Assessing the Cervical Range of Motion in Infants With Positional Plagiocephaly

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Purpose: To determine if infants with positional plagiocephaly have limitations of active and passive cervical range of motion measured with simple and reliable methods.

Methods: The examiners assessed bilateral active and passive cervical rotations and passive cervical lateral flexion. Cervical assessment was performed twice by 2 different physicians to assess intertester reliability. To assess intratester reliability the first investigator performed a second examination 48 hours after the first one.

Results: One-hundred nine subjects were analyzed; 70.7% of the sample had head positional preference on the right, while 29.3% had head positional preference on the left (χ^2 35.52, $P<0.001$). Cervical rotations and lateral flexion showed reliable levels of agreement for intra and intertester reliability.

Conclusions: The most limited range of motion in infants with positional plagiocephaly was cervical active rotation which affected more than 90% of patients. Passive cervical rotations and lateral flexion were limited in more than 60% of patients.

Key Words: Cervical assessment, cervical range of motion, positional plagiocephaly

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Positional or deformational plagiocephaly (PP) is the most common infant cranial asymmetry characterized by an asymmetrical flattening of the skull and it is possibly associated with an increased risk of developmental delays in toddlers.[1] Its incidence has significantly increased over the past few decades[2] following the recommendation of the American Academy of Pediatrics to place infants in a supine sleeping position to avoid Sudden Infant Death Syndrome.[3] Particularly, the incidence in infants at 7 to 12 weeks of age was estimated to be 46.6%[4] and the prevalence varies between 6.1% and 13.0% at birth, 16.0% and 22.1% at age 6 to 7 weeks, 19.7% at age 4 months, 9.2% at age 8 months, and 6.8% at age 12 months.[5]

The risk of PP is increased for infants who required assisted deliveries, are first-born children, are male, have high cumulative exposure to supine positioning, and have neck problems such as infant difficulty turning head, decreased cervical rotation, limited passive cervical rotation, and limited active cervical rotation. The pathophysiologic mechanisms underlying this condition are poorly understood.[6–8]

Abnormal prenatal and/or postnatal external forces acting on the malleable and growing cranium in the first months of life may be a cause unilateral occipital flattening and, in severe patients, even misalignment of the ears on the axial plane and asymmetry of the face. A strong association between congenital muscular torticollis (CMT), positional preference, and the onset of the PP has been suggested.[9] Congenital muscular torticollis is a congenital musculoskeletal disorder characterized by unilateral shortening of the sternocleidomastoid muscle usually presenting during infancy.[10] The presence of muscular damage or an imbalance in the neck after birth might, in fact, determine a "position of comfort" for the head, which could result in persistent rotation, thereby causing a flatten of the head. Alternatively, the presence of PP at birth could induce the development of a postnatal torticollis.[11] To date, uncertainty still exists about the prevalence of cervical range of motion (ROM) limitations in infants with PP.

Measuring cervical ROM in patients with PP should be an essential part of the physical examination to identify factors, such as head rotation preferences to 1 side or decreased active cervical ROM, which might contribute to the development of PP.[12] It is also essential to assess the severity of PP, to provide appropriate nonsurgical or surgical therapeutic interventions as well as to verify their effectiveness.

We hypothesize that a high percentage of infants with PP might have both active and passive cervical ROM restrictions, and that these limitations could be detected by simple and reliable maneuvers during a standard physical examination.

The objective of the present research was, therefore, to determine if infants diagnosed with PP have limitations of active and passive cervical range of motion measured with simple and reliable methods.

METHODS

This study was designed as an uncontrolled, prospective patient series. From January 2011 to June 2013, all infants who referred to our Physical Medicine and Rehabilitation outpatient service for the assessment of cranial asymmetry were screened for eligibility.

Patients of both sex aged between 0 and 18 months with a diagnosis of PP were included in the present study. We excluded infants with a diagnosis of synostotic plagiocephaly, CMT, and suspected dysmorphism of the spine.[13]

All parents agreed to participate in the study and signed an informed consent form in accordance with the Declaration of Helsinki.

