ARTICLE

Management of monocular congenital cataracts

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Abstract

Monocular congenital cataracts have been treated for the past two decades with early surgery, contact lens correction and patching of the unaffected eye. While an occasional patient has had a good visual outcome with this treatment regiment, the majority end up being legally blind in the aphakic eye. Intraocular lenses (IOLs) are increasingly being used as an alternative means of optically correcting aphakia during infancy. A growing body of literature suggests that an IOL correction is associated with an improved visual outcome and a relatively low incidence of post-operative complications. A monkey model has also been used to study the safety and feasibility of correcting aphakia in neonates with IOLs. These studies have revealed that the visual outcome is as good if not better with an IOL correction. A multi-centre clinical trial, the Infant Aphakia Treatment Study (IATS), is being organised in the United States to critically compare an IOL and contact lens correction for infantile aphakia.

Key words Aphakia, Congenital cataracts, Contact lens, Intraocular lens, Myopic shift

Until the 1970s it was generally believed that there was no means of restoring the vision in an eye with a monocular congenital cataract (MCC). However, in 1973 Frey and co-workers[1] demonstrated that a good visual outcome could be achieved in eyes with MCCs, but this only occurred rarely. Subsequently Beller *et al.*[2] demonstrated that excellent visual results could be obtained in selected children with MCCs with early treatment and exceptional contact lens (CL) and patching compliance. They emphasised the importance of initiating treatment during the sensitive period of visual development. While others have also obtained excellent visual results in an occasional child with a MCC treated with early surgery, CL correction and patching of the unaffected eye,[3] the majority of these eyes continue to have a poor visual outcome.[4–8] Obstacles to achieving a good visual outcome include a delay in diagnosis and poor compliance with CL wear and patching therapy. Theoretically a unilateral aphakic spectacle correction could be used to optically correct children who are CL intolerant; however, this generally is not practical due to compliance problems and the severe aniseikonia created.

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Contact lens

CLs have been the preferred means of optically correcting aphakia in infants[9] because they more closely simulate the optics of the crystalline lens than do spectacles. During the first 4 years of life the aphakic infantile eye undergoes a mean decrease in its refractive error of 9–15 dioptres (D).[10–12] Therefore the ability easily to change the power of a CL as the refractive needs of the eye change is a significant advantage. Excellent visual acuities have been obtained with CLs in infants with bilateral aphakia treated during the sensitive period of visual development. CL compliance is usually good for children with bilateral aphakia and if they become CL intolerant, they can be treated with spectacles.

The treatment of infants with monocular aphakia with CLs has been less successful. In published series, only 8–24% of these eyes achieve 20/40 or better visual acuity and the majority see 20/200 or worse.[13–15] Only 4 of 34 (12%) infant eyes treated for monocular cataracts at the Jules Stein Eye Institute during the 1980s achieved a visual acuity of 20/40 or better, whereas 25 (73%) were 20/200 or worse.[14] These poor visual outcomes largely arose from poor CL and patching compliance. The poor visual acuity in the aphakic eye then makes it more difficult to patch the fellow eye, which causes further visual deterioration. Ultimately, many parents abandon patching and CL treatment for their child altogether, due to the difficulty and time demands of this treatment regimen. Assaf *et al.*[15] reported that only 44% of children with unilateral aphakia were wearing their CL when they returned for follow-up appointments. The poor compliance in these patients is multifactorial but lens loss, the difficulty of inserting and removing CLs from a small child and the absence of a discernible visual benefit, since the fellow eye has normal vision, all contribute to poor CL

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compliance. The resulting poor vision in this eye then creates a number of problems for this child. First, there is a greater risk of the normal eye being injured secondary to the child's reduced peripheral vision.[16] Second, if their fellow eye becomes blind secondary to injury or disease later in life they do not have a back-up eye with useful vision. Lastly, more than two-thirds of these patients develop a sensory strabismus which is unsightly and often must be corrected surgically.[17,18]

