Labial Salivary Glands Transplantation for Severe Dry Eye Due to Stevens-Johnson Syndrome

Dr. V. P. Gupta

Aim: Labial salivary glands transplantation (LSGT) was evaluated in patients with severe dry eye due to Stevens-Johnson syndrome (SJS). Methods: 21 eyes (11 patients) with dry eye due to SJS underwent LSGT in superior/inferior fornix. A detailed history and ocular examination including Schirmer I, basic secretion test and BUT were performed before and after surgery. All patients had a preoperative Schirmer I test value of zero. Results: There was a statistically significant improvement in the BCVA, Schirmer I, BST, BUT and corneal transparency in all patients. The use of lubricants was reduced in all cases. All patients reported marked symptomatic improvement in photophobia, foreign body sensation, pain and ease of opening the eyes. Follow-up period ranged from 9-24 months. Conclusion: LSGT resulted in subjective and objective improvement of dry eye. It appears to be a promising new treatment for severe dry eye due to SJS. This is the first report of LSGT in dry eye from India.

Stevens-Johnson syndrome (SJS), also known as erythema multiforme major involves the eye in about two-thirds of cases. Cicatricial abnormalities in the conjunctiva during the healing phase and destruction of the limbal epithelial stem cells, result in symblepharon, entropion, trichiasis, dry eye, keratinisation and corneal neovascularisation. Various surgical options for severe dry eye include permanent punctal occlusion, tarsorrhaphy, oral mucosal grafting, salivary gland duct transposition, amniotic membrane transplantation, submandibular gland transplantation and labial salivary glands transplantation (LSGT).

In 1997, Murube described transplantation of labial salivary glands for the treatment of severe dry eyes. Minor oral salivary glands exist in large numbers in the labial, buccal and palatal mucosa, and account for approximately half the baseline secretion of saliva. Labial salivary glands along with the overlying mucosa as a complex graft has been transplanted to the posterior lamella of the eyelids to increase ocular surface and reduce discomfort in patients with severe dry eye. However there is no report of LSGT for severe dry eye due to SJS from India. The purpose of the present study was to evaluate labial salivary gland transplantation in patients with severe dry eye due to SJS.
MATERIALS AND METHODS
This prospective, non-comparative, interventional case series included 21 eyes (11 patients) with severe dry eye secondary to SJS. Patients were recruited from the Cornea and Oculoplastic, orbital and lacrimal services, department of Ophthalmology, University College of Medical Sciences and GTB Hospital, Delhi–110095. Detailed preoperative assessment including a detailed history and ocular examination with slit lamp biomicroscopy was performed on all patients. Preoperative dry eye work-up including Schirmer I, basic secretion test, tear film breakup time (BUT) and syringing were performed before surgery and postoperatively periodically. All patients were requested to complete a questionnaire on symptoms such as photophobia, foreign body sensation, pain, dryness, eye movements and ease of opening the eyes, pre and postoperatively to compare the improvement following surgery on a scale of 0 to 5 with 0 representing no improvement and 5 representing marked / definite improvement.

Inclusion criteria: SJS patients with severe, constant and disabling symptoms, and signs of level 4 dry eye and a preoperative Schirmer I test value of zero were included. Exclusion criteria consisted of any active infection, corneal perforation or melt, dry eye due to primary or secondary Sjogren syndrome, and xerostomia. Routine laboratory investigations like haemogram and urine analysis and pre anaesthetic check up were done. Photographic documentation was done pre and post operatively. All the operations were done under general anaesthesia by the same surgeon (VPG) after obtaining informed consent.

Surgical procedure
All the patients underwent labial salivary gland transplantation (LSGT) in superior/inferior fornix. Labial salivary glands were obtained en block with the mucous membrane of the patient’s own lower lip, above the orbicularis oris muscle. The labial salivary glands with mucosal graft were placed in the superior / inferior or both conjunctival fornices after creating a pocket above tarsal plate in central or nasal third of eyelid. The graft was sutured using 8-0 polygalactin continuous suture with buried knot.

