Unresolved Congenital Torticollis and Its Consequences: A Report of 2 Cases

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Abstract

Objective: The purpose of this report was to describe the clinical presentation and case management of 2 children with congenital torticollis.

Clinical Features: Two male children (ages 6 and 10 years) presented to a chiropractic clinic with a history of congenital torticollis. They showed signs and symptoms of postural deficiency and developmental delay: posterior plagiocephaly, facial scoliosis, head tilt, compensatory thoracic scoliosis, decreased range of motion in the cervical spine, palpable decreased segmental motion of the upper cervical spine, and an age-related delay in throwing and catching a ball and in one-leg standing.

Intervention and Outcome: Both children received chiropractic care (spinal manipulative therapy) and physical therapy (therapeutic exercises, including neck, back, and coordination exercises). Each patient responded favorably with improvement in both structural (posture) and functional (range of motion of the spine and coordination) deficits.

Conclusions: Both patients responded favorably to the combined therapy. These findings suggest that children with congenital torticollis may benefit from a treatment plan that includes a broad therapeutic approach based on the principles of biomechanics and sensorimotor development. (J Chiropr Med 2017;16:257-261)

Key Indexing Terms: *Congenital Torticollis; Chiropractic; Spinal Manipulation*

Introduction

The incidence of congenital torticollis ranges from 0.3% to 16% and is a condition warranting evaluation and treatment at an early stage.[1,2] Congenital torticollis typically stems from a musculoskeletal problem or an underlying nonmusculoskeletal pathology and is characterized by a head and neck tilt, often combined with a rotational preference of the neck.[3,4]

Classic congenital orthopedic torticollis, mainly referred to as congenital muscular torticollis, is a nonparoxysmal torticollis with involvement of the sternocleidomastoid (SCM) muscle with a pseudotumor in the SCM.[3-5] The literature has reported on nonparoxysmal, musculoskeletal torticollis such as postural torticollis. In this type of torticollis, an imbalance in the neck musculature may be present.[6,7] Furthermore, nonparoxysmal torticollis may be caused by dysfunction in the upper cervical spine, and is sometimes referred to as kinematic imbalance caused by suboccipital strain (KISS).[8] Both postural torticollis and KISS may be observed in infants and have an unknown etiology. Postural torticollis usually has decreased active range of motion (ROM) but normal passive ROM, whereas torticollis with SCM involvement and KISS show decreased active and passive ROM.[2,4,8] All 3 types of torticollis can lead to secondary changes in shape, such as deformational plagiocephaly (DP), facial scoliosis, and infantile scoliosis, and functional problems, including unilateral breastfeeding problems and asymmetrical use of the hands.[3,4,8] In addition to this, in the last decade, the question of whether congenital torticollis can lead to a delay in gross motor and coordinative development in infants, preschool children, and schoolchildren has gained increasing interest among clinicians and researchers.[2,5,8]

The consequences of congenital torticollis that is still present in school-aged children have been only vaguely described in the literature. Therefore, the purpose of this article is to describe the clinical presentation and case management of 2 children with congenital torticollis.

Case Report

The 2 patients were seen at a private chiropractic clinic in Switzerland. The parents gave informed consent to publish this report.

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Case 1

History. A 6-year-old boy presented at a chiropractic clinic complaining of neck pain and stiffness. The mother reported that his head was not "straight" (tilted to the right), and the boy's neck and shoulders appeared tight and inflexible. He was right-handed when performing gross motor movements (eg, throwing and catching a ball). He was a little clumsy, but his fine motor skills were good.

The boy was born through natural delivery at week 40 (weight 3.7 kg and height 52 cm). When the boy was approximately 1 month old, his pediatrician noted torticollis (right rotation, right lateral flexion). His mother did not know what type of torticollis it was. Craniofacial abnormalities were not detected. The torticollis was never treated nor was the case followed up because the family moved to another country for the following 2 years. The mother did not remember if the boy was mostly in the prone position during infancy. He was a "quiet" baby and slept well; he crawled at 11 months, his standing history was unclear, and he started walking at 15 months. At his school entry at 6 years, delayed gross motor skills were detected.

Physical Examination. The chiropractic examination showed right-sided posterior parieto-occipital plagiocephaly, right lateral flexed neck position, left high shoulder, right high pelvic bone with unequal leg lengths (+1 cm on the right), and a left convex curvature in the thoracic spine. He had decreased active and passive ROM in the cervical spine in left rotation (left 70°; neutral 0°; right 90°) and left lateral flexion (left 10°; neutral 0°; right 40°). Difficulty throwing and catching a ball was detected (of 10 attempts, only 4 were successful), as well as difficulty in one-leg standing on the left leg (3 seconds possible on the left side, 8 seconds on the right). The Beighton score (a measure of generalized hypermobility) was 5 of 9. With segmental palpation, decreased joint play and palpable decreased segmental motion were observed at the right C1/C2 and at the right iliosacral joint. A radiograph of the cervical spine showed right lateral flexion of the cervical spine and lateral deviation of C1 on C2 to the right (medial joint space between C1 and C2 was decreased on the left).

