Successful Reversal of Complex Regional Pain Syndrome Type 1 of Both Upper Extremities in Five Patients

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Dear Editor,

Bilateral complex regional pain syndrome (CRPS) appears to be unusual [1–5]. We report details of bilateral CRPS diagnosed by International Association for Study of Pain (IASP) criteria in five patients who presented with florid symptoms (more prominent in one extremity) that persisted despite physical therapy (PT) and medications (Figure 1). They were effectively treated with a novel multimodality treatment regimen (MMTR) developed from our experience in over a hundred patients of CRPS-1 in the last 10 years.

Documented details of the patients included pain severity on verbal rating scale (VRS) at rest and movements; motor features (dystonia and disabilities); swelling; redness; and temperature changes using skin thermometer. Objective details documented included range of motions (ROMs) from shoulder to distal interphalyngeal (DIP) joint by goniometry and hand grip using dynamometer. This allowed scoring on disability of arm, shoulder, and hand (DASH) scale. Ultrasonography (USG) of the forearm muscles was performed pre and post MMTR (Figure 2).

Patients 1–4 were treated with MMTR comprising medication with highest tolerated dose of amitriptyline, pregabalin, and tramadol; stellate ganglion block (SGB) for the less affected extremity; continuous brachial plexus block (CBPB) for 4–5 weeks for the more affected extremity; dry needling (DN) and PT of muscles of both upper extremities. The fifth patient, unable to afford the cost of CBPB, was treated with SGB, DN, and PT in the more affected left extremity and only DN, PT in the right extremity.

CBPB was accomplished via 0.125% bupivacaine 1–2 mL/h infusion from a patient-controlled analgesia (PCA) pump (Ambit®, Sorenson Medical, Salt Lake City, UT, USA) for 21–38 days. Bolus of 3–4 mL of 0.125% bupivacaine, with 2-hour lockout interval, was programmed and could be activated for pain >3VRS during PT and DN. Patients went home with an antibiotic schedule and instructions for PCA pump use. Wound dressing and pump refill were done weekly.

DN of posterior neck muscles and extensor aspects of both extremities alternated with DN of pectorals and flexor aspect of both extremities for 40 days. DN involved slow, incremental advancement of 32 gauge, 25–50 mm long needles over 3–5 minutes. USG-guided DN was done for those specific muscles affected by the stiffness of CRPS. DN was followed by PT. Patients were followed up by telephone interviews up to 1 year.

Results

SGB and CBPB improved pain, swelling, and warmth but not motor impairment. Paradoxically, warmth, redness, and swelling recurred intermittently up to 15 days especially after PT and daily activities. We documented a reduction of these symptoms as well as stiffness after each DN session. This enabled pain-free passive mobilization of the needled muscles. This improved the ROM by 4–5° at several joints. Needling of the digital extensors resulted in a marked improvement in finger flexion, whereas needling of the digital flexors improved finger extension. Patient 5 achieved relief of all symptoms with only DN for his right limb. Successive DN sessions led to a global motor improvement within 5 weeks coinciding with return of normal muscle architecture on USG (Figure 2). All patients progressed from total disability to complete motor recovery (DASH score reduction from 70–95 to 9–15). They continue to maintain their prior lifestyle to date.

Discussion

Each component of MMTR served a specific purpose. SGB reduced pain, vasomotor, and sudomotor changes. CBPB continuously suppressed the same symptoms in the more affected extremity leading probably to reversal of neuronal plasticity. DN and PT, common to all the 10 limbs treated, appeared to specifically address the motor pathology of CRPS-1. DN relaxed the stiffness, making PT
effective. ROMs measured before and after DN showed a predictable and consistently reproducible increase.

CRPS-1 patients struggle to achieve any purposeful movement because of extreme motor stiffness. We propose that this movement difficulty leads to strain on the digital flexor and extensor tendons that traverse snug fibrous tunnels along the fingers [6]. Attempts at movement provoke repeated friction and irritation between the tendons and their surrounding synovial sheaths, leading to
inflammation inside these sheaths. This inflammation, seen as swelling, redness, and pain in fingers and hand affected by CRPS-1, is primary to the hand itself rather than a neurogenic inflammation as proposed previously [7].

Muscle relaxation by DN reduced the inflammation leading to temperature reduction as documented in all patients with skin thermometer. We attribute this effect to the muscle relaxation that reduced the movement-induced friction and inflammation. Muscle relaxation also reduced the excessive motor activity inherent to the spasm/stiffness/contracture, a hallmark of CRPS-1.

Pre-MMTR USG showed that the well-defined distinction between hypoechoic muscle fascicles enveloped by hyperechoic fascia of a normal muscle was lost in our patients and was replaced by uniformly hyperechoic fibrous tissue with no delineation of fascicles, or even of the muscle itself [8]. Restoration of normal movements after successive DN sessions was accompanied by a gradual, discernible return toward the normal appearance of hypoechoic muscle fibers with hyperechoic septae on USG (Figure 2). In addition, USG demonstrated, in real time, the previously described response [9] of a painful muscle to needle introduction viz., the initial muscle twitch and flutter in response to needle introduction followed by quiescence that our patients described as relief of pain and reduction of stiffness. This has been described as a needle-induced reflex relaxation [9], probably mediated by inverse stretch reflex or Renshaw cell-mediated inhibition of extrafusal fibers [10].

To conclude, we propose that motor impairment is the primary feature of CRPS with the other manifestations like pain temperature and swelling being secondary to local inflammation. SGB and CBPB alleviated these secondary but clinically dominant features like pain warmth and

Figure 2  USG of the more affected forearm of patient 4 (right flexors) and patient 5 (left extensors) before (images 1 and 3 on the left) and after treatment (images 2 and 4 on the right). Patient 4 had multimodality treatment regimen with continuous brachial plexus block, stellate ganglion block, dry needling, and physiotherapy. Patient 5 had only stellate ganglion block and dry needling. The images 1 and 3 on the left, documented at the first visit, show a mass of undifferentiated tissues with no delineation of fascicles, or even of the muscle itself. The complete destruction of structure makes it impossible to identity individual muscles. The images 2 and 4 on the right, documented at the end of the treatment, show a clear differentiation of hypoechoic muscle fascicles enveloped by hyperechoic fascia of a normal flexor and extensors.
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swelling, while DN and PT addressed the primary motor pathology. Together, all these interventions (MMTR) were responsible for complete resolution of CRPS including disability, in this small cohort of five patients with bilateral CRPS (10 CRPS-affected limbs).

Sincerely,

LAKSHMI VAS, MD
Interventional Pain Management
Ashirvad Institute for Pain Management and Relief
Mumbai, India

RENUKA PAI, Dip. Anaesthesia
Interventional Pain Management
Ashirvad Institute for Pain Management and Research
Mumbai, India

References


