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Relationship between sonography of sternocleidomastoid muscle and cervical passive range of motion in infants with congenital muscular torticollis

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ABSTRACT

Background: An abnormal sternocleidomastoid muscle in congenital muscular torticollis can be classified into one of the four types via sonography. However, this categorization lacks quantitative measurements. The purpose of the study was to determine quantitative measurements of the sonograms via image analysis.

Methods: Infants younger than 12 months of age suspected of having congenital muscular torticollis were included. Intraclass correlation coefficient estimates for interobserver reliability and a simple regression analysis for criterion validity were calculated. Spearman correlation analysis was then performed. The analyzed parameters included cervical passive range of motion for lateral flexion and rotation, area, brightness, max/min Feret's diameters, and muscular width/thickness.

Results: Of the 29 (4.0 ± 2.6 months) screened infants, 13 (1.9 ± 1.7 months) were included. Nine were male, and 4 were female. Seven infants with mass were ultrasonographically classified into type I, and the other six infants were classified into type II. The affected/unaffected side ratios of cervical passive range of motion for lateral flexion and rotation were 0.92 ± 0.13 and 0.88 ± 0.16 , respectively. The parameters measured on the sonograms were reliable, and the max/min Feret's diameters were valid measurements. The affected/unaffected side ratio of cervical passive range of motion for rotation significantly correlated with the affected/unaffected side ratios of the sternocleidomastoid muscle sonogram on area ($r = -0.62, p = 0.03$) and min Feret's diameter ($r = -0.69, p = 0.01$).

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Conclusions: The area and min Feret's diameter were efficacious parameters for image analysis on sternocleidomastoid sonograms, and the min Feret's diameter would be more suitable than thickness for measuring the thickening SCM in transverse view. A healthy control group, more data and follow-up would be needed to confirm the changes on the SCM sonograms for clinical decision.

At a glance of commentary

Scientific background on the subject

Ultrasound is used to diagnose congenital muscular torticollis, and it can document severity and identify pathologic changes in affected sternocleidomastoid muscles. However, physicians generally classify sonography into one of four types based on echogenicity without quantitative measurements, resulting in variations as per physician's subjective judgment.

What this study adds to the field

This study evaluates sternocleidomastoid sonograms via image analysis. Image analysis can provide physicians with quantitative measurements on sonograms. Of four parameters, two are useful in practice, namely area and min Feret's diameter. The min Feret's diameter would be more suitable than thickness for measuring the thickening sternocleidomastoid in transverse view.

The clinical condition of torticollis seen in infants has been well recognized for centuries [1]. The incidence of torticollis is between 1.3% and 2.0% [2,3] and has even reached 15.7% in newborn infants [4]. A number of disorders can result in torticollis; however, congenital muscular torticollis (CMT), which is caused by a practically painless and contracted cordlike sternocleidomastoid (SCM) muscle, accounts for a vast majority of cases in clinical practice [5]. Approximately 81.6% of children with confirmed torticollis have a congenital muscular etiology [6], and 47.2% of infants with torticollis have a SCM mass [7].

Among various diagnostic techniques, sonography is a valuable tool to document severity and identify pathologic changes in affected SCM muscles of infants with CMT [8–11]. Abnormal SCM muscles could be classified sonographically into four types based on echogenicity [9]. The initial difference in the cervical passive range of motion (PROM) for rotation in type III infants was significantly greater than that in type I infants [11]. Nevertheless, this categorization lacks quantitative measurements, and it may be hard to distinguish between type II and normal, resulting in variations as per physician's subjective judgment.

One common way to obtain quantitative measurements on sonograms is to perform image analysis. Generally, an ultrasound machine with a built-in system has basic measurements on the two-dimensional images, including distance [12–15], circumference [12], and area [15]. Other measurements, such as shape descriptors and pixel value statistics of

user-defined selections, require a standalone image processing and analysis software program [16]. This kind of program has more measurements, such as mean gray value, centroid, bounding rectangle, shape descriptors, and Feret's diameter [17], but some of the parameters have their specific usage. For example, shape descriptors are useful when such measurement is performed for particle shape, whereas they are useless when the measurement is performed for a band.

