Complex Regional Pain Syndrome-Type 1 Presenting as deQuervain’s Stenosing Tenosynovitis

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Aim: To report the presentation of complex regional pain syndrome-1 (CRPS-1) as deQuervain’s stenosing tenosynovitis (DQST).

Case report: A 24-year-old woman presented with a 3-year history of clinical diagnostic criteria (CDC) of CRPS-1. Conservative and surgical treatment for this as DQST had failed to relieve her. We diagnosed the problem as CRPS-1 with CDC as inflammatory manifestations of mechanical tenosynovitis of all her 5 digital tendons caused by movement of the fingers and hand tethered by agonist (flexor)/antagonist (extensor) muscles in co-contraction. Ultrasound guided dry needling (USGDN) relaxed the muscles, replacing the abnormal agonist/antagonist co-contraction with normal agonist/antagonist coordination. Resolution of tenosynovitis reversed the inflammation causing the CDC. Six months later she leads normal personal and professional life, with reduction of scores of painDetect (from 21 to 5), Patient Health Questionnaire (from 13 to 4), Disability of arm, shoulder and hand from 70.8 to 25 and reversal of muscle abnormality characteristic of CRPS-1 on Musculoskeletal Ultrasonography (MSKUSG).

Conclusion: We believe the primary pathology of CRPS-1 to be co-contraction of agonist (flexor)/antagonist (extensor) muscles of digits resulting in tenosynovitis akin to DQST. CDC of CRPS are actually inflammatory manifestations of tenosynovitis amenable to reversal by USGDN which also addresses the disability, a hallmark of CRPS.

Key words: Complex regional pain syndrome-1 (CRPS-1), deQuervain’s stenosing tenosynovitis (DQST), neuropathy, co-contraction, dry needling (DN), ultrasound guided dry needling (USGDN), musculoskeletal ultrasonography (MSKUSG)
conservative and surgical treatment for DQST worsened the pain and disability. We treated the condition as CRPS with co-contraction. Resolution of co-contraction with USGDN reversed not only the co-contraction of the agonist/antagonist muscles responsible for the inflammatory mechanical tendinoses causing the CDC of CRPS but also the disability.

**Case Report**

A lady baker aged 24 years presented with positive Budapest Clinical Diagnostic Criteria. She had pain (6/10 at rest on numerical rating scale [NRS] [31] exacerbating to 10/10 on hand movements), sensory findings like stroke allodynia, hyperalgesia to light touch, pinprick, and cold > warmth over forearm (Fig. 1), 1° temperature increase and swelling (vasomotor and sudomotor features). Motor findings included severe stiffness, heaviness, and flexion/extension restriction at wrist. Elbow movements were normal but painful. Shoulder flexion and abduction beyond 90° were painful. External and internal rotation also produced pain in the axillary folds in the end range of movement. Neck movements were restricted by pain (600 flexion/extension and 20 – 30° lateral flexion). She had no nail/hair changes. Her painDETECT score was 21 (a score of 18 – 38 has a > 90% likelihood of neuropathic pain) (32). Disability of arm, shoulder and hand (DASH) score was 70.8 (33) indicating considerable disability. Personal Health questionnaire (PHQ 9) was 13 indicating a major depressive disorder (34). She was unemployed for 3 years and had expressed suicidal ideation. Validity scale L of Minnesota Multiphasic Personality Inventory

Fig. 1. Pre and post treatment clinical pictures.
Row 1: The right hand shows mild swelling and a temperature difference of 1° compared to the normal left hand. She had stroke allodynia, hyperalgesia to light touch, pinprick, and cold > warmth over the marked areas of distal 2/3 of forearm. The words “allodynia” and “hyperalgesia” on the patient’s right forearm were written after the first dose of midazolam + ketamine prior to DN. There was a definite response of flinching but she allowed it willingly and was quite coherent to indicate the areas to be marked. Care was taken to write very lightly. Movements were severely painful at all the joints of the thumb and moderately painful over the other fingers. Both flexion and extension were restricted at the wrist.
Row 2: Dynamometer reading of right hand was 2 pounds per square inch (psi) as compared to the 6 psi on left. But at 30 days the grip strength has increased to 8 psi. The measurements on this type of dynamometer give a lower reading as compared to the Jamar dynamometer but in this patient both the pre and post measurements were carried out with the same bulb dynamometer hence they reflect the true improvement in this patient. The flexion and extension at the wrist also has become normal.
Her symptoms had started 3 years prior, when she was 21 years with pain at the base of the second and third fingers after a bout of hand overuse as an apprentice baker (after shelling 2 kilos of walnuts). The pain and swelling progressed to the whole hand but mainly centered near the thumb. This remained unresponsive to physiotherapy and various analgesics prescribed by orthopaedic surgeons, rheumatologists, and psychiatrists over the next 2 years. Rheumatoid factor, C-reactive protein, and cyclic citrullinated peptide antibody were normal. Triple phase bone scan had suggested CRPS involving small joints of the right hand and wrist. Two consecutive steroid injections into the abductor and extensor tendons under ultrasound guidance had actually worsened her condition. Surgical release of the first extensor compartment 2 years later relieved her pain only for a month. Gradually over the next year, recurrent episodes of pain and stiffness had remained unresponsive to analgesics, neuromodulators (duloxetine, pregabalin, amitriptyline), and to 2 courses of oral steroids for 7 days with the dose tapered thereafter. She reported that there were intervals of reduced pain when she could use her hand minimally, but this would result in a flare up of activity-triggered pain, swelling, and warmth not only in the hand, but also the elbow and shoulder. At the time of presentation to us, the pain and disability had involved the whole of right extremity.

