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Tremor and Other Hyperkinetic Movements

Case Reports

Integration of Osteopathic Manual Treatments in Management of Cervical Dystonia with Tremor: A Case Series

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Abstract

Background: Cervical dystonia, also known as spasmodic torticollis, is a chronic disorder in which patients exhibit involuntary repetitive contractions of neck muscles resulting in abnormal postures or movements. Occasionally, there is also a dystonic head tremor. The underlying mechanisms for cervical dystonia and dystonic tremor are not clear, and treatments are limited.

Case Report: In the present cases, two females with head tremor starting in adolescence developed worsening symptoms of cervical dystonia with dystonic tremor in their 60s. On osteopathic physical examination, both had a vertical type strain to the sphenobasilar synchondrosis.

Discussion: Vertical strains are more frequently found in patients after head trauma, congenital or later in life, than in healthy patients, and head trauma may have been a precipitating factor in these patients. There were improvements in cervical dystonia symptoms, including tremor, in both patients after osteopathic manual treatment.

Keywords: Tremor, torticollis, osteopathic manipulation, craniocerebral trauma, movement disorders, spine, botulinum toxins

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Introduction

Cervical dystonia, also known as spasmodic torticollis, is a chronic disorder in which patients exhibit involuntary repetitive contractions of the neck muscles resulting in abnormal postures or movements.[1] Owing to variability in seeking diagnosis and physician diagnosis, the epidemiology of cervical dystonia has been difficult to ascertain, but this rare disorder has a prevalence of less than 1%. Cervical dystonia varies in severity and affects men and women between the ages of 30 and 50 years old.[2] The exact causes and clinical presentations of this disorder have yet to be fully characterized.

The main treatment for cervical dystonia is an intramuscular injection of botulinum toxin (BTX) every 90 days to decrease muscle spasm. The severity of symptoms affecting quality of life may be assessed with the validated Cervical Dystonia Impact Profile Scale (CDIP-58).[3] Previous reports of the improvement in CDIP-58 from BTX were a mean total score of 46.0 at baseline and 36.2 measured 4 weeks after the first injection ($p=0.0001$).[4] The severity of cervical dystonia with regards to pain, disability, and range of motion is assessed by the Toronto Western Spasmodic Torticollis Rating Scale (TWSTRS), the original or TWSTRS-2.[5] The effect of BTX typically wears off before the end of the 90-day period. Integrating osteopathic manual treatment (OMT) of the affected areas may improve symptoms and quality of life.

OMT is a hands-on approach to treating illness and injury wherein clinical examination of posture, balance, mobility of body regions, and behavior lead to the diagnosis of specific patterns of restrictions and dysfunctions (referred to as somatic dysfunctions) underlying or related to sub-clinical and clinical illnesses. The goal of OMT is to improve structures that inhibit function or homeostasis. The treatment can be used for muscle and joint pain,[6,7] among other ailments such as

migraines.[8] It has been shown in multiple studies that OMT is as effective as a complement, or even an alternative, to drugs or surgery for symptom relief of various conditions.

The following cases are examples of the use of OMT to improve symptoms associated with cervical dystonia with dystonic tremor.

Case report

Case 1

A 65-year-old female was diagnosed with essential tremor involving a no–no (side-to-side) action tremor of the head that began at the age of 13. At age 63, she started experiencing neck stiffness and pain with backwards pulling of her head. Additionally, she developed foot and ankle cramps. Her initial diagnosis was revised to retrocollis type cervical dystonia with dystonic tremor. In previous treatments with BTX, her neck pain and tremor were markedly improved. The effect of the BTX wore off 3–4 weeks before she could receive her next injections.

Additionally, the patient's medical history was positive for chronic left low back pain. She had a family history of Parkinson's disease in her father. She denied any past surgeries, and was not taking any medications. The patient was never a smoker and denied recreational drug use but occasionally had an alcoholic drink. Alcohol did not alleviate the tremor. She worked in jewelry retail, but the tremor did not interfere with her work. There were no other pertinent positives in her review of systems.

On physical examination, her vital signs were normal, except for slightly elevated blood pressure. Neurological examination revealed positive lateral head tremor (no-no) and bilateral upper extremity tremor greater on the left than the right. Mental status was normal. There were no cranial nerve abnormalities. Motor strength was normal, sensory examination was normal, reflexes were symmetric, and cerebellar signs were absent. She ambulated without assistance and exhibited a normal-based gait.

The patient received BTX (rimabotulinumtoxin B) injections using a 30-gauge, ½-inch needle. She received approximately 500 units into each sternocleidomastoid muscle. She received 2,000 units into the bilateral posterior cervical and capitis muscles. She also received 1,000 units into each upper trapezius muscle. A total of 5,000 units was used. The patient tolerated the procedure well.

