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Impaired Range of Motion of Limbs and Spine in Chronic Fatigue Syndrome

Peter C. Rowe, MD[1], Colleen L. Marden[1], Marissa A. K. Flaherty, MD[1], Samantha E. Jasion[1], Erica M. Cranston[1], Allison S. Johns[1], John Fan, MD[1], Kevin R. Fontaine, PhD[2], and Richard L. Violand, PT[3]

Objective To determine whether adolescents and young adults with chronic fatigue syndrome (CFS) have a greater prevalence of impaired range of motion (ROM) of the limbs and spine than healthy control patients.

Study design Case-control study comparing rates of abnormal ROM in 48 consecutive adolescents and young adults with CFS and 48 healthy control patients matched by sex and joint hypermobility. We examined range of ankle dorsiflexion, passive straight-leg raise, seated slump, upper-limb neurodynamic test, prone knee bend, and prone press-up. Abnormal ROM was defined before the study began. The number of abnormal responses ranged from 0 (normal ROM throughout) to 11 (impaired ROM in all areas tested).

Results The median number of areas with impaired ROM was greater in patients with CFS at the onset of stretch in the involved limb (5 vs 2, $P < .001$) and at end-range (2 vs 0, $P < .001$). Patients with CFS were more likely to have greater than 3 areas of impaired ROM (OR 6.0, 95% CI 2.1-17.3; $P < .001$) and were more likely to develop abnormal symptomatic responses to the individual tests and to the overall assessment (40% vs 4%; $P < .001$).

Conclusions Impaired ROM is more common in subjects with CFS than in healthy adolescents and young adults matched by sex and joint hypermobility. Adding a longitudinal strain to the nerves and soft tissues provoked symptoms in some subjects with CFS. The causes, functional impact, and optimal treatment of these abnormalities warrant further study. *(J Pediatr 2014;165:360-6)*.

Chronic fatigue syndrome (CFS) is a relatively common disorder in adolescence, with an estimated prevalence of 1 per 1000.[1-4] It is a prominent cause of prolonged school absence.[5-7] Cognitive behavioral therapy and graded exercise are the treatments best supported by the evidence in both adolescents and adults,[8,9] but treatment effect sizes are modest.[9,10] Moreover, concerns have been raised about the potential for harm if exercise is pursued too aggressively.[11] Better understanding of the pathophysiology of symptoms is needed to identify more effective treatments.

We have reported an overrepresentation of Ehlers-Danlos syndrome[12] and joint hypermobility syndrome[13] in adolescents and young adults with CFS. More recently, in the clinical care of patients, we have noted that adolescents and young adults with CFS have a greater-than-expected prevalence of postural abnormalities that could be attributable to their joint laxity (such as thoracic kyphosis, increased lumbar lordosis, head-forward posture). Somewhat paradoxically, we have also noted localized impairments in symptom-free range of motion (ROM) of the limbs and spine. Because individuals often report worsening of CFS symptoms during and the day after their initial physical therapy assessment[14] and an improvement in symptoms and daily function after the movement restrictions have been treated successfully, these impairments may have a role in the pathophysiology of symptoms.

The current study was designed to test the hypotheses derived from clinical practice that, compared with healthy control subjects, adolescents and young adults with CFS will have an increased prevalence of restrictions in limb and spine ROM, and ROM assessment, especially end-of-range measures, will be associated with increased symptoms.

Methods

Consecutive subjects with CFS were included if they had been referred to the Johns Hopkins Children's Center Chronic Fatigue Clinic between October 2008 and December 2012, were 10-30 years of age, and met the 1994 criteria by Fukuda et al for CFS.[1] Subjects with CFS entered the study with the expectation that they would be followed and treated clinically for 2 years. Subjects with previous depression who were referred by psychiatrists for evaluation of chronic fatigue were excluded.

Healthy control subjects in the same age range recruited during the same time period were eligible if they reported good, very good, or excellent general health.

CFS	Chronic fatigue syndrome
ROM	Range of motion
SF-36	Medical Outcomes Study 36-Item Short Form Health Survey
SLR	Straight-leg raise
ULNT1	Upper-limb neurodynamic test 1

From the [1]Department of Pediatrics, Johns Hopkins University School of Medicine, Baltimore, MD; [2]Department of Health Behavior, University of Alabama at Birmingham School of Public Health, Birmingham, AL; and [3]Violand and McNerney, PA, Ellicott City, MD

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All participants were excluded if they had conditions or treatments expected to interfere with ROM measurements such as recent fractures or sprains, previous joint surgery, neuropathy, arthritis, developmental anomalies, pregnancy, suspected anorexia (body mass index [kg/m^2] <17), severe obesity (body mass index >40), or recent physical therapy or other manual therapy. Control subjects were excluded if they had self-reported CFS, postural tachycardia syndrome, neurally mediated hypotension, fibromyalgia, recurrent syncope, or other health conditions associated with chronic fatigue (including major depression as measured by a T score >60 on the Child Depression Inventory[15] or a score >13 on the Beck Depression Inventory[16]). The study was approved by the Institutional Review Board of the Johns Hopkins Medical Institutes, and informed consent was obtained from participants or their parents as appropriate.

