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Reflex sympathetic dystrophy: an enigmatic improvement with spinal manipulation

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Reflex Sympathetic Dystrophy (RSD) or complex regional pain syndrome, is an extremely painful and disabling condition commonly seen following trauma. Its early recognition and treatment is most critical for a favorable prognosis. Although its diagnosis and treatments vary, neuroblockade is the treatment of choice. Very little has been reported in the literature in regards to manipulation as an early treatment modality to improve joint mobility and reduce pain and future disability. This case report reviews one case presentation of RSD where dramatic results followed cervical spine manipulation.
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KEY WORDS: reflex sympathetic dystrophy, manipulation, chiropractic, neuromatrix, cervical spine.

L'algodystrophie sympathique (AS) est une affection extrêmement douloureuse et invalidante qui se rencontre souvent après un traumatisme. Son diagnostic et son traitement précoces sont des éléments cruciaux d'un pronostic favorable. Même si les moyens diagnostiques et thérapeutiques varient, le traitement de premier choix est l'anesthésie par blocage nerveux. Il existe très peu de documentation sur les manipulations comme mesure thérapeutique précoce pour améliorer la mobilité articulaire, soulager la douleur et atténuer l'incapacité potentielle. Voici un cas d'AS où les manipulations de la colonne cervicale ont donné d'excellents résultats.
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MOTS CLÉS : algodystrophie sympathique, manipulations, chiropratique, neuromatrice, colonne cervicale.

Introduction

Reflex Sympathetic Dystrophy (RSD) or complex regional pain syndrome, are terms often used to describe a clinical condition presenting with severe pain, hyperesthesiae, vasomotor disturbances and dystrophic changes in the upper and lower extremities. These signs and symptoms begin gradually days or weeks after an injury but may manifest within a few hours. The patient suffers greatly and protects the affected area. This disorder progresses in stages that have variable lengths lasting anywhere from weeks to years.

Scientific literature describing RSD presents a potentially confusing array of syndromes. It was first described in 1864 by S.W. Mitchell in his study of Civil War veterans. He provided the designation of causalgia.[1] In 1947, Evans[2] believed that causalgia and related syndromes were merely various manifestations of one disease entity and suggested the term *reflex sympathetic dystrophy*. Other historical terms used were Sudeck's Atrophy,[3] post traumatic osteoporosis,[4] major causalgia,[5] shoulder hand syndrome[6] and sympathetically maintained pain syndrome[7] to name a few.

The diagnosis of the condition is defined by the clinical picture of continuous pain with one more physical sign of sympathetic over activity which is confirmed by a substantial reduction of pain with a differential sympathetic block.

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The continuous pain was most often described as burning, aching or throbbing. Kozin[8] has proposed clinical criteria to assist in a definitive diagnosis, although Kozin's criteria may be too restrictive. Many clinicians feel only two findings, hyperalgesia and allodynia, are needed to be present to diagnose RSD, and that the trophic changes, sweating and libido reticularis, are not constant findings in all cases.

The incidence of RSD is unknown. Some studies have documented its occurrence in association with various diseases. Incidences in peripheral nerve injuries (2 to 5%),[9] stroke (12 to 21%),[10,11] Colles' fracture (2 to 11%),[12,13] coronary artery disease (1 to 20%)[5,8,14] and trauma in general (0.5%). Studies have shown an even distribution of RSD among all age groups[15] and the male to female ratio is estimated between 1:1 and 1:3. RSD can also occur in children with the earliest reported case to be age 3.

The pathophysiology of RSD is still theoretical. Efferent sympathetic hyperactivity is implied in the physical signs. The theories are both peripheral and central in etiology. Doupe[16] proposed that peripheral nerve trauma leads to discontinuity in the surrounding connective tissue and myelin. This would allow for an "artificial synapse" between sympathetic efferents and somatic sensory afferents. Doupe felt the sympathetic impulse would jump across to the somatic afferent and be perceived as pain centrally.

Some centrally mediated theories such as the "reverberating circuits theory",[17] suggests reverberating circuits in the internuncial pools creating increased sympathetic efferent activity. The Turbulence Theory[18] suggests the modulating affect of the internuncial nerves is altered by changes to the sympathetic ganglia, secondary to trauma from a post ganglionic peripheral sympathetic nerve.

The Wide Dynamic Range Theory[7] (WDR) suggests the activation of "unmyelinated C-fiber nociceptors" in the peripherally injured tissues lower the threshold of the WDR cells in the cord. A response may also be elicited by activation of "A-fiber" mechanoreceptors which may result in allodynia. The only abnormal state exists in the central WDR neurons.

Therefore, to date, no theory fully explains the findings associated with RSD. Perhaps a combination of these peripheral and central theories may provide a more accurate understanding of the etiology of RSD.

