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CLINICAL INVESTIGATION

Sex-specific difference in age distribution of congenital lower eyelid epiblepharon in a Japanese population

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Abstract

Purpose To examine the sex-specific differences in age distribution of Japanese patients with congenital lower eyelid epiblepharon.

Study design A retrospective, observational study.

Methods A total of 291 patients (87 men, 204 women) who underwent modified Hotz for correction of congenital lower eyelid epiblepharon were included in this study. Data on sex, age at surgical intervention, and affected side were collected. Patients' age was classified according to the orbital growth pattern (0-3 years, 4-6 years, 7-12 years, 13-15 years, and ≥ 16 years). The sex-specific difference in age distribution was analysed using Pearson's chi-squared test.

Results Among male patients, 75 (86.2%) underwent surgery during 4-12 years of age. Only 2 (2.3%) boys had surgery at 13-15 years and 1 had it while he was ≥ 16 years. Among female patients, 11 (5.4%) underwent surgery at 13-15 years and 59 (28.9%) at ≥ 16 years. The age distribution was significantly different between sexes ($P < 0.001$).

Conclusion Among patients aged ≥13 years, there were more women compared to men. This implies that congenital lower eyelid epiblepharon is more persistent in females and may reflect slower and less orbital growth from 13 years of age and onwards in females. The results suggest that as long as there are no severe corneal complications and no risk for amblyopia, conservative management may be a good option for males since spontaneous resolution is likely to occur. However, for females aged 13 years or more, the threshold for surgical intervention should be lower.

Keywords congenital lower eyelid epiblepharon · orbital growth · sex-specific difference · age distribution · management

Introduction

Congenital lower eyelid epiblepharon (congenital epiblepharon) is a common eyelid condition among East-Asians wherein the cilia of the lower eyelid are directed upward and inward towards the cornea [1–6]. The weak tarsal attachment of the pretarsal orbicularis oculi muscle and the skin and the failure of the anterior layer of lower eyelid retractors to reach the skin are the main pathophysiologies [4–9]. This condition is frequently observed in patients with a narrow interpupillary distance, epicanthus, fullness around the buccal area, and relatively prominent eyelids [1, 2, 5, 6]. Congenital epiblepharon requires treatment when the lashes rub against the cornea resulting in ocular irritation or corneal erosion [1, 4, 5, 8–10].

Congenital epiblepharon becomes less pronounced with age and mostly resolves within one to two years [1]. However, the condition does not always resolve in all patients and some still retain the condition up to high school [10–12].

Growth of the orbital or facial bones is the main factor influencing spontaneous resolution of congenital epiblepharon [10, 13], and this is markedly different between sexes. This is thought to accompany skin and muscle extension, resulting in spontaneous resolution [10]. A recent study performed in East-Asians shows that the development of the orbit continues until 15 years with a biphasic growth pattern and is greater in boys from 13 years of age [14]. Another previous study in the Japanese also reports that

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periorbital growth usually stops at age 11 in girls [15]. These revelations lead us to a hypothesis that the number of male patients who required surgical intervention for congenital epiblepharon aged 13 years or more is lower than that of female patients aged 13 years or more. Thus, we examined the sex-specific difference in age distribution of patients who underwent surgery for congenital epiblepharon.

Materials and methods

Institutional Review Board (IRB)/Ethics Committee approval was obtained from the IRB of Aichi Medical University Hospital; the study followed the tenets of the Declaration of Helsinki and its amendments. Personal identifiers were removed from the records prior to data analysis.

All Japanese patients who underwent modified Hotz procedure [16] for correction of congenital lower eyelid epiblepharon were reviewed by 2 authors (YT and HK) or by registered doctors supervised by 1 author (YT) between April 2009 and September 2018 at Aichi Medical University Hospital. Surgery was suggested to all patients who were observed to have significant astigmatism or corneal erosions that were not relieved by medical management. We excluded patients with any history of lower eyelid surgery for correction of congenital epiblepharon at other clinics. We also excluded patients with other congenital diseases, such as congenital upper eyelid epiblepharon, congenital ptosis, and congenital nasolacrimal duct obstruction, because these conditions may influence decision-making and lower the threshold for surgical correction.

