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Shear wave sonoelastography in infants with congenital muscular torticollis

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Abstract

Congenital muscular torticollis (CMT) is characterized by shortening or excessive contraction of the sternocleidomastoid muscle (SCM). The main purpose of this study was to evaluate the feasibility of quantifying SCM stiffness using acoustic radiation force impulse (ARFI) sonoelastography in infants with CMT. Twenty infants with an SCM thickness greater than 10 mm with or without involvement of the entire SCM length (limitation of neck rotation passive range of motion [PROM]: group 1S >30°, group 1M = 15°–30°) and 12 infants with an SCM thickness smaller than 10 mm with or without involvement of any part of SCM (group 2) were included. The SCM thickness was measured using real time B-mode ultrasound, and the local SCM shear wave velocity (SWV) and subcutaneous fat layer using ARFI sonoelastography. The neck rotation PROM was significantly greater in group 1S (36.5° ± 5.3°) than in group 1M (18.8° ± 4.9°; $P < .01$); the SWV of the SCM in the affected side (2.96 ± 0.99 m/s) was significantly higher than that in the unaffected side (1.50 ± 0.30 m/s; $P < .01$) in group 1. The SWV of the SCM was significantly higher in group 1S than in group 1M. There was significant correlation between the degree of PROM deficit of neck rotation and the SWV of the affected SCM ($r = .75$; $P < .01$) in all infants. This study revealed a difference in the SWV of the affected SCM in relationship to the limitation of neck rotation PROM in infants with CMT, if there was no difference in SCM thickness among infants.

Abbreviations: ANOVA = analysis of variance, ARFI = acoustic radiation force impulse, CMT = congenital muscular torticollis, ICC = interclass correlation coefficient, PROM = passive range of motion, ROI = region of interest, RTS = real time sonoelastography, SCM = sternocleidomastoid, SWS = shear wave sonoelastography, SWV = shear wave velocity.

Keywords: shear wave sonoelastography, sternocleidomastoid, ultrasound, torticollis

1. Introduction

Congenital muscular torticollis (CMT) of the neck is characterized by shortening or excessive contraction of the sternocleidomastoid muscle (SCM),[1–3] which leads to head tilt, limited head rotation, and/or a palpable mass. The incidence of CMT varies from 0.3% to 2.0%.[4]

Although many hypotheses have been proposed including muscle trauma at birth or chronic repetitive microtrauma such as prolonged poor fetal positioning,[5,6] the true cause of CMT remains uncertain. Regardless of the cause of CMT, endomysial fibrosis with deposition of collagen and migration of fibroblasts around individual muscle fibers are regarded as the finding on pathologic evaluation.[7]

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Muscle fibrosis is defined as an abnormal and chronic over-proliferation of extracellular matrix (ECM) components,[8] and fibrosis causes a loss in muscle function.[9] The increased percentage of fibrous tissue in muscle fibrosis is remarkably similar to the progression of SCM muscle contracture in CMT.[7] In general, the severity of CMT has been assessed by measuring passive range of motion (PROM) of neck with goniometer. However, the reliability of PROM of neck is not high (intraclass correlation coefficient 0.54–0.79) even if in cooperative adults.[10] It is supposed that the reliability may be much lower in infants with severe CMT due to poor cooperation. The classifications of the severity of SCM fibrosis using B-mode ultrasound have been reported.[9,11] B-mode ultrasound was well correlated with PROM of neck,[12] and feasible as it can be quickly implemented in a clinical setting. But it is inevitably subjective, semiquantitative (imprecise), and prone to inter-observer error. It can be useful for measuring the thickness of SCM muscle accurately, but cannot evaluate the muscle stiffness.

