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Description of Mandibular Improvements in a Series of Infants With Congenital Muscular Torticollis and Deformational Plagiocephaly Treated With Physical Therapy

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Abstract

Background: Many infants with congenital muscular torticollis (CMT) have deformational plagiocephaly (DP), and a small cohort also demonstrate mandibular asymmetry (MA). The aim of this retrospective study was to evaluate mandibular changes in these infants with previous computed tomography (CT) scans who underwent physical therapy (PT) to treat CMT.

Methods: A retrospective study included patients presenting to a pediatric plastic surgery clinic from December 2010 to June 2012 with CMT, DP, and MA. A small subset of these patients initially received a 3D CT scan due to concern for craniosynostosis. An even smaller subset of these patients subsequently received a second 3D CT scan to evaluate for late-onset craniosynostosis. Patients were treated with PT for at least 4 months for CMT. Initial CT scans were retrospectively compared to subsequent CT scans to determine ramal height asymmetry changes. Clinical documentation was reviewed for evidence of MA changes, CMT improvement, and duration of PT.

Results: Ten patients met inclusion criteria. Ramal height ratio (affected/unaffected) on initial CT was 0.87, which significantly improved on subsequent CT to 0.93 ($P < .05$). None of the patients were diagnosed with craniosynostosis on initial CT. One patient was diagnosed with late-onset coronal craniosynostosis on subsequent CT.

Conclusions: We identified a small cohort of infants with MA, CMT, and DP. These patients uniformly demonstrated decreased ramal height ipsilateral to the affected sternocleidomastoid muscle. Ramal asymmetry measured by ramal height ratios improved in all infants undergoing PT.

Keywords

mandibular asymmetry, deformational plagiocephaly, congenital muscular torticollis, facial asymmetry, physical therapy

Introduction

Deformational plagiocephaly (DP) refers to an asymmetrical head shape that is the result of pressure on the developing skull in utero and/or from a persistent sleep or resting position after birth. The incidence of DP has increased since the American Academy of Pediatrics began the "Back to Sleep" campaign which was instituted in 1992 (Karmel-Ross, 2012). Deformational plagiocephaly is documented as occurring in 18% to 19.7% of babies (Rogers, 2011). We postulate that the increase in the incidence of DP is multifactorial, including the frequent use of and/or sleeping in reclined positioners and chairs such as bouncy seats, reclined rockers, swings, and car seats and the dramatically decreased frequency of tummy time. Infants are spending much more time supine and in reclined positions both day and night and less time prone than in the past (Dudek-Shriber and Zelazny, 2007). Typically, the diagnosis of DP is determined by physical examination. When assessing the

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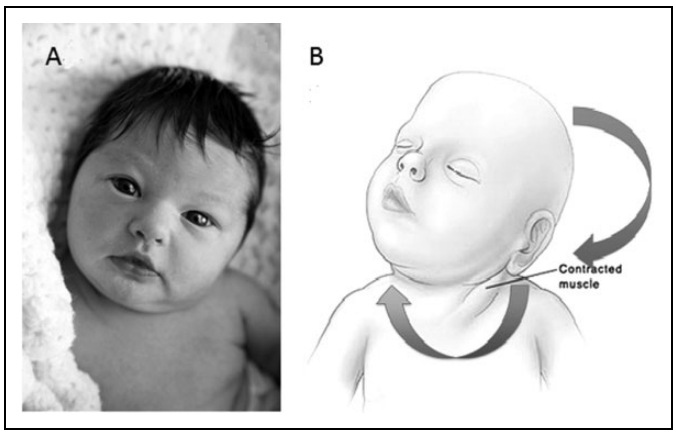

Figure 1. Newborn with congenital muscular torticollis causing the head to tilt toward and rotate away from the affected sternocleidomastoid (SCM) muscle (A). Drawing demonstrating the rotation and lateral flexion in congenital muscular torticollis (CMT; B).

cranial vault, our center utilizes hand caliper measurements of the cranial index (cephalic ratio) and the oblique diagonal difference to determine the severity of DP. However, occasionally, skull X-rays or a 3-dimensional CT scan (3D CT) of the head may be ordered if there is question of craniosynostosis (fusion or premature closure of the skull sutures). Other differential diagnoses may include hydrocephalus and hemi-facial microsomia. Some of the asymmetric facial features observed in children with hemi-facial microsomia may also be seen in children with DP and congenital muscular torticollis (CMT).