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Medical history of patients was obtained from parents and a standard physical examination was performed on each patient before the cervical ROM measurements were taken. The degree of skull asymmetry was evaluated according to Argenta classification.[14,15] It distinguishes 5 types of PP, based on the severity of the asymmetry of the skull, ear position and face as follows: restricted to the back of the skull, adds malposition of the affected ear, adds forehead deformity, adds facial deformity, and adds abnormal vertical growth.[14]

The assessment of active and passive cervical rotations and passive lateral flexion was performed twice on every patient by 2 different physicians (examiner 1 and examiner 2), to assess intertester reliability. The assessors performed their evaluation without being made aware of one another and another's assessment 1 to 24 hours apart.

Furthermore, examiner 1 performed a second examination 48 hours after the first one to assess intratester reliability.

Those who participated in the study did not receive any form of treatment between the first and second examinations.

The infant was placed supine on an examination table with his shoulders in a stabilized position to observe the spontaneous attitude of the head. The assessment of bilateral active and passive cervical rotations was performed taking as reference the baby's chin and the shoulder ipsilateral to the rotation.

The infant was stimulated to perform an active rotation of the head toward both sides using a colored object and a sound or the maternal voice and face. Once the infant reached the maximum degree of active rotation, the physician softly forced the head rotation to obtain the full passive ROM. Moreover, the presence of stress signs in baby's facial expression, crying, and marked opposition to the forced rotation were considered to determine the end of rotation ROM.[16]

Both active and passive cervical rotation ROM were categorized as follows (Fig. 1): full ROM, chin goes beyond the ipsilateral shoulder; mild limitation, chin reaches the ipsilateral shoulder without going beyond it; moderate limitation, chin goes beyond half clavicle but does not reach the should and severe limitation, chin does not go beyond half clavicle.

To assess the passive cervical lateral flexion, the infant's ear was approached to the ipsilateral shoulder, by placing examiner's hand at the level of the occipital bone of the child, with the contralateral shoulder held stable by the examiner. Cervical lateral flexion was categorized as follows (Fig. 2): full ROM, the ear reaches the shoulder; moderate limitation, the ear goes beyond 45° of inclination but does not reach the shoulder; severe limitation, the ear does not go beyond 45° of inclination.

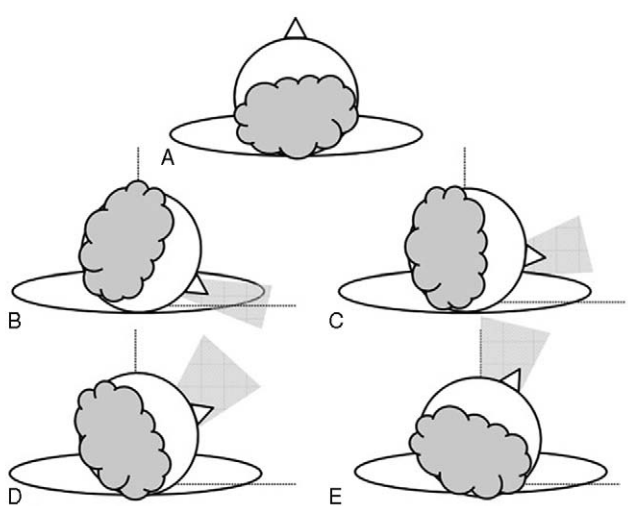

FIGURE 1. Active and passive cervical rotation ROM. (A) Neutral position. (B) Full ROM. (C) Mild limitation. (D) Moderate limitation. (E) Severe limitation. ROM, range of motion.

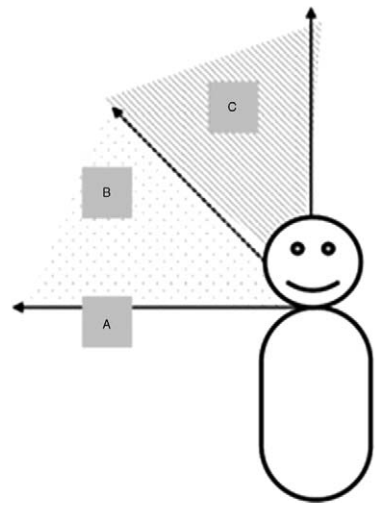

FIGURE 2. Passive cervical lateral flexion. (A) Full range of motion. (B) Moderate limitation. (C) Severe limitation.

