Reversal of Complex Regional Pain Syndrome Type 2 and the Subsequent Management of Complex Regional Pain Syndrome Type 1 Occurring after Corrective Surgery for Residual Ulnar Claw

Dear Editor,

Complex regional pain syndrome type 1 (CRPS-1) that has previously resolved can recur after repeat surgery at the same anatomic site [1]. No existing treatment reverses CRPS-2 or a recurrent CRPS-1 [2–7]. We report the reversal of initial CRPS-2 as well as the subsequent CRPS-1 after surgery for residual ulnar claw with a multi-modality treatment regimen (MMTR) developed by us [8].

A lady of 39 years presented with 3 months of CRPS-2 following surgery for humerus fracture. Electromyography confirmed radial and ulnar nerve involvement. The Disability of Arm, Shoulder and Hand (DASH) score was 90.2 [9]. Ultrasonography (USG) showed hyperechogenicity in CRPS-2-affected muscles (Figure 1). She was treated with MMTR comprising medications (daily nortriptyline 35 mg, pregabalin 150 mg, tramadol 100 mg, and diclofenac as needed), continuous brachial plexus block (CBPB) for 4–5 weeks, dry needling (DN) of muscles of CRPS extremity, and physical therapy (PT).

CBPB was performed with nerve stimulation under USG guidance in operating theater. A patient-controlled

References


analgesia (PCA) pump (CADD-Legacy®, Smiths Medical MD, Inc., St. Paul, MN, USA) maintained infusion of 0.125% bupivacaine at 1–2 mL/h and 3–4 mL bolus with a 2-hour lockout interval. She went home with instructions regarding PCA pump usage: to stop the pump if sensory/motor deficits developed, bupivacaine boluses for pain beyond three Verbal Rating Scale and pump refill every 7–9 days were provided. Oral cefoperazone (500 mg, 1060

Figure 1 (A) Muscles of the normal left upper extremity showing a well-defined distinction between hypoechoic muscle fascicles enveloped by hyperechoic fascia characteristic of a normal muscle. (B) Muscles of CRPS-2-affected right limb flexor carpi radialis (FCR), flexor digitorum superficialis (FDS), flexor digitorum profundus (FDP) show reduction in size and hyperechogenicity indicating fibrosis, whereas pronator teres (PT) is showing normal echogenicity; extensor digitorum (ED), supinator (S), biceps, and brachialis show hyperechogenicity, loss of outline, and reduction in size. (C) FCR, FDS, and FDP showing increase in size and appearance of new muscle fibers replacing fibrosis; ED showing new muscle fibers replacing fibrosis; biceps and brachialis showing new muscle fibers, appearance of outline, and increase in size. A-A, B-B, C-C, D-D are calipers put to measure the thickness of muscles; BI = biceps; BR = brachialis; CRPS = complex regional pain syndrome; EXT or E = extensors; H = humerus; N = normal; PL = palmaris longus; R = radius; Rt = right; U = ulna; V = vessel.
(A) The right upper extremity appears wasted. At presentation, patient’s elbow was fixed in 90-degree flexion and in mild pronation. Weakness restricted the wrist extension to 5 degrees. Thenar eminence as well as the dorsal aspect of the first web space show wasting. (B) First web space wasting was no longer obvious (compared with A). Inset shows the ability to abduct, a function subserved by the intrinsic muscles of the hand supplied by ulnar nerve. The little finger is beginning to develop an ulnar claw. At this time, her only complaint was intermittent stiffness (ranging from 0–1 up to 10, which was described as a “crane-like” extensor pull at the fifth metacarpophalangeal joint [MPJ]) worsened by temperature changes and immobility. (C) The X-ray of hand seen from the side shows a claw with fixed flexion deformity at proximal interphalangeal joint. (D) K wire fixation of the fifth metacarpophalangeal joint (MPJ) joint. (E) The hand after removal of the splint and the K wire showing the increased temperature, swelling, and color changes. She still had continuous infusion and was using the bolus facility through the CBPB regularly to ensure continuous pain relief. (F) The swelling at the MPJ of little finger is very obvious when seen from the side. (G) Operated right extremity showing DN in the forearm above the splint. (H) Forearm showing pre- and post-DN temperature change from 37 degrees to 35 degrees on the extensor aspect of right forearm proximal to the splint. The reduction in temperature is at 40 minutes after DN. Patient reported a corresponding reduction of stiffness and pain after DN. (I) She is able to make an effective fist though the flexion at the last two MPJs remains less than that at the first two MPJs. CBPB = continuous brachial plexus block; DN = dry needling.
twice daily) and weekly wound dressing enabled the maintenance of the tunneled catheter for 5 weeks.

DN targeted muscles of the neck and upper extremity up to 40 days with flexors alternating with extensors every other day. 32G, 25–50 mm long needles were inserted slowly over 5–10 minutes because of pain and resistance of muscles perceptible to the operator as a grating sensation. Needles were removed after 30 minutes. USG-guided DN targeted specifically the individual muscles involved in performing the restricted/painful flexion, extension, abduction, adduction, pronation, and supination at various joints of the extremity.

CBPB reduced the rest pain but had no effect on movement pain, warmth stiffness, or edema. After DN, patient reported reduction of stiffness and movement pain. We documented reduction of temperature, redness, and swelling which would invariably recur intermittently after PT and daily activities in spite of ongoing CBPB. Pain-free passive mobilization after each session improved the range of motion (ROM) by 4–50 at several joints. Active ROM followed later as the patient had to develop the requisite muscle power. Needling of extensors resulted in a marked improvement in flexion, whereas needling of flexors improved extension. By 35 days, she achieved complete functionality including weight training (2 kg). DASH improved to 38.8. This global motor improvement coincided with return of normal hypoechoic muscle, discernible outline, and thickness on USG (Figure 1). However, residual issues like elbow flexion restricted by humerus implant to 1,500; little finger numbness and stiffness persisted with the beginning of ulnar claw. USG showed dense fibrosis around the ulnar nerve.

