Chiropractic Management of Cluster-Tic Syndrome: A Case Report

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ABSTRACT: *Objective:* To report a case of cluster-tic syndrome (CTS), a rare chronic head pain syndrome recognised by the International Headache Society (IHS). This report describes utilisation of chiropractic in a multi-modal management plan for CTS. The case illustrates a cervical association with head pain, a feature that has not previously been reported. The confluence of nociceptive afferents through the trigenminocervical nucleus and trigeminovascular system has been hypothesised to be responsible for the change. *Clinical Features:* This case involves a 61-year-old Caucasian male who has suffered typical cluster headaches for more than 30 years. He recently developed a tic-like pain that overlapped his typical cluster headaches. Cervical spine flexion relieved the headaches and the tic-like pains. *Intervention and Outcome:* The condition was managed with a multi-modal approach that integrated a traditional pharmacological approach and chiropractic, which included cervical and thoracic spinal manipulation therapy (SMT). This approach provided great relief when the pharmacological approach alone did not. *Conclusions:* This paper describes a new form of management for alleviation of cluster-tic syndrome. Further case studies are required to outline the utility of this approach. Future research should investigate the role of the cervical spine in head pain syndromes. Use of allied health professions may provide new opportunities in the management of chronic pain syndromes.

INDEX TERMS: (MeSH): CHIROPRACTIC; HEADACHE; CLUSTER HEADACHE; TIC DISORDERS; MANIPULATION, SPINAL; MANIPULATION, CERVICAL .

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INTRODUCTION

Cluster-tic syndrome (CTS) was first used by Green and Apfelbaum[1] to describe a rare headache that encompassed both a cluster headache (ciliary neuralgia, hemicrania neuralgiformis chronica, migrainous neuralgia of Harris, petrosal neuralgia of Gardiner) and trigeminal neuralgia (tic douloureux). It is recognised by the International Headache Society (IHS) in the most recent classification of headache disorders[2] with categories of headache cluster headache 3.1 and trigeminal neuralgia 13.1 being met for the syndrome to be adequately diagnosed. Cluster headaches are characterised by severe unilateral facial pain located about the orbit, supraorbital area, temporal region, or a combination of these. The duration of attacks ranges from 15 to 180 minutes at a frequency of once to 8 times a day (Table 1). These attacks are accompanied by one or more of the following ipsilateral autonomic symptoms: conjunctival injection, lacrimation, nasal congestion, rhinorrhea, forehead/facial sweating, miosis, ptosis and eyelid oedema. During these attacks, sufferers become highly agitated. The main differentiating features of the cluster portion of this syndrome and other trigeminal autonomic cephalgias are duration and frequency of attacks along with pharmacological relieving agents suggesting a different pathophysiological mechanism.

Classical trigeminal neuralgia is characterised as unilateral electric shock or icepick-like sensations that arrive and end abruptly. They are limited to one division of the trigeminal nerve. These attacks can last from a few seconds to 2 minutes. Trivial stimulation often evokes the attacks, with small areas about the nasolabial fold and/or chin being particularly sensitive.[2] Attacks commonly occur without provocation. A refractory period following attacks exists in which no attack can occur. Trigeminal neuralgia conventionally affects the second and third branches of the trigeminal nucleus.[2] Pain frequently leads to spasm of facial muscles of the affected side.

When cluster headaches and trigeminal neuralgia overlap in the same craniofacial region (area and side), the term *cluster-tic* is applied. The two components of cluster-tic may be present concurrently or exist separately in the same patient.[4] That is, the symptoms of cluster headache may occur simultaneously (concurrently) with trigeminal neuralgia in the same period of time, or may occur separately at different time intervals. Of these two groups of cluster-tic patients, the group without concurrent manifestations appears to be larger at 65% or 28 of 39 patients reported in one paper.[5] The latter group of non-concurrent symptoms has been subdivided further[6] into patients with tic symptoms preceding cluster, coined *neuralgic-vascular type*, and patients with cluster symptoms preceding tic, coined *vascular-neuralgic type.*

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Table 1

DIFFERENTIAL DIAGNOSIS OF CLUSTER HEADACHE AND TRIGEMINAL NEURALGIA[2,3]

