Research Paper

Meta-Analysis of Diagnostic Value of HSY5-Ultrasound in Children with Muscular Torticollis

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Hu *et al.*: Clinical Utility of Ultrasound Elastography

To investigate the potential clinical utility of ultrasound elastography in the diagnosis and management of infants with congenital muscular torticollis. We conducted searches in PubMed, Embase, Web of Science and Cochrane library databases and limited the search time from the database was created in April 2023. Articles were screened according to inclusion and exclusion criteria. RevMan (version5.3) is used for meta-analysis. The study results were presented using either standard mean difference or mean difference as statistical measures to compare the difference between two groups and 95 % confidence intervals for consecutive outcomes. Chi-square test was used to calculate I^2 to detect the heterogeneity between studies and then appropriate effect models were selected according to I^2 or p values. Publication offset was evaluated using funnel plots. Of the 5 studies included. The years of publication ranged from 2013 to 2021, with sample sizes ranging from 20 to 120. Ultrasonic measurements of sternocleidomastoid muscle thickness (mean difference was 0.57, 95 % confidence intervals was -0.42-0.72, $p<0.001$). The A/U ratio of sternocleidomastoid muscle measured by ultrasound was (mean difference was -0.45, 95 % confidence interval was -1.56-0.66, $p=0.42$). Ultrasound imaging may be a useful adjunct to the evaluation of B ultrasound in sternocleidomastoid muscle infants. However, further high quality clinical trial studies are needed to obtain stronger clinical evidence.

Key words: Sternocleidomastoid muscle, computed tomography, muscular torticollis, ultrasound

Congenital Muscular Torticollis (CMT) is a common cervical deformity in children, mainly caused by Sternocleidomastoid Muscle (SCM) contracture, which is typically characterized by a tilted head to the affected side and rotation of the mandible to the healthy side. Currently, the diagnosis of CMT mainly depends on history, physical examination, and imaging[1-3]. However, traditional imaging methods such as X-ray and Computed Tomography (CT) have problems such as high radiation dose, failure to assess soft tissue elasticity and failure to fully assess the extent of lesions in SCM[4-6]. Clinically, the lesion hardness of CMT is positively correlated with fibrosis, and the accurate evaluation of the lesion degree of CMT is very important for the judgment of treatment and prognosis. A non-invasive, low-risk, accurate and repeatable diagnostic method is of great significance for the diagnosis and treatment of pediatric muscular torticollis[7-10].

Traditional imaging methods, such as X-ray and CT, have problems such as high radiation dose, failure to evaluate soft tissue elasticity, and failure to comprehensively evaluate the degree of lesions in SCM[11]. Therefore, it is of great significance to develop a low-cost, noninvasive and repeatable diagnostic method for the diagnosis and treatment of pediatric muscular torticollis[12]. In recent years, ultrasound technology has attracted wide attention as a low-cost, non-invasive and reproducible imaging method[13-16]. It can provide more comprehensive and accurate diagnostic information. In recent years, some studies have shown that Virtual Touch Tissue Imaging Quantization (VTIQ) technique has high accuracy and reliability in the diagnosis of pediatric muscular torticollis, which has important clinical application value in guiding the treatment and monitoring the efficacy of pediatric muscular torticollis. The purpose of this meta-analysis was to evaluate the accuracy and reliability of ultrasound in

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the diagnosis of pediatric muscular torticollis, and to explore its clinical application value. In this context, this paper will systematically comb the existing relevant literature, and summarize and analyze the performance of ultrasonic technology in the diagnosis of pediatric muscular torticollis, in order to provide clinicians with more accurate and reliable diagnostic methods, and provide better guidance for the treatment of pediatric muscular torticollis.

MATERIALS AND METHODS

Search strategy:

We searched the keywords related to "ultrasound", "muscular torticollis" and "children" in PubMed, Embase, Web of Science and Cochrane Library databases. The search time was limited from the establishment of the database to April 2023.

Qualification criteria:

Research types: Only observational studies and Randomized Controlled Trials (RCTs) on ultrasonic diagnosis of pediatric muscular torticollis were included. Non-RCTs, case reports and animal studies, and reviews were excluded.

