RESEARCH ARTICLE

Anatomical success of tectonic keratoplasty in children at a tertiary care eye hospital

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Abstract

Objective: To evaluate the anatomical outcome of tectonic grafts performed in children with perforated or melted corneas.

Method: The prospective study was conducted at the Department of Paediatric Ophthalmology, Al-Shifa Trust Eye Hospital, Rawalpindi, Pakistan, from January to December 2017, and comprised children of either gender aged <16 years who received tectonic grafts owing to desmetocoele or corneal perforation of >3mm. The causes of corneal perforation, size of perforation, post-operative graft outcome and globe integrity were noted. Periodic follow-ups were done till December 2019. Data was analysed using SPSS 22.

Results: Of the 27 patients, 9(33.3%) were girls and 18(66.7%) were boys. The overall mean age was 47.7±43.5 months. The most common indication was corneal perforation 19(70.4%), followed by descemetocoele 8(29.6%). The most frequent cause was infectious keratitis 13(48.2%), followed by trauma 8(29.6%) and vitamin A deficiency 5(18.5%). The mean size of perforation/descemetocoele was 5.5±1.5mm. Post-operatively, graft failure occurred in 5(18.5%) eyes and required repeat grafts. Globe integrity was restored in all 27(100%) cases, and 24(88.8%) eyes maintained their size and intraocular pressure, while 3(11.1%) became phthisical. Post-operative visual acuity was better than 6/36 in 3(11.1%) eyes, between 6/36 and 6/60 in 10(37%) and counting finger in 14(51.8%).

Conclusion: Tectonic graft was found to be a valuable therapeutic option in emergency globe-threatening corneal thinning and perforation and in maintaining the anatomical integrity of eyes.

Keywords: Keratoplasty, Corneal perforation, Corneal grafting.

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Introduction

Corneal melting and perforations are ophthalmic emergencies that can lead to loss of globe and vision. Emergency treatment is warranted to maintain the integrity of the globe and to preserve vision.

Various infectious and non-infectious disorders can lead to corneal perforations. Infectious causes include bacterial, fungal, viral and acanthamoebal keratitis.[1,2] Non-infectious causes include trauma, lagophthalmos, xerophthalmia, keratoconus, atopic keroto-conjunctivitis, Sjogren syndrome and idiopathy.[1-3] According to a study conducted in a tertiary care unit in northern China, trauma was identified as the leading cause of corneal perforation leading to enucleation in children.[4]

Different treatment options are available depending on size, location of perforation and visual prognosis. Small perforations can be treated with bandage contact lenses, corneal gluing, amniotic membrane transplantation (AMT) and conjunctival flaps. But large-sized defects require tectonic grafting and at worst evisceration or enucleation.[1,2]

Ramon Castroviejo described the world's first total penetrating keratoplasty in 1951 as a procedure for replacing full-thickness diseased cornea with donor tissue.[5] Technical advancements have made the corneal grafting possible among paediatric population, but allograft rejection remains an important cause for graft morbidity in the younger age group and significantly influences the clarity and survival time of corneal grafts. Paediatric corneal transplantation has an increased rejection rate because of the more active immune system in younger patients. The use of steroids and other immunosuppressants can significantly decrease the rate of immunological rejection of the graft. Intensive medications are necessary to decrease risk of surgery failure and recurrence of the primary disease, which can cause profound loss of vision or loss of the entire globe.

The current study was planned to evaluate the causes requiring urgent tectonic corneal grafts in children, and to determine the outcomes of tectonic keratoplasty in the paediatric population.

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Patients and Methods

The prospective, interventional study was conducted at the Department of Paediatric Ophthalmology, Al-Shifa Trust Eye Hospital, Rawalpindi, Pakistan, from January to December 2017, while periodic follow-ups lasted till December 2019. After approval from the institutional ethics review board, the sample was raised using convenience sampling technique. Those included were children of either gender aged <16 years who received tectonic grafts owing to descemetocoele or corneal perforation of >3mm secondary to either infectious or non-infectious aetiology. Data, including demographics, causes of corneal perforation, size of perforation, post-operative graft outcome and globe integrity, was noted after taking informed written consent from the parents/guardians of all the subjects. After keratoplasty, any child with follow-up of less 4 months or lost to follow-up was excluded from the study.

