Motor and cognitive development at one-year follow-up in infants with torticollis

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Abstract

Background: Clinical experience suggests infants with torticollis are at risk for developmental delay.
Aims: To examine gross motor (GM) skills at presentation in infants with torticollis; report first-year GM and cognitive outcomes; examine relationship between types of torticollis and above outcomes.
Study design: Prospective follow-up study.
Subjects: Infants referred to 2 regional child development centers from April 2001–December 2002 with torticollis/head tilt and no perinatal complications syndromes were studied.
Outcome measures: GM function was measured using the Alberta Infant Motor Scale and classified as normal (>10%), suspect (5–10% inclusive), or abnormal (<5%). Cognitive function was measured at follow-up using CAT-CLAMS-r Developmental Assessment. Follow-up data obtained between 8 and 15 months of age.
Results: One hundred and one infants with torticollis were seen for initial assessment at mean age 2.9 (SD 1.5) months. Eighteen had sternomastoid tumor, 47 muscular torticollis and 36 postural torticollis. At presentation, 35 (35%) of the 101 infants had suspect or abnormal GM function. 19/66 children with normal GM and 17/35 with suspect or abnormal GM function had postural torticollis (p=0.054). All children received physical therapy. Follow-up assessment of 83 participants, mean age 12.8 (SD 3.6) months, showed 75 had normal GM function and 8 had suspect or abnormal GM function; 11/83 still had torticollis. Cognitive assessment on 66 infants, mean age 14.4 (SD 4.8) months, revealed 57 (87%) had normal cognitive function and 9 (13%) were either delayed or significantly delayed.
Conclusions: Infants with torticollis are at increased risk for early GM delay but most normalize by one year. Torticollis is not associated with delays in early cognitive function.

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1. Background

Congenital Muscular Torticollis (CMT) is a common condition in infancy warranting referral for evaluation and treatment. Cheng et al. [1], in reviewing the literature, reported a prevalence of congenital muscular torticollis between 0.3 and 2.0% and classified infants with CMT into three groups based on clinical examination: a palpable sternomastoid tumor (SMT), muscle tightness on passive head flexion or rotation but without sternomastoid tumor (MUSCULAR), and all the clinical features of torticollis, but lacking tightness or tumor (POSTURAL).

The etiology of CMT remains uncertain. The most prominent explanations are the intrauterine theory and the birth trauma theory [2]. Both propose that mechanical stresses, due to either intrauterine malposition including breech presentation or difficult vaginal delivery, damage the sternocleidomastoid (SCM) muscle, which leads to muscle fibrosis, either directly in the case of intrauterine malposition or indirectly due to bleeding with hematoma formation in difficult vaginal delivery. However, both theories have significant problems. Demirblek & Atayurt reported that only 52% of infants with torticollis had evidence of birth trauma or intrauterine malposition, and they concluded that mechanical/focal injury is not the sole etiologic factor in torticollis [3]. Wei et al. noted that more than 60% of the infants for whom data were available had spontaneous vaginal delivery without complications [4]. These studies suggest that CMT is due, at least in part, to other, unknown, factors.

Our two child development centers are primary sites for assessing and providing physical therapy to infants with torticollis. We have noted in our clinical practice that some infants referred with CMT present with early motor delay and persist with later developmental disabilities long after the torticollis has resolved. While there have been numerous studies examining the management and outcome in infants with CMT, these studies have not reported the presence of developmental disability [1–7]. One study examined this issue indirectly. Miller and Clarren identified a cohort of 174 infants with deformational plagiocephaly [8]. They reported that 84% of this cohort was believed to also have torticollis. In follow-up, using telephone interviews, they contacted parents of 63 of these children to discuss developmental outcomes. They reported that 39.7% of these 63 children had received special help in primary school including special education assistance, and physical, occupational or speech therapy.

We hypothesized that CMT is associated with abnormal central nervous system development, accompanied by an increased incidence of developmental delay. In particular, it is likely that infants with the postural subtype are more at risk for developmental delay, perhaps because of a central neurological dysfunction. Infants in whom torticollis resulted from damage to the SCM (muscular torticollis or sternomastoid tumor) would be less likely to evidence developmental delay.

It is important to investigate the link between CMT and developmental delay in order to intervene more efficiently for follow-up and outcome. Categorizing the potential developmental abnormalities in infants with torticollis in general as well as in these subtypes in particular would allow for more targeted treatment and follow-up with these children, as well as possibly prevent future disability.

