CASE REPORT

Customised simple limbal epithelial transplantation for recurrent limbal stem cell deficiency

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SUMMARY

A 4-year-old boy developed unilateral, total limbal stem cell deficiency (LSCD) following an acid injury. A stable ocular surface was achieved with autologous simple limbal epithelial transplantation (SLET), performed 4 months after the injury. Focal areas of conjunctivalisation were noted in the postoperative period. These were addressed using a novel modification of SLET. A very small piece of limbus from the healthy eye was split into multiple pieces, and applied to the bare corneal stroma with fibrin glue after pannus resection. The surface was covered with amniotic membrane. This resulted in a stable surface, and visual acuity improved to 20/50. The donor remained healthy despite two biopsies being harvested. This case demonstrates that early ocular surface reconstruction may be considered in children with LSCD if amblyopia is a concern. Customised SLET allows surgeons to tackle focal recurrences of LSCD effectively, using minimal limbal tissue from the donor site.

BACKGROUND

Ocular surface reconstruction is generally carried out at least 6 months after the original insult. Simple limbal epithelial transplantation (SLET) is a novel surgical technique that combines the benefits of direct and cultivated limbal stem cell transplantation for limbal stem cell deficiency (LSCD). We present a case that demonstrates the need for early intervention if amblyopia is a concern. A novel modification of SLET is also presented to highlight how it can be customised to treat focal recurrences of LSCD.

CASE PRESENTATION

A 4-year-old boy presented with a gradual decrease in vision over a period of 4 months and inability to open the left eye following accidental exposure to toilet cleaner liquid containing hydrochloric acid. He had earlier undergone amniotic membrane transplantation twice (once on the day following chemical injury, and again 2 weeks later for a persistent epithelial defect) in the left eye following the acute chemical injury. Visual acuity was 20/20 in the right eye and light perception in the left eye. The right eye was unremarkable on examination. In the left eye, the entire surface was covered by a thick, fleshy looking fibrovascular pannus, with symblepharon formation superiorly (figure 1A). There was no view of the cornea, limbus or any intraocular structures.

INVESTIGATIONS

An ultrasound B scan showed no obvious abnormality of the posterior segment in the left eye.

DIFFERENTIAL DIAGNOSIS

Based on the characteristic history and clinical features, a diagnosis of total LSCD in the left eye was reached.

TREATMENT

The favoured option for surgical reconstruction in an eye with unilateral total LSCD is an autologous limbal stem cell transplantation procedure. Techniques have evolved from conjunctival-limbal autografts to cultivated limbal epithelial transplantation and, recently, SLET. We generally prefer to wait for at least 6 months after acute chemical injury, prior to undertaking a reconstructive procedure. In this particular case, considering the age of the patient and the potential risk of development of deep amblyopia, we decided to make an exception and intervene early. Autologous SLET combined with conjunctival autografting was performed utilising <2 o'clock hours of limbal tissue from the right eye. The surgical technique

Figure 1 (A) Left eye with total limbal stem cell deficiency. (B) Same eye, post simple limbal epithelial transplantation.

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Figure 2 (A and B) Left eye with recurrent symblepharon and focal recurrences of limbal stem cell deficiency.

has been described earlier.¹ In brief, a small limbal biopsy and conjunctiva were taken from the healthy eye. Conjunctival autografts were applied after symblepharon release on the superior and inferior bulbar parts of the left eye with fibrin glue. Following resection of fibrovascular pannus from the affected eye, the limbal tissue was cut into multiple small pieces and spread over an amniotic membrane applied over the corneal stromal bed. Fibrin glue adhesive and bandage contact lens were

applied. Postoperative medication included antibiotic eye drops and a topical steroid, tapered over a 6-week period.

OUTCOME AND FOLLOW-UP

This procedure resulted in a stable ocular surface, and visual acuity improved to 20/160 with glasses (-2.50 DC $\times 180^\circ$) (figure 1B). Patching was started for the right eye, and the child was on regular follow-up.