History	Cluster Headache—IHS 3.1	Trigeminal Neuralgia (classical) —IHS 13.1
Location	Unilateral: orbital, supra-orbital, temporal (or any combination thereof)	Unilateral: limited to one or more divisions of the trigeminal nerve
Character	Sharp stabbing, hot and boring	Brief electric shock-like pains, abrupt in onset and termination
Intensity	Excruciating	Severe
Frequency	Once to 8 times daily	Up to hundreds daily
Duration	15-180 minutes	Fraction of second to 2 minutes
Provocation	Possibly alcohol, histamine and nitro-glycerine	Trivial stimuli: washing, shaving, smoking, talking and/or brushing teeth. Can occur spontaneously
Associated Symptoms	Ipsilateral: conjunctival injection, lacrimation, nasal congestion, rhinorrhea, forehead/facial sweating, miosis, ptosis, eyelid oedema	Muscle spasm of facial muscles on the affected side

CASE REPORT

A 61-year-old Caucasian male with a 30-year history of cluster headache presented for treatment of a new variant of his cluster headache. For more than 30 years the patient had suffered typical bouts of cluster headache located about the left orbital and supra-orbital area. Autonomic features of these bouts included rhinorrhea, ptosis and eyelid oedema. The cluster periods occurred with staunch regularity twice a year in June and at Christmas time. The duration of cluster periods averaged 8 weeks, with remission periods on average lasting 6 months. The longest cluster-free period recalled was 6 months. These bouts of cluster attack were controlled with moderate but irregular success by use of ergotamine medication. Approximately 18 weeks earlier the patient became alarmed with the changing nature of his headaches. This led him to seek consultation with numerous health care practitioners including his general medical practitioner and a neurologist. The new characteristics were superimposed on those described above. The new features included a sharp pain located in the left ophthalmic region and jaw. The patient reported that onset of the new features of pain could be provoked by stroking areas about the jaw and forehead. He denied bruxism or other motor anomalies of his jaw. The new headaches were accompanied by exacerbation of the other autonomic cluster features. These included lacrimation, conjunctival injection, nasal congestion and sweating from a small area located proximal to the left eyebrow. Interestingly, the patient reported that cervical spine flexion relieved this headache, a phenomenon not previously noted. It was the associated cervical pain that led the patient to visit a chiropractor. The patient is a builder by occupation. He performs heavy manual work and suffers from periodical cervical spinal pain. He denies a recent traumatic event that precipitated his neck pain.

On examination the patient's face appeared "ashen grey" in nature. He appears to be a robust, fit man. Cervical spine range of motion was full in all directions consistent with age. His ROM demonstrated crepitus in rotation and was painful at the end of range in all directions, particularly extension. He was palpably tender in the suboccipital and trapezii regions, left greater than right. Motion palpation revealed the occiput-C1-C2 was fixated on the left and bilaterally at C5-T1 and T3-6. He demonstrated an increased cervicothoracic kyphosis consistent with an upper crossed syndrome. Further palpation revealed no temporal artery tenderness, but stroking the skin of the ophthalmic division of the fifth cranial nerve reproduced sharp pain. He had no other cranial nerve findings that were positive. His blood pressure was reported as normal. No imaging studies were taken.

Prior to treatment the pain was rated at 10/10 on a visual analogue scale. No other outcome measures were taken. Management followed a multi-modal approach. Ergotamine medication was taken in accordance with the previous 18 weeks of prescription up to and during the first chiropractic treatment session. Chiropractic treatment was provided twice a week for 2 weeks. Manual therapy included chiropractic upper cervical (C1, 2) and thoracic (T1-4) manipulation (diversified technique) after appropriate screening tests for vertebrobasilar insufficiency were performed and returned negative responses. A substantial reduction in pain, to 5/10 on the VAS, was reported after the first treatment. Three

days following chiropractic intervention, medications were changed to include: Verapamil (Isoptin) 1 table 3x/day, Ergotamine/Caffeine (Cafergot) 1 table 2x/day, Prednisone 50mg for 2 days, 25 mg for 2 days, 12.5 mg for 2 days, then stop. This course of medication was utilised only once. A combined approach to treatment continued for a further 3 consultations with complete resolution of the combined pain syndromes. Treatment was discontinued by the patient after that time, as symptoms had resolved.

At 6-month follow-up, there had been no return of the new cluster tick-like symptoms, and the patient had not experienced a cluster headache period. Ongoing suboccipital and cervicothoracic pain has continued on an episodic basis over time. The presentation at 6 months was for the management of the neck pain rather than the cluster variant headache. The patient again presented one year later with suboccipital head pain and mechanical low back pain (non-specific sprain strain injury of the lumbosacral spinal region). He reported that he had no return of the cluster tic symptoms and has not experienced a cluster period since previous contact. He reported that his neck had been relatively good during that period. Follow-up at 18 months revealed no return of cluster tic symptoms or cluster headache periods.