2. Objectives

The objectives of this work were to examine:

1) the gross motor skills of infants referred with torticollis at presentation and one-year follow-up;
2) the cognitive skills of infants referred with torticollis at one-year follow-up; and
3) the relationship between the types of torticollis and the above outcomes.

3. Patients and methods

3.1. Design

Prospective follow-up study. The Director of the Medical Section of the HMO approved this study and written parental consent was obtained.

3.2. Setting

Two regional child development centers in central Israel, which are the primary referral sites in their respective locations for infants with torticollis who belong to the Kupat Holim Meuhedet Health Maintenance Organization, one of four in the country.

3.3. Patients

Infants were referred by physicians or public health nurses for evaluation and were not solicited for this study. From April 2001 through December 2002, parents of all infants who were less than one year old when symptoms of torticollis or head tilt manifested were invited to join the study. Inclusion criteria were gestational age of at least 37 weeks; birthweight >2500 g for singleton births and >2000 g for twin births; and no significant perinatal complications or suspected medical syndromes. Infants with torticollis due to etiologies other than CMT were excluded from the study.

3.4. Measurements

A developmental pediatrician and a pediatric physical therapist examined each infant and completed a preprinted data form prepared for the study. The following data were obtained: gender, gestational age, birth weight, obstetric and birth history including presentation, type of delivery and perinatal course, age of referral, source of referral, maternal report of age of onset, presence of craniofacial deformation (plagiocephaly) [9], presence of head side flexion, head rotation, passive head range of motion, presence of sternocleidomastoid tumor. Parents were asked to obtain hip and neck sonograms of their infants [10–13]. Staff radiologists not involved in the study analyzed the sonograms. A standardized protocol for assessment of the neck sonograms was not used nor was echogenicity of the SCM muscle noted in the results.
Infants were initially classified according to the head position in supine according to the presence of side flexion and/or rotation of the head with further delineation according to which side was involved. Furthermore, range of motion was examined both actively in response to following a visual stimulus (Face) and passively. Using these data, we classified the infants into clinical groups described by Cheng et al. [1]: SMT, MUSCULAR, POSTURAL.

At initial presentation and at follow-up, gross motor function was measured using the Alberta Infant Motor Scale (AIMS) [14,15]. Scores were classified as follows: >10% — normal, 5–10% — suspect, <5% — abnormal. Cognitive function was measured only at follow-up using the CAT-Quotient (DQ) was categorized as normal (DQ > 85) or either of the language or visual/fine motor components abnormal. The relationships between torticollis type defined clinically at presentation and motor and cognitive development at presentation and at outcome were examined.

All parents were invited to bring their children for weekly physical therapy sessions, which consisted of manual stretching and, when motor delay was present, developmental therapy. In addition, parents were instructed on how to employ stretching exercises at home on a daily basis and their ability to perform the exercises was examined at therapy visits. It should be noted that an accepted treatment protocol for torticollis is implemented in our clinic [19]. As the torticollis improved, therapy sessions were reduced in number and home exercises revised. However, those children who continued to show delayed motor development with or without persistent torticollis continued to receive weekly therapy. Persistent torticollis was defined as the presence of an asymmetrical head position while supine or sitting, due to either side flexion or rotation.

4. Results

4.1. Referral and perinatal data

One hundred and one infants, (67 male, 34 female), with a mean referral age of 2.8 (SD 1.5) months were diagnosed with torticollis. The referring sources were primary care physicians or well baby care clinics (83.2%) or orthopedists (16.8%). Forty infants were reported by the mothers to have onset of torticollis within the first week of life; the latest reported onset was at 5 months of age. Mean gestational age was 39.7 (SD 1.3) weeks, and mean birth weight was 3272 (SD 491) g. Seventy-three infants were delivered via vaginal delivery, 9 infants via vacuum/forceps assisted vaginal delivery and 19 infants via caesarian section. Six infants had breech presentation and were delivered by caesarian section. Five infants had oligohydramnios during the pregnancy.

4.2. Torticollis data

Data on head position in supine relative to the presence of neck side flexion and/or rotation are summarized in Table 1. Passive range of motion examination found limitation of head side flexion or rotation in 65 (64.3%) infants and limited active range of motion in 78 (77.2%) infants. Based on these data, we classified the infants into three groups: 18 (17.8%) with sternocleidomastoid tumor, 47 (46.5%) with Muscular, and 36 (35.6%) with Postural. Thirty-one (30.7%) infants had craniofacial deformities, which presented as plagiocephaly.

