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CLINICAL EVIDENCE

Cranial optokinetic reflex and cervical dystonia.

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Abstract: A 45-year-old female presented with acute cervical dystonic myotonic spasms and pain of two-day duration, unresponsive to acupuncture and physical therapeutic interventions. Initially her optokinetic reflex system was stimulated to increase dopamine activity related to specific left mesencephalic ocular stimulation. This required an optokinetic strip motion from the subject's right upper visual field diagonally directed toward their left lower visual field. Directing the optokinetic strip on the diagonal was selected by the extraocular muscles innervation pathways. The selection of diagonal right upper to left lower optokinetic activation was specifically chosen to increase the activation and collateral stimulation of the ascending left mesencephalic reticular system and dopaminergic generating centers. This action was intended to regulate and modulate control through the left cerebral cortical-striatal pathways and through the descending medial longitudinal fasciculus to regain inhibitory balance to the output of her right cervical ventral horn cells. Completion of the intervention was determined when her symptoms were reduced by approximately 75%. Home exercises consisted of a download application of optokinetic strips into her iPhone with instructions to direct the optokinetic stimulus diagonally down and away from the side of cervical tension

Indexing Terms: Cervical Dystonia, cranial, optokinetic reflex, optokinetic strip motion

Introduction

Cervical dystonia (CD), also known as spasmodic torticollis (ST), is a focal dystonia characterised by neck muscles contracting involuntarily causing abnormal movements and awkward posture of the head and neck. CD is the most common form of adult-onset focal dystonia, has a heterogeneous clinical presentation with variable clinical features that often can lead to difficulties and delays in diagnosis. The prevalence of CD in the population is estimated to range from 20–4,100 cases/million. (1)

> *... this case report demonstrates a very low-risk approach to resolving Cervical Dystonia using a cranial optokinetic reflex procedure'*

Defazio et al found that with CD generally, '*studies that relied on service-based and record-linkage system data likely underestimated the prevalence of CD, whereas population-based studies suffered from over-ascertainment. The more methodologically robust studies yielded a range of estimates of 28–183 cases/million. Despite the varying prevalence estimates, an approximate 2:1 female:male ratio was consistent among many studies*'. (1) Of the three studies reviewed in the DeFazio et al systematic review they found an estimated incidence of CD, ranging from 8–12 cases/million person-years. (1)

CD is most often primary (sometimes called idiopathic cervical dystonia (ICD)), though it can also be secondary to other neurological disease. Dystonic movements are typically involuntary, spasmodic, and repeated in a stereotype pattern. '*From onset they are often mild and intermittent,*

but become constant with progression. Turning of the head (torticollis) is most frequent, but lateroflexion (latero-), extension (retro-) and flexion (anterocollis) of the neck are also seen. The range of voluntary neck movement is often reduced'. (2)

If the CD contractions are sustained, they may cause abnormal posture of the head and neck. If the spasms are periodic or patterned, they may produce jerky head movements. The severity of cervical dystonia varies from mild to severe. Movements are often partially relieved by a sensory trick (also known as *geste antagoniste*) such as gently touching the chin, other areas of the face, or back of the head. Cervical dystonia may begin in the neck and spread into the shoulders, but the symptoms usually plateau and remain stable within five years of onset.

The pathogenesis of CD remains obscure. However, genetic factors, trauma, the sensory system, and impaired basal ganglia function may all play a role in the development of this disease. (3) Le Doux et al (4) performed a review of the literature studying secondary CD and determined that 'structural lesions associated with cervical dystonia were most commonly localized to the brainstem and cerebellums with the remaining cases were equally divided between the cervical spinal cord and basal ganglia. Although inconsistent, head rotation tended to be contralateral to lesion localisation' (4). Of importance, they found that the '*relative paucity of basal ganglia pathology and concentration of lesions in the brainstem, cerebellum, and cervical spinal cord in patients with secondary cervical dystonia suggests that dysfunction of cerebellar afferent pathways may be important to the pathophysiology of primary cervical dystonia*'. (4)

