Introduction of Epithelial Cells in the Flap-Graft Interface During Descemet Stripping Automated Endothelial Keratoplasty

Microkeratome-assisted Descemet stripping automated endothelial keratoplasty (DSAEK) is a form of lamellar corneal surgery that allows for the selective replacement of diseased endothelium. The DSAEK procedure is relatively new. So far, few complications have been described; these include graft detachment and graft failure. Recently, epithelial ingrowth in the flap-graft interface after DSAEK has been reported. We report a case of introduction of epithelial cells, originating from the donor tissue, in the flap-graft interface.

Report of a Case. In May 2005, a 67-year-old woman visited with pseudophakic bullous keratopathy in the right eye. Best-corrected visual acuity was 0.08 OD. Because of the vision-limiting bullous keratopathy in the right eye, a DSAEK was performed under general anesthesia. A cornea from a male donor was used.

One week postoperatively, the posterior lamellar graft was detached inferiorly and nasally. An additional air bubble and two 10-0 nylon sutures were placed to fixate the transplant. Thereafter, partial adherence of the lamella was seen.

Four months postoperatively, the graft was attached and best-corrected visual acuity improved to 0.63 OD, with -1.00. Between the 3-o’clock and 6-o’clock positions, anterior synechia were present and the interface was slightly hazy. During regular follow-up, the abnormalities remained stable under fluorometholone, 1%, eye drops. The intraocular pressure remained between 10 and 17 mm Hg during follow-up.

One year after DSAEK, the patient had progressive blurring of vision to hand movements. We observed total corneal decompensation with bullae and Descemet membrane folds. Therapy with prednisolone acetate, 10%, eyedrops (Pred Forte) 6 times a day, prednisolone pivolate, 5%, eye gel (Ultracortenol) 3 times a day, and preservative-free chloramphenicol, 0.4%, eyedrops 3 times a day proved to be unsuccessful. A penetrating posterior mushroom keratoplasty was performed. Six months after surgery, best-corrected visual acuity improved to 0.80 OD with -3.25 – 2.00 x 30°. The corneal graft in the right eye has remained clear.

Histopathologic examination of the removed corneal button demonstrated 2 cysts at the interface between the recipient cornea and the donor stroma. One cyst was lined by squamous epithelium; a smaller, centrally located cyst contained eosinophilic granular material (Figure 1A). The Descemet membrane with some excrescences was incarcerated in the surgical scar. There was bullous keratopathy of the corneal epithelium. On immunohistochemistry, the lining of the cyst as well as the granular material stained strongly positive for the epithelial markers 34Be12 and keratin 5/6 (Figure 1B). The granular material was consistent with degenerated epithelial cells. In situ hybridization with X and Y probes revealed 2 X chromosomes in the nuclei of the epithelium and the stroma of the recipient. The nuclei of the epithelium of the cyst wall contained 1 X chromosome and 1 Y chromosome (Figure 2).

Comment. In our patient, the DSAEK was complicated by introduction of epithelial cells along the interface. Her native Descemet membrane with Fuchs endothelium dys-
tropho was partially incarcerated in the surgical scar, similar to the earlier described case, suggesting an invasive postoperative ingrowth process. The X-Y karyotyping revealed that the epithelial cells were of donor origin. The technique of posterior lamellar keratoplasty is relatively new and technologically difficult, with a surgeon's learning curve. The most critical step is the preparation of the posterior lamellar disc by hand or by use of a microkeratome. We postulate that the donor epithelium was implanted during the preparation of the donor posterior lamellar disc and was introduced intraoperatively. If complete attachment of the donor posterior lamella is accomplished, ectopic epithelial cells in the interface might remain stable without proliferation. In a partially detached donor lamella, ectopic epithelial cells might proliferate along the recipient's posterior cornea.

**Figure 2.** In situ hybridization with X and Y chromosome probes revealed 2 X chromosomes (arrow) in the epithelium and the stroma of the recipient (A) and 1 X chromosome (arrow) in the epithelium of the cyst (B). C, The epithelium of the cyst wall contained 1 Y chromosome (arrows). D, There was no staining of the Y chromosome probe (arrow) in the tissue of the recipient.

**Congenital Melanocytoma Manifesting as Proptosis With Multiple Cutaneous Melanocytic Nevi and Oculodermal Melanosis**

Melanocytomas are benign, pigmented tumors usually seen in adults. They are normally asymptomatic, localized, and unassociated with any systemic features. We report a case of congenital melanocytoma with diffuse ocular involvement, extending into the orbit and causing proptosis, along with oculodermal melanocytosis and multiple congenital melanocytic nevi on the body.

**Report of a Case.** A 6-month-old boy had progressive protrusion of the right eye since birth. He had a proptosis measuring 34 mm with a pigmented subconjunctival mass in the upper fornices and cutaneous pigmentation on the upper eyelid (Figure 1A). Multiple pigmented cutaneous lesions were seen throughout the body, ranging from 1 to 10 mm (Figure 1B).

A computed tomographic scan showed a large lesion infiltrating the right eyeball and posterior orbit, causing scalloped expansion of the orbit (Figure 1C). An incisional biopsy specimen taken through the superior fornix revealed a melanocytoma. Because melanocytomas of such infiltrative nature and progression can become malignant, an exenteration was done. A nevus was also biopsied. Results of a complete metastatic workup were normal. Gross examination showed a pigmented mass measuring 35 × 25 × 25 mm, filling up the globe and extending superoposteriorly into the orbit (Figure 1D). Microscopical analysis showed heavily pigmented tumor cells, of which bleached preparations revealed plump polyhedral nevus cells with abundant cytoplasm and small, uniform nuclei (Figure 2A and B). The tumor had infiltrated the sclera posteriorly and replaced the entire optic nerve. There was no evidence of mitosis or vascular invasion. Staining for S-100 protein and HMB-45 was positive. Staining for Ki-67 showed a proliferative index less than 1%. Features of melanosis oculi in the form of increased dendritic melanocytes were seen in the episclera, sclera, and optic nerve sheath (Figure 2C). Results of the microscopical analysis of the cutaneous lesion were consistent with a congenital melanocytic nevus (Figure 2D).

Three years later, there was no recurrence.

**Comment.** Melanocytoma, although previously often confused with malignant melanoma, is now distinctly recognized by its typical clinical and benign histological features. The tumor in this case was diffuse and infil-