Pediatric Neurology xxx (2016) 1–5

Contents lists available at ScienceDirect

Pediatric Neurology

journal homepage: www.elsevier.com/locate/pnu

Original Article

Transient Motor Asymmetry Among Infants With Congenital Torticollis—Description, Characterization, and Results of Follow-Up

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ABSTRACT

AIM: The purpose of this study was to assess the prevalence of transient functional motor asymmetry in infants with congenital postural torticollis. **METHODS:** This was a retrospective review of the medical records of infants with postural torticollis. We analyzed epidemiological, obstetric, perinatal data, physical therapy, physician assessments, and clinical follow-up for two years after diagnosis. **RESULTS:** Of 173 children, 44 (25.4%, 95% confidence interval = 19.5 to 32.4) demonstrated functional asymmetry. Demographic and obstetrical data did not differ between the asymmetry/nonasymmetry groups. Delayed motor development ($P = 0.01$) and plagiocephaly ($P = 0.032$) were more common in infants with motor asymmetry. No difference was observed in the frequency of referral for further neurological diagnosis between the group with functional asymmetry and that without asymmetry. Among the 44 patients with functional asymmetry, 78% depicted no evidence of torticollis by the age of two years, and the motor asymmetry had disappeared in 82%. **CONCLUSION:** Benign, transient functional motor asymmetry occurred in a quarter of infants with congenital postural torticollis. Transient motor delay was also significantly more common in the asymmetry group. In most cases, motor asymmetry and motor delay disappeared by the age of two years. Plagiocephaly was more common in the asymmetry group. Clinician awareness of this transient asymmetry may have avoided unnecessary diagnostic tests in these infants.

Keywords: congenital torticollis, infants, motor, motor asymmetry, transient

Pediatr Neurol 2016; ■: 1-5

Congenital torticollis has an incidence of 0.3% to 1.35% among healthy newborns,[1] with a prevalence of 8% to 12% among infants younger than six months.[2,3] Torticollis is commonly recognized within the first several weeks of life, although it is evident at birth in 10% to 16%.[4] Some infants have disorders such as sternomastoid (SCM) trauma or swelling, fracture of the clavicle, or oligohydramnios, and torticollis may be associated with other skeletal conditions such as developmental dysplasia of the hip. Most children have no identifiable etiology, and these are considered to be idiopathic or "postural."[2] In general, two groups of congenital torticollis are recognized: (1) congenital muscular torticollis (CMT) attributed to SCM involvement and (2) postural (idiopathic) torticollis.[3]

Following the 1992 recommendation of the American Academy of Pediatrics for infants to sleep on supine or sideward position to reduce the risk of sudden infant death syndrome, an increased incidence of plagiocephaly and torticollis was observed.[5-7] Infants who do not spend enough time in prone position have slower gross motor development than infants who spend a significant part of the day lying prone. Regarding postural torticollis, slightly more than 50% of infants still exhibit asymmetrical head posture six to eight months after the diagnosis of torticollis, and another 25% exhibit varying degrees of torticollis after 24 to 36 months.[3]

Conflicts of interest: None.

Article History:
Received November 28, 2015; Accepted in final form February 6, 2016

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0887-8994/$ – see front matter © 2016 Elsevier Inc. All rights reserved.
http://dx.doi.org/10.1016/j.pediatrneurol.2016.02.005

Data on long-term motor development of infants with torticollis are rather scarce, especially regarding postural (idiopathic) torticollis. As for CMT a study on 82 infants revealed that at least until age ten months, these infants had significantly delayed motor milestones compared with the control group, the amount of time spent on supine being a significant risk factor for motor delay.[4] However, long-term motor development of children with congenital torticollis appears to be good.[8]

Among infants, overt hand preference and functional motor asymmetry between the two sides of the body may raise a suspicion of central nervous system pathology, leading to diagnostic procedures seeking an etiology. However, clinical experience suggests that among infants with congenital torticollis, including those with postural torticollis, some infants exhibit transient motor asymmetry that disappears as the severity of the torticollis decreases (Watemberg N., personal observation).

The aims of our study were to establish the incidence of this transient motor asymmetry among infants with congenital postural torticollis and to determine the natural course of this asymmetry.

Materials and Methods

We reviewed the records of all consecutive infants with congenital torticollis of the postural (idiopathic) type referred to Meir Hospital's Child Development Center. Patients' records were handwritten, but computerized information on clinical diagnosis, initial evaluations, and discharge summaries was available. Cases were identified from these computerized data searching for the term "torticollis." Authorization from Meir Medical Center's Institutional Board Review was obtained before performing the study.

