

Acupuncture for Pediatric Complex Regional Pain Syndrome

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ABSTRACT

Background: Complex regional pain syndrome (CRPS) is a debilitating chronic pain condition that has a complicated pathophysiology and involves sensory, vasomotor, sudomotor, or motor changes. There are various treatments, but no one option is superior. Previous work has shown that acupuncture alleviates symptoms and decreases pain levels in adult patients with CRPS; however, there is limited information on the effects of acupuncture on pediatric patients with CRPS. The purpose of this case series is to examine acupuncture for pediatric CRPS. A retrospective chart review was performed on 3 patients' charts from 2013 to 2015. Demographics, Numerical Rating Scale (NRS), and Brief Pain Inventory results were recorded.

Cases: Three pediatric patients with CRPS were referred to a tertiary pediatric pain clinic for acupuncture treatment. Acupuncture treatment was administered for 5–6 weekly sessions. The treatment outcome was evaluated with the NRS and a Brief Pain Inventory.

Results: After acupuncture treatment, the 3 patients experienced alleviation of pain and reduction of their CRPS symptoms. The mean decrease in the average CRPS-related pain score for the 3 patients was 3.6 of 10.

Conclusions: Acupuncture appeared to reduce pain and improve daily living activities in these 3 patients with CRPS.

Key Words: Electroacupuncture, Neurological Disorders, Pain Management, Complex Regional Pain Syndrome, Pediatrics, Acupuncture

INTRODUCTION

COMPLEX REGIONAL PAIN SYNDROME (CRPS), previously known as reflex sympathetic dystrophy or causalgia, is characterized by continuous, spontaneous, or evoked pain that is disproportionate to the inciting event. CRPS can have multiple etiologies, including damage to tissue or nerves, immobilization, and casting. Some cases have no identifiable causes. The pain typically affects the limbs and is usually not localized to a specific nerve region or sensory dermatome.¹ Patients with CRPS present with abnormalities in the sensory, vasomotor, or motor systems, including edema, changes in sweating, hyperalgesia, allodynia, changes in blood flow, and limited range of motion (ROM) in affected regions.^{1–3} CRPS is complex and poorly understood, leading to few targeted therapies and mixed

results.⁴ The efficacy of commonly performed treatments for CRPS is low: local anesthetic sympathetic blockade provides complete pain relief in less than one-third of patients.⁵ Many patients do not experience full recovery even after receiving all conventional therapies.⁵ Severe complications due to infection, ulcers, chronic edema, dystonia, or myoclonus affect 7% of patients with CRPS.⁶ There is a high incidence of disability in patients with CRPS, such that only 20% of patients resume previous levels of daily activity⁷ and are able to continue their occupations full-time.⁸

Since the National Institute of Health's 1997 consensus statement on the effectiveness of acupuncture,⁹ acupuncture has become increasingly integrated with Western medicine¹⁰ and has been shown to be an effective, noninvasive treatment for chronic pain in adults^{11–13} and children.^{14,15} Research investigating acupuncture for adult patients with

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CRPS has found therapeutic benefit from this treatment modality. Korpan et al. found that, in adult patients with CRPS who received acupuncture, clinical parameters and pain reduction improved to nearly normal levels after 6 months.¹⁶ A case series showed that two soldiers with upper-extremity CRPS had reduction of pain by >80%, decreased sensory changes, and improved extremity function after Chinese scalp acupuncture treatment provided once to twice per week for 1–4 weeks. Twenty months after treatment, the soldiers retained their reduction of symptoms and had no recurrence of CRPS.¹⁷ Finally, a case report of a 34-year-old woman with CRPS who received treatment with acupuncture and laser acupuncture showed lowered pain levels, decreased depression, and improved quality of life (QoL).¹⁸

There is little information, however, on the effects of acupuncture treatment on children with CRPS. A previous case study of a child with CRPS found improved ROM and strength following electrical stimulation at acupuncture points.¹⁹ Another case study described 3 pediatric patients with CRPS who received Seven External Dragons and Devils Acupuncture, administered at GV 20, bilateral BL 11, BL 23, and BL 61 for 30 minutes. Two patients had resolution of pain after 2 acupuncture treatments over 2 weeks and remained pain-free for 10 and 12 months later, respectively.²⁰

The purpose of this report is to describe a retrospective case study investigating the clinical course and treatment response to acupuncture in 3 children with CRPS, who were referred to a tertiary pediatric pain clinic.

