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CASE SERIES

Targeting Sensory Systems in the Treatment of Dystonia: Outcomes from a Case Series

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ABSTRACT

Substantial scientific evidence has been gathered on the pathophysiology of dystonia, leading to proposed theoretical treatment protocols for rehabilitating patients with this disorder. This study aimed to assess patient outcomes following the application of novel functional neurology treatment protocols based on basic neuroscience research.

A consecutive case series of patients diagnosed with dystonia and treated in a private practice setting was studied for their response to neural rehabilitation techniques based on published basic neuroscience research. Sensory stimulation techniques including non-invasive spinal cord stimulation, vestibular nerve neuromodulation based on targeted skull vibration and stimulation of reciprocal inhibitory nerve activity of dystonic muscles was used as novel add on therapy to standard care. The subjects included 6 cases of cervical dystonia and 3 cases of more generalized dystonia. The proposed rehabilitation procedures were explained in detail to the patients, including the investigational nature of these procedures, and written informed consent was obtained from each patient prior to treatment.

Based on the comparison of total dystonia scores upon admission and prior to treatment with total dystonia scores at the completion of a course of care, all 9 patients in this case series demonstrated improvement in their dystonic symptoms after treatment. The group average pre-treatment dystonia score was 11.4, while the post-treatment group averaged dystonia score was 5.2 points. This represents a moderate, but clinically important improvement in global dystonia scores in a series of 9 patients treated in a private practice setting.

Considering that dystonia is highly resistant to treatment, the improvement seen in this case series suggests that the methods involved warrant further investigation in a more formal research setting. The clinically meaningful improvements (approximately 50% for the group average) seen in these cases argue for more research into the role of the sensory systems in both the pathophysiology and treatment of dystonic conditions.

Introduction

Dystonia is a movement disorder causing involuntary muscle contractions and abnormal postures/movements. Current treatments like medications, botulinum toxin injections, and deep brain stimulation have limitations and are not universally effective across all subtypes. Some patients experience incomplete symptom relief, adverse effects, or treatment resistance over time. This case series explores sensory stimulation techniques that can temporarily alleviate dystonic symptoms as a potential adjunctive therapy to address the limitations of existing approaches.

Dystonia can be primary (idiopathic) or secondary (due to an underlying condition or insult to the nervous system). Primary dystonia is categorized by age of onset as early-onset (<20 years) or adult-onset. [1] The distribution of affected body regions also varies, with patterns including focal (single body region), segmental (adjacent regions), multifocal, generalized, and hemi-dystonia (one side of the body). Accurate classification of the dystonia subtype is crucial for guiding appropriate treatment approaches and research efforts aimed at improving outcomes.[1,2]

While considered a relatively rare disorder, dystonia's prevalence has been estimated at approximately 16.5 cases per million individuals, [3] depending on the specific subtype and geographic region studied. [4,5] Early-onset forms of primary dystonia typically manifest during childhood or adolescence, with symptoms appearing before age 20. In contrast, adult-onset cases tend to unfold later in life, after an individual reaches adulthood. Certain demographic patterns have also emerged, such as cervical dystonia being more commonly observed in women compared to men. Geographic and ethnic variations play a role as well, particularly for certain genetic forms that may cluster within specific populations, reflecting the influence of genetic and environmental factors.

The pathophysiology of dystonia is complex and not fully understood, but proposed mechanisms include dysfunction in the basal ganglia-thalamocortical circuits, imbalances in neurotransmitter systems (e.g., GABA, dopamine, acetylcholine), abnormalities in sensorimotor integration processes, and genetic factors. Likely, multiple interacting mechanisms contribute to the diverse clinical manifestations observed across different dystonia subtypes. [2,6,7]

Despite the availability of various treatment modalities, a significant proportion of individuals with dystonia continue to experience inadequate symptom control or undesirable side effects from current interventions. Medications may cause adverse effects and lose efficacy over time, while botulinum toxin injections provide only temporary relief, necessitating repeated treatments that can be painful. Deep brain stimulation, though effective in some cases, is an invasive surgical procedure with inherent risks and may not be suitable or successful for all patients. [3]

This underscores the critical need for ongoing research into novel therapeutic approaches that can more effectively and tolerably manage this complex disorder by addressing the underlying pathophysiological mechanisms. Emerging strategies, such as sensory stimulation techniques and further elucidation of dystonia's pathogenesis, hold promise for uncovering new treatment paradigms and improving outcomes for those living with this challenging condition.

