Case report

The Osteo-odonto-keratoprosthesis to restore vision after severe dog bite injury

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Received 5 June 2018; revised 2 September 2018; accepted 10 September 2018
Available online 29 September 2018

Abstract

Purpose: To present our experience in Osteo-odonto-keratoprosthesis (OOKP) as the only option to restore vision after severe ocular dog bite injuries.
Methods: We describe our results in OOKP performed in two patients with previous severe dog bite facial injuries that required exenteration of one eye and facial reconstruction.
Results: Both cases achieved initially successful anatomical and visual outcome; however, one case suffered retinal detachment after one year. Buccal mucosa (BM) ulcerations and tilted lamina were present as a result of extraocular muscles loss, and both were difficult to treat.
Conclusion: OOKP has the capability to restore vision in the most challenging cases of ocular trauma; however, refractory BM ulcerations and tilted lamina could be expected as a result of an ischemic ocular surface and orbital pulleys loss.

Keywords: Osteo-odonto-keratoprosthesis; Keratoprosthesis; Dog bite ocular injury

Introduction

Of the 44000 facial dog bites that present to emergency in the United States each year, periorbital and orbital injuries occur in only 4%—8% of cases and are more frequently in children.1 Dog bite ocular trauma is mostly limited to eyelids and lacrimal apparatus, but cases of extraocular muscles transaction, corneoscleral laceration, or even eye globe rupture have been reported.2,3 Although the eye globe might not be seriously affected by the dog bite, the loss of the adnexal architecture implicates devastating consequences for protection and maintenance of a wet ocular surface.4 In such cases, despite eyelid repairing procedures, the absence of blink mechanisms and normal lid margins cause dryness, scarring, and chronic inflammation resulting in a hostile environment to maintain a transparent cornea.5 Progression to corneal cloudiness is inevitable, and any type of keratoplasty will result in failure under these circumstances. When end-stage corneal surface failure develops, keratoprosthesis (KPro) is the only option for visual restoration. However, the most commonly employed synthetic devices like Boston KPro are contra-indicated in a dry surface with adnexal anomalies, and only biological KPros can provide long-term visual restoration.6,7
Osteo-odontokeratoprosthesis (OOKP) is a biological and the most enduring type of KPro that offers visual rehabilitation in end-stage ocular surface diseases with severe dryness. The procedure is performed in two stages: in stage 1, an alveo-dental lamina is made from a single rooted tooth (usually the canine) which harbors an optical cylinder on its center. The lamina is then implanted under the orbicularis of the contralateral lower eyelid for nourishment and formation of soft tissue surrounding the bone. At the same sitting, the ocular surface is prepared and buccal mucous membrane is grafted onto the eye. During this stage (usually 2–4 months later), the lamina is retrieved, the mucosa is lifted off the globe followed by removal of the central cornea, iris, lens and anterior vitreous, and the lamina is then sutured to the sclera, which is finally covered by the buccal mucosa (BM).

In this study, we report two cases of dog bite injuries with severe bilateral adnexal and ocular damage in whom OOKP was the only option for visual restoration.

Case report

Two cases that received OOKP surgery at the Sussex Eye Hospital, Brighton, UK are reported. Institutional review board approval and informed patient consent was obtained. The study followed the tenets of the Declaration of Helsinki.

Case 1

A 51-year-old woman with history of injury in the upper face and both eyes caused by her own pet dog was referred for OOKP to restore her left eyesight. She was severely injured five years before her referral while she was home alone and laid unconscious in the floor due to alcohol intoxication. The dog chewed her upper face and periocular areas including her four eyelids, orbital tissues, and her right eye globe. A right exenteration was performed, and she then underwent sequential procedures for facial and eyelid reconstruction using fasciocutaneous flaps and BM grafts. Despite facial and eyelidrepair, her ocular surface developed progressive scarring due to dryness and exposure. On initial examination, her left eye vision was counting fingers. She underwent OOKP surgery, and stage 1 was particularly challenging due to extensive ocular surface scarring. The BM graft had to be sutured mostly to the sclera as we only identified the lateral rectus muscle inserted. Stage 2 was carried out successfully 3 months later. Postoperatively, she achieved a best corrected visual acuity (BCVA) at three months of 20/60 despite inferior tilt of the lamina and optic cylinder. Unfortunately, one year later she developed a total retinal detachment that could not be repaired despite pars plana vitrectomy and silicone oil injection. Her vision decreased then to light perception. In addition, she suffered refractory BM graft ulceration with bone laminar exposure that did not resolve with repeated BM and hard palate mucoperiosteal grafts. She required a facial advancement flap at the end (Fig. 1).

