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A Clinical Comparative Study of Ultrasound-Normal Versus Ultrasound-Abnormal Congenital Muscular Torticollis

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Abstract

Objective: To investigate the clinical features and outcome of outpatient-based physiotherapy (manual stretch) of congenital muscular torticollis (CMT) with passive neck motion limitation ($\geq 10^{\circ}$) according to whether the finding on ultrasonography (US) is normal or abnormal.

Design: Case-control study.

Setting: Institutional practice.

Participants: A total of 149 patients with CMT who met eligibility criteria were included: age at presentation ≤ 6 months, limitation of passive neck rotation (Δ ROT) or lateral flexion (Δ LAT) $\geq 10^{\circ}$, and completion of our outpatient-based physiotherapy program.

Interventions: Patients were allocated to the US-normal or US-abnormal group. Patients underwent physiotherapy and were followed-up monthly until Δ ROT and Δ LAT were $\leq 5^{\circ}$ or did not respond to treatment.

Main Outcome Measurements: Baseline characteristics, initial Δ ROT and Δ LAT, age at presentation, treatment durations, and success rates of physiotherapy were compared between 2 groups. Treatment duration was adjusted for initial Δ ROT, Δ LAT, and age at presentation using analysis of covariance.

Results: Mean initial \triangle ROT and \triangle LAT in US-abnormal (28.5°; 17.0°) were greater than in US-normal (7.9°, P < .001; 12.3°, P = .001, respectively). Mean age at presentation was older in US-normal (3.8 months) than in US-abnormal (1.8 months, P < .001). Treatment duration was shorter in the US-normal (5.1 weeks) than US-abnormal (14.9 weeks, P < .001). Adjusted treatment duration was also shorter in US-normal (9.7 weeks) than US-abnormal (13.8 weeks, P < .05). The success rates of physiotherapy were 95% in US-abnormal and 100% in US-normal. Two of 6 treatment failures in the US-abnormal group underwent surgery.

Conclusions: In CMT with passive neck motion limitation ($\geq 10^{\circ}$), patients in the US-normal group demonstrated lesser passive neck motion limitation and older age at presentation than US-abnormal. It seems that US-normal showed shorter treatment duration irrespective of severity of neck motion limitation and age at presentation. Additionally, manual stretching applied before 6 months of age appears to show generally good outcome regardless of US findings.

Introduction

Congenital muscular torticollis (CMT) is a common disorder in neonates and infants with a tumor or tightness of the sternocleidomastoid muscle (SCM) caused by fibrotic changes [1-4]. Numerous theories have been proposed, but the true etiology of CMT remains unclear. Various causes implicated for CMT include intrauterine crowding or vascular phenomenon, fibrosis from peripartum bleeds, compartment syndrome, primary myopathy of the SCM, and traumatic delivery [5]. The main clinical findings of CMT include a characteristic head tilt, passive neck motion limitation, and/or a palpable neck tumor [6-8]. In the literature, patients with CMT have been classified on the basis of clinical findings into a SCM tumor group (those with a clinical tumor in SCM) or a muscular torticollis group (those with tightness of the SCM but no clinical tumor) [7,9].

It is well known that clinical diagnosis group, initial limitation of passive neck motion, and age at