Sternocleidomastoid ultrasound was obtained in 57 of the 101 (56.4%) cases with 16/57 (28.1%) being abnormal. Ultrasound findings agreed with clinical examination in 30/57 (52.6%) of the cases. Ultrasound examinations were reported as normal in 24 (42.1%) cases with clinical findings of tumor or muscular torticollis. Ultrasound examinations were reported as abnormal in 3 (5.3%) cases where clinical findings were consistent with a classification of postural torticollis. Hip ultrasound was obtained in 80 of the 101 cases with 2 cases being abnormal (2.5%; 95% CI 0.3–8.7).

4.3. Developmental data — initial and follow-up

At the initial motor assessment performed at 2.9 (SD 1.5) months, 66 infants were classified with normal gross motor function, 27 with suspect gross motor function and 8 with abnormal gross motor function. Thus, a total of 35 infants were classified with gross motor function below the normal range (34.7%; 95% CI 25.5–44.8). Follow-up motor assessment was performed at age 12.8 (SD 3.6) months on 83 of the 101 infants in the original cohort. Seventy-five were classified with normal gross motor function, 5 with suspect gross motor function and 3 with abnormal gross motor function (Table 2). Thus, a total of 8 (9.6%) infants were classified with gross motor function below the normal range. The results show a significant clinical improvement in the motor function of this group as compared to motor function

<table>
<thead>
<tr>
<th>Gross motor function</th>
<th>Initial N (101)</th>
<th>%</th>
<th>Follow-up N (83)</th>
<th>%</th>
</tr>
</thead>
<tbody>
<tr>
<td>Normal</td>
<td>66</td>
<td>65.4</td>
<td>75</td>
<td>90.4</td>
</tr>
<tr>
<td>Suspect</td>
<td>27</td>
<td>26.7</td>
<td>5</td>
<td>6.0</td>
</tr>
<tr>
<td>Abnormal</td>
<td>8</td>
<td>7.9</td>
<td>3</td>
<td>3.6</td>
</tr>
</tbody>
</table>
on presentation (P<0.0001). Of the 83 infants at follow-up examination 11 (13.3%), still had torticollis, of whom all except one had normal gross motor function.

Cognitive assessment was performed on 66 of the 101 (64.4%) infants in the original cohort at 14.4 (SD 4.8) months. Language DQ was 98 (SD 13), Performance DQ was 101.2 (SD 18), and Full Scale DQ was 99.6 (SD 13). Fifty-seven (86.4%) infants had normal cognitive function, 8 (12.1%) had delayed cognitive function, and 1 (1.5%) had significantly delayed cognitive function. Thus 9/66 (13.6%, 95% CI=6.4–24.3) had cognitive function below normal, which does not differ significantly from the general population [20] (Table 3).

The low follow-up rate for cognitive assessment, 64%, reflects non-compliance on the part of parents despite specific requests for follow-up. We examined if there were any significant differences between those infants for whom cognitive follow up was performed vs. those lost to follow-up, as this might affect the validity of the findings at follow-up. Initial gross motor scores for normal, suspect and abnormal groups did not differ (P=0.72, Chi-squared test). A second analysis was performed by combining the suspect and abnormal groups and comparing them to the normal group; results were also non-significant (P=0.91, Fisher Exact Test). Furthermore, no significant differences between the groups were found for age at referral, gender, non-singleton births, gestational age, mode of delivery, age of onset of torticollis, torticollis type, or presence of sternomastoid tumor. There were differences in birthweight and craniofacial abnormalities. However, after correcting for multiple comparisons, even these two findings were not considered significant. In summary, no significant differences in relevant variables were found between the groups that had cognitive follow-up and those lost to follow-up.

4.4. Characteristics of torticollis vs. developmental outcome

The relationship between the torticollis type defined clinically at presentation and gross motor function at initial presentation was examined. Of the 35 infants who had suspect or abnormal gross motor function, 17 (48.6%) had postural torticollis as compared to the presence of postural torticollis in 19 of 66 children with normal gross motor function (28.8%). (Table 4) This ratio almost reaches statistical significance (P=0.054, Fisher Exact Test, two-sided).