Over the next 5 months, focal recurrences of conjunctivalisation on the cornea as well as recurrence of symblepharon were observed (figure 2A, B). Options at this stage would be either to wait until the visual axis was threatened by the encroaching conjunctivalisation, or to intervene. Conventional techniques of limbal transplantation require a considerable amount of limbal tissue. However, SLET allows us to tailor the amount of limbal tissue harvested according to the extent of area with LSCD to be treated. This encouraged us to treat the recurrent LSCD, rather than wait.

To address the focal recurrences of LSCD, we decided to slightly modify the originally described technique of SLET (figure 3A–D). A limbal biopsy of <1 o'clock hour in size was harvested from the right eye, carefully avoiding the area of the first biopsy. In the left eye, fibrovascular pannus in the sectors of recurrent LSCD was resected, leaving the central corneal epithelium intact. Fibrin glue was placed over the bare areas, and small pieces of limbal tissue were placed over this. A large piece of amniotic membrane was sutured over the entire ocular surface as a bandage.

Five months postoperatively, the ocular surface remains stable, and visual acuity has improved to 20/50 (figure 4A). Despite limbal tissue being harvested twice, the donor site in the right eye remains healthy (figure 4B).

DISCUSSION

We have previously reported success using SLET in a similar case with ocular surface granuloma in a child.² Contrary to this, success rates of cultivated limbal stem cell transplantation in

Figure 3 Technique of customised simple limbal epithelial transplantation. (A) Eye with focal areas of limbal stem cell deficiency. (B) Pannus removed from surface. (C) Small pieces of limbal tissue placed on bare areas with fibrin glue. (D) Entire surface covered with amniotic membrane.

Figure 4 (A) Stable surface in the left eye, post customised simple limbal epithelial transplantation. (B) Healthy limbus in the right eye, even after two surgeries.

children are poorer, compared to adults.³ This case demonstrates that a stable ocular surface, with significant improvement in visual acuity, can be achieved using SLET even in cases with severe LSCD secondary to ocular surface burns. In our experience, amniotic membrane application in the acute phase makes it easier to find a good plane for removal of pannus during ocular surface reconstruction.

Another important takeaway from this case is the timing of surgery. In situations where amblyopia is a concern, it may be prudent to attempt ocular surface reconstruction early, rather than late. Delay in clearing the visual axis may result in irreversible amblyopia, whereas limbal transplantation using current techniques may succeed even when performed relatively early after the acute insult, as demonstrated by this case.

Our modification of SLET, which we call customised SLET, provides another surgical option for managing focal recurrences

of LSCD, with a few salient advantages. Unlike in repeat cultivated limbal epithelial transplantation, the healthy central corneal epithelium is not disturbed in customised SLET. Targeted placement of limbal tissue on the affected areas means a very small limbal biopsy is required, minimising the chances of iatrogenic donor site LSCD. Using amniotic membrane as a bandage rather than substrate for growing cells obviates the need for a bandage contact lens, which children often find difficult to maintain. In summary, customised SLET allows early management of recurrent LSCD without compromising the donor limbus.

Learning points

- ▶ Early ocular surface reconstruction may be considered in children with limbal stem cell deficiency (LSCD), to prevent irreversible amblyopia.
- ▶ Targeted placement of limbal tissue on corneal stroma after resection of pannus in cases with focal recurrences of LSCD is successful in restoring a stable ocular surface, without the need of amniotic membrane as a scaffold.
- ▶ Customised simple limbal epithelial transplantation for recurrent LSCD requires a minimal amount of limbal tissue, preserves normal corneal epithelium, is easy to perform, does not need specialised facilities for ex-vivo expansion of cells and may encourage surgeons to intervene earlier in such cases.

Contributors JV was involved in the drafting of the manuscript, revisions and final approval. IL was involved in the acquisition of patient images, revisions and final approval. VS was involved in the revision of the manuscript and final approval.

Competing interests None declared.

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