Temporary Piggyback Intraocular Lens Implantation Versus Single Intraocular Lens Implantation in Congenital Cataracts: Long-Term Clinical Outcomes

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SH and DHL contributed equally to the work presented here and should therefore be considered equivalent authors.

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PURPOSE. To report the long-term results of temporary piggyback IOL implantation in congenital cataract and to compare the clinical outcomes of temporary piggyback IOL with those of single IOL implantation.

METHODS. This is a retrospective, comparative, interventional study. The medical records of all consecutive patients who underwent cataract extraction and single or temporary piggyback IOL implantation within the first 3 years of life from 1999 to 2013 at Samsung Medical Center were reviewed. Twenty-eight eyes from 18 patients underwent single IOL implantation (monopseudophakia group), and 32 eyes of 20 patients underwent temporary piggyback IOL implantation in congenital cataract surgery (polypseudophakia group).

RESULTS. The mean age at initial cataract surgery was 15.8 months in the monopseudophakia group and 11.1 months in the polypseudophakia group (P = 0.144). The average follow-up duration was 135 months in the monopseudophakia group and 120 months in the polypseudophakia group (P = 0.36). The best-corrected visual acuity at the last visit was 0.36 logMAR in the monopseudophakia group and 0.55 logMAR in the polypseudophakia group (P = 0.044). Four (14%) and 14 (44%) reoperations for complications within the anterior segment were performed in the monopseudophakia group and polypseudophakia group, respectively (P = 0.042). Four cases (14.3%) in the monopseudophakia group and 13 cases (40.6%) in the polypseudophakia group had a glaucoma-related adverse event (P = 0.086).

CONCLUSIONS. Compared with primary single IOL implantation in congenital cataract, temporary piggyback IOL implantation produced worse visual acuity, higher reoperation rate, and higher risk of secondary glaucoma. Temporary piggyback IOL implantation does not have benefit in congenital cataract.

Keywords: congenital cataract, cataract surgery, piggyback, polypseudophakia

In congenital cataract, early surgical intervention is required to prevent visual deprivation and development of amblyopia,1 and IOL implantation is the appropriate treatment for aphakic rehabilitation.2 Primary IOL implantations are preferred in children over 2-years old. However, patients younger than 2 years are more prone to myopic shift, visual axis opacification, and inflammation.3-5 Therefore, it remains controversial whether primary IOL implantation at the time of cataract extraction is the optimal approach. Several studies have demonstrated comparable visual outcomes of primary IOL implantation in both unilateral and bilateral congenital cataract,6-8 while a recent prospective randomized clinical trial, the Infant Aphakia Treatment Study (IATS), revealed that primary IOL implantation for unilateral congenital cataract at age less than 7 months had no significant difference in visual acuity and higher rate of complications and reoperations compared with those left aphatic.9 Though the IATS discussed not doing IOL implantation, this option is only applicable to unilateral congenital cataract in very young infants. Specific cut-off age and conditions for primary IOL implantation should be established in future research.

Primary IOL implantation in congenital cataract surgery generally refers to single IOL implantation at the time of cataract removal. However, temporary piggyback IOL implantation, also referred to as temporary polypseudophakia, has been proposed for congenital cataract to achieve favorable refraction during visual development. Wilson et al.10 initially described temporary polypseudophakia for the treatment of congenital cataract in 2001. This procedure involves permanent IOL implantation in the capsular bag and temporary IOL insertion into the ciliary sulcus in order to achieve emmetropia. The temporary IOL can be removed or exchanged according to subsequent refractive changes. Thus, during the critical period of visual development, patients are able to avoid hyperopic periods and the need to wear thick spectacles or contact lenses and can maintain a constant image. Although short-term results of temporary polypseudophakia have been reported,10,11 the corresponding long-term results have not been described in the literature. In this study, we assessed the long-term outcomes of
temporary polypseudophakia and compared these outcomes with those of single IOL implantation in congenital cataract.

METHODS
This retrospective comparative interventional study included patients with congenital cataract who underwent cataract extraction and single or piggyback IOL implantation at an age younger than 36 months. The medical records of all consecutive patients who underwent congenital cataract surgery between 1999 and 2013 at Samsung Medical Center were retrospectively reviewed. Patients with any ocular comorbidity other than congenital cataract (e.g., persistent hyperplastic primary vitreous, Peters’ anomaly, coloboma, or morning glory syndrome) were excluded. This retrospective study was performed in accordance with the principles of the Declaration of Helsinki and was approved by the Samsung Medical Center institutional review board.

