Effect of prolonged neck muscle vibration on lateral head tilt in severe spasmodic torticollis

H-O Karnath, J Konczak, J Dichgans

Abstract

Short term vibration of the dorsal neck muscles (10-35 s) is known to induce involuntary movements of the head in patients with spasmodic torticollis. To investigate whether neck muscle vibration might serve as a therapeutic tool when applyed for a longer time interval, we compared a vibration interval of 5 seconds with a 15 minute interval in a patient with spasmodic torticollis with an extreme head tilt to the right shoulder.

Head position was recorded with a two camera optoelectronic motion analyzer in six different test conditions. Vibration regularly induced a rapid change of head position that was markedly closer to a normal, upright posture. After 5 seconds of vibration, head position very quickly returned to the initial position within seconds. During the 15 minute interval, head position remained elevated. After terminating vibration in this condition, the corrected head position remained stable at first and then decreased slowly within minutes to the initial tilted position.

Conclusions-(1) In this patient, muscle vibration was the specific sensory input that induced lengthening of the dystonic neck muscles. Neither haptic stimulation nor transcutaneous electrical stimulation had more than a marginal effect. (2) The marked difference in the change of head position after short and prolonged stimulation supports the hypothesis that spasmodic torticollis might result from a disturbance of the central processing of the afferent input conveying head position information-at least in those patients who are sensitive to sensory stimulation in the neck region. (3) Long term neck muscle vibration may provide a convenient non-invasive method for treating spasmodic torticollis at the central level by influencing the neural control of head on trunk position.

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Keywords: human; cervical dystonia; spasmodic torticollis; neck muscle vibration; transcutaneous electrical stimulation

Idiopathic cervical dystonia or spasmodic torticollis is an abnormal involuntary contrac-

tion of the neck muscles. Although several hypotheses have been put forward, the pathomechanism that causes this disturbance of head control is still unclear.¹ Several authors reported evidence for vestibular abnormalities in patients with spasmodic torticollis, such as vestibulo-ocular reflex hyperreactivity or asymmetry, indicating an involvement of the vestibular system²⁻⁴ or the cervico-ocular reflex⁵ in the pathogenesis of spasmodic torticollis.

Leis et al⁶ investigated the effect of vibration and electrical stimulation on neck muscles in 11 patients with cervical dystonia. The authors found a reduction of the involuntary muscle activity in two patients by applying vibration and in three patients by applying electrical stimulation. Lekhel et al7 likewise noted involuntary movements of the head with vibration of the dorsal neck muscles in nine out of 15 patients with spasmodic torticollis. Both studies used very short vibration intervals, of 10 and 35 seconds, and found effects that lasted only for seconds after terminating the stimulation. To investigate whether neck muscle vibration might serve as a therapeutic tool when applyed for a longer time interval, the present study compared a vibration interval of 5 seconds with a 15 minute interval in a patient with severe spasmodic torticollis who was found sensitive to neck muscle vibration.

Methods

PATIENT

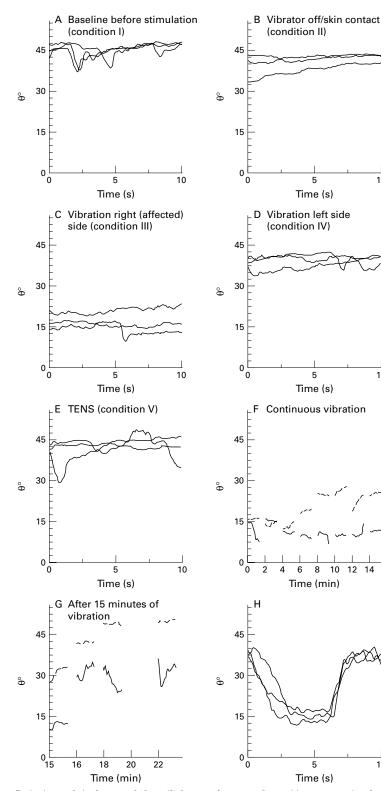
At the age of 48, a presently 54 year old female organist developed a hitherto stabile torticollis spasmodicus of a waxing and waning amount with the head bent to the right and slightly rotated to the left together with elevation of her chin. Scoliosis of the upper trunk to the left and elevation of the left shoulder as well as severe neck pain also developed. Dystonia largely subsided when she was laying in bed and was markedly accentuated, when walking or speaking. Anticholinergic, neuroleptic and benzodiazepine drugs were ineffective. Botulinum toxin A was successfully injected first, but she developed a lasting unresponsiveness after six injections within 1 year. Subsequent denervation of her right sternocleidomastoideus and splenius capitis and the implantation of high frequency epidural stimulation electrodes at C3/4 were of no lasting benefit.