This statement discusses the inclusion and exclusion criteria for a study on pediatric muscular torticollis in individuals aged 0-18 y old. The study includes children with this diagnosis but excludes those with torticollis caused by other medical conditions.

Intervention: This meta-analysis only focused on the diagnostic value of ultrasound technology without limiting intervention measures.

Comparison: In this meta-analysis, no control group interventions were involved and any therapeutic interventions were excluded.

Result measurement: The primary outcome was ultrasound diagnosis of pediatric muscular torticollis. The thickness and stress ratio of the mass in the SCM muscle was evaluated.

Research selection and data extraction:

Research options: When the authors searched the relevant literature in PubMed, Embase, Web of Science, Cochrane Library and other databases, the title and abstract were first screened and the full text was finally screened. Screening criteria included, RCTs or controlled clinical trials; the objective of the study was to evaluate the diagnostic value of ultrasound in pediatric muscular torticollis; data of control group and intervention group were included and provide adequate data. Evaluation of the thickness and stress ratio of the mass in the SCM muscle in the diagnosis of pediatric muscular torticollis by ultrasound. Two evaluators independently performed the screening process and cross-checked against the selection criteria. Differences between the two reviewers are resolved through negotiation, but if consensus is still lacking, a third reviewer is involved to reach an agreement.

Data extraction: Two evaluators independently extracted the publication year, first author name, publication date, study country and sample size of the included articles. Patient characteristics such as age, gender, intervention, methods of ultrasound examination (e.g., type of ultrasound probe, scanning mode, etc.) and results shown by ultrasound (e.g., thickness and stress ratio assessment of mass in SCM muscle, etc.) and ultrasound results. Thickness and stress ratio of mass in SCM muscle in case of differences in data extraction results, consensus is reached through discussion among all authors. All data will be summarized and analyzed to calculate the diagnostic value of ultrasound in pediatric muscular torticollis.

Risk assessment:

The risk assessment method of meta-analysis can be referred to the Cochrane bias risk tool. It mainly includes generating random sequence, including deviation factors, such as generating random sequence, hiding, defining blind method and incomplete result information and other biases. For migration risk assessment, it can be divided into the following aspects; migration type, data source selection, sample selection and processing, data analysis and result presentation. Each aspect can also be classified by risk level as low, unclear or high. The evaluation can be conducted independently by multiple authors and differences can be resolved through discussion. Finally, all evaluation results should be summarized to obtain the overall bias risk assessment results of meta-analysis, and possible bias factors should be discussed and explained in the results to improve the credibility and reliability of the study.

Statistical analysis:

ReviewManager (RevMan) version5.3 is used for meta-analysis. The data were summarized using Standardized Mean Difference (SMD) or Mean Difference (MD) and 95 % Confidence Intervals (CI) for consecutive outcomes. Chi-square test was used to calculate I^2 to detect the heterogeneity between studies, and then appropriate effect models were selected according to I^2 or p values. When $I^2>50$ % or $p<0.05$, indicating the possibility of significant heterogeneity, random effects model was used; otherwise, fixed effects model was used. We then assessed publication bias by observing the symmetry of the funnel plot. $p>0.05$ was considered to indicate the absence of publication bias and the results were presented in the form of pictures.

RESULTS AND DISCUSSION

Using the prepared search strategy, a search was conducted in each database and a total of 534 articles were obtained after the selected study. By screening the titles or abstracts of all articles and the inclusion criteria, 84 studies that could not be included, such as repeated reports 379, conference abstracts, reviews and case reports were excluded,and 60 literatures that could not obtain the full text were excluded. Finally, it was determined that the included 5 studies met the inclusion criteria by browsing the full text. A total of 280 patients were included[11-19] (fig. 1). Of the 5 studies included[17-21], 3 were RCTs and 2 were retrospective studies, 4 were conducted in Korea and 1 was conducted in China. The years of publication ranged from 2013 to 2021, with sample sizes ranging from 20 to 120.The main features of these studies are summarized in Table 1.

Five studies evaluated ultrasonography measures of SCM muscle thickness involving 442 patients. In five studies, the intergroup difference in favor of the experimental group was reported as $p<1.2$ in one study, $p<0.1$, and the final meta results showed that ultrasonic measurements of SCM muscle thickness were (MD: 0.57, 95 % CI: -0.42 to 0.72, $p<0.001$) with moderate heterogeneity ($p=0.8$, $I^2=53$ %, fig. 2).

