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Factors that influence the visual outcome in cases of infantile unilateral cataract

CLARE DEWSBERY DBO(T)

Eye Department, Diana, Princess of Wales Children's Hospital, Birmingham

Abstract

Aim: **To review retrospectively the records of a cohort of patients who presented before 6 months of age with unilateral cataract and identify factors that influenced the visual outcome.**

Method: **The records of children with unilateral infantile cataract, who had been examined since 1993 following the establishment of a new occlusion protocol, were reviewed. For analysis the children were divided into two groups: group A with visual acuity of 0.6 logMAR or better and group B with visual acuity of <0.6 logMAR. Factors examined were referral history, type of cataract, age at surgery, contact lens wear, ophthalmic complications, occlusion therapy, attendance record and distance from home.**

Results: **Thirty-two children met the inclusion criteria; 8 transferred to other hospitals leaving 24 cases for analysis. There were 11 (46%) children in group A with visual acuity ranging from 0.6 to 0.04 logMAR and 13 (54%) children in group B with visual acuity ranging from NPL to 0.74 logMAR. The median age at surgery was 8.5 weeks (13 weeks, group A; 7 weeks, group B). There was a greater incidence of ophthalmic complications in group B. Nystagmus was present in 9 (38%) children, 8 of who were in group B, and this was a significant factor in visual outcome ($p = 0.013$). Compliance with the occlusion protocol was also a significant factor in visual outcome ($p = 0.016$). Further analysis indicated an association between occlusion compliance, visual outcome and distance from home ($p = 0.035$).**

Conclusion: **A high incidence of ophthalmic complications, a delay in establishing daily contact lens wear and a failure to achieve 'good' compliance with occlusion therapy were associated with a poor visual outcome.**

Key words: Congenital, Infantile, Unilateral cataract, Visual outcome

Introduction

It is generally accepted that early diagnosis of unilateral infantile cataract with prompt surgery is imperative to ensure the briefest period of visual deprivation and increase the chances for a good visual outcome.[1–7] Surgery before 6 weeks is reported to increase the chance for a good visual outcome.[8,9] In a case reported by Gregg and Parks where surgery was performed at 1 day of age and a contact lens inserted at 2 days, visual outcome at 8 years was 20/25 (0.1 logMAR) and stereoacuity of 50 sec arc.[10] Birch *et al.* reported that visual deprivation is the only amblyogenic factor in the first few weeks of life.[3] However, if unilateral deprivation is prolonged to 12–30 weeks then unequal binocular cooperation may play a part in the amblyogenesis. Unilateral cataract patients are reported to show reductions in contrast sensitivity similar to those in bilateral cases when treatment is instigated before 8 weeks of age. When treatment occurred later the deficit was greater.[3]

The initial refractive treatment of choice in unilateral cataract in young babies is contact lenses because of their ability to compensate for ocular growth. This is an advantage over intra-ocular lens (IOL) implants.[11] Contact lens compliance can be a problem and has demands on the carer's time.[7] A growing body of literature shows that IOLs, as opposed to contact lens correction of induced aphakia, improves the visual results but increases the risks of complications.[11–14] IOLs are usually the preferred option for toddlers and older children but are being used more in younger babies. Stress levels are reportedly higher in parents of children with contact lenses compared with parents of pseudo-phakic children.[15]

Surgery and refractive correction is followed by occlusion treatment. The period of occlusion of the phakic eye is still a matter for debate and subject to various protocols between centres.[8] Some advocate an aggressive patching regime,[1,4,16] while others advise a less strenuous one to encourage the restoration of binocular single vision in those having very early cataract extraction.[4,5] Occlusion compliance has been found to be a critical factor for a good result,[5,7,9,17,18] with compliance improving when parental preferences and views are considered.[19]

Even with early surgery the visual outcome in infantile unilateral cataract can be disappointing. Some consider vision of 1/60 a significant improvement as a reserve eye.[11] Rahi *et al.* report that the risk of severe visual loss affecting the non-amblyopic eye is much greater than previously thought.[20] The risk of an amblyopic patient becoming blind has been reported to be higher than for the general population.[17] With life