RSD is said to undergo three stages in its clinical progression.[6] These stages are acute, dystrophic and atrophic.

Stage I – Acute
The acute stage begins at the time of injury and lasts several weeks. The pain is disproportionate to the initial injury. There is a burning and aching which is increased by physical contact of the affected part or by emotional upset. The extremity may appear red with a decrease in joint motion. The skin may be cold, cyanotic, or sweaty, warm and dry.

Stage II – Dystrophic
Dystrophic is characterized by edematous tissue with libido reticularis or cyanosis present. Hair loss may occur and decreased nail growth or brittleness may also occur. The pain is constant or increased by any stimulus to the affected part. X-rays may reveal diffuse osteoporosis.

Stage III – Atrophic
The atrophic stage shows a spread of pain and irreversible tissue damage. The skin appears thin and shiny and the fascia becomes thickened with flexion contractures. X-rays show bony demineralization and often ankylosis.

Commonly, the RSD fails to progress through these stages and takes on a partial form in which severe pain is associated with a minimal degree of sympathetic hyperactivity, only slight swelling and a mottling in association with the characteristic burning pain. RSD typically starts distally and spreads proximally, and in some cases spreads to the other extremities without new injury.

The differential diagnosis includes post-traumatic peripheral nerve pain, inflammatory lesions, myofascial pain or fibrositis and vascular disease. In these cases, the pain either remains localized, the sympathetic component is absent or the complete RSD syndrome is not present.

Laboratory tests for skin temperature, skin blood flow, sympathetic response, sweat tests, have not shown a consistent response. X-ray studies may demonstrate a patchy periarticular demineralization in approximately 3 to 6 weeks or even later. These findings, however, may also be found in thyrotoxicosis and hyperparathyroidism. Scintigraphy can confirm abnormalities earlier than routine radiographs.

Neural blockage (sympathetic block) is considered by most to be essential to the diagnosis of RSD. Resolution of symptoms following a successful block is considered diagnostic of RSD. Unfortunately, the response to sympathetic

blockade is not 100%.[19]

Electrodiagnosis is essentially normal in RSD and thermography is of little diagnostic value.[20,21]

The prognosis for patients suffering from RSD is uncertain or guarded at best. No modality to date has proven to be of definitive value. There is agreement in the literature that treatment prior to six months provides a much brighter prognosis. The best study available, using a large sample population was by Subbarao[22] which stated about 60% of patients continued to have pain regardless of treatment.

Treatment of RSD is aimed at blocking the effects of sympathetic hyperactivity, most importantly though, is early intervention. Forms of physical therapy in itself have been shown to be affective in its treatment, with exercises aimed at improving mobility of the affected extremity. Pain is a limiting factor in compliance to the exercise program.

Medically, the most commonly used treatment is sympathetic blockade with anesthetics such a buprivacaine (9.25%) or mepivacaine (0.5%). Its success depends upon the skill of the doctor, the stage of the disorder and the completeness of the blockade. Regional blockade called a Bier Block[23] using 10 to 20 mg of guanethedine has demonstrated some benefit but remains inferior to paravertebral blockade.[24]

Oral medications include the use of corticosteroids[25] or phenoxybenzamine, a sympathetic blocker. These procedures have limited value with pain often recurring following the tapering schedule. Transcutaneous electrical nerve stimulation has had limited value, especially in children, although less value than sympathetic blockade or corticosteroids.[26] Acupuncture has been used in both children and adults with some benefits but a lack of controlled studies exist.[27] Surgical sympathectomy may be used in those who had temporary success for anesthetic blockade. Results range widely from 12 to 97%. Radio-frequency techniques are now being developed.[28]

Case report

The case report pertains to the injuries and subsequent diagnosis of reflex sympathetic dystrophy of a 37-year-old male Caucasian working as a labourer.

The injury occurred four years prior to his presentation to our multidisciplinary chronic pain clinic. The worker described his injury as a crush fracture and laceration to his left hand, which occurred when a nine foot diameter wheel fell onto him. The patient stated that he sustained a laceration in the web of his fourth and fifth digits, which later developed an infection requiring antibiotics. He stated that at the moment of impact, he instinctively attempted to pull his injured hand out from under the heavy wheel, which created traction to his left upper limb. He then immediately attended the emergency department of his local hospital in which radiographs and a preliminary assessment took place. He was advised that he appeared to have a fractured pisiform and the laceration was sutured and he was sent to his family physician. His physician noted increased pain sensitivity, hand edema, discoloration and referred him to a neurologist with a tentative diagnosis of reflex sympathetic dystrophy.

At five months post-injury, he saw a neurologist complaining of a persistently painful left shoulder, intermittent pain and weakness in his left hand, mottling of the skin of his left upper limb, intermittent sweatiness and color changes to his left palm, and pain during movements of his left edematous hand. His bone scan demonstrated a moderate increase in the activity of the left pisiform without any increase in flow and blood pool phase. These findings indicate a subacute healing of the pisiform fracture.