A total of three articles involved ultrasound testing of patients with neck A/U ratio including a total of 94 patients. Forest map results showed that A/U ratio of SCM muscle measured by ultrasonography was (MD: -0.45, 95 % CI: -1.56 to 0.66, $p=0.42$), which had no statistical significance and no heterogeneity ($I^2=0$ %, $p=0.83$), as shown in fig. 3.

For SCM muscle thickness and A/U ratio, SMDS and their associated 95 % CI were combined and analyzed by funnel plot test to assess publication bias. The results are shown in fig. 4a and fig. 4b. Funnel plots of SCM muscle thickness and A/U ratio showed good symmetry.

The meta-analysis was deemed dependable and strong as removing any literature did not considerably alter the results in the sensitivity analysis. Heterogeneity was detected in the meta-analysis results for SCM thickness, with an I^2 of over 50 %, but after excluding the (MI/2020) study, the I^2 decreased to 28 % as shown in fig. 5.

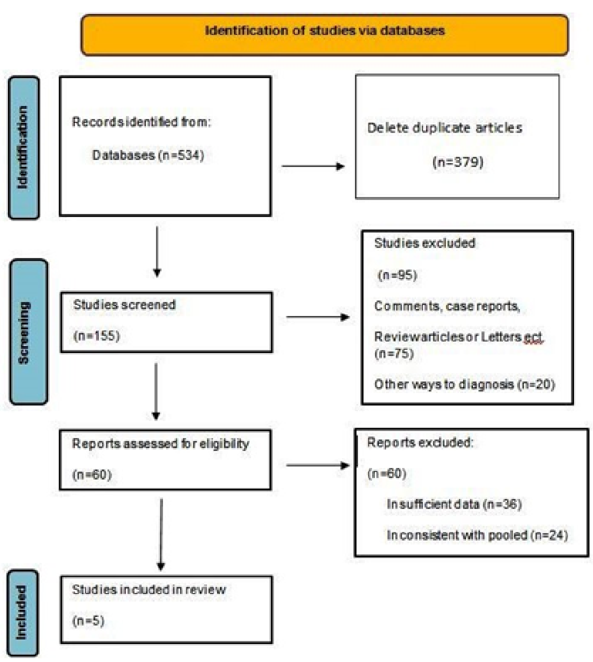

Fig. 1: Flow chart for selection and inclusion of eligible studies

TABLE 1: BASELINE FEATURES INCLUDED IN THE STUDY

Study	Year	Country	Number of cases (experimental/ control group)	Mean age	Male/female
Seong	2016	South Korea	29/24	30.92±21.72	35,18
Mi	2020	South Korea	16041	23.65	50,5
Hyeng	2013	South Korea	67/66	24.21	81,52
Zhang	2021	China	9,10	13.1±77.8	11,8
Gi	2018	South Korea	11,9	69.0±26.2	12,8

