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ORIGINAL PAPER

Surgical intervention for paediatric blepharoptosis: a 6-year case series

Aaron Jamison · Ewan G. Kemp · Suzannah R. Drummond

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Abstract

Purpose To present our experience of paediatric blepharoptosis in a tertiary referral centre and evaluate the effectiveness of surgical intervention.

Methods A retrospective cohort study of all children receiving surgical blepharoptosis correction between 1/1/10 and 29/2/16. Children with pre-operative levator function (LF) $\geq$ 7 mm received levator resection, those with LF $\leq$ 4 mm received brow suspension, and in those children with LF of 5–6 mm, either levator resection or brow suspension was chosen depending on the degree of frontalis recruitment.

Results Ninety-five children (109 eyes, 64 boys) underwent blepharoptosis surgery within the study period. Mean (range) age at surgery was 5.9 (1.2–12.5) years. Seventy-nine (83.2%) had simple levator maldevelopment. Fifteen children were excluded due to inadequate follow-up. Of the remaining 80 children, 41 (51.2%) underwent levator resection, 27 (33.8%) underwent fascia lata brow suspension, and twelve (15.0%) underwent mersilene mesh brow suspension. Margin reflex distance-1 was greatest at 6-week follow-up with a small "lid drop" by 6-month follow-up in both the levator resection (0.9 mm pre-operatively, 3.1 mm at 6-week follow-up, 2.6 mm at 6-month follow-up) and fascia lata brow suspension (0.3 mm, 2.5 mm, 2.2 mm) groups. No immediate complications, and only two serious post-operative complications, were noted. One case of residual blepharoptosis was re-operated (fascia lata brow suspension).

Conclusions Surgical correction of paediatric blepharoptosis is safe and, after an observed lid drop between 6-week and 6-month follow-up (not seen in the mersilene mesh brow suspension group), effect appears to be maintained to 6 months and beyond. Readily accessible orthoptic assessment would help identify children at risk of amblyopia, both pre-operatively and post-operatively.

Keywords Paediatric · Blepharoptosis · Ptosis · Levator resection · Brow suspension

A. Jamison (✉) · E. G. Kemp · S. R. Drummond
Tennent Institute of Ophthalmology, Gartnavel General Hospital, 1053 Great Western Road, Glasgow G12 0YN, Scotland, UK
e-mail: Aaronjamison@gmail.com

E. G. Kemp · S. R. Drummond
Ophthalmology Department, Royal Hospital for Children, Glasgow, Scotland, UK

Introduction

Blepharoptosis can have a large cosmetic, functional and psychological impact on patients. The presentation of this condition in children is further complicated by the risk of poor visual development and amblyopia [1–5]. Further considerations in paediatric blepharoptosis include the inherent difficulty in examination and treatment of children, requiring the use of general

anaesthesia, occasionally in the former, and routinely in the latter.

Although a small collection of excellent papers report experiences in the assessment and surgical management of paediatric blepharoptosis, this cohort is still relatively underreported in the existing literature [6–10]. Several procedures to correct paediatric blepharoptosis exist, and a variety of techniques have been described for each, although no consensus has been reached as to which option is the best. By reporting our surgical outcomes, we hope to add to this ongoing conversation, and in particular, highlight our experience performing fascia lata brow suspension using a modified Fox's pentagon technique, rather than the more commonly performed, and reported, double triangle technique described by Crawford [11, 12].

We report our experience of paediatric blepharoptosis in a tertiary referral centre over a 6-year period and evaluate the effectiveness of our surgical intervention.

Materials and methods

We performed a retrospective case note review of children who underwent surgery for blepharoptosis between January 2010 and February 2016 at the Royal Hospital for Children (RHC), Glasgow.

