Conservative management of congenital microphthalmos with prosthesis

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Conservative management of congenital microphthalmos with prosthesis

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Sung Eun Kim

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ABSTRACT

Conservative Management of Congenital Microphthalmos with Prosthesis

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Congenital microphthalmos is a rare malformation of the eye resulting from abnormalities in the development of the primary optic vesicle. Treatment for this condition aims to avoid the retardation of orbital growth and cosmetically devastating facial asymmetry. This report details the experiences of patients with microphthalmos treated conservatively with prostheses. The clinical characteristics of congenital microphthalmos were investigated, and the effects of prosthesis treatment on orbital symmetry and cosmetic outcome were analyzed.

The medical records of 32 patients were reviewed for the timing of prosthesis treatment, characteristics of the orbits and eyelids, and presence of associated systemic abnormalities. Surveys inquiring about problems experienced during the prosthesis treatment were administered to ocularists and patients' parents. Current prosthesis size was measured and compared with the primary prosthesis. Orbital symmetry and cosmetic outcome were graded in current photographs of patients.

Twenty-six patients (81.3%) had unilateral microphthalmos, and six (18.7%) had bilateral microphthalmos. Mean patient age was 10.7±3.8 years. At initial prosthesis placement, patients

demonstrated eyelid asymmetry (n=12), orbital bone hypoplasia (n=9), and zygomaticomaxillary hypoplasia (n=6). Orbital bone hypoplasia was associated with eyelid asymmetry ($p=0.038$) and zygomaticomaxillary hypoplasia ($p<0.001$). Patient age at the treatment initiation was significantly greater in patients with zygomaticomaxillary hypoplasia than in patients without hypoplasia ($p=0.046$). Cosmetic outcome with the prosthesis was fair to good in all patients. However, patients with either orbital bone hypoplasia or zygomaticomaxillary hypoplasia at primary treatment tended to have remaining orbital bone asymmetry after treatment ($p<0.001$) and to show poorer cosmetic outcomes ($p=0.004$). Additionally, patients with zygomaticomaxillary hypoplasia were more likely to have an insufficient socket after treatment ($p=0.031$).

The management of congenital microphthalmos is challenging. However, treatment with a prosthesis can be effective for most patients, and early initiation of this treatment is crucial to avoid orbital and facial asymmetry.

Key words: Congenital microphthalmos, Prosthesis, Orbital hypoplasia

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I. INTRODUCTION

Congenital microphthalmos is a disfiguring malformation of the eye resulting from abnormal development of the primary optic vesicle. This rare condition has a prevalence of 1.5 to 1.7 per 10,000 births.[1-2] The microphthalmic eye at birth has a diameter of <15 mm (normal range, 15-19 mm) and may exhibit other abnormalities such as coloboma, orbital cyst, cataract, and optic nerve abnormalities.[3]

Although the etiology of microphthalmos has not been conclusively identified, viral infection (e.g., rubella, cytomegalovirus) early in pregnancy, fetal exposure to teratogens (e.g., alcohol, isoretinoic acid), radiation exposure, and diabetes have been suggested as possible causes.[3] Recent reports have also indicated an association between genomic mutations in *SOX2* and *CHX10* and microphthalmos.[4-5]

The impaired development of the globe and the absence of stimulation for orbital bone growth result in underdevelopment of the bony orbit and eyelid. A noticeable hemifacial hypoplasia, including secondary hypoplasia of the zygomaticomaxillary complex and mandible has also been reported.[6]

Treatment for anophthalmos and microphthalmos aims to avoid the retardation of orbital growth, which can lead to cosmetically devastating midfacial asymmetry. Traditional methods have included the use of progressively bigger static acrylic prostheses to enlarge the conjunctival socket and lid, followed by the placement of solid, spherical orbital implants, autogenous dermis-fat grafts, inflatable tissue expanders or, more recently, hydrophilic self-expanding hydrogel expanders to promote orbital expansion.[7-8] Craniofacial surgery to enlarge the orbit may be required at a later stage. Conservative, nonsurgical treatment using progressively larger prostheses can also achieve good cosmetic results, reducing the demands of secondary surgical intervention.[9]

This paper reports an experience with 32 patients with microphthalmos who were treated with the non-invasive prosthesis technique. We investigated the clinical characteristics of microphthalmos, and described the factors affecting orbital symmetry and cosmetic outcome after treatment.

