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Neurosurgical Versus Neuromodulation in Writer's Cramps: A Narrative Review

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Abstract

The authors here reviewed the neurosurgical treatment and neuromodulation techniques used for writer's cramps. The basic aim of this review is to shed a light on the current and previous research done for treating writer's cramp. Surgical treatments and deep brain stimulation has been examined in many trials, with positive results. However, sample sizes in the studies were not large, and higher sample sizes in future studies may help researchers better understand how these treatments work in writer's cramps. Furthermore, procedures are frequently required to alleviate writer's cramps incapacitating effects. Although research has shown that many neurorehabilitation and neuromodulation procedures are effective, most of the studies that evaluated these types of techniques had small sample sizes, limiting their widespread adoption. It is important to remember that these therapies necessitate a personalized approach. There is a requirement for multiple trials with the incorporation of various techniques to determine one treatment method that is most reliable and safe for a patient. Surgical treatments for treating writer's cramps such as magnetic resonance-guided targeted ultrasonic thalamotomy, is good for patients suffering from extreme disabling effects, but it comes with the expense of severe side effects. Recent research on neuromodulation, behavioral methods, and sensorimotor training in the treatment of writer's cramps has yielded encouraging results with few adverse effects, but more research is needed before these interventions can be adopted in practice.

Keywords: Neurosurgery, neuromodulation, writer's cramps

Introduction

The phrase "writer's cramp" (WC) is frequently used informally to describe hand pain that occurs after spending a lot of time writing, by analogy to typical muscle cramps.[1] The phrase describes task-specific, focal hand dystonia (FHD), at least among neurologists.[1] According to the definition of dystonia, it is "a movement condition characterized by persistent or intermittent muscle contractions resulting in aberrant, frequently repetitive, motions, postures, or both." Typical dystonic motions are patterned, twisting, and even tremulous. Overflow muscle activation and voluntary action are frequently linked to the onset or worsening of dystonia.[2] A dystonic bodily component typically has a limited range of motion, slow and laborious movements, and usually remains mobile.[3] Adults most frequently have focal dystonia, which affects the neck (cervical dystonia), the eyes (blepharospasm), or a task (e.g., WC).[3] However, dystonia can also manifest in a generalized pattern, which is more typical in varieties that first appear in children.[3] Dystonic symptoms in severe WC affect the dominant hand while writing or performing additional fine motor tasks.[4]

A fascinating feature of a WC is task specificity, which states that only writing is aberrant, and all other tasks are normal. Nearly any work can be impacted by FHD, which is task-specific and not just limited to WCs. Besides typist's cramp and musician's cramp, which includes pianist cramp and flutist cramp, this hand dystonia is frequently associated with certain tasks. Occasionally, when the disease worsens, the task specificity is lost, and the dystonia can interfere with other tasks or even start on its own.[1] Men are slightly more affected than women when it comes to onset in adulthood, typically during the fourth decade. Because many people suffering from WC do not seek medical help, the condition may go unnoticed and unreported WC has been suggested as a possible trigger for FHD. The link between a hand injury and the onset of WC is uncertain; while limb dystonia has been recorded in the aftermath of peripheral trauma, actual causality is difficult to ascertain. The indications for WC are often minimal at first, developing over months. Patients may experience forearm tightness or stiffness during writing, as well as weariness after lengthy periods of writing. Prolonged writing reduces speed and causes cramps. Predominant flexion patterns are associated with greater pressure on the work surface, and the ability is limited to move in a cursive type of writing, with

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those dominant patterns of extending are associated with flickering, trouble trying to put pen on paper, and fingers/thumb lifting off pen.[5]

Nerve conduction investigations and Electromyography (EMG) could be used to diagnose peripheral nerve issues that may be caused by dystonic posturing (e.g., excessive flexion of the wrist and compression of the median nerve).[5] The therapeutic effectiveness of oral medicines is modest. In addition to helping treat tremors, botulinum toxin is recommended as first-line therapy for localized limb dystonia. Only in situations where medical therapy has failed are surgical options available.[6] It is crucial to talk about the patient's treatment goals and expectations (e.g., functional improvement and improved writing abilities, as well as decreased distress, spasm, aberrant posture, and tremor), as well as the person's demands for the task of writing because the mentioned factors can influence treatment modality chosen.[5]

