

Infants With Torticollis Who Changed Head Presentation During A Physical Therapy Episode

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Purpose: The purpose of this study is to describe infants with congenital muscular torticollis (CMT) who changed head presentation during an episode of physical therapy.

Methods: Data were extracted from electronic medical records between January 2015 and December 2018 to describe infants with CMT who changed presentation.

Results: Eighty-nine infants met criteria (predominantly male, non-Hispanic, White, with private insurance). Torticollis symptoms were noticed by 3 months in 90% of infants with a physical therapy examination before 6 months in 100% of infants. Most infants had early mild CMT with abnormal head shape and greater limitation in active cervical range of motion (ROM) than passive cervical ROM. Clinicians frequently recommended weekly physical therapy that included first-choice and supplemental interventions. Episodes of care averaged 7 visits over 64 days with 73% of infants meeting goals.

Conclusion: Physical therapists should recognize that infants with CMT may change presentation but benefit from physical therapy to resolve symptoms.

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Key words: congenital muscular torticollis, infant, physical therapy

INTRODUCTION/PURPOSE

Congenital muscular torticollis (CMT) is a musculoskeletal condition characterized by unilateral contracture of the sternocleidomastoid muscle. Congenital muscular torticollis affects 3.9%¹ to 16%² of infants. Typical CMT presentation is cervical lateral flexion to one side and cervical rotation to the opposite side. Atypical CMT presentation is cervical lateral flexion and cervical rotation to the same side. Physical therapy examination and treatment is the conservative intervention for infants with CMT.³ The American Physical Therapy Association, Academy of Pediatric Physical Therapy has developed a clinical prac-

tice guideline (CPG) for monitoring, evaluating, treating, and providing best practices in the management of CMT.³

Physical therapists complete a comprehensive physical examination of an infant with CMT.³ In addition to evaluation of the face, head, and neck, this examination includes an assessment of the entire musculoskeletal system, developmental skills, movement patterns, posture, positioning, and feeding routines. Physical therapists rule out sudden onset of torticollis and screen other body systems for nonmuscular causes of abnormal posture to determine whether physical therapy is appropriate. A standard course of physical therapy for infants with CMT involves conservative interventions to address the abnormal head posture identified at their initial assessment.

During an episode of physical therapy, some infants may change their head presentation (Figure 1). A switch in head presentation during an infant's course of physical therapy may be an indication of a differential diagnosis. Benign paroxysmal torticollis of infancy (BPTI) is a spontaneously resolving condition characterized by recurrent, abnormal posture of head and neck with cervical lateral flexion that can last a few days or longer.⁴ Infants with BPTI may demonstrate cervical lateral flexion to the same side with each episode of BPTI or switch head presentation sides.⁴⁻⁶ There are ongoing studies investigating the relationship between BPTI and enlarged vestibular aqueduct (EVA) due to similar participant characteristics.^{7,8} Enlarged vestibular aqueduct is the most common structural temporal bone anomaly associated with congenital hearing loss; the hearing loss can fluctuate, is often progressive, and identified outside the

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Fig. 1. Infant changing head presentation.

perinatal period.^{8,9} Individuals with EVA often have a head tilt in infancy before sensorineural hearing loss is identified, as late as 43 months after onset of torticollis.⁸ Sandifer syndrome is characterized by abnormal movement of the head and the neck associated with an episode of gastroesophageal reflux.¹⁰ The abnormal neck posture associated with Sandifer syndrome is intermittent and may involve changing head presentation sides, which is not related to abnormalities of the sternocleidomastoid muscle.^{10,11} Sandifer syndrome has been documented in the literature to be underdiagnosed; therefore, it is underreported.¹¹ To ensure appropriate diagnosis and treatment of patients with CMT, knowledge of differential diagnoses is important.