All participants completed the following questionnaires: (1) Pediatric Quality of Life Inventory (PedsQL 4.0), which is used to measure health-related quality of life in the preceding month[17] (total score ranges from 0 to 100, with greater scores indicating better quality of life); (2) Functional Disability Inventory, a 15-item self-report instrument for children and adolescents used to assess activity limitations caused by physical health problems during the preceding 2 weeks (total scores ranging from 0 to 60, with greater scores indicating greater disability)[18,19]; and (3) the Medical Outcomes Study 36-Item Short Form Health Survey (SF-36), a self-administered survey that is used to examine health-related quality of life in the preceding month[20] (greater scores indicate healthier function).

All subjects had a general physical examination that included the Beighton score, a commonly used, reliable measure of joint hypermobility, the scores of which range from 0 to 9; greater scores indicate greater joint laxity.[21] We describe the methods for deriving a Beighton score elsewhere.[12,13]

All participants were assessed for ROM with measures commonly used in physical therapy practice, described herein. All ROM testing was performed by a single experienced general pediatrician (P.R.) after >20 hours of training with the study physical therapist (R.V.). The examiner was not blinded with regard to case or control status. A research assistant was present for all measurements. ROM testing was performed with minor modifications according to the methods of Butler.[22,23] Goniometers were used to measure ROM where indicated, with the goniometer for ankle dorsiflexion, straight-leg raise (SLR), and knee bend placed according to the methods described by Norkin and White.[24] We recorded whether the procedure provoked symptoms in the limb tested (eg, stretch, pain, paresthesias) or systemically (eg, increased fatigue, lightheadedness, headache, nausea).

For the seated slump test, the subject sat upright with knees together, legs over the edge of the examining table, and with fingers interlocked comfortably behind the sacrum. The subject was then instructed to collapse/relax the shoulders and upper back (ie, told to slump while keeping the sacrum vertical), then to flex the neck forward to bring the chin toward the chest, and finally to actively extend the knee while in this posture. The angle of knee extension was measured first on one limb then the other as the slump posture was maintained. Full-knee extension was measured as 180°. An abnormal ROM on this test consisted of failure to bring the leg to within 10° of full knee extension. While the leg remained extended, we recorded the subject's symptoms in response to a return of the neck to a neutral, nonflexed position. Stretch in the lower limb was considered a normal response. An abnormal symptomatic response to the slump test (as well as to the ankle dorsiflexion and SLR tests) consisted of provocation of symptoms proximal to the lower limb (eg, back, neck, or head discomfort) or systemic symptoms (eg, fatigue, lightheadedness).

For ankle dorsiflexion, with the subject supine and the knee straight, the examiner held the foot in the subtalar neutral position and the subject actively dorsiflexed the ankle to end-range. Normal ROM was considered to be at least 95° of dorsiflexion on each side.

For passive SLR, while the subject was supine with the hip in a neutral position, the head lying at rest, and the trunk and limbs straight, the examiner raised the straightened leg to the point at which the subject reported stretch along the posterior thigh or leg. The examiner then advanced the SLR to the end-range as limited by pain or by soft tissue tension. An abnormal ROM on this test was considered an onset of soft tissue stretch or end-range of less than 45° in at least one leg.

Upper limb neurodynamic test 1 (ULNT1) also has been referred to as the upper-limb tension test with a median nerve bias.[22] The subject was positioned supine, arms at the sides and with legs together and knees extended. The examiner then abducted one of the subject's arms to 110° while the shoulder girdle was held in a depressed, but neutral position, with the elbow flexed to 90°. The examiner then extended the wrist, thumb, and fingers and supinated the forearm. The shoulder was then laterally rotated to 90°, and then the elbow was extended to the point at which the subject reported stretch along the upper limb. Full elbow extension was measured as 180°. An abnormal ROM consisted of an elbow angle less than 170° in one or both arms. An abnormal symptomatic response consisted of provocation of symptoms other than stretch in the anterior shoulder or antecubital fossa or paresthesias in the hand (eg, discomfort in the back, neck, or head, or lightheadedness).

For the prone knee bend, with the subject prone on the table, the examiner stabilized the hip with one hand, and, with the other, flexed the knee to the angle at which the subject reported stretch in the anterior thigh. We measured the angle between the long axis of the femur and the long axis of the fibula. The examiner then advanced the leg to the end-range of knee flexion. An abnormal test consisted of a knee flexion angle of less than 130°. An abnormal symptomatic response consisted of provocation of symptoms other than anterior thigh stretch.

For the prone press-up, with the subject prone and with hips and feet on the table and the ankles plantar flexed, the

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