In CMT, one of the frequently discussed measurements was SCM thickness. There were excellent interobserver agreements for the SCM thickness and the SCM thickness ratio, and the intraclass correlation coefficients were 0.92–0.99 and 0.95–0.98, respectively [13]. The SCM thickness had not only a statistically significant difference between two sides in an infant with CMT [9,14] but moderate correlation (0.43–0.49) with clinical prognosis [13], and a ratio of the thickness of the affected side (AS) to the unaffected side (UAS) for the sternal head of the SCM muscle of >1.19 showed a diagnostic sensitivity of 97.9% and specificity of 96.4% for abnormal thickening [15]. However, other measurements, such as echogenicity and cross-sectional area, were rarely discussed.

The color scale of most sonograms appears as a monotonic gray scale, so mean gray value could be used for echogenicity analysis. Studies revealed that interobserver reliabilities for echogenicity analysis were 0.84–0.97 and 0.90–0.98 for rectus femoris and diaphragm, respectively [18], and high intrater and interrater reliability for the rectus femoris echogenicity (0.99) still existed even using different software programs [19]. The advantage of mean gray value is to quantify echogenicity of region of interest on sonograms. However, this analysis needs a standard imaging procedure to minimize the effects of instrument settings and transducer positioning. To measure other features of sonograms at the same region of interest in the meantime, area, max and min Feret's diameters, and both length and width of the min area bounding rectangle, which can be set in many image analysis software products, may be useful [17]. The advantage of measuring area is to identify two-dimensional changes, but any tilting scan and contraction will increase the size of the cross-sectional area. The Feret's diameter, also called the caliper diameter, is defined as the distance between the two parallel planes restricting the object perpendicular to a specified direction. Because of the dynamic changes in the SCM muscle, max and min Feret's diameters were preferred rather than the length and width of the min area bounding the rectangle. These parameters can give different views of sonograms other than thickness.

The purpose of the study was to evaluate the result of SCM sonograms via image analysis and to identify whether mean

gray value, area, and max and min Feret's diameters were correlated to the cervical PROM in infants with CMT.

Methods

Study design and setting

This study is a cross-sectional study, and data were obtained from the department of physical medicine and rehabilitation at a regional hospital from February 1, 2013 to September 30, 2015. The protocol of the study was approved by a local institutional review board, and the clinical trial was registered at the ClinicalTrials.gov registration website (identifiers: NCT03266224).

Sample size calculation

The purpose of the study was to identify whether the measurements of the SCM sonograms via image analysis were correlated to the cervical PROM in infants with CMT, so a correlation ρ_1 of 0.75 indicating strong linear relationship was used to estimate *a priori* sample size. The minimum of 11 subjects were required for each parameter analyzed to achieve 80% power with the significance level set at $p < 0.05$ in the two-tailed test using G*Power 3.1.9.2.

Participants

Infants suspected of having wryneck at the time of the first diagnosis were screened in the regional hospital. The inclusion criteria were (1) infants with congenital muscular torticollis and (2) age < 12 months at diagnosis. Exclusion criteria were (1) wryneck caused by other known problems, including strabismus, trauma, neurogenic problems, congenital malformation, or bony deformity (e.g., hemivertebrae of the cervical spine), (2) ultrasonographically normal SCM muscles of both sides, or (3) either difference between both sides or a deficit in cervical PROM $\leq 5^\circ$. Infants whose parents signed informed consent approved by the local institutional review board were enrolled.

Demographics and clinical data

Basic characteristics, including gender, age at diagnosis, and side of wryneck, were collected in a survey questionnaire. Other variables that may affect the ultrasonography of the SCM muscle were also obtained in the questionnaire, such as birth weight, gestational age, neck mass, plagiocephaly, and facial asymmetry.

