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A Prospective Study of Cranial Deformity and Delayed Development in Children

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Abstract: Plagiocephaly, the most common form of cranial deformity, has become more prevalent in recent years. Many authors have described a number of sequelae of poorly defined etiologies, although several gaps exist in their real scope. This study aimed to analyze the effects of physiotherapy treatments and cranial orthoses on the psychomotor development of infants with cranial deformities, complemented by protocolized postural exercises applied by the family. This prospective study on different developmental areas included a sample of 48 breastfeeding infants aged 6 to 18 months who presented with plagiocephaly (flat head syndrome). The Brunet–Lézine scale was used to perform three tests for assessing the psychomotor development of infants, thus offering a measure for global development. The results suggest that plagiocephaly is a marker for the risk of delayed development, particularly in motor and language areas. This delayed development could be improved with physiotherapy and orthopedic treatment, complemented by interventions by the infants´ relatives.

Keywords: plagiocephaly; child development; early intervention; speech development; motor skills disorders

1. Introduction

Deformational plagiocephaly describes one of the most common head shape disorders, which is characterized by occipital flattening and asymmetric cranial vault. It has become a common reason for parents to seek a pediatrician's advice [1]

Presently, its incidence in healthy full-term infants is extremely high [2]. For example, a study in Alberta (Canada) showed that the incidence of plagiocephaly in infants aged 7 to 12 weeks was estimated to be 46.60% in the 440 infants assessed. Among those affected, 78.30% had a mild form and 63.20% were affected on the right side [3]. Various studies have reported a considerable increase in consultations concerning occipital plagiocephaly [4].

The estimated incident rate of posterior plagiocephaly, due to the different available diagnostic criteria, was between 0.3% and 3.0% before 1992 [5] and then showed a steep increase to 48.0% in 2003 [6]. Some authors have indicated that the prevalence of this disorder varies with age; the prevalence decreases as age increases, with around 6.1% to 13.0% at birth, 3.3% at two years of age, and 2.4% at three years of age [7,8].

This increase in the incidence and prevalence of plagiocephaly can be attributed to the prevention campaign of sudden infant death syndrome (SIDS), called "Back to Sleep" by the American Academy

of Pediatrics (AAP) [9]. This campaign was initiated to reduce the rates of SIDS by informing parents. These risk factors for SIDS include prone and side sleeping, soft bedding, bed sharing, inappropriate sleep surfaces (including sofas), exposure to tobacco smoke, and premature birth. Protective factors to prevent SIDS include breast-feeding, pacifier use, room sharing, and immunizations [10]. These efforts were quite successful, resulting in changes in parenting practices and a corresponding decrease in SIDS; however, it is widely believed that an unintended "side effect" of this program was the exponential increase in the rate of plagiocephaly [11,12].

Most relevant studies have described the long-term sequelae in association with the most severe cranial deformities with problems that have no effect on cerebral functions, such as orthodontics and temporomandibular joint disorder syndrome [13]. In addition, this disorder's association with ocular alterations, such as exotropia and psychosocial disorders, has been examined [14,15].

Some studies have emphasized the significance of alleviating the consequences of this disorder [16], claiming that this cosmetic deformity may be corrected, albeit incompletely, over time. Similarly, these studies have indicated that this deformity does not interfere with normal brain growth and cerebral development [4].

The results of several studies suggest that infants with plagiocephaly tend to have below-average cognitive and motor development, in areas such as gross motor, fine motor, problem solving, and personal social skills [17–20]. However, other authors, such as Hussein et al. (2018), concluded that there is no definitive relationship between the severity of plagiocephaly and the degree of developmental delay [21].

The amount of time spent an infant spends in a prone position while awake has been positively correlated with better performance in the motor developmental. Therapists should be aware of the risk of motor delay when evaluating infants in a prone position. It is also important for parents to be informed about the importance of supervised prone playtime to enhance the development of early motor skills [22].