Deformational plagiocephaly is highly associated with torticollis, documented by Rogers in 2011 as 70% to 95%. The most common form of torticollis is CMT, and this affects children worldwide. Congenital muscular torticollis is caused by idiopathic fibrosis of the sternocleidomastoid (SCM) muscle that restricts movement and pulls the head toward the involved side resulting in shortening of the SCM muscle. This muscle acts to laterally flex the head ipsilaterally while simultaneously rotating the head contralaterally (Figure 1A and B). Congenital muscular torticollis is the third most common orthopedic diagnosis in infants (Karmel-Ross, 2012). As with DP, there has also recently been a rise in the occurrence of CMT. Previously, CMT was reported to range from 0.4% to 1.9% (Cheng and Au, 1994; Cheng et al., 1999; Cheng et al., 2001; Do, 2006; Tatli et al., 2006); however in, Stellwagen et al. (2008) reported a prevalence as high as 16%. Congenital muscular torticollis can be associated with several comorbidities including DP, facial and mandibular asymmetry (MA), developmental dysplasia of the hip, developmental delays, ophthalmological abnormalities, and gross motor skill asymmetries. Children diagnosed with CMT are typically treated with skilled physical therapy (PT) services to address weakness, range of motion (ROM) limitations, postural deficits, and altered gross motor skill acquisition. Physical therapy is effective in resolving up to 90% to 99% of cases of CMT; surgical intervention is rarely pursued (Karmel-Ross, 2012).

Congenital muscular torticollis has been documented to have an association with MA that can lead to long-term facial asymmetry (Kawamoto et al., 2009; Karmel-Ross, 2012). Mandibular asymmetry is described as ipsilateral vertical ramal growth restriction causing decreased ipsilateral facial height and contralateral open bite. Although children with CMT demonstrate ROM impairments in both cervical lateral flexion and cervical rotation, we have observed MA to occur more frequently when the lateral flexion component is more pronounced (Figure 2A and B). It is important to address MA as soon as it is identified since it can impact breastfeeding and feeding in general (Wall and Glass, 2006). Alterations in dentition due to MA may lead to the need for orthodontics or contribute to temporomandibular joint issues. Cosmesis and facial asymmetry can also be a direct result of MA (You et al., 2010). All of these concerns can become a burden to the individual, the individual's family, or guardian and result in the need for additional medical care and greater cost to society as a whole. Addressing MA early results in a greater potential for improvement and resolution. Craniofacial asymmetry from neglected CMT definitely becomes more severe with age as noted by Jeong et al. (2015).

The timing and efficacy of PT treatment and its impact on MA in the setting of CMT are currently understudied. We identified a unique small cohort of patients with MA in the setting of CMT and DP presenting in infancy to a major children's hospital pediatric plastic surgery department who were evaluated by the multidisciplinary DP/CMT team clinic. When these patients were seen in follow-up for their DP/CMT, we noticed changes in the MA, thus prompting further investigation regarding how MA changed over time. The aim of this retrospective study was to evaluate mandibular changes in a series of infants, younger than 15 months of age diagnosed with CMT, DP, and MA with CT scans available, who underwent regular PT to treat CMT.

Methods

The institutional review board approved this retrospective study (PRO12080186), which included patients who presented to a large children's hospital pediatric plastic surgery clinic from December 2010 to June 2012 with diagnoses of CMT, DP, and MA. This study was performed in order to evaluate mandibular changes and to determine whether there was an improvement in the MA after undergoing a series of PT to treat CMT.

Patients included in this study presented with DP and active CMT and were treated with PT for a minimum of 4 months. Patients had the option of utilizing early intervention or outpatient PT services. In our state, early intervention (birth to 3) provides PT services to qualifying infants and children for the diagnosis of CMT. During PT evaluation, patients' passive and active cervical ROM are measured bilaterally for both cervical rotation and cervical lateral flexion via goniometry, a standardized measurement tool. Normal passive ROM for cervical rotation in an infant is greater than or equal to 90°; cervical

