

UNDERSTANDING PAIN MECHANISMS IN THE REHABILITATION OF A MISMANAGED POSTTRAUMATIC ADHESIVE CAPSULITIS OF ANKLE WITH COMPLEX REGIONAL PAIN SYNDROME

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Background: Pain has different input mechanisms, a subsequent central processing and an appropriate output mechanism. Accordingly the pain history of a patient varies, and so should the surgical and rehabilitative management.

Case Report: The case report is based on a 20-year-old woman who had a 3-year-old distal tibiofibular fusion surgery done post traumatic interosseous membrane injury with a conservative approach for other multiligamentous injury of the ankle joint mortise. Despite the initial surgery and rehabilitation, she had episodes of fall due to instability even thereafter. Due to a series of unsuccessful interventions over the years, she developed excessive muscle guarding and capsular stiffness in the ankle joint. Her pain persisted for 3 years and she eventually developed complex regional pain syndrome.

Conclusion: This case demonstrates the rehabilitation of a patient based on understanding of the appropriate pain pathways along with the structures involved. It also discusses the etiology of complex regional pain syndrome developed 3 years after her surgery.

Key words: Adhesive capsulitis, complex regional pain syndrome, multiligamentous injury, ankle joint

BACKGROUND

Pain is an aversive sensory and emotional experience typically caused by, or resembling that caused by, actual or potential tissue injury. It has a biological, psychological, and social component (1). Pain has different input mechanisms, a subsequent central processing and an appropriate output mechanism (2). Broadly, there are 5 input components of pain: central sensitization, peripheral sensitization, peripheral nociceptive mechanism, sympathetically dependent mechanism, and cognitive affective mechanism (3,4). Accordingly, the pain history of a patient varies, and so should the surgical and rehabilitative management.

CASE

The case has been compiled keeping in mind the CARE guidelines. A 20-year-old woman came to a tertiary care hospital with the chief complaints of excessive pain in the left ankle with restricted range of ankle motion adding to an altered walking pattern. She rated her pain to be 7 of 10 on a visual analog scale at rest and 9 of 10 on movement, weight-bearing, as well as on touch by another person. She described her pain to be more like a burning type.

She had a history of fall while descending the stairs about 3 years ago, when she developed grade 3 tears of the deltoid, talofibular ligament, and the interosseous

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membrane of the left ankle as shown in the magnetic resonance imaging (MRI) then. The orthopaedic surgeons advised her to undergo a distal tibiofibular fusion with 2 cortical screws to cover up the interosseous membrane rupture. The other ligaments of the mortise were managed conservatively and her left leg was kept in a cast post surgery for 6 weeks. The postoperative investigations and surgical details were not preserved by the patient. Thereafter, she started graded yet vigorous traditional physiotherapy, following which she was almost back to her routine in the next 3 months despite the persistence of pain. Thereafter, she had 3 consecutive falls in the same year and every time she would be treated conservatively using a plaster. Probably due to these repeated falls and plastering, she developed severe muscle guarding and stiffness of the ankle joint. For 2 years, the patient has been undergoing different orthopaedic and physiotherapeutic treatments with no significant relief, which is why she chose to come to our tertiary care hospital in January 2020, where she underwent an MRI; the MRI now revealed interspersed areas of osteolysis and minimal edema in the dome of talus suggestive of probable avascular necrosis. It also revealed a chronic tear with remodelling of the tibiocalcaneal portion of the deltoid ligament, posterior tibiotalar portion of the deltoid ligament, interosseous talocalcaneal ligament, as well as the anterior talofibular, posterior talofibular, and calcaneofibular ligaments.