Pre-operative data collected included details of the underlying aetiology, history of previous surgery, visual acuity and lid measurements. Visual acuity was measured using age-appropriate testing techniques (Snellen acuity where possible) by an orthoptist, but for the purposes of statistical analysis these measurements have been converted to LogMAR visual acuity measurements as described by Holladay [13]. Best-corrected visual acuity (BCVA) was used where available.

The palpebral aperture (PA) and upper margin reflex distance (MRD-1) were measured in distance fixation, while levator function (LF) was estimated based on the maximal upper eyelid excursion between downgaze and upgaze (with frontalis stabilisation). In very young children, measurements were made using a ruler, held vertically as close to the face as tolerated, and recording measurements in primary position, upgaze and downgaze with the use of lights or toys. In cases of poor examination compliance, these measurements were taken with the child's voluntary eye movements. The difficulty of obtaining reliable lid measurements in young children has been acknowledged in our discussion.

All of the operations were performed under general anaesthesia by one of two surgeons (EK, SD). Pre-operative LF measurements were the most important factor used to guide choice of surgery. In patients whose LF was borderline (5–6 mm) and the choice of surgery was between a large levator resection and frontalis brow suspension, MRD-1 played a role in decision-making with those patients with a 4-mm ptosis (MRD-1 of − 1 mm or less) receiving a brow suspension.

- LF $\geq$ 7 mm-levator resection,
- LF 5–6 mm-levator resection or brow suspension,
- LF $\leq$ 4 mm-brow suspension.

All cases of levator resection (LR) used an anterior approach as described by *Collin* [14]. Where appropriate, brow suspension with autogenous fascia lata (fascia lata brow suspension, FLBS) was performed using a Fox's pentagon technique [11]. In children too young to undergo fascia lata harvesting (i.e. those younger than four years old), brow suspension was achieved using mersilene mesh (mersilene mesh brow suspension, MMBS) with a modified Fox's pentagon technique as previously described by *Collin* [14, 15]. Details of any intra-operative complications were recorded.

Children undergoing blepharoptosis surgery at the RHC are routinely offered follow-up appointments at 6 weeks and at 6 months after their operation. Details of their surgical outcome, including visual acuity, repeat lid measurements, and any reported complications were recorded at these visits. Visual acuity was measured as part of an orthoptic assessment at each follow-up time point. Patients/parents were also asked at each follow-up whether they were or were not satisfied with their surgery, and their responses to this question were also recorded.

Statistical analysis of surgical outcomes has been reported at 6-week and 6-month post-operative time points. The visual acuity and lid measurements presented are those of the affected eye in unilateral cases. In bilateral cases, the measurements of both eyes are included in the analysis. Our analysis of pre- and post-operative visual acuity includes the presence of amblyopia, defined by a visual acuity of 6/12 or

worse in the absence of any ocular co-morbidity or improvement with refractive correction.

The National Health Service (NHS) Health Research Authority stated that research ethics committee (REC) approval was not required for this study.

Results

Ninety-five children (109 eyes) underwent blepharoptosis surgery during the 6-year study period, and of these, 64 (67.4%) were boys. The mean age at the time of operation was 5.9 (range 1.2–12.5) years. Seventy (73.7%) of the children were of Caucasian ethnicity, four (4.2%) were Asian and the ethnicity of the remaining patients had not been recorded.

The various underlying aetiologies are presented in Table 1. The majority of cases treated (85.3%) were myogenic in nature, most often due to simple levator maldevelopment (SLM). The male predominance is seen throughout the range of underlying pathologies. One patient had blepharoptosis secondary to congenital myasthenia gravis. They commenced pyridostigmine treatment in 2009, although this was stopped two years later due to poor response and the child underwent unilateral levator resection in 2013, aged six. Two patients had blepharophimosis syndrome. One underwent no other operations, while the other had received bilateral "Z-plasty" medial canthal revisions prior to their blepharoptosis surgery. One patient with third cranial nerve palsy secondary to a visual pathway pilocytic astrocytoma had undergone resection of this tumour five years prior to their blepharoptosis surgery.

