

The Role of Age on Effectiveness of Active Repositioning Therapy in Positional Skull Deformities

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ABSTRACT

Purpose: Non-synostotic positional deformities are currently diagnosed in nearly half of the newborns, however not any evidence-based guidelines are available for management. The aim of this study is to assess the effect of active repositioning treatment at infants with positional skull deformities.

Method: A retrospective data of 158 infants treated with active repositioning as a conservative treatment for at least 2 months were analyzed in this study. Anthropometric 3D scanner measurements of pre-and post-treatment diagonal difference, cranial vault asymmetry index, cranial ratio were evaluated for each patient. Infants were separated to 4 different groups according to their morphologic deformation types as plagiocephaly, brachycephaly, scaphocephaly and combined (brachycephaly+plagiocephaly), and 2 groups according to age at onset of treatment.

Results: In combined group, pre-treatment mean diagonal difference and cranial vault asymmetry index values decreased from 9.38 mm and 6.9% to 6.94 mm and 4.9% respectively. In plagiocephaly group, mean pre-treatment results changed from 10.32 mm and 7.5% to 7.83 mm and 5.5% respectively after treatment. All these changes were statistically significant. Effectiveness of timing of repositioning treatment on different positional skull deformities was analyzed and outcome was found significantly improved when the active repositioning treatment was started before 4 months of age.

Conclusion: Improvement rates of the asymmetry decrease with age due to decreasing skull enlargement rate. Early diagnosis, especially before 4 months of age, more parental education, and close follow-up are important for babies with this condition who may benefit just from repositioning treatment.

Key Words: Positional plagiocephaly, Positional brachycephaly, Cranial deformation, conservative therapy, active repositioning therapy

INTRODUCTION

Neonatal skull is soft and moldable in the natal and newborn periods due to the rapidly growing brain tissue. Skull deformities may be classified as; pathologic type, craniosynostosis, secondary to abnormal suture development; or deformational/positional type secondary to external forces acting upon cranium. Craniosynostosis usually requires surgical intervention, however with early diagnosis, positional skull deformities may be treated with active repositioning, physical therapy and helmet therapy in infants (1-3).

American Pediatric Academy (APA) has started a campaign and suggested that the babies should be at supine position in bed to decrease sudden infant death. (4) Soon after acceptance of supine position in bed campaign in almost all countries, 50% decrease

in sudden infant death syndrome was recorded. (5) However, Argenta et. al in 1996 reported up to 600% increase in the prevalence of cranial asymmetries. Thus, a consensus has been made about the relation between deformational plagiocephaly and supine sleeping position. (6, 7)

Nowadays, skull deformities are diagnosed in 45% of infants, with most common diagnoses being plagiocephaly, brachycephaly and scaphocephaly. Symptoms may be observed initially between 4th and 8th weeks of life. (8-11). Positional plagiocephaly can be recognized as unilateral parieto-occipital flattening with ipsilateral frontal bossing and anterior shift of the ipsilateral ear that results in a parallelogram deformity of the head. Central bi-occipital flattening with an anterior to posterior shortening and medial to

lateral widening of the head is the characteristics of deformational brachycephaly, therefore it is also known as ‘short head’ syndrome (11). Scaphocephaly, ‘narrow head’, is characterized by anterior-posterior elongation and bi-parietal shortening of the skull (12, 13). Besides the cosmetic problems, it is suggested that positional deformities may constitute a risk for temporo-mandibular joint problems, motor skill deficiencies, sleep apnea syndrome, visual field defects, ear infections, difficulties at cognitive functions and academic degrees (14–18).

The first postnatal 4 months are critical for the development of positional skull deformities (PSD), and a peak is observed at the deformation level by the end of 4th month (19). Therefore, in 2008 American Pediatric Academy (APA) proposed that, infants should be positioned in a facedown position 2–3 times for 3–5 minutes, under surveillance, during their awake times to prevent cranial asymmetry, and that this duration should be increased as the child grows older (20). Since the infant skull is easier to mold, early infancy is the most favorable time to prevent PSD.

