Case Report/Series

Split cornea transplantation in anterior lamellar keratoplasty for limbal dermoid surgery: a case report

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ABSTRACT
Limbal dermoid is a rare congenital lesion that can impair vision and raise aesthetic concerns. Surgery is frequently required to reduce discomfort and enhance visual outcomes. A 20-year-old woman presented with a limbal dermoid measuring 4.5 mm in diameter and half the depth of the stroma. Excision was performed with anterior lamellar keratoplasty (ALK) using a post-Descemet's membrane endothelial keratoplasty graft, which resulted in signs of graft failure. Re-surgery was then performed with post-Descemet's stripping endothelial keratoplasty graft. It yielded a clear graft with good visual acuity. The first corneal graft utilized 95% of the graft thickness to cover 55% of the defect, leading to poor host-donor apposition. The second graft employed 55–65% to cover the same portion of the defect. The proportional thickness of the graft is crucial for a successful ALK. Split cornea transplantation produces respectable results; however, the corneal thickness must be carefully considered.

KEYWORDS cornea transplantation, dermoid cyst, host-graft reaction, keratoplasty, lamellar

Limbal dermoid tumors are benign congenital tumors of the neurological tissues, connective tissues, skin, fat, sweat glands, and lacrimal glands. It is most frequently observed in the inferotemporal region, partially across the cornea and sclera. The estimated incidence of limbal dermoid worldwide is 1 to 3 per 10,000.[1] A retrospective study in China in 2017 showed that the prevalence of limbal dermoid was about 0.177 per million people within 10 years.[2]

Anatomically, limbal dermoid is divided into three grades based on size and depth of involvement.[3] Grade I limbal dermoid is a superficial lesion on the limbus less than 5 mm in size, while grade II limbal dermoid is a larger lesion that involves most of the cornea and extends deep into the stroma without affecting Descemet's membrane. Larger lesions covering the entire cornea and extending through the structures between the anterior surface of the eyeball and the pigmented epithelium of the iris are classified as grade III limbal dermoid.[4] Deciding which technique to apply should involve an imaging study to measure lesion depth. Simple lesion excision, lamellar keratoplasty, penetrating keratoplasty, lamellar

keratoplasty with a full-thickness corneal graft, excision with a pericardial patch graft, and fibrin glue-assisted multilayered amniotic membrane transplantation, with or without a limbal allograft, are among the surgical techniques available.[5–8]

Anterior lamellar keratoplasty (ALK) can treat limbal dermoid with a thickness greater than 100 µm stroma depth and/or a diameter greater than 5 mm.[9] Previous studies have shown good results when treating limbal dermoid using the ALK technique with a partial- or full-thickness corneal graft, with minimal complications.[10,11] Shimmura et al[12] in 2004 were the first to propose component surgery for the cornea. This newly discovered surgical procedure involves replacing the affected region of the recipient's cornea with the donor cornea's lamellar buttons. This method, in which single cornea donor can be transplanted to multiple recipients, aimed to alleviate the corneal tissue shortage crisis during transplantation. Therefore, this study aimed to evaluate the surgical outcomes of limbal dermoid excision using a partial-thickness corneal graft based on split cornea transplantation in patients with ALK. This concept was the first to be applied in our setting, thus becoming a learning point for other limbal dermoid cases.

CASE REPORT

A 20-year-old woman complained of a white lump in her left eye since birth, which grew slowly, was painless, and was without visual disturbances. An elevated, well-defined, painless, and vascularized mass was found in the inferotemporal limbus measuring 4.5 mm in diameter (Figure 1a). Anterior segment optical coherence tomography (AS-OCT) revealed a mass in the corneal-limbal-conjunctiva with half the depth of the stroma (Figure 2a). ALK tumor excision was performed using a post-Descemet's membrane endothelial keratoplasty (post-DMEK) corneal graft. The remnant corneal donor after DMEK, previously split by Descemet removal, was used, leaving approximately 95% of the anterior lamellar button. A conjunctival peritomy was performed at the inferotemporal site. The lesion was dissected in a lamellar fashion using a crescent blade and blunt scissors dissection. The remnant corneal graft was then trephined to have the same diameter as the defect without adjusting the graft thickness. The graft was placed on the recipient bed and radially fixed using 12 interrupted 10-0 nylon sutures. The conjunctiva was attached to the limbus and covered half of the graft

