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Surgical outcomes with 360-degree suture trabeculotomy in poor prognosis primary congenital glaucoma and glaucoma associated with congenital anomalies or cataract surgery

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Abstract

Purpose—To evaluate the outcomes of 360-degree suture trabeculotomy in childhood glaucoma with poor prognosis.

Subjects and Methods—A nonrandomized, retrospective chart review was performed on pediatric patients (under 18 years old) treated with a 360-degree suture trabeculotomy for glaucoma. The cases were categorized into the following groups: (1) primary congenital glaucoma with birth-onset presentation accompanied by corneal clouding noted at birth, (2) primary congenital glaucoma with onset or presentation after 1 year of age, (3) primary congenital glaucoma with prior failed goniotomy surgery, (4) infantile-onset glaucoma following congenital cataract surgery, and (5) infantile-onset glaucoma with associated ocular/systemic anomalies.

Results—A total of 45 eyes of 33 patients were analyzed. The mean preoperative intraocular pressure (IOP) was 34.3 ± 6.7 mm Hg on an average of 1.5 medications. Median age at time of surgery was 7 months. Mean final IOP (median last follow-up or failure, 12 months) was 22.2 ± 7.1 mm Hg on an average of 1.5 medications. The probability of success according to time after surgery was 87% at 6 months, 63% at 1 year, and 58% at 2 years. Kaplan-Meier analysis of Groups 1-4 versus Group 5 failed to demonstrate a statistically significant difference ($p = 0.13$). Of 5 eyes with port wine mark-related glaucoma, 2 had a large (>50%), persistent postoperative hyphema and concurrent vitreous hemorrhage.

Conclusions—Children with a wide range of ocular pathologies can be successfully treated with 360-degree suture trabeculotomy. Further evaluation of this surgical technique in primary congenital glaucoma and open-angle glaucoma following congenital cataract surgery is warranted.

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Introduction

Primary congenital glaucoma generally responds well to surgical treatment with goniotomy or trabeculotomy, with reported success rates (with or without glaucoma medication) of 80% to 93%.¹⁻⁴ However, presentation of primary congenital glaucoma at birth, late presentations after one year of age, and eyes requiring more than one goniotomy or trabeculotomy have a worse prognosis for glaucoma control and may require multiple surgical interventions.⁴⁻⁶ Both infantile-onset glaucoma associated with ocular and/or systemic abnormalities and glaucoma that develops after congenital cataract surgery have been noted to be less successful with goniotomy or trabeculotomy surgery, as compared to primary congenital glaucoma that presents after birth and before one year of age.^{4,7-8}

A success rate of 87% to 92% has been noted for 360-degree suture trabeculotomy in cases of primary congenital glaucoma presenting before one year of age, with significantly better results compared to goniotomy in one small, retrospective report.⁹⁻¹⁰ However, no study of the effect of 360-degree trabeculotomy on childhood glaucoma associated with a poor prognosis with traditional angle surgery has been published to date. We report the results of 360-degree trabeculotomy in the management of patients that fall into the following groups: (1) primary congenital glaucoma with birth-onset presentation accompanied by corneal clouding noted at birth, (2) primary congenital glaucoma with onset or presentation after one year of age, (3) primary congenital glaucoma – prior failed goniotomy surgery, (4) infantile onset glaucoma following congenital cataract surgery, and (5) infantile-onset glaucoma with associated ocular or systemic anomalies. As two previous publications address 360-degree trabeculotomy in primary congenital glaucoma presenting after birth and prior to one year of age⁹⁻¹⁰ (best prognosis), this category of primary congenital glaucoma was not evaluated in the present study.