There is little evidence about long-term developmental trajectories. Several recent studies compare cognitive and academic functioning of children with plagiocephaly to non-affected peers at school age [23,24]. Their findings suggest that plagiocephaly is not associated with increased risk of general cognitive and academic difficulties at school age. A close developmental monitoring for infants with moderate to severe forms of plagiocephaly is recommended as this group performed significantly lower on both cognitive and academic measures. These associations do not necessarily to be causal; rather, moderate/severe plagiocephaly may a marker for developmental risk

Controversy exists regarding the management of positional deformities, such as engaging in supervised position changes versus using cranial molding orthotic [25]. Although correcting aesthetic defects is the main purpose of orthotics, their effects on cognitive and motor development are still debated. No differences were noted in the modification of the cranial index when using the management of supervised position changes. Some patients improved spontaneously when they acquired head support and verticalization [14].

In the literature, most studies on infants with plagiocephaly focus on analyzing the effectiveness of the application of different physiotherapeutic techniques via a healthcare professional [26,27]. However, no studies have investigated the usefulness of complementing this therapy with home protocolized exercises performed by the infants' relatives. Therefore, the current study aimed to analyze the effects of the physiotherapy treatments and cranial orthoses on psychomotor development among infants with cranial deformities, complemented by protocolized postural exercises applied by their relatives.

2. Method

2.1. Participants

A prospective cohort-based study was conducted, including a sample of early infants with plagiocephaly. A total of 60 infants, who attended the center for rehabilitation and diagnosis in Burgos

(Spain) only for plagiocephaly and not for another type of deformity, were initially considered. An initial survey was carried out to collect information from the parents. The infants had not received any prior interventions, advice, or preventions before arriving at the rehabilitation center. Extremely premature infants (less than 34 weeks) and those with rare syndromes or chromosomal disorders and other alterations were excluded. Infants with an Apgar score of less than or equal to 7 at one minute and less than or equal to 8 at five minutes were also excluded. The remaining individuals constituted the entire affected population.

One of the sample selection criteria for the infants was to have a normal gestational age at the time of birth (gestation weeks: mean = 38.14, SD = 2.12), thus excluding those with fetal stress whose progress may have compromised their psychomotor development. None of the infants in the sample were in the neonatal intensive care unit or suffered from perinatal hypoxia. In this study, no control group was used due to ethical considerations.

We obtained approval from the appropriate Local and National Ethical Standards Committee (University of Burgos Bioethics Committee Approval IR12/2018). Written informed consent was obtained from all parents before initiating the study.

2.2. Procedures

The infants were referred by their pediatrician to the public rehabilitation center once they were diagnosed with plagiocephaly by a specialist physician based on the appropriate anthropometric measures. Once the infants arrived at the center, they were first assessed before receiving intervention. The infants received the same treatment (frequency and intensity) in the form of physiotherapy exercises, Bobath therapy, cranial–sacral therapy, and postural orientations (increasing the usual developmental recommendations for "tummy time" or upright positioning) for one hour a week, reviewed on a bi-weekly and monthly basis, according to each particular case; in the most severe cases (22 of 48 infants with a Cranial Vault Asymmetry Index (CVAI) >12%), cranial orthoses, such as helmets, were provided for 23 h a day with periodic revisions.

This treatment was complemented by physiotherapeutic techniques applied by the infants´ relatives, which had to be developed in 10-min sessions, repeated three times throughout the day. All infants´ relatives received a 2-h practical session in which they were taught functional exercises and caregiver training. To assess the correct performance of these techniques, direct observation was used. Any error in development was then corrected, obviating any possible doubts of the treatment's efficacy. These techniques consisted of stretching the sternocleidomastoid and trapezium muscles, as well as maintained posture during neck rotation and inclination. In each 10-min period, each exercise was repeated twice every 30 s. These exercises were complemented by a series of postural orientations done by the family, to be performed in all the activities carried out by the infant throughout the day: playing in prone, being held, and sleeping with a tilt and rotation of the head to the opposite side of the plagiocephaly. All the children received the same physiotherapy interventions. To check the adherence of the family members to the home physiotherapy program, a daily self-registration was used.

A total of three waves of tests were conducted. These tests were tested every 6 months by the physiotherapist at the center. The 2nd and 3rd developmental measurements were used to determine the effects of different therapies over time. The physiotherapist was not blind to the type of treatment the infants were receiving.

