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# Medical rehabilitation of a patient with CRPS type I after tibial plateau fracture and sprain of the ankle: A case report and Literature review

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## Abstract

Complex regional pain syndrome is a clinical syndrome characterized by chronic, severe, neuropathic pain, which is associated with sensory, trophic, and autonomic disorders and decreased range of motion of the affected joint. Symptoms begin at a variable interval, at least 6 weeks after a traumatic event or after surgery. The evolution of symptoms is unpredictable, as they can range from complete and self-limiting resolution to significant chronic pain with decreasing limb function and quality of life. Depending on the absence or presence of a well-identified neural lesion, types I and II of CRPS can be observed. Because it is a relatively rare condition, it can be overlooked due to the limited experience that doctors have with this one.

A 61-year-old male patient presented with severe pain 9/10 on the Visual Analogue Scale (VAS) and a significant decrease in mobility after suffering a traumatic fracture with a left tibial plateau fracture 4 months before, which required orthopedic treatment with a plate and screws, and a left ankle sprain. Previously, the patient presented to several emergency services where he received anti-inflammatory and analgesic treatment with gradual aggravation of pain and impaired mobility of the left lower limb, walking being possible only on two axillary crutches. The patient received treatment that combined drug therapy with medical rehabilitation exercises and physical therapy agents, with a favorable evolution. At the time of discharge, the pain was 5/10 on the VAS scale and the patient could move with a single Canadian crutch.

Keywords: CRPS, pain, trauma, fracture, quality of life

Abbreviations: CRPS = complex regional pain syndrome, VAS = visual analogue scale, TENS = Transcutaneous Electrical Nerve Stimulation, NMDA = N-methyl D-aspartate, IAPS = International Association for the Study of Pain

## Introduction

Complex regional pain syndrome (CRPS) is a clinical syndrome characterized by regional pain, which in most cases is severe and chronic, in association with sensorial and trophic skin changes, vasomotor instability, swelling and limited range of motion, symptoms that follows an acute tissular trauma. First, it was described in 1964 by Mitchell et al. and it has been known as reflex sympathetic dystrophy or causalgia [1]. Over time, in literature, the syndrome has been referred to as transient osteoporosis, Sudeck atrophy, algo dystrophy or acute atrophy of bone. CRPS is a neuropathic pain disorder that does not follow a nerve territory or a dermatome, and usually affects the distal part of a limb and is associated with autonomic features [2]. Besides classical neuropathic pain (allodynia, hyperalgesia, intense burning pain), CRPS is accompanied by modifications that suggest autonomic involvement (altered skin temperature, skin color) and local edema [3]. The evolution of the syndrome is unpredictable. It can have a self-limited evolution with mild pain, or it can have a chronic evolution with significant pain and impaired mobility, which requires complex management and sometimes the necessity of using a cane or a wheelchair. All these have a major impact on quality of life with a significant interference in the functional and general activities of the patients [4,5].

As mentioned above, there are two types of CRPS:

- type I, where the nerve injury cannot be identified, usually appearing after a fracture or after surgical interventions and is also known as reflex sympathetic dystrophy; and
- type II, where a specific nerve injury is documented and has been known as causalgia [1]. There is just a traditional diagnostic difference between type I and type II of CRPS because they do not differ in treatment

symptoms and pathophysiological mechanism [3].

## Case report

We present the case of a 61-year-old male patient, hospitalized for movement pain, with a score of 9/ 10 on the visual analogue scale (VAS), with limited range of motion in the left knee and ankle and a modified gait pattern with the impossibility of climbing stairs. Before presentation to our hospital, the patient visited different emergency hospitals, received pain medications, and was sent home. The Anamnesis revealed that four months before, the patient presented a Schatzker II fracture of the left tibial plateau and a sprain of the left ankle secondary to a knee trauma with a blunt object from the lateral part. The tibial plateau fracture was treated orthopedically with plates and screws. The clinical examination of the left knee revealed a supple postoperative scar, edema that extended distal to the knee and with maximal extension at the ankle, was swollen with a positive patellar tap and drawer sign. The skin was thin and pale, with diminished hair with thin hairs. When the joint line was palpated, the intensity of pain was exacerbated. The knee had an irreducible flexion of 10 degrees. Active flexion of the knee was 30 degrees, and the passive flexion was 35 degrees. Quadriceps hypotrophy with a muscular strength of  $\frac{4}{5}$  and hamstring retraction were observed. Ankle examination revealed a limited range of motion, pain that was exacerbated at night, as well as with palpation and mobilization. Walking was possible with the help of two axillary crutches. The patient had an abnormal pattern of walking with an antalgic gait (stance phase was shortened relative to the swing phase) secondary to pain. Biologically, blood tests were normal. Plain X-rays showed bone demineralization most likely due to spotted

