ARTICLE

Lagophthalmos after congenital ptosis surgery: comparison between maximal levator resection and frontalis sling operation

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Abstract

Background/Objectives To compare postoperative lagophthalmos after maximal levator resection (MLR) and frontalis suspension (FS) in congenital ptosis patients with poor levator function (LF).

Methods A cross-sectional study was performed to compare postoperative outcomes in patients with preoperative LF ≤ 4 mm who had undergone MLR or FS at a single tertiary institution, and who had visited the outpatient clinic between February 2017 and August 2018. Main outcome measures were as follows: (1) Preoperative LF and margin reflex distance 1 (MRD1), (2) Postoperative MRD1, lagophthalmos and grade of superficial punctate keratopathy (SPK).

Results Our study comprised 152 eyelids of 122 patients. There were 71 eyelids in the MLR group and 81 eyelids in the FS group. The MLR group had comparable mean postoperative MRD1 (2.8 ± 0.8 mm) to the FS with autogenous fascia lata (AFL) group (3.0 ± 0.7 mm), while the FS with preserved fascia lata (PFL) group had the lowest mean postoperative MRD1 (2.2 ± 1.0 mm). The PFL group had significantly less lagophthalmos (0.6 ± 1.0 mm) than the AFL (1.9 ± 1.4 mm) and maximal levator resection (1.9 ± 1.7 mm) groups. In the MLR group, there was no significant difference in postoperative surgical measurements between the LF 0–2 mm group and LF 2.5–4 mm group in terms of exposure keratopathy, degree of lagophthalmos and MRD1.

Conclusion MLR is an effective alternative to FS in congenital ptosis patients with poor levator function, with the risk of postoperative lagophthalmos related to postoperative MRD1 rather than preoperative LF.

Introduction

The surgical repair of congenital ptosis is particularly challenging because of aesthetic and functional implications [1].

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Although not all children require surgical repair, the timely correction of paediatric ptosis is crucial in maintaining normal visual development and function [2, 3]. There are various controversies pertaining to congenital ptosis surgery, one of which is the ideal surgery for patients with poor levator function (LF).

A number of surgical treatments have been described with the traditional therapeutic algorithm as follows: In case of mild function defect of the levator muscle, a plication could be considered. Moderate deficiency requires a resection, while severe insufficiency of the levator should be treated by frontalis sling (FS) surgery [4–13]. Contrary to traditional recommendations, there have been several papers showing the effectiveness of maximal levator resection (MLR) for congenital ptosis patients with poor LF, with outcomes comparable to those of FS operations and advantages of improved cosmesis, more natural lid contour, no need for implantation of foreign materials, and avoidance of brow scars [14–18].

Lagophthalmos and exposure keratopathy are common concerns after any ptosis surgery. Commonly known risk factors for lagophthalmos after levator surgery are the

severity of the ptosis, LF, and degree of levator complex resection [19, 20]. Hence there is concern that MLR may carry a higher risk of lagophthalmos than FS surgery. However, to the best of our knowledge, there has been no study comparing the postoperative lagophthalmos between MLR or FS operation. Hence the purpose of our study was to evaluate and compare the postoperative lagophthalmos between the two surgeries in patients with severe congenital ptosis with poor LF.

Materials and methods

We performed a cross-sectional study on all patients with unilateral or bilateral simple congenital ptosis who had undergone MLR or FS operation with preoperative $LF \leq 4$ mm at a single tertiary institution, who visited the outpatient clinic between February 2017 and August 2018. Patients with neurogenic, traumatic, or aponeurotic ptosis were excluded, as were patients with less than 6 months of postoperative follow-up or with inadequate data. Patients with ptosis as part of a syndrome such as blepharophimosis or Marcus-Gunn jaw winking were also excluded. The study was approved by the hospital's Institutional Review Board (SMC 2019–11–213) and adhered to the tenets of the Declaration of Helsinki. Informed consent was obtained from all patients in the study.