Methods

A retrospective review of medical records identified all patients who were in the previously described groups and had either an attempted or completed 360-degree suture trabeculotomy between July 1989 and August 2003. Institutional review board approval at Emory School of Medicine was obtained for data review and analysis and the study conformed to the requirements of the United States Health Insurance Portability and Accountability Act. The diagnosis of glaucoma was confirmed by an examination under anesthesia prior to undergoing 360-degree suture trabeculotomy. During the period under study, 360-degree trabeculotomy was performed or attempted for all cases of childhood glaucoma where traditional goniotomy or trabeculotomy was indicated. The term *primary congenital glaucoma* (also known as *primary infantile glaucoma*) was used to refer to all cases with isolated trabeculodysgenesis and clinical findings of glaucoma and ocular enlargement, regardless of age of presentation.¹¹ Primary congenital glaucoma with a history of corneal clouding at birth was placed into Group 1. Primary congenital glaucoma that presented after 1 year of age was placed into Group 2. Primary congenital glaucoma (presentation at any age) that failed an initial goniotomy procedure met the criteria for Group 3. Glaucoma diagnosed in the first 3 years of life that developed following congenital cataract surgery was placed in Group 4. Glaucoma that developed in the first 3 years of life with associated ocular and/or systemic anomalies made up Group 5.

An initial examination under anesthesia was performed in all cases to determine suitability for trabeculotomy surgery. In cases with 3 or more clock hours of peripheral anterior synechiae, 360-degree suture trabeculotomy was not attempted; nor was it attempted in cases with iridocorneal adhesions. Intraocular pressure (IOP) measurement, gonioscopy, slit-lamp examination, A-scan biometry, corneal diameter measurement, and fundus

examination were performed. IOP measurements in this study were performed by handheld applanation tonometry in the office, or by pneumatonometry. (Medtronic Xomed Ophthalmics, Minneapolis, MN) during initial induction of general anesthesia. Central corneal thickness was not assessed during the period of this study.

In all cases a 360-degree suture trabeculotomy was attempted as described in an earlier publication,⁹ with the following variations in surgical technique. Over the period studied, a temporal scleral wound was used instead of a nasal scleral wound. In all cases the goal was passage of a single, curved, 6-0 polypropylene suture fragment over 360 degrees and location of the distal end in the original scleral incision. The operative reports of all cases were reviewed for details of the procedure. However, in cases where the initial suture could not be passed 360 degrees, a second 6-0 polypropylene suture was utilized to cannulate the canal in the opposite direction from the first (leaving the original suture in the canal as a stent), usually allowing 360-degree passage of the second suture. A Lester lens pusher (Katena Products, Inc, Denville, NJ) was utilized occasionally to retrieve the distal end of the suture from a viscoelastic-filled anterior chamber, in cases where dissection to locate the distal end caused premature trabeculotomy at the wound site. Sodium hyaluronate was left in the anterior chamber at the end of the 360-degree trabeculotomy to assist anterior chamber stability and reduce postoperative hyphema formation.

Postoperatively, prednisolone acetate 1% (Pred Forte, Allergan, Irvine, CA) eye drops 6-8 times/day were used initially (tapering over 4-6 weeks), along with antibiotic eye drops 4 times daily for approximately one week, and pilocarpine 2% (Bausch & Lomb, Rochester, NY) solution 4 times daily for approximately one month. Postoperative examinations were performed in the office on the first postoperative day, at 1 week, 1 month, and at 3- to 6-month intervals depending on the clinical situation. Subsequent examination under anesthesia was performed at least twice annually in children under 4 years of age or if adequate examination was not possible in older children.

Surgical success was defined as IOP was less than 22 mm Hg with allowance for glaucoma medical therapy (two consecutive IOP ≥ 22 mm Hg on maximal medical therapy for failure), and stable ocular dimensions and cup/disk ratio (ocular dimensions unchanged or within range of normal growth, cup/disk ratio within 0.1 of initial evaluation), and no further glaucoma surgery performed or recommended.

The cumulative probability of success versus time after surgery was calculated using the product-limit method with the 95% confidence interval determined using the log transformation. The time to failure was compared between groups using the logrank test.