Objectives

The authors wanted to review the neurosurgical treatments and neuromodulation techniques used for WCs. The basic aim of this review is to shed a light on the current and previous research done for treating WC. Patients often want to be treated in a short period with better results or sometimes the symptoms are way more difficult for them to handle. Due to this, they opt for neurosurgical treatment. We want to appraise the articles in which neurosurgical treatments are given and assess their adverse effects. For comparison, we have taken the neuromodulation technique because it has been one of the most understudied techniques with respect to WC. We searched for both invasive and noninvasive neuromodulation technique, which was not previously reviewed. The reviewers wanted to compare the risks and benefits of both neurosurgical and neuromodulation techniques by comparing the current and previous literature (see Figure 1).

Methods

Search Strategy

The search phrases, "writer's cramps" OR "writer cramp," OR "focal hand dystonia" AND "dystonia," AND "neurosurgery" OR "neurosurgical treatment" OR "neurosurgical interventions" OR "surgical treatment," AND "neuromodulation" OR "noninvasive treatment" were used to search PubMed for publications published until January 15, 2023. Randomized controlled trials, case reports, retrospective studies, case series, review articles, and systematic reviews that are published in English are included in this review. Following the literature search, the relevant articles were identified. The research question was further refined through the PICO (Patients, Intervention, Comparison, Outcome) framework which helped in identifying the literature, and Table 1 shows the use of the respective framework.

Main Points

- Magnetic resonance-guided targeted ultrasound thalamotomy and deep brain stimulation are some of the options for treating focal hand dystonia.
- It is important to remember that these therapies necessitate a personalized approach; there is a requirement for multiple trials with the incorporation of various techniques to determine one treatment method that is most reliable and safe for a patient.
- Surgical treatments for treating write's cramps (WCs) include magnetic resonance-guided targeted ultrasonic thalamotomy, is good for patients suffering from extreme disabling effects, but it comes with the expense of severe side effects.
- There is vast evidence showing neuromodulation technique as a treatment with better results and fewer complications in comparison with neurosurgical treatment. Furthermore, there is far more complications seen in patients going through surgery.
- The caveat present with evidence for neuromodulation technique for WCs is that there are not many studies done recently. This is the reason the authors cannot completely conclude neuromodulation as a better option for WC patients.

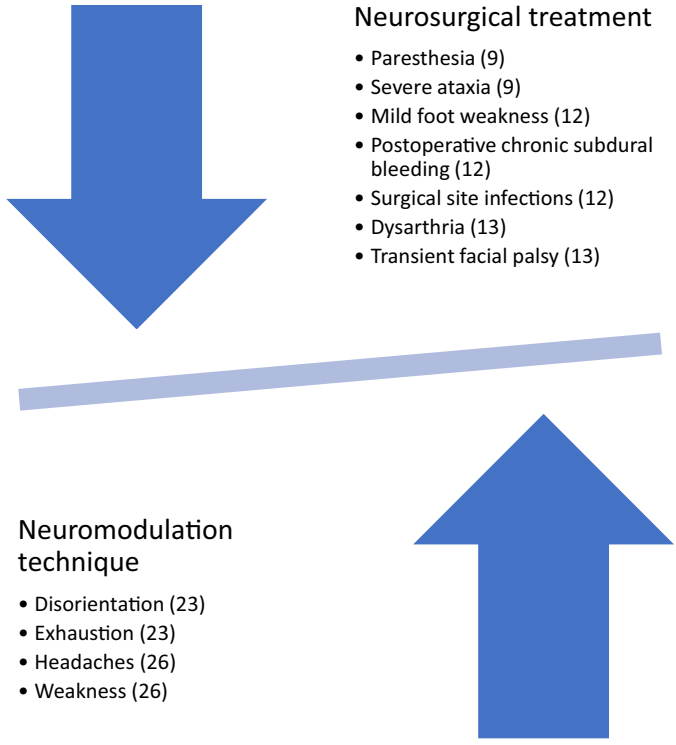

Figure 1. Adverse effects of neurosurgical and neuromodulation treatment.