All the children had not responded to medical treatment. Among the infectious causes, corneal thinning and melting was progressive despite the treatment with topical and systemic antibiotics or antifungals, based on the corneal scrapings and culture reports. Among the non-infectious causes, children with vitamin A deficiency had already received systemic supplements and children with trauma had extensive corneal tissue loss and additional corneal melting due to super-imposed infection. These cases were not suitable for corneal glue or AMT, due to large size of the thinning or perforations and associated extensive necrosis. The clinical picture in all the cases was so aggressive that urgent surgical intervention was needed to preserve the integrity of the eye.

Complete ophthalmic evaluation including brightness amplitude scan (B-scan) ultrasound was performed to assess the vitreous involvement and post-operative prognosis. Systemic evaluation was carried out by a paediatrician regarding the general health and supplemental treatments. A detailed discussion was carried out with the parents regarding the prognosis and risks and benefits of the procedure and the need for multiple surgeries.

All surgeries were performed as an emergency procedure under general anaesthesia (GA) under strict aseptic techniques. The selected donor tissues were the preserved corneal buttons of the keratoconus cases who had undergone penetrating keratoplasty at the same hospital. Corneal buttons had been stored in Optisol GS medium (GS - gentamicin and streptomycin, Bausch & Lomb Surgical, Inc.) and were preserved at 4°C. These buttons were used within 2 weeks of preservation. The donor size and shape were customised according to the size of perforation and was kept 0.5mm larger than the recipient bed. Area of corneal perforation was cleaned, and necrotic tissues were removed off the recipient bed along with a margin of healthy tissue. Graft was sutured with interrupted sutures using nylon 10/0.

Post-operatively, topical antibiotic moxifloxacin 2 hourly, steroid prednisolone acetate 1% 1 hourly, cycloplegic cyclopentolate 1% 8 hourly and lubricants were prescribed and tapered slowly over a period of 6 months. Follow-up examinations were carried out weekly in the first month, fortnightly for 3 months, monthly for 6 months and quarterly afterwards. Visual acuity (VA), intraocular pressure (IOP), anterior segment, and fundus examination and B-scans were performed at each follow-up.

Variables of interest were indications and causes of corneal perforation or melting, size of perforation, outcome of grafts, post-operative VA, and preservation of the globe.

The collected data was analysed using SPSS 22. Mean ± standard deviation values were calculated for age, size of perforation and follow-up duration. Frequencies and percentages were calculated for the different indications for tectonic keratoplasty, causes of corneal perforation, cases with anatomical success and graft failure. Kaplan-Meier survival curve was used to show timing of graft failure (event) during the follow-up.

Results

Of the 27 patients, 9(33.3%) were girls and 18(66.7%) were boys. The overall mean age was 47.7±43.5 months. Of the total, 26(96.3%) patients received corneal grafts, while 1(3.7%) required corneoscleral graft. Mean follow-up duration was 20.7±6.1 months (range: 12-36 months). The most common indication was corneal perforation 19(70.4%), followed by descemetocoele 8(29.6%). The most frequent cause was infectious keratitis 13(48.2%), followed by trauma 8(29.6%) and vitamin A deficiency

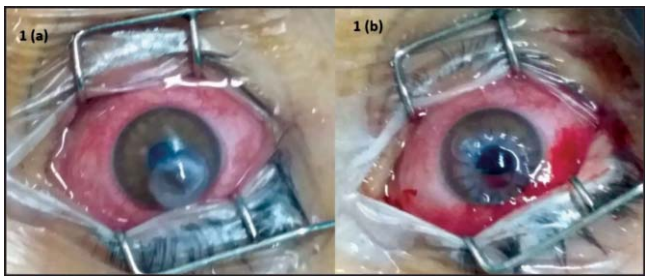

Figure-1: A case with keratitis and desmetocele before surgery (a) and after the tectonic graft (b).

Table: Summary of the post-operative complications and their management.