Clinical data collected included patient demographics, previous eyelid surgeries, and follow-up duration. Preoperative measurements included margin reflex distance 1 (MRD1) and LF, while postoperative measurements included MRD1, degree of lagophthalmos and degree of keratopathy. Preoperative MRD1, preoperative LF and postoperative MRD1 were evaluated from medical records and facial photographs. Measurement of lagophthalmos was made using Image J software (National Institutes of Health, Bethesda, Maryland, USA) as follows: Patients were photographed with their eyes gently closed and jaw raised to ~30° (Fig. 1). An 8-mm diameter circular piece of sticker tape was placed in the centre of their glabella as a guide to measurement. Grading of keratopathy was based on the total area of superficial punctate keratopathy (SPK) after fluorescein staining as described by Miyata [21]: A0 = no punctate staining, A1 = less than one-third of cornea,

A2 = one-third to two thirds of cornea and A3 = greater than two-thirds of cornea.

Surgical technique

All surgeries were performed by a single surgeon (Y-DK). The surgeries were performed under general anaesthesia for children and local anaesthesia for adults. The MLR technique has been described in detail elsewhere [15, 17]. In summary: An upper eyelid incision was made at the desired crease line. A small strip of skin and orbicularis was then excised. The orbital septum was opened and the anterior surface of the levator aponeurosis was exposed to above the Whitnall's ligament. The levator aponeurosis and Müller's muscle were disinserted from the tarsal plate and dissection was carried out superiorly without transecting the medial and lateral horns. Three fixation sutures were placed to obtain the desired lid level and contour. The positioning of the upper lid margin was at the superior limbus in patients with general anaesthesia and 1 mm greater than the contralateral eyelid height in patients with local anaesthesia. This was followed by lid crease formation sutures followed by skin closure with continuous running sutures.

For patients undergoing FS surgery using autogenous fascia lata (AFL), the fascia lata was harvested using a fascia stripper with the surface landmark being the lower quarter of the line connecting the anterior iliac crest and the head of the fibula. The fascia lata was used in a Fox pentagonal configuration [22]. Two stab cuts were made 2 mm from the eyelid margin. Two additional stab cuts were made immediately above the eyebrow, and a third forehead cut was made between the 2 brow cuts to complete an isosceles triangle. Each end of the fascia strip was passed through the cuts and pulled up from the central forehead incision to obtain the desired eyelid height. The target final position of the eyelid margin was at the superior limbus. The overlapping fascia lata were then cross-stitched together and their ends buried and secured superiorly to the frontalis muscle. The procedure was similar in patients who underwent FS surgery using preserved fascia lata (PFL), except a homologous PFL (Tutoplast; Tutogen Medical GmbH, Neunkirchen am Brand, Germany) was used.

Postoperatively, artificial tears were prescribed every hour and lubricating ointment every 2 h. The administration

Fig. 1 Measurement of lagophthalmos from photographs. A patient 1 year after maximal levator resection on the left eye is photographed in primary position (**a**) and with their eyes gently closed and jaw raised to ~30° (**b**).
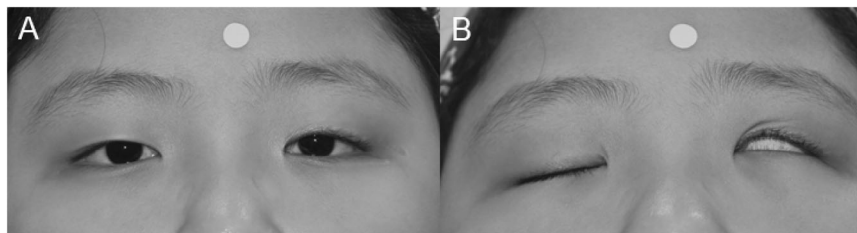
frequency was slowly tapered according to the lagophthalmos and the presence of SPK.

Statistical analysis

Statistical analysis was performed using SPSS software V.18.0 (SPSS, Chicago, IL, USA), where p values < 0.05 were regarded as statistically significant. One-way analysis of variance and Fisher's exact test were employed to analyse demographic data. Linear and generalised linear mixed model were employed to analyse eyelid measurements to compare the outcomes of each surgical procedure. Additionally, we examined the correlation of the degree of lagophthalmos with the LF and the postoperative MRD1 for the MLR group, and for the patients who underwent either type of FS surgery (AFL and PFL groups). The correlation of the degree of lagophthalmos with the LF and postoperative MRD1 were analysed using a linear mixed model.

Results

There were a total of 122 patients and 152 eyes. Table 1 shows the demographics of our study population. MLR was performed on 71 eyelids while FS operation was performed on 81 eyelids (35 AFL, 46 PFL). Overall mean age at operation was 67.7 months (5.6 years). Mean follow-up

duration was 81.2 months (6.8 years). In addition, 19 eyelids (12.5%) had undergone previous eyelid surgery (12 FS operation, 3 levator surgery, 3 unknown, 1 double lid formation).