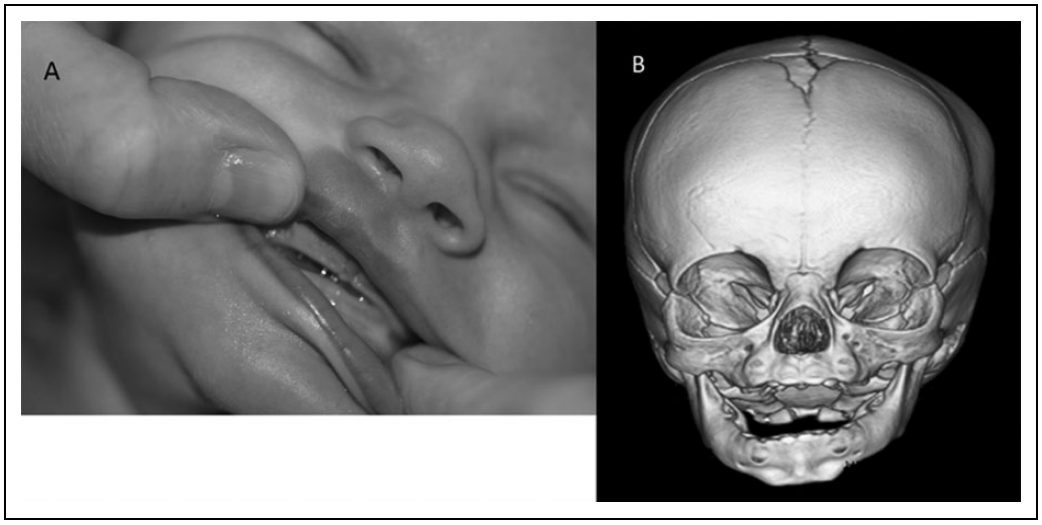

Figure 2. Infant with mandibular asymmetry (A) and correpsonding computed tomography (CT) scan (B) demonstrating shortened ramus associated with significant lateral flexion from congenital muscular torticollis.

lateral flexion is greater than or equal to 40° to 50°. All patients underwent the standard of care for PT in our region for the treatment of CMT which prescribes a frequency of weekly to bimonthly sessions to include stretching, strengthening, developmental, and daily home exercises performed by the caregiver.

Using a coding database, we identified 284 patients from December 2010 to June 2012 who were referred to a large well-established cleft–craniofacial center for evaluation of DP and CMT. Of this group of patients, 213 were examined in a multidisciplinary setting by a single nurse practitioner and a single physical therapist. Consultation with a craniofacial surgeon was available as necessary. Clinical examinations routinely included evaluation for MA by the nurse practitioner via visual and physical examination of the mandible and maxilla. The maxilla and mandible were manually approximated to observe for any degree of asymmetry of occlusion. There is no standardized tool to measure MA; therefore, the documentation was written as "the early closure of the mandible on the side affected by the torticollis and open on the contralateral side," causing a cant of the mandible upward on the side of the torticollis. In addition to their diagnoses of DP and CMT, approximately 10% of the 284 patients referred to the center were found to have MA on clinical examination. A small cohort, approximately 5% (15 patients) with MA underwent 3D CT scans for clinical concerns of craniosynostosis. Ten of these 15 patients demonstrated continued concern for late-onset craniosynostosis, therefore underwent a second 3D CT scan. Computed tomography scans were only obtained when there was a clinical concern for craniosynostosis. The mean time between initial and subsequent CT was 7.5 months (range: 5.5-9.5 months). Both initial and subsequent 3D CT scans were retrospectively reviewed to compare the ramal height asymmetry (calculated ratio: affected/unaffected).

Ten patients met criteria. Data elements collected included demographic information, craniofacial examination for the presence of DP, mandibular/maxillary approximation, PT evaluation, diagnosis of CMT, and both the initial and subsequent 3D CT scan measurements of the bilateral ramal height. These 3D CT scans were reviewed retrospectively. Vitrea software from Vital Images (Minnetonka, Minnesota) was utilized to measure the ramal heights. The ramal heights were measured in millimeters on the side affected by CMT and the nonaffected side at initial evaluation, which was prior to PT intervention, and upon subsequent clinical reassessment, which was after a minimum of 4 months of PT intervention. The ramal height asymmetry ratios were calculated (Figure 3A-C).

