

Review

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Posted Date: 18 September 2025

doi: 10.20944/preprints202509.1610.v1

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Review

Visual Outcomes in Delayed Ophthalmic Procedures After Early and Prolonged Childhood Blindness in Adults

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Abstract

Delayed ophthalmic procedures in amblyopic-age children may lead to irreversible visual impairment. The impact of delayed ophthalmic care in adults after early and prolonged childhood blindness has not been well studied. We conducted a review of historical cases to examine visual outcomes in the setting of delayed ophthalmic procedures after early and prolonged childhood blindness due to bilateral visual deprivation. Based on 11 cases which we identified, the best visual acuity which reasonably can be expected after surgery as an adult for severe visual impairment throughout childhood appears to be about 20/200, following a period of visual recovery and rehabilitation, which can take 1 to 1.5 years. These findings underscore the importance of prompt intervention for individuals with visual impairment and better define the best possible outcome when intervention is delayed.

Keywords: visual outcomes; congenital cataracts; sight restoration

1. Introduction

In 1688, Irish philosopher William Molyneux (1656–1698) asked his colleague, English physician and philosopher John Locke (1632–1704), a question which has stimulated multi-disciplinary scholarly work ever since. Molyneux asked whether a congenitally blind man suddenly obtaining eyesight would be able to visually distinguish a cube from a sphere if he had previously been familiar with these shapes using his sense of touch [1]. Since then, congenital cataract surgery has been the predominant clinical approach towards answering Molyneux's question. Nonetheless, cataract surgery poses certain limitations in this regard. In some instances, surgery can rapidly restore vision in cataracts which are assumed to be congenital, but deeper investigation cannot establish that the cataracts were truly congenital.

Existing studies have examined visual outcomes in delayed ophthalmic procedures, primarily in the pediatric population. For example, delayed surgery for a young child with congenital cataracts may result in irreversible visual impairment due to amblyopia [2]. In a prospective study conducted in India, investigators reported that children who had late treatment of early-onset dense cataracts experienced significant visual improvement [3]. These children all presented with cataracts within 6 months of birth and underwent treatment at 8 years of age or older. Mean logMAR acuity for best corrected visual acuity (BCVA) at distance was 2.19 preoperatively (Snellen: 20/2000) and improved to 1.32 postoperatively (Snellen: 20/250). Similarly, a retrospective multicenter review of shorter delays in treatment of congenital cataracts reported that children whose surgery was delayed past 10 weeks of age had generally worse visual outcomes compared to children whose surgery was

performed sooner [4]. For example, among infants who had surgeries delayed past 10 weeks of age, BCVA was never better than 20/100.

These previous findings highlight that delayed ophthalmic management in amblyopic-age children can improve visual outcomes, although perhaps not as well as treatment performed more quickly. While congenital cataracts generally are addressed urgently, during a longer period in early childhood, up through at least age 7 years, the brain circuitry is plastic, and the visual system continues to develop [5]. The critical period refers to the peak window in which deprivation, strabismus, or anisometropia may result in permanent visual impairment, leading to amblyopia [6]. During this period, children respond best to treatment with patching or atropine drops, and past this period, children have a less favorable response to the same treatment [7]. Nevertheless, most of the existing research has not explored the outcomes of delayed ophthalmic management in adults.

2. Materials and Methods

We conducted a review of 11 historical cases to examine visual outcomes that involved ophthalmic procedures after early and prolonged childhood blindness due to bilateral visual deprivation [8]. We obtained these cases from a single systematic review conducted by May et al. on the impact of delayed access to treatment for blinding conditions [8]. Early blindness was defined as within the first year of life, and prolonged blindness was defined as lasting through age 17 years or longer. From these cases, we obtained clinical information on their visual impairment, procedures that they underwent, baseline visual acuity, and postoperative visual acuity (Table 1).

