ORIGINAL ARTICLE

Occurrence and Severity of Deformational Plagiocephaly in Infants: A Single Center Experience

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ABSTRACT

Objectives: To estimate the occurrence and severity of deformational plagiocephaly among infants.

Methods: A hospital-based, cross-sectional study was done in the pediatric ward of a tertiary care hospital between April 1, 2022 to October 31, 2022. Cranial Vault Asymmetry Index (CVAI) and Argenta Clinical Classification were applied to consecutive infants aged 1 month to 1 year till the calculated sample size was achieved.

Results: 67 infants were recruited and the occurrence of deformational plagiocephaly in the sample was estimated to be 46.3%. Level 2 severity of deformational plagiocephaly was the commonest, while as per the Argenta classification, majority belonged to type I (39.2%). Male gender and developmental delay were the significant risk factors for plagiocephaly with an odds ratio (95% CI) of 3.73 (1.23, 11.26) and 19.25 (2.31, 160.3), respectively.

Conclusion: A high occurrence of deformational plagiocephaly was found in infants studied. There is a need for more studies to further corroborate these findings and study its associated factors.

Keyword: *Cephalometry, Cranium, Intellectual Disability, Prone position*

INTRODUCTION

Deformational plagiocephaly is a condition of cranial distortion leading to a flattening of the skull bones in infants attributed to an external molding force. Although deformational plagiocephaly is a type of cranial distortion and flattening, all kinds of cranial flattening are not deformational plagiocephaly [1]. For diagnosing this condition, various methods like anthropometry, imaging studies, and 3D reconstruction using computer software are available [2]. While craniometry is easier to perform and less resource-intensive, imaging studies and 3D reconstruction have proven more precise. However, in resource-constrained countries like India, imaging studies and 3D reconstruction are not routinely available or affordable, and very few centers offer the expertise needed, hence data on the deformational plagiocephaly among Indian infants is scarce. Extrapolating global data and findings on the Indian population is inappropriate owing to different demographics, risk factors, racial, cultural, and child-rearing practices. The global prevalence of deformational plagiocephaly amongst infants aged 1 month to 1year ranges from 6.8 % to 40.5% [3,4]. Some of the risk factors associated with deformational plagiocephaly are - antepartum (oligohydramnios, multifetal gestation); intrapartum (birth order, presentation at birth, mode of delivery, male gender, and premature delivery) and postpartum (torticollis, reduced tummy time, developmental delay) [3-5]. Traditional methods like massaging, pillow use, especially the ones with mustard seeds and repositioning are expected to reduce the incidence of deformational plagiocephaly [6].

Other than estimating the prevalence, there is also a need to categorize the infants based on the severity as measured by cephalometry. Severity assessment also helps in management as recommended by the Child Healthcare Organization of Atlanta (CHOA). Argenta Clinical Classification has been used as a reliable tool to evaluate cranial deformities and can even help predict the optimal duration of treatment [7]. The current study is aimed to determine the occurence and severity of deformational plagiocephaly among Indian infants and the risk factors for the same.

METHODS

A hospital-based study was carried out on infants aged 1months to 1 year admitted to the in-patient ward of the Department of Pediatrics of a tertiary care institute in Eastern India between April 1, 2022 and October 31, 2022. Infants with underlying conditions that could alter the

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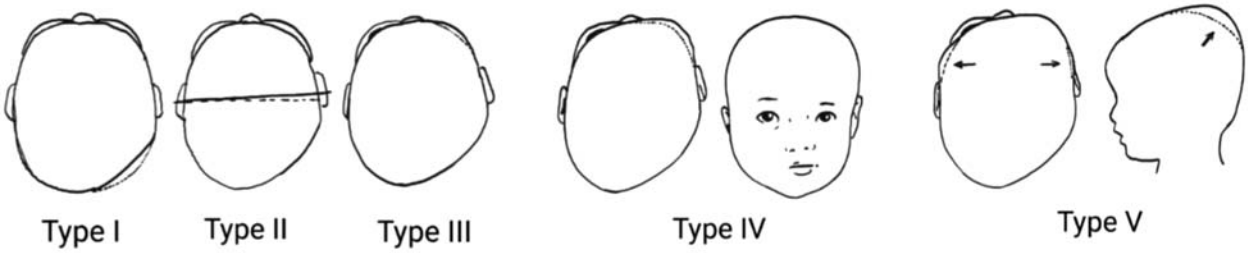
cranium shape, such as craniosynostosis, hydrocephalus, and sick infants were excluded from the study. Institutional Ethics Committee approval was obtained before the start of the study. Informed consent was obtained from parents after explaining the study details.