After taking an in-depth history, she was thoroughly assessed. On observation she had a darker discoloration of skin on the left foot as compared to the right foot, edema, sudomotor changes like patchy sweating, and warmth and a burning sensation on the dorsum of her left foot. Considering the motor, sensory, sudomotor, and vasomotor components of her symptoms, we classified her symptoms as complex regional pain syndrome (CRPS) per the Budapest criteria (5). Her range of motion was limited to between 10 and 15 degrees of plantar flexion, wherein further range came from compensation of the tarsometatarsal and metatarsophalangeal joints rather than the talocrural joint. The inversion, eversion, and dorsiflexion range was nil from the talocrural joint. There was 2-cm swelling when checked using the "figure of 8" method with measuring tape, as compared to her right foot. Further assessment was done using special tests to check for ankle instability; the valgus varus tests revealed no significant opening of the joint. This could have been a false negative owing to the guarding and tightness of the muscles and capsule around that joint with chronic remodelling of the ligaments as shown in the MRI. There

was a possible tightness of the gastrocnemius muscle, but it could not be confirmed because the arthrokinematics of the ankle joint was restricted; hence it could not be moved into dorsiflexion. The end feel of her existing range was firm, indicative of possible capsular involvement. All of this pointed towards adhesive capsulitis.

Her gait pattern was antalgic with minimal weight-bearing on the left leg using an ankle boot brace. Her major complaint was increased pain. She always underwent intense management to increase her muscle strength and proprioceptive response, but her pain was taken care of with painkillers. Previously, she had undergone sessions of dry needling and shockwave therapy for her adhesive capsulitis, but thereafter she reported an excessive increase in pain which was beyond the effectiveness of a pain killer. Therefore there was a need to further investigate to understand her pain.

Therapeutic Intervention

Considering the chronicity of her symptoms, there was a supposed hint of the central component being involved in her pain processing mechanism. She had been in pain for 3 years. There has to be a chain of neuroplastic changes in the brain that makes the pain neurotags more overactive as compared to others (6). Thus we see that an area that was probably the source of earlier symptoms shows increased pain responses, not at all proportional to the stage of tissue healing; and the adjoining areas like calf and knee joint, which are not even involved directly, show pain symptoms. This is because the adjoining areas in the homunculus also get sensitized due to the chronicity of symptoms (6). The first thing included in her therapeutic intervention was counselling and education regarding her condition, possible prognosis, and the importance of engaging in the right kind of systematic treatment (as shown Table 1).

Within 10 sessions, not only her movement improved in range and control, but also her left skin discoloration reduced, as shown in Fig. 1. She came up to 2 degrees of dorsiflexion. Thereafter there was a hard end feel, possibly due to the reduced joint space and avascular necrosis of talus as shown in the MRI reports. At present, the patient has been able to resume her activities of daily living in a rather pain-free way.

DISCUSSION

The novelty of this case lies in the etiology of CRPS in the patient. It occurred 3 years post her lower limb surgery. Recent studies done on CRPS typically mention

Table 1. Systematic chart of treatment progressions over a period of time

Stage of Treatment	Aim of Treatment	Techniques Used
First	Desensitization, tactile discrimination, and pain relief	<ul style="list-style-type: none"> Feeling softer cotton wisp progressing to silk cloth and then rougher textures, to reduce the hypersensitivity to touch on her left leg (10) Transcutaneous electrical nerve stimulation was given for 15 minutes per session to relieve her pain with 2 electrodes placed, one on each malleoli, and the other 2 electrodes placed on calf and sole of foot each. Mirror therapy using a mirror box covering her left foot. The mirror would portray the movements of her right foot and she had to imagine that the mirror image of the right foot was actually her left foot moving (10).
Second	Improving the range	<ul style="list-style-type: none"> Grade 1 Maitland mobilization to the tarsometatarsal, intertarsal, and talocrural joints (11) Immediately after the mobilization she was given cryotherapy using compressed air machine for 10 minutes to reduce the postmobilization soreness. Gentle gastrocnemius-soleus stretching and movement with mobilization for talocrural and subtalar joint (11)
Third	Control and strengthening for the pelvic girdle, the hip, and the knee along with the ankle.	<ul style="list-style-type: none"> Initially non-weight-bearing exercises were started, further progressing to weight-bearing exercises. Proprioceptive and balance training were started, initially using the firm surface and further progressing to foam surface and wobble board (12).



