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Tremor and Other Hyperkinetic Movements

Brief reports

Sensory Tricks Are Associated with Higher Sleep-Related Quality of Life in Cervical Dystonia

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Abstract

Background: Sensory tricks are compensatory gestures that cervical dystonia (CD) patients use to reduce abnormal neck posture and movements. Although sensory tricks are common in CD, little is known about whether trick efficacy changes over time or has effect on quality of life.

Methods: We analyzed clinical data and video recordings from 188 patients with isolated CD. We calculated the duration of CD and assessed the Toronto Western Spasmodic Torticollis Rating Scales and the Cervical Dystonia Impact Profile (CDIP-58).

Results: A longer duration of CD corresponded to a less effective sensory trick ($r(187) = 0.1901$, $p = 0.009$). Patients who demonstrated more effective sensory tricks reported higher sleep-related quality of life than patients with less effective sensory tricks ($r(187) = 0.1680$, $p = 0.0212$). There were no significant relationships between the effectiveness of a sensory trick and the other aspects of quality of life as measured by the CDIP-58.

Discussion: Patients who have had CD longer had less effective sensory tricks consistent with patients' verbal reports of previously having a trick that no longer works. Patients should be apprised of a wide variety of sensory tricks because their previous tricks may lose efficacy over time and because more effective tricks are associated with higher sleep-related quality of life.

Keywords: Sensory trick, adult onset dystonia, cervical dystonia, quality of life, sleep

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Introduction

Cervical dystonia (CD), also known as "spasmodic torticollis," is the most common focal dystonia, identified by involuntary contractions of cervical muscles, producing abnormal head postures sometimes with overlying spasms that may resemble tremor. Many patients develop a "sensory trick," also referred to as an "alleviating maneuver" or geste antagoniste, to control and diminish the severity of the abnormal involuntary movements. In the revised Toronto Western Spasmodic Torticollis Rating Scale (TWSTRS-2),[1] a sensory trick is defined as a touch or other movement that influences the severity of the abnormal movements.

The rating scale item evaluates the degree of improvement when a sensory trick is used. Sensory tricks have been previously classified by location and physical characteristics within a cohort of 138 patients, illustrating their prevalence and role in carrying out common activities of daily living.[2] Our study had two objectives. The first objective was to determine whether patients' sensory tricks have an impact on their quality of life, as measured by the eight subdomains of the Cervical Dystonia Impact Profile (CDIP-58). We chose to evaluate quality of life with the CDIP-58 because it is the most recent rating scale developed to quantify quality of life from the patient's perspective.[3] The second objective was to evaluate the replicability of anecdotal reports and a previous smaller study[4] showing that longer disease duration corresponds to a less effective sensory trick.

Methods

We analyzed data collected from 208 patients with isolated CD enrolled across 10 sites in the Dystonia Coalition's project to develop and validate a comprehensive rating scale for CD severity (http://clinicaltrials.gov/show/NCT01373424). All patients provided informed consent prior to their participation in the study. The protocols for original data collection and subsequent analyses were approved by the Human Research Protection Offices at the Washington University School of Medicine (WUSM), Rush University Medical Center (RUMC), and the University of California, San Diego (UCSD; protocol no. 111255X). All patients were examined and videotaped according to a standard protocol between March 2011 and January 2013. We calculated the duration patients had CD by subtracting their reported age of onset from their age at the time the protocol was administered. The examination protocol included a step in which patients were seated in a chair without head support, feet resting on the floor, and instructed to demonstrate their "most effective sensory trick." If patients were unaware of a trick, they were prompted to try touching their right cheek, left cheek, and back of their head. During the examination, physicians completed the Toronto Western Spasmodic Torticollis Rating Scale (TWSTRS-2) Motor Severity scale which included an item for "Effect of Sensory Trick."[1,5] This item was scored as complete (0), moderate (1), mild (2), minimal (3), or no (4) improvement of posture by one or more tricks. All video recordings were reviewed by a movement disorders neurologist (CLC) and scored for motor severity excluding the trick efficacy, ranging from 0 to 10, referred to hereafter as simply "global motor score."

In addition to the clinician assessments, all patients completed TWSTRS-2 Pain and Disability scales.[6] The TWSTRS-2 Pain and Disability scores were added to the TWSTRS-2 Motor Severity to compute a TWSTRS-2 total score. Patients also completed the CDIP-58. The TWSTRS-2 Motor item "Effect of Sensory Trick" scores were compared with the eight CDIP-58 subscales: head and neck symptoms, pain and discomfort symptoms, upper limb activities, walking, sleep, annoyance, mood, and psychosocial functioning.[3] Each subscale has multiple items, and each item is scored by how much and/or how often patients were "bothered": not at all (1), a little (2), moderately (3), quite a bit (4),or extremely (5) and none of the time (1), a little of the time (2), some of the time (3), most of the time (4), or all of the time (5).