DISCUSSION

There are only 45 reported cases of CTS in the literature.[1,5-7] There are no previous reports of CTS in the chiropractic literature. Leone *et al.*[4] advocate the use of cerebral MRI following diagnosis of CTS to exclude such pathology as pituitary adenoma, vascular ectasia and tumours of the posterior fossa, which have been implicated in causing similar underlying atypical cluster-tic headaches.[4,8,9]

Pathophysiological mechanisms of cluster headache, trigeminal neuralgia and the association of both are known as cluster-tic syndrome. They exist on a theoretical level and are yet to be validated. Theoretical models for cluster headaches revolve about autonomic, neurohormonal, chronobiological, autoregulatory and neuropeptide systems.[10,11] Recognised causes of trigeminal neuralgia include vascular compression of the trigeminal nerve, demyelination of the trigeminal nerve, multiple sclerosis, trigeminal compression due to tumour and cavities of the jaw.[12] Current literature suggests a superimposition of aetiology in cluster-tic syndrome.[5,8,13,14] To date there are no reports of a cervical association in CTS.

Treatment

Under the most recent classification of CTS the IHS states that both the cluster and tic components of the syndrome must be treated for the patient to be pain-free.[2] Management of this case followed this edict and combined pharmacotherapy and chiropractic treatment. Treatment of CTS classically begins with a pharmacotherapy approach and progresses to a surgical intervention if pharmacotherapy is unsuccessful. Carbamazepine (tegretol) is indicated in the treatment of trigeminal neuralgia and has largely been the first-line option in cluster-tic therapy.[5,13-16] It is classed as an anticonvulsant, antimanic, antineuralgic and antipsychotic. Recommended starting dosage is 100mg orally twice a day, with maintenance at 200-400mg twice daily.[17] Aclofen, Phyention/Fosphenyton and Clonazepan have been used where the tic portion was not controlled by Carbamazepine alone. They are classed as muscle relaxants, anticonvulsants and anxiolytics respectivly.[17] Prophylactic treatment of the cluster portion has been *via* lithium carbonate, verapmil, ergotamine-tartrate, sumatriptan and corticosteroids. Surgical intervention in cluster-tic patients has been largely directed to the trigeminal association. Such interventions include Gasserian ganglion thermo-coagulation,[13] microvascular decompression of the trigeminal nerve/Gasserian ganglion,[8.16.18] and subtemporal sectioning of the trigeminal sensory root.[13] Surgical treatment is sought when conservative methods prove ineffective. Thermocoagulation and sectioning techniques invariably aim to damage the sensory roots/parasympathetic ganglion, which carried pain sensation, while microvascular decompression alters the course of aberrant/ectopic blood vessels. It has been suggested that the vascular-neuralgic type of cluster-tic patients as seen in this case may not respond to carbamazepine and conservative surgical procedures.[6,13] To date there are no reports of manual therapy being utilised in the treatment of CTS.

Mechanism

A lesion of the trigeminal sensory pathways has been implicated in cluster-tic attacks.[13] Exetroceptive fibres originate from the anterior portion of the scalp, nasal cavity and dura mater of most of the cranial cavity and terminate in the trigeminal nucleus. Current theory suggests disruption of small myelinated produces the tic-like pains, while disruption of unmyelinated tracts tends to produce cluster headaches.[19] These fibres are part of the trigeminovascular system, a system that is frequently implicated in migraine headaches.[10,11,20] The trigeminovascular system theory assumes nociceptive stimulation of trigeminal pathways modifies the activity of autoregulatory, neuropeptide and autonomic system functions.[10]

The trigeminocervical pathway receives touch and pain input from both the trigeminal distribution and the upper 3 cervical nerves.[3] Nociceptive stimuli arising from skin, muscle, joints and mucous membrane in the trigeminal and/or upper cervical sensory distributions converge on the trigeminocervical nucleus. Nociceptive stimuli arising from trigeminocervical sensory distributions may be a source of nociceptive input into the trigeminovascular system, and nociceptive stimuli from the trigeminocervical nucleus distribution converges on second-order neurons in the trigeminovascular system.[3] This convergence has the potential to represent neck and head pain in the same combined distribution of the head *via* the trigeminocervical nucleus.[3.21] This is not an implausible concept, as other headache syndromes are hypothesised to arise from or produce changes in neck-based somatic structures and distributions.[22]

It has been suggested that this association of cluster and tic symptomatology is more than coincidental,[23] raising attention to the possibility that cluster-tic syndrome is its own clinical entity with a distinct aetiology.[5,8,13,14,24] This new entity is thought to have a slightly larger female predominance,[6] a tendency to affect the first division of the trigeminal nerve, and a larger age range that usually affects those between 20 and 70 years.[13] Large epidemiological studies are required to elucidate exact incidence and prevalence data.