Intraocular lens

Intraocular lenses (IOLs) are now the standard optical treatment for older children with aphakia,[19] but their use during infancy is still controversial because of concerns regarding their safety in a growing eye with an anticipated large myopic shift. Nevertheless, there is a growing body of literature describing favourable outcomes achieved with IOL implantation in infants with MCCs (Table 1). After a 17 year follow-up, Ben Ezra[20] reported 20/60 visual acuity in the pseudophakic eye of a patient who underwent a unilateral lensectomy, posterior capsulotomy and anterior vitrectomy when 10 weeks of age. Surprisingly, the refractive error in this eye only changed from +2.50 to −4.75 D over these 17 years. Dahan and Drusedau[21] recently reported their long-term results after monocular IOL implantation in 17 infants. After a mean follow-up of 7.5 years (range 2–11.5 years), the pseudophakic eyes in these 17 children had a mean visual acuity of 20/60 (range 20/30 to 20/200). The mean initial post-operative refractive error in these pseudophakic eyes was +6.4 D (range +3 to +9 D), whereas the mean last refractive error was −1.0 D (range +3.50 to −8.0 D). The mean myopic shift was 7.4 D (range −2.50 to 12.75 D), which is less than the 9–15 D reported in monocularly aphakic children corrected with contact lenses.[12,22] Interestingly in the 12 infants who had longitudinal axial length measurements, the pseudophakic eyes elongated less (mean 0.46 mm, range 0.14–0.70 mm) than their fellow phakic eyes in 8 of the 12 children.[23] An apparent retardation of axial elongation in these pseudophakic eyes may explain why the myopic shift was less than expected. The follow-up on the 14 other infants with monocular pseudophakia who have been reported in the literature has either been relatively short or the data provided have been incomplete.

In summary, the 23 monocularly pseudophakic infants with 2 years or more of follow-up who have been reported in the literature had a mean visual acuity of 20/60. While the best acuity outcome was 20/30, none of the pseudophakic eyes had visual acuity less than 20/200. Although spectacle overcorrection was needed to compensate for the initial undercorrection and subsequent myopia in these eyes, the magnitude of the myopic shift was less than expected.

Post-operative complications

A variety of post-operative complications have been reported following cataract extraction during infancy. While some of these complications develop during the immediate post-operative period, others may not develop until years later. Opacification of the posterior lens capsule is nearly universal following infantile cataract extraction[24] and for this reason a primary posterior capsulotomy is recommended.[25] Pupillary membranes develop occasionally in aphakic eyes, but occur more frequently in pseudophakic eyes, presumably because the IOL acts as scaffolding for the fibrin. Nevertheless, only 1 of the 32 (3%) eyes summarised in Table 1 developed this complication.[26] The incidence of pupillary membranes is probably influenced greatly by the surgical technique used.

Glaucoma has been reported to have an incidence ranging between 6% and 24% following infantile cataract surgery.[27–30] Angle closure glaucoma, with its onset usually in the immediate post-operative period, is much less common than open angle glaucoma, with its onset usually years after cataract surgery. Simon *et al.*[31] reported a mean interval of 6.8 years from the time of cataract surgery until glaucoma was diagnosed. Microphthalmic eyes are at an increased risk of developing glaucoma post-operatively. Glaucoma has not been reported as a complication following infantile IOL implantation, although with a longer follow-up this complication may be observed more frequently.

Strabismus has been reported to develop in two-thirds or more of children with MCCs treated with CLs.[17,18] Most of these children require strabismus surgery to achieve cosmetically acceptable ocular alignment. In contrast Ben Ezra *et al.*[18] reported that only 10% of children with monocular pseudophakia develop strabismus. The reduced incidence of strabismus in children with monocular pseudophakia compared with monocular aphakia probably stems from the constancy of the optical correction and the improved visual outcome in pseudophakic children.

Lens reproliferation into the pupillary space is a rare complication (< 1%) following infantile cataract extraction.[32] However, it has been reported to occur in 16% of infantile eyes after cataract extraction and IOL implantation. The higher incidence of this complication following IOL implantation during infancy probably stems from the greater amount of lens capsule left in these eyes.

Pupillary abnormalities are fairly common after infantile cataract surgery. The pupil may be damaged intraoperatively or posterior synechiae may cause pupil irregularities post-operatively. Pupillary capture is a relatively common complication following infantile IOL implantation, particularly if the IOL is not placed in the capsular bag.[25] Dahan and Drusedau[21] noted corectopia in 2 of 17 (12%) eyes following infantile IOL implantation. While pupillary abnormalities can be a cosmetic problem in children with light-coloured irides, they generally do not affect the visual outcome.

Table 1. *IOLs in infants less than 6 months of age with monocular congenital cataracts*