Real-time sonoelastography (RTS) is an ultrasound-based technique that facilitates the evaluation of tissue elasticity in real time. This technique is based on the principle that tissue compression produces strain (displacement) in hard tissue to a lesser degree than in soft tissue. Compression RTS has recently been used to assess pathologic tissues affected by various muscle disorders and may be useful for evaluating the stiffness of the SCM muscle.[13–16]

However, the compression RTS technique has several limitations, including an inaccurate compression method, limited operator reproducibility, and semiquantitative methods. To overcome these limitations, shear wave sonoelastography (SWS) using acoustic radiation force impulse (ARFI) imaging has been developed. It is a quantitative and objective imaging method for the analysis of tissue stiffness. It has the advantage

that the tissue displacement response is directly related to the magnitude of the applied force and inversely related to the tissue stiffness. This new SWS is based on so-called shear waves, which are generated within the tissue by using a conventional ultrasound wave that interacts with the tissue and induces horizontally directed shear waves that propagate through the tissue. The velocity of these shear waves can be quantified by using ultrafast algorithms to assess tissue composition and elasticity.[17]

To the best of our knowledge, no other study has attempted to quantify the stiffness of muscle fibrosis in infants with CMT using SWS. Therefore, our aim was to evaluate the feasibility of quantifying SCM stiffness with SWS using ARFI in infants with CMT.

2. Materials and methods

2.1. Participants

This study was designed as a retrospective and cross-sectional study. Thirty-two infants (17 boys, 15 girls; mean age: 0.69 ± 0.26) with a clinical diagnosis of CMT were recruited at the outpatient clinic of the department of rehabilitation medicine at an university medical center. They were divided into 2 groups according to SCM thickness on B-mode ultrasound. Group 1 included 20 infants (11 boys, 9 girls; mean age: 0.71 ± 0.25 months), with SCM thickness greater than 10 mm, and group 2 included 12 infants (6 boys, 6 girls; mean age: 0.65 ± 0.27 months), with SCM thickness less than 10 mm. Group 1 was subdivided into 2 subgroups according to the degree of deficits in PROM of neck rotation: group 1S with severe limitation ($> 30°$) and group 1M with moderate limitation ($15°-30°$). The PROM of neck rotation was measured by a physiatrist using goniometer. On the basis of a previous study,[18] normal passive cervical rotational range of motion was defined as 100°.

Inclusion criteria were: CMT that was verified by a physiatrist who is a specialist in pediatric rehabilitation medicine, palpable neck mass on clinical examination, diagnosis of CMT before 3 months of age, and no previous medical treatment for palpable neck mass. Exclusion criteria were: congenital anomalies of the cervical spine, spasmodic torticollis, and neurogenic and ocular torticollis. The study was conducted after the approval had been obtained from the Institutional Review Board and Research Ethics Committee, and in accordance with the Declaration of Helsinki. A waiver of consent was granted for a chart review without infant contact.

2.2. Procedure

The SCM thickness was measured using a real time B-mode ultrasound, and shear wave velocity (SWV, in m/s) of the SCM and subcutaneous fat using SWS in both groups. The B-mode ultrasound and SWS were performed by a physiatrist with 12 years of ultrasound and 7 years of sonoelastography experience using a commercially available ultrasound system by a linear probe with a 4 to 9 MHz frequency bandwidth (Siemens ACUSON S2000, Siemens Healthcare, Erlangen, Germany).

Ultrasound examination was performed after all infants fell asleep with the aid of their parents. During the procedure, the infant was laid across the couch in such a way that the examiner looked down onto the top of the infant's head. A bolster was used to extend the neck, and the head was rotated contralaterally to the examination side. The ultrasound scanning was discontinued whenever the infant woke up, and was tense and uncooperative.

Both longitudinal and transverse B-mode ultrasound images of both SCMs were scanned for each infant. Moreover, it was determined whether the location of the palpable mass was in the lower, middle, or upper thirds of the SCM. Furthermore, the SCM thickness was measured as the distance from the superficial to the deep aponeurosis at the thickest portion of the muscle using electronic caliper on the transverse ultrasound image (Fig. 1A and D). On a transverse image, 3 regions of interests (ROI) were located in the SCM and one ROI in the subcutaneous fat. The examiner tried to apply manual compression as minimal as possible during the SWS. The color-coded SWV map in which the velocities of the shear waves in user-defined ROI was displayed on a spectrum of colors embracing the relevant SWV values. A color spectrum range was also present on the left side of the screen and indicated the lowest and highest limits of SWVs that were displayed. The highest limit of the SWV value was adjusted according to the SWV of ROI (up to 6.5 m/s) because the maximal highest limit setting makes a very wide SWV mapping spectrum.