CASES

A retrospective chart review was performed from 2013 to 2015 on 3 cases, and patient demographics, Numerical Rating Scale (NRS), and Brief Pain Inventory were extracted. Two pediatric anesthesiologists and pain specialists who are trained in acupuncture medicine administered the acupuncture treatment at a tertiary pediatric-pain clinic. Written parental consent was obtained prior to treatment. An administrative associate collected NRS and Brief Pain Inventory results prior to the acupuncture sessions. Pain scores were not obtained immediately after each acupuncture session. Patients were recommended to undergo 5–6 weekly acupuncture sessions, which were extended to more sessions if requested by the patients.

The acupuncture treatment protocol was based on Traditional Chinese Medicine (TCM). Disposable sterile SEIR-IN[®] acupuncture needles from Shizuoka-shi, Shizuoka, Japan, with tube size No. 02 × 30 mm were used. The depth of the needle insertion was 5–10 mm. The needle was manually manipulated until De Qi was achieved. Acupuncture needles were placed for 15 minutes. At the same time, low-frequency, 2-Hz, electrostimulation using ITO Physiotherapy and Rehabilitation IC-1107+ from Toyotama-Minami, Nerima-ku,

Tokyo, Japan, was performed at ST 36. All acupuncture needles were removed with no apparent complications. Cupping, using valve cups, and *gua sha* were applied at the *Back Shu* points. There were no adverse events.

Case 1

A 15-year-old girl was referred for persistent left-foot pain. Nine months prior to the initial consultation, she experienced a left-foot inversion ankle sprain while playing lacrosse. She described her foot pain as “sharp,” “cutting,” and “digging.” Her pain worsened after 10 minutes of walking, standing, or sitting for long periods, and after exposure to cold temperatures. Removal of her shoes and non-weight-bearing alleviated her symptoms. Her pain score was 6 of 10 on the NRS, and her highest pain score was 8 of 10. On examination, she showed decreased active ROM on left-toe flexion and extension. There was tenderness to palpation at the sole and dorsum of the left midfoot and forefoot, and residual tenderness overlay the anterior talofibular ligament. The patient had active vasomotor changes consisting of tissue edema of the left foot and a dark, bluish discoloration in the left midfoot and forefoot. Her skin was cool to touch. She had allodynia and hyperalgesia over her left ankle. Thus, her examination results met the criteria for CRPS.

The patient used 400 mg of ibuprofen, 1–2 times per week but did not experience much relief. She was placed in a boot for 5 months, received physical therapy, and saw a psychologist for symptoms similar to post-traumatic stress disorder. Overall, the patient missed ~17 days in 1 school year because of her pain. She quit her lacrosse team, which limited her interaction with her friends, resulting in social isolation and depressed mood. Magnetic resonance imaging (MRI) of her left lower extremity revealed a small focus of bone marrow edema along the plantar aspect of the first metatarsal head. There was sclerosis of the subjacent tibial sesamoid without fractures or neuroma.

The patient underwent 10 acupuncture treatment sessions. At her initial session prior to acupuncture treatment, her average pain score was 6 of 10 on the NRS. The primary acupuncture points used for treatment of her CRPS included the following bilateral points: LI 4, LR 3, SP 6, ST 36, GB 34, BL 23, TE 5, LI 11, PC 6, and *Ashi* points. After the first acupuncture session, the patient noted that the treatment was helpful. At the fourth acupuncture session, her pain score decreased to 4 of 10. Her pain was controlled better, and she was able to participate in camp activities. At the sixth session, she reported that her pain decreased to 3 of 10. She felt more comfortable, could participate fully in school without problems, and was able to sleep at night. After her sixth treatment session, she reported that her pain was controlled better compared to the prior session, and she was confident that she was continuing to improve. She was able to walk without difficulty and regained normal motor function. She no longer missed school because of her pain. Over

subsequent treatment sessions, the patient slowly increased her activity level. Overall, she was pleased with her progress and expressed having more enjoyment of her life. The patient returned 4 months later for her last session, during which she experienced a flare-up of symptoms and had an average pain score of 5 of 10.