Table 2 shows the preoperative and postoperative lid measurements for 152 eyes. There was no significant difference in terms of mean preoperative LF and MRD1 for both MLR and FS surgery groups. The mean postoperative MRD1 was significantly ($p = 0.05$) lower in eyelids, which had undergone FS surgery (2.5 ± 1.0 mm) than those which had undergone MLR (2.8 ± 0.8 mm). Mean postoperative lagophthalmos was also significantly less ($p < 0.01$) in eyelids which had undergone FS surgery (1.2 ± 1.5 mm) than those which had undergone MLR (1.9 ± 1.4 mm). The proportion of eyes with SPK was also significantly lower ($p = 0.05$) in FS surgery patients (26.9%) compared to MLR patients (43.4%).

A further analysis comparing postoperative MRD1 and lagophthalmos between MLR, FS with AFL and FS with PFL is shown in Table 3 and Fig. 2. While FS surgery overall had significantly lower mean postoperative MRD1 and lagophthalmos than MLR, a breakdown of FS surgery between AFL and PFL showed that the PFL group had

Table 1 Demographics of study population.

	Maximal levator resection	Frontalis sling operation	Total	p value

Number of patients (eyelids)	67 (71)	55 (81)
		• AFL
		  20 (35)
		• PFL
		  35 (46)
			122 (152)	

Age at operation, months				
• Mean ± SD	72.4 ± 78.7	63.6 ± 65.4	67.7 ± 71.8	0.45[a]
• Range	24–606	8–323	8–606	
Laterality, patients				
• Unilateral: Bilateral	63:4	29:26	92:30	
Gender, patients				
• Male: Female	43:24	32:23	75:47	0.50[b]
Follow-up duration, months				
• Mean ± SD	66.1 ± 51.5	94.5 ± 61.5	81.2 ± 58.6	<0.01[a]
• Range	7–249	6–282	6–282	

AFL autogenous fascia lata, *PFL* preserved fascia lata.

[a]Independent *t*-test.

[b]Chi-square test.

Table 2 Preoperative and postoperative lid measurements for maximal levator resection and frontalis sling surgery.

	Maximal levator resection ($n = 71$)	Frontalis sling surgery[a] ($n = 81$)	Total ($n = 152$)	p value
Preoperative LF, mm				
Mean ± SD	2.2 ± 1.2	2.0 ± 1.3	2.1 ± 1.2	0.44[b]
Preoperative MRD1, mm				
Mean ± SD	−0.1 ± 1.0	0.0 ± 1.2	0.0 ± 1.1	0.56[b]
Postoperative MRD1, mm				
Mean ± SD	2.8 ± 0.8	2.5 ± 1.0	2.7 ± 0.9	0.05[b]
Lagophthalmos, mm				
Mean ± SD	1.9 ± 1.4	1.2 ± 1.5	1.5 ± 1.5	<0.01[b]
Grade of SPK (%)				
A0	40 (58.0%)	58 (74.4%)	98 (66.7%)	
A1	27 (39.1%)	20 (25.6%)	47 (32.0%)	
A2	2 (2.9%)	0	2 (1.4%)	
A3	0	0	0	0.05[c]
NA	2	3	5	
Presence of corneal opacity	1 (1.4%)	1 (1.3%)	2 (1.4%)	

LF levator function, *MRD1* margin reflex distance 1, *SPK* superficial punctate keratopathy.

[a]Includes both frontalis sling with autogenous fascia lata (AFL) and preserved fascia lata (PFL).

[b]Independent *t*-test.

[c]Chi-square test.

Table 3 Preoperative and postoperative lid measurements for maximal levator resection, frontalis sling with autogenous fascia lata and frontalis sling with preserved fascia lata.