Correspondence and offprint requests to: Clare Dewsbery, Eye Department, Diana, Princess of Wales Children's Hospital, Steelhouse Lane, Birmingham B4 6NH. Tel: (0121) 3339468. e-mail: clare.dewsbery@bch.nhs.uk

Total occlusion of the unaffected eye
100% of waking time (except feeding) for 1 month

Occlusion according to the visual difference between the two eyes:

Children < 2 years of age using the Teller Acuity Test

2 octaves (4 lines)	**100% waking time (except feeding)**
0.5–1.5 octaves	**75% waking time**
0.5 octaves	**50% waking time**

Children > 2 years of age *or* when they transfer to other vision tests

<1.0 logMAR	**12 hours**
0.7–1.0 logMAR	**8 hours**
0.4–0.6 logMAR	**6 hours**
0.2–0.3 logMAR	**4 hours**

If the vision remains stable at the same level for three consecutive visits, then occlusion is stopped.
If the vision drops again, then occlusion is restarted using the above regime

Fig. 1. Occlusion therapy protocol following surgery for unilateral cataract adopted by the department in 1993.

expectancy improving there is an increased risk for visual impairment as a result of eye disorders in the elderly. If the disorder affects the phakic eye it would seriously incapacitate an individual who did not have reasonable vision in the aphakic eye.[9]

The aim of this retrospective study of unilateral infantile cataract patients was to identify factors that influence visual outcome and determine any which may influence our current management protocol.

Methods

The case notes of all children who had presented to the department with unilateral cataract since 1993, following the establishment of a new occlusion protocol, were examined. The protocol had been devised following extensive auditing within the department over several years and is shown in Fig. 1. Data for children who had been operated on in the first 6 months of life and been followed up until the age of $\geq$3 years of age were considered for analysis. The age of 3 years was chosen as visual acuity would be assessed using recognition acuity tests.

On the basis of visual outcome of the aphakic eye the children were divided into two groups. Group A comprised children with visual acuity 0.6 logMAR or better and group B comprised children with visual acuity of <0.6 logMAR.

The referral history, type of cataract, age at surgery, associated ophthalmic problems, contact lens history, distance from the hospital and attendance record were examined. The patient's compliance with the occlusion protocol was categorised into good, fair or poor by comparing the amount of occlusion prescribed with the amount the parents reportedly achieved. 'Good' compliance was recorded when the occlusion reported adhered at all times to the occlusion prescribed. The weeks when the parents reported no occlusion were added together and taken as a percentage of the duration of occlusion treatment. 'Poor' compliance was recorded in those children where no occlusion therapy had been obtained for more than 20% of the total occlusion period. Any other compliance history was regarded as 'fair'.

The data for group A and group B were compared. Categorical data were analysed using the Fisher exact test. Comparisons between outcomes of continuous variance were analysed using the Mann-Whitney test.

Results

A total of 32 children met the inclusion criteria. Two had surgery only and returned to the referring hospital. Six cases were transferred to other hospitals before their third birthday. The data for 24 children remained for analysis. Of these children, 13 were female and 11 male and their ages ranged from 3 to 11.3 years.

Data for the children in group A (child A to child K) and group B (child L to child X) are shown in Table 1. The comparison of data for groups A and B is summarised in Table 2.

Visual outcome

There were 11 (48%) children in group A with visual acuity ranging from 0.04 to 0.6 logMAR. Group B contained 13 (54%) children with visual acuity of 0.74 logMAR to NPL. The visual acuity of the unaffected eye ranged from 0.36 to −0.04 logMAR. The median age at the last visual acuity test was 5.92 years (5 years for group A; 6.58 years for group B).

Referral and cataract history

For 9 of 17 children an ocular problem was queried by 1 week of age ($n = 4$ for group A; $n = 5$ for group B).