The relationships between "Effect of Sensory Trick," disease duration, and each of the CDIP-58 subscales were tested using linear regressions. We also used hierarchical linear regressions to control for the potentially confounding effect of global motor severity, age, and disease duration as covariates. All statistical analysis was performed with JMP.[7] We used an alpha level of 0.05 to determine significance.

Results

Of the 208 patients, 20 patients were excluded due to missing data. Demographics, botulinum toxin treatment status, and total TWSTRS scores for the patient cohort are provided in Table 1. All possible rating values of the "Effect of Sensory Trick" score were reported, but the most common score was 1, moderate improvement (n = 83) (Figure 3). The "Effect of Sensory Trick" score was positively correlated with the duration of CD ($r(187) = 0.1901$, $p = 0.009$) (Figure 1), but was not correlated with global motor severity ($r(187) = 0.0140$, $p = 0.8487$) or age ($r = 0.038$, $p = 0.61$). A hierarchical regression model for the Effect of Sensory Trick demonstrated that disease duration makes a significant contribution to trick efficacy even after controlling for global motor severity ($F(2,187) = 3.474$, $p = 0.033$).

Of the eight CDIP-58 subscales, seven had no significant correlation with the effect of sensory trick: head and neck symptoms ($p = 0.7325$), pain and discomfort symptoms ($p = 0.2008$), upper limb activities ($p = 0.4841$), walking ($p = 0.7089$), annoyance ($p = 0.2510$), mood ($p = 0.3389$), and psychosocial functioning ($p = 0.8734$).

The CDIP-58 sleep subscale was positively correlated with the "Effect of Sensory Trick" score ($r(187) = 0.1680$, $p = 0.0212$) (Figure 2). The average CDIP-58 sleep subscale rating was 8.4 (standard deviation [SD] = 4.9), out of a total possible score of 20 (Figure 3). We also found that the CDIP-58 sleep subscale was positively correlated with global motor severity ($r = 0.2059$, $p = 0.0046$). In a hierarchical regression model, although disease duration and global motor severity contribute to the CDIP-58 sleep score ($F(2,187) = 4.2$, $p = 0.016$), the "Effect of

Table 1. Patient Characteristics

Demographics	
Age (SD) (years)	59.7 (10.3)
Sex (F/M)	141/47
Received Botox (Y/N)	129/59
TWSTRS total (possible range 0–98)	
Range	5.00–61.75
Average (SD)	33.0 (13.5)

Abbreviations: SD, standard deviation; TWSTRS, Toronto Western Spasmodic Torticollis Rating Scale.