The aim of this study is to investigate the effects of early conservative treatment in PSD patients on improvement of cranial asymmetry rates.

MATERIALS AND METHODS

A retrospective analyzes of all infants admitted to our outpatient clinics due to skull shape deformities between 2014 and 2018 were performed. The infants who received positional treatment for at least 2 months were determined and included to the study. Parameters including gender, delivery method (vaginal delivery vs caesarian section), gestational age at birth (premature/mature), twin status, age at diagnosis, onset of treatment, treatment duration (days), anthropometric measures of pre-and post-treatment diagonal difference (DD), cranial vault asymmetry index (CVAI), cranial ratio (CR) were evaluated for each patient.

Cranial parameter analyze were made with SmartSoc and Omega Scanner 3D systems (Figure 1). The same instrument was used for all measurements of each individual infant throughout the study. Either of these two systems was used for each patient, they were never used together. The same technician performed the scanning and evaluated the cranial alignment for each infant.

Patients were disintegrated morphologically into 4 groups: Group I (plagiocephaly), infants whose cephalic index was between 78–89 and CVAI was greater than% 3.5; Group II (brachycephaly), infants whose cephalic index was greater than 89 and CVAI was lesser than% 3.5; Group III (scaphocephaly), infants with cephalic index lesser than 78; Group IV (combined: brachycephaly+plagiocephaly), infants whose cephalic index was greater than 89 and CVAI was greater% 3.5. Infants were distributed into 2 groups according to age at diagnosis: Group A, infants aged below 4 months of age and Group B, infants aged 4 months and older.

Statistical analyses were performed using IBM® SPSS® Statistics (version 21.0). Student t-test or Mann Whitney U test were used to compare variables between cohorts. p<0.05 was accepted as statistically significant.

RESULTS

A total of 158 infants were included in the study. Demographics and basic evaluations regarding perinatal and neonatal examinations are detailed in Table 1. A 3D scanner was used to analyze pre-treatment and post-treatment anthropometric measurements of 93 boys (58.7%) and 65 girls (41.3%). Infants were diagnosed as follows: plagiocephaly in 78 (49.3%) infants, combined (plagiocephaly+brachycephaly) in 57 (36.1%), brachycephaly in 16 (10.1%), and scaphocephaly in 7 (4.4%) infants.

In Group I, mean pre-treatment DD results changed from 10.32 mm [±2.74 (range 6–20)] to 7.83 [±3.45 mm (range, 1–19)] after treatment. Pre-treatment CVAI values in this group changed from

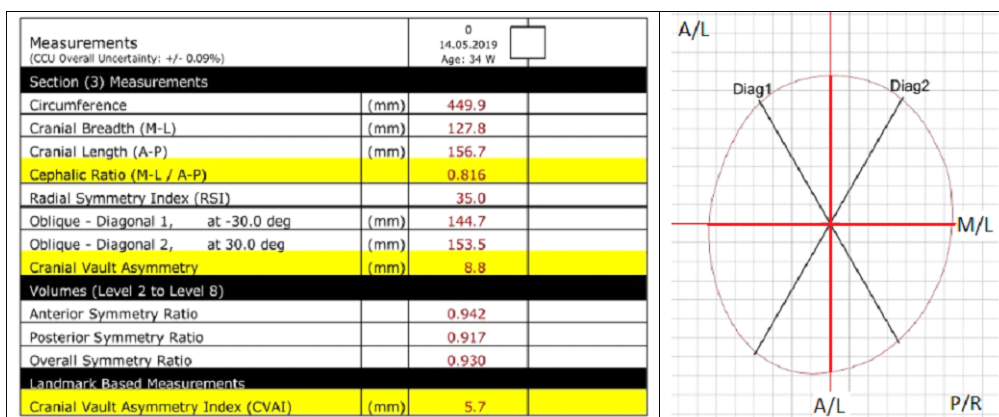


Figure 1. Diagonal diameters (Diag 1 and Diag 2) are measured in millimeters at either 30 degrees (whichever is greater) from the center of the nose to the outer edge of the eyebrow. Glabella and opisthocranion landmarks were used for circumferential measurements and cranial length, which is AP, and right and left ear lobe landmarks were used to measure cranial width, which is ML.