Landmarks and measurements constructed in 3D CT images have been found to be reproducible for the diagnosis of facial asymmetry (Kim et al., 2008). Facial asymmetry studies focused on the mandible, incorporating the use of ramal height ratios, utilized measurements in millimeters to assess MA (Hollier et al., 1999; You et al., 2010). Based on these prior studies, we determined it was clinically relevant to obtain objective measurements of the mandible from the 3D CT scans to demonstrate the improvement in MA and subsequent improvement in occlusion regarding the ramal height changes. In this study, the ramus was measured in millimeters from the superior aspect of the condyle to the inferior aspect of the angle of the mandible (Figure 3A-C). Measurements were performed by a lead radiology technician with an advanced degree specializing in CT and magnetic resonance imaging analysis. This radiology technician was blinded to the purpose of the study and patient data. Statistical comparisons were then performed utilizing IBM SPSS Statistics 21 (Armonk, New York). Mean ramal height ratios for the pre- and posttreatment groups were compared, and a Wilcoxon signed rank test was performed to determine the significance of the *P* value.

Inclusion criteria included infants less than or equal to 9 months of age at initial evaluation with diagnoses of CMT, DP, and MA. Only those patients with all 3 diagnoses who had previously undergone both initial and subsequent CT scans

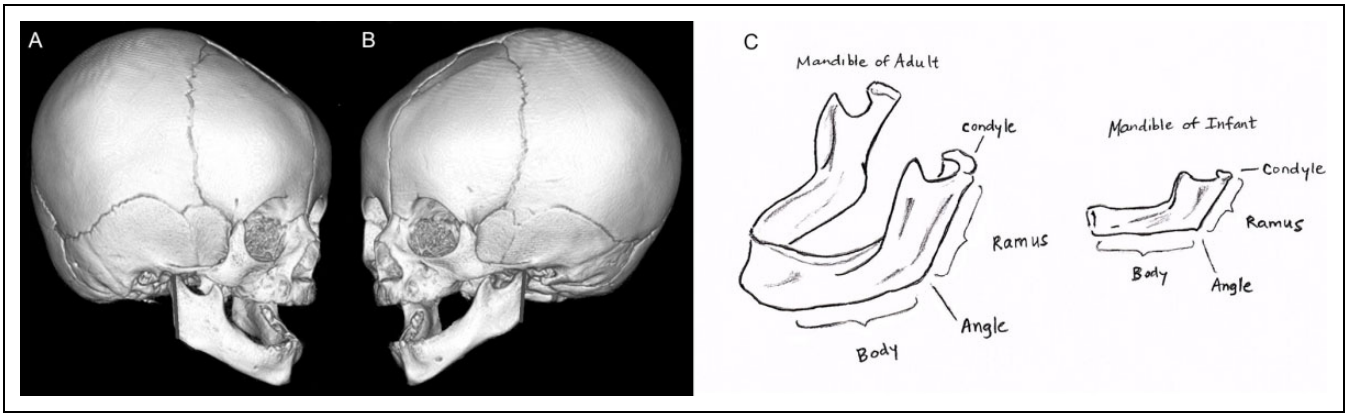

Figure 3. Ramal height of nonaffected side (A) versus the shortened ramal height on the side affected by congenital muscular torticollis (CMT; B). Anatomical landmarks demonstrating the linear ramal height measurement from condyle to angle of body of mandible (C).

were included in the study. Exclusion criteria included infants who were diagnosed with craniosynostosis on initial 3D CT, failed to return for recommended follow-up at our facility, or had significant neurological or neurosurgical diagnoses (such as hydrocephalus, ventriculomegaly, macrocephaly, severe developmental delay, or severe hypotonia). These patients were referred to either the neurology or neurosurgery departments and therefore did not return for further follow-up in our department.