The CPG indicates that an infant with CMT may change presentation, which would warrant further examination with the referring physician.³ The incidence and characteristics of infants who change presentation during an episode of physical therapy have not been recorded in the literature. The purpose of this study is to describe infants with CMT who changed head presentation during an episode of physical therapy.

METHODS

This was a retrospective study based on electronic medical record (EMR) data from a single-site (hospital-based outpatient physical therapy clinic located in the Midwest, United States) clinical registry.^{12,13} The study cohort was infants with CMT who were seen for treatment between January 1, 2015, and December 31, 2018. Infants were included in the study if the treating therapist documented a change in CMT presentation of the preferred direction of head tilt or rotation during physical therapy intervention. Infants were excluded from the registry if they presented with nonmuscular cause of torticollis, ocular dysfunction, abnormal neurological screen, congenital asymmetry (eg, orthopedic conditions such as scoliosis, abnormal

vertebral structure), medically complex diagnosis affecting standard physical therapy care, or were 18 months old at the time of the evaluation and older. Based on these criteria, 89 infants were included in the cohort (Figure 2).

Procedures

The hospital's Institutional Review Board approved this study. Data were de-identified following extraction from the EMR. Demographic data were gender, ethnic group, race, insurance type, age torticollis was first observed, and age at initial physical therapy examination. Infant history variables were gestational position, prematurity, multiple birth, birth weight, birth length, type of delivery, birth order, family history of CMT, maternal complications during pregnancy, infant complications at birth, delivery type, vision and hearing concerns, time in positioning device (eg, swings, bouncers, car seats), sleep in positioning device, and prone tolerance. Comorbidities of interest included sensorineural hearing loss (bilateral, unilateral, vertigo, and EVA), gastroesophageal reflux, and constipation. Baseline clinical characteristics extracted from the EMR were initial CMT presentation, CMT classification grade,^{3,14} plagiocephaly status,¹⁵ passive and active cervical range of motion (ROM),¹³ and cervical lateral flexion muscle function.¹³ Treatment frequency and referral variables provided at the initial examination were mined from the EMR. Physical therapy interventions, number of visits and days to meet treatment goals, and goal status at the end of the episode of care were extracted as described by Strenk et al.¹²

Analysis

Data were analyzed using IBM SPSS Statistics version 25.0.¹⁶ Descriptive statistics were generated for the variables.