The starting and ending positions of the cervical spine were noted for each measurement maintaining the cervical neutral throughout the measurements.

Each motion, passive and active cervical rotation and cervical lateral flexion, was repeated 3 times, and the best measurements (ie, that with the greatest ROM achieved) were thus analyzed.

Statistical Analysis

All the data were analyzed by an individual researcher. After verifying the normal distribution of the analyzed measurements using a Kolmogorov–Smirnov test to verify that the variables were normally distributed, parametric tests were used.

Cohen Kappa was used for the assessment of both intra and intertester reliability of categorical data. Cohen Kappa takes on values between -1 and 1, with 1 indicating perfect agreement between the testers, -1 indicating perfect disagreement, and 0 indicating that any agreement is completely due to chance. It has been suggested that Kappa values above 80% demonstrate excellent agreement, values from 60% to 80% substantial levels of agreement, values from 40% to 60% moderate agreement, and values below 40% poor to fair agreement.[17]

The χ^2 test was used to analyze the prevalence of right or left head positional preference and the side prevalence of the active and passive cervical rotation and the passive lateral flexion. The severity of ROM limitations was assessed by means of χ^2 test.

Correlation analysis was performed to investigate the relationships of Argenta classification with respect to age, sex, and cervical ROM limitations. Correlation analysis was also performed between the degree of cervical imbalance and age at examination.

Correlation coefficients were interpreted as follows: 0.00 to 0.19: very weak correlation; 0.20 to 0.39: weak correlation; 0.40 to 0.69: moderate correlation; 0.70 to 0.89: strong correlation; and 0.90 to 1: very strong correlation.[18]

Statistical Package for Social Sciences (SPSS) version 18 was used for calculations. Differences with P values ≤ 0.05 were considered to be statistically significant, and all results were expressed with a 95% confidence interval.

TABLE 1. Baseline Characteristics of Patients

Number of patients	109
Age (mo)	4.6 ± 2.1 SD (range: 1.5–13)
Sex	70 M, 39 F
Head positional preference	77 R, 32 L
Position assumed during sleep	77 R, 32 L
	21 asymmetry on occipital side
	31 malposition of ipsilateral ear
Argenta classification	30 ipsilateral frontal bone protrusion
	22 asymmetry ipsilateral facial
	5 temporal bulging or abnormal vertical growth
	14% (N = 15) premature
	32% (N = 34) first gravidity
Risk Factors	5.5% (N = 6) breech presentation
	46% (N = 50) assisted delivery
	7.3% (N = 8) Oligoidramnios

F, female; L, left; M, male; R, right; SD, standard deviation.

Sample Size

Power calculations were based on a correlation value hypothesis H1 of −0.46. Assuming a Cohen Kappa coefficient value of 0.6, a 2-tailed α value of 0.05 (sensitivity 95%) and a β value of 0.05 (study power: 95%), we determined that at least 70 subjects were required (G Power3 power analysis program).

RESULTS

One-hundred nine subjects (70 males and 39 females), with a mean age of 4.6 months (standard deviation [SD]: 2.1 months; range [R]: 1.5–13 months) met the inclusion criteria. The baseline characteristics of the sample are reported in Table 1.

Kappa statistics and 95% CIs for the cervical active and passive ROM are presented in Table 2. Intertester reliability was 0.80 for active cervical rotation, 0.83 for passive cervical rotation, and 0.49 for passive lateral flexion, indicating excellent and moderate levels of agreement. Intratester reliability was 0.72 for active cervical rotation, 0.73 for passive cervical rotation, and 0.41 for passive lateral flexion, indicating excellent and moderate levels of agreement.

As far as the direction of detected ROM limitations, in all patients the active and passive cervical rotations were limited contralateral, whereas passive cervical lateral flexion was limited ipsilateral to the side of head positional preference.