Based on a study from New Zealand wherein the prevalence of deformational plagiocephaly at 12 months of age was reported as 6.8% using craniometry [3], and assuming a prevalence of 5% in Indian children, at 95% confidence level and 5% margin of error, the sample size was calculated as 73.

A predesigned proforma was used to collect the demographic details including antenatal, natal, and postnatal history, as well as tummy time of infants. To avoid recall bias, documents like antenatal check-up records, vaccination cards, and ultrasonography reports were cross-checked. Tummy time was defined as an awake-prone position of a child supervised by an adult and was defined as "adequate" if at least three episodes of 10 minutes each or 30 minutes of total tummy time spread throughout the day while awake [8]. The developmental assessment was performed, and developmental delay was adjudged based on Trivandrum Development Screening chart. The developmental quotient was calculated for all infants participating in the study. Cephalometry was performed by the methodology described by Wilbrand et al [9]. Physical examination of the infant for identifying torticollis was done by assessing the active and passive movement of the head and checking sternocleidomastoid (SCM) tightness for a cord like feeling.

Cranial Vault Asymmetry Index (CVAI) was calculated by measuring the difference of the cranial diagonals (A and B, such that A>B) multiplied by 100 and divided by the longer cranial diameter A [9]. All measurements were done using a craniometer within the first 2 days of admission. All the measurements were obtained by a single person to avoid inter-observer variability. In a child with deformational plagiocephaly where there was an anterior ear shift on the side of the deformational plagiocephaly, the transverse diameter was recorded with the eurion being adjusted to the lateral most point on the temporoparietal region lying at a 90-degree angle to the AP diameter measured in line with the normally placed eurion of the opposite side. Based on the CVAI, the severity scoring was graded as, level 1 (< 3.5), level 2 (3.5-6.24), level 3 (6.25 to 8.74), level 4 (8.75 to 10.99), and level 5 (>11) [10]. Infants were classified as per the Argenta Clinical Classification by visually inspecting and clicking clinical photographs for each child (**Fig.1**). An infant was labelled as a case of deformational plagiocephaly only if he had CVAI ≥ 3.5, irrespective of the Argenta Clinical Classification of the infant.

Statistical analysis The collected data were analyzed using JAMOVI ver 2.3.18. Categorical variables were presented as frequency and percentage while the continuous data was presented as mean (SD). Fisher exact or *Chi*-square test was applied to examine the significance of association. Logistic regression analysis was performed to ascertain the risk factors for deformational plagiocephaly and odds ratio (95% CI) were computed. *P* value < 0.05 was considered statistically significant.



Clinical Finding	Type 1	Type 2	Type 3	Type 4	Type 5
Posterior asymmetry	Present	Present	Present	Present	Present
Ear malposition	Absent	Present	Present	Present	Present
Frontal asymmetry	Absent	Absent	Present	Present	Present
Facial asymmetry	Absent	Absent	Absent	Present	Present
Temporal bossing or posterior vertical cranial growth	Absent	Absent	Absent	Absent	Present

Fig.1 Argenta clinical classfication

RESULTS

Out of 69 infants who were assessed for eligibility, two infants who were found to have craniosynostosis were excluded. Out of 67 infants, 31 infants (46.3%) had CVAI ≥ 3.5 and were labeled as cases of deformational plagiocephaly. The mean (SD) age at the time of assessment was 175 (97) days in the deformational plagiocephaly group and 168 (90) days in the non-deformational plagiocephaly group (*P*=0.74).