2.3. Instrumentation

The anthropometric measurements of the plagiocephaly used Cranial Vault Asymmetry (CVA) and the Cranial Vault Asymmetry Index (CVAI). CVA, also known as oblique diagonal difference or transcranial difference, is obtained by subtracting the smaller cranial diagonal from the larger cranial diagonal. The CVAI can be defined as the absolute value of the difference in cranial diagonals (CVA) divided by the smaller diagonal and multiplied by 100 (normal: <3.5%; mild: 3.5%–7.0%; moderate: 7.0%–12.0%; severe: >12.0%) [10,28]. This index allows for a direct comparison between cranial

deformities in infants with varying head sizes. Diagonals are measured from the anterolateral skull to the posterior skull at the level of the greater equator of the skull. These diagonals are measured 30 degrees clockwise and counterclockwise from the midsagittal line. The change in CVAI was calculated as the final CVAI minus the initial CVAI. A completely symmetrical skull will have an index of 0% [28].

We used the revised Brunet–Lézine early childhood Scale of Psychomotor Development for infants aged between 2 to 30 months for data collection [29]. This scale is used to measure psychomotor development during early childhood. It is appropriate for children between the ages of 0 and 30 months and includes complimentary tests for those up to six years of age: one applicable for those aged from 24 months to 5 years and others for those aged 3, 4, 5, and 6 years, with fewer verbal tests. This scale assesses a child's development in four areas: posture control and motoricity; hand–eye coordination and adapting to objects behavior, language, and sociability; and personal and social relations. This scale comprises two parts: one experiential, in which tests are directly administered to the child, and another in which the child's behavior is observed in his or her daily life. This information was collected by asking questions to the parents. Using the scale, the developmental age (DA) and developmental quotient (DQ) were obtained for each area, along with a total score that combined all areas [30] (mean = 100.0 and SD = 15.0). When interpreting these scores, the delay was considered to be mild if the value was less than 1 SD, moderate if it was more than 1 SD and less than 2 SD, and severe if it was more than 2 SD. The test–retest reliability coefficient was 0.689 in the original Brunet–Lézine development scale [29]. In this study, we observed a high-level test–retest reliability coefficient (0.93). The scale version was obtained from the TEA Editions [29].

2.4. Data Analysis

The mean and the standard deviation were used for the descriptive analysis. A chi-square test was used to compare the categorical variables. Given that the distribution was normal according to the Kolmogorov–Smirnov test, a repeated measures analysis of variance (ANOVA) and Student's t-test were used for the numerical variables. The Kolmogorov–Smirnov test showed a normal distribution in the three tests. $p > 0.05$ was obtained via Mauchly's test of sphericity to determine the homogeneity of the covariances, and a repeated measures ANOVA test was also used. To quantify the magnitude of the differences the η^2 test was used. A p-value of <0.05 was considered statistically significant. The statistical analysis was performed using the SPSS statistical software package version 25.0 (IBM SPSS Inc, Chicago, IL, USA).

3. Results

The study sample consisted of 48 infants; 23 were males and 25 were females, with a mean age of 5.60 months (±3.03) in the first test, 10.47 months (±4.87) is the second test, and 15.96 months (±5.86) in the third test. The CVAI at the first evaluation was 9.43% and was 4.71% at last evaluation.

In the first test, a significant delay (2 SD) was observed in global development, with a coefficient of 64.30, which was lower by two standard deviations (15.00) from the mean (100.00). The social area was the most developed (DQ = 71.60), followed by the language (DQ = 64.00) and visual–motor coordination (DQ = 62.30). Motor skill development showed the greatest delay (DQ = 58.40).

The second test was completed after six months. It should be noted that the infants in this test continued to receive treatment since the first test. A greater overall delay in the development quotient was observed during this test (DQ = 80.10), although this value did not reach statistical significance. Once again, the social area was the most developed area (DQ = 84.30), followed by visual–motor coordination (DQ = 80.10) and language (DQ = 78.60), with almost the same scores as the score for motor skills (DQ = 78.00).

