CASE REPORT

Successful simple limbal epithelial transplantation (SLET) in lime injury-induced limbal stem cell deficiency with ocular surface granuloma

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SUMMARY

A 9-year-old boy presented with unilateral, total limbal stem cell deficiency (LSCD) complicated by the presence of a large fibro-vascular ocular surface mass lesion secondary to accidental lime injury. The pathological tissue covering the cornea was excised and simple limbal epithelial transplantation (SLET) was performed using autologous limbal tissue from the fellow eye. Histopathology of the excised ocular surface tissue revealed exuberant granulation tissue interspersed with retained calcium particles. At 6 weeks postoperatively, a focal recurrence of LSCD with symblepharon and forniceal shortening was noted superiorly. This was successfully managed by performing conjunctival autografting along with supplemental SLET. The unaided vision had improved from light perception at presentation to 20/40 at 6 months postoperatively. The fornices were deep and the corneal surface was avascular, epithelised and stable. This case demonstrates the efficacy of SLET in a child with severe ocular burns, highlighting the role of supplementary procedures customised to treat focal recurrences of LSCD.

BACKGROUND

Simple limbal epithelial transplantation (SLET) is a novel surgical technique of limbal stem cell transplantation that requires very little donor tissue and avoids the need for sophisticated laboratory facilities.¹ However, the outcomes of SLET in severe cases of ocular surface burns and in children have not been described previously. It is known that unlike adults, children tend to have more severe ocular surface damage following injury, and greater recurrence of limbal stem cell deficiency (LSCD) with poorer visual recovery following ex vivo cultivated limbal epithelial transplantation (CLET).^{2 3} This case describes the use of SLET to treat a severe case of ocular burn induced LSCD in a 9-year-old child, highlighting the importance of customising supplementary surgery to achieve excellent anatomical and functional outcomes.

CASE PRESENTATION

The father of a 9-year-old boy brought his son to our clinic with visual loss, drooping of the eyelid and a protruding red mass in the child's left eye. These symptoms appeared following accidental injury to the same eye with edible lime (calcium hydroxide) 6 months prior to presentation. On examination, the right eye was essentially normal with a visual acuity of 20/20. Visual acuity in the

left eye was light perception with accurate projection of rays, and intraocular pressure was perceived to be normal on palpation. There was forniceal shortening owing to extensive symblephara extending from the tarsal conjunctiva to the cornea superiorly and the bulbar conjunctiva inferiorly (figure 1A). The limbus was not discernable and the corneal surface was covered with a pannus indistinguishable from the surrounding conjunctiva. A vascular pendunculated mass attached to the corneal surface, which stained positively with fluorescein but negatively with Rose Bengal, protruded through the palpebral aperture (figure 1B).

INVESTIGATIONS

The ocular surface pathology obscured viewing of the intraocular structures and an ultrasound B scan was performed. The B scan revealed that the left eye was phakic with a normal posterior segment.

DIFFERENTIAL DIAGNOSIS

Based on the history and clinical features a diagnosis of unilateral LSCD with an ocular surface, pyogenic or foreign body granuloma following lime injury was made. Other possible causes of such a lesion, namely anterior staphyloma, ocular surface neoplasia or limbal choristoma seemed unlikely.

TREATMENT

The ocular surface pannus along with the mass in the left eye were excised en masse and sent for histopathological examination. SLET was performed using one-clock hour of donor limbal tissue from the right eye. The 3 mm limbal strip was divided into small pieces and fixed to the ocular surface with fibrin glue over an amniotic membrane graft. Histopathology of the excised tissue showed stratified columnar epithelium with goblet cells, dilated hyperaemic vessels with perivascular infiltrate and focally oedematous fibro-collagenous stroma (figure 1C). Histopathology of the mass lesion showed granulation tissue with surface necrosis, proliferating blood vessels, oedematous stroma and mixed inflammatory infiltrates (figure 1D) with focal areas of amphophilic deposits suggestive of retained calcium (figure 1E). The patient was prescribed tapering doses of topical prednisolone acetate 1% eye drops and followed by on day 1, day 7 and 6 weeks postoperatively. At the 6 weeks visit the ocular surface in the child's left eye had re-epithelised completely, the cornea had cleared significantly and the vision had improved to 20/50

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Figure 1 (A) Left eye showing conjunctivalised corneal surface with symblephara superiorly and inferiorly, (B) vascular pendunculated mass attached to the corneal surface, stained with fluorescein, (C) H&E stain ($\times 40$) showing stratified columnar epithelium with goblet cells, dilated hyperaemic vessels (arrows) with perivascular infiltrate and focally oedematous fibro-collagenous stroma, (D) H&E stain ($40\times$) showing granulation tissue with surface necrosis, proliferating blood vessels, oedematous stroma and mixed inflammatory infiltrates and (E) H&E stain ($\times 100$) showing focal areas of amphophilic deposits (arrows) suggestive of retained calcium.