II. MATERIALS AND METHODS

This study included 32 patients with congenital microphthalmos treated with non-surgical expansion of conjunctival socket and orbit with a prosthesis at Severance Hospital between 1998 and 2010. Patients for whom treatment was initiated before 12 years of age and who were followed up for a minimum of 12 months were included in the study. Patients who received any surgical treatment prior to or during the prosthesis treatment were excluded from the study.

The following data were collected from patients' medical records: 1) age at initiation of prosthesis treatment; 2) characteristics of the orbits and eyelids; 3) presence of associated systemic abnormalities; and 4) family medical history. Orbital and zygomatomaxillary hypoplasia of microphthalmic side at initial wearing of the prosthesis was assessed by palpation of bony orbital rim and zygomaticomaxillary complex of both sides and an asymmetric bony development was defined as hypoplasia. Surveys about the problems encountered during the prosthesis treatment were given to the ocularist and to the parents of each patient. Horizontal and vertical lengths and thicknesses of currently worn prostheses were measured and compared with those of the primary prostheses. Photographs were taken of all patients before and after use of the prosthesis, and horizontal and vertical palpebral fissures were measured using ImageJ (National Institutes of Health, Bethesda, MD, USA). Lengths were measured in pixels in both the

prosthetic eye and the normal eye, and relative ratios were calculated against the normal eye. The horizontal palpebral fissure (HPF) was measured as the distance between the most medial point of the medial canthus and the most lateral point of the lateral canthus. The vertical palpebral fissure (VPF) was measured as the distance between the upper eyelid margin and lower eyelid margin, with the center of the pupil as a reference.

Orbital bone symmetry and expansion of the conjunctival socket were assessed with an ophthalmic evaluation by one observer. The symmetry between a microphthalmic orbit and a normal orbit was assessed by measuring the vertical length between upper and lower margin of orbital rim and classified as symmetric (i.e., vertical lengths of normal orbit and microphthalmic orbit are same (B/A=1, Figure 1)), tolerable (i.e., asymmetric but not easily detectable (B/A≥0.95), and asymmetric (i.e., B/A<0.95). Conjunctival socket expansion was evaluated and compared with the conjunctival socket of the normal eye. If the socket was deep enough for the insertion of the prosthesis and the depth was similar to the normal eye, the socket expansion was considered sufficient. Socket expansion was considered fair if the socket was shallower than the normal socket and did not have lid entropion or displacement of the prosthesis. An insufficient socket resulted in displacement of the prosthesis or entropion and required surgical intervention. Final cosmetic outcome at last follow-up was graded subjectively by the parents of each patient as good, fair, or poor.

Data were analyzed using SAS software version 9.1.3 (SAS

Institute Inc., Cary, NC, USA). Comparisons of median values, such as the prosthesis initiation times between two groups, were performed using Wilcoxon rank-sum test, and correlations were analyzed using chi-square test. P values <0.05 were considered statistically significant.

Figure 1. Orbital bone symmetry assessment by measuring the vertical length between upper and lower margin of the orbital rim in the normal orbit (A) and in the microphthalmic orbit (B).

III. RESULTS

Demographic and clinical characteristics of patients

Forty eyes of 32 patients with congenital microphthalmos were analyzed. Twenty-six patients (81.3%) had unilateral microphthalmos, and six patients (18.7%) had bilateral microphthalmos. Mean patient age at last follow-up was 10.7±3.8 years. Seventeen patients were male, and 15 patients were female. Nine of the 32 patients (28.1%) had associated systemic abnormalities; six patients had unilateral microphthalmos and three had bilateral microphthalmos. The associated congenital systemic abnormalities included mental retardation (n=5), cardiac anomalies (n=2), cerebral palsy (n=1), and nephrotic syndrome (n=1). No patient had a family history of congenital microphthalmos. Median patient age at initial prosthesis treatment was 28.5 months (range, 3-120 months). Mean duration of prosthesis treatment was 7.3±3.9 years, and the mean interval between prostheses was 15.9±8.2 months.