The third test was performed six months after the second evaluation. The development coefficients were increased and, despite a slight global delay (DQ = 87.00), were once again not significant. Moreover, the scores became similar across all areas: visual–motor coordination (DQ = 87.30), social

(DQ = 88.40), language (DQ = 87.40), and general motor skills (DQ = 84.50); all areas showed slightly delayed development.

Table 1 shows the data for the three consecutive tests performed to evaluate the different areas of development.

Table 1. Descriptive statistics of the Brunet–Lézine variables measured in the three tests, and the ANOVA test.

Variable	Test	Mean	Std. Dev.	Confidence Interval 95%	
				Lower Limit	Upper Limit
Postural Control $p = 0.001$ $\eta^2 = 0.303$	1	58.81	2.91	52.96	64.66
	2	78.03	2.13	73.75	82.32
	3	84.53	2.12	80.26	88.80
Visual motor Coordination $p = 0.001$ $\eta^2 = 0.270$	1	62.40	2.91	56.54	68.26
	2	80.18	2.36	75.43	84.93
	3	87.33	2.22	82.87	91.79
Language $p = 0.001$ $\eta^2 = 0.182$	1	64.04	3.61	56.78	71.30
	2	78.66	2.65	73.33	83.98
	3	87.43	2.59	82.23	92.64
Socialization $p = 0.001$ $\eta^2 = 0.103$	1	71.60	3.61	64.33	78.88
	2	84.35	2.81	78.69	90.01
	3	88.43	2.73	82.94	93.93
Global $p = 0.001$ $\eta^2 = 0.236$	1	64.32	2.88	58.53	70.11
	2	80.16	2.28	75.58	84.74
	3	87.02	2.27	82.45	91.58

In the multivariate analysis, to determine the improvement between the different assessments, an ANOVA test was used to confirm the significant differences between the three tests. According to the effect size, calculated through η^2, the differences between the three tests were high (for all tests: $p = 0.001$ and η^2 test between 0.103 and 0.303).

A post hoc Bonferroni test was used to determine the significant differences between the tests. A significant difference was observed between the first and second tests in all areas of the development scale ($p < 0.001$) and the global development coefficient. Thus, the total Brunet–Lézine test scored showed a significant difference between the first and second tests ($p < 0.001$).

Furthermore, a significant difference was observed between the second and third tests for the global development coefficient ($p < 0.001$) in all areas of the development scale ($p < 0.001$), except for socialization ($p = 0.053$). The total Brunet–Lézine test score showed a significant difference between the second and third tests ($p < 0.001$). The development coefficient increased in all the areas, particularly in the language area. However, the increase was lowest for socialization.

Our results show significant (more than one standard deviation) differences in all areas of development between the first and third evaluations. These differences were clearly evident during the 12 months that elapsed between the first and the third evaluations in which the infants received treatment. The increase was highest in the postural control area and lowest in the socialization area, which could be due to the intervention focusing more on some of the items and not on all.

4. Discussion

This study showed that plagiocephaly in babies/infants is more strongly associated with the motor skills area and less with the social area, as shown by previous studies [27]. In the first test, the infants

showed a significant developmental delay in all areas, with a global development coefficient of 64.3, which was lower than two standard deviations of the norm [29].

After completing the first test and evaluating the unit of early attention, the infants subsequently received physiotherapy with postural orientations or orthotic treatments in weekly sessions, which were reviewed on a fortnightly and monthly basis by the rehabilitation center doctor, according to the CVAI of each particular case.

A recent systematic review suggests that immediate referral to early intervention services, such as physiotherapy, ameliorates motor delays [27].

A significant difference was observed in all areas and global development coefficients between the first and third tests. The greatest improvement was noted in the motor skills area and the weakest was observed in socialization. Some studies have shown that the type of treatment, predominantly physiotherapy or cranial orthosis, is a key factor influencing motor-related areas [14,31,32].

Therefore, major differences occurred between the first and second evaluations. The areas with the greatest increase were those that had the greatest developmental delays, which evened out in the third test.

These results are in line with those of other descriptive studies without treatment [14,33], showing that differences in psychomotor development are temporal and disappear before 18 months of age.

Several studies have shown the existence of delayed motor development in infants with plagiocephaly [34–36]. According to Pollack [37], children who sleep and remain awake in the supine position have a greater risk of delayed development of their motor skills as a consequence of a potential plagiocephaly caused by the positioning of the children.