The claw at the fifth metacarpophalangeal joint (MPJ) (Figure 2), stiffness, and numbness worsened over next 6 months and was unresponsive to USG-guided injection of triamcinolone (10 mg) into fibrosis around the ulnar nerve above the elbow, a stellate ganglion block with 40 mg triamcinolone (10 mg) into fibrosis around the ulnar nerve, and injection of botulinum A toxin (40U) into the interossei and extensor digiti minimi.

Ten months after recovery of complete functionality and musculoskeletal sonoanatomy, she underwent extensor tenolysis, realignment, and K wire stabilization of the fifth MPJ under repeat CBPB. Surgery corrected the fifth MPJ claw but caused florid CRPS recurrence with swelling, discoloration, stiffness, warmth, and pain in spite of ongoing CBPB (Figure 2). DASH went up to 89.8. DN again relieved stiffness/temperature asymmetry after every session (Figure 2) to completely reverse CRPS in 1 month. DASH became 9.8 indicating complete functional rehabilitation. Two years later, the patient maintains her pre-fracture lifestyle including weight training with 10 kg.

Complete reversal of CRPS-1/2 and the associated disability is unusual [10,11], particularly the return to prior functionality after a recurrence of CRPS-1 triggered by surgery. In this patient, not only was the initial CRPS-2 reversed but also the recurrent CRPS-1 with complete reversal of disability on both occasions. We attribute this to the resolution of the motor impairment with DN. We believe motor impairment plays a pivotal role in CRPS pathogenesis as well as in the disability. We have reported hyperechogenecity of muscles with disruption of muscle architecture on USG in CRPS [12] indicating fibrosis with resultant reduction of muscle elasticity probably with formation of multiple myofascial trigger points (MTrPs). We surmise that the unrelenting stiffness of the finger flexors and extensors that makes movements difficult/painful cause friction between the digital flexor/extensor tendons and the synovial sheaths inside the snug fibrous tunnels along the fingers with resultant inflammation [13]. The ongoing release of inflammatory mediators by the repeated attempts at movement sustains the inflammation, which then presents as the classical clinical features of CRPS in hypertrophic phase like red, hot swollen hand. Persistent pain leads to central sensitization and sympathetic maintenance. The persistent severe pain leads to avoidance of movement. DN is a novel modality that relaxes the MTrPs [14,15] reducing the muscle stiffness. The resultant untethered tendon movement within the synovial sheaths attenuates friction and synovial inflammation, ultimately resulting in a return of normal movements. Thus, DN has the potential to effectively reverse the whole pathology of CRPS, restoring work ability to muscles working across several joints of the extremity. Medications and CBPB of MMTR relieved the severe pain in the hypertrophic phase enabling our patient to make coherent therapeutic decisions. The PCA bolus facility reduced the pain of DN and PT. PT worked synergistically with DN to reverse motor pathology. USG provided an objective corroboration of the motor improvement.

The modalities of MMTR served specific purposes in reversing the CRPS-2 and the recurrent CRPS-1 in our patient. MMTR was developed with the premise that motor impairment forms the primary pathology of CRPS and is amenable to reversal by DN along with PT; CBPB and medications address pain and other features that are secondary.

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Penetration Approach for Repeat Vertebroplasty Through the Previously Impacted Bone Cement

Dear Editor,

Vertebroplasty (VP) and kyphoplasty (KP) are effective treatments for painful vertebral compression fractures (VCFs) [1,2]. Although not common, subsequent refracture in the previously augmented vertebra causes persistent or recurrent pain [3]. Some reports have shown that repeat VP can control pain caused by refractures [3–5]. However, repeat VP is challenging because the previously impacted bone cement blocks the fluoroscopic views and access of procedure needles.

A 57-year-old woman presented with severe pain (visual analog scale [VAS] 8/10) in her lower back and both thighs, which left her unable to sit or walk. She had a history of rectal cancer that had metastasized to the liver and lungs. Six months previously, metastasis to the fourth lumbar vertebra was detected and KP was performed (Figure 1). After the procedure, her pain reduced from VAS 8 to 2, but it recurred a week before her visit to our clinic. Magnetic resonance imaging (MRI) revealed further metastatic progression in the L4 vertebral body, and bone cement was located in front of the left pedicle (Figure 2).

After the failure of conservative treatments, we undertook repeat VP with a unilateral intrapedicular approach through the right pedicle.

The patient lied prone on the procedure table. Under fluoroscopy, we aligned the endplates of the L4 vertebral body. Because the previous bone cement was blocking the fluoroscopic views, we set the target point by comparing the relative locations of the pedicles in the adjacent vertebrae. After local anesthesia with 1% lidocaine, we made a small skin incision, and inserted a 13-gauge bone access needle through the right pedicle of L4. After checking the anteroposterior and lateral fluoroscopic images, the needle was advanced into the vertebral body. We injected 4 mL of polymethylmethacrylate (PMMA), which fully filled the L4 vertebral body in the lateral view, but this failed to fill into the left side of vertebral body.

After deliberation, we chose to insert another needle through the left pedicle to penetrate the previous bone cement. Under fluoroscopic guidance, we inserted the needle, using the same method as with the right side, until