Case reports

Conservative care of temporomandibular joint disorder in a 35-year-old patient with spinal muscular atrophy type III: a case study[☆]

Sébastien Houle DC^a, Martin Descarreaux DC, PhD^{b,*}

^aMaster's Degree Student, Biophysique et biologie cellulaires, Département de Chimie-Biologie, Université du Québec à Trois-Rivières, QC, Canada G9A 5H7

^bProfessor, Département de Chiropratique, Université du Québec à Trois-Rivières, QC, Canada G9A 5H7

Received 25 May 2009; received in revised form 7 July 2009; accepted 9 July 2009

Key indexing terms:

Temporomandibular
joint disorders;
Chiropractic;
Muscular atrophy,
Spinal;
Musculoskeletal
manipulations

Abstract

Objective: This article describes the chiropractic clinical management and therapeutic benefits accruing to a patient with temporomandibular joint (TMJ) disorder and spinal muscular atrophy type III.

Clinical Features: A 35-year-old white man presented at the university chiropractic outpatient clinic with a complaint of masseter muscle pain and mouth-opening restriction. Temporomandibular joint range of motion evaluation revealed restricted opening (11 mm interincisival), and pain was rated by the patient at an intensity of 5 on a pain scale of 0 to 10.

Intervention and Outcome: Chiropractic care was provided and included TMJ mobilization, myofascial therapy, trigger point therapy, and light spinal mobilizations of the upper cervical vertebrae. Final evaluation of TMJ range of motion showed active opening of 12 mm with absence of pain and muscle tenderness of the jaw.

Conclusion: This case suggests that a patient with musculoskeletal disorders related to underlying neurodegenerative pathologies may benefit from chiropractic management adapted to their condition. In the present case, chiropractic treatment of the TMJ represented a viable, low-cost approach with limited adverse effects compared with surgery.

© 2009 National University of Health Sciences.

Introduction

The typical chiropractic patient usually comes into the office with a musculoskeletal disorder, in which case a consensus for standard care already exists. In fact, 76% of patients consulting in chiropractic have a back-related condition.¹ Consequently, the literature

[☆] No funding sources or conflicts of interest were reported for this study.

* Corresponding author. Département de Chiropratique, 3613 Pavillon de Chiropratique, Université du Québec à Trois-Rivières, Trois-Rivières, QC, Canada G9A 5H7. Tel.: +1 819 376 5011x3977; fax: +1 819 376 5204.

E-mail address: martin.descarreaux@uqtr.ca (M. Descarreaux).

regarding chiropractic management of patients with neuromuscular pathology is scarce. However, increasing numbers of these patients consult in complementary and alternative medicine; and studies show that up to 80% of multiple sclerosis patients, 54% of amyotrophic lateral sclerosis patients in Germany, and 40% of Parkinson disease patients report the use of complementary and alternative medicines.² Given their expertise in managing neuromusculoskeletal conditions, chiropractors are likely to be involved in the treatment of patients whose musculoskeletal disorder is related to an underlying pathology.

This article describes the case of a patient with temporomandibular joint (TMJ) disorder and spinal muscular atrophy (SMA) type III who sought chiropractic care. Spinal muscular atrophies, a group of diseases marked by progressive degeneration of spinal motor neurons, are among the most common neuromuscular hereditary disorders, with an incidence of 8 in 100 000.^{3,4} With a prevalence of 1 in 25 000 live births, SMA is the second most common disease in childhood and the most common fatal recessive muscular disease.⁵ Despite some phenotypic variations, the defect in SMAs maps on chromosome 5; but several clinical forms exist.⁶ Spinal muscular atrophy type III is also known as *Kugelberg-Welander disease*, a condition usually characterized by onset after age 18 months, symmetrical muscle weakness of the limbs and trunk, more proximal than distal, and weaker lower than upper limbs. In SMA type III, life expectancy is most likely normal; and the ability to walk can be achieved. The clinical criteria of SMA type III also include tongue fasciculations and hand tremors, as in other types. The clinical criteria of the other types are

described in Table 1.⁷ Other specific findings in SMA include external ophthalmoplegia, facial diplegia, and early respiratory insufficiency in rare cases.⁷ Impairments of the facial musculature can also be found. Such impairments of facial muscles usually result in weakened mastication and TMJ disorders.⁸

