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Wheelchair to walking in 9 months: ABC™ meningeal releases and Stiff Person Syndrome: A case report

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Narrative: Stiff Person Syndrome (SPS) is a neuro-immunological condition most frequently associated with the GAD65 autoantibody. Common treatments are benzodiazepines and intravenous immunoglobulin (IVIG).

The case presented is of a 66-year-old female diagnosed with SPS by a neurologist after more than a decade of worsening muscle spasms and subsequently confirmed with elevated levels of GAD65. Her condition was continuing to deteriorate despite IVIG, baclofen, botulinum toxin, and Chiropractic spinal manipulation.

The introduction of Meningeal Releases as taught as part of the chiropractic technique, Advanced BioStructural Correction™ combined with Chiropractic spinal manipulation was concomitant with progressive improvement in the condition of the patient, to the extent that the patient progressed from wheelchair bound to ambulatory within 9 months of beginning treatment.

Further research to explore the causal relationship between meningeal tension and SPS may inform additional treatment options.

Indexing Terms: Chiropractic; subluxation; ABC™; Meningeal release; Stiff Person Syndrome

Introduction

Stiff Person Syndrome (SPS) is a condition with an incidence of approximately one to two per million. Despite being first recorded in medical literature in 1956 by Dr Frederick Moersch and Dr Henry Woltman, (1) medical understanding of the condition is still evolving.

Classic SPS is a chronic progressive condition characterised by muscle hypertonia and spasms predominantly affecting the trunk and limbs, which can lead to varying degrees of disability. It affects women at least twice as commonly as men, and the onset is often between the ages of late 20s and late 50s. (2) Since its discovery, SPS has been broadened into a condition of multiple phenotypes; the classic phenotype accounting for approximately 70-80% of cases.

> *... within 6-months of commencing ABC™ care this 66yo wheelchair-bound Caucasian woman was able to move from wheelchair to treatment table without support, quickly followed by self-toileting, transitioning from chair to car and then walking her first few steps in years without support, all within a further 2 weeks'*

Stiff limb syndrome or partial SPS is a phenotype that usually affects a lower limb or the torso in a more isolated fashion. SPS-plus includes the classic findings in addition to cerebellar and/or brainstem signs. *Paraneoplastic SPS* has been associated with a variety of malignancies and symptomatically can resemble classic SPS. *Progressive Encephalomyelitis with Rigidity and Myoclonus (*PERM) is similar symptomatically to classic SPS but also includes encephalopathy and therefore garners some debate as to whether it is a separate phenotype or should be included in SPS-plus. Additionally, some consider pure cerebellar ataxia a phenotype of SPS, while others consider it a separate autoantibody associated disorder.

A neuro-immunological condition

SPS is considered a neuro-immunological condition, however its nature is not completely understood. It is diagnosed based on clinical features as mentioned, but also paraclinical features such as electromyographic evidence of the co-contraction of agonist and antagonist muscles, and/or the presence of serum anti glutamic acid decarboxylase 65-kilodalton isoform (GAD65) antibodies. The presence of GAD65 in the serum is recorded in 60-80% of patients, (2) and other auto-antigens have been recorded, but a minority of patients form a subgroup of seronegative SPS. It has been noted that the serum level of GAD65 is not correlated with the severity of the symptoms, but elevated levels are associated with poorer response to immunotherapy. (3) Other research suggests that there is no clear evidence that GAD antibodies are pathogenic in SPS. (4)

Usual treatment for SPS has two aims, the first is symptomatic treatment, and the second is to treat the autoimmune aspect of the condition. Symptomatic treatment is frequently based on benzodiazepines (5) for their GABAergic effect on decreasing nervous system activity, and further medications such as *baclofen* or *botulinum toxin* can be utilised simultaneously. In cases where immunotherapy is indicated, intravenous immunoglobulin (IVIG) is considered the most effective therapy. (6)

Case report

A 66-year-old Caucasian female had been experiencing advancing neurological symptoms for approximately 20 years, and had been diagnosed with Stiff Person Syndrome (SPS) 4 years prior by a neurologist based on symptoms initially, and confirmed 2 years later by serum GAD65 levels of 32IU/ml. No other autoimmune comorbidities were present.