Patient information was extracted from the medical records, including age at the time of surgery, basic biometry, complications after surgery, and reoperation rate. Occurrence of glaucoma or glaucoma suspect was recorded as defined in the IATS.12,13 Glaucoma was defined as IOP greater than 21 mm Hg with one or more of the following anatomic changes: (1) corneal enlargement, (2) asymmetrical progressive myopic shift coupled with enlargement of the corneal diameter and/or axial length, (3) increased optic nerve cupping, defined as an increase of 0.2 or more in the cup-to-disc ratio, or (4) the use of surgical procedures for IOP control. A patient was designated as a glaucoma suspect if he or she had two consecutive IOP readings above 21 mm Hg on different dates after topical corticosteroids had been discontinued or if he or she had received glaucoma medication to control IOP. Periods of amblyogenic hyperopia after cataract surgery were calculated for each eye by thorough inspection of refractive changes after congenital cataract surgery. Amblyogenic hyperopia was defined as spherical hyperopic anisometropia greater than +1.50 diopters (D) or hyperopia greater than +5.00D.14,15

Surgery
Review of the medical records revealed that all surgeries in the current study had been performed in a uniform fashion. Two ophthalmologists (FYC and ESC) had performed all surgeries under general anesthesia. A viscoelastic device was injected into the anterior chamber through a clear corneal incision. After continuous curvilinear capsulorhexis was performed when the predicted postoperative refraction reached emmetropia after removal of the temporary IOL, as described previously.10,17 Under general anesthesia, the surgeon fragmented the temporary IOL using an IOL cutter via a 3-mm temporal clear corneal incision after injection of viscoelastics into the anterior chamber. The pieces were then extracted through the incision using forceps.

Statistical Analysis
Statistical analyses were performed by an independent statistician. Statistical Analysis System software version 9.4 (SAS Institute, Cary, NC, USA) was used for statistical analysis of the data. P values less than 0.05 were considered statistically significant. For analysis of bilateral congenital cataract, generalized estimating equation models were used to account for the correlation of paired eyes. Otherwise, nonparametric statistical analyses including the Mann-Whitney U test and Fisher’s exact test was applied to assess the significance of differences between the temporary piggyback IOL implantation group and the single IOL implantation group.

RESULTS
A total of 105 eyes that underwent congenital cataract surgery at Samsung Medical Center from 1999 to 2013 were reviewed. Of these, the eyes of patients with any other ocular comorbidity, and the eyes of patients who were left aphakic after cataract surgery were excluded from the study. Twenty-eight eyes from 18 patients underwent single IOL implantation (monopseudophakia group), and 32 eyes of 20 patients underwent temporary piggyback IOL implantation (polypseudophakia group). All patients had uneventful cataract surgery with good immediate postoperative IOL placement and no other trauma. Table 1 shows the baseline characteristics of the two groups. The two groups were comparable at baseline regarding cataract laterality, age at cataract surgery, preoperative axial length, follow-up period, and age at the last visit. Detailed distributions of those preoperative parameters are presented in the Figure in the form of a boxplot.

Table 2 shows the postoperative visual and refractive outcomes of the two groups. Best-corrected visual acuity (BCVA) was 0.36 ± 0.31 logMAR (interquartile range, 0.10–0.52) in the monopseudophakia group and 0.55 ± 0.33 logMAR (interquartile range, 0.30–0.82) in the polypseudophakia group. The monopseudophakia group had better BCVA at the last visit compared with the polypseudophakia group (P =
Subgroup analysis was then performed to examine whether outcomes depended on cataract laterality. In unilateral congenital cataract, no significant difference was observed between the two groups. However, in bilateral cataract, the monopseudophakia group had significantly better visual acuity ($P < 0.001$). Subgroup analysis of age at initial cataract surgery in unilateral and bilateral congenital cataract was performed. In unilateral congenital cataract, the age at initial cataract surgery was 12.6 ± 8.7 months in monopseudophakia and 13.7 ± 11.7 months in polypseudophakia; in bilateral congenital cataract the age at initial cataract surgery was 15.1 ± 1.0 months in monopseudophakia and 10.3 ± 7.4 months in polypseudophakia. The age at surgery of monopseudophakia and polypseudophakia was not significantly different in both unilateral and bilateral congenital cataract ($P = 0.846$ and $P = 0.165$, respectively). The monopseudophakia group had a longer amblyogenic hyperopic period of 17.04 ± 21.30 months, while that of the polypseudophakia group was 0.30 ± 1.48 months ($P = 0.019$). Twenty of 32 eyes that underwent piggyback IOL implantation underwent planned piggyback IOL removal during follow-up. The mean age at the time of piggyback IOL removal was 6.12 ± 2.16 years. The mean SE at the time of piggyback IOL removal was $-11.81 ± 3.76$ D, and the mean power of the removed piggyback IOL was $12.40 ± 3.59$ D. At the last visit, the mean SE of the polypseudophakia group was $-6.54 ± 3.90$ D, and the monopseudophakia group had a SE of $-4.71 ± 4.62$ D. In subgroup analysis, the polypseudophakia subgroup of 20 eyes with temporary piggyback IOL removal had a mean SE of $-5.30 ± 3.70$ D, while the polypseudophakia subgroup of 12 eyes without temporary piggyback IOL removal had a mean SE of $-9.01 ± 3.13$ D at the last visit; the latter was significantly more myopic compared with that of the monopseudophakia group ($P = 0.004$). The refractive cylinder at the last visit was $-3.12 ± 1.90$ D in the monopseudophakia group and $-2.50 ± 1.56$ D in the polypseudophakia group ($P = 0.204$).

