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ORIGINAL ARTICLE

Characterization of congenital cataract and postoperative visual outcome in a Pediatric Ophthalmology Unit in a middle-income country

Caracterización de catarata congénita y resultado visual postoperatorio en una Unidad de Oftalmología Pediátrica en un país de ingresos medios

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What do we know about the subject matter of this study?

Understanding of the critical period of visual development has led to considering congenital cataract surgery before 3 months of life (between 6-12 weeks) as necessary for the proper development of binocular vision, thus avoiding a profound and irreversible visual loss and reducing the risk of aphakic glaucoma.

What does this study contribute to what is already known?

In a middle-income country, visual impairment or worse is observed following congenital cataract surgery because its diagnosis and surgical treatment were late and due to high dropout from post-surgery follow-up.

Abstract

The functional visual outcome after cataract surgery is influenced by several factors such as the age at diagnosis and surgical treatment, as well as visual rehabilitation. **Objective:** to characterize congenital cataract patients and assess the postoperative visual outcome in an ophthalmological care center in Guatemala. **Patients and Method:** A cross-sectional retrospective study. Clinical records of all congenital cataract patients who underwent congenital cataract surgery from 2014 and 2015 were reviewed. The following was recorded: postoperative visual acuity, age at diagnosis and surgery, sex, unilateral or bilateral involvement, etiology and morphology of the cataract, preoperative poor visual prognosis factors, and postoperative follow-up. **Results:** Fifty-nine patients underwent pediatric cataract surgery in this period, ages ranging from 1 month to 13 years (median 48 months), and 23 were congenital (38.98%). Fourteen were male, 16 bilateral, and the mean age at diagnosis and surgery was

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7 and 12 months, respectively. Only 13 patients completed 3 years of follow-up visits. Visual acuity was recorded at 3 years after surgery, observing in 11 patients and 8 patients visual impairment or worse, respectively. Delayed consultations occurred in 82.6% of the patients (older than 3 months of age). **Conclusion:** In this series, the diagnosis of congenital cataract and its surgical treatment were delayed, observing a high rate of patients unable to complete follow-up visits. The visual outcome at 3 years after congenital cataract surgery was mostly visual impairment or worse.

Introduction

A congenital cataract is defined as any lens opacity present at birth or in early childhood. Although uncommon, with a prevalence of 4.24 per 10,000 inhabitants[1], it has a significant impact on affected patients due to visual deprivation causing subsequent amblyopia. The etiology of congenital cataracts is variable, with idiopathic being the most frequent (63%), followed by genetic (34%) and infectious (3%)[2]. Worldwide, an estimated 1.4 million children are blind, of which 4% are due to unoperated bilateral cataracts, amblyopia due to late surgery, surgery complications, or other associated anomalies[3]. Visual loss secondary to cataracts represents a huge problem in developing countries regarding morbidity, economic loss, and social burden[3]. Moreover, it continues to be a major cause of preventable blindness in children[4-5].

The management of congenital cataracts is long, complex, and intensive[6-8]. The functional outcome of pediatric cataract surgery is influenced by several factors such as age at diagnosis, age at surgical treatment, and visual rehabilitation[1]. Previous studies of congenital cataracts in other countries have reported an average post-surgery visual acuity (VA) in bilateral cases of 20/50 to < 20/400 and unilateral cases of 20/200 to < 20/400[6-8,9-10]. Table 1 summarizes the findings of these studies.

The objective of this study was to describe the demographic and clinical characteristics of patients with congenital cataracts and to know the post-surgery visual outcome in an ophthalmologic care center in Guatemala.

Patients and Method

A cross-sectional and retrospective study was performed on all cases of congenital cataract seen between 2014 and 2015, at the Pediatric Ophthalmology, Strabismus, and Neuro-Ophthalmology Unit "*Dra. Ana María Illescas Putzeys*", the *Instituto de Ciencias de la Visión* from the *Hospital de Ojos y Oídos "Dr. Rodolfo Robles V."*, and the *Benemérito Comité Pro Ciegos y Sordos de Guatemala*. The study was approved by the Ethics Committee of the *Instituto de Ciencias de la Visión* Of. 45/2020.