Statistical analysis of patients with unilateral microphthalmos

The characteristics and the results of ophthalmic evaluation of the 26 patients with unilateral microphthalmos are summarized in Table 1. At time of prosthesis initiation, eyelid asymmetry (n=12), orbital bone hypoplasia (n=9), and zygomaticomaxillary hypoplasia (n=6) were noted. Orbital bone hypoplasia was generally associated with eyelid asymmetry ($p=0.038$) and zygomaticomaxillary hypoplasia ($p<0.001$). Eleven of the 26 patients (42.3%)

demonstrated good cosmetic outcome with complete satisfaction on symmetry, and 15 of the 26 patients (57.7%) demonstrated fair cosmetic outcome with only partial satisfaction. Orbital bone development after prosthesis treatment was assessed by comparing the orbital symmetry against the normal orbit. Results in the 26 patients indicated symmetry in 16 (61.5%), tolerability in 5 (19.2%), and asymmetry in 5 (19.2%). Twelve of the 26 patients (46.2%) exhibited sagging of the lower lid on prosthetic eye.

Age at initiation of prosthesis treatment

The age at initial wearing of the prosthesis was compared between patients with and without zygomaticomaxillary hypoplasia. In patients with zygomaticomaxillary hypoplasia, the median starting age for treatment was 60 months (range 10-120 months), which was significantly older than that in patients without zygomaticomaxillary hypoplasia (range 3-60 months) ($p=0.046$) (Figure 2). The primary treatment age of patients with orbital hypoplasia also tended to be older than that of patients without orbital bone hypoplasia; however, there was no statistically significant difference ($p=0.117$).

Change in prosthesis size

Measurements of horizontal length, vertical length, and thickness of currently worn prostheses were compared with those of the primary prostheses. The mean horizontal length of the primary prosthesis was 13.0±4.5 mm, and the mean length of the

current prosthesis was 26.2±3.4 mm. The mean vertical length of the primary prosthesis was 9.5±4.0 mm, and the mean length of the current prosthesis was 20.3±5.5 mm. The mean thickness of the prostheses increased from 1.5±0.6 mm to 3.4±0.8 mm. The increase in horizontal length was greater than the increase in vertical length. The increase in the prosthesis size showed no statistical difference between patients with and without orbital bone hypoplasia or zygomaticomaxillary hypoplasia. However, the increase in vertical length was significantly greater in patients with sagging of the lower lid at current evaluation than in those without it (p=0.045).

Palpebral fissure ratio

In each patient with unilateral microphthalmos, the horizontal and vertical palpebral fissures were measured on the photograph, and the ratios compared to the normal eye were calculated. The mean horizontal palpebral fissure ratio was 0.93±0.08, and the mean vertical palpebral fissure ratio was 0.99±0.23. A relatively short lid fissure was identified for microphthalmic eyes compared to normal eyes. In patients with orbital bone hypoplasia at the initiation of treatment, the vertical interpalpebral fissure ratio was significantly lower than that in patients without orbital bone hypoplasia (p=0.033); however, no significant difference was found in the horizontal palpebral fissure ratio (p=0.080).

Difficulties during prosthesis treatment

Surveys on difficulties experienced during the prosthesis treatment identified mucous socket discharge (69.2%), eyelid irritation (50.0%), entropion (46.2%), conjunctival socket insufficiency (34.6%), and pain on wearing of the prosthesis (15.4%). Compared to patients without orbital hypoplasia, patients with orbital hypoplasia experienced more difficulty initiating prosthesis treatment due to the insufficient conjunctival socket and the presence of lid fissure contracture ($p<0.001$). Patients who had asymmetric, short eyelids at the time of prosthesis initiation were apt to complain of eyelid irritation ($p=0.002$), and patients with conjunctival socket insufficiency tended to suffer from eyelid irritation ($p<0.001$) and entropion ($p<0.001$). Patients with zygomaticomaxillary hypoplasia were more likely to have an insufficient socket after treatment ($p=0.031$).

Overall cosmetic outcome

Current subjective cosmetic outcomes with prosthesis were fair to good in all patients. No patient felt the need to cover the eye with the prosthesis or graded his/her overall cosmetic outcome as poor. However, patients who had orbital bone hypoplasia or zygomaticomaxillary hypoplasia at the time of primary treatment tended to retain orbital bone asymmetry after treatment ($p<0.001$) (Figure 3) and to show poorer cosmetic outcomes ($p=0.004$) (Figure 4).

