Refractive Errors and Strabismus In Patients with Orofacial Clefts

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REFRACTIVE ERRORS AND STRABISMUS IN PATIENTS WITH OROFACIAL CLEFTS

ABSTRACT

Purpose: To evaluate the prevalence of refractive errors and strabismus in patients with orofacial clefts.

Methods: This retrospective study analyzed the medical records of 54 patients with orofacial clefts between August 2018 and March 2020. A complete eye examination was performed, including visual acuity assessment on a logMAR scale, anterior biomicroscopy, cycloplegic refraction, eye motility examination, and indirect ophthalmoscopy.

Results: The mean age of the patients at presentation was 9.47 years. Twenty-three (42.59%) patients had isolated cleft palate (CP), 10 (18.52%) had cleft lip (CL), and 21 (38.89 %) had cleft lip and palate (CLP). The mean spherical equivalent was 1.30D (± 1.56) in CL, 0.32D (± 2.24) in CLP, and 0.62D (± 3.76) in CP. The prevalence of refractive error, either spherical or cylinder >0.5 was 88%. The most common refractive error was hyperopia (60%), followed by astigmatism (54%) and myopia (16%). Overall, 52.63% of the patients were prescribed glasses. No statistically significant difference was observed between the groups with respect to the need for prescription of glasses ($p=0.6753$). There were 15 patients with some type of strabismus, and other ophthalmological changes were observed in 13 patients.

Conclusion: In this population with orofacial clefts, the prevalence of refractive errors and strabismus was 88% and 22%, respectively.

Keywords: refractive errors; strabismus; astigmatism; orofacial clefts

INTRODUCTION

Non-syndromic orofacial clefts, which include cleft lip (CL), cleft palate (CP), and/or cleft lip and palate (CLP), comprise a series of disorders that affect the lips and oral cavity. The cause of these orofacial clefts remains largely unknown [1]. They represent the most common congenital deformities of the orofacial region worldwide [2].

The embryological formation of the tissues that comprise the craniofacial system is partly derived from the neural crest, which can consequently affect the development of the brain, ocular, and facial structures that develop from this neural crest [2]. Therefore, effects on speech, hearing, vision, appearance, and cognition can lead to long-lasting adverse effects on health and social integration.

Epidemiological data suggest that environmental risk factors may be important in affected individuals, such as maternal exposure to tobacco smoke and alcohol, folic acid deficiency, viral infection, medications, and teratogens at the beginning of pregnancy [1].

Children with these disorders usually require multidisciplinary care from birth to adulthood and they have a greater morbidity and mortality throughout their lives than unaffected individuals [1]. Medical assistance to these patients generally involves plastic surgery, maxillofacial surgery, otolaryngology, and genetics, among others [3]. Ophthalmological evaluation plays an important role in this context, as visual changes are recognized in this group of patients.

The most common pathology found in regular children is refractive errors, followed by strabismus.

Currently, there are only a few published studies related to this topic [3, 7], it is difficult to know if there is a difference in the frequency with which eye diseases appear in the population with orofacial cleft compared to the population without cleft.

Therefore, this study aims to contribute to the early identification of ocularities associated with this condition, providing immediate intervention, and next, good visual function.

MATERIAL AND METHODS

We retrospectively reviewed the medical records of 54 patients who had orofacial fissures between August 2018 and March 2020. All patients belonged to the Assistance Center for CLP, Curitiba, Brazil. Patient information, such as age, sex, type of orofacial cleft, ophthalmic examination, including best corrected visual acuity, cycloplegic refraction, spherical equivalent, intraocular pressure, anterior segment examination with slit lamp or handheld slit lamp, indirect fundoscopy, and type of strabismus was evaluated. Myopia was considered as a spherical equivalent (SE) $\leq -0.5D$, hyperopia as SE $\geq 0.5D$, and astigmatism as ≥ 0.5 DC. Patients with deformities or other syndromes that did not include orofacial clefts were excluded. The Ethics Committee of the Hospital do Trabalhador, Paraná, Brazil, approved the protocol of this study. The requirement of written informed consent was excepted owing to the absence of ethical infractions, and the study was conducted in accordance with the tenets of the Declaration of Helsinki.

Descriptive analysis and the Kruskal-Wallis test were used for data analysis. A p-value of <0.05 was considered statistically significant.

RESULTS

In the study population, 24 patients (44.4%) were women and 30 (55.5%) were men. Twenty-three (42.5%) patients had isolated CP, 10 (18.5%) had CL, and 21 (38.8%) had CLP. The mean age of the patients at presentation was 9.47 years.

There was a high correlation between spherical equivalent and cylinder of the right eye (OD) and left eye (OS; Spearman's $r=0.85$, and 0.84 , respectively); therefore, we used the OD for refractive error (RE) analysis. Furthermore, only patients with data of both the eyes were considered ($n=50$). The prevalence of RE, either spherical or cylinder >0.5 was 88%. The most common RE was

hyperopia (60%, n=30), followed by astigmatism (54%, n=27) and myopia (16%).