Results

Approximately 10% (25-30) of the patients with DP and CMT had MA on clinical examination. The mean age at presentation was 5.5 months (range: 3-9 months). The mean duration of PT was 6 months (range: 4-9 months), and the mean follow-up was 7.5 months (range: 5.5-9.6 months). Congenital muscular torticollis was left side dominant in 60% of patients and right side dominant in 40%. The patients uniformly demonstrated decreased ramal height ipsilateral to the SCM muscle affected by CMT. Mandibular asymmetry, which was identified as shortening of the vertical ramal height (CT confirmed), correlated with the side affected by CMT in 100% of patients. The initial ramal height ratio (affected/unaffected) was 0.87 mm (range: 0.74-0.96 mm), which improved to 0.93 mm (range: 0.84-1.01 mm) on the subsequent CT. The ramal height ratio change in the affected side on initial CT and subsequent CT was statistically significant with $P < .05$ ($P = .00512$) (Figure 4). All patients demonstrated noticeable improvement in the occlusal plane (clinically and radiologically) after PT intervention (Figures 5A, B and 6A, B). Clinically, the occlusion was assessed by manually approximating the mandible to the maxilla and observing the change in the cant. Clinical observation of improvement in the occlusal cant correlated with the radiological improvement in the ramal hypoplasia. One patient was diagnosed with late-onset right coronal craniosynostosis and underwent surgical intervention with cranial vault remodeling and bilateral frontal orbital advancement. There were no documented complications. Physical therapy initiated shortly after

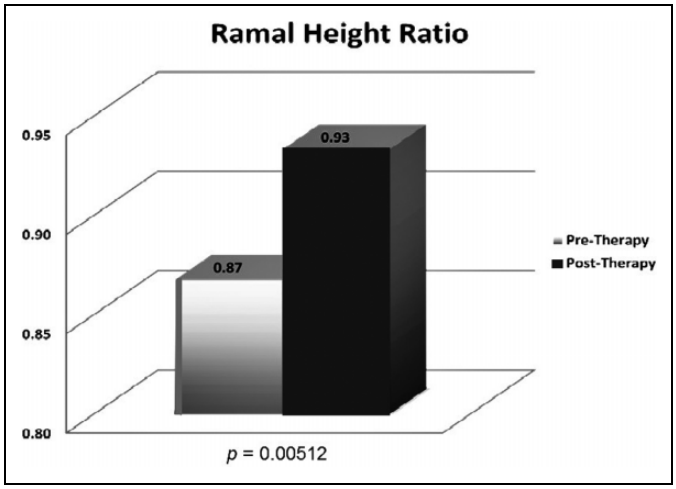

Figure 4. Initial ramal height ratio and subsequent ramal height ratio via computed tomography (3D CT). *P* value was significant; $P = .00512$.

the diagnosis of CMT was associated with improved ramal asymmetry, as demonstrated by calculated ramal height ratios on initial and subsequent CT scans. Mandibular asymmetry improvement is clinically relevant as it is associated with improvement in both malocclusion and facial asymmetry.

Discussion

The incidence of documented CMT has increased and is associated with several comorbidities including DP, facial asymmetry, MA, developmental dysplasia of the hip, developmental delays, and gross motor skill asymmetries. The patients identified in our study all presented with CMT, DP, and MA. A 2002 study by St. John et al supported the clinical observation that MA found with DP is not the result of primary MA, but rather secondary MA due to the rotation of the cranial base. We have observed that MA is more closely associated with CMT rather than in those patients with DP only. We have never identified MA in patients with DP only. We have observed in CMT, when the lateral flexion ROM deficit is more

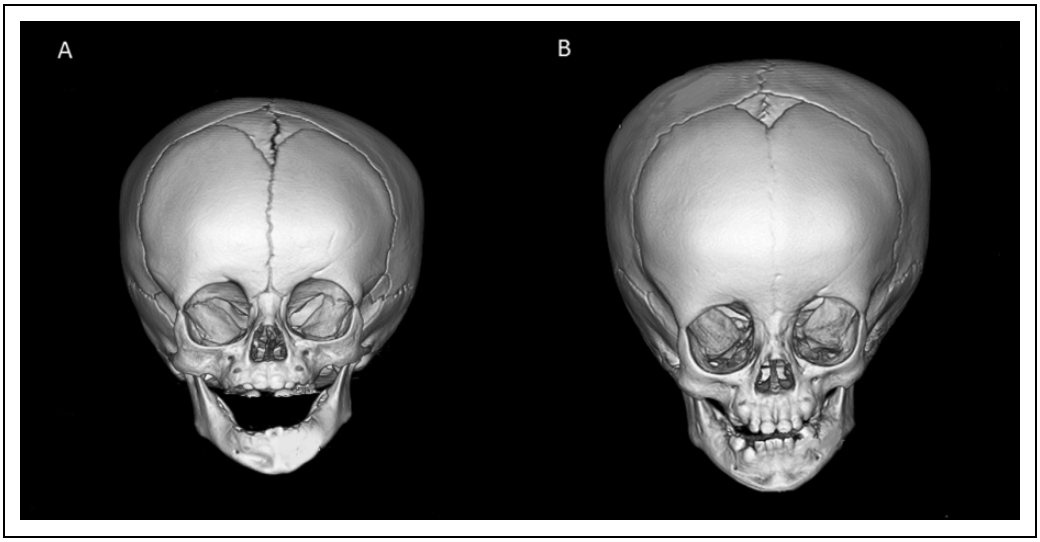

Figure 5. Ramal height asymmetry from congenital muscular torticollis (CMT) prior to physical therapy (A) and the improved ramal symmetry after physical therapy (B).