Cervical passive range of motion measurement

The cervical PROM, the primary outcome measure, of the infant lying in the supine position on a therapy table with the shoulders stabilized and the head in the cervical neutral position was measured with an arthrodiagonal protractor. Because the lateral flexion and rotation of neck had sufficient intra-rater reliability (0.87–0.97) and fair to good criterion validity (0.74–0.90) [20], and the construct validity for severity of CMT [11,21–23] was also existed, only lateral flexion and rotation were measured in the

study. In the study, the rotation of cervical ROM for AS was defined as rotation of cervical ROM toward to AS, but the lateral flexion of cervical ROM for AS was measured by lateral flexion of cervical ROM toward to UAS. The head of the infant was gently rotated to the UAS and then rotated to the AS, ending at the first palpable sign of resistance or at normal degrees. After the rotations, lateral flexion was measured starting with the AS to avoid irritating the infant. The cervical PROM for lateral flexion of 70° was used as the normal reference [24], but rotation of 90° was used instead although an excellent value of 110° could be reached. Cervical neutral was maintained for the measurement through the range. If the infant got irritated, the measurement would be paused until the infant calmed down. A squeaky toy would be played if needed. There were two well-experienced physiotherapists participating in the study, and one infant was evaluated only by one physiotherapist three times according the above standard procedure. Mean value was used for the following statistics.

Sonographic examination

At the time of enrollment, each side of the infant's SCM muscle with head slightly rotated to the opposite side was examined in the supine position using a LOGIQ[®] 9 (General Electric Company, Milwaukee, Wisconsin, USA) with a 14-MHz linear-array transducer for comparison of the lesion side and sound side. A large

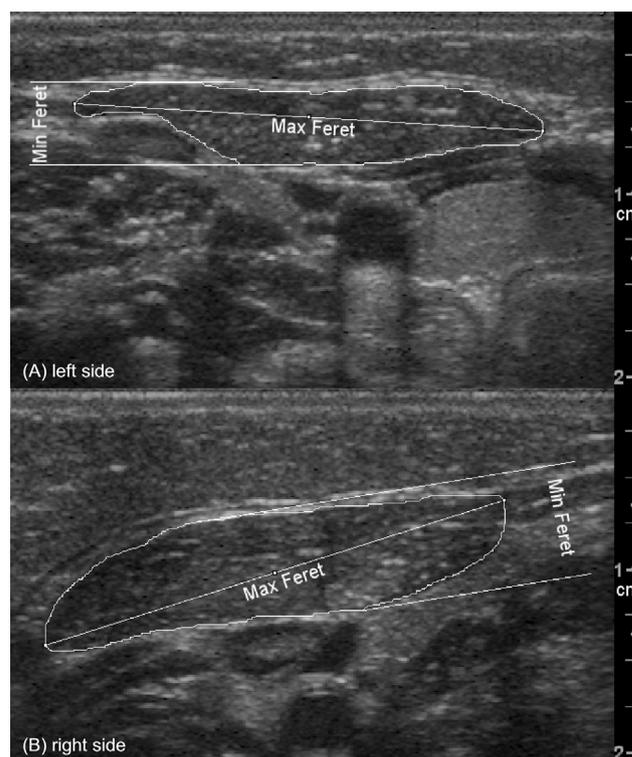


Fig. 1 The region of interest in transverse SCM sonograms of the infant with right-sided CMT in the study. Illustrations of (A) left side and (B) right side both showed max and min Feret's diameters after image analysis [17]. The region of interest on the sonograms was selected via the freehand method.

amount of ultrasound transmission gel was used to let waves transmit directly to tissues beneath and to minimize pressure on the neck. The transducer aligned to the long axis of the neck was gently placed in contact with the skin of the neck and then moved to search the lesion part of the SCM muscle. When the lesion part was positioned, the transducer was rotated 90° then, and the largest cross-section was captured. If no lesion part was found, the largest cross-section was captured at the middle part of the SCM muscle. The sound side was then examined according to the above procedure. If the infant was tense and uncooperative, the scanning was paused. A squeaky toy was played to make the infant calm down if needed. There were four well-experienced physicians participating in the study, and one infant was examined only by one physician according to the above standard procedure.