Functional motor asymmetry was considered to be present in infants who depicted reduced volitional motor activity of one or both limbs on one side of their body but had no evidence of weakness or of spasticity of those extremities.

Data obtained included demographics, pregnancy and delivery information, infant's development, and health status. Regarding torticollis, we reviewed the age at diagnosis, the clinical assessments performed, diagnostic studies obtained, the kind of therapy received, and the infant's gross motor developmental level at the age of two years, as assessed by our physical therapy staff using the Alberta Infantile Motor Scale.[9]

During the three-year period, each infant with postural torticollis was assessed and treated by one of three different physical therapists working in our child development center. The finding of motor asymmetry was recorded by the therapist as part of their routine evaluation. If a neurological evaluation was deemed necessary, the infant was referred to one of the center's three neurologists.

The direction of the head tilt (torticollis) was determined according to the position of the chin. Hence, a chin pointing to the right shoulder was diagnosed as right torticollis, whereas a "chin to the left" was regarded to as left torticollis.

The presence of motor asymmetry was described in the physical therapy's initial evaluation and/or during the follow-up period. Asymmetry was reported when (1) a clear difference was detected in spontaneous/purposeful movements of the limbs between both sides of the body and (2) apparent upper limb preference in older infants.

Exclusion criteria included torticollis of muscular origin, prematurity, known neurological conditions such as brain injury and epilepsy, no evidence of torticollis on the initial physical therapy evaluation, and acquired torticollis.

Statistical analysis

Data are presented as numbers and percentage for nominal variables, and mean and standard deviation for continuous parameters. Differences between patients with vs without asymmetry and parameters such as sex, side, history, type of delivery, and so forth were tested using chi-square tests (χ^2) or Fisher exact test, each when appropriate. Differences of continuous variables (such as age, duration of treatment, age of diagnosis, etc.) between with vs without asymmetry were tested using t test. Difference was considered statistical when $P < 0.05$. Statistical analyses were performed using SPSS—21 software.

Results

A total of 275 infants with suspected torticollis were referred to our center between 2008 and 2010. During the same period, a total of 2136 infants were evaluated by our physical therapy staff. Sixty-three of the 275 torticollis infants were excluded from the study as torticollis was not found during the initial examination or the sign was deemed to be acquired, such as infants with gastroesophageal reflux or those with muscle spasticity.

Of the 212 remaining children, 36 were excluded due to premature birth, one due to cerebral palsy, one due to epilepsy, and one more due to history of brain hemorrhage. The study population included only infants diagnosed with congenital postural torticollis. All infants were referred by a primary care physician and had undergone an orthopedic evaluation and/or neuroimaging studies as necessary. Those with obvious etiologies such as SCM swelling were excluded from the study.

In all, 173 children were included; 44 (25.4%, 95% confidence interval = 19.5 to 32.4) demonstrated functional asymmetry during physical therapy evaluations. Fourty-nine infants were referred by the therapist for evaluation by a neurologist form the development center, with various combinations of signs: all 36 children with plagiocephaly, 9 of 11 with developmental delay, and the two infants with generalized increased muscle tone. Twenty-two of the 44 infants with motor asymmetry were referred for neurological assessment. Table 1 depicts their clinical and neurological findings.

Physical therapy was implemented at 3.9 ± 2.4 months. In 50.9% of infants, torticollis direction was to the right and to the left in 46.8%. For four infants, there was no documentation regarding the direction of torticollis. In 2.4% (4 of 173) of the children, another musculoskeletal defect (such

TABLE 1.
Clinical Features by Distribution Into Groups With/Without Functional Motor Asymmetry

Variables	Without Asymmetry (N = 129), 74.6%	With Asymmetry (N = 44), 25.4%	*P* Value
Age of diagnosis (mo)	3.6 ± 2.5	3.5 ± 1.7	0.938
Treatment duration (mo)	4.0 ± 4.2	6.6 ± 5.0	**0.001**
Torticollis right	48.8% (63)	56.8% (25)	0.464
Torticollis direction not specified	3.1% (4)		—
Family history of torticollis	14.0% (18)	4.5% (2)	0.108
Shoulder girdle weakness	15.5% (20)	18.2% (8)	0.293
Axial hypotonia	6.2% (8)	11.4% (5)	0.334
Increased muscle tone	1.6% (2)	4.5% (2)	0.122
Delayed motor development	9% (11)	30.2% (13)	**0.01**
Plagiocephaly	27.9% (36)	45.5% (20)	**0.032**

Significant *P* values are depicted in bold characters.

as foot deformity or mandible asymmetry) was present. There were no cases with developmental dysplasia of the hip.