The 70.7% (N = 77) of sample had head positional preference on the right, while the 29.3% (N = 32) had head positional preference on the left (χ^2 35.52, $P < 0.001$) (Table 3). Among patients with head positional preference on the right, the 93% (N = 71) had a limitation of active cervical rotation on the left (χ^2 120.42, $P < 0.001$), the 60% (N = 46) had a limitation of passive cervical rotation on the left side (χ^2 77.69, $P < 0.001$), the 62% (n = 48) had a significant limitation of passive lateral flexion on the right side ($\chi^2 = 77.69$, $P < 0.001$). Among patients with head positional preference on the left, the 94% (N = 30) had a limitation of active cervical rotation on the right side (χ^2 120.42, $P < 0.001$); the 75% (N = 24) had a limitation of passive cervical rotation on the right side (χ^2 35.26, $P < 0.001$), the 62.5% (N = 20) had a limitation of passive tilt on the left side (χ^2 26.26, $P < 0.001$).

Correlation analysis showed a not statistically significant relationship between cranial asymmetry and age ($r = 0.102$, $P = 0.40$) and sex ($r = 0.14$, $P = 0.22$). We also found a not statistically significant relationship between the degree of skull asymmetry and active cervical rotation ($r = 0.107$, $P = 0.268$), a significant relationship between degree of skull asymmetry and passive cervical rotation ($r = 0.369$, $P = 0.000$), and a significant relationship between degree of skull asymmetry lateral flexion ($r = 0.319$, $P = 0.000$).

Correlation analysis showed a not statistically significant relationship between the degree of cervical imbalance and age at examination (Table 4).

DISCUSSION

The main results of the present study were that in patients with PP, active and passive cervical ROM limitations are common, being detectable in up to 93% of subjects. As far as we know, this is the first study to report such a high prevalence of cervical ROM limitation in PP. The most limited ROM was cervical active rotation which was decreased in >90% of patients. Moreover, passive rotations and lateral flexion were limited in more than 60% of patients.

Furthermore, the direction of detected ROM limitations was in all patients contra laterally to the side of head positional preference for the rotations, and ipsilateral to the side of head positional preference for lateral flexion. The cervical spine is surrounded by a complex arrangement of muscles that contribute to static and dynamic control of the head and neck. Probably, the head positional preference causes a different morphological evolution between the muscle layers that encapsulate the spine producing a variation in their mechanical effect on the spine.

As far as factors influencing the magnitude of cranial asymmetry, we found a significant correlation between the degree of skull asymmetry and both passive cervical rotation and lateral flexion, but not with active cervical rotation. Age and sex do not appear to influence the cranial asymmetry.

Moreover, although the presence or absence of congenital or acquired torticollis is an important factor that affects gross motor development in infants with plagiocephaly,[19] we found not significant relationship between the degree of cervical imbalance and age at examination. Therefore, on the basis of our data, we cannot definitively determine if this muscular imbalance might represent the cause or the effect of PP; however, we can confirm the data from the previous literature regarding the strong association between these 2 conditions.[5]

Future studies are needed to determine if early evaluations of cervical ROM limitations and physiotherapy, along with tummy time and general positioning rules, could lead to a reduction in the rate of progression of PP over time, as well as to a reduction in the use of cranial orthoses or surgery.[5]

TABLE 2. Intra and Tester-Operator Reliability Coefficients for Cervical Range of Motion Assessment

	Intratester Agreement (Kappa)	SE of Kappa	Intertester Agreement (Kappa)	SE of Kappa
Active cervical rotation	0.72 (0.51–0.93)	0.10	0.80 (0.59–1)	0.09
Passive cervical rotation	0.73 (0.49–0.97)	0.13	0.83 (0.66–1)	0.08
Passive lateral flexion	0.41 (0.35–0.77)	0.18	0.49 (0.11–0.77)	0.07

Mean values of Kappa statistics (95% CI) and standard errors (SE) are represented for each tested movement.