Fifteen patients were excluded from further analysis due to inadequate follow-up. All of the 80 remaining cases (mean age 5.9 years, 56 male, 24 female) were included in subsequent analysis. Seventy children (87.5%) had unilateral blepharoptosis (34 right eye, 36 left eye), and the remaining ten children (12.5%) had bilateral involvement. Five children (6.3%—4 SLM, 1 Marcus Gunn) had previously undergone ipsilateral blepharoptosis surgery.

Of these 80 children, 41 (51.2%) underwent LR surgery (22 left, 19 right) (Table 2). The remaining 39 (48.8%) children underwent brow suspension surgery (27 with autogenous fascia lata, twelve with mersilene mesh). Thirty-eight of 39 brow suspension operations were bilateral, but one child who had previously undergone bilateral brow suspension surgery underwent unilateral (left) brow suspension surgery. Preoperative lid measurements (in millimetres) are summarised for each operation type in Table 2.

All 80 patients attended at least one follow-up appointment: 66 attended at 6 weeks post-operatively, 37 patients attended at 6 months, and only 23 patients attended both. Of the 43 patients that did not attend 6-month follow-up, 28 were discharged at the parent's

Table 1 Blepharoptosis aetiology of patient cohort

Blepharoptosis Classification	Number of patients (%)	Gender
Myogenic	81 (85.3%)	51 M:30 F
Simple levator maldevelopment (SLM)	79 (83.2%)	
Blepharophimosis	2 (2.1%)	
Neurogenic	7 (7.4%)	6 M:1 F
Marcus Gunn jaw-winking	5 (5.3%)	
Myasthenia gravis	1 (1.1%)	
Oculomotor nerve palsy	1 (1.1%)	
Mechanical	1 (1.1%)	1 M
Neurofibromatosis type 1	1 (1.1%)	
Aponeurotic	2 (2.1%)	2 M
Birth trauma	2 (2.1%)	
Syndrome-related	3 (3.2%)	2 M:1 F
Saethre–Chotzen	2 (2.1%)	
Foetal alcohol syndrome	1 (1.1%)	
Apparent	1 (1.1%)	1 M
Microphthalmia	1 (1.1%)	

Table 2 Patient cohort by operation type

	Levator resection	Fascia lata brow suspension	Mersilene mesh brow suspension
Number (%)	41 (51.2%)	27 (33.8%)	12 (15.0%)
Laterality	All unilateral (22 left, 19 right)	26 bilateral, 1 unilateral (1 left)	All bilateral
Age (years)			
Mean	6.1	7.2	2.1
Range	1.7–12.5	4.2–12.0	1.2–3.8
Pre-operative lid measurements (mm)			
Levator function (mean)	8.4	3.8*	2.5
Levator function (range)	2–15	1–6	0–6
MRD-1 (mean)	0.9	0.3	− 0.2
MRD-1 (range)	(− 1) to 2	(− 3) to 2	(−1) to 1

*Excluding one outlier, explained in text

request, one was discharged to their local ophthalmology department for further follow-up, ten did not attend their appointment and were lost to follow-up, and four were still awaited.

Levator resection (LR)

Forty-one LR operations were performed. The follow-up achieved for these patients is described in Table 3. A mean improvement in MRD-1 of 2.2 mm from baseline was seen at 6-week follow-up, although this positive effect reduced slightly (1.7 mm) by 6-month follow-up. All but four patients and/or parents were satisfied with the surgical outcome at 6 weeks, and by 6-month follow-up three of these patients had improved and were satisfied with their surgical outcome by the end of follow-up. The remaining patient went on to undergo FLBS for a residual blepharoptosis following LR (re-operation rate = 2.4%). No serious complications were noted (see Table 3).