3. Results

A systematic review identified cases, from 1826 to the present day, of delayed ophthalmic procedures to improve vision after early and prolonged childhood blindness [8]. The procedures involved cataract surgery, iridotomy, corneal transplantation, and prescription of aphakic spectacles. From this review, we identified 11 patients who: 1) had severely limited visual acuity before the age of 1 year, and 2) had an ophthalmic procedure performed to improve visual acuity at age 17 years or greater (Table 1). All 11 patients had some description of postoperative visual function, though it was not always in a standard form (such as Snellen or LogMAR visual acuity). Only half the patients were reported to see 20/400 or better after intervention. These patients were generally in the range of 20/250, although one patient had visual acuity of 20/100. This patient had spontaneous resorption of infantile cataracts and was corrected with aphakic spectacles at the age of 24 years [9]. Thus, the poor visual acuity was due to refractive error plus amblyopia, rather than media opacity. Based on these reports, the best visual acuity which reasonably can be expected after surgery for severe visual axis opacities throughout childhood appears to be about 20/200, and this follows a period of visual recovery and rehabilitation.

Table 1. Patients with poor vision by the age of 1 years who had corrective ophthalmic procedures from age 17 years onward.

Patient	Clinical Information	Procedure	Pre-Op VA	Post-Op VA
46-year-old female (Wardrop 1826) [10]	Congenital cataract surgery (possibly cataract extraction) bilaterally at age 6 months. After the operation, she "lost the whole globe of one, and the pupil of the other became closed". After her iridotomy at age 46 years, she was able to see that smaller objects, such as a pencil case and a key, were present, even if she could not identify or distinguish them. After three weeks, she was able	Artificial pupil (iridotomy)	Partially sighted. Able to distinguish very light and very dark chamber & gloomy day and sunshine. Unable to perceive objects, have any notion of colors	After first & second iridotomy attempts, improved to perceive more light but could not perceive forms or colors. After third procedure, able to perceive objects but not able to tell what she was seeing.

	to perceive size of objects, motions, depth, and colors.			
17-year-old male (FJ, Franz 1841) [11]	Bilateral congenital cataract couching/division as a toddler. His right eye was "atrophied" after complications from cataract surgery. Nystagmus developed. At age 18 years, the patient claimed to be able to see objects held one inch from the left eye. At age 18 years, a residual lens fragment was extracted from the visual axis of the left eye. Three days after this procedure, upon opening his eye, he had light perception only. Five days postoperatively, he began to see vague shapes and be able to control his ocular muscles slightly. For five weeks, he had sensitivity to bright colors.	Extraction of cataract fragment, monocular, and strabismus correction	Light perception	Improved to recognize light, colors, objects with difficulty in object recognition, depth perception. Also experienced confusion and was overwhelmed by large number of things he saw. He was able to recognize different colors (except yellow and green). He was able to differentiate a 4-inch sphere and cube from each other at 3 feet, though whether more than one trial was performed was not mentioned.
30-year-old male (Latta 1904) [12]	Bilateral congenital cataracts. Before the bilateral cataract extraction under chloroform, he was believed to be able to use his acute sense of hearing, echolocation, and familiarity with his neighborhood to roam around and complete errands. Nystagmus and esotropia were present. After bandages were removed postop, he had less pronounced strabismus and ocular restlessness, but he had little control over his ocular movements. After his procedure, he remained dazed for 10 days but then was able to visualize objects very readily. Once he saw his doctor's face, he was able to quickly recognize what he saw as a face due to hearing his doctor's voice. He was able to notice colors but had difficulty with the color green.	Cataract extraction, both eyes.	Unable to distinguish objects but had light perception	Able to visualize objects but required time or sense of touch and hearing to understand what objects he was looking at. Also could perceive colors but had difficulty doing so in artificial light. Could ultimately distinguish a ball from a toy brick.
52-year-old male (SB, Gregory 1963) [13]	Loss of vision in both eyes at ~10 months of age from keratitis. At the age of 9 years, his vision was recorded as "fingers at one metre", and at age 17 years, vision was "3/60". On examination, as a child, diffuse dense "nebulae" (corneal opacification) in each eye, some vascularization and epithelial xerosis, conjunctivitis with slight discharge, and some scarring of upper tarsus. No nystagmus. He learned to echolocate. Preoperatively as an adult, his vision was hand motions right eye and light perception left eye. While he was an extroverted and high-	Corneal Transplantation, both eyes.	Hand motions, right eye; light perception left eye. "Partially sighted" Limited to bright large surfaces according to family member.	After his bandages post op were removed, he was quickly able to view faces as blurs. After a couple days, he could distinguish between passing trucks and cars. He was able to correctly read the Ishihara color plates. After the first year, he continued to be fascinated by reflections in the window of other objects. His idea of the world arose from touch & his general way of life as a blind man remained with him until his death 2 years later.