The clinical records of patients under 14 years of age with a diagnosis of pediatric cataract operated on in 2014 and 2015 were reviewed. All cases with a diagnosis of cataract of congenital etiology were included for data analysis. The following demographic, clinical, and etiological variables were investigated: age at diagnosis, sex, uni- or bilateral involvement, cataract etiology, type of cataract according to its morphology (nuclear, total, anterior polar, other), preoperative poor visual prognostic factors (nystagmus, strabismus, age at diagnosis, unilateral, age at surgery), post-surgery complications (glaucoma, endophthalmitis, others), IOL implantation (primary, secondary, aphakia), age at IOL implantation, and type of rehabilitation (glasses, amblyopia patching, visual stimulation).

To determine the visual outcome, the best corrected VA was recorded in the better eye in bilateral cases and in the operated eye in unilateral cases, using the Snellen chart at 1-, 3-, and 5 years post-surgery, and classified according to the World Health Organization Prevention of Blindness and Deafness (WHO/PBD) at 3 years post-surgery[11]. For statistical analysis, the data were entered in a spreadsheet in Microsoft® Excel 2017 version 15.37 (170815).

Each patient underwent a complete preoperative ophthalmologic evaluation. Patients who underwent IOL implantation (single-piece non-foldable polymethylmethacrylate in sulcus) underwent biometry at 2 years of age using the IOLMaster® 700 SWEPT Source OCT biometer (Carl Zeiss Meditec) or the AccuSonic a-scan plus® (Accutome) based on the Sanders-Retzlaff-Kraff (SRK/T) formula[12]. The surgical technique used was the one described in the ORBIS Telemedicine, Cyber-Sight manual Cataracts in Childhood[13]. The second eye was operated on one month apart in bilateral cases. Patients were evaluated periodically according to our unit's protocol, which included next-day, 3-day, and 1-week consultations.

All patients were treated with topical fluoroquinolones, topical prednisolone acetate 1%, and oral pred-

Table 1. Postoperative visual acuity (VA) outcome in previously reported congenital cataract extraction

Author	Country	No. of patients	Laterality	Postoperative best corrected VA	Age at surgery
Rong X[6]	China	66	Bilateral	20 / 50	7.45 ± 4.73 months
Khanna RC, et al.[9]	India	107	Bilateral / unilateral	< 20 / 400 ambos	55.2 months
Chack M, et al.[10]	England	153	Bilateral / unilateral	20 / 60 / 20 / 200	3.78 months
Lundvall A[8]	Sweden	22	Bilateral / unilateral	20 / 50 / 20 / 200	0 a 11 months

No.: Number .

nisolone (1mg/kg/day). Visual acuity measurement was performed with the Snellen chart (according to patient collaboration), evaluation of ocular alignment and motility, refraction, the anterior segment with a slit lamp, and posterior segment with indirect ophthalmoscope. In addition, all patients received spectacle rehabilitation, patching for amblyopia in unilateral cases, and visual stimulation, reevaluated every 6 months or as needed.

Results

Over two years (2014-2015), cataract surgery was performed in 59 pediatric patients, 23 (38.98%) of them with congenital etiology, who were included in this study. Table 2 summarizes the characteristics of each of them. 14 patients were males and 16 cases had bilateral cataracts.

Regarding etiology, 13 were of idiopathic cause, 8 were infectious, 2 with clinical features of Hallermann Streiff syndrome, and 1 with an unclassified dysmorphic syndrome. Regarding the cataract morphology, the nuclear type was the most frequent (9 patients), followed by complete (6 patients), polar anterior and subcapsular posterior (2 patients each), one mixed type, and 3 unspecified. Most had preoperative poor visual prognostic factors including late surgery (older than 3 months of age) in 21 patients, strabismus in 12 patients, nystagmus in 14 patients, unilaterality in 7 patients, and microcornea in 2 patients.

The median age at diagnosis was 7 months (range 2-120 months) and the median age at surgery was 12 months (range 2 months-120 months). In 5 patients, the IOL was implanted at the same surgical time, 4 in a second surgery, and 14 remained in aphakia due to a lack of collaboration to perform biometry and lack of the necessary equipment to perform it under general anesthesia, microcornea, and parents in disagreement with a second implantation. The median age of IOL implantation in a second intervention was 5 years (range 2 - 10 years), and it was implanted 2.5 years (range 0-4 years) after the first surgery. Only two patients presented glaucoma as the only complication.