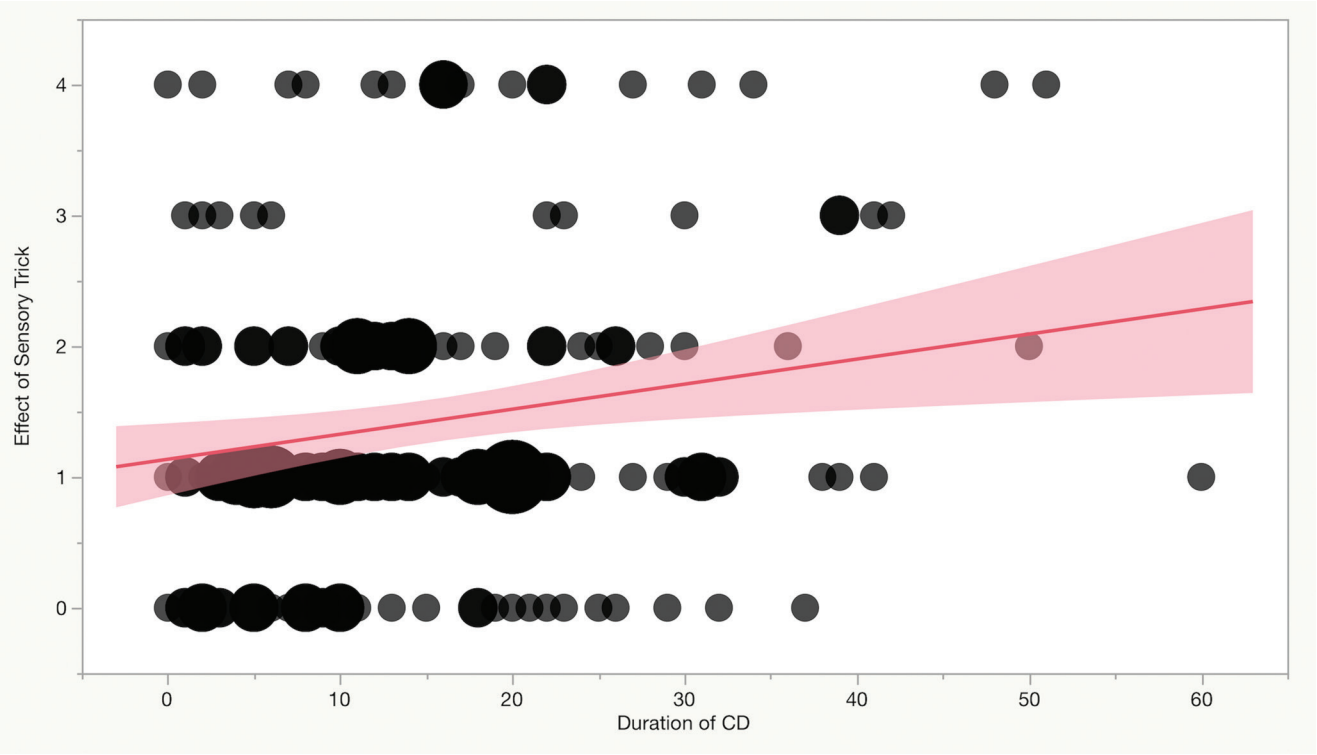

Figure 1. **Scatterplot of Effect of Sensory Trick vs. Duration of CD.** A higher value for "Effect of Sensory Trick" corresponds to a less effective trick. Bubble sizes reflect the number of patients at a given point – the largest bubble including seven patients and the smallest bubble one patient. The shaded region represents the 95% confidence interval.

Sensory Trick" makes a significant contribution to the CDIP-58 sleep score even after controlling for disease duration and global motor severity ($F(3,187) = 4.59$, $p = 0.004$).

Discussion

To our knowledge, this is the first study to systematically investigate whether and how a sensory trick impacts quality of life in CD patients. The results show that a less effective sensory trick corresponds to lower sleep-related quality of life, that is, a higher CDIP-58 sleep subscale score. This score indicates how bothersome CD is for quality of sleep, consisting of the following four items: having trouble falling asleep, experiencing restless sleep, waking up, and not getting enough sleep due to CD symptoms. Our finding of a relationship between trick and no other aspects of quality of life as captured by the CDIP-58 is curious. Other domains in the CDIP-58, such as "head and neck symptoms," "pain and discomfort," "annoyance," "mood," and "psychosocial functioning," would inherently seem to be more affected by a sensory trick. Our cohort of patients reported during their video-recorded examinations the use of sensory tricks while executing activities at work. CD has been shown to reduce productivity by 11 hours during a 40-hour workweek and result in unemployment for 38.5% of patients.[8] Thus, given the significance of work for several aspects of quality of life, we expected the effective use of a sensory trick to improve quality of life in multiple domains. Perhaps the effect of the sensory trick is outweighed by other factors influencing these other aspects of quality of life.

Of course, the fact that we found a correlation between trick efficacy and sleep quality does not bear on the directionality of the relationship between the two. An ineffective trick could lead to a lower quality of sleep. Conversely, lower quality sleep might make a sensory trick less effective. Both scenarios could contribute to a vicious cycle of fatigue and diminished ability to transiently alleviate CD motor symptoms with the trick.

The small effect size in our correlation suggests that the trick may not be a dominant factor in determining sleep-related quality of life. Although we found that trick efficacy is associated with sleep-related quality of life even after controlling for global motor severity, we also incidentally found that sleep-related quality of life and trick efficacy are positively correlated. However, impaired sleep quality in CD patients does not improve with an improvement of motor severity from botulinum toxin treatment.[9] It should also be noted that other factors may influence sleep quality, including oral medications such as benzodiazepines and anticholinergics that are often used to supplement botulinum toxin injections. Taken together, these studies suggest