Image analysis

The sonogram of the SCM muscle was analyzed with ImageJ 1.46r (National Institute of Health, Bethesda, Maryland, USA). The region of interest on the sonograms was selected via the freehand method and is shown in Fig. 1. The area, both max and min Feret's diameters, and mean gray value were set and automatically recorded after image analysis. The area and max and min Feret's diameters were measured in pixels on the original image. Thus, dividing by the pixels of one centimeter or one square centimeter on each sonogram, the unit of the max and min Feret's diameters was converted to centimeters, and that of the area was converted to square centimeters. In addition, the sonograms were grayscale images, and the mean gray value was measured from 0 to 255 in the image analysis software where zero was designated black and 255 was designated white. Therefore, the mean gray value was divided by 255 and then converted to a percentage. In this study, zero percent was designated black, and 100% was designated white and indicated the brightness of the echogenicity on the sonogram. Additionally, the maximal anterior-posterior diameter, the muscular thickness, was measured via the straight line method. Then, the muscular width aligned perpendicularly to the muscular thickness was measured, too. All sonograms of the infants were measured two times with 2-week interval by two trained assistants according to the above standard procedure.

Statistical methods

SAS 9.0 (SAS Institute Inc., Cary, North Carolina, USA) was used for all the statistical analyses in the study. Intraclass correlation coefficient (ICC) estimates were calculated based on a mean-rating ($k = 2$) and the two-way random-effects model for interobserver reliability. ICC values less than 0.5 were indicative of poor reliability, values between 0.5 and 0.75 indicated moderate reliability, values between 0.75 and 0.9 indicated good reliability, and values greater than 0.9 indicated excellent reliability [25]. A simple regression analysis was performed to estimate criterion validity between max/min Feret's diameters and muscular width/thickness. To eliminate differences in infants' growth and development, calculating a ratio of each measurement in the same procedure was a practical method. A ratio of each measurement was defined as the ratio of the AS to the UAS. The Shapiro–Wilk

Table 1 Demographics and clinical data of the study population.

Characteristic	N or Mean \pm SD
Gender, female/male	4/9
Age at diagnosis, months	1.9 \pm 1.7
Birth weight, g	2992.7 \pm 469.6
Gestational age, weeks	38.3 \pm 1.5
Side of wryneck, right/left	6/7
Neck mass	7
Plagiocephaly	8
Facial asymmetry	6
Ultrasonographic type, type I/II ^a	7/6

Abbreviation: SD: standard deviation.

^a Type I denoted a heteroechoic mass in the affected muscles; type II represented diffuse echogenic dots and lines against the hypoechoic background with no detectable mass [28].

test was used to assess the normality of distribution of investigated parameters. Because the AS/UAS ratio of rotational PROM and the AS/UAS ratios of brightness, min Feret's diameter, and thickness of SCM were not distributed normally, the Spearman correlation analysis between the AS/UAS ratio of the cervical PROM and the AS/UAS ratio of the SCM sonogram was performed in the study to investigate whether the cervical PROM was related to the SCM sonogram of an infant with CMT at the time of the first diagnosis. For all regressions and correlations, $p < 0.05$ was considered statistically significant.

Results

Participants and clinical features

In the present study, 29 infants (4.0 ± 2.6 months) suspected of having torticollis were screened. Eight infants with ultrasonographically normal SCM muscles of both sides and 8 infants with either difference between both sides or a deficit in

Table 2 Mean of cervical PROM and SCM sonogram in the study population.