Quality measurement of shear wave image is useful to confirm that an adequate shear wave was formed.[19] Before measuring the SWV values on the color-coded SWV map, the quality of the map was verified on the Quality Map image that was generated automatically by the software. According to the definition of the manufacturer, "Quality Map" indicates the quality and reliability of the shear wave measurements. These correspond to the areas where the shear waves with sufficient good quality for quantification are shown in green color. The SWV values from 3 rectangular ROI were obtained on the color-coded SWV map. The SWV quantification steps of the SCM are illustrated in Figure 1B and E. The mean SWV value of the SCM was calculated for each scan by averaging the 3 SWV values of the 3 ROI in the SCM (Fig. 1C and F). The SCM in the transverse scan was divided into 4 quadrants on the ultrasound image including antero-superior, postero-superior, antero-inferior, and postero-inferior area. The ROI was placed at 3 middle regions among 4 quadrant areas, between the antero-superior and antero-inferior areas, and between postero-superior and postero-inferior areas in the SCM, and the SWV in each ROI was measured.

Seven seconds interframe cooling period was applied during SWS using ARFI to mitigate thermal safety concerns. The SWS was performed twice, and 2 representative SWS images were taken in each scan to check intra-rater reliability. Our study protocol included a standardized color encoding, and the same color scale was used in all infants.

2.3. Statistical analysis

A prior power analysis based on the pilot results determined that 9 subjects would yield a power of 0.8 at a significance level of 0.05. The statistical analysis was performed using SPSS version 14.0 (SPSS, Chicago, IL) with the level of significance set at < 0.05. The interclass correlation coefficient (ICC) was used to examine intra-rater reliability of repeated SWV measurements. Differences in ultrasound parameters among 3 groups were evaluated using one-way analysis of variance (ANOVA) for continuous variables and chi-square test for categorical variable. The Pearson correlation between SWV of the affected SCM and PROM was evaluated. The post-hoc power analysis was performed and the power was $> .95$.