(figure 2A). However, the superior fornix was obliterated with symplepharon reaching up to the clear corneal margin (figure 2B). This was deemed to be a focal recurrence of LSCD. The symblepharon was released and the subepithelial fibrotic tissue was excised. The size of the bare area created on the bulbar surface was measured and a similar sized conjunctival autograft was harvested from the other eye. This autograft that contained a tiny tongue of limbal tissue centrally was glued in its anatomical location in the left eye (video 1). Postoperatively the patient received tapering doses of prednisolone acetate 1% eye drops in both eyes for 1 month and moxifloxacin 0.5% eye drops in both eyes for 1 week.

OUTCOME AND FOLLOW-UP

One month after symblepharon release with supplemental SLET in the left eye the superior fornix was deep (figure 2C). The entire cornea was avascular and epithelised with an unaided visual acuity of 20/40. The donor eye was unharmed and the donor sites had completely epithelised (figure 2D). The steroid

eye drops were discontinued and the patient was asked to use only topical lubricants (carboxymethylcellulose 0.5% eye drops) 2–3 times daily in both eyes. Six months after the last procedure and in the subsequent follow-up visits the child continued to maintain a stable, epithelised and avascular corneal surface in the left eye without any sign of recurrence of LSCD or symblepharon (figure 2E).

DISCUSSION

Edible lime (chuna) injuries in children have been known to cause severe ocular surface burns, and have been reported to have a poor visual prognosis.⁴ This case, to us, represents the severest form of LSCD owing to chemical injury, with extensive symblepharon formation, a completely conjunctivalised surface and no view of the cornea. Of particular interest is the presence of an ocular surface granuloma, later determined to contain residual calcium. This is a rare finding, reported earlier by our group in only 4 out of 29 cases of LSCD caused by chemical injury.⁵ For more than a decade, we have been using autologous

Figure 2 (A) Left eye at 6 weeks postoperatively showing completely epithelised surface with improved corneal clarity, (B) left eye at 6 weeks showing obliterated superior fornix with symblepharon encroaching upon clear cornea, (C) left eye 1 month post 'top-up' SLET showing epithelised and clear cornea with no recurrence of symblepharon, (D) right eye at last follow-up visit showing healthy donor site and (E) left eye at last follow-up visit.

Video 1 Symblepharon release and excision of sub-epithelial fibrotic tissue in the left eye, followed by harvesting of conjunctival autograft with a tiny tongue of limbal tissue in the centre from the right eye, which is subsequently glued into position in the left eye.

CLET for managing cases of total LSCD with a considerable degree of success. However, CLET in paediatric patients is less successful than in adults, with more recurrences.^{2,3} Our group has recently described results of SLET, a technique that utilises minimal limbal tissue from the donor site, akin to CLET.¹ Unlike CLET, however, SLET is a single-stage procedure that obviates the need for expensive ex vivo cultivation of cells. This case illustrates the fact that SLET can be successfully utilised to restore a stable corneal surface with consequent improvement in visual acuity in the face of the severest manifestations of total LSCD. Postoperative events also emphasise the need for rigorous follow-up and meticulous examination for early recurrence of LSCD. It can be argued that focal recurrences, such as in this case, may be observed without intervention as long as the visual acuity remains unaffected. However, the safety and efficacy of a repeat autologous stem cell transplantation procedure have been established.⁶ With SLET it is possible to tailor the amount of limbal tissue taken from the donor site according to the extent of LSCD recurrence. This, coupled with the minimal costs and simplicity of SLET, encouraged us to intervene early. Conceivably, these advantages may make the prospect of repeat, 'top-up' SLET for addressing focal recurrences of LSCD more attractive than repeat CLET. Further experience with more cases

and longer follow-up would help in delineating the role of this novel technique in similar situations.

Learning points

- ▶ Edible lime (chuna) injury can lead to severe ocular surface burns, manifesting as total limbal stem cell deficiency (LSCD) and granuloma with retained calcium particles.
- ▶ Simple limbal epithelial transplantation (SLET) is a novel, single-stage technique that utilises minimal donor limbal tissue. It does not require expensive ex vivo cultivation of cells and can successfully restore the corneal surface, leading to improvement in visual acuity.
- ▶ SLET can be used in paediatric cases of total LSCD secondary to severe ocular surface burns with good outcomes.
- ▶ Careful clinical examination and rigorous follow-up are essential to detect early, focal recurrence of LSCD, which can be managed using supplementary procedures such as targeted, 'top-up' SLET.

Contributors All three authors have contributed in conceptualising and drafting the manuscript and finally approving it.

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