osteoporosis, a plate with screws on the lateral side of the proximal third of the tibia without signs of movement, and calcifications in the projection area of the medial meniscus (Fig. 1,2). Ankle X-ray showed osteosclerosis with an irregular appearance in the internal ankle, possibly post-traumatic, and joint space with normal height (Fig. 3).



Fig. 1 Plain X-ray of the left knee lateral view



Fig. 2 Plain X-ray of the left knee anterior view



Fig. 3 Plain X-ray of left ankle-lateral view

Musculoskeletal ultrasound examination revealed acute patellar tendinopathy, heterogeneity of iliotibial band and collateral ligament (especially lateral), hematoma evolving in the knee joint, heterogeneity of the talofibular, tibiofibular, calcaneofibular ligament, tenosynovitis of peroneal muscles and fluid in the anterior recess of the tibiotalar joint. Following clinical and paraclinical examination, the patient's diagnosis was type I CRPS.

Treatment goals were to soothe the pain, increase the stability of the ankle, improve knee and ankle range of motion, prevent backbone and hip complications, improve the gait pattern, and improve quality of life. To accomplish these objectives, we used a varied treatment scheme, with drugs and physical agents like therapeutic exercises, electromagnetic, thermal, and mechanical agents, polarized light, and adjuvants. We used 100 mg tramadol twice a day for pain control, as well as an intramuscular injection of nonsteroidal anti-inflammatory meloxicam (15 mg/ 1.55 ml) once a day. To soothe the pain at the knee level, we also used Transcutaneous Electrical Nerve Stimulation (TENS) at 110 Hz for 20 minutes, laser therapy, which has, in

addition, anti-inflammatory and trophic effects, and TECAR therapy.

At the ankle level, we used laser therapy for pain and inflammation release. Ultrasound therapy was used to reduce pain, due to its myorelaxant and vasodilator effects. Physical exercises focused on increasing range of motion and decreasing irreducible flexion at the knee level, increasing ankle stability, and preventing complications at the backbone and hip levels.

We tried to increase the amplitude of flexion by passive mobilization of the patella (transversal and longitudinal) followed by actual knee mobilization (the method of stretching the knee under progressive weights). Also, we did active movements: from dorsal decubitus-leg flexions and extensions, ventral decubitus-leg flexion, and extensions (with a small pillow under the knee), sitting-flexions and extensions, with weight on the knee, and in the end, gesture exercises such as walking, going up and down, stepping over, bending over. In the attempt to reduce the knee flexion (determined by posterior capsular retraction, hamstring retraction, as well as the fibrous organization of the lateral capsular-ligamentous apparatus), we used sandbags with progressive weights on the knees (1/3 of the inferior leg being supported on a pillow) and active extension movements.

Treatment also included regaining ankle stability through muscle toning and sensory-motor coordination. We used physical exercises for toning the sural triceps, anterior tibial muscles, extensor digitorum muscles, peroneal muscles, tibial posterior muscles, and flexor hallucis longus muscle. The exercise included dorsiflexion of the foot with resistance on the antero-internal edge of the foot, walking on heels, lifting the foot up on the tip, leaving it (incomplete) on the heel, and returning to the tip. Sensory-motor coordination was promoted by walking on

various routes: flat, uphill, downhill, lateral, on rough terrain, going forward and backward, walking with the feet on the same line or parallel lines, walking with crossed steps, walking on top, on heels, on the edge of the interior and/ or exterior foot.

Occupational therapy was also performed, including cycling, climbing stairs, and obstacle courses. To prevent complications in the hip and backbone, the patient performed exercises to strengthen the paravertebral muscle.

The patient's condition improved, and he reported a decrease in pain when he was discharged. He presented 5 points on the VAS scale. The improvement in the range of motion was remarkable. Active flexion improved from 30 degrees to 70 degrees, and passive flexion increased from 35 degrees to 85 degrees. The knee flexion was also reduced, from 10 degrees to 5 degrees. The gait pattern was improved. The patient went from using two axillary crutches to one elbow crutch.