The boy was sent for neurologic and ophthalmologic examinations to rule out other underlying causes. No neurologic cause for his asymmetry and delayed gross motor skills was found; his eyesight was normal. Moreover, ultrasonography ruled out fibrosis of the SCM.

Diagnosis. The patient was diagnosed with untreated congenital torticollis since infancy caused by decreased segmental motion at the right C1/C2, with secondary plagiocephaly and physiological changes in the thoracic curvature, as well as decreased segmental motion on the right iliosacral joint, with associated leg length difference of 1 cm on the left side. Also noted was delay in gross motor skills, possibly caused by untreated infantile torticollis and tendency to joint hypermobility.

Treatment and Results. Chiropractic treatment consisted of SMT at the right C1/C2 and at the right iliosacral joint. Seven treatments were given over a period of 3 weeks. Within 3 weeks, full ROM in the cervical spine as well as normal joint play at the right C1/C2 and at the right iliosacral joint were achieved. The posture improved, but a slight right lateral flexed head and neck remained. No differences were observed in leg length, and the curve in the thoracic spine was resolved. Furthermore, one-leg standing was equal on both sides (right and left for 8 seconds). After the chiropractic treatment, a physical therapist provided a series of physical therapy exercises for improving coordinative motor skills. Long-term follow-up was done every 6 months until the boy was 9 years old. No regression of the symptoms occurred during that time.

Case 2

History. A 10-year-old boy presented to a chiropractic clinic complaining of neck and upper thoracic pain. The main reason for the consultation was motor delay, tiredness, and poor posture. The mother reported that the boy had a problem with most gross motor skills, such as running, throwing and catching a ball, and jumping. No problems with fine motor skills were reported.

The boy was born at 39 weeks (weight 3.5 kg and height 50 cm) via forceps delivery. At approximately 3 months of age, the pediatrician diagnosed torticollis. The type of torticollis was not explained to the parents, but ultrasonography ruled out hemangioma or fibrosis of the SCM. The mother remembered that the boy always turned his head to the left, and that his head was flat on the left. Physical therapy was initiated, but the mother did not keep the follow-up treatment appointments (a maximum of 8 sessions was undertaken). When asked, the mother reported that the boy spent little time in the prone position. Since infancy, he had had problems falling asleep. He started crawling at 12 months and walking at 18 months. The mother could not recall the age at start of standing and sitting. Delay in gross motor skills was detected when the boy was 5 years old. He was left-handed. He often got tired when performing physical activity. Furthermore, the mother reported problems with concentration, and attention deficit disorder was diagnosed when the boy was 7 years old. A trial of methylphenidate (Ritalin) therapy did not lead to any improvement.

Physical Examination. Chiropractic examination revealed right lateral flexion of the neck and head and rotation of the neck and head to the left. Furthermore, a pronounced left convex facial scoliosis, plagiocephaly with a flattened parietal bone on the left, and a slightly increased thoracic kyphosis were observed. The boy had decreased cervical ROM in active rotation to the right (left 90°; neutral 0°; right 60°) and decreased passive ROM in the cervical spine in right rotation (left 90°; neutral 0°; right 70°) and in left later flexion (left 10°; neutral 0°; right 40°). One-leg standing on the left was possible for 7 seconds and on the

right for 10 seconds. One-leg standing with his eyes closed was not possible on either leg. When throwing and catching a ball, the boy had 5 successful attempts out of 10. With segmental palpation, decreased segmental motion was observed at the C1/C2 level on the left. A radiograph of the cervical spine showed right lateral flexion of the cervical spine, lateral deviation of C1 on C2 to the left (medial joint space between C1 and C2 decreased on the right), and decreased atlanto-occipital space on the left (occiput in left lateral flexion). A neurological examination had been performed by a pediatric neurologist just prior to the chiropractic examination, but no explanation for the asymmetry and developmental delay could be found.

Diagnosis. The diagnosis was partly untreated congenital torticollis caused by dysfunction in the upper cervical spine since infancy resulting from decreased segmental motion at C1/C2 on the left with secondary plagiocephaly and facial scoliosis. Also noted was delay in gross motor skills, possibly secondary to infantile torticollis and attention deficit disorder.