Diagonal Difference (DD)=Diagonal 1-Diagonal 2

Cephalic Ratio (CR)=(M-L/A-P) X 100

Cranial Vault Asymmetry Index (CVAI)=(Diagonal 1 - Diagonal 2)/(Diagonal 1 or Diagonal 2) X100

7.5% [$\pm 1.98\%$ (range 3.8–14.92)] to 5.5% [$\pm 2.45\%$ (1.05–14.25)] in the post-treatment period. Both these differences were statistically significant ($P_{DD}=0.0001$ and $P_{CVAI}=0.0001$) (Table 2).

The mean pre-treatment and post-treatment values of Group II and Group III were evaluated and no statistically significant differences were found (Table 2).

In combined group, Group IV, pre-treatment mean DD values were found 9.38 mm [± 2.67 (range 5–14)], whereas post-treatment values decreased to 6.94 mm [± 3 (range 1–12)]. The pre-treatment

CVAI values were recorded as 6.9% [$\pm 1.99\%$ (range 3.56–11.37)] and post-treatment CVAI values were recorded as 4.9% [$\pm 2.08\%$ (range 0.5–9.08)]. All these changes were statistically significant ($P_{DD}=0.0001$, $P_{CVAI}=0.0001$) (Table 2).

In plagiocephaly deformation types, difference between pre-and post-treatment CVAI and DD levels were statistically significant in both groups of diagnostic age, Group A and Group B. However, in combined deformation types, the regression of CVAI and DD levels were not statistically significant for Group B infants (Table 3). In combined deformation types, absolute change in both DD and CVAI levels after treatment were significantly different at Group A compared to Group B infants (Table 4). [DD (-3.4 \pm 2.6 versus -1.25 \pm 1.45, $p=0.001$); CVAI (-2.7 \pm 1.99 versus -1.14 \pm 0.99, $p=0.0001$)] (Table 4). In plagiocephaly group, although it is not statistically significant, the regression rates at mean DD and CVAI levels after treatment of age <4 months infants were better than the age ≥ 4 months infants, [(-3.03 \pm 2.61 versus -2.04 \pm 2.83, $p=0.209$); CVAI (-2.29 \pm 1.83 versus -1.73 \pm 1.7, $p=0.213$)] (Table 4).

DISCUSSION

Our study analyses the impact of conventional treatment (repositioning) on the management of skull deformities. We found that when started at infants <4 months old they responded better to treatment compared to the infants ≥ 4 months old. This was shown with statistically significant better reduction in DD and CVAI values after treatment in the former group. In general, our results indicate that the repositioning treatment efficacy is related to the age at onset of treatment, and the outcome is significantly improved when the treatment is started before 4 months of age.

Table 1. Demographic characteristics of 158 infants Means \pm SDs or n (%)

Gender	
Males	93 (58.7%)
Female	65 (41.3%)
Birth age (gestational week)	
Mature (born at ≥ 38)	128 (81%)
Premature (born at <38)	30 (19%)
Method of delivery	
Vaginal	49 (31%)
Caesarian-section	109 (69%)
Vacuum assisted delivery	
	3 (2%)
Presentation at delivery	
Occiput anterior	99 (63%)
Occiput posterior	32 (20%)
Breech	27 (17%)
Incubator follow up after delivery	
	19 (12%)
Maternal age (year)	
	26.4 \pm 3.2
Primiparous mother	
	37 (23.4%)
Twin sibling	
	25 (15.8%)