## Discussions

The diagnosis of CRPS type I is made based on Budapest criteria that were discussed later, and the pathogenesis is complex and not completely known. Because it is a relatively rare condition it can be overlooked if the physician has few experiences. Physical therapy and Medication use have reduced pain and functional disability related to CRPS.

### Pathogenesis

The pathogenesis and etiology of CRPS are not fully disclosed, but there are multifactorial causes that are involved in the development of this syndrome: dysfunction of the sympathetic nervous system, classic and neurogenic inflammation, central sensitization, brain plasticity and, respectively, glia activation [6]. There are several hypotheses that consider psychological factors to have an important

involvement in the pathophysiology of CRPS [7].

### **Dysfunction of the sympathetic nervous system**

The impaired involvement of the sympathetic nervous system is suggested by abnormal skin coloration, temperature difference between the limbs, and abnormal exocrine secretion. Moreover, this theory is sustained by the observation that chemical or surgical denervation of the sympathetic nervous system relieves pain in patients with CRPS. This fact leads to the conclusion that there is an increased rate of efferent sympathetic nerve impulses to the affected extremity [8,9]. However, several studies have shown that, in fact, the rate of efferent sympathetic impulses at the affected extremity is rather decreased than increased. Low levels of noradrenaline and neuropeptide Y were found in plasma of the affected area compared to the levels found in the healthy extremity. The unusual interaction between the sympathetic nervous system and sensory nerves is explained by the fact that sympathetic denervation increases the sensitivity of blood vessels to catecholamines [10]. Drummond et al. demonstrated that in the skin of the affected CRPS area, the number of alpha 1-adrenoceptors is higher than in unaffected areas [11].

### **Classic and neurogenic inflammation**

In cerebrospinal fluid and locally in the affected area, patients with CRPS have increased levels of TNF-alpha, IL-6, IL-1beta, and IL-2 and, respectively, reduced levels of IL-4 and IL-10 [12,13]. TNF-alpha induces pain and is upregulated after bone fracture and cast immobilization, while IL-4 and IL-10 are analgesic cytokines [14,15]. Higher levels of proinflammatory cytokines are a physiological response after trauma.

Neuropeptide called P substance is also found in excess in the affected area of the affected limb. This neurotransmitter is considered a cardinal mediator in neurogenic inflammation and has a direct vasodilating effect through its binding to endothelial cells and an indirect vasodilating effect because it binds to mast cells and promotes the histamine release. The signs and symptoms that are characteristic to neurogenic inflammation are edema, redness and warming of the affected area.

### **Central sensitization**

Nociceptive neurons increase their excitability due to the intense activity of afferents terminations from the damaged tissue, this phenomenon being called central sensitization. The substances involved in this process are neuropeptides like P substance and bradykinin and amino acids like glutamate. An important role is played by N-methyl D-aspartate (NMDA) receptors from the spinal column [16]. In the end, central sensitization is expressed by an exaggerated response to the nociceptive stimuli (hyperalgesia) and pain response to non-painful stimuli like light touch (allodynia) [17].

### **Brain plasticity**

A cortical reorganization with a reduction in the somatosensory cortex of the affected limb was observed in patients with CRPS [18]. These alterations are reversible and, after successful treatment and rehabilitation of CRPS, turn to normal [16].

### **Glia activation**

The discovery that spinal cord glia (microglia and astrocytes) has a major impact on pain modulation has changed the view about chronic pain. The sensation of pain is modulated in the spinal cord dorsal horns where it can be suppressed or amplified. This fact has a major implication in using different



types of drugs to release pain. Glia can synthesize and release a large range of neurotransmitters and, also has receptors that are responding to neurological stimulation [20]. TNF-alpha and IL-6, cytokines that are abundant in CRPS, create exacerbation of pain due to their action on the spinal cord.

### **Psychological factors**

Psychological distress is in most cases associated with increased catecholaminergic activity and, as a result, certain personality traits are prone to be more susceptible to the development of CRPS. It is known that depression is associated with increased pain sensation in patients with CRPS [18].

### **Epidemiology**

The incidence of this syndrome is higher in the age interval of 61-70 years and women are more susceptible than men to develop CRPS after an injury [19]. Fractures, sprains, crush injuries and surgery are conditions that are more likely to be associated with the development of CRPS.

### **Clinical symptoms**

The most common symptoms of CRPS include pain and tenderness, autonomic symptoms, trophic changes, and motor disorders. In most cases, the symptoms appear 6 weeks after the traumatic event and one limb is affected, but there are special patients who are resistant to treatment and can develop multi-limb CRPS [20,21].