One week later, the patient reported that the BTX markedly improved her pain and posture. The patient presented for osteopathic manipulative medicine to further improve the neck range of motion, which she felt was limited from full flexion and side-bending to the right. Her CDIP-58 total score was 37.9, and her TWSTRS score was 17. Musculoskeletal examination revealed somatic dysfunction in the head with a superior vertical strain and left torsional strain of the sphenobasilar synchondrosis treated with cranial OMT. Somatic dysfunctions in the neck included C1 rotated$_{left}$, C2 extended-rotated-side-bent$_{right}$, C3-7 side-bent$_{left}$-rotated$_{right}$, and bilateral hypertonic paraspinal muscles. In the thoracic region, T1–10 were side-bent$_{left}$-rotated$_{right}$, with type II dysfunctions of T2 and 3 extended-rotated-side-bent$_{right}$ and T7 flexed-rotated-side-bent$_{right}$. The superior thoracic aperture and shoulder girdle were side-bent$_{right}$-rotated$_{left}$. In the lumbar area, L1–5 were side-bent$_{left}$-rotated$_{right}$. There was a left quadratus lumborum spasm. The spinal dysfunctions were treated with balanced ligamentous tension (BLT), muscle energy, and myofascial release. Her left rib 1 was elevated posteriorly and anteriorly. Her left rib 2 was restricted from going into exhalation motion, which is an osteopathic inhalation dysfunction. Her ribs were treated with muscle energy. She had left abdominal wall muscle hypertonicity that was treated with myofascial release. A left superior innominate shear was treated with BLT followed by muscle energy. A right unilateral sacral flexion was treated with BLT. Restricted left foot mechanics preferred supination and calcaneal inversion that were treated with Still's technique.

Directly after OMT, the patient reported improved comfort in all areas treated, and her neck active range of motion in side-bending to the right had improved. At the 1-week follow-up, the patient reported a decrease in her head tremor and overall pain beyond what she previously experienced with BTX alone. Her CDIP-58 and TWSTRS scores decreased to 32 and 13.

Case 2

A 67-year-old female presented to neurology with a history of essential tremor since she had been a teenager. Since her last neurology appointment, she had several near falls when walking. She recently experienced cramping in her forefingers. She felt the tremor most when lying in bed with her head turned to the left. The patient denied any upper extremity tremor.

The patient's medical history was also positive for lumbar degenerative disc disease leading to chronic back pain, resolved L3–5 herniated nucleus pulposus, prolapsed uterus, and esophageal reflux. The chronic low back pain had been managed with OMT for more than 10 years. She denied any past surgeries or hospitalizations. She had a family history of essential tremor from her father. She was taking the following medications: dexlansoprazole daily, multivitamin daily, vitamin D3 1,000 IU daily, probiotics one capsule three times daily, calcium 1,000 IU daily, vitamin C 500 mg daily, B complex 50 as directed, and ibuprofen as needed. The patient last smoked over 10 years ago. She did not drink alcohol, and her tremor did not improve with alcohol. The patient was a retired physical education teacher. The review of systems was otherwise negative.

Physical examination revealed normal vital signs. Neurological examination revealed prominent head tremor (yes–yes and no–no). There was an increase in head tremor while independently ambulating. She exhibited a normal-based gait with good arm swing bilaterally. There was also an increase in tremor when her head was turned to the right, and a decrease in tremor when turned to the left, both upright and lying down. Mental status was normal and there were no cranial neuropathies. Motor strength, bulk, and tone were normal, and the sensory examination was unremarkable. Reflexes were 2/4 bilaterally, except 1/4 at the Achilles tendon bilaterally. Cerebellar signs were positive for a slight postural tremor on finger-to-nose action bilaterally. The results of general, head and face, eyes, neck, pulmonary,

and cardiovascular examination were unremarkable. The diagnosis was changed to cervical dystonia with dystonic tremor. She refused medications or BTX at the time. Reducing caffeine intake and a trial of OMT for cervical dystonia was recommended.