Table 1. Visual outcome and other results for group A (child A to child K) and group B (child L to child X)

Child	LogMAR VA of aphakic eye	LogMAR VA of unaffected eye	?Linear VA test	Age at last VA test (years)	Initial observations	Cataract type or description	Age at extraction (weeks)	Distance to home (miles)	Appointments missed	Ophthalmic factors	Age at which daily wear of contact lens achieved (months)	Time where no lens in eye (months)	Age occlusion ended (years)	Reversal of amblyopia	Compliance with occlusion therapy
Group A															
A	0.04	–0.04	Yes	9.33	Leucocoria at **1 day**	Nuclear lamellar cataract	8	35	0		6	0	3.08	Yes	Good
B	0.14	0.1	Yes	10.83	GP at **6 week** check	Dense cortical cataract	10	45	0	Loose suture	11	0	7	Yes	Good
C	0.3	0.225	Yes	4	No red reflex – GP at **8 weeks**	Cortical lens opacity	13	20	0		6	0.25	Still	No	Fair
D	0.36	0.0	Yes	7.17	Poor red reflex at **9 weeks**	Nuclear cataract and very slight microphthalmos	16	45	0	Slight microphthalmos, astigmatism	13	0	5.25	Yes	Good
E	0.375	0.2	Yes	4.42	Leucocoria ? when	Dense nuclear and posterior cortical cataract	11	12	3		5	0	Still	No	Fair (atropine)
F	0.425	0.25	Yes	3.58	? cataract at **4 days**. Mum ? red reflex at **17 weeks**	Dense cataract	18	21	0		13	0	Still	No	Good
G	0.475	0.15	Yes	3.75	Lens opacity – postnatal check at **few days**	Central posterior cataract	7	30	2		3	0.25	Still	No	Good
H	0.48	0.16	Yes	5.83	Mum felt LE 'odd' from infancy	Post-subcapsular lenticular opacity	26	100	0	**Nystagmus,** loose suture, astigmatism	11	0.5	Still	Yes	Good
I	0.5	–0.075	Yes	5	Cataract at **4 weeks**	Small nuclear cataract– patched. By 13 weeks thickened– white pupil. Surgery delayed–ill	20	30	0		13	1.75	Still	Yes	Good
J	0.55	0.05	No	3.66	No red reflex – GP at **8 weeks**	Posterior subcapsular cataract	11	15	0		12	2	Still	No	Fair
K	0.6	0.1	Yes	6	Opacity – paediatrician at **1 week**	Total zonillar cataract	18	25	0	Microphthalmos	?	?	6	No	?

Group B															
L	0.74	0.18	Yes	10.66	No red reflex – GP at **8 weeks**	Dense cataract	14	10	0	**Nystagmus**	5	0	4.33	Yes	Fair (atropine)
M	0.85	0.25	Yes	4.33	?	Dense central opacity	4	15	0	**Nystagmus,** slight PHPV	5	0	Still	No	Fair
N	1.26	0.36	Yes	10.42	Leucocoria at **6 weeks**	Dense cataract	8	34	0	**Nystagmus**	4	0	5.5	Yes	Fair
O	1.34	0.1	Yes	7.75	Leucocoria at **6 weeks**	Dense nuclear lens opacity	9	5	3	Slight PHPV, pupil anomaly, meibomium cyst, pupillary membrane, vitrectomy	3	4.25	6.25	No	Fair (atropine)
P	1.5	0.0	No	3.5	?	Dense lens opacity	9	10	6	Slight macular haemorrhage post-operatively	No – IOL	4.75	3.5	No	Poor (atropine)
Q	1.6	0.3	Yes	7.25	No red reflex at **birth**	Congenital cataract	5	30	0	**Nystagmus,** loose suture	16	2	5	No	Fair (atropine)
R	1.62	0.04	Yes	6.58	No red reflex at **1 day**	Congenital posterior lens opacity	8	15	2		9	1.5	5.92	No	Fair
S	1.7	0.075	No	3	Leucocoria at **birth**	White cataract	5	2	5	**Nystagmus,** abscess keratitis	22	0.5	? moved	No	Poor (atropine)
T	1.9	0.12	Yes	6.58	No red reflex at **2 days**	Significant PHPV on back of lens	3	109	1	**Nystagmus**	5	0.5	5.25	No	Good
U	CF	0.0	Yes	11.33	?	Dense cataract	7	40	2	**Nystagmus,** PHPV, slight glaucoma, microphthalmos	17	41	3.92	No	Fair
V	PL	0.12	Yes	3.17	?	Dense cataract	5	10	10		38	23.5	3	No	Poor
W	PL	0.1	Yes	6.25	Leucocoria ? when	Dense cataract	13	3	8	**Nystagmus,** microphthalmos	12	>10	3.42	No	Poor (atropine)
X	NPL	0.125	Yes	4.83	Poor red reflex at **few days**	Dense cataract	7	12	0	Glaucoma	No		2.42	No	Good