An intriguing clinical feature of dystonia is a phenomenon known as the geste antagoniste or "sensory trick." It is characterized by temporary alleviation of dystonic symptoms through specific sensory input, such as gentle touch, pressure vibration, [8] eye movement,[9] potentially TENS [10] as well as galvanic vestibular stimulation[11] applied to the body.

Another frequently reported finding in cases of dystonia is evidence of abnormal inhibitory modulation [6] which has been reported as defective GABAergic function [12] expressing as faulty inhibition in the brain, [13] spine cord [14] and in the abnormal expression of reciprocal inhibition between agonist-antagonist muscle groupings. [15 16]

A novel hypothesis offers a unifying explanation that accounts for these various abnormalities. It proposes the existence of a "neural integrator" for head motor control, analogous to the well-established integrator circuits that hold the eyes steady after a saccadic movement. Just as abnormalities in the ocular integrators lead to nystagmus and drifting eye movements, it is proposed that faulty head neural integrators could be the core deficit in cervical dystonia. [17–19]

Informed by these and similar research findings, the author sought to translate this data into novel treatments for patients presenting for neural rehabilitation of dystonia signs and symptoms. Patients suffering with intrusive dystonic symptoms that had failed to respond to traditional methods of dystonia treatment were offered a novel sensory stimulation approach as an add-on therapy to traditional rehabilitation. The investigational nature

of the treatment was explained and written informed consent was obtained prior to treatment.

Materials and Methods.

Nine cases that had a well-established diagnosis of dystonia and a substantial history of inadequate therapeutic response to medications, botulinum injection and or physical therapy are included in this consecutive case series. They include six cases of cervical dystonia (three male, three female). One female case of juvenile onset hemi-dystonia, a male patient with generalized dystonia and a female patient with generalized paroxysmal dystonia. The age range was 39 to 67 years old. Eight of the nine cases completed at least a six-week course of personalized neurorehabilitation as described below. No adverse events were reported, none of the participants reported worsening of their dystonia. The one drop-out was due to an unrelated medical condition.

Standard physical, orthopedic, and neurological examinations were conducted on each patient. Based on anatomical modeling and physiological assessment of dystonic muscles, we developed a framework to identify primary dystonic muscle dysfunction. By combining published literature on the anatomy of cervical dystonia that relied on use of PET scanning, [20] we formulated specific exercises for individual patients.

The key muscles evaluated through visualization and palpation were the levator scapula, sternocleidomastoid (SCM), posterior neck, and anterior scalene muscles. Abnormal activity in these muscles guided the selection of antagonistic muscle groups to target in the exercises. In those cases of more extensive muscle involvement the same principals were applied to effected regions of the body.

To quantify each patient's baseline presentation, we used a 5-point visual analogue scale to assess pain levels. Posture was assessed using a wall mounted grid and plumb bob, rating postural deviation 1-5. A head-mounted laser (Senmocore TM) was used to track the patients' involuntary dystonic head and neck movements. Patients were positioned eight feet from a vertical target and instructed to keep the laser pointer on the target while being recorded at 20-second intervals. This allowed for a graphical representation of head excursion to be obtained (see Figure 1). The displacement of the laser from the central target was used to measure the amplitude of involuntary head movement on a scale of 1 to 5. These three scores were combined to give an overall symptom score.