Table 1. Presentation of 26 patients with unilateral microphthalmos

No.	Sex	Age	Side	Age at primary treatment (months)	Prosthesis change interval (months)	Follow up (months)	Evaluation at time of primary treatment						Current status evaluation					
							Eyelid asymmetry	Insufficient Socket	Orbital bone hypoplasia	Zygomaticomaxillary hypoplasia	Congenital Systemic abnormalities	Prosthesis Initiation	VPF ratio	HPF ratio	Cosmetic outcome	Lower lid sagging	Socket expansion status	Orbital bone symmetry
1	F	8	RE	14	12	82	−	−	−	−	Nil	Easy	0.83	0.95	Good	−	Sufficient	Symmetric
2	M	7	RE	36	9	48	−	−	−	−	Mental retardation Developmental delay	Easy	1.78	1.00	Good	−	Sufficient	Symmetric
3	F	9	LE	8	8	100	+	+	−	−	Nil	Tolerable	0.85	0.85	Fair	−	Fair	Tolerable
4	F	12	RE	8	8	136	−	−	−	−	Nil	Easy	1.13	1.00	Fair	+	Fair	Symmetric
5	F	5	RE	11	12	49	+	+	−	−	Nil	Easy	1.00	1.00	Good	−	Sufficient	Symmetric
6	M	9	RE	24	12	84	+	+	−	−	Nil	Tolerable	1.00	0.83	Fair	−	Insufficient	Symmetric
7	F	9	RE	25	12	83	+	+	−	−	Nil	Tolerable	1.08	0.89	Fair	+	Fair	Symmetric
8	M	17	LE	11	12	193	−	−	−	−	Nephrotic syndrome	Easy	1.00	0.89	Good	+	Sufficient	Symmetric
9	M	7	RE	6	12	78	+	+	−	−	Nil	Tolerable	1.00	0.95	Good	_	Fair	Symmetric
10	F	10	RE	6	12	114	−	+	−	−	Cardiac anomaly	Tolerable	0.89	0.97	Fair	+	Fair	Symmetric
11	F	7	RE	12	18	72	−	−	−	−	Nil	Tolerable	1.09	1.00	Good	+	Sufficient	Symmetric
12	M	12	RE	3	12	141	−	+	−	−	Cerebral palsy	Tolerable	1.13	1.00	Fair	+	Insufficient	Symmetric
13	M	16	RE	45	18	147	−	−	−	−	Nil	Easy	1.18	1.00	Good	−	Sufficient	Symmetric
14	M	8	RE	60	18	36	−	−	−	−	Mental retardation	Easy	1.21	1.04	Fair	−	Sufficient	Symmetric
15	F	12	RE	60	36	84	−	−	−	−	Nil	Easy	1.00	0.98	Good	−	Sufficient	Symmetric
16	M	19	RE	12	12	216	−	−	−	−	Nil	Tolerable	0.95	0.89	Good	+	Sufficient	Symmetric
17	M	14	RE	48	24	120	−	−	−	−	Nil	Easy	0.88	0.96	Good	−	Sufficient	Symmetric
18	F	6	RE	12	9	60	+	+	+	−	Nil	Difficult	1.11	0.90	Fair	+	Fair	Tolerable
19	F	2	RE	9	11	15	+	−	+	−	Nil	Tolerable	0.88	0.92	Fair	−	Fair	Asymmetric
20	M	10	RE	32	12	88	+	−	+	−	Nil	Difficult	0.40	0.88	Fair	−	Sufficient	Tolerable
21	M	17	LE	120	24	84	+	+	+	+	Nil	Difficult	0.67	1.00	Fair	+	Insufficient	Asymmetric
22	M	9	LE	15	6	93	+	+	+	+	Nil	Tolerable	0.94	0.92	Fair	_	Insufficient	Tolerable
23	M	15	RE	60	24	120	−	−	+	+	Nil	Difficult	1.00	1.00	Fair	+	Sufficient	Asymmetric
24	M	13	LE	60	18	96	−	−	+	+	Nil	Tolerable	0.88	0.95	Fair	+	Fair	Tolerable
25	M	11	RE	10	24	122	+	+	+	+	Cardiac anomaly	Difficult	1.00	0.72	Fair	+	Insufficient	Asymmetric
26	F	9	LE	72	18	36	+	+	+	+	Nil	Tolerable	0.84	0.80	Fair	−	Insufficient	Asymmetric