We explained to the patient about our concept of CRPS manifesting as mechanical tendinoses and the logic of treatment with USGDN. She refused stellate ganglion and continuous brachial block for pain relief but consented for USGDN with sedation. Possible complications of pain/heaviness during the USGDN and ecchymosis after USGDN were explained. She chose to defer psychological counseling for her depressive symptoms until after her pain was controlled. She attended 2 psychological counselling sessions while she was asked to challenge her ability (like manual preparation for baking bread > 5 kg cakes, playing carom etc.) to eliminate any residual disability. She continues with a normal professional life at 6 months with painDETECT score of 3, PHQ of 1, DASH of 6.3 (90%), and has presently gotten engaged to be married.

**Discussion**

This case presentation exemplifies our surmise that the movement difficulty of CRPS is primarily because of abnormally co-contracted digital flexor/extensors and that it is the basic pathology of CRPS. All the clinical manifestations that form the CDC of CRPS are actually manifestations of a global mechanical tendinoses of the flexor/extensors of all the fingers including the thumb. Our patient had originally presented at 21 years of age while the normal age of presentation of DQST is 30 – 55 years of age (36-41). One DQST survey shows an age category of greater than 40 as a rate of 2.0 per 1,000 person-years compared to 0.6 per 1,000 in persons under 20 years (40). The results of steroid injections in DQST are reported to be good for about 4 months (42,43) and DQST surgery also reports good results (44,45). Our patient’s symptoms had worsened with both conservative and surgical treatment of DQST which had left the hand useless with a DASH score of 70.8 (33). It was only after recognition of the condition as a mechanical tendinoses resulting from the use of
Fig. 2. Muscle ultrasonography shows the wasted muscles in the right extremity.
Row 1: The combined bulk of the flexor carpi radialis (FCR) and palmaris longus (PL) is 0.90 cm compared to the left hand FCR size of 0.97 cm. The pronator teres (PT) is 0.67 cm compared to the left hand PT which is 1.4 cm. The other muscles have indiscernible borders and hence were not measured. The features of wasting in CRPS differs from that of disuse in that muscle with disuse wasting have clear-cut muscle outlines, and the typical sonographic signature of a normal hypoechoic appearance with bright streaks of intramuscular septae. Whereas in CRPS patients like this, the muscle outlines are blurred and the contrast of hypoechoic muscle with hyperechoic septae becomes replaced by hyperechogenicity as seen in FCR and FDP. The same right hand after 45 days of dry needling shows an obvious increase in muscle bulk as well as the return of definition to muscles. Pronator teres size has increased to 0.70 cm and FCR and PL together measure 0.49 + 0.53 = 1.2 cm. The flexor digitorum superficialis (FDS 1.59 cm) and flexor digitorum profundus (FDP 1.06 cm) are well defined. R: radius; U: ulna; FCU: flexor carpi ulnaris.
Row 2: The first image show the sustained increase in size of flexors at 3 months. The second image shows the hyperechogenicity of pectoralis minor (P. MIN) which has become normal by 45 days. PM: pectoralis major; BR ARTERY: brachial artery.
Row 3: Shows the extensors. At 45 days, the extensors and supinators also show an increase in size. BR: brachioradialis; S: supinator; ED: extensor digitorum; SUP: supinator.
Row 4: Shows the synovial effusion of the right hand extensors compared to the normal left hand tendons. The effusion around the extensor tendons has resolved by 45 days. 2, 3, 4: bases of the numbered metacarpals respectively; C: carpals; T: tendon.
abnormally co-contracted flexor/extensors of CRPS that specific treatment with USGDN could reverse the motor impairment as well as the CDC of CRPS.

