

Long-term effects of electrotactile sensory substitution therapy on balance disorders

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This clinical research investigated whether a new type of rehabilitation therapy involving the use of a vestibular substitution tongue device (VSTD) is effective for severe balance disorders caused by unilateral vestibular loss. Sixteen patients with postural imbalances because of unilateral vestibular loss underwent training with VSTD. The VSTD transmits information on the head position to the brain through the tongue as substitutes for the lost vestibular information. The device's electrode array was placed on the tongue and participants were trained to maintain a centered body position by ensuring the electrical signals in the center of their tongue. All participants completed 10 min training sessions 2–3 times per day for 8 weeks. Functional gait assessments and the dizziness handicap inventory were, respectively, used to evaluate participants' dynamic gait function and their severity of balance problems before and after the training period. All examined parameters improved after the 8-week training period. These changes were maintained for up to 2 years

after the termination of the training program. Short-term training with VSTD had beneficial carry-over effects. VSTD training might represent a useful rehabilitation therapy in individuals with persistent balance disorders and might lead to long-term improvements in their balance performance and ability to perform daily and social activities. *NeuroReport* 27:744–748 Copyright © 2016 Wolters Kluwer Health, Inc. All rights reserved.

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Introduction

Peripheral vestibular disorders cause severe vertigo, nausea, nystagmus, and postural imbalances. These characteristic symptoms usually improve spontaneously with time by a behavioral recovery process known as vestibular compensation [1,2]. However, in some patients, a lack of central vestibular neuroplasticity renders the central vestibular compensation mechanism ineffective, limiting the extent of any recovery in their balance performance. Such patients usually do not respond to standard therapies such as medical and vestibular balance rehabilitation therapy to facilitate vestibular compensation [3]. Therefore, these patients have difficulty in standing and walking without aids, such as canes and walkers, resulting in significant decreases in their quality of life and difficulty in performing daily and social activities [4,5].

In this study, we propose a new therapy involving the use of a vestibular substitution tongue device (VSTD; BrainPort balance device, Wicab Inc., Middleton, Wisconsin, USA [6,7]) for the treatment of patients with intractable balance disorders caused by vestibular deficits. Several studies found that training with a VSTD produced favorable short-term results in patients with intractable vestibular disorders [7–9]. However, it has not

been determined whether the acute beneficial effects of VSTD continue in the absence of the device after the training session. The long-term effects of VSTD will determine its clinical utility as a rehabilitation tool because some patients have difficulty in continuing the training program using such devices for a long period.

Therefore, to evaluate the practical utility of VSTD in patients with intractable balance problems because of peripheral vestibular etiologies, this study examined whether the improvement produced by VSTD training persisted after the end of the training period.

Methods

Participants

This study was approved by the Clinical Research Ethics Board of Nara Medical University Hospital (Approval no: 06-033). Sixteen patients with unilateral vestibular loss because of vestibular neuritis, Ramsay Hunt syndrome, labyrinthitis, or acoustic nerve resection provided their written informed consent before participating in this study. They ranged in age from 29 to 79 years (mean: 59.8 years) and included five men and 11 women. They had presented with chronic dizziness and postural imbalance that had lasted for over 5 years and markedly interfered with their daily and social activities.

All participants fulfilled the following selection criteria:

- (1) Significant unilateral horizontal semicircular canal dysfunction (defined as a maximum slow phase velocity of less than 3 degrees per second during irrigation with ice water) detected before and 2 years after the training period.
- (2) The participant's balance disorder assessed by gait function test had not improved at all after 6 months or longer of conventional vestibular balance rehabilitation therapy.
- (3) There was no current or previous evidence of functional or morphological pathologies of the central nervous or musculoskeletal system, as assessed by questionnaires and brain imaging such as MRI or computed tomography.

Vestibular substitution tongue device

The device consisted of an intraoral device and a controller, which were connected to each other through a tether (Fig. 1a). The intraoral device contained a sensor (Fig. 1b) for detecting the participant's head position and an electrode array (a 24×24 mm matrix with a diameter of 1.5 mm, containing a 100-point gold-plated electrode) (Fig. 1c). The sensor, which contained a two-axis accelerometer for sensing head tilting and motion in the anterior–posterior and left–right directions, was mounted on the back of the electrode array. The accelerometer collected head position information and sent it to the controller for processing. The controller then translated the information and converted it into electronic signals, which were delivered to the electrode array positioned on the participant's tongue, resulting in an electrotactile stimulus. In this method, the device is used to replace the lost vestibular input with sensory inputs from the tongue and transmit the patients' balance information to their brain.