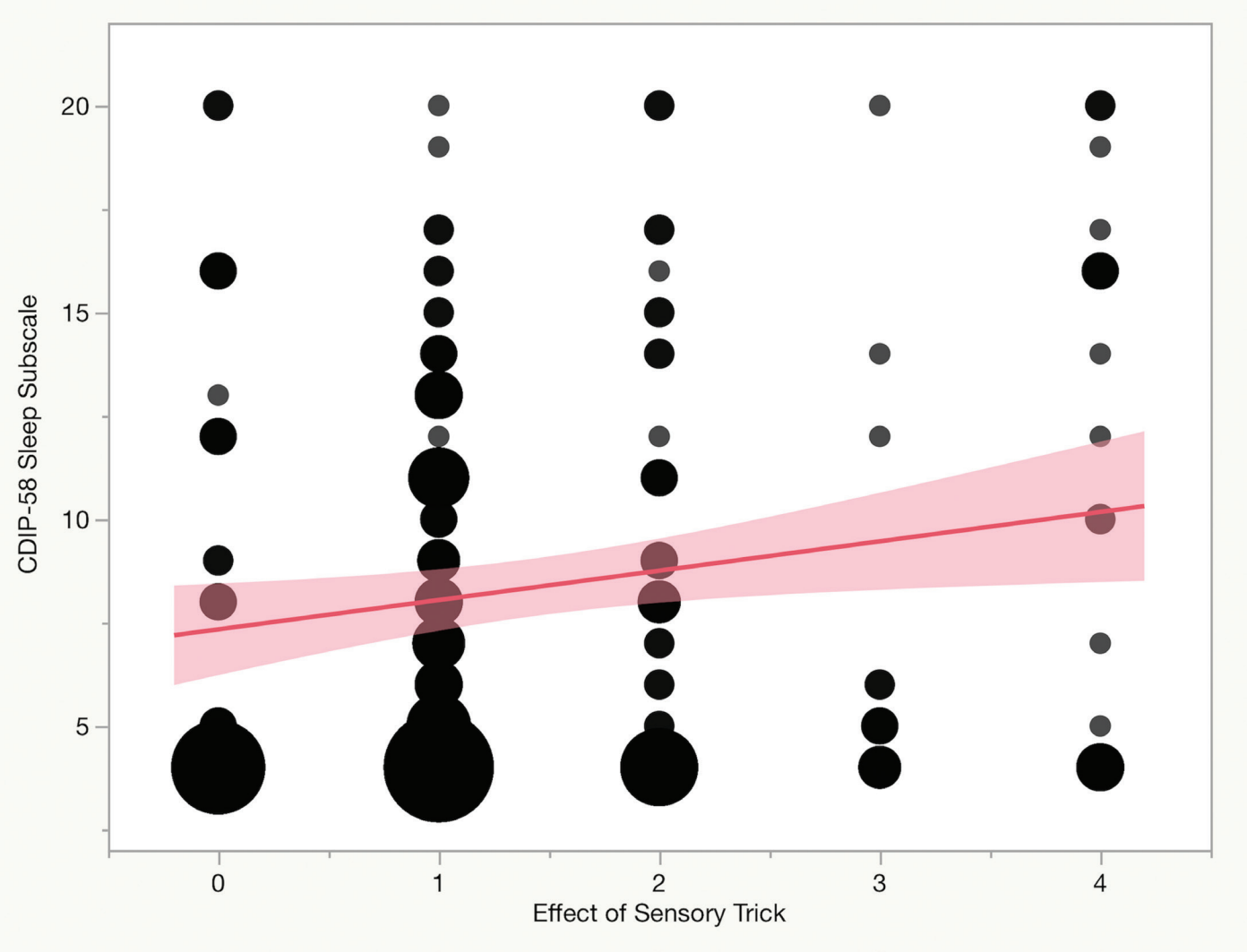

Figure 2. **Scatterplot of CDIP-58 Sleep Subscale Scores vs. the Effect of a Sensory Trick.** Bubble sizes reflect the number of patients at a given point – the largest bubble including 26 patients and the smallest bubble 1 patient. The shaded region represents the 95% confidence interval.

that other aspects of CD may play an important role in sleep perturbations. Future research should differentiate the degree of impact from motor and non-motor factors on sleep-related quality of life in CD patients.

Patients who have had CD longer tend to have less amelioration of abnormal movements with the use of sensory tricks, even after controlling for global motor severity. These results confirm and extend anecdotal reports and a previous smaller study.[4] In their report on the temporal discrimination threshold in CD that included results on the relationship between disease duration and trick efficacy, Kagi et al.[4] showed that nine patients with complete trick efficacy had shorter disease duration than 17 patients with only partial efficacy. Our results with 188 patients are consistent with, and give further statistical power to, their report. Although the mechanism for this remains unclear, it does not seem to be age, because at least in our cohort the effect of the trick was not correlated with age. Regardless, sensorimotor circuit plasticity is widely believed to decrease with time, and the plasticity of these circuits could play an important role in mediating the effect of sensory tricks.[10] This is further supported by a recent case study demonstrating an association between the effect of sensory trick and physiological characteristics of the parietal cortex, which is known for its role in sensorimotor integration.[11] Perhaps gradually decreasing plasticity in the sensorimotor mappings, in a manner specific to CD pathology and not just age-related, makes the sensory trick less effective over time.