Data on sex, age at surgical intervention, affected side, and presence or absence of corneal opacity were collected. We classified patients' age according to the orbital growth reported previously: 0-3 years, 4-6 years, 7-12 years, 13-15 years, and ≥ 16 years [14]. The sex-related difference in the distribution of patients' age was analysed using Pearson's chi-squared test. The percentage of patients with unilateral epiblepharon was compared among the age groups using Pearson's chi-squared test in each sex group. All statistical analyses were performed using SPSS™ ver. 22 software (IBM Japan). A P-value of < 0.05 was considered statistically significant.

Results

A total number of 367 patients with congenital lower eyelid epiblepharon were reviewed but we excluded 76 patients (history of lower eyelid surgery, 29 patients; congenital upper eyelid epiblepharon, 42 patients; congenital ptosis, 5 patients; congenital nasolacrimal duct obstruction, 4 patients; 4 patients had two of these conditions); 291 patients (87 men and 204 women; age range, 0 to 47 years) met the inclusion criteria for this study.

The distribution of patients' age in each sex group is shown in Table 1. Among the men, 75 patients (86.2%) underwent surgery within the age range of 4-12 years. Only 2 (2.3%) required surgery at 13-15 years and 1 patient (18-year-old, 1.1%) at ≥ 16 years. On the other hand, although 116 of 204 women (56.9%) underwent surgery within the age range of 4-12 years, 11 (5.4%) required surgery at 13-15 years and 59 patients (28.9%) at ≥ 16 years. The distribution was significantly different between sexes ($P < 0.001$).

The percentage of patients with unilateral epiblepharon in each sex group is shown in Table 2. Among male patients, although none of the patients aged >13 years showed unilateral epiblepharon, the percentage of patients who underwent surgery for unilateral epiblepharon was largest in the age

Table 1 The distribution of patients who underwent surgery for congenital lower eyelid epiblepharon according to age and sex

	0-3 years	4-6 years	7-12 years	13-15 years	≥ 16 years	Total	*P* value
Male (%)	9 (10.3)	39 (44.8)	36 (41.4)	2 (2.3)	1 (1.1)	87	< 0.001
Female (%)	18 (8.8)	53 (26.0)	63 (30.9)	11 (5.4)	59 (28.9)	204	

Table 2 The distribution and percentage of patients who had surgery for congenital lower eyelid epiblepharon according to laterality, age and sex

Male	0-3 years	4-6 years	7-12 years	13-15 years	≥ 16 years	*P* value
Bilateral (%)	8 (88.9)	37 (94.9)	27 (75.0)	2 (100)	1 (100)	0.147
Unilateral (%)	1 (11.1)	2 (5.1)	9 (25.0)	0	0	
Total	9	39	36	2	1	
Female	0-3 years	4-6 years	7-12 years	13-15 years	≥ 16 years	*P* value
Bilateral (%)	15 (83.3)	47 (88.7)	54 (85.7)	7 (63.6)	41 (69.5)	0.041
Unilateral (%)	3 (16.7)	6 (11.3)	9 (14.3)	4 (36.4)	18 (30.5)	
Total	18	53	63	11	59	

group of 7-12 years. Among female patients, the percentage of patients who required surgery for unilateral epiblepharon was noted to be highest after 13-15 years of age. Although the difference in the distribution of male patients with unilateral epiblepharon did not reach statistical significance (P = 0.147), that of female patients was statistically significant ($P = 0.041$).