RESULTS
21 eyes (11 patients) with severe dry eye due to SJS were included in the study. Age of the patients ranged from 22-45 years with mean age of 30.25 years. There were 8 males and 3 females. None of the cases had undergone any similar surgery in the past. All patients except one underwent bilateral LSGT. The graft sizes were 15mm x15 mm to 20mm x15 mm. No intraoperative complications except for intraoperative bleeding while harvesting the graft were observed. Lid edema, mechanical ptosis and inability to open the eyes were noted in all cases for first 3-4 days postoperatively. All patients had level 4 dry eye with a
preoperative Schirmer I test value of zero. There was a statistically significant improvement in the BCVA, Schirmer I, BST, SPK and corneal transparency/neovascularisation in all patients. The use of lubricants the frequency as well as number, was reduced in all cases. All patients reported symptomatic improvement (2-4 in scale of 0-5) in photophobia, foreign body sensation, dryness, ocular motility, pain and ease of opening the eyes. Follow-up period ranged from 9-24 months. All grafts have been intact till date. No donor site complications were noted.

**DISCUSSION**

Labial salivary glands transplantation (LSGT) is a recent modality of treatment to substitute tears for severe dry eyes. Experience with this technique is still very limited. This is the first report of LSGT in dry eye secondary to SJS from India. Murube’s technique was further modified with the use of Ellman high-frequency/low-temperature radiosurgical device for harvesting the graft without damaging the glands. Soares and França described a successful outcome with the same surgical treatment in 21 cases of severe dry eye, 12 (57.1%) of which occurred secondary to SJS. In our study, LSGT was performed in dry eye due to SJS only. The results of the questionnaire showed symptomatic improvement (2-4 on scale of 0-5) in photophobia, foreign body
sensation, dryness, ocular motility, pain and ease of opening the eyes. There was a significant improvement in the BCVA, Schirmer I, BST, SPK and corneal transparency / neovascularisation in all patients. The use of lubricants was reduced in all cases. All grafts have been intact during 9-24 months follow-up. However, the long-term viability of LSGT needs to be established.8

LSGT is believed to improve the outcome of limbal and corneal graft in patients with SJS by improving lubrication of the ocular surface.7 Raus (2003) reported improvement in the Schirmer test and tear break-up time following LSGT from lower lip in 10 cases of dry eye, although the etiology of dry eye was not mentioned.3 Similar results of improvement in their ocular discomfort, BCVA, corneal staining, Schirmer’s test, break-up and reduced need of lubricants have been reported by others after LSGT for severe dry eye due to SJS.1,7

The graft size was 15mm x15 mm to 20mm x15 mm in the present study. Soares and França5 described an elliptical incision in the labial mucosa which was 2-3 cm long and 1cm wide. Geerling et al4 harvested a minor salivary glands graft of approximately 2.5 x 2 cm by means of a surgical knife or radiosurgical device. The graft was cut in two strips of approximately 2.5 x 1 cm size. In our opinion, the size of the graft and the number of the glands should play a vital role in determining the amount of secretion of “salivary tears”. The flow rate of saliva from human labial salivary glands (LSG) has been reported as 0.05 – 4.8 μl/cm²/min.6 The data for single gland gives a comparable figure of 0.1 μl/min/gland.6 Thus larger graft having more number of LSG or even two grafts one in each fornix may further improve the success rate. In a recent study greater tear production was observed in patients who received submucosal grafts containing more than 10 glands.7 We feel LSGT may be more effective in patients with prior punctal / canalicular occlusion preventing the drainage of “salivary tears”.

We had excluded the cases having Sjögren’s syndrome and xerostomia that affect the salivary glands leading to poorer results. Additionally, the possibility of the graft being involved by a recurrent inflammation in the late postoperative period caused by the disease which originally produced the dry eye still remains a major concern. No relapse of dry eye occurred during follow-up between 6 and 27 months.1 Dry eye recurred in two eyes with Stevens Johnson syndrome and one eye with pemphigoid during 4 year follow-up after LSGT.5 Repeat surgery may have difficulties due to cicatricial changes in the eyes, oral mucosa with chance of harvesting less number of minor salivary glands leading to suboptimal outcome. However, successful results been reported with repeat LSGT.5,7

LSGT resulted in subjective and objective improvement of dry eye. It appears to be a promising new treatment for severe dry eye due to SJS.
REFERENCES