In case of mean visual acuity, a high correlation was observed between the OD and OS when considering patients who had measurements of both the eyes (Spearman's $r = 0.81$, $n=47$).

In table I is shown the mean of age, visual acuity, and spherical equivalent and cylinder of the right and left eye, and in Figure I the relationship of this information, with each group of orofacial clefts.

Table I: Mean age, visual acuity, and spherical equivalent and cylinder of the right and left eyes.

	N	Mean	SD
Age	54	9.87	7.61
BCVA OD (LogMAR)	44	0.22	0.39
BCVA OS (LogMAR)	48	0.30	0.52
Spherical equivalent OD	50	0.64	2.83
Spherical equivalent OS	53	0.62	3.59
Cylinder OD	50	0.85	1.13
Cylinder OS	54	1.06	1.35

BCVA, best corrected visual acuity; OD, right eye; OS, left eye; SD, standard deviation.

Figure I: Mean visual acuity and spherical equivalent and cylinder of the right and left eyes, according to each group of orofacial clefts.

CP, cleft palate; CL, cleft lip; CLP, cleft lip and palate; OD, right eye; OS, left eye; VA, visual acuity

A mean of 52.6% of the patients were prescribed glasses, of which six were in the CL group, 13 in the CLP group, and 11 in the CP group. No statistically significant difference was observed between the groups with respect to the need for prescription of glasses (p=0.6753).

Table II shows the cases associated with strabismus, and in Table III others ophthalmic findings.

Table II: Cases of strabismus associated

Exotropia	8	
Esotropia	Isolated	1
	DVD	2
	Hyperopia	1

Nystagmus	1

DVD	2

DVD, dissociated vertical deviation

Table III: Other ophthalmic findings

Microcornea	2

Trichiasis	2

Anophthalmia	3

Lagophthalmos	2

Microphthalmia	1

Lacrimal punctura agenesis	1

Nasolacrimal duct obstruction	1

Ptosis associated with ectropion	1

DISCUSSION

Only a few studies describing the ophthalmic findings in patients, especially in children, with orofacial clefts are available in the literature [3, 7].

Hashemi et al [8] reported that astigmatism was the most common RE in children and adults worldwide, followed by hyperopia and myopia. In 2015, Ferraz et al. [9] published a similar study in which astigmatism was the most prevalent RE in the Brazilian population. In this study, the most frequently found RE was hyperopia (60%), followed by astigmatism (54%) and myopia (16%). With-the-rule axis of astigmatism was frequently observed in young participants and against-the-rule axis was frequently observed in older individuals.

In a study by Cronemberger et al. [10] the prevalence of RE in a specific population with a Möbius sequence was evaluated. They reported a 42.4% prevalence of RE, from the spherical equivalent, with 40% of the total participants having astigmatism greater than or equal to 0.75D, corroborating

with the statistics of the general population. In this study, astigmatism was considered as ≥ 0.5 DC, which justified the higher incidence.

In the study by Anchlia et al. [3] of 27 patients with CL/CLP and ocular abnormalities, eight (17%) had strabismus and seven (14%) had RE. In this study, a higher prevalence of RE (88%) and strabismus (22.2%) was observed, and we found that the most common RE was astigmatism (64.8%) and among strabismus types, exotropia (57.1%) was most commonly observed. The results of RE are consistent with the results found worldwide, nationally, and in groups with similar characteristics, such as the Möbius sequence. Comparison of the strabismus data was not possible, owing to the scarcity of studies addressing this topic.

Previously published case reports describe ocular abnormalities in patients with orofacial clefts, resulting from the abnormal fusion of the embryonic optic fissures, such as ocular coloboma, ectopia lentis, ectopia pupillae, and bilateral optic disc pit with maculopathy [4, 6]. These changes were observed in patients with CL/CLP without systemic or neurological involvement, excluding syndromic associations. The chief complaint of these patients was low visual acuity reported or observed in an ophthalmic examination, reinforcing the importance of ophthalmic follow-up, early diagnosis, and immediate intervention for a good visual prognosis [4, 6].

Finally, despite being scarce, the cited studies corroborate the importance of ophthalmological follow-up for patients with non-syndromic CLP, because of the high prevalence of REs, strabismus, and structural/anatomical changes [3, 7].

CONCLUSION

The study concluded that the prevalence of REs and strabismus was 52.6% and 22.2%, respectively, in patients with CL and CLP. Early referral and regular ophthalmological monitoring of this group is required to achieve good visual development.

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CONFLICT OF INTEREST

All authors certify that they have no affiliations with or involvement in any organization or entity with any financial interest or non-financial interest in the subject matter or materials discussed in this manuscript.

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ETHICS APPROVAL

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