Types of Treatment Modalities

Neuromodulator stimulatory techniques, neurosurgical, immobilization, intense practice, sensory retraining, adaptive devices, occupational therapy, and physical therapy are only a few of the nonpharmacological treatments for WC and FHD that have been proposed.[6] The treatment methods most often used in the neurological treatment of WCs are Gamma Knife radiosurgery, thalamotomy, surgical targeting with deep brain stimulation (DBS), and Magnetic Resonance-guided Focused Ultrasound Surgery (MRgFUS). According to Bledsoe et al,[7] noninvasive neuromodulation is transcranial magnetic stimulation (TMS), transcranial direct

Table 1. Research Objective Identified Through the PICO Framework

Patients	People suffering from focal hand dystonia (writer's cramps)
Intervention	Neurosurgical treatment
Comparison	Neuromodulation technique
Outcome	Benefits and adverse effects

PICO, Patients, Intervention, Comparison, Outcome.

current stimulation (tDCS), and transcranial alternating current stimulation.

Efficacy

Research has been done on noninvasive stimulatory effects, like tDCS, TMS, or neurosurgical treatments, such as lesions or DBS, for the treatment of WC.[6] According to the International Neuromodulation Society, "To treat disease, neuromodulation uses cutting-edge medical device technology to either increase or decrease the activity of the nervous system. These technologies include implantable and nonimplantable tools that administer electrical, chemical, or other agents to reversibly alter brain and nerve cell function. Unlike pharmaceutical treatments, which are systemic throughout the body, neuromodulation therapies are highly targeted to certain regions of the brain or spinal cord. Highly reversible, permitting doctors to stop treatment right away by taking out the stimulating device in comparison to methods that rely on set intermittent doses, continuous therapeutic compliance"[16] Direct current stimulation and TMS are noninvasive neurostimulator techniques that are utilized in the treatment of a wide range of neurological and mental disorders.[6] The premotor cortex, primary somatosensory cortex, and primary motor cortex have all been targeted using repetitive transcranial magnetic stimulation (rTMS).[6] Both these neurostimulation techniques have the potential to yield not only new insights into the pathophysiology of WC, along with cortical neuroplasticity and general neuromodulation but also make it a noninvasive treatment option for WC.[6] Type-A botulinum toxin was found to be the most prevalent treatment for FHD. It is low risk in comparison to other treatments, and 10%-56% of people stick with it for a long period.