Out of 31 infants who were found to have deformational plagiocephaly, 20 infants (64.5%) had level 2 severity, 8 infants (25.8%) had level 3 severity, 2 infants (6.5%) had level 4 severity, and 1 infant (3.2%) had level 5 severity. Based on the Argenta Clinical Classification, 11 (16.4%) infants did not show any physical deformation. Out of the remaining 56 infants, 22 (39.2%), 13 (23.2%), 18 (32.1%), and 3 (5.4%) infants were classified as Argenta Type I, II, III and IV respectively. We did not find any infant with the Argenta Type V severity (**Fig. 1**). Overall, 56 infants (83.6%) were found to have posterior asymmetry, 34 infants (50.7%) were found to have malposition of the ear, 21 infants (31.3%) were found to have forehead asymmetry, and three infants (4.48%) were found to have facial asymmetry.

Amongst the various risk factors for deformational plagiocephaly, male gender was significantly associated with developing deformational plagiocephaly (OR 3.73, *P* = 0.01) (**Table I**). Developmental delay was observed in 12 infants, and 11 (91.7%) among them had deformational plagiocephaly (OR 19.25, *P* < 0.01). There were 9 boys who had developmental delay and deformational plagiocephaly. There was no significant association observed between parity, mode of delivery, presentation at delivery, oligohydramnios or preterm delivery with deformational plagiocephaly as shown in **Table I**. Though reduced tummy time is seen in 91% of infants, we did not find any significant association between reduced tummy

Table I Risk Factors and Deformational Plagiocephaly

Factors	*Group*	*n (%)*	*Deformational Plagiocephaly*		*Odd's Ratio (95% CI)*	*P value*
			Present	*Absent*		
Gravida	Primigravida	32 (47.8)	18	14	2.17 (0.82-5.79)	0.12
	Multi-gravida	35 (52.2)	13	22		
Parity	Nullipara	39 (58.2)	20	19	1.63 (0.61-4.35)	0.33
	Multipara	28 (41.8)	11	17		
Oligohydramnios	Present	7 (10.5)	4	3	1.69(0.35-8.24)	0.51
	Absent	59 (88.5)	26	33		
	Not Known	1 (1.5)	1	0		
Type of gestation	Multifetal	3 (4.5)	1	2	0.56 (0.05- 6.57)	0.65
	Single	64 (95.5)	30	34		
Presentation at Birth	Vertex	26 (38.9)	12	14	NA	NA
	Breech	1 (1.5)	1	0		
	Not Known	40 (59.7)	18	22		
Mode of Delivery	Normal vaginal delivery	35 (52.2)	16	19	0.95 (0.36-2.5)	0.92
	Cesarean	32 (47.8)	15	17		
Preterm Delivery	Preterm	14 (20.9)	7	7	1.2 (0.37-3.93)	0.75
	Term	53 (79.1)	24	29		
Gender	Male	44 (65.6)	25	19	3.73 (1.23-11.26)	0.02
	Female	23 (34.4)	6	17		
Torticollis	Present	5 (7.5)	3	2	1.82 (0.28-11.67)	0.52
	Absent	62 (92.5)	28	34		
Reduced Tummy Time	Reduced	61 (91)	27	34	0.39 (0.07-2.33)	0.29
	Normal	6 (9)	4	2		
Developmental delay	Present	12 (17.9)	11	1	19.25 (2.31 -160.3)	< 0.01
	Absent	55 (82.1)	20	35		
Traditional Method usage	Yes	37 (55.2)	13	24	0.36 (0.13 - 0.98)	0.04
	No	30 (44.8)	18	12		

Data presented as n (%). DP Deformational Plagiocephaly, NVD Normal vaginal delivery

time and deformational plagiocephaly (**Table I**). The parents of 37 infants (55.2%) were using some traditional methods to maintain the cranial shape, which significantly reduced the occurrence of deformational plagiocephaly in our study (OR 0.36, and $P = 0.042$) (**Table I**).