Except for functional motor asymmetry, including asymmetric crawling, 71.7% had normal neurological examination. The remaining infants mostly depicted delayed motor milestones, shoulder hypotonia, and increased limb muscle tone. Of these findings, delayed motor development was significantly more common in infants with functional asymmetry ($P = 0.01$). No signs of spasticity were detected. Occipital plagiocephaly was detected in 32.4% (56 of 173). This finding was significantly more prevalent in the group with functional asymmetry than that among infants without asymmetry (45.5% vs 27.9%, $P = 0.03$). None of the infants with plagiocephaly required surgical intervention.

Infants with functional asymmetry received longer physical therapy treatment than those without asymmetry (6.6 ± 5 months vs 4.0 ± 4.2 months, $P = 0.001$).

Table 2 depicts the main demographic data and obstetric history for both functional asymmetry and no asymmetry groups. No significant differences were detected between the two groups, except for a history of abnormal amniotic fluid volume (either oligo- or polyhydramnios) that was more common in the functional asymmetry group. Nevertheless, the number of cases with abnormal amniotic fluid volume was quite small, in spite of suggesting a statistical difference.

Further etiologic evaluation of torticollis was performed in 70.5% of patients (Table 3). There was no statistical difference between the two groups regarding the proportion of infants undergoing diagnostic procedures or the type of tests performed; imaging (brain and/or spine) studies were rarely obtained. Hip joint ultrasound to exclude hip dysplasia was the most frequently performed procedure for both groups.

TABLE 2.
Demographic and Obstetric Distribution Groups With/Without Functional Asymmetry

Demographic/Obstetric Data	Without Asymmetry (N = 129), 74.6%	With Asymmetry (N = 44), 25.4%	*P* Value
Sex			
Male	54.3% (70)	54.5% (24)	0.974
Female	45.7% (59)	45.5% (20)	
Age of diagnosis (mo)	3.6 ± 2.5	3.5 ± 1.7	0.938
Number of fetuses			
Single	97.7% (126)	95.5% (42)	0.602
Twins	2.3% (3)	4.5% (2)	
Type of delivery			
Spontaneous vaginal delivery	67.4% (87)	70.5% (31)	0.298
Vacuum	7.8% (10)	13.6% (6)	
Caesarean section	24.8% (32)	15.9% (7)	
Head	96.9% (125)	97.7% (43)	1.00
Breech	3.1% (4)	2.3% (1)	
Gestational age (wk)	39.3 ± 1.3	39.3 ± 1.4	0.30
Birth weight (g)	3209.2 ± 518	3113.1 ± 384	0.212
Oligo-/polyhydramnios	0.8% (1)	4.5% (2)	**0.048**
Other pregnancy complications	11.6% (15)	9.1% (4)	0.784
Fetal distress	3.9% (5)	2.3% (1)	1.00

Significant *P* values are depicted in bold characters.

TABLE 3.
Prevalence of Additional Diagnostic Procedures by Asymmetry

Diagnostic Procedure	Without Asymmetry (N = 129), 74.6%	With Asymmetry (N = 44), 25.4%	*P* Value
Brain imaging (all normal)	6.2% (8)	11.4% (5)	0.262
Spine imaging	3.1% (4)	4.5% (2)	0.645
Hip joint ultrasound	59.7% (77)	70.5% (31)	0.203
Ophthalmology evaluation	25.6% (33)	31.8% (14)	0.422
No diagnostic procedures	33.3% (43)	18.2% (8)	0.454

A number of other findings were present among the 44 children who indicated motor asymmetry: the preferred body side was the right one in 75% and the left one in 22.7% (one child depicted asymmetry that changed sides during physical therapy). As mentioned previously, torticollis direction was to the right in 50.9% and to the left in 46.8%. Asymmetric crawling was observed in 43.6% (17 of 39). There was not enough information in the patients' charts concerning their visual field preference.

On examination of the correlation between the direction of torticollis and the direction of asymmetry (Table 4), 100% (25 of 25) of the children with torticollis to the right (i.e., chin pointing to the right) exhibited a preference for the right side of the body. In addition, 100% (10 of 10) of children with left side preference exhibited torticollis to the left. However, 18.2% of infants with left torticollis exhibited preference for the right side of the body, but no child with chin to the right had a left-sided preference ($P = 0.008$).