Author (year)	Country	No. of eyes	Surgical technique	IOL power (D)	Post-operative complications	Follow-up interval (years)	Initial refractive error (D)	Final refractive error (D)	Final visual acuity	Initial axial length (mm)	Final axial length (mm)
Markham *et al.* (1992)[49]	England	2	Lens aspiration only	24, 25	Posterior capsular opacification	0.25	+6.00, +3.00	N/A	N/A	N/A	N/A
Sinskey *et al.* (1994)[26]	USA	1	Limbal PC	24.5	Retrolenticular membrane Lens reproliferation into pupillary space	1.5	+8.50	+5.50	N/A	19.1	N/A
Vasavada and Chauhan (1994)[25]	India	5	Limbal PC	21–28	Posterior capsule opacification Pupillary capture Pigment deposits on IOL IOL decentration	2+	N/A	+0.50 to −5.00 (mean −3.25)	20/80–20/200	16.0–22.7 (mean 18.4)	N/A
Thouvenin *et al.* (1995)[50]	France	4	Limbal PC + anterior vitrectomy	26	Fibrous anterior chamber reach	1.2	N/A	+1 to +8 (mean +5)	N/A	N/A	N/A
Knight-Nance *et al.* (1996)[51]	Ireland	2	Lens aspiration only	N/A	Posterior capsular opacification	1.0	+1.50	−3.50	N/A	18.0, 16.4	22.6, 19.2
BenEzra (1996)[20]	Israel	1	Limbal PC + anterior vitrectomy	26	None	17	+2.50	−4.75	20/70	N/A	N/A
Dahan and Drusedau[21] (1997)	South Africa	17	Limbal PC + anterior vitrectomy	25–27	Corectopia (2) Lens reproliferation into pupillary space (4)	Mean 7.5 (range 2–11)	Mean 6.4	Mean −1.0	Mean 20/60 (range 20/30–20/200)	Mean 18.41	Mean 22.57

PC, posterior capsulotomy.

Cystoid macular oedema is a common complication following cataract extraction in adults, but occurs only rarely following infantile cataract extraction with or without IOL implantation.[33,34]

IOL decentration may develop following IOL implantation during infancy.[25] The type of IOL used and the placement of the IOL probably influences the incidence of this complication. Intracapsular placement of the IOL probably reduces the incidence of this complication.

Other post-operative complications such as endophthalmitis, retinal detachment and corneal oedema occur rarely following infantile cataract extraction.[35] IOL implantation does not seem to influence the relative frequency of these complications.

The adverse psychological effects of the treatments prescribed for MCCs are not known. Robb and co-workers[36] discontinued patching after noting reclusive behaviour in one child undergoing occlusion therapy. Smith and co-workers[37] were unable to demonstrate using two standardised parental report measures (The Minnesota Child Developmental Inventory and Child Behaviour Checklist) a difference between 22 children with monocular aphakia and 18 normal siblings. While impaired reading has been reported in children with bilateral aphakia and reduced vision,[38] the cognitive ability of children with unilateral aphakia is considered generally to be normal.

Monkey model of monocular aphakia/pseudophakia

In contrast to the limited data on outcomes following the correction of infantile monocular aphakia in humans with IOLs, the Vision Section at the Yerkes Regional Primate Research Center has published extensively on the visual outcomes, post-operative complications, effects on ocular growth and histopathological findings associated with both CL and IOL correction of infantile monocular aphakia using a Rhesus monkey model in which treatments were standardised and compliance carefully monitored. Since the Rhesus monkey has eyes anatomically similar to human eyes, albeit approximately 20% smaller, and nearly the same visual acuity as humans, these findings should be highly predictive for similar studies in human infants.

Patching and visual outcome

Patching therapy was critical to an excellent visual outcome in both CL- and IOL-corrected monocularly aphakic monkeys.[39] While the mean logMAR acuity of the pseudophakic eyes in the IOL group treated with 70% occlusion therapy of the fellow eye (mean = 0.66, $n = 10$) versus no patching (mean = 0.93, $n = 9$) was not significantly different ($p > 0.05$) at 32 weeks of age using forced choice preferential looking (FPL),[40] when retested at 2–4 years of age using a more reliable operant testing procedure the acuity of the pseudophakic monkeys receiving patching therapy was significantly better (mean = 0.67 (Snellen equivalent ~20/80), $n = 6$ versus mean = 1.2 (Snellen equivalent ~20/230), $n = 3$; $p < 0.01$).[41] In the CL group, a significantly better visual outcome was obtained in the aphakic monkey eyes that received part-time occlusion of their fellow eyes compared with those that did not at 32 weeks using FPL.[39] Unfortunately operant testing was only completed on 2 of the CL group monkeys when they were older and both received occlusion therapy, so it is not known whether this difference increases over time. For the monkeys that had 70% occlusion therapy, the difference in visual acuity using operant testing was not significant (largely because of the small sample size).[42] Studies using different patching regimens suggest that the best visual outcome can be achieved with as little as 50–70% patching therapy of the fellow eye.[39] Stereopsis could not be demonstrated in any of the monocularly aphakic or pseudophakic monkeys.[43,44]