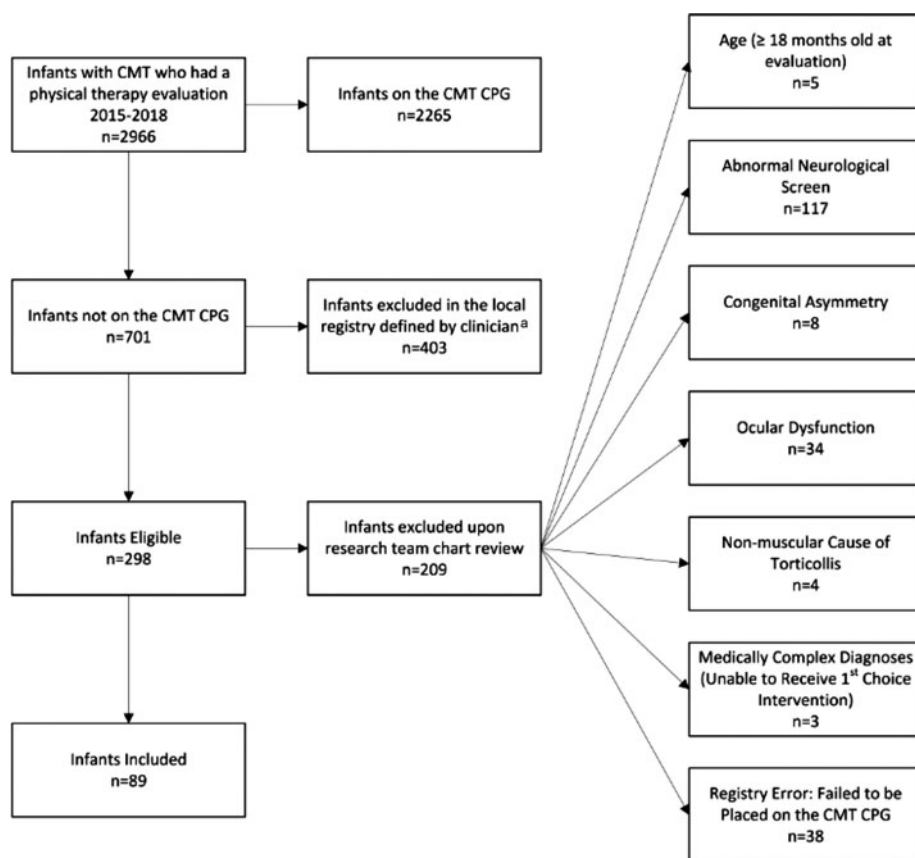


Fig. 2. Flowchart for identifying infants with CMT in this study. CMT indicates congenital muscular torticollis; CPG, clinical practice guideline. ^aNew documentation allowed clinicians to select discrete exclusion criteria.

Continuous variables were described using mean and standard deviation or median. Categorical variables were described using frequencies and percentages. A subgroup analysis of baseline clinical characteristics for infants born premature and infants born full-term was completed using independent *t* tests, χ^2 test, and Mann-Whitney tests depending on data distribution.

RESULTS

Data from 2966 infants with CMT seen between 2015 and 2018 were reviewed, with 89 (3%) infants found to have changed CMT presentation during the course of physical therapy treatment and met conditions for this retrospective study (Table). Of the 89 infants from this cohort, 58% were 0 to 3 months of age at the initial examination and 56% were male. Ninety-six percent of the infants were non-Hispanic, 80% were White, and 55% had private insurance. Some infants (38%) had mothers with maternal complications during pregnancy, including bed rest, gestational diabetes, and low amniotic fluid, and 34% of infants had abnormal gestational position. Several infants (44%) were firstborn children, but 10% of those with siblings had a family history of CMT. Infant birth history included 30% prematurity, 16% multiple births, 55% vaginal delivery, and 28% infant birth complications. The mean birth weight was 5 lb 6 oz and the average birth length was 19 in. No infant had a comorbidity of sensorineural hearing loss, but 29% had gastroesophageal reflux and 9% constipation. Vision and

hearing concerns involved only 8% and 3% of infants, respectively. Positioning equipment was primarily used for less than 1 hour per day during waking hours but 49% of the time for daytime sleeping. Prone tolerance was 5 minutes and less in 47% of the infants.

Torticollis was first noticed by a caregiver or referring physician at 0 to 3 months (90%) in most infants; however, only 58% of infants were referred before 3 months. Presentation at initial examination was left CMT (34%) and right CMT (30%), with the remaining infants having atypical CMT. The majority of infants (78%) had an early mild CMT classification grade. Baseline passive cervical ROM was limited by a mean of 5° (SD: 9°, median: 0°) for rotation and 7° (SD: 9°, median: 5°) for lateral flexion. Active cervical rotation was limited by an average of 19° (SD: 22°, median: 10°), and muscle function of the cervical lateral flexors was an average of 1 point different between sides (SD: 1 point, median: 1 point) at baseline. Most infants (78%) had an abnormal head shape. Physical therapists recommended weekly visits for 72% of infants, with an average number of 7 (SD: 4, range: 2-42) visits completed over 64 (SD: 57, range: 9-264) days. Physical therapy included stretching, strengthening and symmetrical development (91%), kinesiotaping (6%), and myofascial release (3%). More infants (73%) completed a physical therapy episode meeting all established goals compared with infants (27%) not meeting goals (Figure 3).