Long-term follow-up data were available for 120 of the 173 children. At age two years, 78.3% had no evidence of torticollis. Among the other children (21.7%), 3.33% had torticollis of the same severity as at the time of diagnosis, and in 18.3% the torticollis had improved compared to the time of diagnosis. A comparison between infants with and without the asymmetry did not reveal statistical significance regarding the presence or absence of torticollis at age two years.

Follow-up documentation was available for 33 of 44 children with motor asymmetry. Between the ages of one and two years, this asymmetry had disappeared in 22 of the 33 infants (66.6%). Motor asymmetry in 27.2% was of the same severity as at the time of diagnosis, and various degrees of improvement were observed in 6% compared with the time of diagnosis.

Discussion

Clinical experience suggests that a transient, benign motor asymmetry may occur in some infants with congenital postural torticollis. We detected transient motor

TABLE 4.
Differences Between the Direction of Torticollis and Motor Asymmetry in 44 Infants With Functional Asymmetry

Direction of Torticollis	Right Hand Preference	Left Hand Preference
Chin to right	25	0
Chin to left	8	10

asymmetry in one quarter of our 173 patients with congenital postural torticollis.

Among healthy infants without torticollis, the supine head-orientation preference correlates with hand-use preference at the age of 16 and 22 weeks, particularly for right handedness.[10] Visual field preference was also related to supine head-orientation, and a certain degree of asymmetry in hand use was observed.[11] In older children with a history of CMT, an association between ipsilateral chin-pointing and visual field preference was reported, and investigators also raised the possibility of some degree of motor asymmetry during infancy in these patients.[12] Healthy five-month-old infants indicate immediate hand preference for the hand that remains visible when the other hand is out of the field of vision.[13] In our study, we lacked retrospective information on visual field preference in our patients with and without motor asymmetry. However, we believe that visual field preference as a result of restricted head and neck motion likely plays a significant role in the phenomenon of transient motor asymmetry. Our findings suggest that torticollis, by restricting head movement and probably limiting eye–hand contact on one side of the body, may be a determinant factor in developing early hand preference and motor asymmetry in these infants.

Delayed motor development has been described in infants with congenital torticollis. In fact, 34.7% exhibit motor function below the normal range, but by 1 year, this delay decreases to only 9.6%.[14] Nevertheless, at school age the same research group reported a higher prevalence of neurodevelopmental disorders such as attention deficit disorder, developmental coordination disorder, and autism in their original cohort of torticollis patients.[15] Conversely, motor development outcome at preschool age has been described as normal.[16] In our study, 14.5% of all infants exhibited delayed motor development, but this delay was more common among the motor asymmetry group ($P = 0.001$). It is possible that functional asymmetry may be a contributing factor to worsening, even temporarily, of the motor delay described in infants with torticollis. In two thirds of our 44 patients with motor asymmetry the asymmetry had disappeared by the age of two years. Although we lack follow-up information at later ages for the remaining one third, none had been diagnosed with chronic neurological or motor deficit (such as cerebral palsy). Because our child development center is the only referral center for these patients, it would be very likely for these children to have been referred back to us.

Plagiocephaly, a deformity of the skull not associated with synostosis, is often the result of torticollis in infants born with normal head shape. It is usually more prominent in the occipital region, and about 40% of children who develop plagiocephaly were reported to need further educational support in elementary school.[17] During infancy and early childhood, many of these children exhibit abnormal muscle tone and developmental test scores at the lower normal range.[18] Whether there exists a true association between plagiocephaly and later learning disabilities needs further evaluation, as diverse etiologies for plagiocephaly exist. Robinson and Proctor suggested that there is not enough evidence to suggest that plagiocephaly by itself is a risk factor for delayed development in infants.[19] Indeed, the study by Schertz et al.,[14] describing normalization during early childhood of motor development in delayed infants with congenital torticollis, does not address the potential influence of plagiocephaly in their patients. Conversely, 61% of congenital torticollis infants demonstrating normal motor development at preschool age also had plagiocephaly, suggesting that this acquired skull deformity has no long-term implications for motor development in these patients.[16] In our study, occipital plagiocephaly occurred in about one third of all torticollis cases, although the skull deformity was significantly more prevalent among the infants with motor asymmetry.

The association between congenital torticollis and subsequent plagiocephaly is well recognized,[6] and as previously mentioned, spending longer periods on a supine or on a sideward position has increased the prevalence of this association. Boere-Bonekamp and van der Linden-Kuiper[3] coined the term “postural preference” for this condition, which often leads to referrals, additional diagnostics, and treatment. These and other authors describe “symptomatic asymmetry,” referring to asymmetric posturing and limited range of passive movement of the head and neck.[2,3] Motor asymmetry, that is, preference in hand use and leg movement in these infants, was not reported.