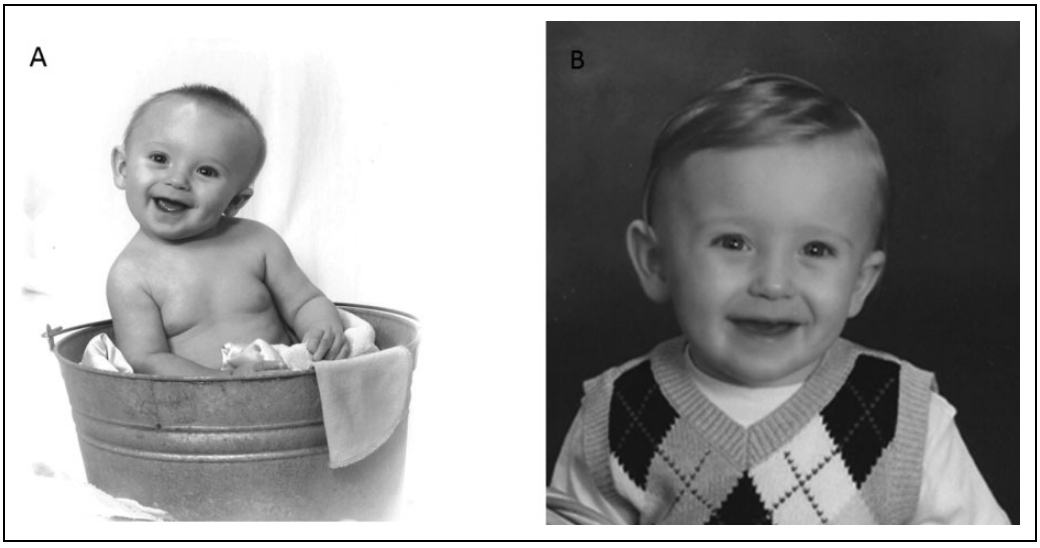

Figure 6. Infant with significant lateral flexion from congenital muscular torticollis (CMT) prior to physical therapy (A) and after at least 4 months of physical therapy (B).

pronounced than the rotational ROM deficit, there appears to be more likelihood of MA. More research is needed to determine the degree of lateral flexion deficit that contributes to MA. Stellwagen et al (2008) observed jaw tilt to occur in 13% of the infants in their research. Among children diagnosed with CMT and DP at our center, we have identified MA in approximately 10%. We observed these patients to subsequently demonstrate clinical improvement in their MA after receiving PT. Although this reflects our clinical observation, this finding may not be representative of this population across the country. Unfortunately, due to the risk of radiation to the developing brain of the infant, CT scans cannot ethically be performed for the sole purpose of documenting changes in MA. Therefore, although clinical improvement has been noted in these patients, obtaining radiological evidence was not clinically nor ethically appropriate.

Currently, the major contributing factor of MA is unknown. In our patient population, MA was observed to occur more frequently in patients with CMT who demonstrated a greater degree of resting cervical lateral flexion in the supine or sitting positions and/or when there was a large discrepancy when comparing left and right cervical lateral flexion passive ROM. We speculate that, in addition to the previously discussed ROM limitations, MA is also adversely impacted by the infant's asymmetrical neck muscle strength and function. This results in an asymmetric ability to actively move the head and neck back to midline. In patients with CMT, the tight, restricted SCM causes a constant torsional pull on the glenoid fossa. This

aberrant force, in turn, translates to the mandible. The condylar region has a huge influence on the growth of the mandible (Yu et al., 2004). If this aberrant torsional force continues, it can result in greater severity of asymmetrical ramal growth.