Vestibular substitution tongue device training

The electrode array was placed on the anterior center portion of the tongue, forming a human–machine interface, during the training sessions (Fig. 1d). A representative training scene is shown in Fig. 1e. The electrotactile signals delivered to the tongue provide the participants with information about their posture through their tongue. For example, if the patient sways to the right, an electrotactile stimulus is delivered to the right side of the array. Therefore, the patient would realize that his/her posture deviates to the right. The patient would then attempt to adjust their posture so that the signals were delivered back to the center of their tongue. This is the way patients were trained to maintain a centered body position by ensuring that the electrical signals were being delivered to the center of their tongues. While VSTD was in place, patients underwent training in the Romberg or the Tandem-Romberg position with

their eyes open or closed, according to their skill level and their ability to balance.

After receiving instructions on the training method, patients underwent two initial training sessions ending with a 10 min trial at the clinic and 2–3 consecutive training sessions per day at home for an 8-week period. The patients visited our clinic weekly and we confirmed their compliance with the training.

Study metrics

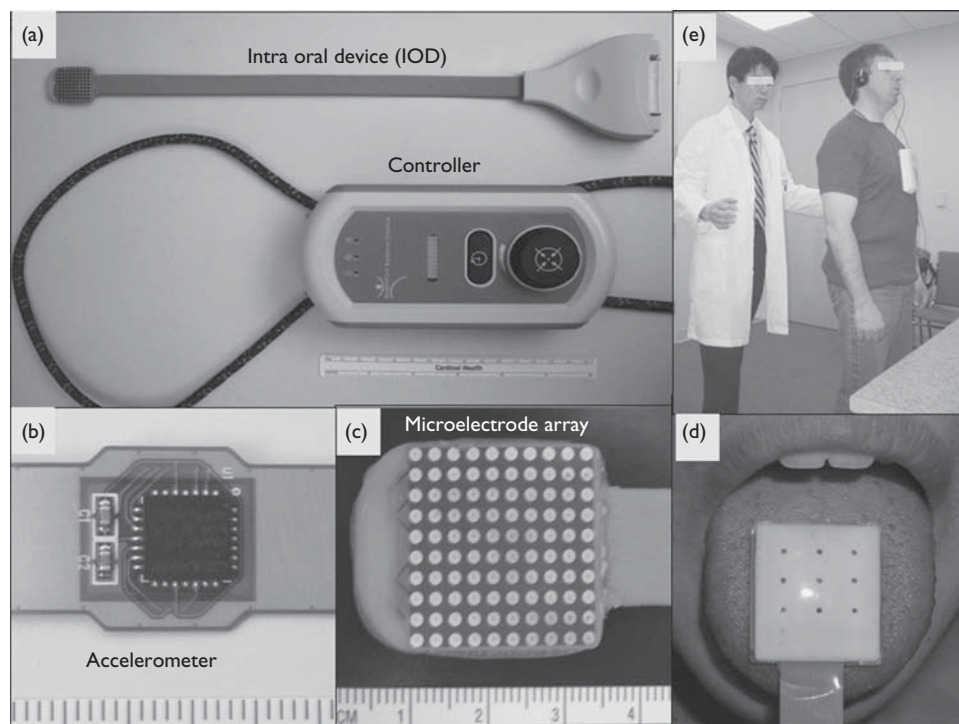
The participants' dynamic gait function was examined using functional gait assessments (FGAs [10]). The dizziness handicap inventory (DHI) [11] (quality-of-life indices) was used to subjectively examine the severity of the participants' dizziness and imbalance. The above parameters were assessed in all patients before VSTD training (BT); after the 8-week training program (AT_0); and at 1 (AT_1), 3 (AT_3), 6 (AT_6), 12 (AT_{12}), 18 (AT_{18}), and 24 (AT_{24}) months after the cessation of the program. The significance of differences between pre-treatment and post-treatment time points was analyzed using the one-way repeated-measures analysis of variance and Dunnett's test. All tests were subjected to third-party evaluations that were not blinded.

FGA [10] is designed to measure an individual's ability to perform dynamic balance tasks while walking 20 feet. FGA consists of 10 different gait tasks, and the participants' performance during each task is rated from 0 (poor) to 3 (excellent), resulting in a total gait performance score ranging from 0 (worst) to 30 (best). All FGA were videotaped for further review and observational gait analysis.

DHI [11] is used to determine the perceived extent of disability experienced by an individual in daily life because of dizziness. DHI is a standardized questionnaire consisting of 25 items related to the emotional, physical, and functional components of vestibular dysfunction. Individuals are required to answer 'yes,' 'sometimes,' or 'no' to each question, which are then awarded the scores of 4, 2, and 0, respectively. Thus, the total DHI score ranges from 0 (best) to 100 (worst).

