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Prevalence and characteristics of pediatric hypertelorism: insights from Ugwolawo, Kogi State, Nigeria

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Abstract

Background: Hypertelorism is a medical condition characterized by an abnormally increased distance between two organs or structures, particularly the eyes.

Objectives: The study aims to investigate the prevalence and characteristics of hypertelorism among 8–10-year-old children in Ugwolawo community of Kogi State, Nigeria.

Methods: Three hundred children (150 males and 150 females) were included in the study. Data on inner canthal distance (ICD), interpupillary distance (IPD), and outer canthal distance (OCD) were collected through physical examinations and questionnaires. Standardized photographs were taken to aid in further analysis and confirmation of hypertelorism. The data were analyzed using GraphPad Prism version 8, employing t-tests for comparisons involving variables such as gender, and Spearman's correlation coefficient to assess the association between age and the facial indices. The prevalence of hypertelorism in the study population was assessed, and potential risk factors were explored.

Results: The mean ages for male and female participants were 9.0±0.8 and 9.49 ± 0.5 years, respectively, while the overall mean age for all subjects was 9.25 ± 0.7 years. IPD, ICD, and OCD were assessed, and no statistically significant differences were found between males and females in terms of these indices. Additionally, Spearman's correlation coefficient indicated a significant correlation between age and OCD, but no significant correlations were observed for IPD and ICD. The medical history analysis showed that none of the participants had been diagnosed with hypertelorism, but 4% reported a family history of hypertelorism or craniofacial abnormalities.

Conclusion: The absence of diagnosed cases of hypertelorism in the studied population suggests a relatively low prevalence of the condition. However, the presence of family history in 4% of the participants highlights the potential role of genetic factors in hypertelorism. Larger-scale studies are recommended for deeper exploration of these findings.

Keywords: Hypertelorism, Prevalence, Child, Population Characteristics, Nigeria.

Introduction

Hypertelorism is a medical condition characterized by an abnormally increased distance between two organs or structures, particularly the eyes. This condition can be present at birth or can develop later in life due to various factors. Congenital hypertelorism is often associated with certain genetic disorders or syndromes, such as craniofacial disorders like Apert syndrome, Crouzon syndrome, or Pfeiffer syndrome.[1,2] Hypertelorism is a physical manifestation observed in numerous craniofacial anomalies, but it does not constitute a syndrome on its own.[3] Hypertelorism is a rare condition, with a low estimated incidence rate. According to some sources, the incidence is estimated to be approximately in 20,000 births.[3]

The degree of hypertelorism can vary widely, ranging from mild to severe. Hypertelorism is typically measured using the intercanthal distance, which refers to the distance between the inner corners (medial canthi) or outer corners (lateral canthi) of the eyes.[4] The

measurement of Inter-pupillary distance (IPD) is regarded as the most precise and dependable method for quantifying ocular hypertelorism because it remains unaffected by the soft tissues surrounding the eye, making it highly accurate.[5,6] The normal range for intercanthal distance can vary based on ethnicity and age. For example, the average intercanthal distance for Caucasians is approximately 28-34 mm, while for African Americans, it is around 30-36 mm. These measurements may serve as a reference point when assessing hypertelorism.[7] True orbital hypertelorism is identified when measurements exceed the 95th percentile on standard anthropometric reference values. Orbital hypertelorism represents lateralization of the orbits, meaning increased interorbital and outer orbital distances.[8] Hypertelorism can affect the appearance and function of the eyes and may be associated with other facial abnormalities.

Hypertelorism is believed to result from disruptions in the embryological development of the face, specifically occurring between weeks 4-8 of gestation. The frontonasal prominence, which gives rise to the forehead and nose, plays a crucial role during this period. In typical development, the frontonasal prominence undergoes lateral movement of the eye sockets (orbits) followed by inward migration. However, if there is a disruption in the normal progression of frontonasal prominence development, the space intended for the orbits may be occupied by primitive brain tissue. This hinders the normal inward migration of the orbits, resulting in their positioning in a more lateral or wide-set configuration.[9] Another etiology of hypertelorism is the premature ossification of the lesser wings of the sphenoid bone, which hinders the inward movement of the eye sockets, causing them to remain in a foetal position. Additionally, certain craniosynostosis syndromes, characterized by the premature fusion of cranial sutures, can disrupt the normal migration and development of the orbits, contributing to hypertelorism.[10]