Table 3 presents the postoperative complications and rates of additional surgery for complications. Reoperation for anterior segment complications such as visual axis opacification, IOL dislocation, and glaucoma are included for analysis. Eyes with temporary polypseudophakia had a higher rate of additional surgery (14/32 eyes, 43.8%) compared with the

<table>
<thead>
<tr>
<th>Parameter</th>
<th>Monopseudophakia</th>
<th>Polypseudophakia</th>
<th>$P$ Value</th>
</tr>
</thead>
<tbody>
<tr>
<td>Number of eyes</td>
<td>28</td>
<td>32</td>
<td></td>
</tr>
<tr>
<td>Number of patients</td>
<td>18</td>
<td>20</td>
<td></td>
</tr>
<tr>
<td>Male:female</td>
<td>8:10</td>
<td>8:12</td>
<td>$&gt;0.999^*$</td>
</tr>
<tr>
<td>Unilateral:bilateral</td>
<td>8:10</td>
<td>8:12</td>
<td>$&gt;0.999^*$</td>
</tr>
<tr>
<td>Age at diagnosis, mo</td>
<td>13.4 ± 9.9</td>
<td>10.7 ± 8.6</td>
<td>0.650†</td>
</tr>
<tr>
<td>Age at operation, mo</td>
<td>15.8 ± 10.7</td>
<td>11.1 ± 8.6</td>
<td>0.144‡</td>
</tr>
<tr>
<td>Preoperative axial length, mm</td>
<td>19.8 ± 1.3</td>
<td>19.4 ± 0.9</td>
<td>0.124‡</td>
</tr>
<tr>
<td>Preoperative keratometry, D</td>
<td>44.8 ± 1.6</td>
<td>44.1 ± 1.5</td>
<td>0.187‡</td>
</tr>
<tr>
<td>Mean follow-up period, mo</td>
<td>153.1 ± 54.5</td>
<td>119.5 ± 31.6</td>
<td>0.391‡</td>
</tr>
<tr>
<td>Age at last visit, y</td>
<td>12.4 ± 4.8</td>
<td>10.9 ± 2.9</td>
<td>0.193‡</td>
</tr>
</tbody>
</table>

Values are presented as mean ± SD unless otherwise indicated.

* Fisher’s exact test.
† Mann-Whitney $U$ test.
‡ Generalized estimating equation model.
monopseudophakia group (4/28 eyes, 14.3%; \( P = 0.042 \)). Among the four eyes that underwent additional surgery in the monopseudophakia group, two required anterior vitrectomy, one underwent IOL rectification, and the other one had IOL rectification with pupilloplasty. In the polypseudophakia group, the anterior segment complications that necessitated the additional surgery consisted of iris-IOL synechiae, trans pupillary membrane, temporary piggyback IOL dislocation, and interlenticular opacification (ILO). Three eyes underwent IOL rectification with synechiolysis, two had IOL rectification with synechiolysis and anterior vitrectomy, four had synechiolysis alone, one had pupilloplasty, and four had ILO removal.

The overall development rates of glaucoma and glaucoma-related adverse events (glaucoma + glaucoma suspect) in the current study were 3/60 (5.0%) and 17/60 (28.3%), respectively. The polypseudophakia group had three cases (9.4%) of glaucoma and 10 cases (31.3%) of glaucoma suspect, whereas the monopseudophakia group had only four cases (14.3%) of glaucoma suspect. The rate of glaucoma-related adverse events together was higher in the polypseudophakia group (\( P = 0.086 \), effect size = 0.550; 4/28,14.3%, cases in the monopseudophakia group and 13/32, 40.6%, cases in the polypseudophakia group). Furthermore, two eyes in the polypseudophakia group underwent filtering surgery due to uncontrolled IOL at 6 and 7 months after primary cataract surgery, respectively.