Aside from primary or secondary types of CD there are also discriminations between whether the CD appeared post-traumatically and if the onset was acute or delayed. In a study by Tarsy, (5) '*patients with cervical dystonia attending a movement disorder clinic were reviewed for history of trauma before onset of symptoms. Patients with symptom onset within 4 weeks of trauma were compared with patients who developed symptoms between 3 months and 1 year after trauma. Acute-onset cervical dystonia was characterized by markedly reduced cervical mobility; prominent shoulder elevation with trapezius hypertrophy in most patients, absence of involuntary movements, sensory tricks, or activation maneuvers; and poor response to botulinum toxin injection. By contrast, delayed-onset cervical dystonia was clinically indistinguishable from non-traumatic idiopathic cervical dystonia*'. (5)

The most common therapeutic option for CD is botulism toxin, (6, 7) but this is not without risks (8, 9, 10) and if it is not adequate to control the patient's CD, anticholinergics, benzodiazepines, *baclofen* and other medications can be used as adjunctive therapy. (11) Low-risk physical modalities that have shown some promise include '*EMG biofeedback training, muscular elongation, postural exercises and electrotherapy*'. (12) Two chiropractic studies found some improvement in CD with one utilising chiropractic manipulative care (13) and the other using a form of Optokinetic reflex therapy, (14) similar to what was used in this current case. CD patients who are refractory to conservative methods, botulism toxin injections, and medications may respond higher risk procedures such as deep brain stimulation (15) and surgical interventions. (11)

While conservative methods offer low-risk options to a patient presenting with CD and pharmaceutical and surgical options offer greater risk, there is a risk to no treatment at all. A review of the literature by Konrad et al (16) found that there are orthopedic and neurological complications that can arise from untreated CD: '*cervical spine degeneration, spondylosis, disk herniation, vertebral subluxations and fractures, radiculopathies and myelopathies*'. (16) They cautioned that awareness '*of the frequent occurrence of complications and screening for symptoms of radiculomyelopathy in patients with dystonia is essential to avoid irreversible spinal cord damage. Complications of cervical dystonia need to be taken into consideration when weighting risks and calculating costs of the disease and its treatment*'. (16)

Case

A 45-year-old female presented with acute cervical dystonic myotonic spasms and pain of two-day duration, unresponsive to acupuncture and physical therapeutic interventions.

Methods/Intervention

The patient's complaint was acute pain and stiffness in her right posterior cervical musculature. Through preliminary examination, her right-sided pain was determined to be registered in her left parietal sensory cortex within the post-central gyrus. From this examination it was considered that her right cervical dystonic spasms were associated with a mismatch between either her left motor-basal ganglionic association or pathways pre- or post- to this system.

The left motor-basal ganglionic association or pathways was chosen because the Parietal lobe is the sensory cortex of the brain and all sensations that are brought to a conscious level can only occur if the parietal lobe is activated. Somatosensory pain or activity in the right side of the body can only be brought to a conscious awareness by the subject if their left parietal Somatosensory region (post-central gyrus) is activated.

Initially her optokinetic reflex system was stimulated to increase dopamine activity related to specific left mesencephalic ocular stimulation. This required an optokinetic strip motion from the subject's right upper visual field diagonally directed toward their left lower visual field. Directing the optokinetic strip on the diagonal was selected by the extraocular muscles innervation pathways.

The selection of diagonal right upper to left lower optokinetic activation was specifically chosen to increase the activation and collateral stimulation of the ascending left mesencephalic reticular system and dopaminergic generating centers. This action was intended to regulate and modulate control through the left cerebral cortical-striatal pathways and through the descending medial longitudinal fasciculus to regain inhibitory balance to the output of her right cervical ventral horn cells.

In general the amount of time and intervals of the optokinetic strip stimulus was based on the patient's metabolic capacity to receive the stimulus. This is commonly determined by pulse rate, blood pressure readings, ability to concentrate on optokinetic stimulation acceptance, activation of blithrospasm manifestations around the eyelids or increased sweating. In this patient's case, the application was administered until sweating was induced and each application was administered one second less than the time it took to induce perspiration.