13 patients were rehabilitated exclusively with glasses, 7 required patching for amblyopia (because of unilaterality), and 100% were referred to visual stimulation therapies. 10 patients dropped out of follow-up before 3 years post-surgery, with an average follow-up duration of 17.28 months. 13 patients completed the evaluations at 3 years post-surgery and 9 at 5 years post-surgery. In 11 patients their quantitative visual acuity was evaluated at 3 years post-surgery, and more than half of them (8 patients) obtained a classification of visual impairment or worse. Table 3 shows the characterization of visual outcome according to WHO/PBD classification in patients with 3-year post-surgery follow-up.

Discussion

Congenital cataracts can affect visual development due to visual deprivation and interocular competition in unilateral cases and compromise visual development in bilateral cases[14] due to the absence of foveal stimulation by inadequately focused images (amblyopia ex anopsia)[15].

The understanding of the critical period of visual development has led to consider that a surgical intervention before 3 months of age (between 6-12 weeks) is necessary for the proper development of binocular vision, thus avoiding a profound and irreversible visual loss[7,16-18] and reducing the risk of glaucoma in aphakia[19]. In this series, 21 patients underwent surgery after 3 months of age due to late consultation, probably because of low socioeconomic status, low schooling, lack of access to health services, and some cultural aspects as described by Schwering in a demographic, sociocultural, and socioeconomic characterization of 53 patients in Malawi[20]. In this study reports low awareness of cataract diagnosis, its causes, and treatment, as well as remoteness from health centers, consultation with traditional healers, fear of surgery, and supernatural beliefs[20].

Table 2. Characteristics of patients operated on for congenital cataract

Patient No.	Age at diagnosis / age at surgery	Sex	Laterality	Cataract etiology	Cataract morphology	Poor prognostic factors prior to surgery	IOL implant	Other factos that influenced the visual outcome	Best corrected VA in the best eye at one year	Best corrected VA in the best eye at 3 years	Best corrected VA in the best eye at 5 years
1	2 months / 3 months	M	Bilateral	Infectious	Nuclear	Nystagmus	Aphakia	Parents in disagreement with secondary implantation and delay in performing preoperative tests	C	C and M	20/100
2	2 months / 2 months	M	Bilateral	Infectious	Anterior polar	None	Aphakia	PMR (non-cooperation to perform biometrics) and aphakia glaucoma	C and S	C and S	No C, no S no M
3	3 months / 4 months	M	Bilateral	Idiopathic	Nuclear	Nystagmus	Aphakia	Microcornea	C	20/400	Dropout
4	4 months / 6 months	M	Bilateral	Idiopathic	Nuclear	Nystagmus, strabismus and late consultation	Secondary	Parental delay in performing preoperative tests	C	C and M	20/100
5	5 months / 8 months	M	Bilateral	Idiopathic	Total	Nystagmus and late consultation	Aphakia	PMR (non-cooperation to perform biometrics) and parental delay in performing preoperative tests	C	Dropout	Dropout
6	6 months / 8 months	M	Bilateral	Genetic	Total	Nystagmus and late consultation	Aphakia	PMR, Hallermann Streiff SD, parental delay in performing preoperative tests and abandonment before 2 years of age	C	Dropout	Dropout
7	7 months / 11 months	M	Bilateral	Idiopathic	Total	Nystagmus, strabismus and late consultation	Aphakia	PMR, non-cooperation to perform biometrics, Hallermann Streiff SD and parental delay in performing preoperative tests	C	C	Not quanti-fiable
8	24 months / 24 months	M	Bilateral	Idiopathic	Anterior polar	Late consul-tation	Aphakia	Non-cooperation to perform biometrics	C, S and M	C, S and M	Dropout
9	24 months / 36 months	M	Bilateral	Infectious	Total	Nystagmus, strabismus and late consultation	Primary	Parental delay in performing preoperative tests	C, S and M	Dropout	Dropout
10	60 months / 60 months	M	Bilateral	Infectious	Total	Nystagmus and late consultation	Primary	None	Dropout	Dropout	Dropout
11	72 months / 72 months	M	Bilateral	Idiopathic	Total	Late consul-tation	Primary	None	20/40	Dropout	Dropout