Discussion

We reported the distribution of patients who underwent surgery for congenital epiblepharon according to age and sex. Only 2 (2.3%) male patients underwent surgery at 13-15 years and 1 (1.1%) at >16 years; on the other hand, there were 11 (5.4%) female patients who underwent surgery at 13-15 years and 59 (28.9%) at >16 years. The disparity between the male and female population of our study coincides with Wei's and Furuta's studies, which reported that periorbital growth usually slows down from age 13 or stops at age 11 in females, resulting in smaller orbital volumes in females compared to males at age >13 years [14, 15]. These findings of Wei and Furuta might explain why in our study, more female patients who are >13 years of age, were noted to have congenital epiblepharon compared to males.

Our results also show that majority of boys required surgery for unilateral congenital epiblepharon at 7-12 years of age compared to the younger age group. On the other hand, majority of girls required surgery for unilateral congenital epiblepharon by age 15. A previous study demonstrated that 21% of patients with epiblepharon were unilateral cases [11]; in our experience, we also encounter patients with bilateral epiblepharon in whom the severity of epiblepharon differs between sides. These findings also suggest that congenital epiblepharon on a milder side is more likely to resolve as patients grow older based on the orbital growth pattern [14, 15].

From these findings, we believe that thresholds for management of epiblepharon should be different for boys and girls. Threshold for surgical intervention could be set lower for girls aged >13 since the chance of spontaneous resolution is low and they are more likely to have complications. On the other hand, it is probably prudent to be more conservative in managing boys with epiblepharon, if they don't have severe corneal damage and if the risk of amblyopia is low, since spontaneous resolution can be expected and complications are less likely to occur.

Our results indicate that majority of patients underwent surgery for repair of congenital epiblepharon at the age range of 4-12 years. Similar to our findings, Kim et al.'s study on Korean children with congenital epiblepharon reveals that the mean patient age at time of surgery was 6.95 ± 2.52 years [10], which coincides with the 4-12 year age group.

A previous study performed on Japanese children reports that 13 of 683 boys aged 16 to 18 years (1.9%) had epiblepharon [11]. That study showed no sex-related differences in incidence of epiblepharon in the respective age groups [11]. Although the number of boys included in the previous study was larger than our population, it included both patients with epiblepharon involving the upper and lower eyelids and those with the upper eyelid only [11]. Unlike patients with lower eyelid epiblepharon, we encounter patients with upper eyelid epiblepharon in any age or sex group. In addition, as the previous study examined patients on routine ophthalmic screening, it may have included patients with mild epiblepharon not requiring surgical correction, particularly older men [11]. None of the symptoms in 78% of patients included in the previous study suggest the mildness of the condition [11]. The differences in our inclusion criteria could probably explain the discrepancy.

Several factors are usually considered in deciding whether to pursue surgery. These include fear of potential complications from general anesthesia, as well as the surgery itself, socioeconomics, severity, age at presentation, and cultural issues. We acknowledge that fear of potential complications is a possible factor that could have influenced the parents' decision in agreeing to or refusing surgery for congenital epiblepharon. Some parents may not want to subject their children to general anesthesia, taking into consideration the risk vs. benefit ratio of such a procedure. Furthermore, the complications arising from the surgery itself might hinder the parents from allowing their children to undergo epiblepharon surgery. In terms of cultural issue, women exhibit more concern for their appearance. In our results, there were more girls having surgery within the age range of 0-12 years (84 boys vs. 134 girls). It is possible that a bias may have occurred, with parents of girls wanting to have the congenital epiblepharon addressed at a young age in order to resolve the cosmetic issue before their children reached their teens. This might have skewed our results to display a larger number of girls aged 0-12 years undergoing surgery.

The age at which patients sought consultation with an ophthalmologist should be considered in a study regarding the sex-related difference in age distribution of congenital epiblepharon. In Japan, all children receive serial eye examinations by law. Mandatory eye examination starts at age of 3 to 4 years, before entering elementary school (at 6 years), and every year from the 1st to the 6th grade. Therefore, it is thought that there was no sex-related difference in the age of consultation with an ophthalmologist in the subjects of this study.