Treatment and Results. The chiropractic treatment comprised 10 treatments over 5 weeks with SMT for C1/C2 on the left. Simultaneously, a physical therapist provided exercises for improving coordinative motor skills. After 5 weeks, full ROM in the cervical spine was achieved. Joint play at C1/C2 on the left was improved, although it did not become fully normal. There was no change in plagiocephaly and facial scoliosis. Gross motor skills were slightly better; one-leg standing on the left was possible for 12 seconds. The chiropractor tried to encourage more physical activity. The boy continued to complain of extreme tiredness. Long-term follow-up was done every 6 months until the boy was 12 years old. No regression of the cervical ROM and worsening of the spinal lesion and delay in gross motor skill occurred over time.

Discussion

The 2 children described in this article responded positively to combined chiropractic care and physical therapy. The treatment outcome consisted of improvements in posture, increased active and passive ROM in the neck, and improved motor and coordinative skills. This suggests a possible spinal mechanical cause of the torticollis in these patients. Furthermore, it was suspected that there could be a relationship between developmental delay at school age and congenital torticollis.

Some authors have considered an association between infantile torticollis and delayed gross motor skills and developmental coordination disorder. Schertz et al[2] found that 19.1% of a cohort of 68 children with congenital torticollis were at risk for developmental coordination problems and delayed gross motor skills when the children were 9.6 years old, even though they had been treated for torticollis during the first year of life or longer. The follow-up prospective multisite study on congenital torticollis at 1 year of age used both congenital torticollis with SCM involvement and positional torticollis as inclusion criteria. The motor skills assessment, based on the Movement Assessment Battery for Children (MABC-2) or the Bruininks-Oseretsky Test of Motor Proficiency, was carried out by a trained pediatrician or physical/occupational therapist.

The findings in the study by Schertz et al[2] were contradictory to those of other studies, which found gross motor delays in children with infantile torticollis resolved mostly when the children were 12 to 18 months of age.[1,9,10] In an earlier study, a prospective follow-up study of 83 infants with congenital torticollis assessed at 2.9 and 12.8 months of age, Schertz et al[9] had found early motor developmental delay in the majority of infants with torticollis at 2.9 months but only in a minority of them at 12.8 months; all were assessed with the Alberta Infant Motor Scale. In a similar study by Ohman et al,[10] a group of children with congenital torticollis, all treated at a department of physiotherapy in a hospital in Sweden, scored significantly lower in motor development assessment at 2 and 6 months of age in comparison to the control group, but there were no differences between the groups at 18 months. Here, motor development was also assessed with the Alberta Infant Motor Scale. Furthermore, Öhman and Beckung[6] undertook a follow-up study in which the parents and children from the first study were asked to participate again, and interestingly, the findings of the second study revealed no difference in motor task skills in 3.5- to 5-year-old children who had congenital torticollis in comparison with the control group. Gross and fine motor developments were evaluated according to the MABC-2.

Similar findings, but with other parameters for defining gross motor development (ie, rolling over skill and crawling and sitting without arm support assessed by an experienced physiotherapist and a pediatric neurologist), were reported in a study by Cabrera-Martos et al,[1] revealing that in infants with DP and torticollis, crawling and standing skills were delayed in comparison with infants with DP without torticollis, after adjusting for the severity of DP.

As indicated by the findings of the abovementioned studies, there seems to be a somewhat contradictory observation in that children with torticollis and developmental delay at 2 and 6 months seem to "recover" from their developmental delay when they are 12 to 18 months of age, but that a further delay can be observed at school age. The reason for this observation may be that developmental delay caused by torticollis in infancy is "hidden" in the toddler years and becomes apparent again in school-aged children. Demographic surveys have shown that children are most commonly brought to chiropractic clinics during infancy and then again at school age.[11,12] The reason for this is unknown, but it is possible, hypothetically, that school-aged children experience and complain of more pain, and that postural deficits are more obvious in infants and in school-aged children than in toddlers. In addition to this, it is possible that delay in gross motor skills and coordination is more difficult to detect in toddlers than in preschool-aged children and school-aged children because

of the development of visual–motor coordination and the ability to perform complex motor activity, which increases from 5 years of age.[13]

The findings in the 2 cases in the present article support the findings of the abovementioned studies; both patients had congenital torticollis, and they both showed delayed motor skills as infants and then again at school age, but between 2 and 5 years of age, there seemed to be a gap in visible developmental delay. Although we could find only 1 study on congenital torticollis and developmental delay at school age[2] in our literature search, the findings in this case series support a connection.

