iris and appearance of ipsilateral cataracts. Additionally, the patient reported increased intraocular pressure (IOP) in the OD after the episode.

Topical use of dorzolamide hydrochloride 2% eye drops every 12 h, timolol maleate 0.5% every 12 h, and an oral dose of acetazolamide 250 mg every 8 h was initiated. The patient had no personal or familial complaints. The left eye (OS) did not show any changes.

Best visual acuity with no light perception in OD and 20/25 in OS. The IOP was 16 mmHg OD and 12 mmHg OS. Anterior biomicroscopy in OD revealed prominent eyelid sulcus of the tear trough, bulbar conjunctival hyperemia 1+/4+, transparent cornea, plateaued and hyperchromic iris in relation to the contralateral eye, superior temporal chorectopia, 360° anterior synechia with the presence of neovessels in the iris, and white cataract. (Figure 1A and B) Biomicroscopy performed in the OS with no alterations showed a honey-brown iris. Performing fundoscopy was impractical for OD due to opacity of the lens; however, fundoscopy revealed the absence of alterations in OS. Ultrasonography of the OD with evident excavation and anteroposterior diameter slightly decreased in relation to the OS without alterations.

Keratopigmentation associated with retrobulbar injection of hyaluronic acid under sedation and retrobulbar block was indicated. We opted for the Automated Surface Keratopigmentation technique, which uses an electronic device (Ultra Pen Electric Ink® rotary tattoo machine) to perform superficial micropunctures in the stroma in an automated manner (after complete removal of the epithelium) using cartridges with one and three tip needles. The colors were added by matching the appearance of the contralateral eye to achieve the appropriate shade of the iris (Electric Ink® moss green, dark skin, and Easy Glow® monkey brown tattoo inks were used), and the pupil was made using a circle (Diameter, 3 mm) with the color Electric Ink® Black Line. (Figure 2A and B)

Finally, retrobulbar injection was performed with 2 mL of hyaluronic acid Saypha® Volume Plus, using a technique similar to that described by Sung *et al*. (2020) [4-6], through the inferotemporal transcutaneous introduction of a 25-gauge needle into the intraconal space, slowly and cautiously observing the anterior projection of the eyeball. (Figure 2C)

After two months, the patient presented with excellent corneal coloration, but she presented with a prominent and persistent lower eyelid pocket, suggesting displacement or excessive injection of retrobulbar hyaluronic acid. (Figure 3A) 1 mL of hyaluronidase (300 IU) was then applied superficially, directly to the temporal bulging region. Two days after the procedure, significant improvement was observed, but a small remaining lower nasal bag persisted. Therefore, a further 0.1 mL (30 IU) of the same solution was applied, with resolution of the condition and an excellent final aesthetic result. (Figure 3C)

Discussion

This case report describes the treatment approach for a patient with enophthalmia due to phthisis bulbi, unilateral hyperchromia of the iris associated with irreversible blindness, and total white cataract. The patient had no visual prognosis for lens extraction, and facectomy for aesthetic reasons was contraindicated because of the increased risk of evolving or accelerating the process of phthisis bulbi [7].

Keratopigmentation involves corneal stromal implantation of micropigments of different colors to change the coloration. Currently, the four most widely used techniques are superficial and stromal techniques. Superficial techniques can be classified into Manual and Automated (involving the use of a rotary tattoo machine). Furthermore, intrastromal techniques include Manual Intrastromal Keratopigmentation, which involves manual dissection of a stromal tunnel for pigment implantation, and Femtosecond Laser-assisted Intrastromal Keratopigmentation [1, 2, 8].

In blind eyes with a clear cornea, manual intrastromal keratopigmentation should be the first choice because it is less aggressive to the ocular surface, and through this technique, the epithelium is preserved and only a few incisions are made for lamellar dissection of the cornea. However, this technique limits tonal variation because only one color is injected into the stroma. Therefore, for the iris to be reproduced more reliably, we opted for the Automated Superficial KTP technique to enable the gradual application of different colors and a final result that was very close to the adelphic eye.

To perform the automated superficial technique, total de-epithelialization is recommended at the beginning of the procedure, ensuring that the pigment is introduced into the stroma and not into the epithelium. However, this results in a more painful postoperative period and longer epithelialization time, although it may present better aesthetic results.

The use of dermal fillers in the periocular/retrobulbar area is becoming more common for both functional and aesthetic indications. The balance between the volume and content of the orbital soft tissue determines the position of the globe in the orbit. A disturbance of this balance creates asymmetry and enophthalmos, which can be challenging for the surgeon to correct [4-6]. The use of hyaluronic acid for this purpose offers an alternative to some surgical procedures. However, its success depends on the judicious selection of patients, products, and techniques to achieve favorable outcomes.

Hyaluronic acid use in soft tissues was approved by the U.S. FDA in 2003 [9] and it has become an important and emerging tool used by oculofacial plastic surgeons. Synthetic hyaluronic acid achieves its expected function when stabilized after cross-linked synthesis, which alters its half-life and viscosity and can also vary the uniformity, particle size, and concentration [10].

One of the main complications of retrobulbar filling with hyaluronic acid is hematoma, which usually occurs immediately after the direct puncture of vessels. The risk of hematoma formation can be minimized by avoiding antithrombotic agents before treatment and by using larger caliber cannulas, including 25G, which reduce the chance of perforating vessel walls, compared with needles or cannulas with smaller diameters [10].