The laser-target setup was also used for rehabilitation exercises. Patients were instructed to move the laser dot (head) along patterns printed around the central target. This allowed the author to determine the specific patterns and direction of movement impairments for each patient, as well as observe the effects of various sensory stimulations on normalizing their dystonic movements.

Based on published reports, the author applied vestibular stimulation, [11] tonic eye positions, [9] and non-invasive direct current spinal cord simulation which may increase inhibitory activity within the neuro axis. [21] Recording of these movement tasks under various stimulation conditions made it possible to document any reduction of involuntary muscle contraction and demonstrate any improved motor control attributable to individual or combined sensory stimulations for each case. Any stimulation that appeared to improve motor control was incorporated into an individualized rehabilitation program.

Vestibular skull vibration was applied using a smartphone app (Tone Pacer Pro) and bone conducting headphones (Aftershotz). Different frequencies (100 Hz, 500 Hz) and locations of vibration were observed to see how they affected each patient's dystonic symptoms. The parameters that improved motor control were then incorporated into the rehabilitation program.

Oculomotor assessments were also conducted. When specific eye movements (e.g. smooth pursuit, saccades or tonic gaze).[9,22] were found to modulate a patient's dystonic symptoms, those movements were noted and included in the individualized treatment plan.

Finally, transcutaneous spinal cord stimulation was trialed using a high-voltage galvanic unit (CS Medical Systems CS6102). Electrodes were placed on the patient's back, with the anode at the T11 level and cathode placed over the midline of the sacrum. The intensity was adjusted to the individual's tolerance. Patients were evaluated to determine if this neuromodulation technique improved their posture and/or reduced involuntary movements. [23]

During the physical examination, muscle palpation and visualization of involuntary movements were used to assess and evaluate the affected muscles. Dystonic activity (involuntary muscle contractions) was noted in the patient's chart.

A case-specific rehabilitation strategy was developed based on the principles of reciprocal inhibition and agonist-antagonist muscle

relationships. This approach was used to address the patient's specific impairments.

To address hypertonic (increased tone) in the levator scapulae muscle, the patient was instructed to activate the latissimus dorsi muscle, which is an antagonist muscle. This aims to inhibit the overactive levator scapulae.

For the sternocleidomastoid (SCM) and posterior cervical muscles, the deep cervical flexor muscles, which are known antagonists, were preferentially activated. This helps to counteract the overactivity in the SCM and posterior neck muscles.

The scalene muscles were addressed by having the patient maintain a static posture while activating the semispinalis capitis, semispinalis cervicis, and pectoralis muscle groups. The serratus anterior muscles were also activated, as they are antagonists to the scalene muscles.

Rehab sessions were typically divided into two parts. The first part involves using a head laser setup to train the patient's deficient head and neck movements. The patient is asked to move the laser along preset patterns, and the specific head movements that are determined to be deficient for that individual case are targeted.

Where appropriate, these head movement exercises were augmented by applying a trans-spinal direct current stimulating current during the exercises and or by providing vestibular stimulation through skull vibration.

The combination of therapies that demonstrated the best outcome in terms of improving motor control for the individual patient was then used repeatedly throughout the remaining rehabilitation sessions.

The second phase of rehabilitation involved exercises designed to inhibit hypertonic muscles through activation of their paired antagonist as described above. Similarly, agonist/antagonist muscle exercises were performed, with the addition of either skull vibration or spinal cord stimulation, and or eye movements as described above. The specific combination that showed the superior effect at restoring normal motion was repeated during the course of the rehabilitation sessions.

The specific scoring for each of 9 cases is presented in Table 1. The cases represent 6 cases of cervical dystonia, one case of generalized paroxysmal dystonia, one case of generalized dystonia and one case of hemi dystonia. Based on the comparison of total dystonia scores upon admission and prior to treatment compared with total dystonia scores at the completion of the course of care, all 9 patients in this case series demonstrated improvement in their dystonic symptoms after treatment.