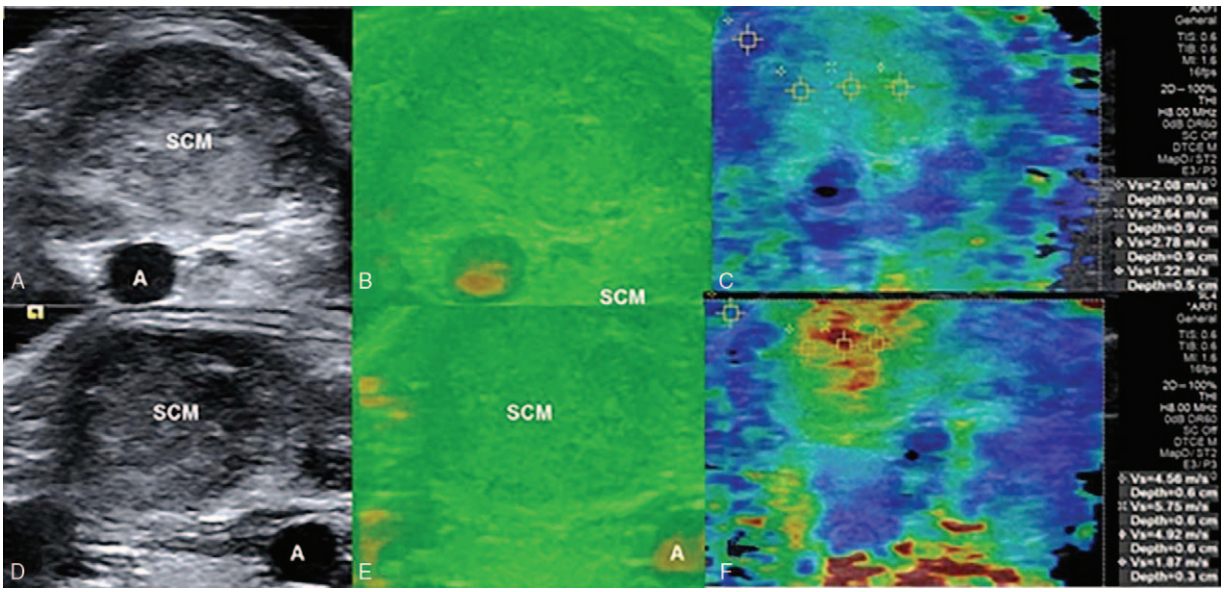

Figure 1. Representative transverse shear wave sonoelastography image of sternocleidomastoid muscle in group 1M (A–C) and 1S (D–F). (A, D) B-mode ultrasound. (B, E) The quality map of the shear wave sonoelastography showed the green color that indicated the high quality image. (C) In group 1M, the mean shear wave velocity (2.50 m/s) was measured in the region of interest of the sternocleidomastoid muscle on shear wave sonoelastography. (F) In group 1S, the mean shear wave velocity (5.07 m/s) was measured in the region of interest of the sternocleidomastoid muscle on shear wave sonoelastography. Group 1S, severe passive range of motion deficit of neck rotation (>30°); group 1M, moderate passive range of motion deficit of neck rotation (15°–30°).

3. Results

There was no significant difference in the demographic data between both groups (Table 1). The mean PROM deficit of neck rotation is 36.5° ± 5.3° in group 1S, 18.8° ± 4.9° in group 1M, and 2.1 ± 2.5° in group 2.

B-mode ultrasound revealed that the entire length involvement of the SCM was observed in 2 infants, and the middle and lower thirds involvement in 9 patients of group 1S. In group 1M, B-mode ultrasound revealed the middle and lower third involvement in 6 infants and the lower third involvement in 3 infants.

The mean affected SCM thickness was 11.5 ± 0.7 mm in group 1, and 7.2 ± 1.8 mm in group 2. However, there was no significant difference of the affected SCM thickness between groups 1S (10.9 ± 2.1 mm) and 1M (11.0 ± 1.0 mm), and the unaffected SCM thickness between groups 1S (5.2 ± 1.5 mm) and 1M (5.0 ± 1.2 mm).

In group 1, the mean SWV of the affected SCM (2.96 ± 0.99 m/s) was significantly higher than that of the unaffected SCM (1.50 ± 0.30 m/s; $P < .01$). The mean SWV of the affected SCM in group 1 (2.96 ± 0.99 m/s) was significantly higher than that in group 2 (2.08 ± 0.36 m/s; $P < .05$). Moreover, the mean SWV of the affected SCM in group 1S (3.65 ± 0.75 m/s) was significantly higher than that in group 1M (2.07 ± 0.36 m/s; $P = .001$) (Table 2, Fig. 1). In group 1S, the mean SWV was 4.98 ± 0.12 m/s in the 2 entire affected SCMs and 3.36 ± 0.41 m/s in 9 nonentirely affected SCMs.

In group 1, the SWV of the affected SCM was positively correlated with the degree of PROM deficit of neck rotation in the affected side ($r = .77$; $P < .01$). There was significant correlation between the degree of PROM deficit of neck rotation and the SWV of the affected SCM ($r = .75$; $P < .01$) in all infants.

There was no significant difference in the mean SWV of the subcutaneous fat in the affected side among the 3 groups (Table 2). The interclass correlation coefficient of the repeated SWV measurement in the affected/unaffected SCM was 0.923/0.912.

Table 1

Demographic data.

