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Safety and Long-term Outcomes of Congenital Ptosis Surgery: A Population-Based Study

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Abstract

Purpose—To report the long-term outcomes of childhood ptosis surgery in a population-based setting over a 46-year period.

Methods—In this population-based cohort study, the medical records of all patients who were residents of Olmsted County, Minnesota, diagnosed as having blepharoptosis and having undergone surgical management prior to 19 years of age (between January 1, 1965, and December 31, 2010), were retrospectively reviewed. Age at time of surgery, type of surgery, duration of follow-up, number and nature of revisions, degree of amblyopia, and postoperative lagophthalmos and dry eye were documented.

Results—Forty-seven children meeting inclusion criteria underwent ptosis surgery. The median age at time of first surgery was 5.6 years (range: 1.5 to 17.7 years). Fifteen of 47 (31.9%) patients required a second procedure. Three of 47 (6.4%) patients underwent three procedures. The median time was 1.1 years (range: 0.03 to 7.8 years) between the first and second surgery and 6.0 years (range: 0.3 to 6.1 years) between the second and third procedure. Seven of 47 (14.9%) patients had amblyopia. Nineteen of 47 (40.4%) patients were noted to have lagophthalmos and 3 of 47 (6.4%) presented for symptomatic dry eye postoperatively.

Conclusions—In this population-based setting, more than half of children with ptosis required only a single surgical procedure, although a significant proportion required two procedures. Postoperative lagophthalmos is common, but symptomatic dry eye is rare.

INTRODUCTION

Surgical management of childhood ptosis is challenging because levator maldevelopment can lead to unpredictable results. Previous studies have reported on the surgical results of congenital ptosis surgery at tertiary referral centers or compared a variety of surgical techniques.[1–9] Long-term complications associated with surgical management of childhood ptosis in a population-based setting have not been reported.

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The purpose of our study was to evaluate the long-term outcomes of childhood ptosis surgery in a population-based cohort. We assessed reoperation rates, amblyopia, rate of lagophthalmos, dry eye, and other complications.

PATIENTS AND METHODS

With the approval of the Mayo Clinic Institutional Review Board, the medical records of all patients younger than 19 years who were residents of Olmsted County, Minnesota, and who underwent ptosis surgery between January 1, 1965, and December 31, 2010, were reviewed. The review was in accordance with the Heath Insurance Portability and Accountability Act (HIPPA). The cases were identified using the resources of the Rochester Epidemiology Project, a system designed to capture data on any patient–physician encounter in Olmsted County, Minnesota.[10] The population of this county is relatively isolated from other urban areas and effectively all childhood ptosis surgery and follow-up in the county is performed at Mayo Clinic or Olmsted Medical Center.

Inclusion criteria included all pediatric (younger than 19 years) patients with ptosis who were treated surgically and were residents of Olmsted County, Minnesota. Three patients with severe craniofacial trauma and 2 patients with an anophthalmic socket were excluded. Data abstracted from the medical record included dates and type of procedure performed, underlying etiology for ptosis, the patient's age at time of surgery, ocular comorbidities, amblyopia, strabismus, postoperative lagophthalmos at final follow-up, and any other complications.

RESULTS

Forty-seven residents of Olmsted County meeting the inclusion criteria underwent surgery for ptosis between January 1, 1965, and December 31, 2010. There were 28 boys and 19 girls. Surgery was performed on the right upper eyelid for 15, the left upper eyelid for 28, and both upper eyelids for 4 patients, for a total of 51 eyelids of 47 patients. The median age at the time of first surgery was 5.6 years (range: 1.5 to 17.8 years). The median time was 1.1 years (range: 0.03 to 7.8 years) between the first and second surgery and 6 years (range: 0.3 to 6.1 years) between the second and third procedure. No patients required more than three eyelid procedures. The median eye clinic follow-up duration was 3.3 years (range: 0.01 to 22.9 years). Of 47 patients, 29 had greater than 2 years' follow-up from initial surgery (median: 8.0 years; range: 2.0 to 22.9 years) and 18 patients had less than 2 years of follow-up (median: 0.27 years; range: 0.01 to 1.86 years). Of the 18 patients with less than 2 years of follow-up data available, the median duration of time between initial ptosis surgery and last known time residing in Olmsted County was 6.1 years (range: 0.9 to 20.1 years).