RE, Right eye; LE, Left eye; VPF, Vertical palpebral fissure; HPF, Horizontal palpebral fissure

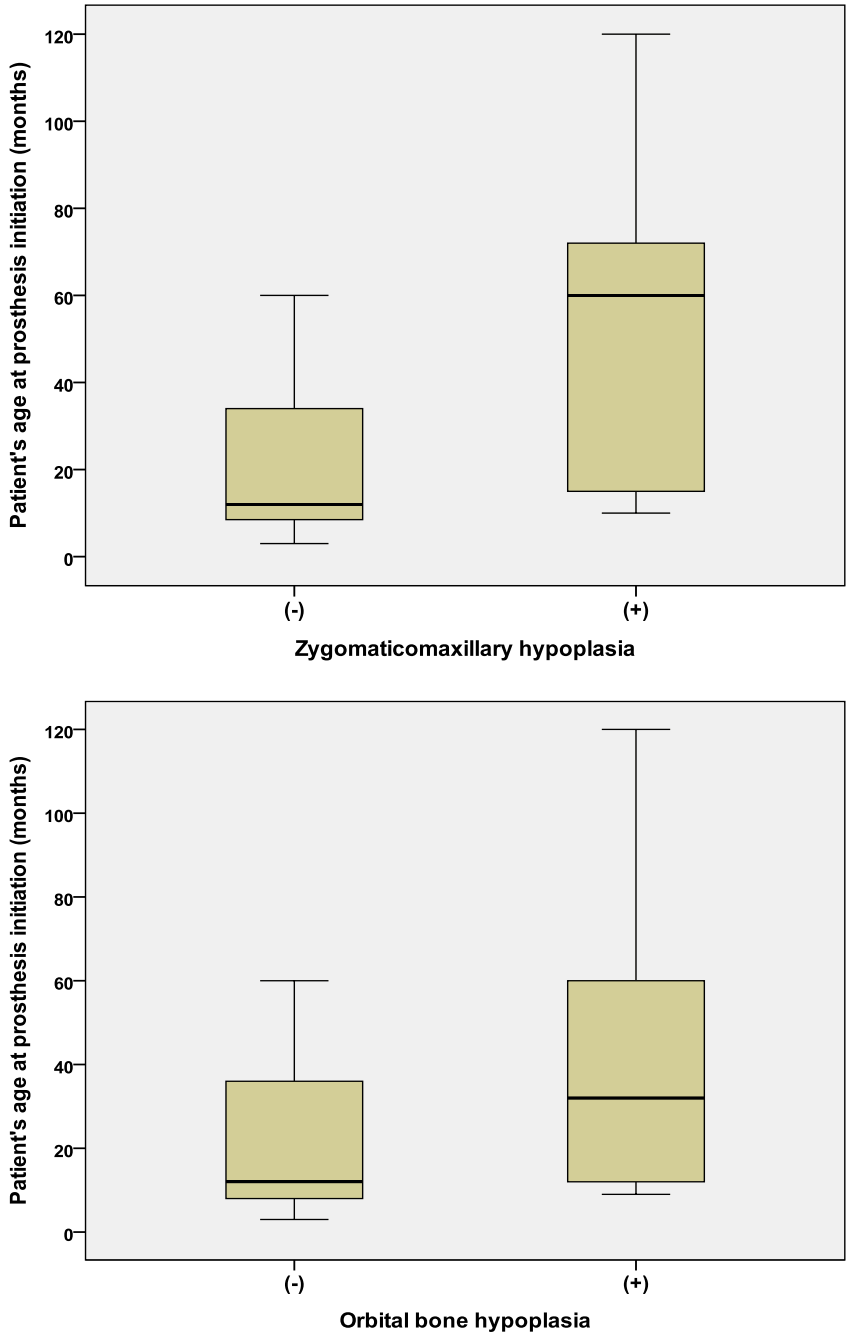

Figure 2. Comparison of the patient age at prosthetis initiation

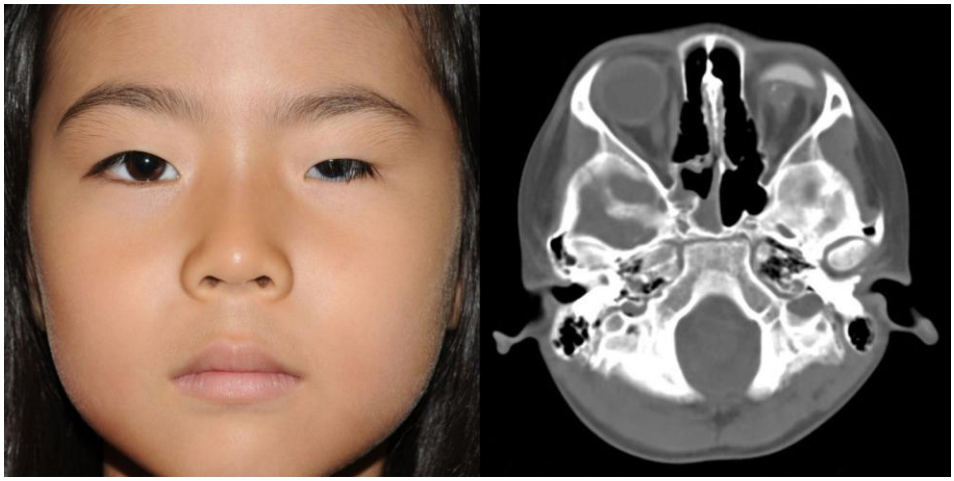

Figure 3. A Photograph and a computed tomography scan of a patient with orbital hypoplasia and facial asymmetry who started an initial wearing of the prosthesis at 72 months of age (Case 26)

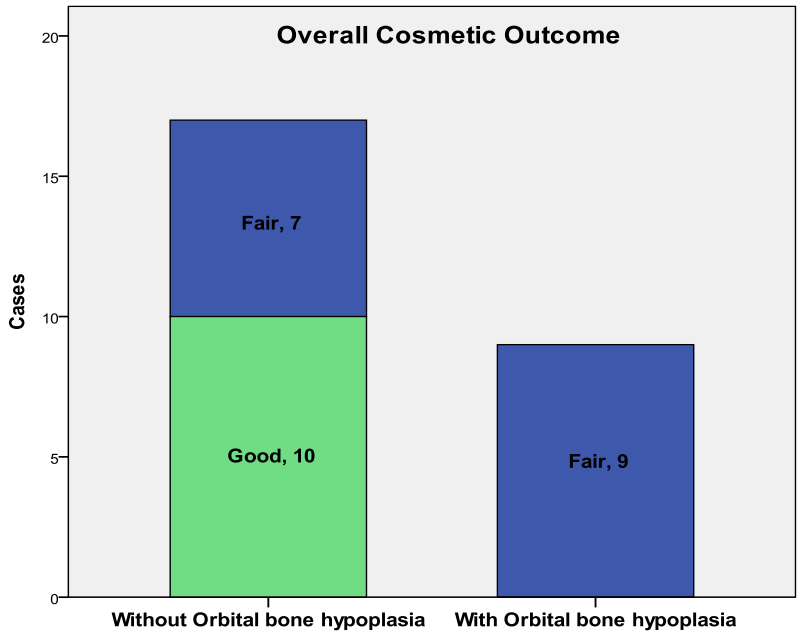

Figure 4. Cosmetic outcome after treatment in patients with or without orbital bone hypoplasia at initial wearing of the prosthesis

IV. DISCUSSION

This study analyzed 32 patients with congenital microphthalmos before and after prosthesis treatment. The inclusion of a relatively large number of microphthalmic patients compared to previous reports allowed a review of the general features of congenital microphthalmos. The effects of the prosthesis treatment and the factors affecting orbital symmetry and cosmetic outcome after treatment were assessed.

Several patients had associated systemic abnormalities. Six of the 26 patients (23.1%) with unilateral microphthalmos and three of the six patients (50%) with bilateral microphthalmos experienced mental retardation, developmental delays, cardiac anomalies, cerebral anomaly, or nephrotic syndrome. This finding is comparable to the 20.6% incidence of systemic abnormalities reported by Tucker et al.[10] It is also similar to that reported by Schittkowski et al, who showed a predominance of cerebral anomalies in patients with bilateral microphthalmos compared to those with unilateral microphthalmos.[11] Therefore, a child presenting with suspected microphthalmia, especially bilateral microphthalmia, requires early assessment by both an ophthalmologist and a pediatrician to identify underlying systemic abnormalities.