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Projection angle in the coronal plane (θ) between the two marker positions, representing the longitudinal axis of the patient's head, and the earth vertical. In conditions I to V (A-E)the solid lines represent the three trials that were recorded for a duration of 10 seconds each In condition VI (F–H), the dashed line represents the data of the first recording session; the solid line refers to the second recording. (A) No stimulation. (B) Haptic stimulation without vibration of the affected, right splenius muscle. (C) Vibration of the affected splenius muscle. (D) Vibration of the unaffected, left splenius muscle. (E) Transcutaneous electrical stimulation of the affected splenius muscle. (F) During a vibration interval of 15 minutes of the affected, right splenius muscle, head position was recorded every 2 minutes over a period of 2 seconds each. (G) Directly after this prolonged vibration period, head position was again recorded for 2 seconds. In addition, interval recording (every 2 min for 2 s each) of head position continued for another 7 minutes. (H) 5 second vibration interval of the affected splenius muscle. Head position recording started with the begin of vibration and continued for 10 seconds. The solid lines represent the three trials recorded.

APPARATUS AND PROCEDURE

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Head position was recorded with a two camera optoelectronic motion analyzer (ELITE) at a sampling frequency of 100 Hz. Two infrared light reflective markers were used, one attached to the tip of the nose, the other at the nasion to record head posture. Time position data of each marker were filtered applying the automatic model based bandwidth selection procedure LAMBDA.8 As a measure of head tilt, we subsequently computed the projection angle in the coronal plane (θ) between the two marker positions, representing the longitudinal axis of the head, and the earth vertical. The patient sat on a normal chair and was instructed to hold her head upright facing straight ahead.

Head position was recorded under six conditions. In conditions I to V we recorded three trials for a duration of 10 seconds each. In condition I, the patient's head position was recorded with no additional stimulation. In condition II, the tip of the experimental vibrator, a flat disk (2.3 cm in diameter), was placed on the right splenius muscle without vibration. In condition III, vibration was applied with the frequency fixed at 80 Hz and an amplitude of 0.4 mm. The tip of the vibrator was placed on the same position as in condition II. The recording period started 5 seconds after the beginning of each muscle vibration. In condition IV, the unaffected, left splenius muscle was vibrated. Condition V applied transcutaneous electrical stimulation instead of muscle vibration. The location of stimulation was identical with that in condition III. The stimulator used superficial electrodes with a pulse frequency of 100 Hz. The recording period started 5 seconds after the beginning of stimulation.

In condition VI neck muscles were continously vibrated for 15 minutes while the head position was recorded every 2 minutes for a 2 second interval. Directly after this vibration period, head position was again recorded for 2 seconds. In addition, interval recording (every 2 min for 2 s each) of head position continued for another 7 minutes to determine retention. To contrast the change of head position that followed the long vibration interval of 15 minutes, after 1 hour of resting the patient's neck muscles were vibrated for only 5 seconds. Head position recording started with the beginning of muscle vibration and continued for 10 seconds. Three trials were recorded.

Results

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The figure shows the projection angle in the coronal plane (θ) between the two marker positions, representing the longitudinal axis of the head, and the earth vertical. In baseline condition I, her head was tilted to the right with an average angle of 45° (fig A). Head position was not rigid but oscillated in an angular range of 37°-48°. Placing the vibrator tip on the right splenius muscle without vibrating it (condition II; fig B) seemed to reduce head oscillation but had no relevant influence on the head position which showed an average angle of 40°. This changed markedly with neck muscle vibration of the right side (condition III; fig C). With an average angle of 17°, the patient's extreme head

tilt to the right was substantially reduced. By contrast, vibration of the splenius muscle on the unaffected, left side (condition IV; fig D) had no relevant effect on head position (average angle=39°). By contrast with vibration of the right side (condition III; figure C), nonspecific stimulation by transcutaneous electrical stimulation at the same anatomical site (condition V; fig E) had only a marginal effect. The patient's head was still tilted to the right with an average angle of 42°, which fell within baseline range.

To document the replicability of the vibration effect that we were able to find clinically each time the patient was seen in our department, we formally tested her twice within a 3 month time interval. Figure F presents the results of both recordings during a 15 minute interval of continuous vibration (condition VI). Compared with the initial tilted head position with an average angle of 45° in the first and 50° in the second session, head position remained relatively stable and more upright during continous stimulation. The average angle was 21.5° (SD 5.2°) in the first and 11.2° (SD 2.0°) in the second session (fig F). After the 15 minute vibration interval (fig G), the patient's head position in the first session slowly returned to the initial tilted head position within minutes. In the second session, even 7 minutes after vibration, the head was only tilted 30° to the right (angular range=26°-37°).