Results

All 16 participants showed pronounced improvements in their walking balance performance after the 8-week training program. The participants' mean FGA score increased significantly from 13.1 ± 1.8 before the training program to 23.2 ± 1.3 immediately after the training program. The improved FGA score was maintained until 12 months (22.3 ± 1.5) after the end of the training program, and then gradually but not significantly decreased by 24 months (19.7 ± 1.6). The scores during the 2-year post-training course were significantly better than the pretraining scores (Fig. 2a).

Fig. 1

Aspect of a vestibular substitution tongue device. (a) An intraoral device and a controller. (b) A sensor (an accelerometer) for detecting the position of the head of the participant. (c) An electrode array. (d) The electrode array placed on the anterior center portion of the tongue. (e) The representative training scene.

Improvements were also noted in the participants' quality-of-life indices, as shown by their DHI scores. The participants' mean DHI scores improved significantly from 65.2 ± 4.8 to 39.1 ± 5.4 during the 8-week training period. The improved score remained significantly stable until the end of the 2-year follow-up period (38.4 ± 5.3) (Fig. 2b).

Thus, improvements in the participants' gait and ability to perform daily activities, which were induced by VSTD training, were maintained throughout the post-training follow-up period (Table 1).

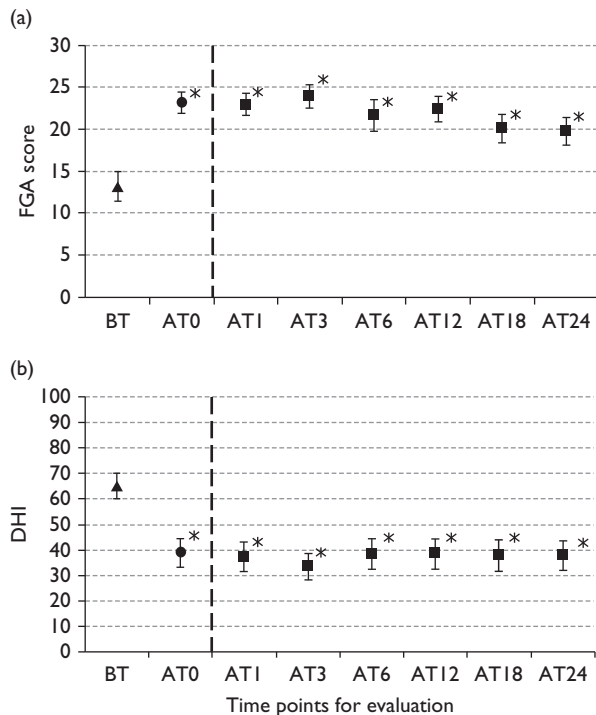
Discussion

Technology based on sensory substitution has the possibility of transmitting impaired sensory information to the brain by another working sensory system [12,13]. Thus, patients with single sensory deficits are able to experience the sense that they have lost using a sensory substitution system. VSTD used in this study transmits lost vestibular information to the brain through the mechanoreceptors on the tongue [6,7]. It is considered that the tongue has several features that make it an appropriate site for the device's interface. The brain region that receives neuronal projections from the tongue is approximately as wide as the region that receives neuronal projections from the fingertips and thus is able

to receive a large amount of tactile information [14]. The voltage required to stimulate the mechanoreceptors on the tongue is only 3% of that required for fingertip stimulation as the tongue is covered with saliva, which is electrically conductive [14]. Therefore, it is possible to transfer a large amount of information to the brain using a low current tactile interface covering a small region of the tongue.

In this study, patients with unilateral vestibular loss and a lack of central vestibular plasticity were trained to use a sensory substitution system that delivered electrotactile stimulation to the tongue as a substitute for vestibular information [6,7]. There have been a few trials for the treatment of balance disorders in which balance information was transmitted through other sensory routes. Several studies described systems for transmitting balance information of the patients' medial-lateral trunk tilt and body movements in pitch and roll acquired by an acceleration-gyro sensor, respectively, to the lower back [15,16] and the outer periphery of the waist [17] in the form of vibrations. In another auditory-vestibular substitution system, stereophonic pitch and volume were used to provide information on the direction and extent of body swaying, which were obtained by a sensor that measured linear acceleration in two dimensions and was attached to a waist belt [18,19]. These studies suggest

Fig. 2



Changes in the FGA score (a) and DHI (b) in patients before VSTD training (BT); after the 8-week training program (AT₀); and at 1 (AT₁), 3 (AT₃), 6 (AT₆), 12 (AT₁₂), 18 (AT₁₈), and 24 (AT₂₄) months after the cessation of the training program. The highest and best obtainable score is 30 in the FGA score. The highest and worst obtainable score is 100 in DHI. Values are expressed as mean \pm SE. * $P < 0.05$ compared with the value at BT. DHI, dizziness handicap inventory; FGA, functional gait assessment; VSTD, vestibular substitution tongue device.

that substitution systems in which vibrotactile sensations or sound are used as substitutes for vestibular information can be used to restore patients' balance. In this study, patients with intractable balance disorders showed improved balance performance after a short period of the VSTD training. As this study obtained results similar to those reported previously [16,17,19], it is suggested that electrotactile stimulation of the tongue is a useful method

for delivering balance information to the brain involving the central vestibular system.