Individuals with hypertelorism may have several ocular and non-ocular manifestations. Ocular complications can include problems with binocular vision, strabismus (misalignment of the eyes), reduced visual acuity, and exposure of the eyes due to a widened nasal bridge.[11,12] Non-ocular manifestations can involve the skull, face, and other body systems, depending on the underlying cause. Hypertelorism was identified solely in patients with neurofibromatosis.[13]

Objectives

This research aims to investigate the occurrence and features of hypertelorism among 8–10-year-old children in the Ugwolawo community of Kogi State, Nigeria. Understanding its prevalence and characteristics in this specific age group is essential for early detection and intervention. By focusing on children in this age range, the study seeks to capture important developmental milestones and gain valuable insights into the condition during a critical period of growth. Furthermore, conducting the research in the Ugwolawo community of Kogi State will contribute vital data to the limited existing literature on hypertelorism in this region.

Methods

Study Design

The research utilized a cross-sectional study design, allowing for the collection of data from the target population within a specified period. This design enabled the assessment of hypertelorism prevalence and associated risk factors among the selected age group. Before commencement of the study, informed consent was obtained from all participants legal guardians via consent form. The form clearly explained the purpose of the study, the procedures involved, and the voluntary nature of participation. Legal guardians of the participants had the opportunity to ask questions and make an informed decision about their child`s involvement in the study.

Study Population

The study population consisted of 8–10-year-old children residing in Ugwolawo, Kogi State, Nigeria. Inclusion criteria encompassed children within the specified age range and residing in the study area during the data collection period. Exclusion criteria included children with a known diagnosis of syndromic hypertelorism or those unwilling to participate.

Sample Size

Three hundred children (150 males and 150 females)

with age range of 8-10 years old were selected to gather data on inner canthal distance (ICD), interpupillary distance (IPD), and outer canthal distance (OCD). The subject's data were obtained at hospitals, homes, and schools in Ugwolawo Community. Sample size was determined based on various factors, including the estimated prevalence rate of hypertelorism in similar populations, anticipated effect size, and desired level of precision.

Data Collection

The study employed several data collection methods to gather comprehensive information:

a. Physical Examination

Each participant underwent a thorough physical examination, with specific focus on facial features. Measurements of ICD, IPD, and OCD were taken to assess for hypertelorism accurately. This method allowed for the objective assessment of physical characteristics. Every participant was positioned in a relaxed manner on a chair, where their head was aligned with the examiner's head and positioned 40 cm in front of it. The subject's face received ample lighting. To conduct the measurement, the examiner closed one eye, instructed the patient to concentrate on a distant target, and positioned the zero end of a plastic ruler on the patient's forehead at the outer edge of their left eye. The examiner then measured the distance from there to the inner edge of the patient's right eye, which represents the IPD.

In order to measure the ICD, a plastic ruler was placed on the bridge of the nose, with the zero-mark aligned with the inner corner of the right eye. While the examiner closed the right eye, the distance between the two inner corners of the eyes was measured. The rationale for closing the right eye by the examiner was to help in more precise readings. To measure OCD, the plastic ruler was placed at the outer corner of one eye, and the distance between the two corners of the eyes was measured while the examiner closed one eye. All the measurements were taken twice by a single examiner in order to ensure accuracy and eliminate any potential inconsistencies [Figure 1].

b. Questionnaire

A structured questionnaire was administered to collect demographic information, medical history, family history of hypertelorism, and potential risk factors. This approach provided insights into various aspects that could be associated with the prevalence of hypertelorism.

c. Photographic Documentation

Standardized photographs were taken of each participant's face to facilitate further analysis and confirmation of hypertelorism. These photographs allowed for a more detailed examination.

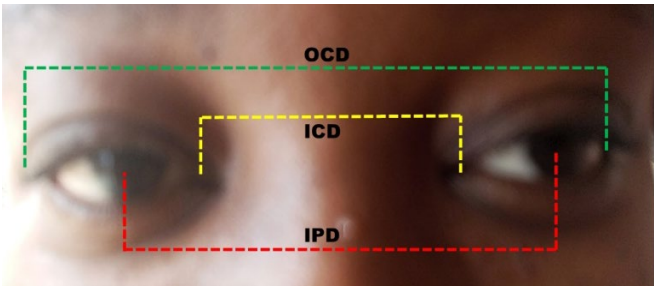

Figure 1. Anthropometric measurements of human eye showing inner canthal distance (ICD), interpupillary distance (IPD), outer canthal distance (OCD)

Inclusion and Exclusion Criteria

The inclusion criteria included children within the specified age range and residing in the study area at the time of data collection. The exclusion criteria included children with a known diagnosis of syndromic hypertelorism or those who were unwilling to participate.