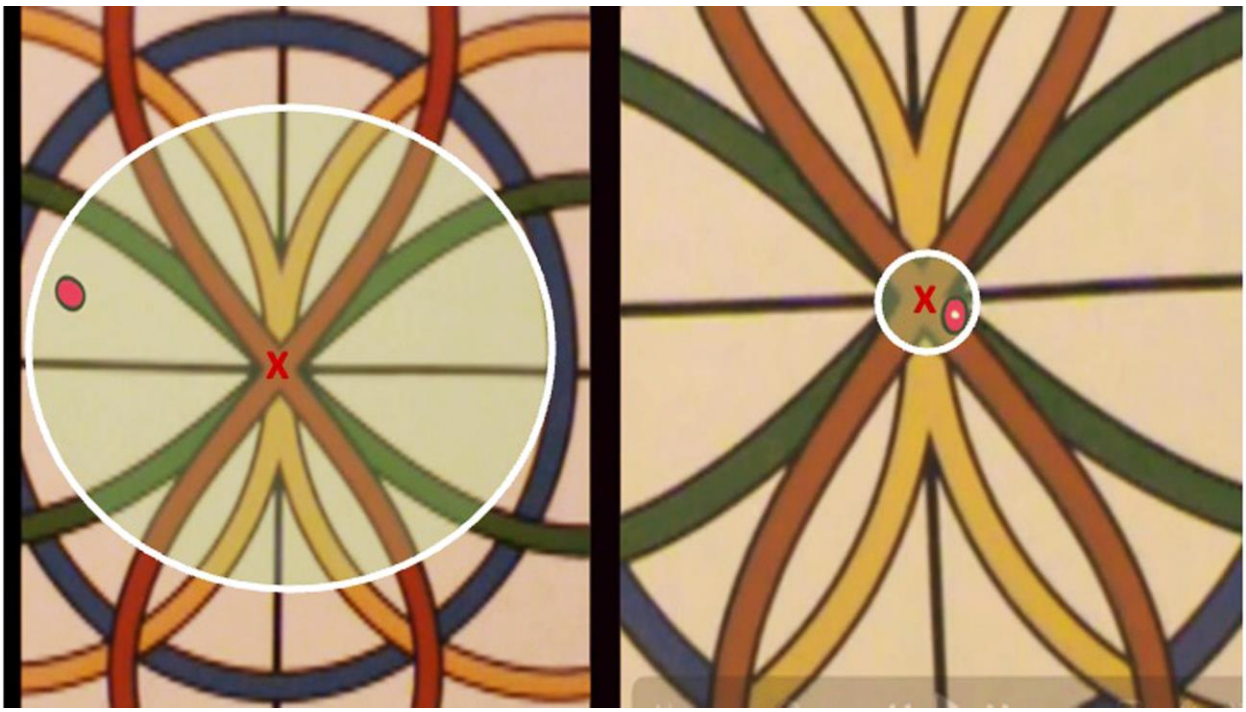

Figure 1: Involuntary head excursion as visualized using a head mounted laser, left head excursion rated 4, same patient after treatment, head excursion rated one. See text for detailed explanation.

		Pre-treatment- a				Post-Treatment -b				
Case#	Type	Posture	Involuntary Movement	Pain	Dystonia Score-a	Posture	Involuntary Movement	Pain	Dystonia Score-b	Delta
1	CD	1	5	1	7	1	3	1	5	-2
2	CD	3	5	5	13	1	3	5	9	-4
3	CD	5	5	5	15	1	1	3	5	-10
4	GPD	1	5	5	11	1	1	3	5	-6
5	CD	3	5	5	13	1	3	5	9	-4
6	GD	5	5	3	13	1	1	1	3	-10
7	CD	3	3	3	9	1	1	1	3	-6
8	CD	3	5	3	11	1	1	1	3	-8
9	HD	3	5	3	11	1	3	1	5	-6
					11.4				5.2	-6.2

Table 1: Five-point scale for abnormal posture, involuntary movement and pain. One is minimal, three is moderate, five is severe. Dystonia score = posture + involuntary movement +pain. Dystonia Score- a is pretreatment, Dystonia Score -b post treatment. CD= cervical dystonia, GPD = generalized paroxysmal, GD = generalized dystonia, HD = hemi dystonia. Delta = Pretreatment-a minus Post-Treatment-b or change in condition after treatment.