DISCUSSION

The occurence of deformational plagiocephaly in our study was found to be 46.3%, which was higher than the 40.5% reported by Rocco et al [4] in the same age group and much higher than the 6.8% reported by Hutchison et al [3] in one-year-old children. There is great variability in the actual prevalence of deformational plagiocephaly in infants, which is dependent on the age at the time of assessment, tool and classification system used for evaluation. In our study, the mean (SD) age at the time of assessment was 171 (94) days and we have recorded a higher occurence as compared to that in studies where it is done within 8-12 weeks of age. Among the cases of deformational plagiocephaly, most infants (64.5%) had level 2 severity, followed by 25.8%, 6.5%, and 3.2% having level 3, 4, and 5 severities, respectively. Amongst the infants identified with plagiocephaly based on the Argenta Classification, most belonged to type I (39.2%), followed by type III (32.1 %), type II (23.2%), and type IV (5.4%). This differed from the findings of Branch et al [7] who found a maximum prevalence of type III followed by type II, type IV, type I, and type V. This may be because the study by Branch et al was a retrospective study among the infants presenting to a plastic surgeon for corrective surgery. Hence, infants with less severe type I may have been missed as they were self-limiting not requiring referral.

In our study, the significant risk factors for the development of deformational plagiocephaly were the male gender and developmental delay, with an odds ratio of 3.73 and 19.25, respectively. This was similar to the results observed by van Vlimmeren et al [6]. Though there is no concordance of risk factors associated with deformational plagiocephaly in infants in most studies but male gender and supine positioning have been associated in more than 50% of the studies [10]. We could not find any significant association of deformational plagiocephaly with parity. Similar results had been reported by van Vlimmeren et al [6] and Miyabayashi et al [11]. Unlike Solani et al [12], we could not find any significant association of oligohy-dramnios or multifetal pregnancy with deformational plagiocephaly. No significant association was observed between the mode of delivery or the type of presentation at birth in our study, similar to a study by van Vlimmeren et al. Assisted delivery has been believed to be associated with deformational plagiocephaly due to application of an instrument over the skull leading to a deformity. In our study, we could not obtain a history of assisted delivery, hence the results cannot be generalized for the same. A positive association between preterm delivery and torticollis with deformational plagiocephaly was observed in the current study though not statistically significant. This may be due to a small sample size. Preterm babies with softer skull bones may have a higher risk of positional deformity as compared to term babies [13]. Further prospective studies need to be designed specifically to look for the same in preterm babies. No significant effect of reduced tummy time was observed in developing deformational plagiocephaly in the current study which is in concordance with findings of van Ballardini et al [14]. Reduced tummy time was observed in almost 91% of our babies, which may be due to lack of awareness among parents. Traditional methods like massage, use of mustard pillow and repositioning were found to be beneficial for maintaining a normal cranial shape and hence may be recommended to parents. These traditional methods have been in practice for generations to stabilize the position of the head, which is considered one of the risk factors. Infants with positional preference of right or left during sleeping or feeding had four times higher risk of developing deformational plagiocephaly [15]. Infants with frequent change of head position while sleeping have lesser risk of having deformational plagiocephaly.

The strength of our study was the accurate measurements using a craniometer by a trained user. The limitations of the current study include a hospital-based set-up, a small sample size, the developmental assessment done in hospitalized children, recall bias and the lack of 3D cephalometry. Further prospective cohort studies are needed to ascertain the risk factors for deformational plagiocephaly.

Nearly, half of the infants in our study had deformational plagiocephaly; most cases were of severity level 2 by CVAI. Developmental delay and male gender increased the risk for deformational plagiocephaly, although parity, mode of delivery, presentation at birth, oligohydramnios and preterm delivery were not found to be associated with deformational plagiocephaly.

Ethics clearance: All India Institute of Medical Sciences, Bhubaneswar, Odisha; Institutional Ethics Committee No. IEC/AIIMSBBSR/STS/2021-2022/02 dated March 29, 2022.

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WHAT THIS STUDY ADDS?

- The occurence of deformational plagiocephaly in our study was 46.3%, much higher than that reported globally.
- Male gender and developmental delay were the most significant risk factors for deformational plagiocephaly.

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