Table 2

DIFFERENTIAL DIAGNOSIS BASED ON GENDER AND AGE[6,10,11,13]

History	Cluster Headache	Trigeminal Neuralgia	Cluster-Tic Syndrome
Sex (M:F)	5:1	1:2	2:3
Age at onset (yrs)	20-40	50-60	20-70

This case reports a CTS patient who is classed as a vascular neuralgic type. The symptoms began non-concurrently, and the cluster pain preceded the tic symptoms. This type of patient is thought not to respond to carbamazepine and conservative surgical procedures.[6,13] As reported by others,[6,13] our patient experienced an exacerbation of autonomic symptoms with the introduction of the tic headache. Unique to this case presentation was the fact that the patient reported that cervical spine motion was an alleviating factor to cluster-tic symptoms. This is the first time that such an alleviation of symptoms has been reported in the literature. This observation raises further questions regarding the pathophysiological mechanisms of the syndrome and adds indirect evidence to claims that at least some of the mechanism is cervicogenic in nature. Indeed, this case may have revealed a very rare variant of cervicogenic CTS. Somatic referral has been suggested as the most common cause of head pain.[2] Its role in complex pain syndromes should be further explored.

Efficacy of Treatment

To our knowledge this is the first reported case of cluster-tic syndrome that has responded favourably to a combination of pharmacological and manual methods. Previous osteopathic methods of managing trigeminal neuralgia have been reported as helpful.[25] Two recent publications advocate different management strategies. These report the use of transcutaneous electrical nerve stimulation (TENS) in the management of trigeminal neuralgia and facial pain.[26,27] It is thought that this modality is effective in closing the theoretical pain gate first proposed by Melzack and Wall in 1965.[7,28] Thorsen and Lumsden[27] suggest this form of therapy should be considered first before medication and surgery, as there are no likely adverse effects to non-invasive therapy compared with the pharmacological and surgical interventions. Large clinical outcome studies are required to validate such an approach.

In addition to other physical therapies, the efficacy of exercise should be further explored in chronic head pain syndromes, particularly where a cervical association is suspected. It is thought that the combination of manipulation/mobilisation and exercise is more effective than manipulation/mobilisation alone in treating mechanical neck disorders with or without headache.[29]

A recent Cochrane review[30] all but validated the efficacy of physical treatments, particularly SMT, in the treatment of chronic/recurrent headaches. It was stated that physical treatments may be as effective as prophylactic treatments. The headaches reviewed were migraine, tension-type, cervicogenic, a mix of migraine and tension-type and post-traumatic headache, all of which are thought to have a cervical association. In this case a combined approach of prophylactic pharmacotherapy and manual therapy makes it difficult to ascribe the chiropractor's intervention as the decisive treatment, as only 50% reduction of symptom intensity was noted prior to medication change. It should be anticipated that the medications had some effect. In addition to the patient's correlation of neck pain and headache that led the patient to seek chiropractic care may have biased the patient's expectations for manual therapy in a positive fashion. As the longest cluster-free period for the patient in this case was 6 months, the possibility that the headache just went into remission is unlikely. Given the efficacy of physical therapy in the management of headaches with a cervical association, a trial of manual therapy for CTS patients with a cervical association seems warranted. Future efforts should be directed towards the clinical effectiveness and cost effectiveness of pharmacotherapy techniques *versus* manual therapy.