Because of the prevalence of premature infants (30%), a subgroup analysis between infants born premature and infants born

TABLE

Infants With CMT Who Switch CMT Presentation Demographic and Baseline Clinical Characteristics

	Total Sample, n (%)
Number of subjects	89 (100)
Gender	
Male	50 (56)
Female	39 (44)
Ethnicity	
Non-Hispanic	85 (96)
Hispanic	2 (2)
Not recorded	2 (2)
Race	
White	71 (80)
African American	9 (10)
Asian	1 (1)
Other	2 (2)
Not recorded	6 (7)
Insurance type	
Private	49 (55)
Government assistance	30 (34)
Private and government assistance	9 (10)
Self-pay	1 (1)
Family history of CMT	
Yes	9 (10)
No	53 (60)
Not applicable (only child)	24 (27)
Not recorded	3 (3)
Maternal complications	
None	55 (62)
Gestational diabetes	5 (6)
Bed rest/bleeding	4 (4)
Low amniotic fluid	1 (1)
Other	24 (27)
Gestational positioning	
Unremarkable	48 (54)
Breech	9 (1)
Drop early	4 (5)
Other	17 (19)
Unknown	11 (12)
Prematurity	
No	61 (69)
Yes	27 (30)
Not recorded	1 (1)
Birth weight	
Mean (SD)	5 lb 6 oz (2 lb 5 oz)
Birth length	
Mean (SD)	19 in (2.6 in)
Delivery type	
Cesarean delivery	29 (33)
Vaginal	49 (55)
Unknown	11 (12)
Multiple births	
No	65 (73)
Yes	14 (16)
Unknown	10 (11)
Birth order	
Firstborn	39 (44)
2	24 (27)
≥3	21 (23)
Not recorded	5 (6)
Complication at birth	
None	64 (72)
Respiratory	9 (10)
Hip dysplasia	3 (4)
Jaundice	2 (2)
Cardiac	1 (1)
Foot deformity	1 (1)
Vascular	2 (2)
Other	7 (8)

(continues)

TABLE

Infants With CMT Who Switch CMT Presentation Demographic and Baseline Clinical Characteristics (Continued)

	Total Sample, n (%)
Infant comorbidities	
None	42 (47)
Gastroesophageal reflux	26 (29)
Constipation	8 (9)
Sensorineural hearing loss	0 (0)
Other	13 (15)
Time in positioning device	
<1 h	34 (38)
1-3 h	29 (32)
4-6 h	12 (14)
<6 h	12 (14)
Unknown	2 (2)
Age torticollis detected	
Birth to 3 mo	80 (90)
4-6 mo	7 (8)
7-12 mo	1 (1)
Not recorded	1 (1)
Age at initial physical therapy examination	
Birth to 3 mo	52 (58)
4-6 mo	31 (35)
7-12 mo	5 (6)
≥13	1 (1)
Initial CMT presentation	
Left CMT (left tilt, right rotation)	30 (34)
Right CMT (right tilt, left rotation)	27 (30)
Left atypical (left tilt, left rotation)	14 (16)
Right atypical (right tilt, right rotation)	17 (19)
Right lateral flexion only	1 (1)
CMT classification grade	
Early mild (1)	69 (78)
Early moderate (2)	11 (12)
Early severe (3)	3 (3)
Late mild (4)	3 (3)
Late moderate (5)	1 (1)
Late severe (6)	2 (2)
Plagiocephaly status	
None	18 (20)
Type I	14 (16)
Type II	15 (17)
Type III	27 (30)
Type IV	9 (10)
Type V	1 (1)
Unknown	2 (2)
Other (brachycephaly)	3 (4)
Proposed treatment frequency	
Weekly	64 (72)
Biweekly	24 (27)
Other	1 (1)
Referrals	
Plagiocephaly clinic	58 (65)
Neurology	1 (1)
Ophthalmology	0 (0)
Orthopedics	0 (0)
Physical therapy intervention	
Stretching, strengthening, and symmetrical development	68 (77)
Stretching, strengthening, symmetrical development, and supplemental interventions	9 (10)
Strengthening and symmetrical development	4 (5)
Stretching and strengthening	3 (3)
Stretching, strengthening, and supplemental interventions	2 (2)
Stretching and symmetrical development	1 (1)
Strengthening	1 (1)
Positioning	1 (1)
Physical therapy goals at end of episode	
Met all goals	63 (73)
Did not meet all goals	24 (27)

Abbreviation: CMT, congenital muscular torticollis.