Results

Case Number 1: The patient was a 39-year-old male. Upon examination of this patient and the observation of a sensory trick that completely abolished his involuntary movements, he was diagnosed with cervical dystonia. This patient presented with mild, grade 1 resting posture abnormality, his involuntary movement was rated 5 severe, and this pain was rated mild 1, for a total score of 7. Hypertonicity and involuntary motions were observed/and palpated in the SCM muscles bilaterally. Vestibular skull vibration stimulation with tone bursts failed to modulate the patient's dystonic symptoms. Ts-DC modulated his dystonic activity. Eye movement failed to modify his dystonic symptoms. Generalized eye-head movement exercises were prescribed as detailed above in the above. Exercises were administered daily for 2-3 days per week in office and performed as described at home. In office neuromodulation in the form of ts-DC was applied during exercises. Upon completion of this 6-week treatment program the dystonic rating scores were as follows: posture 1, involuntary movement 3, pain 1 for a total score of 5 yielding a 2 point improvement.

Case Number 2: The patient is a 65-year-old male with a 17-year history of cervical dystonia which failed to respond to medical management including botulinum injections. He presented with the resting posture of grade 3 moderate distortion directed to the right of midline. His involuntary movement was rated severe grade 5, he rated his pain as severe grade 5 for a total score of 13. Eye movements with gaze fixed to the patient's upper left field completely stopped dystonic activity, as did 500 Hz skull vibrations bilaterally. Primary dystonic muscle determined through inspection/palpation yielded: Levator scapula muscle bilateral, SCM muscle right, Posterior neck muscles left. In office and home exercises were prescribed as above. During in office sessions ts-DC was applied as described above. Eye movement exercises biased towards the patient's left upper visual field were instituted and combined with 500hz skull vibrations. The patient was seen 18 sessions over a 6 week period. Post-treatment scores were 1, 3 and five, totaling 9 and representing an improvement of a 4 point reduction in symptoms.

Case number 3: The patient is a 49 year-old female with a 4 year history of cervical dystonia that failed to respond to medical management and several courses of neural rehabilitation previous to reporting for treatment at the office. She presented with a severe postural distortion grade 5, deviation to her right. Her involuntary motion score was also severe grade 5 and she rated her pain as severe grade 5, total 15. Her Primary dystonic muscles visualization/palpation were Levator scapula muscle right, SCM muscles left, Posterior neck muscles right, and Anterior scalene muscles right. Trans-spinal direct current diminished dystonic symptoms, as did 500hz skull vibration bilaterally. Eye-movement biased toward her left diminished her symptoms. Treatment was administered 2-5 visits per week for 6 weeks, with in-house and at-home exercises. Post-treatment dystonia scores

were 1, 1, 3 totaling 5 and representing a 10 point reduction in symptoms.

Case number 4: The patient is a 57 year-old female with a 10 year history of cervical dystonia that evolved into generalized paroxysmal dystonia. The patient had inadequate response to medical management including botulinum injections. Resting posture was rated grade 1 mild, her involuntary motion was rated 5 severe because it involved involuntary posturing in the entire body, her pain scores were rated 5 severe. Pretreatment total score of 11 points. Primary dystonic muscles: Levator scapula muscle right, SCM muscles bilateral, Posterior neck muscles bilateral. Eye-movement: Noncontributory. Response to ts-DC positive. Response to 500hz bilateral tone burst vestibular skull vibration, positive. Patient was seen in office and did home therapy 12 times over a 4-week period but dropped out of care due an unrelated medical issue. Post-treatment scores were: 1,1,3 totaling 5 and representing improvement of 6 points.