Statistical analysis

The data obtained in this study were entered and analysed using GraphPad Prism version 8. For comparisons involving variables such as gender, t-test was employed. Median, mean, and standard deviation were used to report the quantitative outcome variables, namely IPD, ICD, and OCD Indices. To assess the association between age and the IPD, ICD, and OCD indices, Spearman's correlation coefficient was calculated. A significance level of $p < 0.05$ was considered as statistically significant.

Ethical considerations

This study was approved by Adeleke University Ethical Review Committee (09-2022). The study was conducted in accordance with the Declaration of Helsinki. The present study did not interfere with the process of diagnosis and treatment of patients and all participants signed an informed consent form.

Results

In Table 1, it is observed that the average age of male participants was 9.0±0.8, with the maximum and minimum ages recorded as 0 and 8 respectively. Similarly, for female participants, the mean age, along with the maximum and minimum ages, were 9.49±0.5, 0, and 9 respectively. Overall, when considering all subjects regardless of gender, the mean age was 9.25±0.7, with the maximum and minimum ages being 0 and 8 respectively, as shown in Table 1.

Table 2 presents the findings for IPD, revealing the mean, maximum, and minimum values as 5.74 ± 0.3 cm, 6.3 cm, and 5.2 cm, respectively. Additionally, the mean, maximum, and minimum values for ICD are reported as 2.76±0.37 cm, 3.5 cm, and 2. cm, respectively. Furthermore, the mean OCD in this study is recorded as 0.39 ± 0.7 cm, while the maximum and minimum values for OCD are .4 cm and 9. cm, as shown in Table 2.

Table 3 presents a comparison of the IPD, ICD, and OCD indices between male and female participants in this study. The analysis revealed that there were no statistically significant differences between males and females in terms of IPD (p-value=0.4926), OCD (p-value=0.950), and ICD (p-value=0.0894).

Table 4 presents the calculation of Spearman's correlation coefficient to evaluate the association between age and the indices of IPD, ICD, and OCD. The results indicate that there was no correlation observed between age and certain variables, specifically IPD and ICD, with correlation coefficients of 0.066 (p=0.2577) and -0.023 (p=0.6875) respectively. However, a significant correlation was found between age and OCD, with a correlation coefficient of -0.25 (p<0.000) [Table 4].

Table 1. Gender and age distribution of study participants (n=300)

Gender (N)	Mean ± SD	Median (Min-Max)
MALE (150)	9.0±0.8	9 (8-10)
FEMALE (150)	9.49 ± 0.5	9 (9-10)
Both gender (300)	9.25 ± 0.7	9 (8-10)

Table 2. Interpupillary Distance (IPD), Inner Canthal Distance (ICD) and Outer Canthal Distance (OCD) indices of study participants (n=300)

Variable	Mean ± SD	Median (Min-Max)
Interpupillary Distance (cm)	5.74 ± 0.3	5.7 (5.2-6.3)
Inner Canthal Distance (cm)	2.76 ± 0.37	2.8 (2.1-3.5)
Outer Canthal Distance (cm)	10.39± 0.7	10.4 (9.1-11.4)

Table 3. Comparison of Interpupillary Distance (IPD), Inner Canthal Distance (ICD) and Outer Canthal Distance (OCD) indices among male and female study participants (n=300)

Variable	Gender	Mean ± SD	Median (Min-Max)	p-value
Interpupillary Distance (cm)	Male	5.75 ± 0.3	5.75 (5.2-6.3)	0.4926
	Female	5.73 ± 0.2	5.73 (5.4-6.2)	
Inner Canthal Distance (cm)	Male	2.8± 0.4	2.8 (2.1-3.5)	0.0894
	Female	2.72± 0.37	2.75 (2.1-3.3)	
Outer Canthal Distance (cm)	Male	10±0.57	10.0 (9.5-11)	0.950
	Female	10±0.73	11.0 (9.1-11)	

Table 4. Correlation between age of subjects and IPD, ICD and OCD Indices (n=300)

Variable	Pearson r	R squared	P- value
Age vs IPD	0.066	0.0043	0.2577
Age vs ICD	0.023	0.00054	0.6875
Age vs OCD	-0.25	0.062	<0.000

Table 5 showcases the analysis of the medical history of the study participants. Among the total of 300 participants, 5.7% had received a diagnosis for at least one medical condition, while 3% had undergone surgeries or medical interventions. None of the participants had previously been diagnosed with hypertelorism or any other facial abnormalities. However, 4% of the participants reported that a family member had a history of hypertelorism or other craniofacial abnormalities.