Temporomandibular joint disorders (estimated to be prevalent in 20% of the general population) seem to have a higher incidence in SMA patients because one third of them present feeding-related issues, limited mouth opening, and mastication problems.⁸⁻¹⁰ Pain at the TMJ level and across the masticatory muscles is the most common symptom of TMJ disorders; the pathophysiology and etiology of chronic symptoms are still obscure.¹¹ Spinal muscular atrophy patients have abnormal craniofacial growth patterns usually associated with weak jaw muscles.³ Prior studies have shown that the masticatory muscles of SMA patients are weakened, less efficient, and fatigued more rapidly than those in unaffected control subjects.¹⁰ Moreover, mandibular movements seem to be limited in SMA patients compared with control subjects.¹⁰ Mandibular restrictions and weaknesses affect the biting and chewing abilities of SMA patients and can lead to prolonged mealtimes and fatigue.¹² In severe cases, mandibular problems can be responsible for serious conditions, such as chronic malnutrition that can lead to exacerbation of the neuromuscular disease.¹³ Usual chiropractic management of the TMJ might include chiropractic manipulative therapy, the myofascial release technique, activator adjustments, hot and cold packs, and soft tissue massage. Diet, lifestyle advice, and some exercises of the jaw are usually prescribed.¹¹ Activator adjustments have seemed to correct aberrant

Table 1 Clinical criteria of SMA types⁷

Type	Age at Onset	Muscle Weakness	Other Associated Features	Course	Life Expectancy
I	Prenatal to 6 mo	Trunk and limb muscle weakness Symmetrical Wasting is often not conspicuous	Tongue fasciculations and hand tremors	Arrested development of motor milestones Never able to sit without support	Less than 2 y
II	Before 18 mo	Trunk and limb muscle weakness Symmetrical	Tongue fasciculations and hand tremors Hand tremors are frequently observed in SMA types II and III.	Arrested development of motor milestones Unable to stand or walk without aid	Adolescence or adulthood
III	After 18 mo	Trunk and limb muscle weakness Symmetrical	Tongue fasciculations and hand tremors Hand tremors are frequently observed in SMA types II and III.	Ability to walk will be achieved.	Most likely normal

biomechanics of the TMJ in several cases.¹⁴ In addition, chiropractic manipulations have been effective in the treatment of TMJ disorders such as anteriorly displaced temporomandibular disk with adhesion.¹⁵ Studies have reported that manual therapy improves oral opening and diminishes pain in patients with arthrogenous TMJ disorders.¹⁶ To our knowledge, no data are currently available regarding the chiropractic clinical management of TMJ disorders related to neurodegenerative diseases. Therefore, the purpose of the study was to describe the chiropractic clinical management and therapeutic benefits accruing to a patient with TMJ disorder and SMA type III.

Case report

A 35-year-old white man presented to the university chiropractic outpatient clinic with a complaint of

Fig 1. Joint mobilization of the TMJ in distraction. This mobilization is done by pulling down the mandible with contact on the lower teeth and without muscular contraction by the patient.

Fig 2. Joint mobilization in lateral deviation. This mobilization is performed by applying a lateral-medial and downward force vector to the mandibulae with contact on the ramus mandibulae.

masseteric pain and mouth-opening restriction. The pain appeared insidiously and was constant for the last 8 years. The patient described his jaw as being generally stiff, and active mouth opening was restricted. Pain was rated by the patient at an intensity of 5 on a pain scale of 0 to 10. Aggravating factors included long speeches, mastication, eating, and fatigue. The patient was relieved by celecoxib 100 mg, a nonsteroidal anti-inflammatory drug, and rest. He had not received any earlier medical or chiropractic care for his TMJ condition. His medical history revealed SMA type III (Kugelberg-Welander disease). Family history was normal.