Results

A total of 49 eyes of 36 patients were evaluated for inclusion into this study. In 4 eyes of 3 patients (2 with prior angle surgery, 2 with congenital ocular anomalies), 360-degree suture trabeculotomy could not be performed due to inability to successfully pass the suture, and Harm's trabeculotomes were used to perform a standard trabeculotomy procedure. In 3 additional eyes, a second scleral incision was used to retrieve the distal end of the suture (1 case with prior high peripheral anterior synechiae from cataract extraction, 1 case each of Lowe syndrome and port wine mark-related glaucoma), treating from 270 to 330 degrees of the angle. Suture trabeculotomy was successfully performed over 360 degrees from one scleral incision in 42 of 49 eyes (85.7%). The 4 eyes (3 patients) which had a standard trabeculotomy all failed and were excluded from statistical evaluation since a suture trabeculotomy was not performed.

Thirty-three eyes of 33 patients (first treated eye of each patient) were included for statistical evaluation in the study. The mean ($\pm$ standard deviation) of preoperative IOP was 34.3 ± 6.7 mm Hg (range, 23-51 mm Hg) on an average of 1.5 medications (range, 0-4). Mean final IOP (last follow-up or failure) was 22.2 ± 7.1 mm Hg (range, 13-41 mm Hg) on an average of 1.5 meds (range, 0-4). Median age at surgery was 7 months (interquartile range, 3-14 months; range, 0.25-92 months). The median follow-up or time to failure was 12 months (interquartile range, 6-42 months; range, 2-85 months). The number of medications used after surgery was: 0 (36%), 1 (15%), 2 (21%), 3 (21%), or 4 (6%). The overall the probability of success according to time after surgery (95% confidence interval) was 87% (69%-95%) at 6 months, 63% (41%-78%) at 1 year, and 58% (36%-74%) at 2 years, (Figure 1).

The results for each of the 5 patients groups are summarized in Tables 1 and 2. Although the sample sizes of each group were too small for formal statistical evaluation of the individual groups, the following observations were made: 360-degree suture trabeculotomy was most successful in Groups 1-4, with 13 of 17 eyes successful (77%), with 8 of 17 eyes (47%) on no medications, as compared to Group 5, with 7 of 16 eyes successful (44%) and 4 of 16 eyes (25%) on no medications (Tables 1, 2). Kaplan-Meier analysis of Groups 1-4 versus Group 5 failed to demonstrate a statistically significant difference ($p = 0.13$; Figure 2).

There were 16 eyes assigned to Group 5, the most varied group in terms of etiology. Two eyes had congenital anterior segment anomalies (one isolated sclerocornea, and one Peters anomaly), 4 had congenital glaucoma with systemic syndromes, 1 had congenital glaucoma associated with retinopathy of prematurity, 4 had concomitant iris anomalies (iris stromal abnormalities, ectropion uveae, poor pupillary reactivity), and 5 had port wine mark-related glaucoma. Of the 5 eyes with port wine mark-related glaucoma, 4 failed (time to failure, 11-42 months), with the 1 success having only 5 months of follow-up (lost to follow-up).

Transient hyphema lasting less than one week is commonly noted after trabeculotomy surgery¹¹ and was not considered a complication in this study. Of the 5 eyes with port wine mark-related glaucoma, 2 had a large (>50%), persistent postoperative hyphema and concurrent vitreous hemorrhage. One eye required 2 surgical interventions for the hyphema and vitreous hemorrhage due to amblyopia concerns. No cases of Descemet's tears, iridodialysis, persistent hypotony or subretinal suture passage were noted.^{12,13}

Visual acuity information was limited by the large number of preverbal children (median age at surgery, 6 months) included in the study. Limited follow-up was noted in some cases due to family relocation or long follow-up distance. Snellen or Allen visual acuity was available for 14 eyes following suture trabeculotomy, ranging from 20/20 to counting fingers. The case of port wine mark-related glaucoma that required two surgical interventions due to hyphema and vitreous hemorrhage, was noted to have 20/100 best-corrected vision with an early posterior subcapsular cataract.