Goals Achieved During PT Episode of Care

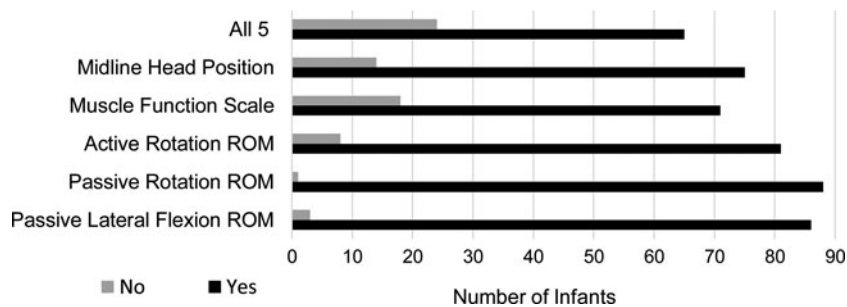


Fig. 3. The number of infants achieving all 5 goals and each individual goal during the physical therapy episode of care. PT indicates physical therapy; ROM, range of motion.

full-term was completed for baseline clinical characteristics. A statistically significant difference was found between groups for birth weight ($P = .049$). No significant differences were found between groups for all additional baseline clinical characteristics: age torticollis symptoms were first observed, age at evaluation, CMT presentation, CMT classification grade, difference in muscle function of the cervical lateral flexors, difference in active cervical ROM, or difference in passive cervical ROM.

DISCUSSION

Infants who change head presentation during the course of therapy are able to meet physical therapy goals. While switching head presentation during the course of treatment for CMT symptoms deviates from following the CPG,³ our findings indicate that infants are still able to achieve resolution of symptoms. Infants who change head presentation appear to have less passive cervical ROM limitations compared with infants who did not switch presentation in a previous torticollis registry.¹² Because of infant head presentation changes, the infant has cervical ROM available and activates muscles from both the ipsilateral and contralateral sides, indicating more of a postural asymmetry than true ROM or strength deficits.

Thirty percent of the infants were born premature, which is higher than the US preterm birth rate ranging from 9.63% in 2015 to 10.02% in 2018.¹⁷⁻²⁰ In our study, infants born premature did not have different baseline clinical characteristics compared with infants born full-term. The only exception was birth weight, which would be expected, given the preterm birth. Prematurity and birth weight are not identified by the CPG as risk factors for CMT.³ In addition, infants evaluated at a young age prior to developing sufficient strength to hold their head upright against gravity may present with a head tilt in the same direction as their rotational preference. Spending extended time in reclined positioning devices such as swings and bouncy seats in addition to limited head control may also contribute to atypical CMT posture. It is also plausible that late change head preference in infants is a result of excessive focus on positioning and interaction to the nonpreferred side or imbalance of muscle function.

Atypical presentation warrants a physician consultation per the CMT CPG as it may indicate a nonmuscular cause of torticollis.³ It is vital that therapists are knowledgeable in the

differential diagnosis of CMT, particularly when treating infants who switch head presentation. The number of infants with an atypical CMT presentation at evaluation was higher in our study (35%) than the number of infants who did not change presentation in a previous cohort (15%).¹³ None of the infants in our study demonstrated sensorineural hearing loss related to EVA, migraines or vomiting indicative of BPTI, or reflux with pain, eye deviation, and dystonic posture typical for Sandifer syndrome, which could contribute to a change in CMT presentation. Ninety-two percent of the infants were younger than 43 months; therefore, the infants were likely too young to have sensorineural hearing loss identified on their problem list at the time of data extraction.