Surgical Indications

Forty-one of 47 patients had congenital ptosis, including one with bilateral hypoplastic optic nerves and one with Marcus Gunn jaw wink. One patient each had blepharochalasis, blepharophimosis, Crouzon syndrome, Noonan syndrome, congenital third nerve palsy, and acquired ptosis secondary to contact lens wear.

Six of 47 (12.7%) patients had ptosis surgery due to amblyopia and 2 for threatened amblyopia, and 1 was noted to be mildly amblyopic following successful ptosis surgery, for a total of 7 patients with amblyopia. Of the 6 patients with amblyopia diagnosed preoperatively, 4 had a margin reflex distance 1 (MRD1) of 0 and the other 2 had an MRD1 of 1 mm before surgery. The patient noted to have mild amblyopia in the left eye postoperatively had surgery at age 5 years with a preoperative MRD1 of 2 to 3 mm. Postoperatively, her eyelid position was documented as “2 mm of left upper eyelid ptosis.” Characteristics of the patients with amblyopia are highlighted in Table 1. Eight patients (17%) were documented to have strabismus: 2 with exotropia, 2 with esotropia, 1 with congenital third nerve palsy, 1 with congenital fourth nerve palsy, and 2 without documented details of the misalignment, 1 of which was a child with Noonan syndrome.

Initial Surgical Procedure

Initially 51 ptosis procedures were performed on 47 patients (Tables 2–3), with 37 eyelids undergoing levator resection procedures, 8 having frontalis suspension procedures, and 6 undergoing either Mueller muscle conjunctival resection or Fasanella-Servat procedures.

Reoperation With Less Than 2 Years of Follow-up

A total of 18 patients underwent ptosis surgery and had less than 2 years of postoperative data available. All had unilateral surgery. Their initial procedures and reoperations are summarized in Table 2. Two of 18 (11%) patients with less than 2 years of postoperative data required reoperation, 1 following levator resection and 1 following levator resection plus superior tarsectomy. Both of these patients underwent levator aponeurosis advancement, and at final follow-up both were noted to be slightly undercorrected but with good cosmesis.

Reoperation With Greater Than 2 Years of Follow-up

Reoperations for those with greater than 2 years of follow-up are summarized in Table 3. Overall, 14 of 33 (42.4%) eyelids, or 13 of 29 (44.8%) patients, required a reoperation. Seven of 13 (53.8%) eyelids that initially underwent levator aponeurosis resection required a second procedure. All seven revision procedures after levator resection involved additional levator surgery, from either an anterior or posterior approach. Five of 9 (55.6%) eyelids that initially underwent unilateral levator resection with superior tarsectomy required a second procedure. Only 1 of 4 (25%) patients (2 of 7 [28.6%] eyelids) who initially underwent frontalis sling placement required a second procedure; this was a patient with blepharophimosis whose initial procedure used autogenous fascia lata. The 6 eyelids that underwent Muller muscle conjunctival resection or Fasanella Servat procedures as the initial procedure did not undergo revision.

The indication for the second procedure was residual ptosis in 8 of 14 (57.1%), recurrence of ptosis in 5 of 14 (35.7%), and upper eyelid entropion for 1 of 14 (7.1%) eyelids. A third procedure was required by 3 of the initial 29 patients (10.3%) on 3 of the initial 33 eyelids (9.1%). The indications for the third procedure were residual ptosis (1 case), recurrence of ptosis (1 case), and ptosis overcorrection (1 case).

Surgical Specialty

Of procedures included in this review, all but 3 were performed by either an oculoplastic surgeon or pediatric ophthalmologist. The 3 not performed by an ophthalmologist were the initial and 2 revision levator aponeurosis resections on the same eyelid of a patient with Crouzon syndrome.