presentation are the most important predictors of the outcome of physiotherapy [7]; however, the clinical diagnosis is not always accurate. Thus, high-resolution ultrasonography (US) increasingly has received attention because of its diagnostic abilities to demonstrate a tumor or fibrotic lesion in the SCM [10-14]. In terms of CMT with abnormal US findings, several studies have been conducted on the relationship between clinical features of CMT and relevant US abnormalities, which have found a close association of the severity of abnormal US findings with treatment course or prognosis [13-17]. In particular, more severe fibrosis of the SCM on US is associated with greater neck motion limitation, longer treatment duration, and more likelihood of need for surgical intervention. In terms of CMT with normal US finding, one study by Tatli et al [12] reported that patients without a palpable mass and with normal US findings showed better conservative treatment outcomes than those with a clinically palpable tumor and any relevant US abnormalities of the SCM; however, they did not describe the severity of initial passive neck motion limitation before treatment and did not suggest protocol of the physiotherapy program, which also are associated intimately with the outcome of physiotherapy [18-20]. Not uncommonly, we find US-normal CMT with passive neck motion limitation (difference of passive neck motion between both sides) $>10^{\circ}$ in routine clinical practice, who are considered to need outpatient-based physiotherapy on the basis of previous studies [7,15,20]; however, we still don't have much information about such patients. To our knowledge, no previous study has addressed patients with normal US finding but whose clinical finding shows clinical features of CMT considered to need outpatient-based physiotherapy: passive neck motion limitation $\geq 10^{\circ}$. Thus, the present study was undertaken to investigate the clinical features and outcome of outpatient-based physiotherapy (manual stretching program) in US-normal CMT group whose passive neck motion limitation were more than 10° and to conduct a comparative analysis with an US-abnormal CMT group whose passive neck motion limitation were more than 10° .

Material and Methods

Subjects

A total of 318 patients with clinically suspected CMT visited the torticollis clinic at our hospital from 2007 to the first half of 2013 (boys: 182, girls: 136). Of these, a consecutive series of 149 infants (boys: 72, girls: 77) who met all study inclusion criteria were included in the study. These criteria were as follows: (1) an age younger than 6 months; (2) limitation of passive neck rotation (Δ ROT) or lateral flexion (Δ LAT) $\geq 10^{\circ}$; and (3) completion of our outpatient-based physiotherapy program. Δ ROT or Δ LAT was defined as difference of passive neck

motion (rotation or lateral flexion) between both sides. In addition, the following exclusion criteria were applied: (1) congenital anomaly of the cervical spine; (2) ocular torticollis; and (3) neurogenic or spasmodic torticollis. All patients underwent plain spine radiography. An ophthalmologist was consulted when an ocular problem was suspected. A total of 162 patients that visited our torticollis clinic did not meet the study inclusion criteria. Of these patients, 27 patients presented after the age of 6 months and 36 patients showed both $\triangle ROT$ and $\triangle LAT < 10^{\circ}$. Forty patients did not undergo our physiotherapy program for various reasons, such as financial problems or far distance from home. The data of 59 patients were missing at follow-up. In addition, 7 patients were excluded for strabismus (2 patients), which was diagnosed by an ophthalmologist, and for other diseases (5 patients), such as, lymphadenitis, tic disorder, or lipoma. The study was approved by our institutional ethics review board, which waived the requirement for informed consent because of the retrospective study design.

US Evaluation and Classification

The 149 patients were divided into the US-abnormal group and US-normal group on the basis of US findings. The US-normal group had normal findings on US but showed clinical features of CMT that were considered to need outpatient-based physiotherapy (passive neck motion limitation $>10^\circ$) (Figure 1A). The US-abnormal group also had clinical features of CMT considered to need outpatient-based physiotherapy (passive neck motion limitation $>10^\circ$) and had any relevant US abnormalities of the SCM, such as tumor, hypertrophy, heterogeneous echotexture, and hyperechoic lesion in SCM muscle (Figure 1B-D). US evaluations were performed by 2 radiologists and 1 physiatrist using a HDI 5000 (Philips Medical Systems, Bothell, WA), a Voluson 730 (GE Healthcare, Zipf, Austria) or a LogiqE9 (GE Healthcare, Milwaukee, WI) equipped with a broadband linear-array transducer.

Physiotherapy Protocol

Physiotherapy was conducted via a standardized protocol (manual stretching exercise for about 30 minutes 3 times per week) modified from Cheng et al [7,15,20], which was performed by an experienced physical therapist a few days after diagnosis of CMT for all patients with a Δ ROT or a Δ LAT of $\geq 10^{\circ}$. The manual stretching exercise consists of 4 directional neck motion exercises, including rotation, flexion, lateral flexion, and extension (Figure 2). Each directional motion exercise involved 3 repetitions of 15 manual stretches for 1 second with a rest period of 10 seconds between stretches. In addition, parents were educated to carry out only active symmetric neck

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