TABLE 3. Cervical ROM Limitations in Patients With Positional Plagicephaly

	Right Head Positional Preference (N = 77)			Left Head Positional Preference (N = 32)		
Cervical ROM	Side	χ^2	*P* Value	Side	χ^2	*P* Value
Active rotation	Left	120.42	<0.001	Right		0.001
Mild (N)	44	43.14	0.001	16	15.60	0.001
Moderate (N)	19			12		
Severe (N)	8			2		
Passive rotation	Left	77.69	<0.001	Right		0.001
Mild (N)	33	15.69	0.001	21	47.62	0.001
Moderate (N)	13			1		
Severe (N)	–			1		
Pasive lateral flexion	Right	77.69	<0.001	Left		0.001
Mild (N)	39	35.04	0.001	16	12.10	0.001
Moderate (N)	9			4		
Severe (N)	–			–		

N, number.

Our data contrasts with a previous study conducted by Rogers et al[20] which measured cranial asymmetry using an anthropometric spreading caliper. This study found statistically significant negative correlation between age and head rotational asymmetry ($r = -0.200$; $P = 0.004$) and a significant positive correlation between cranial asymmetry and head rotational asymmetry ($r = 0.142$; $P = 0.043$). However, according to the interpretation of the Pearson correlation coefficient,[18] they showed weak and very weak correlations.

We considered the cervical ROM examination with simple maneuvers that classified the neck limitations as mild, moderate, and severe, in terms of anatomical landmarks as described above.

Health care professionals use a variety of methods to evaluate cervical spine ROM, but in order for a measure of ROM to be clinically useful, it must be first reliable; this refers to the consistency of a measurement across time, patients, or observers.[21] In our study the active and passive cervical rotations demonstrated the highest level of intra and intertester reliability. In particular, for the active and passive cervical rotations, substantial and excellent levels of agreement were found for intra and intertester reliability, respectively. Moreover, for passive side bending moderate agreement was found for both intra and intertester reliability.

Cervical passive ROM rotation is considered to be normal at 110°, while in lateral flexion the variation is 40° to 90° in children with congenital muscular torticollis.[22,23] However, the references of ROM values in healthy infants are insufficient. Indeed, only Ohman and Beckung[24] evaluated in 38 healthy infants the cervical passive rotation and lateral flexion, using an arthrodial protractor. The infants were laying supine on the examination table with the shoulders held stable and the examiner supported the head and neck in the neutral position over the edge of the examination table. Neck rotation ranged from 100° to 120° and lateral flexion from 65° to 75° in infants aged 2 to 10 months. Cheng et al[22] assessed the cervical rotation on 821 consecutive patients with congenital muscular torticollis, classified as palpable sternomastoid tumor, muscular torticollis, and postural torticollis. The reproducibility of rotation measurements had an intertester reliability correlation coefficient of 0.71. Likewise, Klackenberg et al examined the lateral flexion in 23 infants with CMT by a goniometer for rotation and a protractor for lateral flexion. This method was found to have high intratester reliability with the interclass correlation coefficient reported as 0.94 to 0.98.[24]

TABLE 4. Correlation Between the Degree of Cervical Imbalance and Age of Subjects

	Right Head Positional Preference		Left Head Positional Preference		
	Age	*P* Value		Age	*P* Value
Left active rotation	−0.15	0.12	Right active rotation	−0.26	0.07
Left passive rotation	−0.11	0.27	Right passive rotation	−0.31	0.03
Right lateral flexion	0.07	0.43	Left lateral flexion	−0.15	0.28

The main limitation of the present study was the absence of a control group. Moreover, we did not perform a goniometric assessment of cervical ROMs to compare our clinical evaluation with an instrumental gold standard. Finally, follow-up examinations could be interesting to understand how these patients evolve over time.

Infants with PP are likely to have cervical ROM limitations, even in the absence of any structural neck musculature damage. From this point of view, assessing active and passive cervical ROM should be a part of the routine physical examination in patients with PP. Cervical ROM limitation may be detected by means of easy and reliable maneuvers, such as those described in the present paper.

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The Effect of Cigarette Smoking on the Healing of Extraction Sockets: An Immunohistochemical Study: ERRATUM

In the article that appeared on page e397 of the July 2014 issue of the *Journal of Craniofacial Surgery*, an author's name did not appear. The full list of authors and affiliations is as follows:

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