Complications

Following surgery, 19 of 47 (40.4%) patients, or 21 of 51 (41.2%) eyes, experienced lagophthalmos at their final visit: 15 of 37 (40.5%) eyelids following one or two levator resection procedures, 6 of 9 (66.7%) eyelids following a frontalis sling, either as the primary or secondary procedure, and none of the 6 patients undergoing Mueller muscle conjunctival resection or Fasanella-Servat procedures. Three of 47 (6.4%) patients presented for care related to symptomatic dry eye at any point during their follow-up. All three had lagophthalmos. Two patients with symptomatic dry eye had isolated congenital ptosis and both had undergone levator resection procedures. The first patient was 15 years of age at the time of bilateral levator aponeurosis advancement, which was followed by a repeat levator aponeurosis advancement for undercorrection. She presented for evaluation of dry eye symptoms 11 years later, at 26 years of age. The second patient was a 4-year-old girl initially overcorrected with a second levator aponeurosis advancement. Subsequent upper eyelid retraction repair 14 months later led to resolution of dry eye symptoms. The third patient who experienced symptomatic dry eye postoperatively had blepharophimosis and had undergone bilateral autogenous fascia lata frontalis sling procedures twice, at which point it was decided the achieved eyelid height was a compromise between normal eyelid position and the risk of exposure keratopathy. No patient was documented to have granuloma formation or extrusion of sling material. No other complications were documented and no serious corneal sequelae developed due to exposure.

DISCUSSION

During the 46-year period between January 1, 1965, and December 31, 2010, 47 Olmsted County children underwent surgery for blepharoptosis. Most required only a single surgical procedure. Slightly less than one-third underwent a second procedure and a small minority underwent a third revision. Most of the reoperations were performed for residual ptosis in the immediate postoperative period. Of the 6 patients who had preoperative amblyopia, all but 1 ultimately had visual acuity of 20/50 or better postoperatively. Despite high rates of postoperative lagophthalmos, patients in our series seldom experienced symptomatic dry eye, and there were no cases of serious corneal or other complications.

Our population-based study evaluating long-term outcomes of childhood ptosis surgery builds on Griepentrog et al.'s incidence study of childhood ptosis in Olmsted County.[11] Griepentrog et al.'s series found 107 children diagnosed as having childhood ptosis between January 1965 and December 2004, for an incidence of 7.9 of 100,000 children younger than 19 years. Although our inclusion criteria included children undergoing surgery through December 2010, only 5 of the 47 children received a diagnosis of ptosis after 2004. We

therefore estimate that slightly less than half of the children with ptosis in Olmsted County underwent surgical management over the course of this study.

The reoperation rate in our series, 16 of 51 (31.4%) eyelids in 15 of 47 (31.9%) patients, is somewhat higher than the 10% to 20.8% reoperation rate reported in previous case series.[1–3] This may reflect the continuity of care afforded in a population-based cohort in a relatively isolated medical setting and patient and surgeon factors that promote critical evaluation of surgical results and reoperation. We do not believe that this reoperation rate reflects surgeon inexperience. Although we report on only 47 eyelids undergoing surgical repair over 45 years, this sample does not reflect our total surgical volume. The cohort reported herein represents a small fraction of the total number of cases of congenital ptosis surgical procedures performed at Mayo Clinic during the study period because patients who did not reside in Olmsted County, Minnesota, at the time of surgery were excluded from this study. The exclusion of these cases minimized referral bias and bias from loss of follow-up. This methodologic design feature strengthens our study because it allows us to assess outcomes and complications in patients who might be excluded from a tertiary referral practice in which patients may not maintain long-term follow-up because of the time and cost associated with travel.

Reoperation rates as a function of upper eyelid excursion would have been interesting, but many older records excluded quantification of upper eyelid excursion. The highest reoperation rates of children with greater than 2 years of follow-up were among the levator aponeurosis resection, with slightly more than half undergoing revision. Of those undergoing levator resection and ultimately requiring revision, 12 had documentation of upper eyelid excursion, of whom 4 of 12 (33.3%) were less than 4 mm. This may reflect a reluctance to perform frontalis sling procedures as an initial procedure, due to physician or family concerns about cosmesis, asymmetry, or an effort to try a non-sling type procedure prior to proceeding with a sling. This may contribute to a higher revision rate. However, none of the patients requiring revision after initial levator aponeurosis surgery underwent subsequent frontalis sling procedure.