Notably, in the case described, a possible displacement of the product or possible hypercorrection may have occurred because hyaluronic acid was applied after keratopigmentation performed under retrobulbar anesthesia. This may have caused the surgeon to lose the parameters at the time of the procedure, or even the retrobulbar anesthetic had “opened the way” for possible displacement of the product. Ideally, this procedure should be performed in combination with KTP under general anesthesia or a second time after pigmentation.

Vascular occlusions and consequent vision loss are among the most serious complications of facial filling, with an estimated incidence of up to three per 1,000 injections. Between 1906 and 2019, more than 190 cases of blindness caused by aesthetic injectable treatments were reported in the literature. Most of these cases occur as a result of autologous injection of fat and retrograde embolism into the ophthalmic and central arteries of the retina [10].

Hyaluronidase can rapidly break down hyaluronic acid in the tissue and has been reported to diffuse through the walls

of some vessels, and it can reverse skin ischemia in the region affected by embolism or compression. However, recent studies have reported a lack of penetration through the arterial walls and optic nerve sheath, thereby casting doubt on the role of retrobulbar hyaluronidase in the treatment of vision loss caused by embolism with hyaluronic acid injection [10]. To the best of our knowledge, no study has reported contralateral blindness caused by retrobulbar injection of hyaluronic acid in already-blind eyes.

In the present case, despite a slight hypercorrection that was resolved with hyaluronidase injection, the patient was very satisfied with the final aesthetic result. Keratopigmentation presented the expected result, and retrobulbar filling improved symmetry between the eyes, reducing the prominent eyelid sulcus. Although complications can occur, ophthalmologists are among the best prepared professionals to perform this procedure; however, the risks and benefits should be widely discussed beforehand. Although not widespread, both techniques have proven to be relatively safe, less painful, and with satisfactory aesthetic results. Further studies are warranted to better evaluate the behavior of retrobulbar hyaluronic acid and its possible complications.

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Fig 1: Ectoscopy with different light incidences showing: A) Heterochromia with iris hyperchromia in the OD; B) Prominent tear trough palpebral sulcus and enophthalmos in the OD

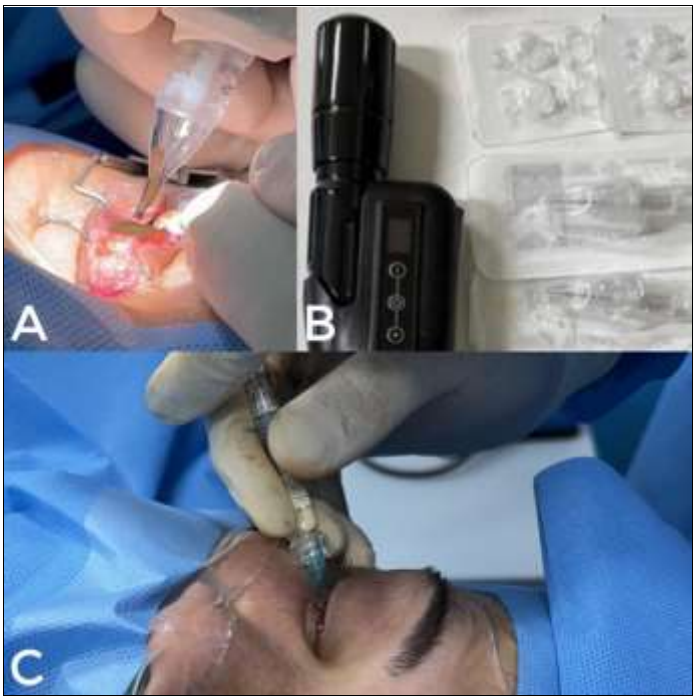

Fig 2: A) Superficial Automated KTP in progress; B) Dermograph and cartridges; C) Hyaluronic acid retrobulbar filling in OD

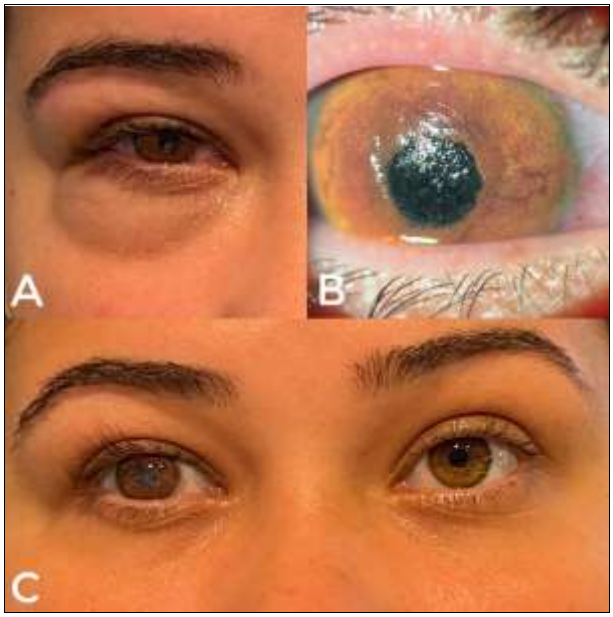

Fig 3: A) Ectoscopy with prominent lower eyelid pocket, suggesting displacement or excessive injection of the retrobulbar hyaluronic acid after two months; B) Slit lamp final aspect; C) Final ectoscopy after retrobulbar hyaluronic acid filling

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