Table 6 shows correlation between family history of hypertelorism with three facial indices: IPD, ICD, and OCD. The results showed very weak positive correlations

for all three variables. For IPD, the correlation coefficient (r) was 0.087, explaining only 0.53% of variance (R squared). The correlation for ICD had r of 0.029, explaining 0.05% of variance. OCD had r of 0.034, explaining 6.2% of variance. However, none of these correlations were statistically significant, suggesting the observed relationships may have occurred by chance.

Table 7 illustrates the evaluation of hypertelorism among the study participants, as reported by the parents. Out of the 300 participants' parents who responded, 7.7% expressed concern regarding their child's eye spacing, either personally or based on feedback from healthcare professionals. Furthermore, 20.3% of parents noticed a greater distance between their child's eyes compared to other children of the same age. Additionally, 0.7% reported that someone had made comments about their child's eye spacing or overall facial appearance.

The analysis of the participants' lifestyle and certain environmental factors [Table 8] reveals that among the 300 individuals included in the study, 9% had encountered facial trauma or injury at some point. Additionally, 6% were engaged in contact sports or activities that could potentially elevate the chances of facial injuries, and 6.3% had been exposed to environmental factors or substances that might be linked to craniofacial abnormalities.

The evaluation of the overall health and development of the 300 study participants indicates that 7.3% of them have encountered developmental delays or concerns pertaining to speech, motor skills, or cognitive abilities. Furthermore, % of the participants had issues with vision or hearing, while 5.3% had encountered various other health conditions or concerns [Table 9].

Table 5. Medical history study participants (n=300)

Variable	Yes (%)	No (%)
Child been diagnosed with any medical conditions?	7 (5.7%)	283 (94.3%)
Child undergone any surgeries or medical interventions?	4 (.3%)	296 (98.7%)
Child ever been diagnosed with hypertelorism or any other facial abnormalities?	0 (0%)	300 (00%)
Any family member (parents, siblings, etc.) with a history of hypertelorism or other craniofacial abnormalities?	2 (4%)	288 (96%)

Table 6. Correlation between family history of hypertelorism and subject`s IPD, ICD and OCD Indices (n=300)

Variable	Pearson r	R squared	P-value
Family history vs IPD	0.087	0.0053	0.3677
Family history vs ICD	0.029	0.00050	0.8865
Family history vs OCD	0.034	0.062	0.682

Table 7. Hypertelorism Assessment Amongst Study Participants (n=300)

Variable	Yes (%)	No (%)
Has your child's eye spacing been a concern to you or any healthcare professional?	53 (7.7%)	247 (82.3%)
Have you noticed any increased distance between your child's eyes compared to other children of the same age?	6 (20.3%)	239 (79.7%)
Has anyone ever commented on your child's eye spacing or facial appearance?	32 (0.7%)	268 (89.3%)

Table 8. Lifestyle and Environmental Factors Amongst Study Participants (n=300)

Variable	Yes (%)	No (%)
Has your child ever experienced any facial trauma or injury?	27 (9%)	273 (9%)
Does your child participate in any contact sports or activities that may increase the risk of facial injuries?	8 (6%)	282(94%)
Has your child been exposed to any environmental factors or substances that may be associated with craniofacial abnormalities?	9 (6.3%)	28 (93.7%)

Table 9. General Health and Development of Study Participants (n=300)

Variable	Yes (%)	No (%)
Has your child had any developmental delays or concerns in areas such as speech, motor skills, or cognitive abilities?	22 (7.3%)	278 (92.7%)
Has your child had any vision or hearing problems?	3 (%)	297 (99%)
Has your child experienced any other health conditions or concerns?	6 (5.3%)	284 (94.7%)

Discussion

Hypertelorism is an uncommon condition, with a relatively low estimated incidence rate. Based on certain sources, the occurrence is approximately in 20,000 births.[14] Hypertelorism is commonly assessed by measuring the ICD, or outer corners (outer canthal distance) of the eyes. In this study, the mean ICD was measured to be 2.76±0.37 cm. In males, the ICD was found to be 2.8±0.4 cm, while in females, it was 2.72±0.37 cm, showing no significant difference between the genders. A previous study conducted by Laestadius et al.[13] reported the mean normal value of ICD in males to be 2.6 cm and 2.8 cm in females, within the 8-10 age range. Despite some variation in the study population between Laestadius et al.'s research and this study, the outcomes were found to be similar.