Five of 18 patients (27.8%) in the monopseudophakia group, and 6 of 20 patients (30.0%) in polypseudophakia group underwent strabismus surgery. There was no significant statistical difference between the two groups (\( P > 0.999 \)). None of the cases in the study required additional retinal surgery.

### DISCUSSION

Hyperopia is much more amblyogenic than myopia.\(^{18}\) Hyperopic anisometropia greater than 1.00 to 1.50 D is considered to produce anisometric amblyopia, and hyperopia greater than 4.00 to 5.00 D is generally thought to induce refractive

<table>
<thead>
<tr>
<th>Parameter</th>
<th>Monopseudophakia</th>
<th>Polypseudophakia</th>
<th>( P ) Value</th>
</tr>
</thead>
<tbody>
<tr>
<td>Additional surgery for complications</td>
<td>4/28 (14.3%)†</td>
<td>14/32 (43.8%)†</td>
<td>0.042</td>
</tr>
<tr>
<td>Anterior vitrectomy</td>
<td>3</td>
<td>2</td>
<td></td>
</tr>
<tr>
<td>Intraocular lens rectification</td>
<td>2</td>
<td>5</td>
<td></td>
</tr>
<tr>
<td>Pupilloplasty</td>
<td>1</td>
<td>1</td>
<td></td>
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<tr>
<td>Synechiolysis</td>
<td>0</td>
<td>9</td>
<td></td>
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<tr>
<td>Interlenticular opacification removal</td>
<td>0</td>
<td>4</td>
<td></td>
</tr>
<tr>
<td>Trabeculectomy</td>
<td>0</td>
<td>2</td>
<td></td>
</tr>
<tr>
<td>Glaucoma</td>
<td>0/28 (0.0%)</td>
<td>3/32 (9.4%)</td>
<td></td>
</tr>
<tr>
<td>Glaucoma-related adverse events‡ (glaucoma + glaucoma suspect)</td>
<td>4/28 (14.3%)</td>
<td>13/32 (40.6%)</td>
<td>0.086</td>
</tr>
</tbody>
</table>

Bolded values are statistically significant with \( P < 0.05 \).

* Generalized estimating equation model.

† Total number of cases that underwent additional surgery due to complications. Multiple surgical procedures could have been conducted during the same episode. Strabismus surgery is not included.

‡ Glaucoma-related adverse event = glaucoma and glaucoma suspect together.
piggyback IOL in adults 21,22 and is thought to be associated with longer follow-up period.

anterior segment complication rate were exacerbated in our study. Wilson et al. 10 reported that ILO developed in 4 of 32 (12.5%) of the polypseudophakic group. None of the cases in the monopseudophakia group had these complications. Because unilateral congenital cataract was small, so it was difficult to reveal differences; larger samples might have been able to present differences in unilateral congenital cataract.

The additional surgery frequency was significantly higher in the polypseudophakia group compared with the monopseudophakia group. The main causes of additional surgery in the polypseudophakia group were posterior synechia, ILO, and glaucoma. None of the cases in the monopseudophakia group had these complications. Wilson et al. 10 reported that piggyback patients and single IOL patients had comparable rates of anterior segment complications that required additional surgery (26% and 22%, respectively). Complications in piggyback patients consisted of IOL tilting, IOL capture in the pupil, and posterior cortex rephoration. 10 Differences of anterior segment complication rate were exacerbated in our study compared with the previous study, possibly due to our longer follow-up period.

ILO has been reported frequently after implantation of piggyback IOL in adults 21,22 and is thought to be associated with two IOLs in the bag. We expected that ILO could be avoided as long as the piggyback IOL was located in the ciliary sulcus as hypothesized by Wilson et al., 10 and therefore performed cataract surgery according to the method described. However, despite meticulous capsular polishing, pearl-type ILO developed in 4 of 32 (12.5%) of the polypseudophakic cases. This result is inconsistent with the result of Wilson et al., 10 in which none of the cases had ILO postoperatively. Because glaucoma is a well-documented and serious complication after congenital cataract removal, all patients in the study were monitored closely for development of glaucoma during the follow-up period. According to a previous study, eyes with primary IOL implantation have a lower risk of developing glaucoma compared with eyes left aphakic. 23 However, retrospective reports might be biased and thus inaccurately conclude that IOL has a protective effect. 24 In a recent prospective randomized clinical trial (IATS 5-year outcome study), the incidence of glaucoma was similar in groups with primary IOL implantation and in those left aphakic. 9,12,13 The current study revealed that the polypseudophakia group had a higher rate of glaucoma-related adverse events (40.6%) compared with the monopseudophakia group (14.5%), and this is as expected, because postoperative elevation of IOP, pigment dispersion syndrome, and secondary glaucoma after piggyback IOL implantation have been reported in adults.9,20 However, the difference of glaucoma-related adverse event was not significantly different in the two groups (P = 0.086, effect size = 0.550). With the sample size of our study, large effects of at least 0.89 can produce a statistical power greater than 80%; we believe that more samples would have proven the significance. Due to the retrospective nature of our study and the lack of gonioscopic and sonographic angle evaluation, it is not possible to specify the exact mechanism of glaucoma development. Further studies are warranted to examine this point. To this end, we are planning further patient follow-up that includes angle evaluation.