Outcomes

Gamma Knife radiosurgery was performed on the patient with intracranial hemorrhage inside the left cerebrum. Left-handed WC resulted from arteriovenous malformation. When the cramping got worse, he could not write. The patient partly responded to a sensory trick, reporting that it was easier to write when a little object was placed on his left forearm. He said he had no problems using the keyboard or chopsticks.[8] According to Writer's Cramp Rating Scale (WCRS), there are scores from 0 to 30 with 2 subscores, one is the writing movement score (WMS) with a maximum total of 28 scores measuring elbow, wrist, and first 3 fingers movement (2 + 2 + 6), multiplied by the latency of dystonia,[8] adding of writing tremors score (2) multiplied by 2, and other is writing speed score with 2 scores. Higher scores are a sign of more severity. The writer in this case received a 21/30 on the cramp rating scale. He had normal pharyngeal function. Magnetic resonance imaging (MRI) of the partial internal capsule, left temporal lobe, and basal ganglia, revealed bleeding damage. Immediately following surgery, the symptoms subsided. According to an MRI took 1 week after surgery, the target loca tion was coagulated.[8] Another case with no past limb injuries and no relatives with dystonia or tremor was registered with WC. He described using his right hand frequently for handwrit ing and firing weapons. Hand cramping and odd handwriting when writing was the first symptoms, he experienced in his early 1930s. The symptoms were persistent and obtrusive, and they had hampered his ability to write with his dominant hand for the past 9 years. Based on the literature analysis, the patient care conference decided to pursue the ventralis intermedius (VIM) nucleus with surgical targeting of the ventralis anterior/ventra lis posterior complex with DBS. Writer's Cramp Rating Scale motor scores improved significantly from (a baseline score of 18) through intraoperative programming (score of 5), and 4 weeks after surgery (score of 4). At 10 months after surgery, VIM DBS provided long-term benefits.[9] There was single-center case series conducted in Osaka, Japan, including 4 patients with focal dystonia of the right upper limb (1 man and 3 women; ages 25-44) that had ventral-oralis (Vo) complex thalamotomy. All the patients had been experiencing symptoms for more than 2 years, and they were resistant to conventional medical treatment. Neither of the patients consumed their medications as prescribed.[10] According to Sheehy and Marsden's[11] categorization, 2 patients had dystonic WC, while the other 2 had simple WC. All patients' preoperative MRI revealed no abnormal abnormalities. The Vetralis oralis anterior-ventralis oralis posterior (Voa–Vop) nuclei junction was the principal target of contrast-enhanced MRI images of the thalamic Vo nucleus. Following targeted lesioning of the thalamic Vo nucleus, all 4 patients with WC experienced no dystonia symptoms. Preoperative WMS was 23.3 on average (range: 22-26). The patient's symptoms were significantly reduced within 7 days after surgery, as confirmed by neurologi cal evaluations which used the WMS and writing tests. The writing speed improved dramatically, and the WMS results in all patients eventually dropped to 0. Across the follow-up phase (1-2 years), no resurgence of symptoms was recorded.[10] In a retrospective study of 219 individuals with WC and other occupational dystonia who had unilateral Vo-thalamotomy at a single center (Tokyo Women's Medical University Hospital) between October 2003 and February 2017, 138 patients responded well (80.2%), 30 responded partially (17.5%), and 4 did not respond at all (2.3%). After the Vo-thalamotomy, 18 patients (10.5%) had symptom recurrence.[12] In pilot research conducted in Japan from April 2017 to May 2018, 10 patients with FHD were included. In 10 individuals with FHD, they used MRgFUS to thalamotomy the Vo nucleus. At baseline, 3 and 12 months after treatment, they assessed the WCRS.[13] The WCRS score was lowered by 71.4% (range: 0%-100%) after 12 months in 9 patients with WC (6.1 ± 2.9 at baseline vs. 1.8 ± 3.3 after 12 months, $P = .011$).[13] Another case series of 8 patients with medically refractory FHD received Vo nucleus thalamotomy (7 patients) of globus pallidus inter nus (GPi) DBS (1 patient) at an Indian institute between August 2012 and September 2015. All the patients struggled to do their most basic duties. Symptoms lasted anywhere from 6 months to 12 years. All patients experienced immediate surgical alleviation from dystonic symptoms, which lasted throughout the follow-up period.[14] The WMS (range 0-28) recovered from a mean of 14.5 before surgery to 2, while the symptom severity scale(SSS; maximum 43 and minimum 10) increased from a mean of 15.3 before surgery to 2. There were no problems, morbidity, or mortality from the surgery.[14] In 8 patients with focal arm and hand dystonia, stereotactic selective VIM–Vo thalamotomy was performed with depth microrecording.[15] Following VIM lesioning, there was a significant reduction in electromyographic tonic discharges in focal dystonia. The benefits of selective VIM–Vo thalamotomy on focal dystonia were good and long-lasting[15] (see Table 2).

For patients with WC, newer therapy techniques such as noninvasive brain stimulation are being researched. Repetitive TMS and tDCS are the modalities being studied[16-23] In rTMS experiments, researchers activated distinct motor brain areas such as the primary motor cortex, premotor cortex, and supplementary motor area in a single day or 5-day sessions with improvements in handwriting and pen pressure reduction.[17-23] Various study designs were used, primarily with sham control, crossover, or parallel randomization, and were either single- or double-blinded. To improve cortical inhibition, all trials used low-frequency subthreshold rTMS as

Table 2. Neurosurgical Interventional Studies for Writer's Cramps (WCs)

Authors	Interventions	Results
Doshi et al[14]	Ventrooralis thalamotomy (VoT), globus pallidus internus deep brain stimulation (GPi DBS)	Writing movement score (WMS) improved from a mean of 14.5 before surgery to 2, whereas the SSS improved from a mean of 15.3 before surgery to 2 at the last follow-up
Shimizu et al[10]	Selective lesioning of the thalamic Vo nucleus, bilateral Vo DBS, or unilateral Vo DBS with unilateral VoT	The symptoms of dystonia present in 4 patients with WC completely disappeared
Horisawa et al[12]	VoT	7 patients achieved improvement after going through the VoT procedure again
Asahi et al[8]	Lesioning in the contralateral hemisphere (right)	Left hand WC resolved
Hirt et al[9]	Ventralis intermedius (VIM) DBS	There was a noticeable improvement in the WCRS motor scores in refractory WC. At 10 months after surgery, ventralis intermedius (VIM) DBS provided long-term benefits
Horisawa et al[13]	Unilateral magnetic resonance-guided focused ultrasound VoT	Significantly improved the symptoms of FHD, as measured by the WCRS, TMDS, and ADDS