Case 2

An 11-year-old girl with an accessory navicular bone of the left foot had an acute onset of left-medial foot pain during ballet class, during which a plantarflexion movement gave her considerable pain and residual pain thereafter. Pointing her left foot became difficult and elicited sharp pain. She experienced immediate swelling and could not bear weight on the left side. She was placed in a cast for ~2 weeks, but her pain worsened, and she rated her pain as 8 of 10 on the NRS. She complained of a persistent burning sensation and allodynia to bed sheets, socks, and shoes. She chose to wear softer shoes to prevent rubbing along the medial aspect of her foot. On examination, it was noted that the patient had purplish discoloration on her left distal leg and that her pain worsened with walking. An MRI of her left foot revealed edema at her accessory navicular bone. There was focal bony edema involving the plantar talus and calcaneus and patchy marrow edema within the cuboid, talus, and calcaneus.

Physical therapy achieved some minimal improvement, but desensitization exercises worsened her pain. Her medications included lidocaine ointment, Voltaren[®] gel, and Neurontin[®]. The patient was referred for acupuncture treatment with the diagnosis of CRPS.

At her initial acupuncture session, the patient reported sharp pain and sensitivity to light touch over the medial aspect of her left foot and had an average pain score of 8 of 10 on the NRS. The primary acupuncture points used for treatment of CRPS included the points GV 20 and *Yin-Tang* and the bilateral points LI 4, LI 10, LI 11, LR 3, SP 6, KI 3, KI 7, and ST 36. After her first session, she reported reduced pain for ~3 days with an average pain score of 7 of 10. At her third session, she reported an average pain score of 5 of 10 and had decreased pain for a few days after treatment. She also stated that her pain interfered less with her general activity, mood, relations with other people, and sleep. Over the next treatment sessions, she continued to report benefit from the acupuncture and returned with no new complaints. At the sixth session, her average pain score was at 6 of 10, and she stopped using Voltaren[®] gel and lidocaine ointment regularly. Upon examination, it was noted that she no longer had discoloration along the medial aspect of her left foot and ankle. Compared to the prior week, she reported that her pain interfered less with her general life activities, walking ability, and sleep. She had no adverse side-effects from the acupuncture treatment. Overall, the 6 sessions of acupuncture helped alleviate her symptoms of CRPS and decrease her pain score from 8 to 6 of 10.

Case 3

A 12-year-old boy was diagnosed with a nondisplaced left tibial plateau fracture after a skiing accident. He was given crutches, placed in a hinged ©Bledsoe knee brace, and was non-weight-bearing for 6 weeks. Thereafter, he underwent physical therapy, and his knee pain was reduced. Two months after his tibial injury, he started lacrosse season and had an insidious onset of significant pain in his left ankle and foot. The pain was localized near the base of his left fifth metatarsal. He had difficulty ambulating with an antalgic gait on his left side. He used crutches and stayed off his left foot for a couple of days. He complained of dull, achy pain at 8 of 10 on the NRS and was unable to run or participate in sports. He wore a boot for a month without improvement. Ice and rest helped to decrease his discomfort, but walking and increased activity elicited more pain. He expressed frustration because he lost an opportunity to participate in sport-related activities for 5 months. On examination, it was noted that he had dysesthesia related to temperature changes along the lateral part of his left ankle 3" above and 2" below the left lateral malleolus. In addition, he had a slight limp with ambulation.

Other than physical therapy and water therapy, he received no treatments. An MRI of his left ankle showed diffuse bone marrow edema throughout the ankle and midfoot bones, and he was diagnosed with early CRPS of the left ankle. Three months after his initial ankle sprain, he was referred for acupuncture.

At his initial acupuncture treatment session, the patient rated his average pain as 7 of 10. The primary acupuncture points used for treatment of CRPS included the points GV 20 and *Yin-Tang*, and the bilateral points LI 4, LR 3, GB 34, ST 36, and *Ashi* points. At the second session, his average pain decreased to 3 of 10, and he was feeling better with reduced CRPS symptoms. At the third session, he reported that he had been doing very well and that his pain no longer interfered with his general activity or enjoyment of life. His average pain was 2 of 10. He was able to participate in baseball and planned to participate in another upcoming game. At the fourth session, the patient's pain decreased to 1 of 10 on NRS. He noted that his pain no longer impeded his walking ability and that he was able to continue playing baseball over the summer. He was also getting ready to train for lacrosse tryouts at the beginning of the school year. At the fifth session, the patient reported that his left ankle continued to do well and that he began to play soccer. He denied having any discoloration or temperature changes in his leg or CRPS-related pain.