12	2 months / 5 months	F	Bilateral	Infectious	Nuclear	Nystagmus and strabismus	Aphakia	Parental delay in performing preoperative tests and non-cooperation to perform biometrics	C and M	C and M	20/200
13	4 months / 6 months	F	Bilateral	Idiopathic	Nuclear	Nystagmus, strabismus and late consultation	Aphakia	Parental delay in performing preoperative tests and non-cooperation to perform biometrics	C	Not quanti-fiable	Dropout
14	6 months / 24 months	F	Bilateral	Idiopathic	Without specification	Late consultation	Secondary	Aphakia glaucoma OD, parental delay in performing preoperative tests	C, S and M	20/400	20/400
15	6 months / 12 months	F	Bilateral	Genetic	Nuclear	Nystagmus, strabismus and late consultation	Aphakia	Microcornea, microcephaly, brain atrophy, not cataloged dysmorphic SD and parental delay in performing preoperative tests	C and M	20/200	20/200
16	12 months / 72 months	F	Bilateral	Idiopathic	Nuclear	Strabismus and late consultation	Primary	Parental delay in performing preoperative tests	20/60	20/40	Dropout
17	7 months / 9 months	M	Unilateral	Infectious	Nuclear	Strabismus, unilateral and late consultation	Aphakia	Abandonment before 2 years, parental delay in performing preoperative tests and failed amblyopia treatment	C	Dropout	Dropout
18	18 months / 18 months	M	Unilateral	Idiopathic	Posterior subcapsular	Strabismus, unilateral and late consultation	Aphakia	Failed amblyopia treatment	C and S	Dropout	Dropout
19	60 months / 60 months	M	Unilateral	Idiopathic	Without specification	Nystagmus, strabismus, unilateral and late consultation	Secondary	Failed amblyopia treatment	Dropout	Dropout	Dropout
20	9 months / 14 months	F	Unilateral	Infectious	Posterior subcapsular	Nystagmus, unilateral and late consultation	Aphakia	Abandonment before 2 years, parental delay in performing preoperative tests and failed amblyopia treatment	Dropout	Dropout	Dropout
21	12 months / 12 months	F	Unilateral	Idiopathic	Nuclear	Strabismus, unilateral and late c onsultation	Aphakia	Failed amblyopia treatment	C and S	10/200	1/200
22	12 meses / 12 meses	F	Unilateral	Infectious	Without specification	Nystagmus, strabismus, unilateral and late consultation	Secondary	Failed amblyopia treatment	No C, no S no M	Dropout	Dropout
23	120 months / 120 months	F	Unilateral	Idiopathic	Mixed	Unilateral and late consultation	Primary	Failed amblyopia treatment	20/150	20/150	20/150

VA: visual acuity, F: female, M: male, IOL: intraocular lens, C: central, S: steady, M: mainteined, SD: syndrome, PMR: psychomotor retardation, OD: right eye, OS: left eye.

In our study, the median age of diagnosis was 7 months, which could explain the poor visual outcome in this series. The median age at surgery was 12 months, which is higher than that found in other studies (table 1)[6,8,9-10]. Because patients first present for pediatric evaluation, the American Academy of Pediatrics (AAP), the American Academy of Ophthalmology (AAO), and the American Academy of Pediatric Ophthalmology and Strabismus (AAPOS) have established that, at least, an eye examination with the Brückner test (red reflex) should be performed at birth to rule out leukocoria and immediately refer the patient to a pediatric ophthalmologist for evaluation[21-22]. A congenital cataract carries a 20-fold increased risk of poor visual outcomes compared with a developmental cataract[9].

In this study, congenital cataract of infectious etiology was secondary to congenital infections as in other studies[23]. 13 (56.52%) of the patients with congenital cataract were of idiopathic etiology, similar to that reported in the literature (60%)[24]. The literature reports that 6-7% of congenital cataracts are related to syndromes[23]. In this study, 2 patients were associated with Hallermann Streiff syndrome and unclassified dysmorphogenetic syndrome.

Preoperative strabismus and nystagmus are poor visual prognostic factors described in children with congenital cataracts, with reported frequencies of 27-100% and 71%, respectively[6]. These data are similar to what was observed in this study where strabismus was found in 52.17% of the cases and nystagmus in 61%.