Five of the eight infants who had either suspect or abnormal gross motor function at follow-up had presented initially with postural torticollis. The only child to have abnormal cognitive function at follow-up had postural torticollis. In contrast, of the 8 children who had suspect cognitive function and 1 child who had abnormal cognitive function, 4 had postural torticollis, which is not more than expected for this group.

We conducted several secondary analyses of the data: the relationship between the presence and type of neck side flexion and rotation at presentation with motor function at both presentation and follow-up; and with cognitive function at follow-up. Results were found not to be significant. Similarly, we found no association between the presence of plagiocephaly and motor and cognitive function.

5. Discussion

Congenital muscular torticollis including postural torticollis is a localized asymmetry in infancy with preferential posture of the head and asymmetric cervical movements [21]. Etiology remains unclear though in most cases of CMT it appears to involve damage to the SCM. This study found that infants with CMT were generally at increased risk for concomitant early gross motor delay, and those with the postural torticollis subtype appeared to be at greatest risk. Follow-up data indicate that most infants with motor delay normalized by one year of age. However, it should be noted that the infants in our cohort received physical therapy. Among the few that persisted with motor delay, there appears to be an association with postural torticollis on presentation. In contrast to the motor development findings in this study, early cognitive function does not appear to be affected by torticollis. Further study will reveal whether minor developmental problems manifest later. Our findings provide a basis for communicating cautious optimism to parents and pediatricians of newborns with torticollis.

To our knowledge, this is the first study that examined and demonstrated a relationship between CMT and early infant motor development. Miller and Clarren found that school age children who had plagiocephaly as infants were at greater risk for developmental difficulties [8]. As noted before, the overwhelming majority of their population had associated torticollis. In contrast, our study did not find cognitive developmental abnormalities in infants with torticollis. This may be due to the different ages of the samples examined.

The relationship between CMT and delayed early motor development is still unclear. However there is mounting evidence from various aspects of child development suggesting an interaction between posture and early gross motor development. Hopkins et al. reported that the maintenance of an upright head position was preceded by a marked improvement in postural stabilization, defined as the ability to hold the head upright when seated in an infant chair [22]. Cioni and Prechtl found that normal infants developed the ability to maintain head position in the midline in the supine position by 12 weeks of age [23], and Hylton noted that the presence of postural torticollis and tone dysfunction affected antigravity postural control and balance [24].

### Table 3
**Torticollis: cognitive function at follow-up**

<table>
<thead>
<tr>
<th>Cognitive function</th>
<th>N (66)</th>
<th>%</th>
</tr>
</thead>
<tbody>
<tr>
<td>Normal</td>
<td>57</td>
<td>86.4</td>
</tr>
<tr>
<td>Delayed</td>
<td>8</td>
<td>12.1</td>
</tr>
<tr>
<td>Significantly delayed</td>
<td>1</td>
<td>1.5</td>
</tr>
</tbody>
</table>

### Table 4
**Torticollis type vs. gross motor function at initial presentation**

<table>
<thead>
<tr>
<th>Torticollis type</th>
<th>Normal</th>
<th>Suspect or abnormal</th>
</tr>
</thead>
<tbody>
<tr>
<td>SMT or muscular</td>
<td>47</td>
<td>18</td>
</tr>
<tr>
<td>Postural</td>
<td>19</td>
<td>17</td>
</tr>
</tbody>
</table>

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Philippi et al. [25] found that more than half the infants in their study had trunk convexity predominance together with the cervical rotation deficit characterizing infantile postural defects. They found that infants with only a single apparent sign of asymmetry have actually a much more generalized disturbance. In a study by Michel and Harkins [26] the complexity of postural asymmetry was found to influence the development of motor preferences as shown in the development of dextral bias in handedness development.

Van Vlimmeren et al. [21] reported that asymmetries in infancy have become more prevalent in the past decade. This has been particularly noted since the recommendation for supine sleeping due to the finding that prone sleep position was associated with sudden infant death syndrome [27]. Furthermore, the sleep position also significantly influences gross motor development. Jantz et al. [28] reported that supine sleep position caused a delay in achieving the milestone of rolling over but was not influential for other motor milestones. Majnemer and Barr [29] found that supine sleep position was associated with delayed motor development by age 6 months.