	spirited man when he first visited his ophthalmologists' office, his demeanor shifted after his operation. His doctors formed an impression that "his sight was to him almost entirely disappointing."			
23-year-old female (Carlson 1983) [14]	Born 3 months premature, weighing 1330 g, and diagnosed at 5 months with retrolental fibroplasia (retinopathy of prematurity). Throughout her life, she was on pilocarpine treatment due to elevated intraocular pressure. Fundus exam revealed nasal avascularity of the peripheral retina plus atrophic small discs in each eye. She was able to count fingers when she was 6 years old. At age 9 years, vision was light perception in the right eye, and no light perception in the left eye. Nystagmus was present. After cataract extraction at age 23 years, she did not have visual rehabilitation immediately. At first, she only had light perception. One month after the operation, she began to see colors. Post-operative examination findings were not reported.	Cataract extraction with iridectomy OD	Light perception	Nine months postop, she still was behaviorally blind while using her cane and not having the ability to recognize objects without sense of touch. Based on the contrast acuity function, her visual acuity appeared worse than 20/400. While she could objectively see more objects, it did not aid much in her activities of daily life. After 1.5 years total of active rehabilitation, she was able to learn how to use vision in her day-to-day life such as by seeing traffic lights and recognizing objects like mailboxes.
20-year-old male, SH (Mochizuki 2005) [15]	Bilateral congenital cataracts. Preoperatively, he had hand motions vision, and could identify almost all basic colors, distinguish horizontal from vertical stripe, and discriminate between two sizes of a circular 2-D shape.	Cataract extraction both eyes.	Hand motions. Light perception, colors, slight 2-D shapes	11 months after his right eye procedure, he was able to distinguish 3-D figures and distinguish the differences in height for objects. With his left eye, he was only able to locate objects but not discern their shapes.
24-year-old male (SK, Ostrovsky 2009) [9]	Congenital cataracts were spontaneously resorbed, leaving him aphakic. He had an inability to fixate and lack of visually guided behaviors by age 4.	Eyeglasses	Visual acuity 20/900	Visual acuity 20/120. Two weeks after he wore glasses, he had near-normal ability to discriminate among colors, luminance, and motion directions. He enumerated geometric shapes on their own but had difficulty when shapes overlapped. At 18 months after treatment, he demonstrated improvement in visual skills.
17-year-old male (BG, Held 2011) [17]	Dense congenital bilateral cataracts. Two days after surgery, he was able to use tactile stimuli to identify objects tactilely (touch-to-touch tasks) and use visual stimuli to identify objects visually (vision-to-vision tasks). However, he had more difficulty completing touch-to-vision tasks. One week after the procedure, he was re-tested and showed vast	Cataracts extraction.	Light perception	VA: 20/400

improvement in touch-to-vision tasks.				
21-year-old female (dc-c, Bottari 2016) [18]	Congenital but not dense cataracts. Fundus visibility in best eye was hazy but visible. Nystagmus was present.	Cataract extraction.	Finger counting from 0.5 m away in better eye	VA: 20/250
19-year-old male (cc-n, Bottari 2018) [19]	Congenital cataracts. Nystagmus present preop.	Cataract extraction.	Able to perceive light and report the location of light in better eye	VA: 20/250
18-year-old male (CC9, Rajendran 2020) [20]	Congenital cataracts. Nystagmus present.	Cataracts removal	Able to perceive light and report the direction of light rays	VA: 20/240

4. Discussion

Among the 11 patients we identified with limited visual acuity by age 1 year and ophthalmic procedures from age 17 years or older, eight patients had detailed reports of visual rehabilitation after their procedures, and four patients had detailed reports of visual rehabilitation for one year or greater. After removing post-operative bandages, these patients generally were able to immediately experience increased vision in the form of recognition of faces and light perception. Upon trying to observe their environment in the early days and weeks post-operatively, many of these patients reported that they felt overwhelmed. Though many could visualize objects, they reported difficulty identifying what objects they were seeing and whose faces they were looking at without using other senses, such as hearing the person's voice.