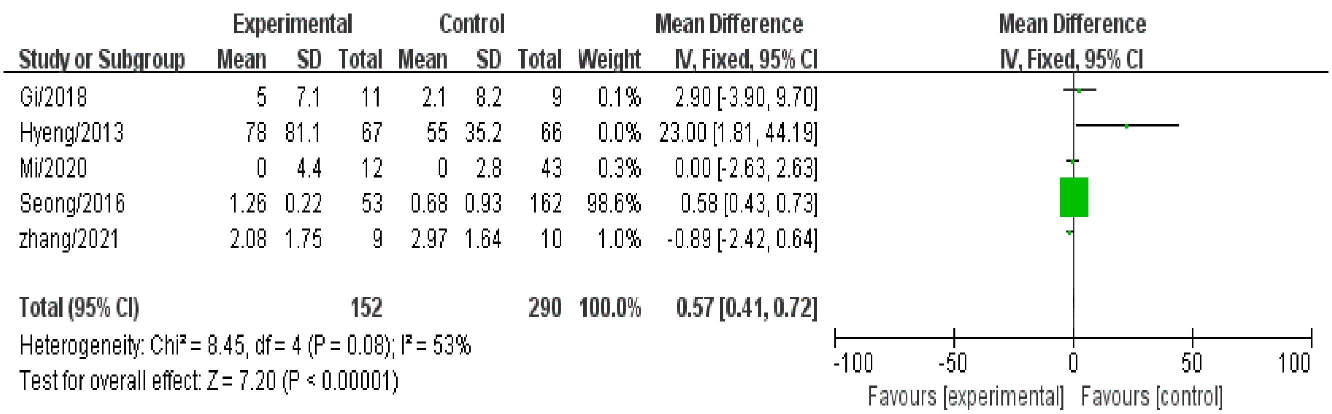

Fig. 2: Forest map of ultrasonic measurement of SMD thickness

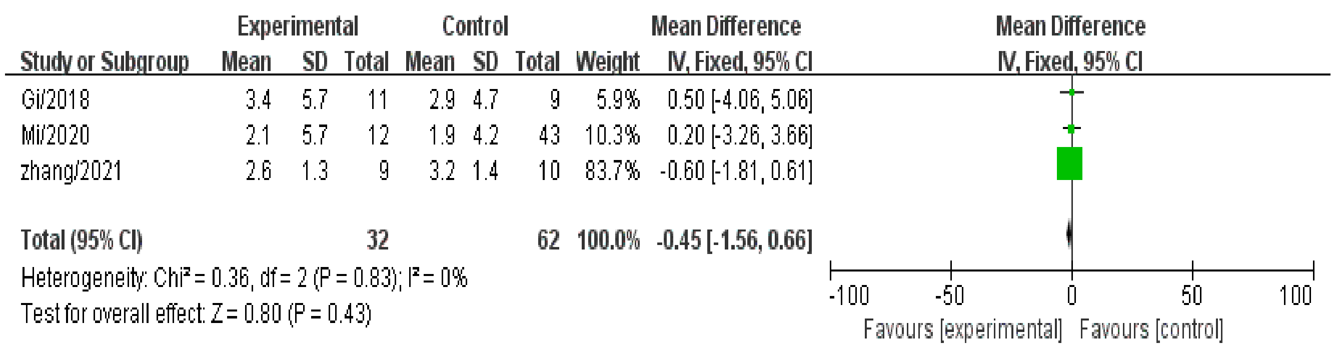

Fig. 3: Forest map of A/U ratio tested by ultrasound

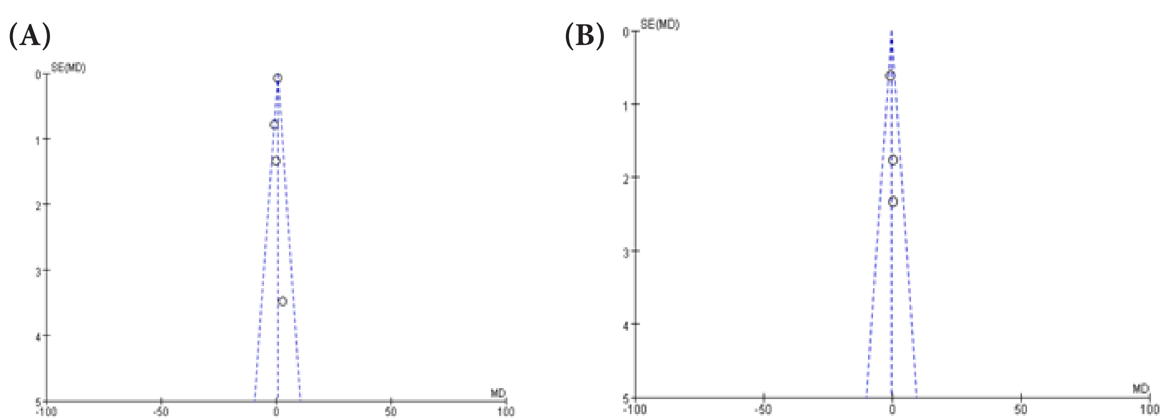

Fig. 4: Funnel plot of (a): SCM muscle thickness and (b): A/U ratio

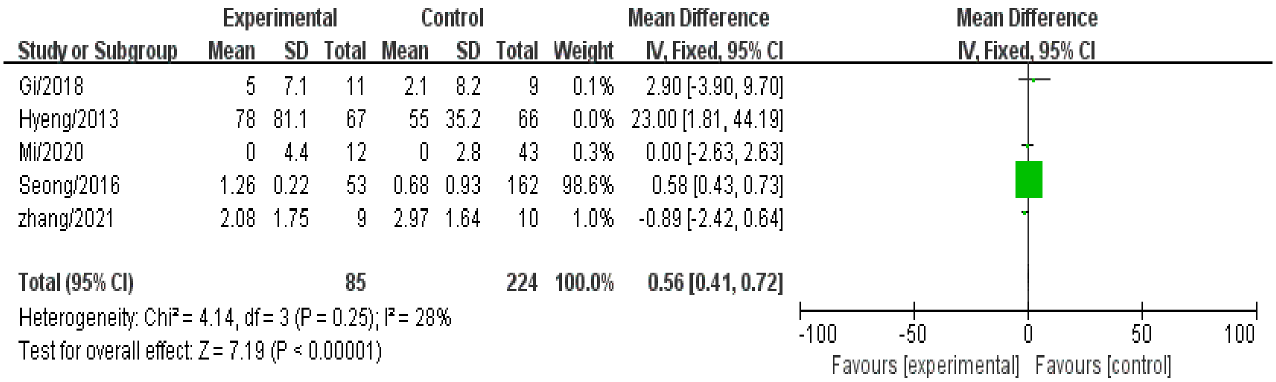

Fig. 5: Meta-analysis results for SCM thickness

CMT is a common childhood neuromuscular disease. The main manifestations of the children are head tilt to the affected side after birth, hard tumor-like mass in the involved SCM[22-24], and crooked or twisted neck. If not treated in time, the patient may have head tilt, facial asymmetry and other symptoms, leading to other secondary deformities[25]. It may affect children's life and study. Therefore, early diagnosis and treatment are very important. Ultrasound is a non-invasive, non-radioactive imaging method that can detect changes in neck muscles and nerves[26-28]. It is the primary diagnostic tool for CMT, which can detect the thickness of SCM and muscle fiber thickening. In this paper, a meta-analysis was conducted on the diagnostic value of ultrasound in pediatric muscular torticollis, and its clinical significance[29]. Limitations and suggestions for further research discusses the use of VTIQ technology for diagnosing and monitoring children with CMT[30-33]. VTIQ is a real-time elastography technique that measures tissue elasticity in response to back pressure. The study found that the Square Wave Voltammetry (SWV) of the affected side in children with CMT was significantly greater than that of the healthy side, and the VTIQ technique can provide a more objective and accurate means of diagnosis. Furthermore, the mean SWV was positively correlated with head deviation and muscle hardness, suggesting that VTIQ can also evaluate the effectiveness of treatment for CMT[34-37].