Table 2. Pre-treatment and post-treatment values of plagiocephaly, brachycephaly, scaphocephaly and combined deformities

	DD, mm			CVAI, %			CR, %		
	onset of treatment	end of treatment	p	onset of treatment	end of treatment	p	onset of treatment	end of treatment	p
Plagiocephaly	10.32 \pm 2.74	7.83 \pm 3.45	0.001	7.5 \pm 1.98	5.5 \pm 2.45	0.001	83.8 \pm 3.02	85.16 \pm 3.1	0.006
Brachycephaly	2.37 \pm 1.25	2.62 \pm 1.66	0.635	1.63 \pm 0.79	1.86 \pm 1.32	0.558	93.6 \pm 1.9	92.3 \pm 3.1	0.160
Scaphocephaly	3.42 \pm 1.39	2.42 \pm 1.71	0.256	2.17 \pm 0.82	1.79 \pm 1.11	0.502	74.3 \pm 2	76.6 \pm 3.5	0.070
Combined	9.38 \pm 2.67	6.94 \pm 3	0.001	6.9 \pm 1.99	4.9 \pm 2.08	0.001	92.3 \pm 2.33	92.1 \pm 2.63	0.680

Combined, brachycephaly+plagiocephaly; CR, cranial ratio; CVAI, cranial vault asymmetry index; DD, diagonal difference; p, p-value.

Table 3. Difference between pre-and post-treatment CVAI and DD levels in groups of diagnostic age

	DD, mm			CVAI, %		
	onset of treatment	end of treatment	p	onset of treatment	end of treatment	p
Plagiocephaly						
<4 months	10.67 \pm 2.51	7.63 \pm 3.28	0.0001	7.78 \pm 1.93	5.49 \pm 2.31	0.0001
≥ 4 months	9.91 \pm 2.87	7.87 \pm 3.83	0.042	7.18 \pm 2.18	5.44 \pm 2.72	0.018
Combined						
<4 months	10 \pm 2.72	6.63 \pm 3.14	0.0001	7.44 \pm 2.03	4.74 \pm 2.25	0.0001
≥ 4 months	8.54 \pm 2.39	6.94 \pm 3	0.104	6.19 \pm 1.72	5.05 \pm 1.86	0.099
All patients						
<4 months	10.41 \pm 2.6	7.24 \pm 3.24	0.0001	7.65 \pm 1.97	5.20 \pm 2.3	0.0001
≥ 4 months	9.22 \pm 2.7	7.58 \pm 3.34	0.009	6.68 \pm 2	5.25 \pm 2.31	0.001

Combined, brachycephaly+plagiocephaly; CVAI, cranial vault asymmetry index; DD, diagonal difference; p, p-value.

Table 4. Absolute change in both DD and CVAI levels after treatment in age groups

Age (onset of treatment)	Combined			Plagiocephaly			Total		
	<4 months	≥4 months	p	<4 months	≥4 months	p	<4 months	≥4 months	p
Duration of treatment (days)	66.2±7.4	68.5±14.2	0.477	65.2±8.2	64.9±6.9	0.915	66.7±11.2	65.5±7.9	0.524
CVAI, %									
Absolute change	(-) 2.7±1.99	(-) 1.14±0.99	0.0001	(-) 2.29±1.83	(-) 1.73±1.7	0.213	(-) 2.4±1.89	(-) 1.4±1.41	0.0006
DD, mm									
Absolute change	(-) 3.4±2.6	(-) 1.25±1.45	0.001	(-) 3.03±2.61	(-) 2.04±2.83	0.209	(-) 3.2±2.58	(-) 1.6±2.26	0.0006

Combined; brachycephaly+plagiocephaly, CVAI; cranial vault asymmetry index, DD; diagonal difference, p; p-value