Side Effects of Treatment

This report demonstrates the successful integration of allied health modalities in the successful management of the complex pain of cluster-tic syndrome. Methods utilised by manual therapists, such as massage, mobilisation, exercise and TENS present few side effects. Another method of management commonly used by manual therapists (particularly chiropractors) is cervical spine manipulation. This method of management is associated with the potential for serious complications.[31-35] Serious complications are considered relatively rare, being estimated at 1 in 20,000 to 5 in 10 million.[36] Raising significant attention within the recent literature is the association of cervical manipulation with vertebrobasilar artery dissection. This is estimated to occur once in 1.3 million treatment sessions.[33] Powell suggests 6 risk factors that can be associated with complications of SMT. These include misdiagnosis, failure to recognise onset or progression of neurological signs or symptoms, improper technique, SMT performed in the presence of a coagulation disorder or herniated nucleus pulposus, and manipulation of the cervical spine.[34] These risks should be balanced with the potential for risk with current pharmacological and surgical methods. Serious adverse effects, including fatality, have been reported during use of carbamazepine.[37-42] Carbamazepine has adverse effects on almost every body system, ranging from bone marrow function, hepatic function, kidney function, ophthalmic change, plasma levels, dermatologic effects, urinary retention and increased intraocular pressure, cardiovascular and musculoskeletal effects.[17] It is also associated with a rare (1 in 1,000 to 1 in 10,000) potentially fatal hypersensitivity reaction,[38] potentially fatal drug interactions,[42] potentially fatal in cases of overdose,[39,40] and is identified with birth defects when administered to pregnant women.[17] Reports of surgical complications are relatively

mild in comparison. They include temporary and permanent paresis and/or paraesthesia with or without change in head or face pain.[43,44] Future management strategies of CTS patients should consider the risk factors associated with therapy option.

CONCLUSION

This paper presents a case of chiropractic co-management of CTS. It introduces the concept of a multi-modal (pharmacotherapy and manual therapy) approach in the management of this debilitating condition. Current literature on CTS is limited to reporting the outcome of a small number of case reports. The pathophysiological pathways of CTS are not well understood or validated, and this case suggests the possibility that nociceptive input from the trigeminocervical pathway may contribute to this chronic pain syndrome. Current management suggests pharmacotherapy is the therapy of choice, but it is associated with unwelcome side effects, particularly in high dosage. The combination of traditional pharmacotherapy and manual therapy in this case provided significant relief and resolution to the syndrome, while the previous pharmacotherapy approach alone seemed to be less effective. Future efforts should investigate the aetiology and management of head and facial pain. Given the small number of reports on this syndrome and its management in the literature, further studies of an epidemiological (survey) and clinical outcome (case control) are required to validate the changes noted in this report. Based on this report, a trial of manual therapy should be considered, in addition to the normal interventions, in management of CTS.

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Letter to the Editors

APPRECIATING A LEGACY WITHOUT PREJUDICE

To the Editors:

I was quite pleased by and agreed with most of what appears in your recent editorial, *Appreciating a Legacy Without Prejudice*.[1] In particular, I think that you said a lot when relating the two-edged sword of loyalty and heroism, on the one side, to "dogged fundamentalism" on the other. It seems to me that the passion and commitment to principle that permitted the chiropractic profession to survive despite intense persecution from political medicine has indeed become a liability in this age of accountability and at a time when doctors of chiropractic seek integration within the health care systems of various nations.

I also felt, however, that your editorial "crossed the line" and perhaps epitomises the problem of seeing what we wish to believe when you discuss the outcomes of the B.J. Palmer Clinic, which operated for more than 25 years. You wrote, "Results were impressive, as the collection of discarded braces, casts and mobility aids adorning some of the clinic walls attested." I am reminded of the sceptic who visited the cathedral at Lourdes, France. His reaction to the numerous crutches and canes adorning the pillars of this impressive house of worship was to ask, "Where are the crutches of those who were not healed?" I suggest that displays of discarded crutches and canes suffer the same selection bias as do case studies. Valuable as hypothesis-generators and possibility-illustrators, case reports of chiropractic care prepared by chiropractors nonetheless tend to over-report positive outcomes, while case reports of chiropractic care prepared by allopathic doctors tend to over-report negative outcomes. It is only human nature, I suppose, to report that evidence which serves to buttress our beliefs (hence the value of random sampling and random assignment). It seems not only true that seeing is believing, but also that believing is seeing. I advise against being unduly impressed by collections of crutches and canes.

Given available outcome data from the B.J. Palmer Clinic, I am unable to confirm or refute the benefits attributed to the care provided therein. My sense is that Dr Palmer never bothered to determine the rules of evidence in clinical research before setting out to collect the mountaian of outcomes data that constitute the legacy of that facility.[2,3] It can be fairly argued that the rules of evidence we employ today were embryonic in his time,[4] but it seems also the case that Palmer ignored what was available.[5] For example, I found no controlled clinical trials in Palmer's 1951 *Chiropractic Clinical Controlled Research*;[6] I suspect that B.J. was often more concerned with the sizzle than the steak. It may be that some or all or none of the patients seen in this facility during 1935-1961 came in limping and departed dancing *because* of the care received—but such attributions are not justified by the data I have encountered. Neither can I assert that any of them were harmed by chiropractic adjustment. We just don't know.

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