Benign paroxysmal torticollis of infancy is one of the diagnoses that causes an atypical CMT presentation.⁴⁻⁶ Current limitations to diagnosing BPTI in real time include the fact that it is not always possible to identify the symptoms that set it apart from CMT in young infants. For example, it may be difficult to distinguish the vomiting associated with migraines and vestibular involvement from typical infant reflux. Having the ability to identify BPTI at an earlier age within this population would be beneficial in guiding treatment strategies and referrals to specialists as well as potentially providing primary care physicians and therapists with additional diagnostic criteria that can be applied to young infants. In addition to BPTI, there is the possibility that there may be additional factors influencing the presentation of CMT in infants who switch side of tilt or rotation beyond those captured in this study. Those influencing factors may not have been specifically documented in the EMR or analyzed. Therefore, there may be links between switching sides and additional diagnoses that were not identified in this study. We plan to continue to follow this cohort to determine whether symptoms, new diagnoses, or other comorbidities are identified with maturation, which may provide useful information in establishing a differential diagnosis at an earlier age.

To improve accuracy in identifying infants with CMT who switch presentation, we altered our documentation. Items were added to specifically document whether a CMT presentation changed during the course of treatment, and physical therapists were educated on the importance of recognizing a change in presentation. Improving the ease of documenting changes in presentation and stressing the importance to clinicians may result in an increased identification of

infants who change CMT presentation during the course of treatment.

Limitations

Data for infants with CMT who changed head presentation during physical therapy at this Midwestern hospital cannot be generalized to all facilities. This number may be lower than the actual incidence within the population as it is possible that not all infants who changed presentation were identified within the registry. Identification was dependent upon accurate data entry into the EMR by the treating therapist. Documentation error may have contributed to an inability to track every incident. Another limitation to this study is the lack of details recorded within the EMR describing the change and frequency in CMT presentation and the infant's age and number of physical therapy visits before the change in presentation occurred. In addition, factors such as type of change in CMT presentation and frequency of change (eg, once versus multiple times during the episode) could be related to differential diagnoses such as BPTI, EVA, and Sandifer syndrome. These differential diagnoses may have also been missed because the extracted data looked only for diagnoses included in the problem list of the EMR.

Future Research

Additional research is needed to understand infants with CMT who change head presentation. Identifying patterns in presentation, such as a one-time change in head posture versus multiple times, switching preference of rotation rather than tilt, or starting with an atypical versus typical CMT presentation, may allow for greater understanding of associative factors. Establishing subgroups could potentially inform earlier differential diagnoses and guide treatment planning focused on the relationship of excessive time spent in positioning devices prior to the development of head control. None of the subjects in this study received a differential diagnosis at the time of data extraction, but long-term follow-up is needed to fully appreciate missed symptoms (eg, headaches, vomiting, hearing loss) that may be related to differential diagnoses. Following this population longitudinally would provide the opportunity to determine whether infants are diagnosed with BPTI or EVA once they are older. Review of medical records beyond infancy could allow for identification of comorbidities, which may link a change in CMT presentation and other diagnoses. This could inform differential diagnoses as well as guide referrals for this patient population. Furthermore, following a prospective cohort of infants with CMT would be beneficial to study the incidence of changing head presentation during a physical therapy episode of care as well as compare baseline characteristics with a case-control group of infants with CMT.

CONCLUSION

Some infants with CMT change head presentation during an episode of physical therapy. Many of these infants have an atypical presentation of CMT at initial examination with little

passive cervical range of motion deficits resulting in a classification of early mild CMT. The majority of infants who change head presentation achieve resolution of CMT symptoms during an episode of physical therapy. Physical therapists should be aware that a change in head presentation can occur in infants with CMT without identified differential diagnosis, but it is recommended that physical therapy continue until symptoms resolve.

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