Table 2. Comparison of results between groups A and B

Criteria	Whole study ($n = 24$)	Group A ($n = 11$)	Group B ($n = 13$)
Sex	13F, 11M	5F, 6M	8F, 5M
VA of aphakic eye (logMAR)			
Range	0.04 to NPL	0.04 to 0.6	0.74 to NPL
VA of phakic eye (logMAR)			
Median		0.10	0.12
Range		−0.08 to 0.25	0.00 to 0.36
Age at last vision test (years)			
Median	5.92	5	6.58
Range		3.55–10.83	3–11.33
Age cataract first observed (weeks)			
Median		4	0.39
Range		0.14–9	0.0–8
Age at extraction (weeks)			
Median	8.5	13	7
Range	3–26	7–26	3–14
No. of children having surgery < 12 weeks of age	16	5 (46%)	11 (85%)
Distance from hospital (miles)			
Median	20.5	30	12
Range	2–109	12–100	2–109
No. of patients attending all appointments	14 (58%)	9 (82%)	5 (39%)
No. failing >4 appointments	4	0	4
Average distance to home (miles)	6.25		6.25
Nystagmus	9 (38%)	1 (9%)	8 (62%)
Glaucoma	2 (8%)	0	2 (8%)
Strabismus	24 (100%)	11 (100%)	13 (100%)
No. of ophthalmic complications	29	7 (24%)	22 (76%)
No. of children with no complications	9 (38%)	7 (64%)	2 (16%)
Age at which daily wear of contact lens achieved (months)			
Median		11	9
Range	3–38	3–13	3–38
Time without contact lens (months)			
Median	1.87	0.13	1.75
Range	0–23.5	0–2	0–23.5
No. of children reporting 'good' compliance with occlusion protocol	9 (39% of 23)	7 (76% of 10)	2 (16%)
No. of children reporting 'poor' compliance with occlusion protocol	4 (17%)	0	4 (17%)
No. of children having atropine occlusion	7 (29%)	1 (9%)	6 (46%)
No. of children showing reversal of amblyopia	7 (29%)	5 (45%)	2 (15%)

Records were incomplete in 7 cases. One child (child I) had a history of an initial small cataract that became dense during management. In child F a cataract was initially suspected at 4 days old and dismissed, but later a poor red reflex was noted at 17 weeks of age.

Surgical history

Sixteen (67%) children had cataract surgery younger than 12 weeks of age ($n = 5$ for group A; $n = 11$ for group B). The median age at surgery was 8.5 weeks, but extraction was performed on average later in group A than group B (13 weeks for group A; 7 weeks for group B).

Ophthalmic complications

There was a greater incidence of ophthalmic complications in group B ($n = 11$, 85%) compared with group A ($n = 4$, 36%). Complications included microphthalmos, loose sutures, astigmatism, pupil anomaly, abscess keratitis and posterior hypoplastic primary vitreous (PHPV). Two cases (8%) had glaucoma, one severely. All children had strabismus. Nystagmus was reported in 9 (38%) children, 8 of whom were in group B, and this was statistically significant for visual outcome ($p = 0.013$, Fisher exact test).

Contact lens wear

All children had soft contact lenses fitted at surgery or soon after. One of the patients had an IOL fitted later. Glasses with +3.00 reading add to the aphakic eye were used during the occlusion period at approximately 3 years of age, when the contact lens power prescribed was changed from an intermediate to a distance correction.

The mean age at which daily insertion and removal of contact lenses was achieved was 9 months for group A compared with 11 months for group B. The parents of 2 children in group B never achieved daily insertion and removal. The median time children were without their contact lens was 0.13 months for group A and 1.75 months for group B.