Completion of the intervention was determined when her symptoms were reduced by approximately 75%.

Home exercises consisted of a download application of optokinetic strips into her iPhone with instructions to direct the optokinetic stimulus diagonally down and away from the side of cervical tension. She was instructed to apply the stimulation for approximately 10-12 cycles and then recheck her neck tension. This procedure was to be reapplied 4-5 times per day.

Results

The patient was treated with cranial optokinetic reflex procedure and had 75% improvement within 2-minutes. She was given the procedure to perform at home and within two-days her CD had completely resolved and has not returned in the past 12-months, as determined at a follow-up assessment.

Discussion

Conservative treatment for primary and secondary CD that offers low risk would be preferred to more high-risk procedures such as surgery, deep brain stimulation, and medications. Generally options for care are weighted toward low-risk and incrementally adding therapy with greater risk as the patient appears refractory to lower-risk options. It is clear that untreated CD also has its risk (16) so extended *'watching and waiting'* may be problematic.

In this report optokinetic stimulation therapy was used to treat acute onset idiopathic cervical dystonia in a 45-year-old female. The hypothesis suggests that the improvement of the patient's CD was related to a re-circuiting of the neural pathways in the basal ganglia, substantia nigra, pontine brainstem, and cerebellum. (17, 18) In Bova and Sergent's therapeutic approach (14) they also similarly employed optokinetic stimulation to achieve a reduction in a patient's cervical dystonic condition.

The rational for the specific care utilised in this specific patient's case involves reviewing some specific neurological pathways(19) to help explore why the optokinetic stimulation appeared to have a positive effect on this patient's CD. Left optokinetic stimulation is believed to be a combined action of left pursuits and right re-fixation saccades. Left pursuits are known to activate the left parietal cortex (sensory cortex). Right refixation saccades utilise the left frontal cortex, which initiates from the frontal eye field and feeds into the basal ganglia as well as the left supplementary and primary motor cortical regions. The right-to-left optokinetic reflex also feeds from olivary climbing fibres directly into the right cerebellum and projects through the efferent copy feedback pathways to the left midbrain dopaminergic generator cells of theSubstantia nigra and ventral tegmentum while in route back to the left cerebral cortex. Therefore, in this patient, the direction of right-to-left optokinetic stimulation was initially selected.

A neurological association has been found between the extra-ocular muscles of the eyes as they relate with nuclei embedded within the brainstem (20) and cerebellar-striatal-cortical regions. Optokinetic stimulation can also affect head position and spatial neglect (21) possibly associated with CD. Utilising these various pathways involved in the optokinetic reflex offers a possible explanation how, in a subset of patients with CD, optokinetic stimulation may help effectively modulate muscular tone. (22, 23)

As with any case report, caution must be used when considering the results of the clinical encounter since with single subject studies we cannot rule out placebo or ideomotor effects, regression to the mean, and other confounders. What is compelling, however, is that the patient was unresponsive to prior interventions (acupuncture and physical modalities) and responded within minutes to the optokinetic reflex therapy.

Conclusion

CD is a life-altering traumatising condition lasting 24 hours a day without relents. Patients with CD are seeking low-risk options that may offer therapeutic value. In this case a patient with early-onset CD unresponsive to acupuncture and physical modalities was treated with cranial optokinetic therapy and had an immediate positive response without any relapse during the ensuing 12 months. While this intervention is not considered an option for CD caused by genetic or diseased atrophy of the basal ganglia, it is hoped that future study will find specific subsets of CD patients who present with spastic conditions caused by diminished or aberrant pathway connectivity that can be helped with optokinetic therapy.

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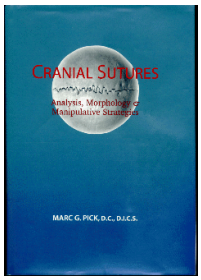

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