The patient was confined to a wheelchair because of his clinical condition and limited mobility. Therefore, postural evaluation was not formally conducted; and neurologic examination was limited to cranial nerve assessment. A brief physical examination revealed that the patient also had neuromuscular scoliosis.

Temporomandibular joint range of motion (ROM) evaluation disclosed painless, restricted opening

(11 mm interincisival). Generally, an opening less than 30 mm is considered as malocclusion.¹⁷ Assessment of the trigeminal nerve motor portion (cranial nerve V) showed fasciculation of the left masseter muscle and decreased mouth-closure force on the left side. Evaluation of the accessory nerve (cranial nerve XI) divulged bilateral weaknesses of the upper trapezius and sternocleidomastoid muscles, and examination of the hypoglossal nerve (cranial nerve XII) demonstrated bilateral fasciculation of the tongue with normal tongue strength. No other significant changes were noted during cranial nerve examination. Muscle palpation detected tenderness and hypertonicity of the masseter muscle, the anterior fibers of the temporalis, lateral pterygoid, and digastric and suboccipital muscles on both sides of the joint. The patient was diagnosed with a chronic TMJ disorder associated with multiple bilateral muscle hypertonicity of the TMJ.

Chiropractic care was provided and included joint mobilization of the TMJ in distraction (Fig 1), lateral deviation (Fig 2), protrusion, and also in retrusion (Fig 3). Active ROMs were also targeted, with the same vectors. To address general jaw stiffness and muscle hypertonicity, myofascial therapy, including proprioceptive neuromuscular facilitation technique

Fig 4. Proprioceptive neuromuscular facilitation technique of the TMJ muscles. The patient performs isometric contractions of the TMJ muscles (movement in distraction, lateral deviation, protrusion, and also retrusion) followed by passive stretching of the same muscle group.

Fig 3. Joint mobilization of the TMJ in protrusion and retrusion. This mobilization is performed by applying anterior-posterior and posterior-anterior force vectors to the mandibulae with contact on the mental protuberance.

(Fig 4) and active stretching of the masseter, temporalis, lateral pterygoid, digastric and suboccipital muscles, was performed at each visit. Trigger point therapy (ischemic pressure) of the same muscles was also conducted. Light spinal mobilizations of the upper cervicals were included as part of the treatment to increase mouth opening because studies have demonstrated that craniocervical junction position modifies jaw position and its ROM.¹⁸ Considering the higher incidence of cervical pain in patients with TMJ dysfunction, upper cervical spine mobilization could prevent further involvement of the neck.¹⁹ The patient was treated twice a week for 4 weeks.

Follow-up evaluation of the condition was performed at the eighth visit. At that time, the patient reported less TMJ pain and subjectively less stiffness of the jaw. Pain was rated at an intensity of 2 on a pain scale of 0 to 10. At that time, physical examination showed no change in mouth opening; and active mouth opening was still 11 mm interincisival. Based on the moderate clinical changes observed at follow-up evaluation, it was decided to continue care twice a week. At the 12th treatment, the patient reported further improvement and said that he

could insert a tobacco pipe between his teeth, something that he could not for years. The final evaluation of TMJ ROM showed active opening of 12 mm and absence of pain and muscle tenderness of the jaw. At this point, it was believed that maximal therapeutic benefit was achieved; and the patient decided to continue with elective care. He received a total of 22 treatments over a period of 4 months and was subsequently referred for evaluation of maxillo-facial surgery, as his was an unusual clinical case.