The impact of associated plagiocephaly on motor development in torticollis infants is unclear; in our series, plagiocephaly was significantly more common in infants with motor asymmetry. We believe that plagiocephaly indicates the presence of more serious torticollis, and thus the risk of functional asymmetry increases in these cases. Hence, plagiocephaly in itself would not play a role in the development of motor asymmetry but it is rather associated with the degree of head movement limitation from the torticollis. A recent study revealed that in very preterm infants, in whom plagiocephaly commonly develops, sleeping in the supine position rather than plagiocephaly was predictive of motor asymmetry at the corrected age of 6 months.[20]

Another objective of our work was to investigate the impact of motor asymmetry on the proportion of diagnostic procedures obtained. There was no difference in the extent of etiologic evaluation, if any, between the two groups. Infants with asymmetry were not referred for neurological evaluation more often than infants without asymmetry. Perhaps the reason for this is that the various therapists, especially the expert physiotherapists, are aware of the existence of temporary asymmetry and therefore do not find the need to refer for a neurological examination.

Table 4 shows the lack of correlation between the direction of torticollis and the direction of asymmetry. There are several possibilities to explain this finding: the research of Ocklenburg et al.[11,12] suggested that among infants with torticollis to the right, the dominant hand was the right hand in 100% of infants, whereas among infants with torticollis to the left, the dominant hand was the right hand in 78%, less than that in the general population but not fully correlating with the direction of torticollis. In our study, there was 100% correlation between the direction of torticollis and the preferred hand when the torticollis was to the right but not when it was to the left. It is possible that these results from a higher incidence of right-hand dominance are expressed at an early stage. Another possibility is that differences among observers were found during review of the medical records in the definition of the torticollis, with

some conventionally defined by the direction of the chin and some defined by the neck tilt direction. Thus there may be cases in which the lack of correlation between the direction of torticollis and the direction of asymmetry is not real but derives from faulty definition.

Long-term follow-up results of torticollis indicate findings similar to those of Boere-Boonekamp and van der Linder-kuiper[3] who followed infants with torticollis for two years. Although treatment duration was significantly longer for infants with asymmetry, the proportion of toddlers still showing torticollis at the age of 2 years did not differ between the two groups.

Epidemiologic and congenital data were similar for both groups, conversely to previous reports.[1,2] We did not identify a higher incidence of uterine anomalies, multiple pregnancies, or breech presentation among mothers of infants with torticollis. Regarding the amount of amniotic fluid, (oligo- or polyhydramnios), this complication was reported in only 3 of the 173 cases, two among the asymmetry group and one in the nonasymmetry group ($P = 0.048$). Interestingly, no cases of developmental hip dysplasia were found in our series in spite of a reported prevalence of up to 4% of developmental hip dysplasia among patients with congenital torticollis.[1,13]

Our study has several limitations. First, being based on chart annotations from different physicians and therapists, there may have been differences in the clinical assessments and clinical progress documentation between patients. However, as our physical therapy team uniformly uses the Alberta Infantile Motor Scale test, the level of motor performance of these infants was evaluated in a similar fashion, thus reducing interexaminer differences. Second, and most important, as therapists and physicians documenting data did so in a practical, routine manner, some data such as assessment of visual field preference was scarcely observed. Moreover, patients were monitored at diverse stages; hence, information on clinical progress was not uniformly documented. Third, data on long-term motor outcome beyond the age of two years was not available for the one third of children who still exhibited some degree of motor asymmetry at this stage. As previously stated, we are confident that had abnormal neurological findings been noticed, most of these patients would have been referred back by community pediatricians to our child development center, as our institution is the only referral center in our area.

In conclusion, transient motor asymmetry occurred in one quarter of our patients with postural torticollis and had disappeared in two thirds of the patients by age two. Our findings support our hypothesis that this is a benign phenomenon that does not require additional evaluation or special treatment except for standard treatment of torticollis. We detected a higher incidence of plagiocephaly and delayed motor development in the group with functional motor asymmetry. These findings suggest that the presence of asymmetry and the presence of plagiocephaly indicate a more severe torticollis. Perhaps this explains the higher incidence of transient motor disabilities in a certain percentage of infants with congenital postural torticollis, as described in the existing medical literature.

The authors thank Nava Jelin for her assistance with statistical analysis.

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