Characteristic	AS	UAS	AS/UAS Ratio
Cervical PROM			
Lateral flexion, degrees	60.9 \pm 8.6	66.3 \pm 6.0	0.92 \pm 0.13
Rotation, degrees	75.5 \pm 14.8	85.7 \pm 5.3	0.88 \pm 0.16
SCM sonogram			
Area, cm ²	1.30 \pm 0.47	1.10 \pm 0.19	1.24 \pm 0.52
Brightness, %	24.43 \pm 7.51	24.58 \pm 6.73	0.99 \pm 0.14
Max Feret's diameter, cm	2.53 \pm 0.43	2.54 \pm 0.33	1.00 \pm 0.10
Min Feret's diameter, cm	0.73 \pm 0.21	0.63 \pm 0.12	1.21 \pm 0.45
Width, cm	2.33 \pm 0.42	2.35 \pm 0.35	1.00 \pm 0.15
Thickness, cm	0.70 \pm 0.21	0.58 \pm 0.13	1.26 \pm 0.46

Abbreviations: PROM: passive range of motion; SCM: sternocleidomastoid; AS: affected side; UAS: unaffected side. Data were mean \pm standard deviation. N = 13.

Table 3 Reliability of SCM sonogram of the study population measured two times with 2-week interval.

Characteristic	AS	UAS
Area	0.88	0.73
Brightness	0.99	0.98
Max Feret's diameter	0.81	0.70
Min Feret's diameter	0.98	0.95
Width	0.78	0.90
Thickness	0.71	0.99

Abbreviations: SCM: sternocleidomastoid; AS: affected side; UAS: unaffected side. Data were calculated based on a mean-rating ($k = 2$) and the two-way random-effects model, ICC (2, k). $N = 13$. Two repeated measurements were done by 2 trained assistants.

Table 4 Criterion validity between max/min Feret's diameters and muscular width/thickness of SCM sonogram of the study population.

Dependent/Independent variables	Side	Intercept	Slope	r^2	p
Max Feret's diameter/Width	AS	0.38	0.93	0.81	<0.001
	UAS	0.62	0.82	0.76	<0.001
Min Feret's diameter/Thickness	AS	0.12	0.88	0.74	<0.001
	UAS	0.09	0.94	0.94	<0.001

Abbreviations: SCM: sternocleidomastoid; AS: affected side; UAS: unaffected side. The validity was performed using simple regression analysis. $N = 13$.

Table 5 Correlation between AS/UAS ratios of cervical PROM and SCM sonogram of the study population.

AS/UAS Ratio of SCM Sonogram	AS/UAS Ratio of Cervical PROM			
	Lateral flexion		Rotation	
	r	p	r	p
Area	-0.22	0.47	-0.62	0.03
Brightness	-0.31	0.30	0.04	0.90
Max Feret's diameter	0.01	0.97	0.07	0.81
Min Feret's diameter	-0.15	0.61	-0.69	0.01
Width	-0.14	0.65	-0.48	0.10
Thickness	-0.30	0.32	-0.55	0.05

Abbreviations: PROM: passive range of motion; SCM: sternocleidomastoid; AS: affected side; UAS: unaffected side. The correlation was performed using Spearman correlation test. $N = 13$.

cervical PROM $\leq 5^\circ$ were excluded. Thirteen infants (1.9 ± 1.7 months) were enrolled and analyzed. Nine of the 13 infants were male, and 4 were female. Seven infants had mass at the AS and were ultrasonographically classified into type I, and the other six infants were ultrasonographically classified into type II [Table 1]. Eight had plagiocephaly, and 6 had facial asymmetry.