Because the design of our study is cross-sectional, we are measuring the effect of each patient's *current* sensory trick. Some patients may have changed their trick over time. In these cases, if the efficacy of the initial trick waned over time and those patients were assessed with only their initial trick, our result – that longer disease duration is associated with

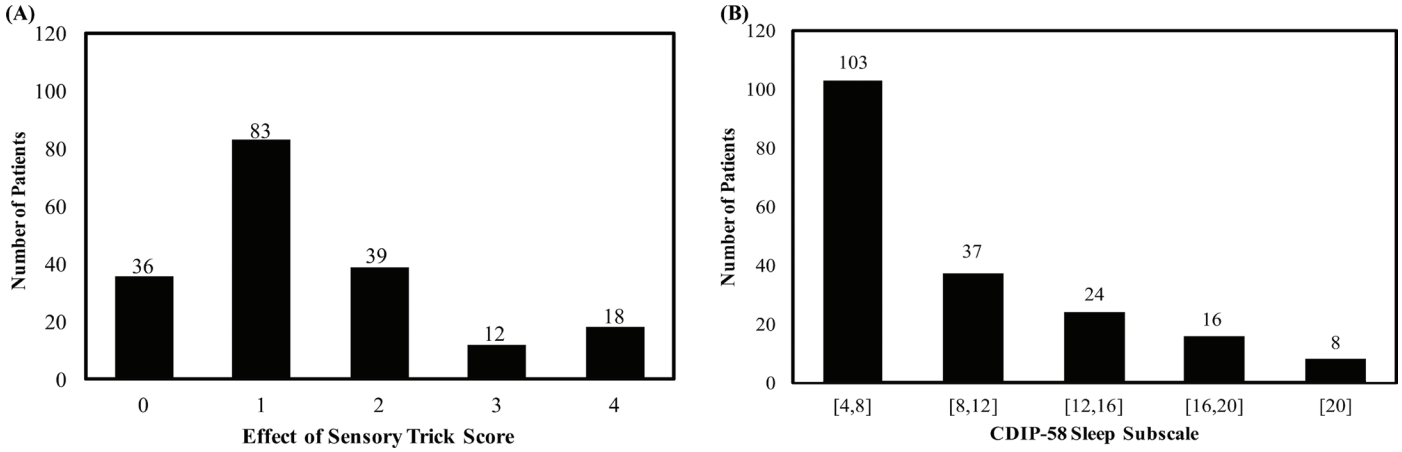

Figure 3. (A) Distribution of the TWSTRS-2 Motor Item Rating the "Effect of Sensory Tricks" (B) Distribution of the CDIP-58 Sleep Subscale. (A) Physicians rated the degree of improvement when a sensory trick is used on a scale of 0–4, with 0 indicating complete improvement of posture and 4 indicating no improvement. (B) The total sleep subscale score is calculated by the sum of the four individual items: having trouble falling asleep, having restless sleep, waking up, and not getting the amount of sleep needed. Each item is scaled 1–5 based on the frequency with which it occurs as a result of CD, 1 indicating none of the time and 5 indicating all of the time.

lower trick efficacy – would be even stronger than reported. Future studies should include structured formal inquiry about any changes over time that patients have made in their use of tricks as well as assessments of the efficacy of not only currently used but also previously used tricks.

Our study has some limitations that should be considered when interpreting the results. First, the scales used to measure severity of both effective use of sensory trick and impairment of sleep within the TWSTRS-2 and CDIP-58 ratings are intrinsically subjective. Second, although sensory tricks are observed during a protocol when specifically prompted, not all tricks may be reported. One reason may be due to the inconsistency of the use of the term "sensory trick" in referring to a physical movement or sensation that improves abnormal head movements. This can cause confusion on behalf of the patient regarding the definition. Third, there may be circumstances in patients' daily lives that facilitate an effective sensory trick but which cannot be easily replicated in the clinic. Finally, because the CDIP-58 asks about patients' quality of life over only the past 2 weeks, and because most patients were evaluated at the end of their injection interval (a minimum of 3 months), we are unable to capture the potential week-to-week dynamics of the relationship between trick and quality of life.

In summary, this study provides the first quantitative analysis of the improvement of head posture attributable to the use of sensory tricks and its association with overall health-related quality of life for CD patients. Further exploration of the magnitude of improvement due to the use of a sensory trick on the health impact of a CD patient's life with more precise and comprehensive clinical examinations and questionnaires could help maximize the therapeutic potential of sensory tricks in CD.

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