Discussion

Patients with musculoskeletal disorders related to underlying neurodegenerative pathologies can benefit from chiropractic management adapted to their condition; in the present case, chiropractic treatment of the TMJ represented a viable, low-cost approach with limited adverse effects compared with surgery.²⁰ In fact, as mentioned by Hawk et al (2007),²¹ for all age groups and conditions, the adverse effects in chiropractic care are rare and if they occur, these effects are transient and not severe. With the lack of success in conventional care, for which they have little to offer for debilitating conditions like neurodegenerative disorders, chiropractic management is a low-risk alternative.²¹ In SMA patients, abnormal craniofacial growth patterns and poor masticatory muscle function might contribute to limited ROM of the jaw. As mentioned previously, maximum masticatory muscle strength is diminished in SMA patients. These muscles are less efficient and fatigue up to 30% faster than those in healthy control subjects.⁸ Muscle therapy and joint mobilizations seem to limit and reverse such effects. Other therapies have proven to be relatively effective and should be considered in future studies. For example, programs involving relaxation techniques and biofeedback, electromyographic training, and proprioceptive reeducation have been suggested as being potentially effective in the past. These therapies could be more effective than placebo and occlusal splints in decreasing pain and increasing mouth opening in acute or chronic muscular TMJ disorders.⁹ It is known in SMA patients that the main gastrointestinal and nutritional problems include feeding and swallowing difficulties, gastrointestinal dysfunction, as well as growth and nutrition deficiencies.²² Effective assessment and treatment of SMA patients' TMJ dysfunction are likely to contribute to improvement of their nutritional, gastrointestinal, and general health quality. The actual consensus statement for standard care in SMA states "the evaluation of oral

structures that influence feeding efficiency and consideration of the effect of positioning and head control on feeding and swallowing are essential."¹² Although the clinical guidelines for SMA recognize the importance of addressing masticatory and oral structures in the evaluation, precise intervention recommendations are limited and only include interventions, such as changing food consistency, optimizing oral intake, and proactive nutritional supplementation.¹² Evaluation of the TMJ and the masticatory apparatus should be included in standard care of this condition as well as in management related to their dysfunction. Early chiropractic clinical evaluation and prophylaxis management retaining maximum functional abilities of the TMJ should be investigated. Other suggestions include case series and clinical trials on conservative care of TMJ disorder in patients with SMA. Prior studies have shown interesting findings, indicating a considerable willingness to engage in complementary and alternative medicine trials among patients with craniofacial conditions and TMJ disorders. Willingness has also been observed among individuals who have not previously used alternative medicine.²³ However, because of their restricted mobility, patients with neurodegenerative conditions may have limited accessibility to such care.

One of the limitations of this case study is the lack of objective measures. Tools, such as questionnaires evaluating the general quality of life and TMJ function, should be considered for future reports and studies. Finally, this patient was treated for a short period; and evaluation of long-term care may have shown either more improvements or stabilization of the condition. Further investigations on ongoing care needed by the patient and evaluation of the best interval between visits are needed. The effects of a home exercise program combined with chiropractic care should also be tested.

Conclusion

A patient with chronic TMJ disorder and SMA type III showed subjective and functional improvement after chiropractic clinical management of his condition. The clinical information stemming from this case work reveals an interesting future for chiropractic care in the treatment of neuromuscular disorders and in the comanagement of such conditions. Few studies have investigated the role of complementary and alternative medicine, such as chiropractic, in neurodegenerative and neuroimmunologic diseases. Because most of these patients seek care through complementary and alterna-

tive medicine, the relevance of chiropractic care should be addressed and carefully explored.²