Fascia lata brow suspension (FLBS)

Twenty-seven children underwent FLBS. A mean improvement in MRD-1 of 2.2 mm was noted at 6-week follow-up with a reduction of effect to 1.9 mm by 6 months. All but two patients and/or parents were satisfied with the surgical outcome at 6 weeks. By 6 months, both of these patients had improved and were satisfied with their outcome.

One patient developed a late recurrence of their blepharoptosis, 47 months post-operatively, although no further surgery has yet been planned. The child had undergone a brow suspension in another health board, six years prior to "redo" FLBS within our service. The cause of this second recurrence of blepharoptosis is difficult to identify due to poor clinical information relating to their initial surgery, and a long period without eye examination between their post-operative follow-up and their late recurrence 47 months post-surgery. Other than this patient, one patient post-operatively developed lagophthalmos and exposure keratopathy which required referral to the local corneal team for further management resulting in successful resolution. No other serious complications were noted. No re-operations were required in this patient cohort (re-operation rate = 0%).

Mersilene mesh brow suspension (MMBS)

Twelve patients underwent MMBS. A mean improvement in MRD-1 of 2.2 mm was noted at 6-week follow-up and 2.7 mm at 6 months. All patients and/or parents were satisfied with the surgical outcome at 6 weeks and at 6 months. One patient who underwent bilateral MMBS for a right blepharoptosis developed left-sided lagophthalmos and had their banding released. A further patient developed a mild late

Table 3 Post-operative details by operation type

	Levator resection	Fascia lata brow suspension	Mersilene mesh brow suspension
Attended follow-up (*n*)			
6 weeks	33	21	12
6 months	17	16	4
MRD-1 (mm)			
Pre-operative	0.9	0.3	− 0.2
6 weeks (change from baseline)	3.1 **(+2.2)**	2.5 **(+2.2)**	2.2 **(+2.4)**
6 months (change from baseline)	2.6 **(+1.7)**	2.2 **(+1.9)**	2.5 **(+2.7)**
Satisfactory eyelid contour?			
6 weeks	29 Yes, 0 No, 4 Not recorded	19 Yes, 0 No, 2 Not recorded	9 Yes, 0 No, 3 Not recorded
6 months	16 Yes, 0 No, 1 Not recorded	16 Yes, 0 No, 0 Not recorded	4 Yes, 0 No, 0 Not recorded
Patient/parents satisfied?			
6 weeks	29 Yes, 4 No	19 Yes, 2 No	12 Yes, 0 No
6 months	16 Yes, 1 No	15 Yes, 0 No	4 Yes, 0 No
Complications			
Immediate	None reported	None reported	None reported
During follow-up	3 mild lagophthalmos, 2 mild post-op swelling at 6 weeks (one resolved fully by 6 months, the other resolved by 6 months to reveal a residual ptosis)	2 mild lagophthalmos, 1 further lagophthalmos with exposure keratopathy (referred to local corneal team)	2 mild lagophthalmos
Late	1 residual ptosis (as above)	1 mild recurrence (no further surgery planned)	1 contralateral lagophthalmos (banding released), 1 mild recurrence (no further surgery planned)
Re-operation required?	1 × fascia lata brow suspension	None	None
Re-operation rate	*2.40%*	0%	0%

recurrence of their blepharoptosis, 26 month post-operatively, believed to be due to dehiscence of the non-autologous material. No further surgery has yet been planned. No other serious complications were noted. No re-operations were required in this patient cohort (re-operation rate = 0%).

Visual acuity

Visual acuity was generally maintained post-operatively (Table 4). Children receiving LR surgery appear to have a small mean reduction in LogMAR VA from 0.2 pre-operatively to 0.3 post-operatively, although when two patients who were noted to have significant drops in their VA (mentioned later) are removed, this effect is no longer apparent. Children receiving brow suspension operations had stable mean VA at 6 months.