The rates of reoperation and adverse events were lower in this study than in the IATS. Overall development rates of glaucoma and glaucoma-related adverse events in our study were 5.0% and 28.3%, respectively. In the IATS 5-year outcome study, the IOL implantation group had a 19% glaucoma rate and a 28% glaucoma-related adverse event rate. The rate of glaucoma-related adverse events in our study is comparable to that of the IATS, but the glaucoma rate is lower. Regarding additional surgery, in addition, the IOL implantation group in the IATS had a 72% reoperation rate, whereas we demonstrated a reoperation rate of 30.0%. We speculate that these lower glaucoma and reoperation rates are due to age differences of participants and the retrospective nature of our study. The IATS only enrolled infants younger than 7 months of age, whereas the mean age of the children in our study was 11 months at the time of surgery. Because the risk of complications is known to be higher in young infants, difference of baseline age may have produced lower rates of complications and associated additional surgeries in the current study. In addition, regarding glaucoma, our study was a retrospective chart review, and axial length and corneal diameter were not routinely examined, so possible glaucoma cases might have been categorized as glaucoma suspect rather than glaucoma.

The amblyopic hyperopic period was significantly longer in the monopseudophakia group than in the polypseudophakia group during the critical period of visual development. However, somewhat paradoxically, the final vision of the polypseudophakia group was worse. This might have been produced by several factors. First, the higher rates of anterior segment complications and visual axis opacification such as synechia or ILO could have interfered with image focusing and aggravated the development of amblyopia in the polypseudophakia group. Second, although this hypothesis has not been proven, polypseudophakia might have produced severe higher order aberration that affected the quality of vision and eventually deteriorated into amblyopia. Because it is almost impossible to exactly align the anterior temporary IOL and the posterior permanent IOL at the central axis point, temporary polypseudophakia is more likely to induce higher order aberration compared with monopseudophakia. In addition, the monopseudophakia group was aggressively treated with glasses for amblyogenic refractive error, so they might have had less amblyopia with better visual outcomes than expected.

To the best of our knowledge, this is the first study to report long-term outcomes of temporary piggyback IOL in congenital cataract. The strengths of this study are the long follow-up period, the similar baseline characteristics of the eyes in each group, and the uniformity in surgical procedures, as evidenced by the uneventful surgeries of all cases. Because the study shows long-term clinical outcomes and considering that ages at the last visit in both groups were above 10 years, the visual acuity and refraction of our study are accurate and reliable. A previously reported comparative study of polypseudophakia and monopseudophakia for congenital cataract followed patients for only 2 years and was unable to accurately analyze visual acuity due to the young age of the study participants; this merits deeper study.

The current study had certain limitations. First, the study was designed to be retrospective. Hence, our study is prone to compounding factors, such as indication, time, and surgeon experience; this is an inherent limitation of this type of study. However, the baseline characteristics of age at diagnosis, age at operation, preoperative axial length, and keratometric value were not significantly different in the two study groups; this at
least suggests that the indication of surgery did not greatly differ between those two groups. Additionally, the two groups did not seem to be different regarding time and surgical expertise; the median date of cataract surgery was November 5, 2004 in the monopseudophakia group and September 8, 2005 in the polypseudophakia group. Hence, we believe that surgeon experience for these procedures would not have significantly influenced the differences of surgical outcomes between the two groups. Another limitation is the small sample sizes of the study groups. Even though we included the largest number of piggyback IOL implantations for congenital cataract reported in the literature, the sample size was still relatively small. This, in addition to the retrospective nature of the work, small sample size is a limitation to interpretation, and future prospective studies with larger sample sizes will be required to confirm the results.

In conclusion, our study suggests that temporary piggyback IOL implantation in patients with congenital cataract yields worse final BCVA, more additional surgeries, and a higher rate of glaucoma development compared with single IOL implantation. Temporary polypseudophakia does not have benefit in congenital cataract and single IOL implantation.

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References