Cervical PROM and SCM sonogram

The infants with CMT had cervical limitation for lateral flexion ($60.9 \pm 8.6^\circ$) and rotation ($75.5 \pm 14.8^\circ$). The AS/UAS

ratios of cervical PROM for lateral flexion and rotation were 0.92 ± 0.13 and 0.88 ± 0.16 , respectively [Table 2]. The SCM sonogram of the study population measured two times with 2-week interval had moderate to excellent reliability (0.70–0.99) [Table 3]. The area, min Feret's diameter, and thickness at the AS were higher than those at the UAS. The AS/UAS ratios of area, min Feret's diameter, and thickness were 1.24 ± 0.52 , 1.21 ± 0.45 , and 1.26 ± 0.46 , respectively. Criterion validity demonstrated good agreement (slope = 0.88 to 0.94) between the min Feret's diameter and the thickness [Table 4]. The AS/UAS ratio of cervical PROM for rotation significantly correlated with the AS/UAS ratios of the SCM sonogram on area ($r = -0.62$, $p = 0.03$) and min Feret's diameter ($r = -0.69$, $p = 0.01$) [Table 5].

Discussion

The study revealed that area and min Feret's diameter were useful in image analysis and could give different views of sonograms other than thickness. Furthermore, the result showed that the AS/UAS ratio of the min Feret's diameter had higher correlation ($r = -0.69$, $p = 0.01$) with the AS/UAS ratio of the cervical PROM for rotation than the AS/UAS ratio of the thickness did ($r = -0.55$, $p = 0.05$) [Table 5]. It implied that the min Feret's diameter would be more suitable than thickness for measuring the thickening SCM in transverse view.

Nowadays, there were some semi-automatic or fully automatic image segmentation methods to improve efficiency of delineating the boundaries of an object or tissue regions [26,27]. By combination with suitable segmentation methods, the region of interest could be selected and then automatically analyzed [27]. The data of area and min Feret's diameter on the SCM sonograms could be collected more easily. It would enhance our understanding the clinical changes of the SCM in infants with CMT.

The study was the first to measure the min Feret's diameter of the SCM in transverse view, and this measurement was reliable (ICC = 0.95–0.98) [Table 3] and had good agreement (slope = 0.88 to 0.94) with the traditional measurement of the thickness [Table 4]. For measuring the thickness of muscles on longitudinal ultrasound images, there was another suitable method to achieve a 100% automatic segmentation success rate and a excellent reliability (ICC = 0.98–0.99) with mean differences between the automatic and manual measurements in the range of 0.006–0.045 cm [27].

Although more severe rotational deficits would be accompanied by a higher proportion of hyperechogenicity [10], the AS/UAS ratio of the brightness was not correlated to the AS/UAS ratio of the cervical PROM for rotation ($r = 0.04$, $p = 0.90$) in the study [Table 5]. It might be due to only type I and type II SCM fibrosis of the infants with CMT enrolled and type III and type IV of cases absent. Studies showed that increased echogenicity without hypoechoic background happened in the type III and type IV [9], the mean age of the patients with type III were 4.4 ± 3.9 years old [28], and few infants younger than one year had type III or type IV fibrosis [9,11]. Hence, the inclusion criterion of age <12 months at

diagnosis and small sample size were the limitation in the study. Moreover, the study lacking a healthy control group was another potential limitation. In addition, the two well-experienced physiotherapists and the four well-experienced physicians examined these infants according to the standard protocol, but the observer bias might be considered.

In further research, wider age distribution would be considered to confirm the efficiency of brightness, or a longitudinal follow-up might be needed to understand the change of brightness on the SCM sonograms of infants with CMT. A healthy control group might be helpful to determine normal reference values for these parameters and to make clinical diagnosis.

Conclusions

This study clearly revealed that the area and min Feret's diameter were efficacious parameters for image analysis on the SCM sonograms, and the min Feret's diameter would be more suitable than thickness for measuring the thickening SCM in transverse view. They provided clinicians and researchers with both objective and quantitative measurements. Nevertheless, a healthy control group, more data and follow-up would be needed to confirm the changes on the SCM sonograms for clinical decision.

Conflicts of interest

The authors have no conflicts of interest relevant to this article.

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Appendix A. Supplementary data

Supplementary data to this article can be found online at <https://doi.org/10.1016/j.bj.2018.10.001>.

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