Case number 5: The patient is a 55-year-old female with a 12 year history of cervical dystonia which failed to respond to medical management including botulinum injection. Her case was complicated by injuries sustained in a motor vehicle accident. Primary dystonic muscles: Levator scapula muscles bilaterally, SCM muscle right, Posterior neck muscles bilaterally. Eye-movement; upper right to lower left suppressed her involuntary head movement. Positive response to 500hz tone and ts-DC. Dystonia score pre-treatment was 3,5,5 totaling 13 and 1,3, 5 post-treatment equaling 9 with a reduction of symptoms of 4 points. Patient was treated 4 times per week for six weeks.

Case number 6: The patient is a 43 year-old male with a decades long history of generalized dystonia which failed to respond to medical management. He required a mechanical assistance device to ambulate. Primary dystonic muscles: Levator scapula muscles bilaterally, SCM muscle right, piriformis and thigh adductor muscles. Eye-movement; non-contributory. Positive response to 500hz tone and ts-DC. Dystonia score pre-treatment was 5,5, 3 totaling 13 and 1,1,1 post-treatment equaling 3 with a reduction of symptoms of 10 points. Patient was treated 3-4 times per week for 6-weeks. Discontinued use of ambulatory assistive device after completing rehabilitation.

Case number 7: The patient is a 67-year-old female with a 9 year history of cervical dystonia which failed to respond to medical management. Primary dystonic muscles: Levator scapula muscles bilaterally, SCM muscle right, Posterior neck muscles bilaterally. Eye-movement; non-contributory. Positive response to left sided 500hz skull vibration and ts-DC. Eye Head and upper body exercises as described above. Dystonia score pre-treatment was 3,3,3 totaling, 9 and 1,1,1 post-treatment equaling 3 with a reduction of symptoms of 6 points. Patient was treated 3-4 times per week for 6-weeks.

Case number. 8: The patient is a 58-year-old male with a 5 year history of cervical dystonia which failed to respond to medical management. Primary dystonic muscles: SCM muscles left, Posterior neck muscles bilaterally. Eye-movement up and to the left, suppressed dystonic muscle activity. Positive response to 500hz skull vibration bilaterally and ts-DC. Dystonia score pre-treatment was 3,5,3 totaling 11 and 1,1,1 post-treatment equaling 3 with a reduction of symptoms of 8 points. Patient was treated 2-4 times per week for 6-weeks.

Case number 9: The patient is a 67-year-old female with a life long history of hemi-dystonia which failed to respond to medical management including botulinum injections. Primary dystonic muscles, right sided hemi-dystonia. Positive response to 500hz skull vibration bilaterally, Eye movements up and to her right and ts-DC. Dystonia score pre-treatment was 3,5,3 totaling 11 and 1,3,1 post-treatment equaling 5 with a reduction of symptoms of 6 points. Patient was treated 2-3 times per week for 6-weeks. Remarkable clinical outcome, able to open door with one hand for the first time since childhood.

The group average pre-treatment dystonia score was 11.4, post-treatment the group averaged dystonia score was 5.2 points. This represents an average group improvement of 6.2 points. Representing clinical improvement of approximately 50% in global dystonia scores in a series of 9 patients treated in a private practice setting.

In. general the sensory neuromodulation stimulation techniques employed in this series of dystonia cases appeared to improve motor function to a greater degree than reducing pain, this was an unexpected observation.

Discussion:

Since dystonia is classified as a movement disorder, it is not too surprising that treatment has been mainly directed towards the motor portion of the nervous system. That sensory input can modulate motor control in dystonia has been appreciated since the sensory trick phenomenon was initially reported in the medical literature. Perhaps because

it was reported as a trick rather than neuromodulation through sensory stimulation, interest in the importance sensory component of the disease has been minimized.