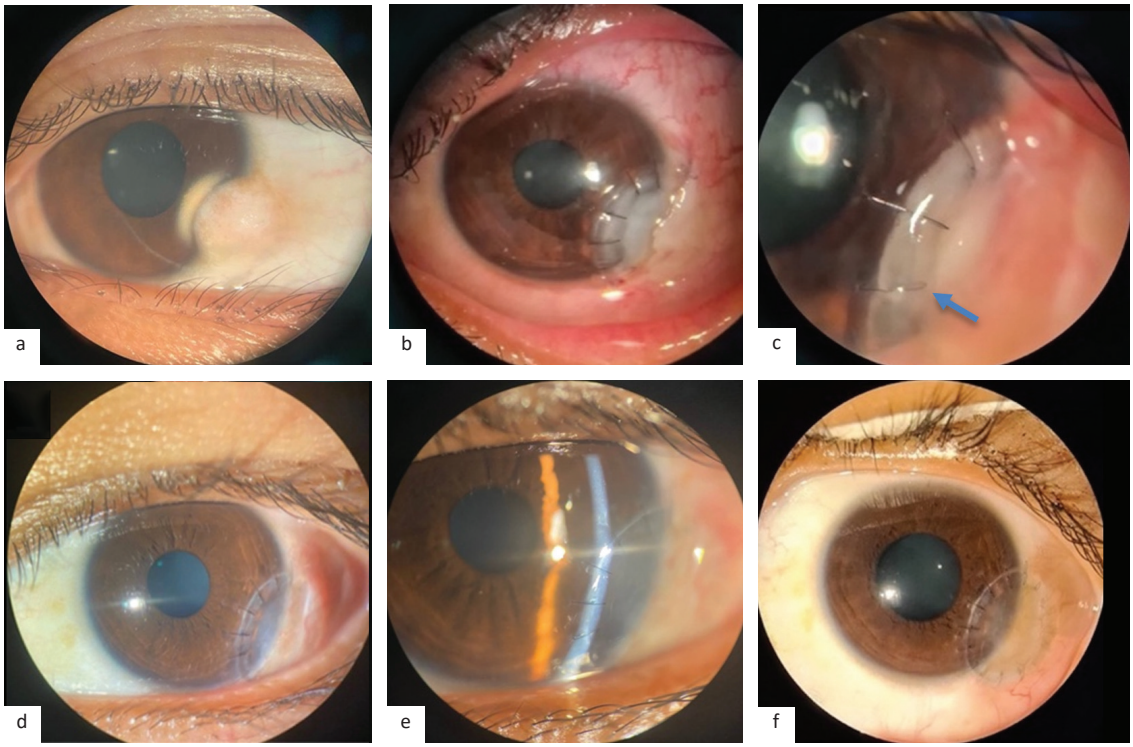

Figure 1. Slit-lamp examination of the patient's left eye. (a) Limbal dermoid before surgery; (b and c) 5 weeks after the first surgery, blue arrow showing loose suture; (d and e) 4 weeks after re-surgery, well-apposed host-graft junction; (f) 3 months after re-surgery

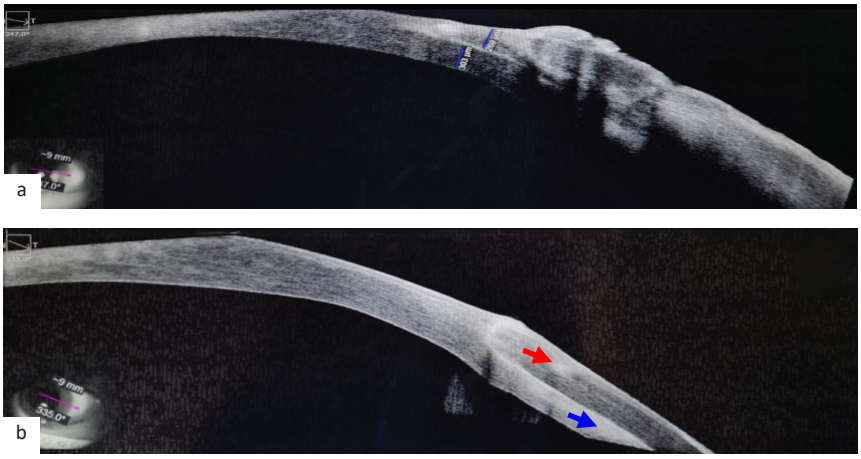

Figure 2. Anterior segment optical coherence tomography (AS-OCT) examination before surgery. (a) The thickness of extending tumor on the cornea measuring 269 µm; (b) 3 months after release and regraft showing good donor-host apposition (red arrow: donor; blue arrow: host)