Complications	No of cases (n=27)	Treatment of Complications	Outcome of Treatment		Repeat Surgery	Outcome of Repeat surgery
No Complications	22		Graft Haze	Anatomical success		
Rejection	3	Topical & Systemic Antibiotics And Topical & Systemic Steroids	Severe Graft Rejection & Melting of Cornea	Graft Failure	Repeat tectonic graft	2 Anatomical Success & 1 Phthisis
Recurrence of Keratitis	1	Topical & Systemic Antibiotics	Graft Thinning & Perforation	Graft Failure	Repeat tectonic graft	Phthisis
Trauma	1		Traumatic Perforation of Graft	Graft Failure	Repeat tectonic graft	Phthisis

Figure-2: A case with keratitis and desmetocele before surgery (a) and after the tectonic graft (b).

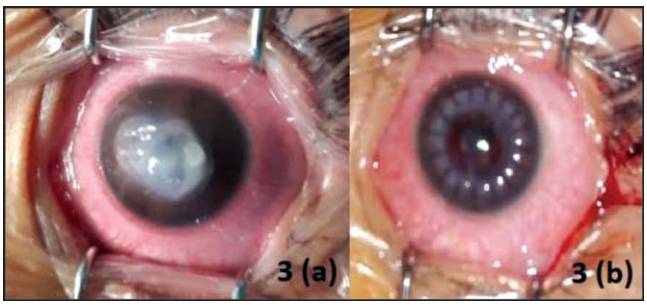

Figure-3: A case with keratitis and desmetocele before surgery (a) and after the tectonic graft (b).

5(18.5%). The mean size of perforation/descemetocoele was 5.5±1.5mm (range: 3.5-10mm).

Globe integrity was restored in all 27(100%) cases (Figure 1-3). Post-operative VA was better than 6/36 in 3(11.1%) eyes, between 6/36 and 6/60 in 10(37%) and counting finger in 14(51.8%) (variance 0.5).

Post-operatively, graft failure occurred in 5(18.5%) eyes and required repeat grafts. Graft rejection was the main cause leading to graft failure in 3(11.1%) cases, followed by infectious keratitis in 1(3.7%) and traumatic graft rupture in 1(3.7%). Complications such as shallow anterior chamber, wound leakage, cataract, graft dehiscence, graft melting, glaucoma or endophthalmitis were not seen in any of the cases (Table).

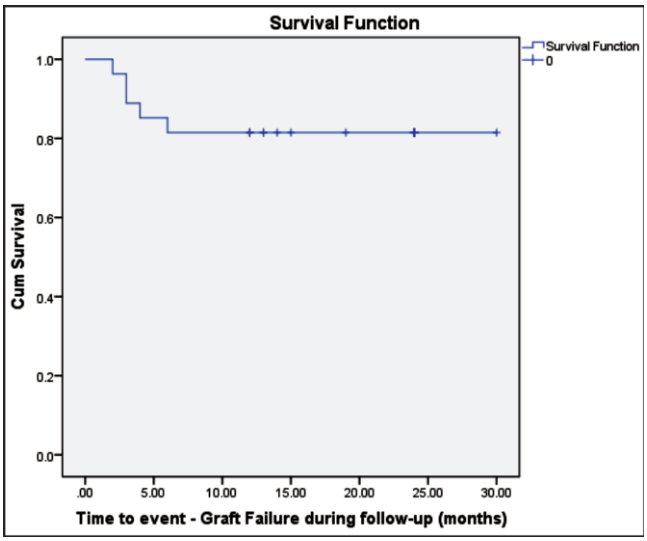

Figure-4: The Kaplan-Meier graph showing the timing of graft failure (event) during follow-up.

Kaplan-Meier graph recorded the timing of graft failure (event) during the follow-up (Figure 4).

During the follow-up, 24(88.8%) eyes maintained their size and IOP, while 3 (11.1%) became pthisical (variance 0.1). There was no mortality during the intervention and follow-up phases.

Discussion

In the current study, tectonic grafts were performed in children with corneal perforation or impending perforation. Corneal desmetoceles and perforations were >3mm in size and had associated corneal melting and tissue loss. All these cases were not suitable for conservative treatments, like bandage contact lenses, corneal gluing, or amniotic membrane transplantation. Similarly, primary corneal repair and suturing was also not possible because of the large size, tissue-loss and necrosis. Owing to the aggressive condition, these children were managed in emergency as waiting for

elective optical keratoplasty was not possible. If they had not been treated urgently, these children would have developed severe infection, eventually requiring evisceration, which is traumatising for the children as well as their parents. Therefore, one of the factors determining the prognosis in these children was the urgency of treatment.