Pain is usually characterized by a burning sensation located in the deep structures (bones and muscles). It can also be described as a constriction or stabbing sensation that is exacerbated by movement. Other important characteristics of pain are allodynia and hyperalgesia.

Autonomic and trophic changes in the affected area are also present. Pallor, hyperemia, and temperature changes can be

observed depending on the stage of the syndrome. Patients may also have different hair and nails growth compared with the contralateral limb. According to some studies, 70% of patients have edema at the time of their clinical examination [22].

The range of motion is limited in approximately two-thirds of patients. The limitations of the movements are based on reduced muscle stretch, but also on the presence of edema, pain, and contractures [23].

### **Diagnostic criteria for CRPS**

For the first time, the diagnosis of CRPS was based on the criteria of the International Association for the Study of Pain (IASP), but, in 2003, due to the low specificity leading to overdiagnosis, the Budapest criteria were proposed to have higher specificity and equal sensitivity [24]. They include the presence of incessant pain, disproportionate to the initial traumatic event and appear after a period, most commonly at 6 weeks. The patients should have at least one symptom of three of the following categories: sensory - hyperesthesia or allodynia; vasomotor - temperature or color asymmetry; sudomotor or edema - changes in sweating or asymmetry, edema; motor or trophic - decrease in range of motion, motor dysfunction (weakness, tremor, dystonia), trophic changes (skin, hair, nails). The patient must associate a sign with at least two of following at the time of evaluation: sensory - evidence of hyperalgesia or allodynia; vasomotor - evidence of temperature asymmetry, skin COPD case report color changes, or asymmetry; sudomotor or edema - evidence of edema, sweating changes, or asymmetry; sudomotor or edema - evidence of edema, sweating changes, or asymmetry; sudomotor or edema - evidence of edema, sweating changes, or asymmetry; motor or trophic - evidence of decreased range of motion, motor dysfunction

(weakness, tremor, dystonia), trophic changes (hair, nails, skin). It is considered that there is no other diagnosis to explain the signs and symptoms of CRPS [25,26]. Besides clinical diagnosis criteria, physicians can use plain X-rays for diagnosis, which show patchy osteoporosis, but the sensitivity of this investigation is low, less than 30% of patients developing this sign. Imaging by magnetic resonance can be used for differential diagnosis, but not for diagnosis [27].

### **Principles of therapy**

Due to the complex pathogenesis and the multitude of affected structures, the management of CRPS should include a multidisciplinary approach. The goal of therapy is to reduce pain with the minimum use of analgesic drugs, restore function in the affected limb, and offer the patient the quality of life that he had before the traumatic event [28]. To accomplish these results, the treatment includes, besides drugs, other therapies like patient education, physical therapy, and psychological assessment.

Physical therapy is the first line of treatment for CRPS, and the participation of the patient in rehabilitation sessions should be supported by an explanation of the disorder.

Medication treatment for CRPS includes multiple drugs, but their efficacy is limited. Medications can include calcitonin, bisphosphonates, corticosteroids, intravenous immunoglobulin, antihypertensive, alpha-adrenergic antagonists, and pain relief medications. In cases with refractive pain, administration of anticonvulsants or even antidepressants may be necessary (gabapentin, carbamazepine) [21].

### **Conclusions**

In conclusion, CRPS is a complex condition that has multiple causes and is characterized especially by long-lasting pain, accompanied

by sensory, motor, sudomotor, and trophic changes. These can be overlooked especially if the physician has little experience. Often, anti-inflammatory, and analgesic drugs are not enough, and physical therapy, which is the gold standard for this disease, must be added. The success of the therapy is conditioned by a multidisciplinary, long-term approach and can restore the functionality of the affected limb, as well as the reduction of pain.

### **Conflict of Interest statement**

The authors declare that they have no conflicts of interest regarding the publication of this manuscript.

### **Informed Consent and Human and Animal Rights statement**

Informed consent has been obtained from all individuals included in this study.

### **Authorization for the use of human subjects**

Ethical approval: The research related to human use complies with all the relevant national regulations, institutional policies, is in accordance with the tenets of the Helsinki Declaration, and has been approved by the review board of Department of Rehabilitation and Physical Medicine, National Institute of Rehabilitation and Physical Medicine and Balneoclimatology, Bucharest, Romania.

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### **Author Contributions**

All authors have contributed equally.

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