He was also sent for electrodiagnostic consultation as he complained of a reduced sensation in digit five. An earlier electrodiagnostic test revealed a normal ulnar sensory evoked response. This second electrodiagnostic testing found him having difficulty squeezing his left hand and unable to distinguish hot from cold in the left fifth digit. The digit turned a purplish red, but not white in cold. This second electrodiagnostic testing revealed a Tinel's sign over the left ulnar nerve at the elbow, but not over the median nerve at the wrist. Muscle bulk, tone, and power were normal except for left abductor digiti minimi, which did not fully recruit and appeared to be grade four. Pain on flexing the distal interphalangeal joint of his left fifth digit was present. All reflexes were +2 and symmetric throughout. He had decreased pinprick sensation on the dorsum of his hand in both dorsal ulnar cutaneous and radial distributions, the ulnar palm and left digits four and five. The nerve conduction tests conclusion was that his symptoms were related to a "functional" disturbance of the nerve on cold exposure rather than "structural" deficit of the nerve.

He was subsequently referred to an orthopedist who concurred the diagnosis of reflex sympathetic dystrophy and sent him to an anesthetist for a series of nerve blocks

using Quinethidine, which provided no lasting relief of the pain and disability.

Finally, after developing the injury in 1993, neurological and orthopedic examinations in 1994, and a series of 10 nerve blocks from 1994 through 1996, he presented at our multidisciplinary chronic pain clinic in early 1997. At this time, his presenting complaints were a sensation of cold, cramping, throbbing with sharp shooting pains in the left upper limb. He claimed that the limb felt heavy and tried. He also now presented with restricted left glenohumeral range of motion and pain.

Previous treatments prior to admission to our rehab clinic consisted of both active and passive physical therapeutic modalities. Passive therapy consisted of T.E.N.S., the application of pulsed ultrasound to his wrist, and left upper limb mobilizations.

Active physical therapy consisted of submaximal multiple angle isometric strengthening of his elbow and shoulder and grip strengthening isotonic exercises. The left upper limb was later strengthened with passive, passive assisted, active and active resistive movements utilizing PNF movement patterns. He was also performing active pool exercises and light cardiovascular conditioning using a treadmill.

Massage therapy was administered utilizing spray and stretch techniques to the wrist flexors and extensors, as well as left upper trapezius and anterior scalenes. The goal of massage therapy at that time was to eliminate local trigger points in the muscles. Cross fiber massage in the form of deep friction massage was applied to the rotator cuff muscles as well.

Medications used included the use of tricyclic antidepressants, various anticonvulsant medications such as Neurontin (Gabapentin) and nonsteroidal anti-inflammatories. As stated earlier, he received 10 nerve blocks using Quinethidine. The patient also received several acupuncture treatments as well.

The success of his multiple modality therapies prior to admission to the pain clinic was limited at best. Treatment prior to admission to our multidisciplinary pain clinic was pain focused in nature and no functional capacity evaluations were performed. The FCE was performed at admission to our clinic.

Physical examination

At our pain clinic, he demonstrated restrictions in mobility in the cervical spine. He had only 30 degrees of left lateral bending and 40 degrees of left cervical rotation with reduced cervical extension to 50 degrees. Pain occurring at the limited end range of movement. All other cervical ranges of motion were within normal limits. His left glenohumeral active range of motion was 85 degrees flexion, 48 degrees abduction, 20 degrees external rotation and normal internal rotation. Passive movements were similar, although 55 degrees of abduction was attainable.

Wrist range of motion was 42 degrees extension and 70 degrees flexion both actively and passively. Pain occurred at end range in both flexion and extension.

Muscle strength testing was 5/5 on the right upper limb and 4/5 on the left side throughout testing individual muscle groups. All muscle strength testing was done while the shoulder was in a pain free zone and in a neutral position. He had difficulty gripping the hand dynamometer due to pain in the hand.

During his eight week tenure in our pain program, the patient had undergone active physical therapy including active muscle energy techniques, McKenzie exercises, strength and conditioning training, proprioceptive retraining, cardiovascular reconditioning, cervical traction, thoracic vertebral mobilizations, pool exercise therapy, occupational work hardening and vocational counseling. He also received extensive in-program pain management education from the staff psychologist.

Functional capacity evaluation was conducted by the staff occupational therapist. The results revealed no limitations in sitting, weight bearing, walking and climbing. The patient was able to perform low level work with reduced ability to perform over shoulder work and his manual handling abilities were greatly reduced. The occupational therapy summary suggested that the major work limitations were in doing overhead work with reduced manual handling using left hand up to nothing more than 10 pounds.