The primary objective of the tectonic grafts was to attain the structural integrity of the globe, and avoidance of evisceration or enucleation with visual rehabilitation afterwards. Structural integrity of the globe was restored in 88.8% eyes and elective penetrating keratoplasty could then be considered, depending upon the prognosis in each case. Phthisis developed in 11.1% cases due to inflammation and graft failure. None of the children required evisceration or enucleation.

In the study, the common conditions causing corneal perforation or descemetocoele and requiring emergency tectonic grafts were infectious keratitis followed by trauma and vitamin A deficiency. Various infectious and non-infectious corneal diseases can lead to corneal melting and perforation, resulting in loss of vision and globe, if not treated promptly. In a single-centre Danish study, out of all the paediatric keratoplasties performed over 40 years, 41% were carried out due to tectonic indications.[6]

In the current study, infectious keratitis leading to corneal perforation was the most common indication requiring tectonic keratoplasty. In another Pakistani study, corneal scars post-keratitis and trauma were the third most frequent indication after keratoconus and corneal dystrophies even for elective paediatric keratoplasties.[7] In a Malaysian study, infective keratitis was the main indication (56.2%) for the paediatric keratoplasties performed over 10 years.[8] In an Indian study, the common indications for therapeutic keratoplasty were perforated corneal ulcers (56.1%) and non-healing corneal ulcers (24.6%).[9] In a United Kingdom study, out of 1330 emergency corneal grafts performed over 6 years, 30.5% patients had severe infection, 28.5% had threatened perforation and 65.9% had actual perforation.[10]

Ocular trauma was the second most common indication in the current study. Ocular trauma is common in children, affecting approximately 6 million children worldwide every year.[11] Out of those, 5% children sustain severe globe injuries necessitating hospital admission.[11] In an earlier study at the same hospital where the current study was conducted, 509 cases of ocular trauma in children were reported over 5 years.[12] In the current study as well as in various other studies, ocular trauma was more frequent in boys,[12] which can be due to the difference in the nature of the activities of boys and girls. In a few studies, keratoplasty was indicated due to ocular trauma in 12.5%[8] to 27%[13] children.

Vitamin A deficiency was the third frequent cause in the current cases. Xerophthalmia due to severe vitamin A deficiency is one of the leading causes of preventable blindness in children.[14] Severe subclinical deficiency of vitamin A prevails in Pakistan because majority of population belongs to low socioeconomic group with limited availability of food rich in vitamin A. Vitamin A deficiency is further exaggerated by measles, diarrhoea and respiratory tract infections.[14] The most vulnerable age group is that of children, especially of age <6 years, with blinding xerophthalmia, corneal xerosis, corneal ulcers and keratomalacia in about 61% of these children.[15]

Keratoplasty is a technically challenging procedure, especially in cases of corneal perforation, or impending perforations. Corneal thinning, tissue necrosis, collapsed anterior chamber and hypotony make it difficult. Results are unpredictable due to the infectious process, inflammation and corneal vascularisation. In children, the situation becomes more complex due to decreased scleral and corneal rigidity as well as severe inflammation, with increased chances of graft rejection.[16]

In most of the current cases, guarded prognosis was expected due to the advanced and aggressive nature of the underlying disease. The main purpose was globe salvage, which was successfully achieved in 88.8% patients and future keratoplasty for optical indications could be planned, based on the anatomical outcome of each case. The probability of graft survival for patch grafts is observed to be lower than penetrating keratoplasties performed for optical indications.[17] In a study,[6] globe preservation was also achieved in 70% paediatric graft cases. In another study,[18] therapeutic-tectonic keratoplasty resulted in globe salvage in 97.6% patients with perforated infectious corneal ulcers. In one study,[9] therapeutic penetrating keratoplasty was performed in 57 patients aged 2-76 years. After keratoplasty, anatomical success (restoration of integrity of globe), therapeutic success (eradication of primary infection) and functional success (preservation of vision) was observed in 85.96%, 89.47% and 70.17%, respectively.[9] A study observed that in cases with open globe injury, after penetrating keratoplasty, graft survival at 1 and 5 years was 80.4% and 41.7%, respectively.[19] One study reported graft survival at 1, 2 and 5 years to be 78%, 66% and 47%, respectively, in cases of emergency keratoplasties.[10]

Corneal perforation in majority of the current cases was large, and full thickness corneal grafts were carried out according to the size of perforations. Due to the variability in the size of perforation, standard techniques of trephination were not possible in all cases, and the donor and recipient were customised according to the perforation. Eccentric corneal perforations can be treated with tectonic grafts, but increasing decentration of the graft is associated with more risk of glaucoma and corneal haze.