Post-operative complications

Surgically removing the crystalline lens and implanting an IOL in a neonatal monkey eye resulted in more post-operative complications that simply removing the lens. A pupillary membrane, the most common complication, developed in 100% of 21 pseudophakic eyes and 55% of 40 aphakic eyes ($p < 0.01$).[43] Presumably, the IOL acts as scaffolding for fibrin, increasing the incidence of this complication. These membranes generally develop within 1 week of surgery. Lambert *et al.*[43] have used a variety of surgical techniques to open these membranes including needle discission, injecting tissue plasminogen activator into the anterior chamber, surgical excision and YAG membranectomy. YAG membranectomy was found to be the most effective treatment, although multiple treatments were often required. The severity of this complication was reduced by adding heparin to the infusion solution and avoiding any manipulation of the iris. Lens regeneration into the pupillary space also occurred in a higher percentage of pseudophakic than aphakic eyes (28 vs 6%, $p < 0.05$), presumably because more lens epithelial cells were left in the pseudophakic eyes. This complication was treated either by aspirating the lens material through a limbal incision when the regenerated lens material was in the anterior chamber or by a pars plana vitrectomy when the lens material was in the vitreous chamber. Glaucoma developed in a similar percentage of eyes with PMMA IOLs and aphakic eyes (9.5% vs 12.7%, $p = 0.34$). However, glaucoma developed in 7 of 9 (83%) eyes after the implantation of an acrylic IOL. Other complications developing in pseudophakic eyes included papillary capture of the IOL and haptic breakage.[43] These complications are largely preventable by careful attention to surgical technique and IOL design.

Ocular growth

Ocular growth was retarded by the removal of the crystalline lens during the neonatal period.[45] The effect was similar whether the eye was left aphakic or an IOL was implanted. After a 2 year follow-up, pseudophakic

Table 2. *Pros and cons of intraocular lens (IOL) versus contact lens (CL) correction of neonatal monocular aphakia*

Intraocular lens	Contact lenses
Arguments for:	
1. Closely approximates the optics of the crystalline lens	1. Power can easily be reduced as the eye grows
2. Full-time partial optical correction is guaranteed correction	2. Secondary IOL can be implanted when the child is older when the refractive error of the eye is stable
3. Monkey data show better visual outcome than CL	
4. Limited human data show better visual outcome than CL correction	
5. Lower incidence of sensory strabismus	
Arguments against:	
1. Their long-term safety in a growing eye has not been established	1. Fifty per cent of patients with this treatment have visual outcome $\leqslant$ 20/200
2. The surgery required to implant an IOL is technically more difficult	2. Poor compliance with CL wear may reduce patching compliance
3. An overcorrection with spectacles or CLs is needed initially or after the eye is fully grown	3. Lenses are frequently lost and there may be a delay in their replacement
4. Limited human data show higher complication rate	4. Maintenance takes time each day and can be stressful for patients and parents

eyes were 2.0 ± 0.2 mm shorter than their fellow eyes compared with a difference of 2.3 ± 0.2 mm between aphakic eyes and their fellow eyes. Part-time occlusion of the fellow eye and visual outcome were not correlated with the degree of axial elongation. To minimise the refractive error of these monkeys once they were adults, they were initially undercorrected with the IOL and given a CL overcorrection. After a 4-year follow-up, the pseudophakic eyes experienced a mean myopic shift of 10.7 D (range 5–20 D).[46]

Histopathology

One of the primary advantages of a monkey model of aphakia and pseudophakia is that the eyes can be studied histopathologically. Almost all the changes observed were in the anterior segment. They included angle closure in the eyes developing glaucoma, Soemmerring rings, haptic erosion into the iris root and a small reduction in the corneal endothelial cell count.[47,48] The only change reported in the posterior segment was axonal loss in the monkeys developing glaucoma.

In summary, an improved visual outcome was obtained in the monkeys that were corrected with IOLs. In addition, the myopic shift was smaller than anticipated due to a retardation of axial elongation in these eyes. While more complications developed in the pseudophakic eyes, the incidence of these complications was reduced to an acceptable range by improvements in surgical technique and IOL design. When the pseudophakic eyes were examined histopathologically, the IOLs appeared to be well tolerated.

Conclusions

Both monkey and human studies show an improved visual outcome with IOL treatment. While the ability to change the power of a CL to compensate for ocular growth is a significant advantage, the difficulty of achieving full-time CL compliance and the increased time demands of this treatment are significant disadvantages (Table 2). Certainly, if more complications develop in the eyes with IOLs, either because the surgery is technically more difficult to perform or because the IOLs are poorly tolerated in these infantile eyes, then the relative costs and benefits for each of these treatments will have to be weighed against each other. The Infant Aphakia Treatment Study (IATS) is currently being organised as a multi-centre clinical trial in the United States to compare these two treatments.

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