Occlusion

The duration of occlusion was generally more than 2½ years. In group A, 7 (76%) of 10 children (the notes of 1 child were incomplete) had achieved 'good' compliance with occlusion therapy and had a visual acuity of 0.5 logMAR or better. Only 2 children in group B had 'good' compliance and all cases of 'poor' compliance were in group B. The reported compliance with occlusion was significant for visual outcome ($p = 0.016$, Fisher exact test).

Table 3. Association of distance from home, missed appointments and occlusion compliance

Reported occlusion compliance	'Good'	'Fair'	'Poor'
Median distance from home (miles)	35	15	7
Median no. of appointments missed	0	0	7

Atropine occlusion (instilled once daily) was used in 7 cases when parents were having problems with conventional occlusion and was continued until they were able to revert back to the conventional occlusion; only 1 case was in group A.

Seven children had episodes of reversal of amblyopia during their occlusion therapy. This was established by accurate visual assessment and/or reversal of deviation seen on cover test. Five of the cases were in group A and 2 in group B.

Distance from home

The children in group A lived from 12 to 100 miles from the hospital, with a median of 30 miles. The distance for group B ranged from 2 to 109 miles with a median of 12 miles. When comparing distance from home and reported occlusion compliance it was found that the median distances were 35, 15 and 7 miles in the good, fair and poor groups respectively (Table 3).

Attendance

Fourteen children attended all appointments, 9 (82%) in group A and 5 (39%) in group B. Four children, who lived an average of 6.25 miles from the hospital, failed 5 or more appointments and all these children fell into group B. When comparing attendance with reported occlusion compliance it was found that the median of the number of missed appointments was 7, 0 and 0 in the poor, fair and good groups respectively (see Table 3).

A statistically significant association between distance from home, reported occlusion compliance and visual outcome was found ($p = 0.035$, Mann-Whitney test).

Discussion

A retrospective review of 24 patients with unilateral infantile cataract who presented before 6 months of age revealed that 46% of the children attained a good visual outcome with a visual acuity of 0.6 logMAR or better. Eight children were still having occlusion therapy, so the final outcome visual acuity is not known. It is hoped the vision in these children may improve further. It is generally considered that 0.6 logMAR or better is a good result in these cases.[2,9] Birch *et al.* reviewed 19 studies and found that of the 362 patients reviewed, 36.8% of children over the series of studies achieved this level of acuity or better.[9]

Early surgery is reported to increase the chance for a good visual outcome.[8,9] All but 2 children in group B had their surgery by 8 weeks of age, but group B children still had a poor visual outcome. Secondary ophthalmic complications can be an obstacle to a good visual result[18,21] and this was evident in this case series. There was a lower incidence of complications in group A and these were less serious than the complications noted in group B.

Two (8%) children in this case series developed glaucoma and they both showed very poor visual outcome. Secondary glaucoma in children having early surgery is reported in the literature. Vishwanath *et al.* examined a cohort of 80 patients having a total of 128 lensectomies for congenital cataract within the first few weeks of life. They found that 5 years after surgery there was a risk of glaucoma in 15.6% of the eyes.[22] Egbert *et al.*'s prospective study considered the prevalence of ocular hypertension and glaucoma 5 years after lensectomy for unilateral cataract and found that of the 40 children examined 13% had glaucoma and 45% had ocular hypertension.[23]

Watts *et al.* examined the prevalence of post-operative complications in 55 cases of congenital bilateral and unilateral cataract having surgery within the first 12 weeks of life.[24] They reported nystagmus in 33% of the cases. The incidence in this case series is similar, with 7 (29%) children showing nystagmus within the same extraction time criteria, although a total of 9 cases had nystagmus. Only one of the 9 cases had a good visual outcome and analysis indicated nystagmus was a statistically significant factor in visual outcome.

Contact lens wear is less successful in the management of unilateral cataract than in bilateral cataract.[12] This may be because there is little visual incentive when the fellow eye has normal vision. Furthermore there may be loss of lens, infections may ensue and there may be considerable problems with insertion and removal. Lambert quotes Assaf *et al.*, who reported only 44% of children wearing their contact lenses at return appointments.[12] However, in this case series only 3 children showed significant problems with contact lens wear. It appeared easier to learn insertion and removal when the child was a baby and could be easily restrained, than later when an active toddler. The children whose parents learnt insertion and removal techniques later had more infections which interrupted the occlusion regime, which in turn may have caused resistance to patching.