	Maximal levator resection (n = 71)	Autogenous fascia lata (n = 35)	Preserved fascia lata (n = 46)	p value
Preoperative LF, mm				
Mean ± SD	2.2 ± 1.2	1.9 ± 1.4	2.2 ± 1.1	0.60[a]
Preoperative MRD1, mm				
Mean ± SD	−0.1 ± 1.0	0.0 ± 1.0	0.0 ± 1.3	0.84[b]
Postoperative MRD1, mm				
Mean ± SD	2.8 ± 0.8	3.0 ± 0.7	2.2 ± 1.0	<0.01[b]
Lagophthalmos, mm				
Mean ± SD	1.9 ± 1.4	1.9 ± 1.7	0.6 ± 1.0	<0.01[b]
Grade of SPK (%)				
A0	40 (58.0%)	24 (68.5%)	34 (79.1%)	
A1	27 (39.1%)	11 (31.4%)	9 (20.1%)	
A2	2 (2.9%)	0	0	0.14[c]
A3	0	0	0	
NA	1	0	2	
Presence of corneal opacity	1(1.4%)	0	1 (2.3%)	

LF levator function, *MRD1* margin reflex distance 1, *SPK* superficial punctate keratopathy.

[a]Kruskall–Wallis test.

[b]One-way analysis of variance.

[c]Linear by linear association.
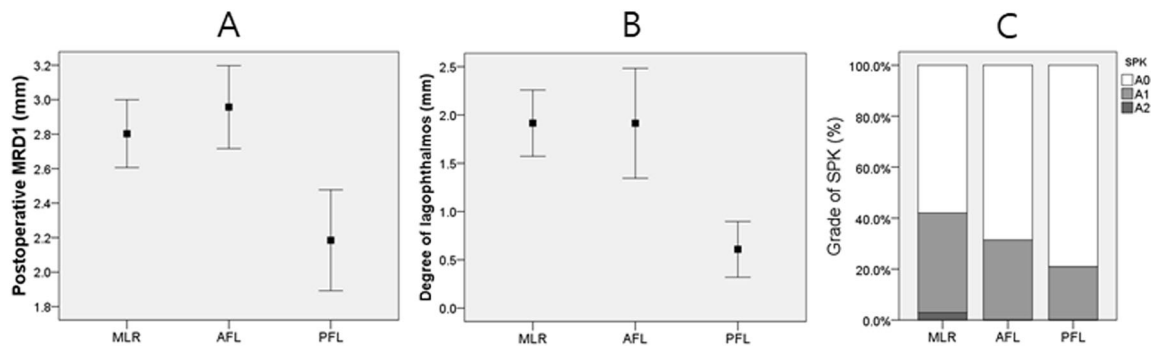
Fig. 2 Comparison of MRD, lagophthalmos and SPK. Comparison of postoperative margin reflex distance 1 (MRD1) (**a**), degree of lagophthalmos (**b**), and grade of superficial punctate keratopathy (SPK) (**c**) in each surgical group.

significantly lower mean postoperative MRD1 (2.2 ± 1.0 mm) than the MLR and AFL group, while the AFL group had a comparable mean postoperative MRD1 (3.0 ± 0.7 mm) to the MLR group (2.8 ± 0.8 mm). Similarly, the PFL group had significantly less lagophthalmos (0.6 ± 1.0 mm) than the AFL (1.9 ± 1.4 mm) and MLR (1.9 ± 1.7 mm) groups. The proportion of patients with keratopathy was highest in the MLR group (42.3%), followed by the AFL group (31.4%) and lastly the PFL group (21.7%), although the keratopathy was mostly mild (A1: SPKs less than one third of the cornea).

Table 4 compares the preoperative measurements and postoperative outcomes of eyelids treated with MLR divided according to the LF preoperatively: 0–2 or 2.5–4 mm.

The preoperative LF and MRD1 was significantly different between the two groups, with the LF 0–2 mm group having significantly lower LF (1.3 ± 0.7 mm) and MRD1 (−0.3 ± 1.1 mm) than the LF 2.5–4 mm group (3.4 ± 0.5 and 0.2 ± 0.9 mm, respectively). However, there was no significant difference in postoperative surgical measurements between the LF 0–2 mm group and LF 2.5–4 mm group in terms of exposure keratopathy, degree of lagophthalmos and MRD1.

Discussion

Our study compared MLR and FS surgery for congenital ptosis with poor LF (<4 mm) and found that MLR and FS

Table 4 Comparison of preoperative measurements and surgical outcomes in patients treated with maximal levator resection.