Our greater than 40% rate of postoperative lagophthalmos is higher than that described in previous series. Berry-Brincat and Willshaw reported that 20% of patients undergoing levator resection and 18% undergoing brow suspension experienced “lagophthalmos and superficial punctate erosions.”[3] It is unclear if the authors excluded patients with lagophthalmos without corneal changes. Our study included all patients with documented lagophthalmos, regardless of corneal status. It is possible that lagophthalmos resolved after last documentation or clinical follow-up in our patients.

The indication of ptosis surgery for 6 of 47 (12.7%) patients was amblyopia. This does not represent the incidence of amblyopia in all patients with congenital ptosis. Griepentrog et al. reported a 14.9% incidence (16 of 107 patients) of amblyopia in children with ptosis using the same population database but a different time frame.[12] The incidence of amblyopia in children with ptosis has previously been reported in a non-population-based setting to range from 17% to 48%.[13–21] The higher incidence in those series may reflect referral bias at tertiary care centers, and emphasizes the importance of population-based studies.

We acknowledge limitations to our series. Reliance on clinical records limited our ability to quantitatively assess the preoperative and postoperative eyelid position and function and the decision-making process leading to selection of a particular procedure. However, the goal of this study was not to report on surgical results defined by MRD1 or other eyelid height parameters. Another potential limitation is that some patients had a brief followup interval. Patients with shorter follow-up may differ from patients with longer follow-up. Specifically, patients with shorter length of follow-up may have more satisfactory outcomes and therefore no need for multiple postoperative visits. Alternatively, those with shorter length of follow-up may have unsatisfactory outcomes or low patient satisfaction and therefore seek care elsewhere. The latter is unlikely to happen regularly, given the lack of other subspecialty ophthalmology practices in proximity to the study area. Additionally, we addressed this limitation by reporting patients with shorter follow-up separately from those with longer follow-up, and found that those with longer follow-up had a higher reoperation rate. Finally, this was a population-based study rather than a cohort study. It is conceivable that some patients did leave Olmsted County for further care and eyelid surgery, and that subsequent care would not have been captured by our database.

The findings of this study provide population-based data regarding the outcomes and long-term complications of childhood ptosis surgery. Most patients required only a single procedure, although nearly one-third did undergo reoperation. Postoperative lagophthalmos was noted in nearly half of these children, whereas symptomatic dry eye was rare.

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TABLE 1

Visual Outcomes of Patients With Amblyopia Undergoing Childhood Ptosis Surgery

Preoperative VA	VA at Final Follow-up	Age at Time of Surgery (y)	Other Amblyopia Therapy
20/20[a]	20/25	5.3	Patching
20/20	20/20	13.0	Patching
20/30	20/30	5.0	None
20/200	20/200	3.6	Patching, high anisometropia
20/60	20/50	4.1	Patching, anisometropia
Central steady maintained	20/20	1.5	Patching
Fix and follow	20/30	2.5	Patching

VA = visual acuity

[a]Amblyopia diagnosed after surgery.

TABLE 2

Summary of Procedures and Reoperations for Patients With < 2 Years of Follow-Up Data

1st Procedure	No. of Patients	2nd Procedure	No. of Patients
Unilateral levator resection	8	Unilateral levator resection	1
Unilateral levator resection plus superior tarsectomy	7	Unilateral levator resection	1
Unilateral polypropylene sling	1		
Unilateral Fasanella-Servat	2		
Total	18		2

TABLE 3

Summary of Procedures and Reoperations for Patients With > 2 Years of Follow-up

1st Procedure	No. of Patients	2nd Procedure	No. of Patients	3rd Procedure	No. of Patients
Unilateral levator resection	11	Unilateral levator resection	4	Unilateral levator resection	2
		Unilateral levator resection plus superior tarsectomy	1		
		Unilateral Muller muscle conjunctival resection	1		
Bilateral levator resection	1	Unilateral levator resection	1		
Unilateral levator resection plus superior tarsectomy	9	Upper eyelid entropion repair	1		
		Banked fascia frontalis sling	1		
		Unilateral levator resection	3	Upper eyelid retraction repair	1
Bilateral autogenous fascia lata sling	1	Bilateral autogenous fascia lata sling	1		
Unilateral silicone sling	1				
Bilateral silicone sling	1				
Bilateral polypropylene sling	1				
Unilateral Muller muscle conjunctival resection	1				
Unilateral Fasanella-Servat	1				
Total	29		13		3