Analysis showed that those who complied with occlusion treatment had a better visual outcome than those who did not. The period of occlusion prescribed for aphakic children is still a matter of debate and subject to varying protocols between centres. It is reported that occlusion of up to 8 hours daily from early infancy is possible without producing detrimental effects on the sound eye.[9,16] Parental support with occlusion therapy is paramount.[5] A study by Chua *et al.* found that the duration of treatment was not associated with patching compliance.[25] Parkes stated families of children with dense and moderate amblyopia appeared to be less compliant with occlusion therapy than those with milder amblyopia.[26]

Some parents in this case series had described the importance of establishing an 'occlusion habit' that became part of the child's daily regime. This was especially important in the toddler stage when compliance with occlusion was often more difficult. It was at

this age more than any other that atropine occlusion was used. Occlusion therapy began when the child was only a few months old when compliance by the child was not an issue. If the vision improved sufficiently by the time the child reached the difficult toddler stage and the 'occlusion habit' had been established the child would experience less visual reduction during patching. If this habit was broken, the amblyopia increased, the resultant visual 'difficulty' during occlusion was increased and thus compliance reduced.

Those children who lived further away from the hospital had a better outcome than those who lived closer. This may be a socio-economic factor, i.e. inner city versus suburban family, and/or the parents of these children were more motivated and distance to travel was not an issue. It was unclear whether the 6 patients who were transferred to other hospitals did so because the distance was a problem. Those children with the better attendance record achieved a better visual outcome. Some of those with the worst attendance records lived closest. These factors are features of compliance and analysis indicated a significant statistical association between those living further away, good attendance, 'good' compliance with occlusion and a good visual outcome.

There is a concern that the vision of the phakic eye may be compromised by aggressive and protracted occlusion regimes. Some studies have confirmed this.[5,27] Lewis *et al.* reported that the vision sensitivity of the 'good' eye of children treated for unilateral congenital cataract is on average slightly reduced even in cases with minimal patching.[28] Therefore the deficit discovered after aggressive occlusion may not have been caused by occlusion amblyopia. Lloyd *et al.* showed there was no obvious reduction of visual acuity of the phakic eye.[29] The children in this case series who achieved a vision of 0.2 logMAR or less in the unaffected eye were either quite young or had nystagmus. The children who showed 'good' compliance to the occlusion protocol had good vision of the phakic eye. Seaber and Buckley comment that maintenance occlusion is essential throughout childhood to cement visual gain.[1] Our occlusion protocol has recently been changed to include low levels of maintenance occlusion.

As all the children reported developed a squint it is possible that a further review of our occlusion protocol is required. Other studies have used less aggressive occlusion regimes to encourage the development of binocular single vision. However, the number of children in this series with vision of 0.6 logMAR or better is well above the average reported in the literature. Therefore the potential for good visual outcome needs to be balanced against the potential for developing binocular single vision.

Reversal of amblyopia during occlusion occurred very early in treatment and in all cases the resultant amblyopia of the phakic eye was retrievable. One child (child B) showed five separate incidences of reversal and another (child A) showed reversal for several months. Both children have essentially normal vision in both eyes. Reversal of amblyopia still makes orthoptists and ophthalmologists anxious; however, all but one child who showed reversal of amblyopia had vision of 0.74 logMAR or better in the aphakic eye. The high incidence of reversal amblyopia suggests that the occlusion protocol should be reviewed or the frequency of visits for visual acuity monitoring increased in the early stages of treatment.

Conclusions

The factors that influenced visual outcome were multifactorial. Whilst early surgical intervention is important, the children who had surgery by 8 weeks did not have a good visual outcome. A high incidence of ophthalmic complications, a delay in establishing daily contact lens wear and a failure to achieve 'good' compliance with occlusion therapy were associated with a poor visual outcome. The importance of maintaining an 'occlusion habit' cannot be underestimated, as interruptions break the routine of patching and make returning to occlusion much harder.

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