Seven of the 41 children receiving LR had VA equal to or worse than 6/12 pre-operatively. One of these children had previously suffered a penetrating eye injury and had a best-corrected VA of counting fingers, but the six remaining children (14.6%) had no ocular co-morbidities and were assumed to be amblyopic. Four of these children remained 6/12 or worse following surgery, although two showed improvements to better than 6/12, both by first follow-up.

Two children (4.9%) who underwent LR developed VA of 6/12 or worse as a new finding post-operatively. These children were aged three and four at the time of

Table 4 Visual acuity (VA) and amblyopia

	All operations (n = 80)	Levator resection (n = 41)	Fascia lata brow suspension (n = 27)	Mersilene mesh brow suspension (n = 12)
LogMAR VA				
Pre-operative	0.2	0.2	0.1	0.6
6 weeks	0.2	0.2	0.2	0.2*
6 months	0.1	0.3	0.1	0.2*
Amblyopia				
(1) ≤ 6/12 pre-operatively (n)	**11 amblyopic**	**6 amblyopic** (+1 with ocular co-morbidity)	**1 amblyopic** (+1 with ocular co-morbidity)	**4 amblyopic**
Remained ≤ 6/12 post-operatively	*(6)*	*(4)*	*(0)*	*(2)*
Improved post-operatively	*(5)*	*(2)*	*(1)*	*(2)*
(2) ≤ 6/12 post-operatively only (n)	**4**	**2**	**2**	**0**

*Improvement in VA seen at 6-month follow-up is an artificial result due to the non-attendance of two patients with poor vision

operation. Neither of these patients had any reported complications following surgery, had any ocular co-morbidities or improved with best correction, and so both were diagnosed with amblyopia. Of all of the cases presented in this paper, these were the only two children to suffer a loss of vision of 0.2 LogMAR units or greater following surgery.

Two of the 27 children receiving FLBS had VA of 6/12 or worse pre-operatively. One of these children had microphthalmia associated with Peters anomaly and had a best-corrected vision limited to perception of light only, but the remaining patient (3.7%) had no ocular morbidities and was assumed to be amblyopic. This child improved from 6/12 pre-operatively to 6/9 post-operatively.

Two children (4.9%) who underwent FLBS developed VA of 6/12 or worse a new finding post-operatively. Neither of these children had any reported complications following surgery or improved with best correction. One of these children, aged twelve, had a long-standing blepharoptosis and had previously received a MMBS (aged three). The other child, aged eight, had Marcus Gunn jaw-winking but no other reported ocular co-morbidities. Both children were diagnosed with amblyopia.

Four of the 12 children (33.3%) receiving MMBS had VA of 6/12 or worse pre-operatively. All of these children had no ocular morbidities and were assumed to be amblyopic. Two of these children remained ≤ 6/12 following surgery, although two showed improvements to better than 6/12.

None of the children who underwent MMBS developed VA of 6/12 or worse as a new finding post-operatively.

Discussion

Our patient cohort shares a similar demographic to those found in the existing literature. In particular, we present a wide range of underlying aetiologies which highlight the different mechanisms at play and the care with which each individual case must be approached. Although many of the presently reported cases of blepharoptosis were congenital in nature, the average age at the time of operation (mean 5.9 years) suggests a delay before children are listed for surgery. The reasons for this are likely to include observation at a local ophthalmology department until referral to our tertiary referral centre was required, or delay in presentation until a time when a child's parents felt correction was most appropriate (such as prior to starting school) or when an older child is prompted by cosmetic and/or social concerns [6].

The choice of surgery is largely guided by the child's pre-operative levator function: LF ≥ 7 mm, levator resection; LF ≤ 4 mm, brow suspension. In those children with LF of 5-6 mm, either levator

resection or brow suspension was chosen depending on the degree of frontalis recruitment. If there is no frontalis recruitment on the ptotic side in a child with poor function ptosis, frontalis sling procedures become less predictable, and one may consider supramaximal levator resection in their place [16].