She had been wheelchair bound for approximately 5 years with constant muscle spasms creating trunk, hip, and knee flexion as well as ballistic spasms of both upper limbs and neck muscles frequently. Additionally, severe spasms of the lower leg muscles and intrinsic muscles of the foot led to a constant state of severe muscle contraction resulting in complete constant flexion of all toes, and venous insufficiency in the form of oedema and discolouration of the feet if the feet were lowered beneath the level of the hips. IVIG, *baclofen*, *benzodiazepine*, *dantrolene*, *nabiximols*, and *botulinum toxin* treatments provided some relief with declining effectiveness over time, *botulinum toxin* treatment was ceased after 3 treatments as it became ineffective.

Chiropractic spinal manipulation consistently provided relief from both muscle spasms and pain consistently over the previous 17 years, regularly achieving a relaxed state on completion of the appointment, however the effect was temporary, often worsening again over 24-48 hours. Physiotherapy treatment was not tolerated as prescribed exercises triggered muscle spasms.

Intervention and outcome

Meningeal Releases as taught in the *Advanced Biostructural Correction*™ (ABC™) technique were introduced and combined with Chiropractic spinal manipulation. Treatment was provided twice weekly. Immediately muscle spasms reduced through the body, and voluntary movement in the feet started to return. Within a month of beginning treatment spasmodic flexion in the trunk,

hip, and knee was intermittent, and the patient could lie supine with ease. Two months after beginning treatment hip and knee spasm was almost eliminated, and she started to put some weight on her legs from a seated position.

Within 4 months of beginning treatment, voluntary movement of her knees had returned, and she could flex and extend her legs while lying supine. At this stage she was progressing weight bearing on feet and performing abduction/adduction movements of her hips. By the 6-month mark, she was able to transition from wheelchair to treatment table without support, which was quickly followed by self-toileting, transitioning from chair to car and then walking her first few steps in years without support, all within a further 2 weeks.

The patient quickly noticed she could walk backwards for the first time in memory. She then recommenced physiotherapy exercises and could tolerate them without the spasms being triggered as they were previously. She was walking with a 4-wheel support frame for 3 months before being able to walk without support approximately 9 months after beginning treatment. At this point, her serum GAD65 levels were re-tested, and despite the improvement in her symptoms, her levels were 455IU/ml, significantly elevated over the initial results 2 years prior, which were 32IU/ml.

Discussion

Evidence is emerging that explains the impact of chiropractic spinal manipulation on neuromuscular function. (8, 9) That evidence may shed some light as to how spinal manipulation provided temporary relief of muscle spasticity consistently for over 17 years. Despite the temporary relief provided by spinal manipulation, and the introduction of medical interventions, the SPS symptoms continued to progress to a point of severe disability.

A thorough understanding of SPS in the literature appears to be lacking. While GAD65 is the most common autoantibody associated with the condition, others have been recorded in SPS patients. The existence of seronegative SPS frustrates the description of SPS as an immunoneurological syndrome. Additionally, dissociation of serum antibody levels and symptomatology is inconsistent with our current understanding of the nature of SPS. SPS' status as an incurable condition and subsequent symptomatic treatment completes a picture of a syndrome that's poorly understood. There is some promise in reports of improvement through autologous haematopoietic stem cell therapy (auto-HSCT), though this treatment has only been used in a small number of cases. (9)

The rarity of SPS (approximately 1-2 per 1 million) is such that a randomised controlled trial is not practical, so the observations made in a case study as in the auto-HSCT study could be more informative for future treatment considerations. This case study, while anecdotal in nature, was concomitant with such consistent improvement over the 9-month course of treatment that it warrants further research into the potential of meningeal releases as a treatment for SPS.

The meningeal release

The aim of a meningeal release is to reduce mechanical tension caused by adhesions in the meninges. According to the work of Dr Alf Breig (7) these adhesions can occur between layers of the meninges, or between the meninges and the spinal cord itself, which can theoretically induce spinal cord compression and/or tension within the dural system.

Given the immunoneurological nature of SPS, a mechanism as to how the meningeal release can be concomitant with progressive, consistent, and significant improvement in SPS symptoms is not known. Therefore, further research into the impact of meningeal tension on SPS and other immunoneurological conditions may yield additional avenues of treatment to explore.

The response in this case to manual intervention, suggests a neuromuscular physiological mechanism may be involved.

Conclusion

The aetiology of Stiff Person Syndrome (SPS) is unclear and the management is often of limited success. ABC™ is a type of manual therapy, predominantly employed by Chiropractors, which aims to influence mechanical tension in the central nervous system by decreasing dural tension. This cases responded well to the implementation of meningeal releases which are part of the ABC™ protocol.

We recommend further research into the neurophysiological effect of ABC™.

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