The results of this study showed that ultrasonic measurements of SCM muscle thickness (MD: 0.57, 95 % CI: -0.42 to 0.72, p<0.001). A/U ratio of the SCM muscle measured by ultrasound was (MD: -0.45, 95 % CI: -1.56-0.66, p=0.42). The affected SCM mass grows in response to environmental changes after birth, undergoes differentiation and maturation, and then produces CMT. In this study, no evidence of neck or SCM injury was found in the physical examination screening of 442 newborns. In addition, ultrasound examination found no evidence of neck injury in infants with CMT. This raises questions about the quality of ultrasonic SCM. Overall, our results provide new insights into ultrasonic inspection SCM. These findings may help improve ultrasound diagnosis and treatment of SCM, thereby helping to reduce patient suffering and improve quality of life.

There are some limitations to this meta-analysis. First, due to resource and time constraints, ultrasound cannot be performed on every child with pediatric muscular torticollis and some patients may be missed. Secondly, because some patients or their parents do not comply with the study protocol or are difficult to follow, some data may be missing or follow-up may be lost. In addition, as this meta-analysis is a retrospective study, there may be some selection bias, reporting bias or other possible bias, which may affect the accuracy of the conclusions. In addition, the diagnostic criteria for muscular torticollis may vary, which may also influence the results. Therefore, these limitations need to be noted when interpreting the results.

In conclusion, ultrasound imaging may be a useful adjunct to the evaluation of B-type ultrasound in SCM infants, especially in predicting the recovery outcome of SCM infants.

Conflict of interests:

The authors declared no conflict of interests.

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