Fig. 1. Showing the progression of skin colour discoloration over 10 sessions.

its occurrence maximum one year post surgery or fracture (7). Even if her subsequent falls are to be considered as triggering factors, they were 2 years prior to the time she experienced symptoms (which fit the Budapest's criteria of clinical diagnosis). Thus the only causative factor could be the microtraumas that her limb received over a period of time, the last one being the session of dry needling and shockwave therapy for the adhesive capsulitis of her ankle. Probably the induced inflammation leading to the altered

vasomotor responses, along with the disproportionate sympathetic activity in the body with brain neuroplastic changes owing to the chronicity of her pain, may have been the major cause of her CRPS (7).

The above case report gives us insights into 4 very important aspects of treatment. Firstly it focuses on the correlation of the stage of tissue healing and the parameters of the symptoms the patient is presenting (8,9). If we treat simply based on the radiographic and clinical picture

it will not yield optimum results. Just like in the above case, chronicity of symptoms do lead to central sensitization, which has to be managed accordingly. Considering these factors, patient education forms an integral part of the management; the clinician needs to make the patient aware of the reason for different techniques being used and their effectiveness in that specific case. According to a study done by Pollard et al (10), there is emerging evidence for techniques including mirror therapy, tactile discrimination training, graded exposure therapy, and virtual reality in the management of CRPS. Hence we used these same techniques, described in Table 1, to tackle the symptoms of the patient. Treating her pain first becomes important, as in CRPS there are ongoing changes in vasomotor and sympathetic activity in the body. Hence subjecting her to other forms of stimulatory or noxious treatments for her adhesive capsulitis without controlling her pain would not give the same results as otherwise; on the contrary, this would exaggerate her symptoms. Secondly, there was Maitland mobilization given to the ankle joints, as studies have shown a significant improvement in the dorsiflexion ranges with talocrural and distal tibiofibular joint mobilization followed by gentle active stretching (11). Thirdly, with regards to management involving strengthening of the muscles proximal to the ankle joint as mentioned in Table 1, it is very important to do so to bring about stability in the lower limb kinematic chain, as many studies have shown that people with ankle sprains have proximal muscle weakness (12). Lastly, there is a need to optimize the type of surgery being done in such patients, as fusion of the distal tibiofibular joint without any ligamentous repair would not meet the functional requirements. Ac-

cording to a study done by Romero et al (13), there should be syndesmotic screw fixation, suture-button dynamic fixation, or repair of the anterior talofibular ligament with direct sutures, suture anchors, or screws with washers. In her case, there was no repair of the ligaments done; hence she kept having the falls, as all her ligaments surrounding the ankle joint mortise were dysfunctional. Her persistent pain caused guarding in the surrounding muscles, preventing them from functioning as stabilizers around the ankle joint. The tibiofibular fusion further caused a reduction in the talocrural range of motion owing to a reduction in the distal tibiofibular splay and reduced inferior and superior glide of the fibula with superadded unrepaired ligaments. Thus there is a future need to reexplore her ankle joint complex for long-term relief and recovery.

CONCLUSION

The case report discusses the probable etiology of CRPS in the patient and the various interventions applied to manage a patient with a postsurgical chronic pain condition. It highlights the occurrence of CRPS 3 years after surgery, unlike a typical CRPS presentation. Hence further research is needed to see if CRPS occurs so long after the inciting event or if it can also be caused by microtraumas over a period of time from the actual inciting event.

A holistic approach must be adopted in making the diagnosis and management of CRPS. Also, any treatment plan should always be built upon the goal of improving the patient's quality of life and must be designed keeping in mind the patient's expectations and their individual pain pathways.

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