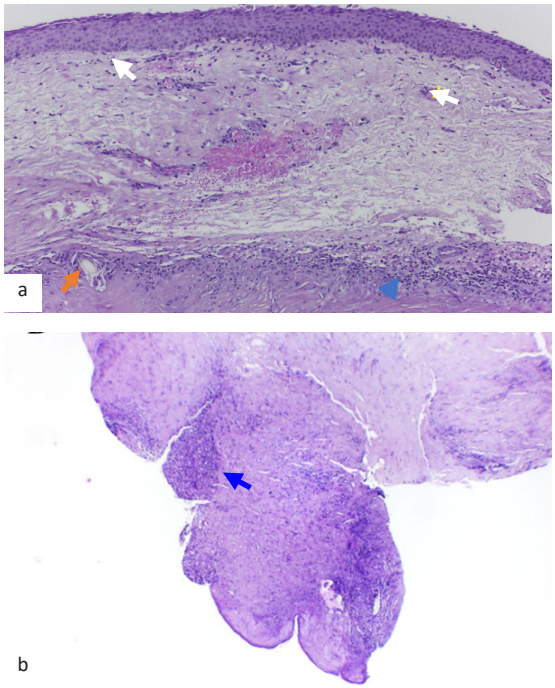

Figure 3. Histopathological examination of the first corneal graft. (a) Corneal grafts lined by hyperplastic squamous epithelial layer and subepithelial consisting of loose and fibrotic tissue, with neovascularization (white arrows), multinucleated foreign-body giant cells (orange arrow), and acute and chronic inflammatory cells (blue arrow) (H&E, 100× magnification); (b) area with dense infiltration of acute inflammatory cells (blue arrow) (H&E, 40× magnification). H&E=hematoxylin and eosin

area. Prednisolone acetate and levofloxacin eye drops were given every 3 hours postoperatively.

After 5 weeks, the graft showed overlapping signs of failure and infection (Figure 1, b and c). The patient complained of redness and foreign-body sensation in the left eye. Edema of the corneal graft, loosening of the suture with infiltration, and graft melting were noted on slit-lamp examination. An immediate plan for release and regrafting using the remnant cornea from post-Descemet's stripping endothelial keratoplasty (post-DSEK) was performed for re-treatment. The corneal tissue surrounding the host corneal defect was dissected using a crescent blade and blunt corneal scissors. The corneal donors were trephined to a defect size of 6.5 mm in diameter. The graft was placed on the recipient bed and secured using a 10-0 nylon suture for 16 radial sutures. The conjunctiva was sutured using vicryl 8-0 around it. The released graft was sent to the Department of Pathological Anatomy for histopathological examination (Figure 3).

Four weeks after re-surgery, good apposition of the host-graft junction with minimal conjunctival hyperemia was observed (Figure 1, d and e). The result at 3 months follow-up was a clear graft and well-apposed graft-host junction on slit-lamp examination (Figure 1f) and in AS-OCT (Figure 2b). Good visual acuity was observed: 0.00 logMAR with –0.50 cylindrical lens correction.

DISCUSSION

Surgery is usually recommended for grade II and III limbal dermoids because they frequently cause refractive or occlusive amblyopia. Surgical techniques include simple excision to the lamellar, penetrating keratoplasty with a corneal relaxing incision, a corneal-limbal-scleral graft transplant, and surgical

excision followed by sutureless multilayered amniotic membrane transplantation or pericardial patch graft, depending on the depth, size, and placement of the lesion.[4–8] The limbal dermoid, in this case, was categorized as deeper-Grade I since it was 4.5 mm in diameter (<5 mm) and extended half the depth of the stroma (>100 µm).[9] We performed partial thickness corneal splitting, followed by lamellar delineation and partial thickness corneoscleral graft implantation in accordance with the recommendation from Pirouzian et al's[9] study on pediatric corneal limbal dermoid management. Lamellar excision of large limbal dermoid with matching partial thickness corneoscleral grafts is safe and provides good cosmetic and tectonic stability without postoperative astigmatism.[10] However, simple keratectomy with or without tattooing may be sufficient if the dermoid is relatively superficial. However, in this case, the dermoid was lodged in the cornea, and simple keratectomy was insufficient. Penetrating keratoplasty is indicated when the dermoid involves the anterior chamber.

Deep anterior lamellar keratoplasty (DALK) has several theoretical advantages over penetrating keratoplasty, including retention of the recipient's healthy endothelium, maintenance of Descemet's membrane integrity, and reduce intraocular surgical complications, which minimizes allograft endothelial rejection and reduces the need for immunosuppression. Lamellar keratoplasty of the limbal dermoid also decreases the incidence of postoperative sequelae, such as corneal opacity, neovascularization, and pseudopterygium. However, DALK is a more time-consuming and technically challenging procedure that might cause interface irregularity and scarring, resulting in lower best-corrected visual acuity and a higher risk of early postoperative interface haze and inflammation.[13,14]

In the present case, during observation from the first day after surgery until 3 weeks follow-up, the graft was clear without edema. However, at 5 weeks follow-up, it showed signs of graft failure, possibly due to graft infection or rejection. According to the literature, the process of graft rejection starts at 3 weeks (10 days) in a successful clear graft. During these periods, follow-ups every week or 2 weeks are advisable to detect signs of failure earlier. Because the corneal endothelium is not transplanted in the ALK technique, endothelial rejection cannot occur and usually responds to corticosteroid therapy.[15] However, in this case report, the etiology of failure was doubtful, stroma rejection showed significant haze, neovascularization occurred, and the graft was released.