Despite their contradictory findings with regard to late gross developmental delay, the abovementioned studies suggested that congenital torticollis may be associated with early gross motor developmental delay. Thus, the questions are "why" and "how." There is some evidence from child development studies that suggests an interaction between midline posture and early gross motor development.[14] It has been reported that the maintenance of a midline head position was preceded by a marked improvement in *postural stabilization*, defined as the infant's ability to hold the head upright when seated in an infant chair. Hylton[15] reported that the inability to move the head into the position of flexion (chin tuck) in the midline leads to inefficient stabilization of the rib cage and shoulder girdle, which, in turn, disturbs the ability of the oblique abdominal muscles to stabilize the midline posture. Furthermore, Hylton[15] noted that the presence of torticollis and tone dysfunction affected the balance of the infant. Ideally, infants should develop the ability to maintain head position in the midline in the supine position within 12 weeks[13] and in the prone position with the neck stretched (tummy time) within about 16 weeks.[13] Most likely, infants with torticollis are not able to do this because of the fixation of the head in lateral flexion and contralateral rotation, which would disturb sensory information about positional symmetry and balance as well as the weight-shifting movements necessary for transitioning from one posture to another. Ohman et al[10] found that infants who spend more time in the prone position when awake show earlier development of gross motor skills compared with infants who spend no or little time in this position. Furthermore, she found that the group with congenital torticollis spent less time in the prone position compared with the control group. The prone position boosts upper body strength used in the acquisition of many infant motor milestones and activates motor development.[10] It is possible, therefore, that in infants with torticollis, a vicious cycle of not maintaining the midline in the prone position decreases the amount of tummy time, which then causes developmental delay.

There is evidence that early interventions for infants with congenital torticollis result in better outcomes.[16] Manual stretching is the most common evidence-based treatment for congenital torticollis.[16] However, in addition to neck mobility, the overall development of the infant must be considered, such as neck and trunk active ROM and development of symmetrical movements.[16] In addition to the therapeutic interventions discussed earlier, environmental adaptations and parent education are recommended as part of the treatment plan.[16]

Another interesting observation in the 2 case studies presented here was the dominant right-handedness in the 6-year-old boy, who had head rotational fixation to the right in infancy, and the left-handedness in the 10-year-old, who had left rotational fixation of the head in infancy. A secondary functional deficit in congenital torticollis is asymmetrical use of the hands.[8] Öhman and Beckung[6] reported an interesting incidental finding in their study. Of those children who had preferred to rotate to the left as infants, twice as many preferred to use their left hand in the motor assessment test. However, because hand preference for grasping in the construction task does not develop until 4 years of age[6] and the children in their study were 3.5 to 5 years of age, it cannot be defined as left-handedness in many children. Nevertheless, an early preference to rotate the head in one direction might have some influence on hand dominance caused by the visual experience of one hand during infancy. More studies are needed to disprove or confirm this.

Limitations

The author did not use any standard gross motor and coordinative tests, such as the MABC-2, when assessing the 2 children's coordinative and gross motor development. Furthermore, this case series was limited to only 2 patients, and thus the results cannot be extrapolated to others. This case series cannot prove that the developmental delay in the 2 cases was caused only by congenital torticollis. Further studies are required to understand the secondary complications of congenital torticollis, especially delayed gross motor skills and other developmental delays in preschool-aged and school-aged children. Future research should investigate if there is a possible connection between congenital torticollis caused by dysfunction in the upper cervical spine and delayed gross motor and coordination development in school-aged children.

Conclusions

Two children with congenital torticollis, both of whom displayed delay in gross motor skills at school age, responded positively to combined chiropractic care and physical therapy with improvements in posture, increased active and passive ROM in the neck, and improved motor and coordinative skills. This case series suggests that children with congenital torticollis may benefit from a treatment plan that includes a broad therapeutic approach based on the principles of biomechanics and sensorimotor development, including midline head position, transitional movements, balance, and visual inputs.

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Contributorship Information

Concept development (provided idea for the research): M.H.S.
Design (planned the methods to generate the results): M.H.S.
Supervision (provided oversight, responsible for organization and implementation, writing of the manuscript): M.H.S.
Data collection/processing (responsible for experiments, patient management, organization, or reporting data): M.H.S.
Analysis/interpretation (responsible for statistical analysis, evaluation, and presentation of the results): M.H.S.
Literature search (performed the literature search): M.H.S.
Writing (responsible for writing a substantive part of the manuscript): M.H.S.
Critical review (revised manuscript for intellectual content, this does not relate to spelling and grammar checking): M.H.S.

Practical Applications

- These cases suggest that there may be possible motor developmental delay as a secondary consequence of partly treated or untreated congenital torticollis.

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