Maximal levator resection (n = 71)	Levator function 0–2 mm	Levator function 2.5–4 mm	p value
No. of eyelids (patients)	41	30	
Age at operation, months			
Mean ± SD	74.2 ± 94.5	69.9 ± 51.4	0.82[a]
Preoperative measurements			
LF, mm	1.3 ± 0.7	3.4 ± 0.5	<0.01[a]
MRD1, mm	−0.3 ± 1.1	0.2 ± 0.9	0.02[a]
Postoperative measurements			
Exposure keratopathy	13(33.3%)	16(53.3%)	0.10[b]
Degree of lagophthalmos, mm	1.9 ± 1.4	1.9 ± 1.5	0.94[a]
MRD1, mm	2.7 ± 0.9	2.9 ± 0.7	0.26[a]
Follow-up duration, months			
Mean ± SD	61.0 ± 49.5	73.2 ± 54.2	0.33[a]

SD standard deviation, *LF* levator function, *MRD1* margin reflex distance 1.

[a]Independent *t*-test.

[b]Chi-square test.

with AFL resulted in similar postoperative lagophthalmos and MRD1. FS with PFL, however, resulted in significantly less lagophthalmos and a lower MRD1 than both MLR and FS with AFL.

Poor LF (<4 mm) is a hallmark of typical myogenic congenital ptosis whereby a proposed mechanism is anomalous innervation of the muscle during embryogenesis resulting in levator muscle dysgenesis [23]. Although generally nonprogressive, ptosis may be associated with abnormal visual development and function [24]. Surgical repair is indicated in instances where the upper eyelid interferes with the visual axis, resulting in stimulus deprivation or astigmatism that is amblyogenic, or when the ptosis results in cosmetic disfigurement [25]. The best-suited surgical approach is primarily based on LF and degree of ptosis. For poor LF of 4 mm or less, the FS is the most commonly used method [2]. However, the use of MLR for congenital ptosis patients with poor LF has also been described with good results [15, 18]. Nonetheless, a concern remains that in the setting of poor LF, MLR should be avoided because of the risk of overcorrection, lagophthalmos and corneal exposure [23].

To our knowledge, there has been no study comparing the postoperative lagophthalmos and keratopathy incidence between the two surgeries for congenital ptosis with poor LF. A criticism of the MLR is that when large sections of the levator palpebrae superioris are resected, the elastic properties of the eyelid are severely impaired [14]. Downward saccadic movements are restricted, which provokes

eyelid lag and lagophthalmos. Our study showed that MLR for congenital ptosis with poor LF had comparable mean lagophthalmos and rate of exposure keratopathy to the FS with AFL group. While the FS with PFL group had lower mean lagophthalmos and rate of keratopathy than the other two groups, this correlated with lower mean postoperative MRD1 in the PFL group.

While patients who underwent MLR tended to have more SPKs than those who underwent FS surgery, it was not statistically significant different. The keratopathy that was observed in the MLR group was not progressive on postoperative follow-up and all improved with lubrication, including the patient with corneal opacity. A possible explanation for lower SPK rates in patients undergoing FS is the movement of their eyebrows during eyelid closure. The connection between the tarsal plate and frontalis muscle made during the FS surgery makes this muscle responsible for eyelid relaxation upon downgaze. Baccega et al. [26]. reported that brow motion accounted for 45.2% of lid descent in the patients who underwent FS surgery, while only for 3.3% in the controls, which may explain why exposure keratopathy after FS surgery seems to be easily compensated by lowering of the eyebrows. In contrast, the main source of the upper lid movement that accompanies downward saccadic movement of the eye in normal subjects or patients who have undergone MLR is the relaxation of the levator palpebrae superioris muscle and orbicularis oculi contraction, without the compensatory movement of the eyebrows [27]. Nonetheless, development of SPK is multifactorial and further studies may be necessary to determine the exact influence by each type of surgery.

Frontalis suspension has been frequently recommended for congenital ptosis with poor LF, although the use of AFL subjects the patient to donor-site morbidity and is difficult to harvest in infants, and the possibility of permanent thigh scars makes it an unattractive choice to some parents [15, 28, 29]. Alternative substances are readily available and can shorten operative times, but have also been associated with higher rates of infection, graft exposure, rejection and granuloma formation [15]. Unsatisfactory results can also be encountered in patients with unreliable brow function [30, 31].