The main purpose of our study is to analyze the efficacy of early repositioning treatment using photogrammetric methods. In this study of 158 infants, we observed that, in the group of infants diagnosed and treated before 4 months of age, mean pre-treatment DD of 10.4 mm and CVAI of 7.65% improved to mean DD of 7.24 mm and CVAI of 5.2% after treatment. For the group of infants diagnosed and treated after the age of 4 months, mean pre-treatment DD of 9.2 mm and CVAI of 6.68% improved to mean DD of 7.58 mm and CVAI of 5.25% at the post-treatment period. Comparison of improvements at DD and CVAI measurements in both groups revealed that the significance of improvement was much more prominent in the treatment Group A, which are similar to those previously reported in literature. Shweikeh et al. reviewed 15 articles in literature and investigated the efficacy of current skull deformity management guidelines. They concluded that parents should be informed as early as possible about positional skull deformity (PSD) and that the education by means of close surveillance is the center of prevention and management of this disorder (21).

Craniofacial measurements are quite important in the diagnosis and evaluation of these patients (22). Previous studies investigated various techniques and skull shape measurements for the diagnosis and follow-up of PSD, however there is no consensus on a practical clinical method to measure the intensity and the change of deformity (23). Radiologic diagnostic techniques are barely helpful in these patients, and although plain radiographs and computerized tomography (CT) scans were performed in the past for these patients, these are not recommended as routine diagnostic tools for patient evaluation. The CT scans are not preferred for long-term follow-up in infants and children since the patient is exposed to high dose radiation, and it requires sedation to immobilize the patient to obtain optimum images. However, CT may be preferred in the differential diagnosis between deformational disorders and craniosynostosis, if there is suspicion after clinical evaluation (24). Nevertheless, 3D measurement devices provide non-invasive, effective, reliable and low-cost evaluation of skull asymmetries. Furthermore, this technique is compatible with the gold standard 3D CT technique in the diagnosis and follow-up, and may even provide more detailed and accurate shape information. (25).

Neglected cranial deformations may lead to negative outcomes in a child's future life. Previous studies reported association of

skull deformities and abnormal language development, visual-perception deficits, and delayed intellectual and motor development skills (13, 26, 27). Therefore, children at school age usually require supportive education and speech therapy, physical therapy, and work education. These patients are also prone to astigmatism. Thus, it is common for these children to wear prescription glasses and they need to wear proper protective helmets to do some sport activities like snowboard, bicycle riding (28). Miller et al. reported that deformational infants with plagiocephaly consist of a high risk group for developmental difficulties at school age (29). Recently, a study using Bartley's developmental scale III on 6-month-old plagiocephaly infants showed that these babies are at high risk for delayed neurologic development (15). Steinbok et al. reported that 33% of infants with skull deformities needed educational support and 14% were located at special needs class (30). Thus, these patients need to be diagnosed early by neonatologists and general pediatricians not only to prevent aesthetic deformations but to prevent psychomotor developmental retardation, as well.

CONCLUSION

Recently, PSD prevalence has been on the rise. It is important that the pediatricians are able to evaluate the severity of the problem and establish an early diagnosis in these cases. Improvement rates of the asymmetry decrease with age due to decreasing skull enlargement rate by age. Early diagnosis and close follow-up are quite important so that the infants with this condition may benefit from conservative management.

Informed Consent: Informed consent was obtained from all of the families of individual participants included in the study

Compliance with Ethical Standards: This study is a retrospective data analysis. However, All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

Peer-review: Externally peer-reviewed.

Author Contributions: Concept - AO,SI,SC; Design - AO,SI,SC; Supervision - AO; Fundings - AO; Materials - AO; Data Collection and/or Processing - AO,SI,SC; Analysis and/or Interpretation - AO,SI,SC; Literature Search - SI,SC; Writing Manuscript - SI,SC; Critical Review - AO

Conflict of Interest: No conflict of interest was declared by the authors.

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