RESULTS

Three pediatric patients presenting with CRPS received acupuncture treatment for CRPS-related symptoms. Over the recommended 5–6 weekly sessions of acupuncture, the changes in CRPS-related pain for the 3 cases were as follows: for Case 1, from 6 to 3 of 10; for Case 2, from 8 to 6 of 10; for

TABLE 1. CHANGES IN NRS PAIN SCORE

Case	NRS Pain Score		
	Baseline	Post-treatment	Change
1	6	3	3
2	8	6	2
3	7	1	6

NRS, Numerical Rating Scale.

Case 3, from 7 to 1 of 10 (Table 1). The mean decrease in average CRPS-related pain scores for the 3 patients, when comparing the first acupuncture session with the recommended fifth or sixth acupuncture session, was 3.6 ± 1.2 points (mean \pm standard error of the mean). As described by the patients' Brief Pain Inventory, after the acupuncture treatments, all 3 patients reported alleviation of CRPS symptoms. Common improvements included increased mobility and physical activity, decreased usage of medications, and return to social activities and school.

DISCUSSION

Chronic pain is a major health burden in the United States. Approximately 100 million adults in the United States have chronic pain, and it costs our nation \$560–\$635 billion every year.²¹ Moreover, pain is not only a biologic problem, as it can lead to significant emotional and cognitive effects for patients. Effective treatment and management of chronic pain are often not available for patients.²¹ The Institute of Medicine has declared that there must be tremendous revisions to the understanding, assessment, and treatment of pain in the United States.²¹

CRPS is a chronic pain condition that causes great pain and suffering and hinders QoL significantly in both adults and children. In children with CRPS, the recurrence rate is 50%,²² and relapses may even be more severe than the original CRPS occurrence.²³ For children who experience high relapse rates or whose conditions are refractory to conventional treatment, Low et al. suggest that other treatment modalities should be considered.²⁴

People diagnosed with difficult-to-treat chronic disease conditions, such as CRPS, are increasingly turning to complementary medicine.²⁵ In 2007, nearly 40% of American adults used complementary and alternative medicine in the year prior to being surveyed.²⁶ Acupuncture is one therapy that is most commonly used for pain relief.²⁵

The TCM perspective provides mechanisms by which acupuncture can be utilized to relieve pain and the debilitating symptoms of CRPS. Acupuncture is administered to correct the abnormal flow of Qi in the disease state. As described by Bäcker and Hammes, in TCM, CRPS is classified by pain of the four extremities and is characterized by exacerbated obstruction of the meridians and of the Luo.²⁷ The researchers noted that CRPS can be caused by wors-

ening stagnation of Qi and Blood Stasis, which can be elicited by extended immobilization of the affected limb. Possible mechanisms involved in the development of CRPS include the following: Damp-Heat obstructing the Luo, which causes the CRPS symptoms of reddening, warmth, and swelling of the extremity with burning pain; Damp-Cold obstructing the Luo, which leads to the CRPS symptoms of paleness, coolness, swelling, and painful sensations that are deep and exacerbated by pressure; Qi stagnation and Blood Stasis, which lead to the CRPS condition of painful tension and cold sensations on the distal extremities and fixed, constant pain; and finally, a combination of Blood and Yin Deficiency, which engenders CRPS-related weakness, atrophy, occasional contracture, tingling, and numbness of the affected limbs.²⁷ Acupuncture corrects the disruptions in harmony and helps balance Yin and Yang.²⁸

Selection of acupuncture points for the 3 pediatric patients with CRPS in this report was based on TCM principles. Regional acupuncture points can be used to help patients who have CRPS symptoms. Corresponding points on the contralateral side may also be used if the regional point is at a painful site.²⁹