ADDS, Arm Dystonia Disability Scale; DBS, deep brain stimulation; FHD, focal hand dystonia; GPi DBS, globus pallidus internus deep brain stimulation; SSS, symptom severity scale; TMDS, Tubiana Musician's Dystonia Scale; VIM, ventralis intermedius; Vo, ventral-oralis; VoT, ventrooralis thalamotomy; WC, writer's cramp; WCRS, Writer's Cramp Rating Scale; WMS, writing movement score.

an intervention. For proof of concept, early investigations used single-session inhibitory rTMS and assessed the outcome immediately after the intervention.[18,24] More studies[21-25] used extended sessions lasting 5 days or longer and investigated the effect at many time points. Despite the mild results of a single session, it appears that repeated sessions over several days are needed for apparent therapeutic effects. There was case series in which WC has been improved with low-frequency rTMS, but the results were short-lived and contentious.[26] The effects of a long-term low frequency rTMS paradigm on WC symptoms are described in a study done in Italy.[26] The study included a 25-year-old man who had been diagnosed with uncomplicated WC. The WCRS and a 1-minute writing test were used to assess him objectively. They also used EMG wireless equipment to record muscle activation in the upper limb while writing. For a total of 6 months, the series of stimulations was repeated 4 times.[26] With the advancement of the therapy, the patient's clinical condition gradually improved. After completing rTMS, WC symptoms vanished com pletely, and all clinical scores improved significantly. The effects of the treatment continued for up to a year. Additionally, using a paired associative stimulation procedure, researchers discovered a long-term improvement in sensorimotor plasticity. Findings suggested that long-term application of 1 Hz rTMS to PMC is a safe and potentially useful tool for reversing dysfunctional plasticity mechanisms inside the sensory-motor areas of the hemisphere contralateral to the dystonic hand, most likely by regressing dysfunctional plasticity mechanisms inside the sensory-motor areas of the hemisphere contralateral to the dystonic hand.[26] One tDCS study found that anodal, cathodal, or sham tDCS targeting the cerebellum improved writing kinematics and reduced cerebellar inhibition.[27] Neuromodulation approaches may be viable therapy choices for people with FHD, according to the existing data. Park et al[28] investigated the role of combined treatment of WCs patients with botulinum toxin type-A and occupational therapy, comparing it to treatment with botulinum toxin type-A alone. They discovered no differences between the groups in the primary endpoint of patient-rated subjective scale scores at 20 weeks. However, they showed considerable objective improvement in the combined treatment group, with a 28% decrease in WC impairment scale (WCIS) ratings compared to botulinum toxin type-A alone on secondary outcomes.[28] Patients with FHD have received help from rehabilitative treatments such as transcutaneous electrical nerve stimulation (TENS). Tinazzi et al[29] used TENS on 10 patients with WC and found that 3 of the 4 assessments of dystonic dysfunction in all patients improved significantly and that the improvement lasted for 3 weeks (see Table 3).

Complications

Surgery with DBS showed that increasing the amplitude provided even more benefit but at the expense of paresthesia and severe ataxia.[9] Even though none of the patients had any surgical morbidity or death, they all reported temporary (4 days to 1 month) and mild paresthesia of the lower extremities contralateral to lesions, which did not cause any walking difficulties. There was also no evidence of postoperative speech dysfunction.[10] In 6 patients, long-term adverse effects occurred (3.5%).[12] Mild foot weakness (n = 2) was one of them. In 28 patients, temporary negative effects occurred (16.4%). There were no signs or symptoms of brain bleeding. Two patients suffered postoperative chronic subdural bleeding and 2 more developed surgical site infections. Following Vo-thalamotomy, 18 patients had recurrent dystonic symptoms.[12] Three patients experienced temporary dysarthria, while 1 patient's dysarthria lasted for a year. Transient facial palsy affected 2 patients. The internal capsule was encroached on in both cases, resulting in facial palsy.[13] One patient showed signs of transient dysarthria.[15]

One patient suffered from disorientation and exhaustion as these were the 2 mild side effects observed in the rTMS study, both of which lasted less than 1 day.[22] In another study after the rTMS session, 2 patients experienced slight headaches and 1 patient felt weakness.[25] In a randomized controlled study, a prolonged session of cathodal DCS on the motor cortex contralateral to part suffering from WC had no positive clinical effects.[29]