	Group 1 (n = 20)				
	Group 1S (n = 11)	Group 1M (n = 9)	$P^{\dagger}$-value	Group 2 (n = 12)	P^{*}-value
Corrected age, months	0.71 ± 0.25	0.68 ± 0.30	.756	0.65 ± 0.27	.674
Weight, kg	3.35 ± 0.46	3.44 ± 0.29	.812	3.32 ± 0.75	.725
Sex (boy/girl)	6/5	5/4	.914	6/6	.765

Values are presented as mean ± standard deviation.

Corrected age, age of the child from the expected date of delivery.

Group 1, SCM muscle thickness of greater than 10 mm; group 1M, moderate passive range of motion deficit of neck rotation (15°–30°); group 1S, severe passive range of motion deficit of neck rotation (>30°); group 2, SCM thickness of less than 10 mm; SCM, sternocleiodomastoid muscle.

* *P*-value, independent *t*-test or chi-square test between group 1 and group 2.

† *P*-value, independent *t*-test or chi-square test between group 1 and group 2.

Table 2

Comparison of shear wave velocity of affected sternocleidomastoid muscle in infants with congenital muscular torticollis.

	Group 1S (n = 11)	Group 1M (n = 9)	Group 2 (n = 12)	*P*-value
SWV of SCM muscle (m/s)	3.65±0.75	2.07±0.36	1.50±0.30	.001
SWV of subcutaneous fat (m/s)	1.51±0.18	1.39±0.26	1.42±0.12	.292

Values are presented as mean ± standard deviation.

Group 1, SCM muscle thickness of greater than 10 mm; group 1M, moderate passive range of motion deficit of neck rotation (15°–30°); group 1S, severe passive range of motion deficit of neck rotation (>30°); group 2, SCM muscle thickness of less than 10 mm. SCM = sternocleiodomastoid, SWV = shear wave velocity.

P-value, one way analysis of variance.

4. Discussion

The key results of this study were that the SWV of the affected SCM in a severe group (PROM deficit > 30°) was significantly greater than that in a moderate group (PROM deficit 15°–30°) in infants with CMT, whose SCM thickness was greater than 10 mm. To the best of our knowledge, this is the first study to quantify the stiffness of the SCM with SWS using ARFI in infants with CMT. In addition, the SWV of the affected SCM in infants with CMT revealed a positive correlation with the degree of PROM deficit of neck rotation. These findings indicate that the SCM stiffness with severe PROM deficit of neck rotation was significantly greater than that with moderate deficit despite of the similar SCM thickness in infants with CMT.

Park et al[12] reported that the thickness of SCM in B-mode ultrasound was well correlated with PROM of neck. In clinical settings, however, physicians encountered several infants who showed similar thickness of SCM, but different PROM of neck, like group 1S and 1M in current study. According to the previous study, we assumed that severe PROM deficit was caused by excess fibrosis of SCM and it reflects the severity of CMT.[2,3] Measuring PROM of neck by goniometer is not difficult but sometimes it is inaccurate and takes a long time in infant because the measuring made the infant tense and uncooperative. In the present study, SWV was significantly correlated with the PROM deficit, therefore SWV is useful to evaluate the severity of CMT.

A previous study has revealed that the increased muscle stiffness associated with CMT was qualitatively measured using compression sonoelastography.[16] In our study, the SWS using ARFI was used for quantitative assessment of the SWV in the affected SCM to overcome the qualitative measurement of muscle stiffness. Liu et al[20] found that the SCM in the patients with postirradiation fibrosis was stiffer than in controls, using SWS. In addition, they demonstrated that SWS was an objective and quantifiable imaging method to reflect the severity of neck fibrosis.

Although B-mode ultrasound has been regarded as the gold standard for the diagnosis of CMT,[4,21] its role seems to be limited to predict the prognosis of CMT since the basic pathological finding in CMT was the muscular fibrosis that cannot be assessed using B-mode ultrasound.