Case 2

A 58-year-old man was referred for left eye OOKP. He suffered severe dog bite facial trauma eight years ago when he was unconscious due to alcohol toxicity. He lost all four eyelids, bilateral orbital tissues, and his right eye globe. He also required a total right exenteration. Similar to our first case, he underwent facial and eyelid reconstruction procedures. However, anatomical repair did not provide the functioning blink mechanism for a normal tear flow. He developed progressive scarring of the ocular surface and suffered repeated episodes of bacterial keratitis. A cataract also developed later which was treated with phacoemulsification, but his vision continues to decrease due to progressive corneal scarring. Our initial examination revealed visual acuity of hand movements and an opaque cornea with central thinning. Stage 1 OOKP was successfully followed by stage 2 three months later. During stage 1, the superior rectus muscle could not be identified. He achieved a BCVA of 20/80 three months after surgery improving to 20/30 at six months. Despite successful OOKP, his lamina and optic cylinder showed progressive tilting downward as a result of rotation of the eye globe. Re-suturing of tilted lamina was carried out 3 years after surgery, but the beneficial results were short lived. He also developed BM ulceration with bone laminar exposure that was successfully repaired by advancement and rotational BM flaps, a fresh BM graft and medial tarsorrhaphy. His vision with spectacles remains 20/30 four years after OOKP surgery (Fig. 2).

Discussion

Dog bite facial injuries can cause permanent damage to the adnexal tissues. These injuries are usually unilateral and typically affect the lid margins, canaliculi, lacrimal gland, levator muscle, and trochlea. The eye globe itself is rarely affected by the canine bite possibly due to the blink reflex that spares the globe from direct trauma. However, there are reports in the ophthalmic literature of corneoscleral lacerations, eye globe rupture, or even cataract formation in patients who suffered dog bites.

Both of our patients suffered severe facial injuries with bilateral ocular involvement that occurred under depressed consciousness. Bergmann et al. reported one similar case of a patient that suffered “degloving” bite injury of the upper face and one orbit when she was unconscious after attempting suicide. Under these circumstances, the damage of the facial tissues by dog bites can be devastating and can potentially result in bilateral blindness.

Our cases were exceptional due to the challenging surgical repair of the complex lesions that included the loss of one eye globe, four eyelids, orbital pulleys, conjunctiva, and extraocular muscles. Both cases underwent adnexal repair with fasciocutaneous flaps and BM replacing the bulbar and tarsal conjunctiva. Despite good anatomical results, these procedures cannot restore the lacrimal system and cannot provide the complex natural function of eyelid blinking. In the long run,
both resulted in severe dryness and corneal scarring. In this scenario, the hostile environment of the ocular surface carries a poor prognosis for corneal transplantation or even Boston KPro Type 1.\(^6,^9\) Having only one eye, both cases were referred to our center because only biological KPro could provide long-term anatomical and visual success.

To our knowledge, these are the only reported cases of OOKP after bilateral dog bite ocular trauma. OOKP was challenging in both cases due to the demanding surgery in eyes with previous multiple adnexal and ocular surface procedures. The absence of extraocular rectus muscles represented a drawback for the BM graft as they provide vascular supply and reduce the risk of ulcerations.\(^8,^9\) Both of our cases developed BM ulcerations being more severe in our first patient in whom we only identified the lateral rectus. BM graft or hard palate periosteal grafts are usually successful to treat this complication, but as in our first case, it may not suffice in an ischemic eye surface. With no other options, our first patient required skin facial flaps to cover the entire eye to prevent laminar bone resorption and endophthalmitis.

We found that downwards tilt of the lamina and cylinder was a particular complication in both cases. This was also difficult to correct and resulted from the rotation of the eye globe secondary to loss of orbital tissues and the superior rectus. Both patients, however, adopted a compensatory head posture to achieve a better field of vision. Despite initial successful visual outcomes, our first patient developed a total retinal detachment. This complication is known to have a poor prognosis in OOKP due to the challenges for surgical repair. The main problems are the limited visualization through a 4 mm diameter optical cylinder (as in our case) and the need of laminar removal for a temporary KPro that carries even more risks of complications.\(^11\) Endoscopic vitrectomy has been suggested as an option for vitreoretinal complications in OOKP, but has the limitations of depth perception and that is not widely available.\(^12\)
In summary, we present our experience in two cases with an only eye and previous dog bite facial injuries occurred under similar circumstances. In such complex injuries, facial and eyelid anatomic repair can be achieved with different surgical approaches; however, blink efficiency cannot be restored, and these cases will progress to corneal scarring. Although it is known the capability of the OOKP to restore vision in the most challenging cases of ocular trauma, refractory BM ulcerations and tilted lamina and optic cylinder could be expected in patients that suffered loss of the orbital pulleys and extraocular muscles.

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