He continued to have slight strength gains during the first seven weeks of our pain program, but resolution of the pain and increased motion in the left upper limb was minimal at best. During his final week in program, the patient began receiving manual chiropractic cervical manipulative therapy. The manipulations were performed in a seated position using a diversified thrusting technique with doctor contact located on the patient's left anterolateral transverse process at the level of the seventh cervical ver-

tebrae. After propositioning, a quick dynamic thrust was delivered in an inferior to posterior direction. Excellent joint cavitation occurred with no patient discomfort.

Two days postmanipulative treatment, the patient had improved in left shoulder flexion to 160 degrees and 170 degrees abduction, pain free. He described his pain as 2/10 on the visual analog scale, in the entire left upper limb.

A second manipulation was performed in the same manner as the first on the next day with continued pain reduction to 1/10 on the VAS, and a full 180 degrees flexion, and 180 degrees abduction and full pain free circumduction in his left shoulder. His cervical biomechanics (i.e. range of motion) had also revealed a springy end feel and increased range of motion. After a total of three cervical manipulations, the patient felt completely ready for full work duties and promptly returned to his previous employment within two weeks of discharge from the program, after being off work for four years. He was advised to continue with his exercises in a home-based program and to attend his local chiropractor for periodic manipulations on an as needed basis.

Discussion

Very little has been published in the literature in regards to the use of manipulation as a treatment for reflex sympathetic dystrophy. Blumberg,[33] in his paper on this topic states that it is accepted today that there is a spectrum of conditions that have as their basis altered activity of the sympathetic nervous system and conform to the overall category known as "sympathetically maintained pain syndromes".

Duncan,[29] in 1988 published a study in which manipulation was used with a patient under Bier Block anesthesia and found significant improvement in range of motion from 46 to 81% from a preblock mean.

It is well established that the central nervous system adapts and has plasticity influenced by proprioceptive and nociceptive inputs. Shipton[30] states in his editorial on Multimodal Analgesia, that injury can induce both peripheral and central hypersensitivity and has important consequences for pain control strategies. He speaks of a class of unmyelinated primary afferent fibers called "silent nociceptors" that respond to inflammatory and chemical sensitization. They discharge vigorously during ordinary movement. Nociceptor stimuli develop alterations in the processing of sensory information at the cord level called "plasticity".

He states with nociceptive input, there is not a simple stimulus response relationship, but a "wind up" of spinal cord neuron activity. This central sensitization is perceived as allodynia and hyperpathia, the two key components in reflex sympathetic dystrophy. This central sensitization allows recruitment of low threshold mechanoreceptor afferent input with produces pain, something they normally never do.[31]

Many clinicians believe a patient must demonstrate a favorable response to sympathetic blockade before a diagnosis of RSD can be made. Chemical sympathectomy, however, has a 12 to 97% success rate and may be too variable as a diagnostic criterion. Tahmoush[34] had proposed that only the two findings of allodynia and hyperpathia need to be present to diagnose RSD.

In this case, the patient did not respond to the neural blockades, but presented with allodynia and hyperpathia. This demonstrates, as stated by Vernon[35] in his paper on RSD and chiropractic, that it is important to recognize the spectrum of pain syndromes associated with central nervous system, including causalgia, RSD, sympathetically maintained pain and reflex sympathetic dysfunction.

In this case, it was medically unanimous that the patient suffered from a Sympathetically Maintained Pain syndrome labeled as RSD. It is difficult to determine why the patient responded so dramatically to the cervical manipulation. Perhaps it was centrally mediated neurological sequelae, but it is important to establish treatment in a multimodal sense for analgesic and functional purposes early post-injury to prevent the full development of these previously mentioned neural changes.

As shown in models by Melzak,[32] proprioceptors work in fashion at the cord level to increase inhibition to the nociceptive input. It is certainly worthy of further investigation to determine the role that chiropractic manual manipulation plays in affecting the pain neuromatrix and its neural plasticity.

Conclusion

The patient diagnosed with reflex sympathetic dystrophy remains a difficult clinical challenge. The clinical idea that a person diagnosed with RSD needs to have a persistent painful lesion, a personality disorder and abnormal sympathetic nervous system is presently being challenged.

This patient with a diagnosis of RSD poses complex and

difficult physical and psychological sequelae. These patients need early intervention and are at best served in a multidisciplinary setting to address all the four main components of pain; these being emotional, behavioral, perceptual and nociceptive. Due to the complexity and unknown factors of the pathophysiology of RSD, it is most critical that a multimodality approach be initiated as soon as possible to avoid advancement into its later stages. This case report demonstrates that chiropractic manipulation, in conjunction with active rehabilitation, aimed at restoring normal vertebral biomechanical function and perhaps affecting the pain neuromatrix, peripherally and/or centrally, be considered a valuable modality to be included early in the care of this patient population.

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