A limitation of our study was the retrospective nature of our study design. Hence in the future, it would be better to have a large, prospective, randomized study design, which could control and limit confounders ensuring better validity. Second was the absence of an objective evaluation

system for the cilia-induced-corneal irritation as well as the subjective nature of assessment as to the need for surgical intervention of the patients in this study.

In conclusion, our findings suggest that conservative management may be a good option for boys with congenital epiblepharon as long as there are no severe corneal complications and no risk of amblyopia as spontaneous resolution is likely to occur. However, for girls aged >13 years, the threshold for surgical intervention should be lower since spontaneous resolution is less likely to occur and complications are more likely to develop when the epiblepharon is left to persist.

Author contributions All authors qualify for authorship based on contributions to the conception and design (YT and HK), acquisition of data (YT), literature search (MRPV and YT), and analyses and interpretation of data (MRPV, YT, SN, and HK). All authors contributed to drafting the article and revising it critically for important intellectual content and final approval of the version to be published.

Contributors No one contributed to the work who did not meet our authorship criteria.

Conflicts of interest M R. P. Valencia, None; Y. Takahashi, None; S. Nakakura, None; H. Kakizaki, None.

References

1. Levitt J. Epiblepharon and congenital entropion. Am J Ophthalmol. 1957;44:112–3.
2. Johnson CC. Epiblepharon. Am J Ophthalmol. 1968;66:1172–5.
3. Kakizaki H, Leibovitch I, Takahashi Y, Selva D. Eyelash inversion in epiblepharon: is it caused by redundant skin? Clin Ophthalmol. 2009;3:247–50.
4. Seo JW, Kang S, Ahn C, Esmaeli B, Sa HS. Non-incisional eyelid everting suture technique for treating lower lid epiblepharon. Br J Ophthalmol. 2018;102:1504–9.
5. Young SM, Kim YD, Woo KI. Modified everting sutures: an alternative treatment for mild to moderate epiblepharon. Br J Ophthalmol. 2018;102:1510–4.
6. Shin DH, Woo KI, Kim YD. Relationship between lower eyelid epiblepharon and epicanthus in Korean children. PLoS One. 2017;12:e0187690.
7. Preechawai P, Wong A, Sundar G. Refractive changes in epiblepharon. Am J Ophthalmol. 2007;143:835–9.
8. Kim N, Yoo YJ, Choung HK, Khwarg SI. Epiblepharon in congenital glaucoma: case-control study. Br J Ophthalmol. 2017;101:1654–7.
9. Naik MN, Pujari A, Ali MJ, Kaliki S, Dave TV. Nonsurgical correction of epiblepharon using hyaluronic acid gel. J AAPOS. 2018;22:179–82.
10. Kim JS, Jin SW, Hur MC, Kwon YH, Ryu WY, Jeong WJ, et al. The clinical characteristics and surgical outcomes of epiblepharon in Korean children: a 9-year experience. J Ophthalmol. 2014;2014:156501.
11. Noda S, Hayasaka S, Setogawa T. Epiblepharon with inverted eyelashes in Japanese children. I. Incidence and symptoms. Br J Ophthalmol. 1989;73:126–7.
12. Hayasaka S, Noda S, Setogawa T. Epiblepharon with inverted eyelashes in Japanese children. II. Surgical repairs. Br J Ophthalmol. 1989;73:128–30.
13. Wright KW, editor. Pediatric ophthalmology for primary care. 2nd ed. Ilinois: American Academy of Pediatrics; 2003. p. 257.
14. Wei N, Bi H, Zhang B, Li X, Sun F, Qian X. Biphasic growth of orbital volume in Chinese children. Br J Ophthalmol. 2017;101:1162–7.
15. Furuta M. Measurement of orbital volume by computed tomography: especially on the growth of the orbit. Jpn J Ophthalmol. 2001;45:600–6.
16. Kakizaki H, Selva D, Leibovitch I. Cilial entropion: surgical outcome with a new modification of the Hotz procedure. Ophthalmology. 2009;116:2224–9.