On consultation for osteopathic manipulative medicine, the musculoskeletal examination revealed a forward head carriage posture with increased cervical lordosis and thoracic kyphosis. There was no evidence of scoliosis. Somatic dysfunctions in the head included suboccipital muscle spasm, left mastooccipital suture restriction with overlying myofascial hypertonicity, superior vertical strain of the sphenobasilar synchondrosis, and left temporal bone exhalation dysfunction with temporalis muscle hypertonicity. The motion of the head on C1 (OA (occipito-atlanto) joint) was limited to a flexed-side-bent$_{left}$-rotated$_{right}$ position. C1 was rotated$_{right}$. C2 was flexed-rotated-side-bent$_{right}$. C5 was extended-side-bent-rotated$_{left}$. There was marked hypertonicity of the sternocleidomastoid muscles and the cervical myofascia. In the thorax, T1 was flexed-side-bent-rotated$_{right}$, T2 and 3 were flexed-rotated-side-bent$_{right}$, and T4–6 and T10–12 were side-bent$_{left}$. There were bilateral rhomboid muscle spasms. The right ribs 10–12 were posteriorly positioned and tender to palpation. There was rib 12 inhalation dysfunction. In the lumbar area, L1 was flexed-side-bent-rotated$_{right}$, L5 was extended-side-bent-rotated$_{left}$, and the right psoas muscle was in spasm. In the pelvic girdle, there was an anterior sacral base, a left posteriorly rotated innominate with superior shear/displacement of the left pubic symphysis, a right iliotibial band spasm, and right piriformis muscle spasm. She had bilateral shoulder internal rotation. In addition, there was paravertebral muscle spasm or hypertonicity in all regions.

The somatic dysfunctions were treated with BLT, myofascial release, muscle energy, fascial unwinding, facilitated positional release, cranial OMT, sacroilio gapping, inhibition, counterstrain, and lumbosacral decompression. The patient was instructed to perform at-home pelvic tilt exercises and use heat as needed for treatment of back muscle spasms.

Directly after OMT, the patient reported a decrease in pain. On the 1-week follow-up, she reported that her family noticed a decrease in her head tremor while playing softball, and she noticed less tremor when laying down to rest.

Discussion

In comparing the two cases, both patients experienced a head tremor beginning in their teenage years. They were not disabled by it, nor did they use any medications to treat it. These patients were able to perform fine motor tasks for activities of daily living and their occupations without difficulty. In addition, they did not experience relief from the tremor with alcohol. Typically in essential tremor, the patient begins experiencing a regular bilateral postural or kinetic tremor of the upper extremities in their 40s.[9] The tremor disappears with a change to supine position and markedly improves or disappears with alcohol.[9] Both patients likely were incorrectly diagnosed with essential tremor. Their diagnosis was adjusted to cervical dystonia with dystonic tremor because of the presentation of a cervical muscle spasm with tremor. Additionally, neither patient was able to identify any beneficial alleviating maneuvers, or sensory tricks, which are often used by people with cervical dystonia to improve head and neck posture.[5] Both patients also had chronic low back pain. Patient 2 had previously benefitted from OMT for her low back, but had not had OMT directed at her neurological symptoms. Patient 1 had a family history of Parkinson's disease in her father, whereas Patient 2 had a family history of essential tremor in her father. It is possible, because of the previous ambiguous nature of the standards for diagnosis for head tremor, that both patients' parents may have had cervical dystonia with tremor as well.

Both patients sought complimentary treatment of OMT for their head and neck symptoms. Notable findings on the osteopathic physical examination included the presence of a vertical strain to the sphenobasilar synchondrosis. Vertical strains occur when rotation of the sphenoid and occiput occur in the same direction about their respective transverse axes. This strain is described for the direction of the superior or inferior shift of the sphenoid base in relation to the adjacent occiput.[10] Of 142 healthy subjects, 3% had vertical strain (1% inferior and 2% superior).[10] However, they have been more frequently reported in adult concussion[11] or traumatic brain injury.[12] Vertical strains were also more prevalent in term and preterm neonatal intensive care unit patients (36.8%), than in healthy newborns (<5%).[13] In the present study, the cervical spine segments with the greatest restrictions were C1, C2, and C7. The normal connective tissue attachments to the cervical spinal dura mater originating from the ligamentum nuchae and rectus capitis posterior minor muscle occur between vertebrae C1–C2 and the occipital bone and C1, respectively.[14] Somatic dysfunction at C2 is associated with somatic dysfunction of the upper thoracic vertebra[15] and sigmoid colon abnormalities.[16]

As expected, the feeling of neck stiffness, tightness, and or pain was improved with OMT because of the relief of muscle spasms. The underlying mechanisms of tremor in cervical dystonia are not known, and the reasons for further improvement in tremor with OMT in both cases are unclear. Cervical dystonia is a neural network disorder that may include messaging from the mechanoreceptors in the joint connective tissues. These patients may have inherited a genetic susceptibility to cervical dystonia that clinically manifested as a result of a vertical-strain-inducing mild trauma occurring at birth or later. Further research is necessary to determine the underlying mechanisms in cervical dystonia with tremor as well as how OMT can be used most effectively to improve symptoms.

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