The simultaneous co-activation of both agonists and antagonists impedes all hand movements. Moving the constrained muscles leads to friction, inflammation, and mechanical tendinoses at all 5 digits. Dolor of inflammation causes severe pain and rubor and calor manifest as CDC. Persistent muscle nociception, tendinoses, and pain lead to peripheral and central sensitization leading to proximal extension of CRPS features.

Muscle nociceptive activity has been described in Myofascial Pain Syndrome (MPS) to activate dorsal horn neurons, causing central sensitization, hyperalgnesia, and referred pain. Shortening of the contracted taut painful bands in muscles restricts movements. Impaired reciprocal inhibition results in co-contraction of agonists and antagonists, thus interfering with fine motor control and coordination. Autonomic disturbances like changes in skin temperature and color, and piloerection can accompany myofascial trigger point (MTrP) activation (46).

Once specific treatment with USGDN addressed the abnormally co-contracted flexor/extensors, her pain and motor impairment disappeared as well as the swelling, warmth (within 10 days), and disability. DASH score became 25 (64.69% improvement) within 45 days, enabling her to return to her baking profession. The logic of USGDN was that it would relax both the groups of co-contracted muscles replacing the abnormal agonist/antagonist co-contraction with the normal agonist/antagonist coordination. The movements of the coordinated muscles are no longer impeded and thus, there would be little cause for friction at the flexor-extensor tendons. The tendinoses responsible for causing the CDC (warmth, swelling, color, nail and hair changes if present) would in turn disappear with resolution of the inflammation.

DN has been described as a specific treatment for MTrPs. Since DN appears to resolve the motor impairment of CRPS, it is logical to assume that MTrPs in both agonists and antagonists might be involved in the mediation of co-contraction. We have emphasized the role of the myofascial system in the causation of chronic post surgical pain through the mediation of motor neuropathy/neuromyopathy which also responds to USGDN (47).

The pathophysiology of MTrPs, the role of a compromised local blood supply, hypoxia, and acidosis at the MTrPs in causing muscle pain and dysfunction in
The hypothesis of co-contraction and tendinoses in CRPS is no doubt novel and unusual but is based on our clinical experience of uniform reversal with complete resolution of disability in 150 patients presenting in various phases of CRPS over the last 12 years (28-30). However, the concept of myofascial pain (MPS) in CRPS is not new. MPS has been reported in 61% of a series of 41 CRPS patients (57). EMG abnormality of antagonist co-activation has also been previously reported in CRPS (58). It is just that our hypothesis considers MPS as the initiating pathology that gives rise to all the other manifestations of CRPS. The severe pain of acute tendinoses recruits the sympathetic response which eventually leads to sympathetic maintenance of pain. It is only after contractures develop in the co-contracted muscles to prevent movements of the hand that the tendinoses abates, to give rise to the cold extremity of very late CRPS.

There are 2 reports on the histopathology of CRPS-1 affected muscles which report muscle fiber atrophy. One reports both type 1 and type 2 fiber atrophy (59) with extensive changes in muscle tissue, such as fatty degeneration, fiber atrophy, and nuclear clumping, which was not related to duration of CRPS-I prior to amputation of the CRPS limb. Another reports only type 1 fiber atrophy and nerves showed no consistent abnormalities of myelinated fibers but in 4 patients, the C-fibers showed electron microscopic pathology (60).

MSKUSG provides objective information of loss of myoarchitecture and of effusion in the digital synovial sheaths. We propose a correlation between MSKUSG findings with movement difficulty, weakness, and stiff-
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neness of CRPS. The vasomotor and sudomotor changes are actually a manifestation of inflammation in the tendinoses. In our experience, both stellate ganglion block (SGB) and continuous brachial plexus block (CBPB) are useful, but only for pain relief and neither procedure has any effect on motor impairment. Physiotherapy is often overwhelmed by the recurrence of pain and inflammation from the co-contraction which persists in spite of SGB and CBPB. But when DN is added, CRPS reversal becomes routine. However in patients with hyperalgesia CBPB/SGB/oral ketamine, midazolam analgesia have proven to be useful.

Conclusion

We propose that CRPS could be mistaken for DQST since both are tendinoses presenting with inflammation. The CDC of CRPS are actually inflammatory manifestations of multiple mechanical tendinoses secondary to an abnormal co-activation of the agonist/antagonist muscles of digital movements. This appears to resolve with USGDN which reverses the co-contraction, resulting in resolution of CDC as well as disability; thereby indicating a role for primary myofascial pathogenesis in CRPS.

References