To contrast the change of head position that followed a long vibration interval of 15 minutes, a short vibration interval of only 5 seconds was applied. Figure H demonstrates that the head position very quickly (~ 2 s) reached the basal level under this condition.

In conditions I and III, three healthy men, aged 30, 36, and 39 served as controls. No difference was found between both conditions; the average angle of all three subjects was 2° . Head position oscillated in an angular range of 0°-8° in condition I and 1°-3° in condition III.

Discussion

The present results confirm the finding⁵ that the response of head posture to sensory stimulation at the neck region is selective. In the patient an improvement was only obtained with vibration of the contracted neck muscles. This selectivity argues against the assumption of a non-specific arousal effect leading to the change of head posture but rather for an involvement of neck proprioception in the pathogenesis of spasmodic torticollis.

The aim of the present study was to compare the effects of short and prolonged vibration of neck muscles in spasmodic torticollis. We found that after a short stimulation interval of 5 seconds head position returned to the initial position within seconds. By contrast, after terminating the 15 minute interval, the corrected head position remained stable at first and then decreased slowly within minutes to the initial tilted position. The marked difference in the change of head position after short and prolonged stimulation supports the hypothesis that spasmodic torticollis might result from a disturbance of the central processing of the afferent input conveying head position information-at least in those patients with spasmodic torticollis who are sensitive to sensory stimulation in the neck region. The delay of involuntary change of head position after offset of the 15 minute stimulus argues against a peripheral disturbance of proprioceptive afferents from the neck muscle spindles and from the upper cervical joints,9 but for the involvement of a centrally controlled compensatory torsion of the head due to the altered peripheral proprioceptive input.

An effective, although only symptomatic method to treat spasmodic torticollis was introduced with the injection of botulinum toxin type A or F in neck muscles.^{10 11} Whereas the use of botulinum toxin aims at the efferent component of head control, Freckmann et al9 found a positive effect by treating an afferent component by microsurgical lysis of nerve root anastomoses. By contrast with these two therapies, long term muscle vibration may provide a convenient non-invasive method for treating spasmodic torticollis at the central level by influencing the neural control of head on trunk position.

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- 1 Dauer WT, Burke RE, Greene P, et al. Current concepts on
- Branch W.; Starker K.; Aretiology and management of idiopathic cervical dystonia. *Brain* 1998;121:547-60.
 Bronstein AM, Rudge P. Vestibular involvement in spas-modic torticollis. *J Neurol Neurosurg Psychiatry* 1986;49: 290 - 5
- 3 Diamond SG, Markham CH, Baloh RW. Ocular counterrolling abnormalities in spasmodic torticollis. Arch Neurol 1988;45:164-9
- 4 Huygen PLM, Verhagen WIM, van Hoff JJM, et al. Vestibu-Iar hyperreactivity in patients with diopathic spasmodic torticollis. J Neurol Neurosurg Psychiatry 1989;52:782–5.
 Stell R, Gresty M, Metcalfe T, et al. Cervico-ocular function
- in patients with spasmodic torticollis. J Neurol Neurosurg Psychiatry 1991;54:39-41.
- 6 Leis AA, Dimitrijevic MR, Delapasse JS, et al. Modification for cervical dystonia by selective sensory stimulation. Journal of the Neurological Sciences 1992;110:79–89.
- Lekhel H, Popov K, Anastasopoulos D, et al. Postural responses to vibration of neck muscles in patients with idiopathic torticollis. Brain 1997;120:583-91.
- 8 D'Amico M, Ferrigno G. Comparison between the more recent techniques for smoothing and derivative assessment
- in biomechanics. Med Biol Eng Comput 1992;30:193-204. Freckmann N, Hagenah R, Herrmann H-D, et al. Bilateral microsurgical lysis of the spinal accessory nerve roots for the treatment of spasmodic torticollis. *Acta Neurochir* (*Wien*) 1986;**83**:47–53.
- 10 Tsui JK, Eisen A, Mak J, et al. A pilot study on the use of botulinum toxin in spasmodic torticollis. Can J Neurol Sci 1985;12:314-16.
- Greene PE. Fahn S. Use of hotulinum toxin type F 11 injections to treat torticollis in patients with immunity to botulinum toxin type A. Mov Disord 1993;8:479-83.