Gupta et al.[12] found that gender did not have any significant impact on the OCD, ICD, and IPD values in the normal Indian population, for both children and adults. In their study, Bhalla et al.,[6] observed that male children had relatively higher OCD, ICD, and IPD values compared to females of the same age. This study reveals that the mean OCD in males is 0.39±0.57 cm, whereas in females, it is 0.39±0.73 cm. A comparable study carried out by Laestadius et al.[13] on Caucasian individuals reported a normal male OCD of 0.8 cm and 0.3 cm in females. Like Laestadius et al.'s study, no significant difference was observed between male and female OCD in this study. IPD refers to the distance between the centers of the pupils of the eyes. The measurement of IPD is essential for various purposes. In fact, individuals with hypertelorism may have several ocular manifestations, including problems with binocular vision, strabismus (misalignment of the eyes), and reduced visual acuity.[11] Typically, the IPD of the average adult falls between 54 to 74 mm, whereas children generally have slightly smaller IPDs. In this study, the male IPD is 5.75±0.3 cm, while in females, it is 5.73±0.2 cm. No significant difference was found between the genders. Taken together, this result suggests that IPD, OCD and ICD in children between 8-10 years in Ugwolawo are independent of gender.

Certain studies have reported sex-based distinctions in orbital measurements Evereklioglu et al.,[14] while other researchers have not identified statistically significant differences between male and female children concerning the mean age.[15] This study indicates that there was no correlation between age and IPD, as well as between age and ICD. However, a significant correlation was found between age and OCD. These outcomes indicate that, as children aged between 8-10 years in Ugwolawo, there was no significant change in their interpupillary distance and inner canthal distance. In other words, the size or distance between their pupils and inner corners of their eyes did not vary significantly with age during this developmental stage. Furthermore, this suggests that as the children progressed through this age range, there was a consistent pattern of change in their outer canthal distance. The rationale for this is not fully understood now but will form the basis for future investigation.

The medical history of the participants in this study showed that none of the participants had prior diagnoses of hypertelorism or facial abnormalities, but a noteworthy number (4%) reported a family history of hypertelorism or other craniofacial abnormalities. The outcome of correlation between family history of hypertelorism and facial indices (ICD, OCD & IPD) suggest that there is no statistically significant correlation between family history of hypertelorism and the subject's IPD, ICD, or OCD. The low correlation coefficients and insignificant p-values indicate that these facial indices do not appear to be strongly influenced by a family history of hypertelorism but rather occurred by chance.

Conclusions

The study provides valuable data on the prevalence and characteristics of hypertelorism among 8–0-year-old children in the Ugwolawo community of Kogi State, Nigeria. The absence of diagnosed cases of hypertelorism in the studied population suggests a relatively low prevalence of the condition. However, the presence of family history in 4% of the participants highlights the importance of considering genetic factors in future research. The correlations between age and certain facial indices add to our understanding of the developmental patterns in this age group. Overall, the study's findings shed light on important aspects of hypertelorism in the Ugwolawo community, contributing to further research

and potential interventions for this condition. We recommend other studies with larger sample size.

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Competing interests

The authors declare that they have no competing interests.

Abbreviations

Inner Canthal Distance: ICD; Interpupillary Distance: IPD; Outer Canthal Distance: OCD.

Authors' contributions

NA: Designed the experiment. EA: carried out literature review. RA and AA: Analyzed data. All authors collected data, read, and approved the final manuscript. All authors take responsibility for the integrity of the data and the accuracy of the data analysis.

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Role of the funding source

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Availability of data and materials

The data used in this study are available from the corresponding author on request.

Ethics approval and consent to participate

This study was approved by Adeleke University Ethical Review Committee (09-2022. The study was conducted in accordance with the Declaration of Helsinki.

Consent for publication

By submitting this document, the authors declare their consent for the final accepted version of the manuscript to be considered for publication.

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