Limitations

Different studies have investigated lesioned procedures and DBS, with positive results. However, the size of the sample in these trials was limited. Furthermore, surgeries are mostly required to counter

Table 3. Neuromodulation Interventional Studies for Writer's Cramps (WCs)

Authors	Interventions	Results
Siebner et al[18]	1 Hz repetitive transcranial magnetic stimulation (rTMS)	Transient improvement in 6 patients
Murase et al[19]	rTMS over the premotor cortex (PMC)	Improved handwriting in the patients with WC
Tinazzi et al[29]	TENS	Showed a significant improvement that persisted for 3 weeks in 3 of the 4 measures
Havrankova et al[21]	1 Hz rTMS of the primary somatosensory cortex	Handwriting improved in most patients, as well as the subjective benefit lasted for 2-3 weeks
Benninger et al[27]	Cathodal transcranial direct current stimulation	Failed to restore normal handwriting kinematics and cortical inhibition
Huang et al[22]	Short-latency Intracortical inhibition (SICI) by rTMS	The session was no different after 5 daily sessions. In contrast, multisession stimulation modulated the motor networks more extensively than a single session
Kimberley et al[23]	Multiple session rTMS	It caused a prolongation of the cortical silent period (CSP) after 3 days of intervention and pen force was reduced on days 1 and 5, leaving other measures unchanged
Bradnam et al[24]	Cerebellar anodal tDCS	Appeared to improve the kinematics of handwriting and circle drawing tasks in FHD
Kimberley et al[25]	TMS paired with sensorimotor training	The findings further suggested a potential longitudinal benefit associated with 2 bouts of rTMS separated by 1 month, as indicated by subjective rating improvements
Naro et al[26]	Long-duration 1 Hz rTMS to PMC of the hemisphere contralateral	Suggested a potential efficacy of long-duration 1 Hz rTMS to PMC of the hemisphere contralateral to the affected hand to improve WC's symptoms

CSP, cortical silent period; FHD, focal hand dystonia; PMC, pre-motor cortex; rTMS, repetitive transcranial magnetic stimulation; tDCS, transcranial direct current stimulation; TENS, transcutaneous electrical nerve stimulation; TMS, transcranial magnetic stimulation; WC, writer's cramp.

the disabling effects of WC. Although studies have indicated that many neurorehabilitation and neuromodulation procedures are beneficial, most of the research evaluating these techniques also had a limited sample size, which limits their widespread use. Larger sample sizes in future studies could help researchers better understand the function of these treatments in FHD. It is crucial to remember that these types of treatments require a tailored approach, so numerous trials with different procedures may be required to find the one that is most suited to a patient's needs. Patients who took botulinum toxin type-A reported a 20%-90% improvement in their symptoms in studies. Inadequate botulinum toxin type-A doses, loss of effectiveness over time due to disease development, the establishment of botulinum toxin type-A resistance, reporting of subjective outcomes, and increasing patients' expectations over time are some of the factors in explaining the recorded wide efficacy range. Despite the widespread use, objective criteria to evaluate clinical outcomes with type-A botulinum toxin are absent, and further studies are needed to create valid and standardized techniques to assess botulinum toxin type-A treatment outcomes. Its efficacy in WCs has been studied in multiple randomized trials, but data on other kinds of FHD are scarce. Its involvement in various types of FHD will need to be investigated more in the future. Deep brain stimulation and direct current stimulation are techniques that have been better for patients to tolerate. The noninvasive nature has also been more suited to patients. Complete resolution of symptoms of WC was seen in neurosurgical treatment.

Conclusion

Magnetic resonance-guided targeted ultrasound thalamotomy and DBS are some of the options for treating FHD. Recent research on neuromodulation, behavioral methods, and sensorimotor training in the treatment of WCs has yielded encouraging results with few adverse effects, but more research is needed before these interventions can be adopted in practice.

There is vast evidence showing the neuromodulation technique as a treatment with better results and fewer complications in comparison with neurosurgical treatment. Furthermore, there are far more complications seen in patients going through surgery. The caveat present with evidence for neuromodulation technique for WCs is that there are not many studies done recently. This is the reason the authors cannot completely conclude neuromodulation is a better option for WCs patients.

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