One of the most consistently observed sensory abnormalities in dystonia patients is impaired somatosensory temporal discrimination – the ability to perceive and distinguish the timing of tactile stimuli. [12,24–27]

Through various experimental approaches, researchers have sought to uncover the mechanisms underlying this temporal discrimination deficit in dystonia.

Electrophysiological studies have revealed reduced inhibitory activity within the primary somatosensory cortex, which is thought to contribute to the impaired processing of sensory inputs in the time domain.[12] Functional neuroimaging has further implicated disruptions in brain regions like the putamen and frontal areas, which are likewise involved in temporal discrimination tasks. [25]

Interestingly, this sensory processing deficit appears to be a generalized feature across different types of focal dystonias, affecting various body regions regardless of the distribution or severity of motor symptoms. [26] Moreover, abnormal temporal discrimination thresholds have been observed in unaffected first-degree relatives of dystonia patients, suggesting a potential endophenotype or heritable trait associated with the disorder. [28]

In an intriguing twist, researchers have found that dystonia patients exhibit a paradoxical response to high-frequency repetitive sensory stimulation, which typically enhances cortical inhibition and improves temporal discrimination in healthy individuals. In contrast, dystonia patients demonstrate reduced inhibition and worsening of temporal discrimination following such stimulation. [29] This finding has led to the hypothesis that dystonia patients may have abnormal homeostatic mechanisms regulating inhibitory plasticity within the sensorimotor cortex.

However, low-frequency repetitive sensory stimulation has shown promise in normalizing sensorimotor inhibition and ameliorating temporal discrimination deficits in dystonia patients. This suggests that modulating afferent cortical inputs through specific patterns of sensory stimulation could potentially counteract the sensory processing abnormalities observed in dystonia. [30]

Notably, while deep brain stimulation (DBS) of the globus pallidus internus has been effective in improving motor symptoms in cervical dystonia, it does not appear to correct the underlying temporal discrimination deficits. [31]

This finding underscores the persistence of sensory processing abnormalities despite successful motor treatment and highlights the need for therapeutic approaches targeting sensory deficits directly.

Recent studies the sensory trick in dystonia have shed some light on its mechanisms.

The sensory trick, which involves a combination of active movement and tactile or proprioceptive stimulation, is often the most effective, albeit temporary, remedy for counteracting dystonic postures and movements. This observation strongly suggests that altered sensory processing and sensory-motor integration play a central role in the pathophysiology of dystonia.[8]

Resting-state functional MRI (fMRI) studies have revealed alterations in functional connectivity within the sensorimotor network in CD patients. Patients who experience relief from sensory tricks (CD-trick) exhibit decreased connectivity in the sensorimotor network during the execution of the sensory trick maneuver.

Furthermore, CD-trick patients demonstrate increased activation in the cerebellum during the imagination of sensory trick execution. This finding suggests a potential role for the cerebellum in modulating cortical activity and contributing to the ameliorative effects of sensory tricks. [32]

Electroencephalography (EEG) CD patients with effective sensory tricks exhibit greater alpha and theta desynchronization in the sensorimotor and posterior parietal cortices during the execution of the sensory trick maneuver. These spectral changes correlate with decreased activity in the contralateral sternocleidomastoid muscle.[33–35]

Additional research suggests that input from the sensory trick aids in correcting abnormal posture or movement by providing peripheral sensory feedback. Another proposed mechanism involves the normalization of impaired motor preparation in dystonia, leading to improved dystonic symptoms through modulation of the premotor and primary motor areas. [36]

The observed increase in gamma frequency bands during sensory trick execution may indicate enhanced GABAergic activity, which could contribute to the reduction of dystonic symptoms. [37]

Neuroimaging and electrophysiological studies have highlighted the involvement of sensorimotor networks, the cerebellum, and cortical oscillatory patterns in the efficacy of sensory tricks. Better understanding of these mechanisms may pave the way for the development of novel therapeutic strategies targeting sensory-motor integration in dystonia.