These reports clearly suggest a relationship between head posture, torticollis and motor milestone delay. Abnormal function of the sternocleidomastoid muscle resulting from muscle abnormality as seen in the SMT and MUSCULAR subtypes and its consequential effect on posture could affect the gross motor development of the infant. Postural torticollis though lacking involvement of the SCM could mediate a delay in gross motor milestone development by the presence of abnormal tone and posture as manifested in the sleep position studies [28,29]. Golden et al. [30] posit that positional torticollis is probably caused by muscular imbalance of the bilateral SCM with the muscles to the side to which the head is turned being weak. Logically it follows that such weakness could affect motor development. Our study found a greater relationship between postural torticollis and early gross motor delay as compared to torticollis of a muscular nature. The comment of Golden et al. [30] provides a possible explanation for this increased risk of gross motor delay by suggesting that the muscle weakness affects motor function as compared to the muscular torticollis which has no such underlying deficit. Our findings raise an important question as to which is the primary problem muscle weakness or postural torticollis. The answer will have implications for intervention and follow-up.

A number of our findings regarding torticollis in general merit some discussion. Two out of 80 (2.5%) infants in our cohort showed hip dysplasia, which is similar to the rate of 4.1% reported by Cheng et al. In a series of 1086 cases [2] and that of von Heidenek et al., who reported a rate of 3.7% [31]. Taken together, these findings suggest that the published estimated range for the incidence of developmental dysplasia of the hip in infants with torticollis, namely 0 to 20% [10], should be revised.

The literature reports that there appears to be a slight male predominance for CMT with a relative ratio of approximately 3:2 [32,33]. Our findings of a 2:1 ratio are consistent with male infants being more affected than females. The literature is inconsistent as regards to which side is affected in CMT. Somnez et al. [34] found that 46% of the infants had right CMT while 54% of the infants had left CMT. In contrast Boere-Boonenkamp and van der Linden-Kuijer [32] found that 68% had a right sided preference and 27% had a left sided preference. In our study we found that 31% of infants had a right side preference vs. 62% with a left side preference. The remaining 7% lacked side flexion.

Physical therapy for children with torticollis is considered standard care with good evidence to show the efficacy of such an approach for almost all infants with CMT [2,7,35–38]. Review of the literature found similar treatment programs for intervention. All authors used a program of stretching the neck muscles in three different planes of movement. The exercise programs involved anterior flexion/extension of the neck, lateral flexion to both right and left sides and rotation of the neck to right and left sides. The treatment protocol required that the infants be placed in the supine position and while the muscles were being stretched the shoulders were maintained in a fixed position on the supporting surface. Some differences were noted between the authors on the number of times the stretching procedures were carried out and the length of time that the new position was held. The exercise protocol carried out in our study is consistent with those principles described as above in the literature.

This study has limitations. It did not assess the effects of physical therapy either on the outcome of torticollis or development. While physical therapy intervention could affect the motor development at outcome and act as a confounding factor, given the clear evidence for its efficacy in treating torticollis it would have been unethical to withhold this treatment. Furthermore, the physical therapy should theoretically only affect the gross motor measure at outcome (AIMS). It definitely did not affect gross motor function at presentation as this occurred prior to treatment.

No data support the effect of physical therapy on cognitive outcome.

The infants were examined in two different clinical sites by several clinicians with extensive experience in examining infants with torticollis and all used the same method and form for collecting the clinical data. However, inter-rater reliability was not examined. Another limitation was the low correlation between the clinical and ultrasound findings of the sternocleidomastoid muscle as compared to other studies [10,11,13]. In our study, examiners working in different clinics without a standardized protocol performed the ultrasound examinations and did not note echogenicity in the ultrasound examination, whereas Cheng et al. stressed the need to note the presence of echogenicity of the SCM on ultrasound [13]. In our clinical experience with ultrasound of the SCM, correlation between the clinical findings and ultrasound has not been as clear as that reported in the literature.

In summary, our study has shown that infants presenting with torticollis are at increased risk for early motor delay. Since this is one of the first studies to examine this connection, more studies are needed to corroborate our findings. Van Vlimmeren et al. [21] reported that unequal cervical posture can affect shape and movement of different body parts therefore warranting systematic diagnostic management of asymmetry in infancy. The findings in our study point out the need for a motor assessment at presentation in infants presenting with torticollis.

The results of our study have implications that are very relevant to clinical care. The treating clinician should be cognizant of the relationship between torticollis and early motor delay and pay special attention to early motor
development in children presenting with torticollis. The clinician can provide reassurance to the parent that the great majority of these infants progress to have normal motor function though probably require physical therapy treatment. Early cognitive function appears to be within normal limits, though ongoing follow-up will be necessary to examine if problems manifest at a later time.

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