In this study, children undergoing LR had better pre-operative LF (mean = 8.4 mm) than those receiving brow suspension surgery (mean = 3.8 mm [FLBS] or 2.5 mm [MMBS]). Only three patients did not adhere to the suggested operation choices based on LF. Two children with pre-operative LF of 2 mm underwent LR, in both cases, due to the wishes of the parents. One child with good pre-operative LF (11 mm) underwent brow suspension surgery as this was a re-operation following failed LR. This case has been excluded from the summary statistics in Table 2, marked with an (*).

Children who required brow suspension surgery were allocated to the use of either fascia lata or mersilene mesh in a consistent and appropriate manner. FLBS was performed in children ranging from 4.2 to 12.0 years old (mean = 7.2 years). Mersilene mesh was adopted in those children under four years old (range 1.2–3.8 years, mean = 2.1 years), as they have usually not grown enough to allow successful harvesting of sufficient fascia lata from the thigh.

In the cases of both LR and FLBS, the maximal effect was seen at 6-week follow-up when considering MRD-1 as the primary outcome measure, with a lid drop noticed between 6-week and 6-month appointments. This effect has been previously postulated to correspond to the suture absorption rate [17]. The opposite trend was noticed in those who underwent MMBS, with a positive effect at 6-week follow-up increasing further by 6 months, although the reliability of this result is limited by low numbers of patients in this group attending their 6-month follow-up.

Eleven of the total 80 children (13.8%) were considered to be amblyopic pre-operatively; higher than the estimated 3% prevalence in the general population, but similar to that previously reported for children with blepharoptosis [3–5]. Following blepharoptosis surgery, five of these children improved to better than 6/12 in their affected eye. Four children were found to have a reduction in vision post-operatively that was felt to represent new amblyopia, given the absence of any complications, ocular co-morbidities or improvement with best correction. Given that blepharoptosis surgery can lead to refractive error changes and the onset of strabismus, both of which are potentially amblyogenic, post-operative orthoptic and optometric assessment would be highly desirable in every case [1–5, 10].

Three cases of lagophthalmos were noted in each of the three groups, contradicting previous suggestion that post-operative lagophthalmos is associated with the extent of levator complex resection [18]. Two of these cases were considered to be serious complications as they required onward referral or further surgical intervention (Table 4). No other serious complications were noted post-operatively. This suggests that surgical correction of blepharoptosis is a safe operation in the hands of our two specialist surgeons.

One patient who underwent LR (aged 2.8 years at the time of surgery) had a residual blepharoptosis post-operatively and required re-operation (LR re-operation rate = 2.4%). One mild recurrence occurred in each of the brow suspension groups (FLBS and MMBS), although neither progressed to further surgery during the follow-up period (FLBS and MMBS re-operation rates = 0%). These rates are considerably lower than those previously reported by Berry-Brincat et al. (19.89% overall), Lee et al. (13% overall), Whitehouse et al. (24.7% overall) and O'Reilly et al. (5.6%, FLBS only) [6, 7, 10, 19].

Our results are limited to some extent by incomplete follow-up. In many cases, this occurred as patients received their operation at our tertiary referral centre but were then transferred back to their referring hospital for follow-up, usually at the patient's request. Our follow-up data can be improved in future by sending referring hospitals a specific follow-up form to complete and send back when they review such patients.

The authors also note a degree of variability inherent in the measurement of lid position and levator function in young children who are less likely to fully comply with examination. Given that 21 of our 80 analysed patients were aged under four at the time of their operation, this factor is often relevant when planning surgery.

Surgical correction of paediatric blepharoptosis is safe and, after an observed lid drop between 6-week and 6-month follow-up (not seen in the MMBS group), effect appears to be maintained to 6 months and

beyond. Readily accessible orthoptic assessment would help identify children at risk of amblyopia, both pre-operatively and post-operatively.

Compliance with ethical standards

Conflict of interest The authors declare that they have no conflict of interest.

Ethical approval For this type of study, formal consent is not required.

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