In one patient in the current study, who had cornea-scleral perforation, in addition to corneal button, scleral ring was also used as a patch. Corneo-scleral tectonic patch graft was done in 1 case, but large number of cases are required to assess the outcome of sclero-keratoplasty in children.

The mean age of the children in the current study was 47 months, and majority of the children were aged <4 years. According to various studies, the younger age group, especially <5 years, was associated with poor graft survival and prognosis, with more chances of graft failure.[6,20]

In 18.5% cases in the current study, there was graft failure due to graft rejection, keratitis, and traumatic graft rupture. In one study, corneal grafts were hazy in 68.7% children after keratoplasty and was secondary to graft rejection, glaucoma and graft failure.[8] Graft failure has been documented in 52.6% of therapeutic grafts in cases with advanced infectious keratitis.[9] In cases with open globe trauma, after patch grafts, risk factors for graft failure were graft rejection, retinal detachment and endophthalmitis.[19] According to literature, factors associated with increased risk of graft failure include young age, corneal vascularisation, inflammation, rejection, infective keratitis, secondary glaucoma, large graft size and trauma.[8,21-23] Anterior vitrectomy, combined surgeries, additional procedures after graft and repeat grafts also add to the risk of graft failure.[8,21-24] According to a study,[16] factors associated with reduced graft survival in paediatric keratoplasty include ocular surface disease, corneal vascularisation, previous active inflammation, and presence of glaucoma drainage device.[16]

In the current study, globe anatomy was restored in 88.8% eyes after tectonic keratoplasty. While globe was salvaged, corneal clarity and vision was limited due to the aggressive nature of the primary disease and ongoing inflammation post-operatively. Post-operative VA was better than 6/36 in 11.1% cases, between 6/36 and 6/60 in 37% and counting finger in 51.8% cases.

Poor visual outcome has been documented after keratoplasty in eyes with ocular trauma.[6,13] In a study,[19] after tectonic grafts in eyes with open globe injury, ambulatory vision was gained in 35.7% only.[19] Factors associated with poor visual outcome, after corneal grafts in eyes with ocular trauma, are graft rejection, retinal detachment and endophthalmitis and injury outside the workplace.[19]

Variable and limited visual improvement have been documented after keratoplasty in eyes with keratomalacia. A study[25] performed emergency tectonic keratoplasty in 15 children with acute keratomalacia and corneal melting. Out of 15 eyes with total corneal sloughing, globe was salvaged in 73.3% and phthisis developed in 36.7%. Only 33.3% eyes had clear corneas after a mean follow-up of 7.3 months. Poor visual outcome was due to persistent ocular surface problems and graft rejection, with none of the cases achieving vision better than 6/60.[25]

The current study has limitations as tectonic grafts were used. Donor buttons used in tectonic cases are not clear corneal grafts. In addition, due to the aggressive nature of primary disease, there was a high risk of post-operative infection, inflammation, graft rejection and reduced corneal clarity. So guarded visual prognosis was expected. But the main purpose was to save the integrity of eyes with perforations and to prevent evisceration and enucleation, which was achieved after which elective keratoplasty for optical reasons could be planned.

Conclusion

Infectious keratitis, ocular trauma and vitamin A deficiency were the common causes in children leading to corneal thinning and perforations requiring tectonic keratoplasty. Tectonic graft in children is technically difficult, but is an extremely valuable therapeutic option in emergency sight-threatening situations of corneal thinning and perforation. It helps to restore the anatomical integrity of the globe and evisceration can be circumvented. It results in preservation of vision, depending on the extent of damage to other ocular structures, and buys time so that further procedures can be performed to improve vision afterwards.

Disclaimer: We declare that the abstract has not been previously presented or published in a conference and the manuscript was not part of a research, PhD or thesis project.

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