Normal facial and orbital development is affected by the reduction in ocular volume. In cases of anophthalmia and moderate to severe microphthalmia, there is often underdevelopment of the

bony orbit, the eyelid, and even the zygomaticomaxillary complex. Without intervention, the socket remains underdeveloped and the asymmetry becomes more pronounced as the child grows. This was shown experimentally by Cepela et al using an enucleated cat model.[12] Eighteen cats had unilateral enucleation at 2 weeks of age and then received either expandable orbital implants, solid 8-mm or 12-mm silicone implants, or no implant. Significant orbital growth was stimulated by the use of large or expandable volume implants.

Various orbital expansion methods are currently used for anophthalmic or microphthalmic patients, including the use of progressively larger prostheses, solid orbital implants, autogenous dermis-fat grafts, and inflatable or self-expanding tissue expanders. Early removal of the defective eye and implantation of a ball implant or dermis fat graft is one of the methods used to treat congenital microphthalmos. Another approach involves preserving the microphthalmic eye even if there is no visual potential. Potential advantages of this approach include stimulation of the palpebral aperture, stimulation of socket growth, and avoidance of early invasive surgery.[8] Price et al reported that four severely microphthalmic eyes treated with cosmetic scleral shells demonstrated symmetrical ocular appearance without the need for surgery.[9] Oberhansli et al also concluded that early use of scleral shells induced orbital growth and gave satisfactory cosmetic results.[13]

In this study, prosthetic treatment produced a good cosmetic outcome in most patients. However, if prosthesis treatment is delayed, orbital and facial underdevelopment occurs, which induces orbital asymmetry and a poorer cosmetic outcome. When orbital and facial underdevelopment is noted, these could not be overcome solely with prosthesis treatment and was likely to remain even after prosthesis treatment. Furthermore, in patients with zygomaticomaxillary hypoplasia, socket expansion was also insufficient and necessitated surgical correction. Initiation of prosthesis treatment or an equivalent orbital expansion therapy as soon as possible is crucial for the proper management of congenital microphthalmos.

Recently, self-expanding hydrophilic osmotic expanders have been introduced, and some reports have described promising results.[14] These expanders decrease the risks of tissue ischemia, infection, and extrusion seen with the previous inflatable balloon expanders. However, many ophthalmologists have not had experience with these new expanders and prefer to use the traditional prosthesis treatment.[15] Although prosthesis treatment demonstrates satisfactory results, it alone may not be enough for severe cases of microphthalmos with emerging orbital asymmetry. Therefore, an effort to balance new treatment methods and surgical treatment with prosthesis treatment is necessary.

The measurement of prosthesis size showed that the change in horizontal length was greater than the change in vertical

length. Larger changes in vertical length were associated with lower lid sagging. An overly increased vertical length could raise the vertical pressure on the lower eyelid causing lid laxity and sagging. Furthermore, a deep upper eyelid sulcus, as well as accelerated downward movement of the prosthesis, could occur. Therefore, increases in the vertical length of the prosthesis require attention.

Most patients in this study were tolerable in wearing prostheses. However, several patients complained of mucus discharge, eyelid irritation, and entropion; most of these patients had short eyelids or conjunctival sac insufficiencies. In such cases, the prosthesis brings greater pressure to the surrounding eyelid and conjunctiva, and the increased mechanical stimuli may accelerate inflammatory reactions leading to pathologic conditions such as giant papillary conjunctivitis.[16] Maintenance of proper prosthesis hygiene and instillation of anti-inflammatory drops or artificial tears will be helpful in these patients. Socket expansion with replacement of gradually larger bearable prostheses often will be better tolerated than an excessively large prosthesis.

This study analyzed a significantly large number of microphthalmic patients with a fairly long follow-up period. One comparable report on the prosthetic experience studied 13 cases of microphthalmos over 4 years. Our study also assessed the effects of the prosthesis treatment technique as well as the factors affecting orbital symmetry and cosmetic outcome after treatment

with objective methods. This represents the first report to measure the size of the prosthesis before and after treatment, and to perform a precise photographic measurement of the palpebral fissures of microphthalmic patients.