References

1. Coulter ID, Shekelle PG. Chiropractic in North America: a descriptive analysis. *J Manipulative Physiol Ther* 2005;28(2):83-9.
2. Apel A, Greim B, Zettl UK. Complementary and alternative medicine and coping in neuroimmunological diseases. *J Neurol* 2007;254(Suppl 2):II112-5.
3. Guillot N, Cuisset JM, Cuvellier JC, Hurtevent JF, Joriot S, Vallee L. Unusual clinical features in infantile spinal muscular atrophies. *Brain Dev* 2008;30(3):169-78.
4. Iannaccone ST. Modern management of spinal muscular atrophy. *J Child Neurol* 2007;22(8):974-8.
5. Houston K, Buschang PH, Iannaccone ST, Seale NS. Craniofacial morphology of spinal muscular atrophy. *Pediatr Res* 1994;36(2):265-9.
6. Kasper DL, Harrison TR. Harrison's principles of internal medicine. 16th ed. New York: McGraw-Hill, Medical Pub. Division; 2005.
7. Zerres K, Davies KE. 59th ENMC international workshop: spinal muscular atrophies: recent progress and revised diagnostic criteria 17-19 April 1998, Soestduinen, the Netherlands. *Neuromuscul Disord* 1999;9(4):272-8.
8. Willig TN, Paulus J, Lacau Saint Guily J, Béon C, Navarro J. Swallowing problems in neuromuscular disorders. *Arch Phys Med Rehabil* 1994;75(11):1175-81.
9. Medlicott MS, Harris MR. A systematic review of the effectiveness of exercise, manual therapy, electrotherapy, relaxation training, and biofeedback in the management of temporomandibular disorder. *Phys Ther* 2006;86(7):955-73.
10. Granger MW, Buschang PH, Throckmorton GS, Iannaccone ST. Masticatory muscle function in patients with spinal muscular atrophy. *Am J Orthod Dentofacial Orthop* 1999;115(6):697-702.
11. Vinjamury SP, Singh BB, Khorsan R, Comberiati R, Meier M, Holm S. Chiropractic treatment of temporomandibular disorders. *Altern Ther Health Med* 2008;14(4):60-3.
12. Wang CH, Finkel RS, Bertini ES, et al. Consensus statement for standard of care in spinal muscular atrophy. *J Child Neurol* 2007;22(8):1027-49.
13. Tilton AH, Miller MD, Khoshoo V. Nutrition and swallowing in pediatric neuromuscular patients. *Semin Pediatr Neurol* 1998;5(2):106-15.
14. DeVocht JW, Schaeffer W, Lawrence DJ. Chiropractic treatment of temporomandibular disorders using the activator adjusting instrument and protocol. *Altern Ther Health Med* 2005;11(6):70-3.
15. Saghafi D, Curl DD. Chiropractic manipulation of anteriorly displaced temporomandibular disc with adhesion. *J Manipulative Physiol Ther* 1995;18(2):98-104.
16. McNeely ML, Armijo Olivo S, Magee DJ. A systematic review of the effectiveness of physical therapy interventions for temporomandibular disorders. *Phys Ther* 2006;86(5):710-25.
17. Karasu HA, Okcu KM, Ortakoglu K, Bayar GR, Aydintug YS. Treatment of temporomandibular joint ankylosis with temporalis superficial fascia flap. *Mil Med* 2005;170(2):167-71.
18. Mansilla-Ferragut P. Immediate effects of atlanto-occipital joint manipulation on active mouth opening and pressure pain sensitivity in women with mechanical neck pain. *J Manipulative Physiol Ther* 2009;32(2):101-6.
19. Alcantara J, Plaugher G, Klemp DD, Salem C. Chiropractic care of a patient with temporomandibular disorder and atlas subluxation. *J Manipulative Physiol Ther* 2002;25(1):63-70.
20. Kalamir A, Pollard H, Vitiello AL, Bonello R. Manual therapy for temporomandibular disorders: A review of the literature. *J Bodyw Mov Ther* 2007;11(1):84-90.
21. Hawk C, Khorsan R, Lisi AJ, Ferrance RJ, Evans MW. Chiropractic care for nonmusculoskeletal conditions: a systematic review with implications for whole systems research. *J Altern Complement Med* 2007;13(5):491-512.
22. Kostova FV, Williams VC, Heemskerk J, et al. Spinal muscular atrophy: classification, diagnosis, management, pathogenesis, and future research directions. *J Child Neurol* 2007;22(8):926-45.
23. Schneider J, Vuckovic N, DeBar L. Willingness to participate in complementary and alternative medicine clinical trials among patients with craniofacial disorders. *J Altern Complement Med* 2003;9(3):389-401.