14 patients remained in aphakia at 3 years post-surgery due to non-cooperation for biometry (33.33%), microcornea, abandonment of follow-up before 2 years of age, and parental decision. However, aphakia is not reported to be a poor prognostic factor for functional visual outcome[25].

In patients with 3-year follow-up, the only complication observed in our series was glaucoma (2 patients), an incidence lower than previously reported (17-58.7%)[17, 24, 26]. Since posterior capsulotomy and anterior vitrectomy were performed as part of the surgical technique, posterior capsule opacification was not observed as a complication, unlike other studies that report an incidence of 30.9%[26].

Visual rehabilitation after congenital cataract surgery is long, complex, and intensive[6-8]. Long-term post-surgery follow-up is important to obtain a better visual outcome, especially in congenital cataract[9] as this allows rehabilitation with periodic refractive error correction, treatment of amblyopia, and visual stimulation therapies[7-8]. In this series, the percentage of abandonment from follow-up before 3 years was high (43.47%). However, the mean duration of follow-up was 17.28 months, which is longer than other studies that reported a mean follow-up of 13.1 months[9]. In our study, the longer the post-surgery period, the higher the dropout rate, which could be explained by the socioeconomic condition, the distance from their residence, the language barrier, and the lack of schooling of the parents and/or caregivers.

Table 3. Characterization according to the WHO/PBL classification to determine the visual outcome in patients with a 3-year postoperative follow-up[11]

WHO classification	VA	Bilateral	Unilateral
Blindness	NPL to < 20/400	0	1
Severe visual impairment	< 20/200 to 20/400	2	0
Visual impairment	< 20/60 to 20/200	4	1
No impairment	20/60 or better	1	0
No quantificable	No central, no steady and/or no mainteined	3	0
	Central, steady and mainteined	0	1
Total		10	3

WHO/PBL: World Health Organization Prevention of Blindness, VA: visual acuity, NPL: no perceivable light.

13 patients completed a 3-year post-surgery follow-up, 10 were bilateral cataracts and 3 were unilateral. Of these, VA was measured in 11 patients (9 bilateral and 2 unilateral). 6 of the bilateral and 2 unilateral cases were categorized as visually impaired to severely visually impaired. This result is worse than reported in the literature in bilateral cases (20/50, no visual impairment according to WHO/PBD classification)[6-7,10] and similar in unilateral cases (20/200 and < 20/400, severe visual impairment or worse according to WHO/PBD classification)[6-7,9-10].

An important limitation of this study is that it was not prospective, and follow-up was not standardized. In addition, it is a small series from a referral hospital, so the conclusions cannot be applied to other populations. It would be interesting to perform a study that includes the population of other pediatric ophthalmology care centers in the region, with standardized prospective follow-up.

Conclusions

In our series, diagnosis and surgery of congenital cataracts were late, probably due to delay in consultation. Most patients 3 years after congenital cataract extraction have a visual outcome of visual impairment

or worse. Post-surgery rehabilitation dropout is high and is directly proportional to the time elapsed.

Recommendation s

Post-surgery visual outcome and rehabilitation follow-up show concerning data in this series. Early consultation and better adherence to post-surgery treatment are essential to achieve a better visual outcome. There are multiple reasons why this does not happen, and many of them are challenging to solve. Nevertheless, we can suggest the following recommendations:

1. Raise awareness among pediatricians and physicians about the early referral of patients for ophthalmologic evaluations according to the recommendations given by the AAP, AAO, and AAPOS[21,25].
2. Create and promote informative campaigns for early ophthalmologic consultation in the population.
3. Strengthen the follow-up of post-surgery treatment through blindness prevention programs by contacting patients through telephone calls to their parents.

Ethical Responsibilities

Human Beings and animals protection: Disclosure the authors state that the procedures were followed according to the Declaration of Helsinki and the World Medical Association regarding human experimentation developed for the medical community.

Data confidentiality: The authors state that they have followed the protocols of their Center and Local regulations on the publication of patient data.

Rights to privacy and informed consent: The authors have obtained the informed consent of the patients and/or subjects referred to in the article. This document is in the possession of the correspondence author.

Conflicts of Interest

Authors declare no conflict of interest regarding the present study.

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