However, some limitations of the study deserve consideration. Because we had such a small number of bilateral microphthalmos patients, they had to be excluded from the statistical analysis. Imaging modalities such as orbital computed tomography might be more helpful in the evaluation of orbital symmetry and treatment effect. Future studies should focus on such considerations.

V. CONCLUSION

Congenital microphthalmos presents clinical challenges for oculoplastic surgeons and ocularists. Specific evaluation of the initial characteristics, including orbital volume, socket features, and the orbital and zygomaticomaxillary bony structures will lead to optimum reconstructive plans. Early expansion of the microphthalmic socket may help avoid orbital and facial asymmetry.

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ABSTRACT(IN KOREAN)

선천성 소안구증 환자에서 보존적 치료의 효과

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김 성 은

선천성 소안구증으로 인한 안구의 용적 감소는 안와골 뿐만 아니라 주위 골구조의 성장과 발달에도 영향을 미친다. 이에 소안구증의 일차적 치료는 의안을 착용하여 안와뼈의 성장을 유도하고, 대칭적인 안면성장을 이루는 데에 그 목적이 있다. 본 연구는 선천성 소안구증 환아의 질병양상을 조사하고, 의안을 이용한 보존적 치료의 효과를 객관적으로 분석하며, 치료 시작 시기 및 당시의 안와 및 주변 골구조의 양상이 치료 후 안와와 안면 성장에 어떠한 영향을 주는지 분석하고자 하였다.

선천성 소안구증으로 진단되어, 경성 의안을 이용하여 안와골 및 결막낭의 확장을 유도해 온 환아 32명 38안을 대상으로 하였다. 의무기록 분석을 통하여 의안 착용 시작시기와 당시의 안와 및 안검의 양상과 전신장애 유무를 조사하고, 의안사 및 보호자에 대한 설문조사를 통하여 의안 치료 시작 시의 착용 어려움 정도와 치료 중의 문제점을 조사하였다. 현재 의안 착용 후 외모 대칭성에 대한 만족도를 조사하고, 결막낭의 확장 정도와 안와 비대칭성 정도를 3단계로 나누어 기록하였다. 의안 착용 전후의 정면 사진을 찍고, 수평눈꺼풀길이와 수직눈꺼풀틈새를 측정하여 정상안과 비교하였다. 현재 착용 중인 의안의 가로길이, 세로길이, 두께를 측정하여, 의안 착용 시작 시의 의안의 크기와 비교하였다.

선천성 소안구증 환아 32명의 평균 나이는 10.7±3.8세였으며, 단안 선천성소안구증이 26명(81.3%), 양안 선천성소안구증이 6명(18.7%)이었다. 초진 당시 안와뼈 성장장애와 눈꺼풀 비대칭이

있던 경우는 각 9명, 12명으로, 동반되어 존재하는 경우가 많았고 ($p=0.038$), 광대뼈 비대칭이 있던 경우 6명 또한 안와뼈 성장장애와 동반되는 경우가 많았다($p<0.001$). 의안 착용 시작 시기는 광대뼈 비대칭이 있었던 환아에서 중간값 생후 60개월이었고, 광대뼈 비대칭이 없었던 환아의 중간값 12개월보다 통계적으로 의미있게 늦은 것으로 나타났다($p=0.046$). 의안 착용 초기에 안와뼈의 성장장애나 광대뼈의 비대칭이 있었던 경우, 현재에도 안와뼈 비대칭이 남아, 의안 착용 후 외모 비대칭을 초래하는 경우가 많았으며, 통계적으로 유의한 상관관계를 보였고($p<0.001$), 또한 광대뼈의 비대칭이 있었던 경우, 현재 결막낭 확장 정도도 충분하지 못한 경우가 많았다($p=0.031$).

결론적으로, 선천성 소안구증에서 의안을 이용한 치료는 대부분의 환아에서 만족할만한 미용적 치료 효과를 보였으나, 의안착용 시작 시기가 늦어지면 안와 및 안면 성장장애와 안면 비대칭이 발생할 수 있고, 이후 의안을 착용하여 안와 성장을 유도하더라도 안면 비대칭이 남을 수 있으므로, 빠른 치료의 시작이 중요하다고 하겠다.

핵심되는 말: 선천성 소안구증, 의안, 안와 성장장애