Discussion

Successful control of primary congenital glaucoma by means of 360-degree suture trabeculotomy has been reported to be 87% to 92%^{9,10}; however, these reports almost exclusively pertain to cases of primary congenital glaucoma with the best prognosis for surgical success, namely, presentation after birth and before 1 year of age.¹¹ This retrospective study evaluated the results of 360-degree suture trabeculotomy in poor prognosis primary congenital glaucoma (birth onset or presentation after 1 year of age, prior failed goniotomy surgery, and glaucoma associated with congenital anomalies or cataract surgery).

Previous reports on surgical treatment of primary congenital glaucoma have noted excellent results with goniotomy or trabeculotomy procedures^{11,14}; however, some authors have noted a worse prognosis in primary congenital glaucoma associated with birth onset, presentations after 1 year of age, and prior angle surgery.²⁻⁶ Akimoto and colleagues⁵ noted a success probability of 60.3% with trabeculotomy in primary congenital glaucoma presenting before 2 months of age, 76.4% success with presentation after 2 years of age, as compared to 96.3% success with presentation from 2 months to 2 years of age.¹ Russell-Eggitt and colleagues² noted that eyes presenting at or near birth and those requiring more than one goniotomy procedure were more likely to fail treatment with goniotomy surgery.

In our study, 360-degree suture trabeculotomy had relatively good success (77%) in primary congenital glaucoma for all three groups (Table 1) but not quite as good as the 92% success noted with primary congenital glaucoma presenting after birth and before 1 year of age.¹⁰ However, the results were best with birth onset primary congenital glaucoma, Group 1 (3 of 3 eyes successful [100%], Table 1). Possible explanations for this discrepancy are the small sample size of Group 1, placement of birth-onset glaucoma with associated congenital iris anomalies into Group 5 (3 of 4 eyes failed, 25% success), or an enhanced effect of 360-degree suture trabeculotomy on this subgroup of primary congenital glaucoma. Walton⁵ reported a success rate of 15% using multiple goniotomies for 35 patients with newborn primary congenital glaucoma, but noted congenital iris anomalies in 89% of these patients, with 11 of 35 patients having CYP1B1 mutations consistent with primary congenital glaucoma. Iris hypoplasia associated with congenital glaucoma has been termed iridotrabeculodysgenesis by Hoskins and colleagues,¹⁶ as distinct from isolated trabeculodysgenesis (primary congenital glaucoma). Inclusion of the 4 eyes with congenital iris anomalies with birth onset presentation into the birth onset primary congenital glaucoma group would change the success rate to 60% (6 of 10 eyes). Regardless of how these eyes are classified, birth-onset glaucoma associated with congenital iris anomalies appears to portend a lower chance of success with trabeculotomy or goniotomy surgery.

There were 5 cases of congenital glaucoma with port wine mark-related glaucoma (Group 5). Management of glaucoma associated with Sturge-Weber is notable for its refractory nature and increased incidence of surgical complications such as choroidal detachment.^{7,17-18} In this study, no long-term success was noted in any of the 5 eyes treated with 360-degree suture trabeculotomy. The only significant postoperative complications in this study were 2 patients with port wine mark-related glaucoma who had persistent hyphema and concurrent vitreous hemorrhage. Although the sample size of treated eyes is small, 360-degree trabeculotomy appears to offer no advantage over traditional trabeculotomy or goniotomy surgery in port wine mark-related glaucoma, and may have a higher risk of hyphema complications.

Glaucoma is commonly noted following congenital cataract surgery, usually with an open-angle mechanism, and frequently requiring surgical intervention.¹⁹ Only a few reports of goniotomy or trabeculotomy surgery for this indication are noted in the literature, with very low success rates of 13% to 16%.^{7, 19} Trabeculectomy with mitomycin and tube-shunt procedures are more commonly used to treat aphakic glaucoma, with success rates of 25% to 72% but with the risk of serious complications, such as retinal detachment, hemorrhagic choroidal detachment, phthisis,²⁰⁻²¹ and guarded long-term success rates.¹⁹ The results of this study for infantile-onset glaucoma following congenital cataract surgery (3 of 4 eyes successful, no serious complications; Table 1) are encouraging, but must be tempered by the very small sample size. However, our findings are consistent with those of Bothun and colleagues,²² who noted 57% success with one or more goniotomy or trabeculotomy surgeries in 14 eyes of 11 children with aphakic glaucoma. Further evaluation of 360-degree suture trabeculotomy in patients with open-angle glaucoma following congenital cataract

surgery is warranted, given the frequently refractory nature of glaucoma in these children.
7-8,19