GB 34 was used; it is an influential *Hui* point for tendons.³⁰ As Sun stated, it is a Sea point that eliminates Damp and clears Heat; GB 34 is also a gathering point for the tendon and regulates circulation of Qi.³¹ The following descriptions are based on Sun's explanations.³¹ TE 5 and LI 4 were used to dispel external pathogenic Wind, Cold, and Damp and to relieve external symptoms. LI 4 also promotes circulation of Qi in the channels. SP 6, the crossing point of the Kidney, Spleen, and Liver channels, was used to regulate the Blood and Qi and to relieve leg pain. LI 11 is another Sea point that helps LI 4 regulate the Qi. ST 36, a Sea point, activates the Spleen and Stomach, tonifies the Qi, and eliminates Phlegm-Damp. *Yin-Tang* and GV 20 were used to calm the Mind and regulate circulation of Qi. PC 6 is the connecting point of the Pericardium channel. LR 3 is the Stream and Source point of the Liver channel. Both PC 6 and LR 3 are used for regulating Qi circulation. BL 23 is the Back Transporting point of the Kidney, and it tonifies the Kidney and warms the Yang-Qi to dispel Internal Cold. KI 3, the Stream and Source point of the Kidney channel, tonifies the Kidney-Yin. KI 7, the Metal point, promotes secretion of Body Fluids in the Kidney channel and nourishes the Kidney-Yin. Local *Ashi* points were used to eliminate Damp, regulate Qi and Blood in the channels, and relieve leg pain.³¹

From the Western perspective, acupuncture might modulate CRPS-related pain through involvement of the nervous system at multiple levels of information processing.³² Insertion of acupuncture needles activates peripheral afferent-nerve fibers,³³ as well as various chemicals in the central nervous system (CNS).³³ Naloxone, an endorphin antagonist, blocks acupuncture-induced analgesia, suggesting the release of endogenous endorphins.^{34,35} Pain may also be modulated by activation of the serotonergic,³⁶ histaminergic, dopaminergic,

and noradrenalinergic systems.³⁷ Other potential mechanisms of acupuncture analgesia for CRPS relate to functional MRI (fMRI) findings. Neuroimaging research has shown that CRPS is characterized by central nervous system changes³⁸; for example, a fMRI study showed that there are abnormal activation patterns following evoked mechanical or cold stimuli in pediatric patients with CRPS.³⁹ fMRI can also be used to investigate the intrinsic activity of the brain and resting-state networks for insight into the disease state of many pain conditions.^{40–42}

Researchers have found that CRPS resting state connectivity is altered in children.^{43–45} The default-mode network (DMN)—a network that is activated at rest and deactivated during goal-directed behavior⁴⁶—has increased connectivity in CRPS compared to the healthy brain.⁴³ Acupuncture alters the brain's resting state and leads to deactivation of the DMN.⁴⁷

Both Eastern and Western literature suggest several possible mechanisms to explain the therapeutic effects of acupuncture on CRPS. This article described on an integrative approach for treating pediatric CRPS. The 3 pediatric patients reported having positive healing experiences from acupuncture while simultaneously receiving conventional medical therapy. None of the patients in this case series experienced any adverse effects from acupuncture treatment. Successful management of CRPS utilizes a multidisciplinary regimen,²² such as pharmacologic and interventional treatments,⁴⁸ physical therapy,²² and cognitive-behavioral therapy.²² Acupuncture is an appealing adjunct to conventional therapies because of its low risk profile and its potential value in modern pain therapy for children with CRPS.

As a retrospective chart review, this report has limitations. There were no control patients, so it was not possible to compare patients who received acupuncture with patients who did not receive acupuncture. The small number of patients included in the case series also limits the generalizability of this report. Further research on a larger population of pediatric patients with CRPS is required to ascertain more definitively the effect of acupuncture treatment on recovery from pediatric CRPS.

CONCLUSIONS

While some studies have shown that adult patients with CRPS who receive acupuncture treatment have decreased pain levels, improved QoL, and functional improvement,^{16–18} there is a paucity of data on the effectiveness of acupuncture in the pediatric CRPS population. The 3 children with CRPS in the present case series had decreased pain scores and symptomatic reductions following acupuncture treatment integrated with conventional treatments. Acupuncture is safe to implement in children,⁴⁹ and the patients in this case series had no adverse side-effects. The results in this report suggest that acupuncture is a

promising adjunctive treatment for pediatric CRPS. Future research and prospective, randomized controlled trials on a larger pediatric population are necessary to understand and evaluate further the clinical effectiveness and viability of acupuncture for managing pediatric CRPS.

AUTHOR DISCLOSURE STATEMENT

No competing financial interests exist.

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