Less commonly, MLR has been described to be an effective option for ptosis with poor LF. Levator resection offers the advantages of preserving the dynamic blink, giving a more natural contour to the eyelid, allowing more symmetry between the two eyes in cases of unilateral ptosis, and also reduces the reliance on the frontalis muscle action to aid eyelid elevation [32]. Our study shows that MLR gives excellent surgical outcomes comparable to that of FS operation with AFL and superior to that of PFL, adding to the increasing literature on the role of levator resection in congenital severe ptosis. Epstein and Putterman [33]

achieved 75% acceptable cosmetic results in eight patients with severe unilateral ptosis. Mauriello et al. [30] reported excellent results in 87.5% of 32 patients with levator excursion of 2 mm or less. Press and Hubner [18] reported 36 of 44 cases (81.8%) that underwent levator resection and had successful lid levels in moderate-to-severe congenital ptosis with less than 2 mm of LF. In addition, Cruz et al. [14] showed MLR to effectively reduce the positional asymmetry between eyelids in unilateral congenital ptosis. Mete et al. [16] performed MLR with preservation of the horns of the Whitnall's ligament and obtained successful results in 14 patients (71.43%) and satisfactory results in two patients (21.42%). Lee et al [15]. in a recent publication concluded that MLR is an effective procedure in congenital ptosis, even in patients with poor LF of 0–2 mm, with satisfactory results (excellent or good result) obtained in 93% of the patients.

To date, the impact of LF on postoperative outcome in levator resection appears to be uncertain [34, 35]. A concern that many surgeons have performing MLR on patients with poor LF is a risk of lagophthalmos and resulting exposure keratopathy. Our study showed that the degree of post-operative lagophthalmos in MLR was related to post-operative MRD1 rather than preoperative LF. Comparison of patients with preoperative LF of 0–2 or 2.5–4 mm (Table 4) showed no significant difference in postoperative surgical measurements between the two groups in terms of exposure keratopathy, degree of lagophthalmos and MRD1. Our findings also contradict previous reports that a poorer LF is associated with undercorrection [12, 36], but reaffirms Goncu et al.'s [37] findings that levator resection surgery is an effective treatment for congenital ptosis, including severe ptosis with poor LF. A possible explanation for the latter is a significant and permanent improvement in postoperative LF, which might have an additive effect on surgical success, especially for those with poor LF. The mechanical improvement observed may be provided by excision of dystrophic tissue, shortening of muscle, and relieving some abnormal dystrophic attachments of the levator complex [37].

There were a few limitations in our study. First, this was a cross-sectional non-randomised study with its inherent biases. There were patients with less than 6 months of follow-up who were excluded from our analysis. Nonetheless, the number of patients included in our study is a respectable number compared to other studies on congenital ptosis with poor LF. Information on the amount of resection of levator complex was not collected due to inaccuracy in measuring the distance of levator aponeurosis dissected from the superior border of the tarsal plate in view of its contractile nature and the authors' belief that quantification of levator resection may not be crucial to the surgical success in such patients. We also did not consider the influence of Bell's phenomenon on the rate of exposure keratopathy, as it was difficult to accurately assess it in all patients especially the very young children.

In conclusion, our study has shown that MLR and FS with AFL have comparable surgical outcomes in terms of postoperative MRD1, degree of lagophthalmos and risk of exposure keratopathy. Frontalis suspension with PFL appears to have the lowest risk of postoperative lagophthalmos and exposure keratopathy among the three groups, but is also associated with the lowest postoperative MRD1. Our results support the assertion that MLR is an effective alternative to FS in congenital ptosis patients with poor LF, with the risk of postoperative lagophthalmos related to postoperative MRD1 rather than preoperative LF.

Summary

What was known before

- Traditionally, the ideal surgery for congenital ptosis with poor levator function is frontalis sling (FS).
- Recently there have been several papers showing the effectiveness of maximal levator resection (MLR).
- There has been no study comparing the postoperative outcomes between MLR or FS operation.

What this study adds

- Our study found that MLR and FS with autogenous fascia lata (AFL) have comparable surgical outcomes in terms of postoperative lid height, degree of lagophthalmos and risk of exposure keratopathy.
- Frontalis suspension with preserved fascia lata (PFL) appears to have the lowest risk of postoperative lagophthalmos and exposure keratopathy among the three groups, but is also associated with the lowest postoperative lid height.
- Our results support the assertion that MLR is an effective alternative to FS in congenital ptosis patients with poor LF, with the risk of postoperative lagophthalmos related to postoperative lid height rather than preoperative LF.

Compliance with ethical standards

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