Ideally, a histological examination should be performed for every tissue removed from the human body for a definitive diagnosis.[16] Histopathological examination of the first graft should be performed to determine whether the failure was caused by infection or an immunological reaction. The full-thickness donor tissue was lined with hyperplastic squamous epithelium and sub-epithelium consisting of loose and fibrotic tissue. The graft tissue is also infiltrated with mild-to-moderate acute and chronic inflammatory cells, along with foreign-body giant cells and blood vessel proliferation. These findings correlated with possible graft rejection. Graft rejection after ALK/DALK is likely caused by infection (herpetic keratitis, bacterial infection, or fungal infection) or immunological reactions.[13,16,17] Microscopic investigation of corneal transplant failure demonstrates loss of the epithelial basement membrane with invasion of monocytes, lymphocytes, plasma cells, and fibroblasts into the stroma. Alterations in stromal keratocytes may occur when they come in contact with lymphocytes. New capillary development was observed in the anterior and middle layers of the stroma, with immunoblast-like cells in the rejection region. Lymphocytes and plasma cells are predominantly found outside of the endothelial capillary wall.[15] The presence of multinucleated giant cells in the graft can result from persistent inflammation caused by foreign material presence or ongoing infections.[18,19] The donor corneal cells do not appear to be involved in corneal graft healing; instead, healing appears to be a function of the recipient corneal cells. Impairment of this delicate equilibrium during corneal wound healing can result in pathological corneal vascularization.[20–22] Lymphatic vessels form near the corneal limbus under both normal and pathological conditions. Lymphatic vessels can emerge abruptly in the corneal stroma and are not related to the lymphatic vessels in the limbus. Thus, graft failure is associated with interfacial vascularization and stromal opacification.[23]

Previous case reports have successfully demonstrated that a single-donor cornea can be used for multiple recipients in various types of corneal transplantations, including ALK, posterior lamellar keratoplasty, and limbal stem cell transplantation.

The techniques for splitting the cornea vary among studies.[24–27] Sati et al[28] used a manual dissection technique from a single-donor corneal button for three recipients, while Sharma et al[25] dissected the corneal donor with a microkeratome. Manual dissection is considered inferior to microkeratome dissection, but both corneal splitting techniques yield successful visual acuity and cosmesis outcomes.[25,28] Shen et al[11] used full-thickness central corneal grafts in 10 patients with limbal dermoids undergoing lamellar keratoscleroplasty. The results showed good cosmetic outcomes with minimal postoperative complications. All patients had relatively deep lesions, and lamellar dissection was performed until a clear cornea was reached, accounting for approximately 70–80% of the corneal thickness. Owing to the depth of the excised hole, lamellar dissection of the central corneal graft was not performed, and no tissue overriding occurred after surgery.[11]

In the present study, the first corneal graft was obtained from a post-DMEK graft and dissected manually. It left approximately 95% of the corneal thickness to fill 55% of the recipient's corneal defect, resulting in poor host-donor apposition. Regrafting a post-DSEK graft of 55–65% thickness yielded smooth host-donor apposition, good cosmesis, and minimal postoperative astigmatism. Complications after the first surgery possibly occurred because of the thickness difference between the donor graft and the host cornea, which hampered the healing process. Disturbance of the corneal epithelium and anterior stromal integrity during subsequent wound healing promoted the formation of new vessels inside the donor stroma and hastened transplant rejection. It is essential to determine the depth of corneal penetration by the tumor before deciding on the surgical approach. If lamellar keratoplasty is performed, the donor cornea must be trimmed to match the remainder of the patient's cornea after tumor removal.

To date, no case reports have described the use of split-corneal donors in Indonesia. This case report will be useful for other ophthalmologists applying this method. Subsequently, this may reduce the use of donor corneas, which is helpful as most donor corneas in our setting are donated overseas. Split cornea transplantation is a promising strategy for alleviating the corneal tissue shortage crisis in Indonesia. Future studies with longer follow-up durations are needed to evaluate the stability of corneal grafts.

Conflict of Interest

The authors affirm no conflict of interest in this study.

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