For decades, the underlying cause of cervical dystonia, the most common form of dystonia, has remained elusive. Traditional theories have implicated the basal ganglia, while more contemporary views suggest roles for the cerebellum and proprioceptive feedback.

Shaikh proposed a head neural integrator model by which input signals related to head velocity are converted into signals encoding head position. For stable head positioning, this integrator requires convergent input from the basal ganglia, cerebellum, and proprioceptive feedback from the neck muscles and joints.

If this proposed neural integrator is impaired, either intrinsically or due to abnormal input from any of these sources, it would manifest as the characteristic head drifts and jerky corrective movements seen in cervical dystonia. This unifying theory can reconcile the seemingly disparate basal ganglia, cerebellar, and proprioceptive involvement.[17,18]

Importantly, the neural integrator hypothesis suggests a novel therapeutic approach, modulating the integrator's abnormal activity by altering its faulty sensory signals. If validated, such integrator modulation could provide relief for the millions suffering from this debilitating movement disorder.

One potential sensory interface to the neural integrator is the vestibular system. The literature suggests vestibular stimulation can influence the pathophysiology and reduce the clinical signs and symptoms of cervical dystonia. [11,38–40]

Studies indicate cervical dystonia patients prioritize proprioceptive over sensory information, shifting their reference from head to trunk. This suggests the vestibular system may be involved in the condition's pathophysiology. [39]

Patients with cervical dystonia show greater deviation in the perception of subjective straight-ahead, reflecting body-centered rather than visual-spatial perception. [38,41]

A case report describes a patient who developed cervical dystonia after ear surgery causing vestibular hypofunction, suggesting vestibular dysfunction's role in susceptible patients. [40]

Rosengren demonstrated directly that non-invasive neuromodulation of the vestibular system, via galvanic or acoustic methods, could modulate cervical dystonia symptoms. Acoustic stimulation reduced neck muscle activity by 16-44%, and galvanic stimulation reduced head acceleration by 22.5%, with patients reporting subjective improvement. [11]

These studies form the basis for using vestibular stimulation as an adjunct treatment for cervical dystonia. Stimulation in this case series was achieved via tone bursts of 100 or 500Hz skull vibration, delivered through bone-conducting headphones.

Finally, enhancing GABA mediated neural inhibitory nerve activity by application of direct current stimulation of the skin over the spinal cord was demonstrated by Bocci,[21] suggesting another potential sensory interface with the neural integrator.

Drawing conclusions from a case series has inherent limitations and pitfalls that must be acknowledged. Firstly, case series lack a control group, making it challenging to establish causality or determine the true effect of an intervention. This absence of comparison limits the ability to attribute observed outcomes solely to the intervention under investigation. Additionally, case series often involve a small and potentially biased sample size, which can skew results and limit generalizability to broader populations. Furthermore, the retrospective nature of many case series introduces the risk of recall bias and reliance on incomplete or inaccurate medical records. These limitations underscore the necessity of interpreting findings from case series cautiously and highlight the importance of further research, including controlled trials, to validate observed outcomes and guide clinical decision-making.

Conclusions

The pathophysiology of dystonia remains incompletely understood. Substantial evidence is accumulating that suggests that multiple sensory abnormalities contribute to the symptomatology of the condition and may represent novel avenues for treatment. This case series describes the use of sensory stimulation as an adjunct to standard medical care and rehabilitation in a series of patients who had inadequate response to standard

therapies alone. A clinically meaningful improvement (approximately 50% for the group average) seen in these cases argue for more research into the role of the sensory systems in both the pathophysiology and treatment of dystonic conditions. Considering the fact that dystonia is very resistant to treatment, the improvement seen in this case series suggests that the methods involved warrant further investigation in a more formal research setting.

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