Several researchers have shown an association between the alterations of child language and learning disorders with neuropsychological deficits [38,39] and between psychomotor development and verbal and nonverbal intelligence [40].

Moreover, in line with our results, studies using the Alberta Infant Motor Scale and the Peabody scale [22] reported a motor development coefficient of 84.1 and a global development coefficient of 86.8 for children with deformities between ages 3 and 7.5 months.

Moreover, our study found that children with cranial deformations had temporal alterations in their psychomotor-related development, consistent with previous studies [33]. The results of these studies tailed off before the children were 18 months of age, although their motor skills at age 5 and 6 years were less precise.

Nevertheless, controversy persists over the repercussions of plagiocephaly on child development. Early researchers believed cranial deformities not to cause cerebral dysfunction with delayed psychomotor development, as only occasional cases of ocular visual and auditory delays had been observed [13]. Others have pointed out the lack of scientific evidence of other associated alterations that are not related to cranial and facial deformities [4].

In previous studies, a significant neurodevelopmental delay was observed in children with deformational plagiocephaly, although no definitive relationship between the severity of deformation and the degree of developmental delay was found (the psychomotor development index was only affected by congenital anomalies) [41]. On the other hand, a systematic review showed a positive association between plagiocephaly and developmental delay in 13 of 19 studies, including four of five studies with a "strong" methodological quality [27]. Moreover, this delay was less frequent in studies with children more than 24 months of age, and motor skill development was the most commonly affected area, similar to the results of our study.

The risk factors most frequently reported in the etiology of plagiocephaly are male gender, a supine sleep position, limited neck rotation or preference in head position, being firstborn, a lower level of activity, and a lack of tummy time [41]. Studies reporting the possible etiologies of plagiocephaly have analyzed a wide range of environmental and biological factors but have included few suggestions on the potential influence of everyday baby care environments [41]. The concordance between these studies is very poor for most exposures.

Some recent studies have indicated that pediatricians frequently face positional plagiocephaly associated with a supine sleeping position, head position preference, and an older maternal age [2,21,42]. There is a need to prevent both sudden infant death syndrome and positional plagiocephaly by providing small suggestions to influence the everyday care environment of infants through messages to parents, prenatally and postnatally, by different health professionals [21,41]. One review suggests that plagiocephaly is a marker of elevated risk of developmental delays and that clinicians should closely monitor infants with plagiocephaly for this marker. Prompt referral to early intervention services, such as physiotherapy, may ameliorate motor delays and identify infants with longer term developmental needs [27]. Finally, playing in the prone position and a child´s balance could be affected by this deformity.

The motor skills area should be considered, as it was the most affected area in all evaluations and is crucial in the subsequent evolutive development of a child. Notably, these aspects should also be controlled after discharge when the child enters a new social setting, such as school, where continued evaluation and interdisciplinary monitoring may be necessary.

Early developmental assessment should be considered in the examination of infants with plagiocephaly to improve results in the medium and long term, especially in the most serious cases [22,23]. Even a modest effect can have significant cumulative public health consequences when an exposure or condition is highly prevalent [43]. In this way, delays in cognitive and academic development in later ages can be avoided [23].

The main limitation of this study was the absence of a control group to compare the results, but ethically, this was not feasible. However, these observational data were obtained from a prospective database created for the purpose of this study, without any prespecified hypotheses at the time of data collection, thereby reducing the sample selection bias. Descriptive studies may help to produce early diagnoses to ameliorate motor delays and identify infants with long-term developmental needs.

The association between plagiocephaly and different areas of development does not necessarily indicate a causal relationship. This is the reason why the future studies of early development and head shape from birth should help to clarify whether motor deficits precede or follow skull deformation.

In conclusion, the results suggest that infants with plagiocephaly are more at risk for developmental delays, particularly in the motor and language areas. However, these results should be considered with caution, as this study design does not allow us to draw a cause–effect relationship between plagiocephaly and developmental delays. Delayed development could be improved with physiotherapy and orthopedic treatments complemented by interventions by the infants´ relatives.

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