Surgical procedures other than goniotomy or trabeculotomy have been utilized for the initial treatment of primary congenital glaucoma and infantile onset glaucoma associated with ocular and systemic anomalies, most notably combined trabeculotomy and trabeculectomy. Mullaney²³ noted a success rate of 78% using combined trabeculotomy and trabeculotomy with mitomycin in 49 eyes with primary congenital glaucoma, as compared to a 45% success in 18 eyes with congenital glaucoma with associated anterior segment anomalies. Mullaney's study is consistent with our study, noting a reduced chance of successful control in the setting of associated ocular anomalies.

The limitations of this study are its retrospective nature, arbitrary categorization of patients into groups, and limited sample size. Although the best success rates were noted for patients with primary congenital glaucoma (birth onset, presentation after 1 year of age, prior goniotomy surgery) and infantile glaucoma after congenital cataract surgery, a statistically significant difference was not demonstrated between Groups 1-4 and Group 5; nevertheless, 360-degree suture trabeculotomy can be successfully performed in children with a wide range of ocular pathologies. The iScience iTrack catheter (Menlo Park, CA) is a new option for performing 360-degree trabeculotomy, with the advantage of a lighted, flexible catheter to assist with circumferential passage. Sarkisian²⁴ has demonstrated successful use of the illuminated microcatheter to perform 360-degree trabeculotomy in congenital glaucoma.

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FIG 1.
Cumulative probability of success for the 33 eyes in the study (solid line) with 95% confidence intervals (dashed lines).

FIG 2.
Cumulative probability of success of Groups 1-4 compared to Group 5 (dashed line).

Table 1

Summary of glaucoma groups

Group	No. eyes	Age at surgery (months) ^a	Follow-up duration (years) ^a	Pre-op IOP (mm Hg) ^b	Final IOP (mm Hg) ^b	Success n (%)
1 (primary congenital, birth onset)	3	1.5 (0.5, 2.0)	1.7 (0.8, 5.0)	35.3 ± 6.3	21.0 ± 6.0	3 (100%)
2 (primary congenital, onset after 1 year old)	6	38 (24.8, 79.3)	1.0 (0.4, 4.5)	43.0 ± 5.7	22.8 ± 9.5	4 (67%)
3 (primary congenital, failed goniotomy surgery)	4	7.0 (6.0, 12.5)	0.4 (0.2, 5.4)	25.8 ± 4.1	20.1 ± 9.8	3 (75%)
4 (infantile onset, post-congenital cataract surgery)	4	5.0 (3.0, 14.5)	1.6 (0.3, 3.7)	33.0 ± 7.2	21.8 ± 5.3	3 (75%)
5 (infantile onset, associated anomalies)	16	6.0 (1.1, 7.8)	1.0 (0.5, 3.1)	33.8 ± 5.0	22.9 ± 6.7	7 (44%)

Pre-op IOP, preoperative intraocular pressure^a median (25th, 75th percentiles)^b mean ± standard deviation

Table 2

Number of medications used in glaucomatous eyes after 360-degree suture trabeculotomy according to group

Group	No. eyes	0 n (%)	1 n (%)	2-4 n (%)
1 (primary congenital, birth onset)	3	3 (100%)	0	0
2 (primary congenital, onset after 1 year old)	6	1 (17%)	2 (33%)	3 (50%)
3 (primary congenital, failed goniotomy surgery)	4	2 (50%)	0	2 (50%)
4 (infantile onset, post-congenital cataract surgery)	4	2 (50%)	1 (25%)	1 (25%)
5 (infantile onset, associated anomalies)	16	4 (25%)	2 (13%)	10 (62%)