In previous studies examining VSTD [8,9], improvements were observed during the device training sessions, but it was not confirmed whether improvements persisted after the cessation of the training period. In the present study, VSTD training exerted long-term effects on the participants' balance ability, which were maintained for up to 2 years even after the cessation of the training period. It is reported that auditory and tactile stimuli result in the recruitment of the visual cortex, in addition to the auditory and somatosensory cortices, in blind patients [20,21]. Indeed, neuroimaging using positron emission tomography or functional magnetic resonance imaging showed that electrotactile stimulation of the tongue as a substitute for visual information resulted in the activation of the visual cortex in blind patients [22]. These reports suggest that cross-modality interactions among sensory cortices are involved in the recruitment of brain regions related to a lost sensory modality during the processing of substitute information coming from another modality. Tactile information from the tongue is transmitted through the trigeminal nerve directly to the trigeminal nuclei in the brainstem, which are located close to the vestibular nuclei in the brainstem. It is reported that mutual synaptic connections exist between the trigeminal and the vestibular nuclei [23], and that some trigeminal nerve terminals are present within the vestibular nuclei and connect to neurons that project into the vestibular spinal tract [24]. Therefore, it is suggested that close informational links exist between the trigeminal and vestibular nervous systems, leading to crossmodal plasticity in the brain. The crossmodal plasticity might enable the development of a functional network between the central vestibular system and the somatosensory pathway from the tongue, resulting in the reorganization of the vestibular system. Furthermore, patients could learn about the sensorimotor aspects of the vestibular balance system and develop new behavioral strategies for achieving postural stability and gait control. Consequently, the favorable effects of such training

Table 1 Outcome measures of the FGA score and DHI in patients before VSTD training (BT); after the 8-week training program (AT₀); and at 1 (AT₁), 3 (AT₃), 6 (AT₆), 12 (AT₁₂), 18 (AT₁₈), and 24 (AT₂₄) months after the cessation of the training program

Parameters	Time points for evaluation								Repeated-measures ANOVA test (P -value)	
	BT	AT ₀	AT ₁	AT ₃	AT ₆	AT ₁₂	AT ₁₈	AT ₂₄	BT-AT ₂₄	AT ₀ -AT ₂₄
FGA										
Mean (SE)	13.2 (1.8)	23.2* (1.3)	22.9* (1.3)	23.9* (1.4)	21.6* (1.9)	22.3* (1.5)	20.1* (1.7)	19.7* (1.6)	$P < 0.001$	NS ($P = 0.398$)
P -value vs. BT	–	< 0.001	< 0.001	< 0.001	0.001	< 0.001	0.014	0.022		
DHI										
Mean (SE)	65.2 (4.8)	39.1* (5.4)	37.4* (5.7)	33.6* (4.9)	38.6* (5.8)	38.8* (5.7)	38.4* (5.5)	38.4* (5.3)	$P < 0.003$	NS ($P = 0.994$)
P -value vs. BT	–	0.006	0.003	0.001	0.005	0.005	0.003	0.004		

The values of all parameters in post-training periods improved significantly compared with those at BT. No statistically significant differences in the values in each parameter were observed among post-training periods of AT₀, AT₁, AT₃, AT₆, AT₁₂, AT₁₈, and AT₂₄.

–, not measured/not calculated; DHI, dizziness handicap inventory; FGA, functional gait assessment; NS, not significant; VSTD, vestibular substitution tongue device.

* $P < 0.05$ (Dunnett's test): this indicates a significant improvement compared with the value at BT.

might be maintained for long periods after the cessation of the training period.

We cannot exclude the possibility that our findings were because of natural remission because this study did not include a control group. However, before VSTD training, the participants' balance disorders had lasted for 5 years or longer and had not improved at all with conventional vestibular rehabilitation strategies to facilitate vestibular compensation [3]. This indicates that the favorable results obtained in this study are not likely to have been because of natural remission, but were probably because of the effects of VSTD training.

Conclusion

Short-term training with VSTD led to long-term improvements in the balance performance and the daily and social activities of patients with intractable chronic balance dysfunctions. VSTD training might represent a useful rehabilitation therapy in patients with persistent balance disorders because of vestibular deficits and a lack of central vestibular neuroplasticity. However, further research is required to confirm these preliminary results.

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Conflicts of interest

There are no conflicts of interest.

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