Lin et al[11] have claimed that the extent of fibrotic change in the affected SCM determined the prognosis of CMT. It has been well known that the early detection and treatment for CMT leads to resolution of CMT in the majority of infants.[4,22,23] In some cases of CMT, however, it does not respond to conventional physical therapy and surgical release is needed.[4,24] A previous study revealed that magnetic resonance imaging (MRI) finding was correlated with histopathological finding and was useful to determine whether surgical treatment was needed in infant with CMT.[25] Therefore, the early and quantitative measurement of muscle fibrosis in the affected SCM plays an important role in the therapeutic plan, and SWS is useful for such purpose. MRI is also useful but it is expensive and the sedation necessary for MRI in infants is associated with risks.

A previous study revealed that the addition of microcurrent therapy to therapeutic exercise and ultrasound significantly decreased the duration of treatment in infants with CMT involving the entire SCM muscle.[26] Therefore, in infants with severe SCM fibrosis on SWS, more intensive rehabilitation treatment strategy involving microcurrent therapy is required before surgery.

Interestingly, the SWV of the affected SCM in 2 infants with entire length involvement of the SCM was higher than that of 9 infants with partial length involvement in infants with severe PROM deficit of neck rotation.

This finding indicates that the SCM with entire length involvement is severely fibrotic and stiff. Therefore, there was a difference of the affected SCM stiffness according to the extent of involvement in infants with severe PROM deficit of neck rotation.

The intra-rater reliability of the SWV of the affected and unaffected SCM was high (0.923/0.912). Given that the examinations were conducted by 1 physiatrist due to the uncooperative attitude of the infants, the inter-rater reproducibility of SWS was not assessed. The elasticity of the target volume is the only determinant of the SWV values measured with SWS. The speed at which the shear waves propagate through the medium is strongly influenced by the measurement depth and external compression. The SWV of an object at a deep position is significantly lower than that of an object that is positioned superficially, given that a progressive attenuation of the pulses generating the shear waves occurs as they travel within tissues.[27] In our study, there was a minimal influence of SWV measurement according to object depth since the SCM in infant is a very superficial structure. The stronger the manual compression exerted on the transducer, the higher the tissue density becomes, which increases the speed of propagation of the shear waves.[27] In our study, the examiner tried to apply no additional manual compression during the SWS.

Previous studies have found that SWS might provide the most reliable material property measurement by aligning the transducer's orientation parallel to the long axis of the muscle.[28,29] In our study, the SWS was performed according to the perpendicular scan on the SCM fiber orientation due to several reasons. First, the size of our linear transducer was too large to apply the whole length of the SCM longitudinally in infants. Second, the extent of fibrotic change of the SCM at 3 different levels has a significant value in determining prognosis.[11] Third, the orientation of the ultrasound transducer on the SWS is crucial in order to obtain meaningful results in skeletal muscles.[30] Therefore, perpendicular SWS scan is useful in assessment of SCM muscle stiffness in infant. In the case of perpendicular transducer orientation

relatively to the long axis of muscle fibers, the measurement of SWV rather than shear modulus was recommended (in kilopascals).

This study has several limitations. First, a small number of infants were examined. Second, double blindness was not feasible due to the nature of the B-mode ultrasound and SWS technique. Third, we could not confirm the relationship between the pathological findings and the imaging findings. Fourth, the transducer position was perpendicular, not parallel to the long axis of the SCM due to the above-mentioned reasons. Fifth, the inter-rater reliability of the SWV was not evaluated because of the uncooperativeness of children. Last, the Young modulus of the SCM was not calculated. Further investigation with a larger number of infants is a prerequisite for validating the usefulness of SWS using ARFI in infants with CMT.

5. Conclusion

Our study demonstrated an increased SWV of the affected SCM in infants with severe PROM deficit of neck rotation despite of the similar SCM thickness on B-mode ultrasound and significant correlation between the degree of PROM deficit of neck rotation and the SWV of the affected SCM in all infants. Therefore, SWS using ARFI may be a feasible adjunctive imaging method to B-mode ultrasound to quantify the stiffness of the SCM, and is likely to help us plan the treatment in infants with CMT.

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