Muscular torticollis may lead to cranial base asymmetry resulting in DP and facial asymmetry. A tight, restricted SCM may cause cranial base distortion and lead to a favored sleep or resting position because of the head tilt. This constant posterior pressure on the malleable skull contributes to DP. Lower facial asymmetry is typically a slow, gradual progression with increasing mandibular distortion that may lead to an occlusal cant or mandibular deviation (Kawamoto et al., 2009). Previous studies on facial asymmetry report that the mandible appears to be the dominant factor causing facial asymmetry (You et al., 2010). Further investigation regarding the relationship of MA to CMT, rather than solely DP, is needed. Determining the major contributing factor of MA in the setting of CMT is paramount in determining the best treatment options for these infants.

Neither the age of onset nor the degree of MA has been clearly identified in this patient population. In our study, the youngest patient in which MA was identified was 3 months of age; however, we have clinically evaluated infants as young as 4 days of age in which MA was identified. In terms of resolution of MA, we have noted nearly complete resolution of MA in patients as young as 7.5 months of age. Many questions regarding age of onset, age of resolution, degree of resolution, and prediction of severity of MA remain unanswered from this study and prior studies. Is there a correlation between the age of the infant at initiation of PT with the degree of improvement in MA? What degree of improvement, if any, can be achieved if PT intervention is initiated much later or not at all?

There are many other points of discussion that surface regarding the long-term implications of MA if CMT is not diagnosed, is misdiagnosed, if treatment is not initiated in a timely manner, or if it is untreated (Jeong et al., 2015). We question the impact of MA on the infant's ability to feed optimally and whether mastication is impeded as the infant's feeding skills progress. Jaw tilt can interfere with breastfeeding, especially in the instance of torticollis when head rotation is restricted (Wall and Glass, 2006). We speculate that some of these long-term implications could also impact feeding progression. Furthermore, it is unknown as to what degree MA associated with CMT in infancy can contribute to orthodontic issues into childhood, adolescence, and adulthood. In cases of untreated CMT, malocclusion may be the first clinically presenting complaint which would subsequently be identified during an orthognathic examination as asymmetry with a laterally deviated mandible. Occlusion may vary, with an occlusal cant or a more involved anterior and lateral crossbite. Attributing a laterally deviated mandible to CMT or DP may be challenging (Yu et al., 2004; Kawamoto et al., 2009). Additionally, temporomandibular joint dysfunction could be a late result of untreated MA (St. John et al., 2002).

Limitations of the current study include the retrospective nature, lack of a control group, lack of effective clinical measuring device to approximate the degree of MA, inherent selection bias, small patient cohort, and short-term follow-up. Utilizing only clinically obtained CTs for analysis could be another limitation. Selection bias may account for the improved results as this cohort of patients were compliant with follow-up imaging and appointments and completed a minimum of 4 months of PT. Computed tomography scans are a deterrent to monitoring MA due to radiation exposure. Validated tools to measure MA clinically would be a reasonable target for further work. Although the patient cohort was small and the follow-up time was short, it was sufficient to reach statistical significance.

Deformational plagiocephaly and CMT are highly correlated, and the incidences of both diagnoses have increased significantly over the past few years. Congenital muscular torticollis has been documented to have an association with MA, which has the potential to lead to long-term facial asymmetry (Stellwagen et al., 2008; Kawamoto et al., 2009). Physical therapy is known to be effective in conservatively treating and resolving 90% to 99% of cases of CMT (Karmel-Ross, 2012). The efficacy of PT treatment to improve MA associated with CMT is currently understudied. This small retrospective study proposes there is a potential role of PT in improving mandibular symmetry in the setting of CMT. In many instances, the DP is the diagnosis that triggers a child to be referred for evaluation; therefore, we believe it is imperative that when patients are evaluated for DP they are also assessed for CMT. If CMT is identified, patients should be referred to PT as soon as possible, and the provider should also evaluate the occlusion to assess for any degree of MA. Future studies are needed to identify and monitor the course of MA. It would be challenging to design a study with a comparator group that did not receive PT since this would present ethical concerns for withholding treatment that is proven to be effective for CMT. Unresolved MA may contribute to feeding difficulties and/or lead to the need for future orthodontics or surgical intervention later in life, as well as undesired cosmesis. Physical therapy may potentially prevent the need for these interventions or undesired facial asymmetry.

Declaration of Conflicting Interests

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