In the detailed reports that longitudinally examined recoveries, patients seemed to acquire most of their visual benefit within 1 to 1.5 years post-operatively. Some patients underwent visual therapy in that time, such as Carlson's patient regularly worked with a special teacher of visual rehabilitation for 1.5 years through a series of tasks to train visually guided hand movements [14]. Obtaining visual benefit may have been delayed due to the mechanisms of neuroplasticity for adult vision. Higher levels of neuroplasticity and response to amblyopia treatment are seen in early development while lower levels of neuroplasticity are observed later in life [21]. For instance, children with amblyopia who undergo management with either atropine or patching will generally experience improved visual acuity within weeks to months [22]. In contrast, a retrospective study reported that adults with monocular amblyopia whose non-amblyopic eyes suffered from disease experienced improvement in BCVA in their amblyopic eye, with peak improvement after 12 months [23]. Thus, while adults may respond to amblyopia treatment, their responses are often delayed and not as strong as those seen in children.

During this period of rehabilitation, patients reported improved vision in terms of properly perceiving colors, depth perception, and scattered objects. For instance, Gregory (1963) followed his patient, SB, who underwent cornea transplantation at 52 years of age until his death two years later. Likewise, Carlson's patient, who underwent cataract removal with iridectomy at 23 years of age, had visual improvement within the first 1.5 years after her procedure. Carlson (1983) reported that this patient had slow visual improvement from seeing only light and colors within the first month and still did not have the ability to recognize objects without touch after nine months. After 1.5 years, she was able to incorporate her improved visual function into her day-to-day life.

Nevertheless, not every patient had a gradually progressive improvement in visual acuity post-operatively. This was the case for SK, the patient described by Ostrovsky (2009). After noting visual improvement within the first 6 months after receiving aphakic spectacles, his visual performance remained similar from 6 to 12 months after treatment. Even so, he was noted to have improvement when followed up at 18 months after treatment.

Another interesting case did not qualify for our review because the visual impairment presumably began at age 21 months of age when the patient's mother noted whiteness in both his pupils. Fine (2002) cared for a patient, PD, who began to have denser central nuclear opacification as he grew older [24]. Though he had difficulty seeing curbs and recognizing people, his doctor

managed his cataracts conservatively with atropine treatment because the lens periphery was clear. Once PD was 43 years of age, he underwent cataract removal with intraocular implantation. His baseline visual acuity of 20/80 improved to 20/40 postoperatively. In contrast to our cases of interest, PD had better baseline and postoperative visual acuity. Given that PD's visual impairment became apparent past 1 year of age, his case may reflect the significance of early visual development despite prolonged visual impairment and delayed management.

5. Conclusions

Based on these cases, the best visual acuity which reasonably can be expected after surgery for severe visual impairment throughout childhood appears to be about 20/200, following a period of visual recovery and rehabilitation. This limited visual improvement is consistent with the irreversible visual impairment from amblyopia in individuals with early and prolonged blinding conditions. Despite delays in ophthalmic procedures, patients continue to experience benefits in their best vision. These findings underscore the importance of intervention for individuals with visual impairment, even if treatment must be delayed.

Author Contributions: Conceptualization, C.L.; methodology, C.L, S.P.; writing—original draft preparation, S.P., C.L., S.G.S.; writing—review and editing, S.P., C.L., S.G.S.; supervision, C.L. All authors have read and agreed to the published version of the manuscript.

Funding: This research received no external funding.

Institutional Review Board Statement: Not applicable.

Informed Consent Statement: Not applicable.

Conflicts of Interest: The authors declare no conflicts of interest.

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