Case Report

Complex Regional Pain Syndrome with Bilateral Lower Limb Involvement in an Adult

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ABSTRACT

Background: Complex regional pain syndrome [CRPS] has been reported and studied in adults. However, it is a less acknowledged as a disorder in bilateral lower limb. We present a rare case of CRPS in an adult having bilateral lower limb involvement along with its physiotherapy management. This case report gives an insight about early diagnosis, treatment in the form of aggressive physiotherapy, and improvement that can be achieved in a CRPS case.  
Key Words: CRPS, reflex sympathetic dystrophy, pain, physiotherapy, adolescent

INTRODUCTION

In recent years there has been an increase in the reporting of complex regional pain syndrome [CRPS] in children and adolescents. The term “reflex neurovascular dystrophy” [RND] is commonly used in these populations. (1) Complex regional pain syndrome, formerly known as “reflex sympathetic dystrophy”, refers to debilitating neuropathic pain disorder. The pathophysiology of CRPS is not fully understood; however, it is believed to be a systemic disease involving both the central and peripheral nervous systems. (2) If left untreated, the symptoms can become chronic, spread to other parts of the body, and persist for years. The earlier it is recognized and treated, the better the prognosis. Hence it becomes essential to diagnose and manage these cases early to reduce patients’ morbidity.

Complex regional pain syndrome can present with a wide range of symptoms, but the hallmark is pain out of proportion to the inciting event. It usually occurs in the hands or feet, which are rich in nerve innervations, but can present anywhere in the body. The initial injury may be a sprain, dislocation, fracture, laceration, stroke, myocardial infarction, surgery, or it may even follow an intramuscular injection. (3) Pain is generally described as constant, burning, aching, and/or throbbing. (4) Symptoms can consist of allodynia [pain evoked by a mechanical or thermal stimulus that usually does not cause pain, such as light touch], hyperalgesia [exaggerated response to a normally nonpainful stimulus], and hyperpathia [abnormal painful reaction to a stimulus]. The pain can start within hours to weeks of the inciting event and lasts well beyond the expected time to recover. (5) It
can be exacerbated by immobilization, as with a splint or cast. (6) Thus early mobilization should be instituted in the form of early physiotherapy. It has been concluded by most specialists that the earlier treatment is instituted, the better the outcome. The main goals of therapy are pain relief and restoration of function. In adults, aggressive pharmacologic intervention has been the standard. Additional therapies in adults include transcutaneous electrical nerve stimulation, physical and occupational therapy, cognitive-behavioral therapy, psychotherapy, spinal cord stimulation, sympathetic nerve block, and sympathectomy. (4)

CASE REPORT
A forty-five year old male was admitted with an eight-week history of both lower limb pain following bilateral tibia fracture treated with internal fixator 3 months back. Though the internal fixator was put to immobilize the fracture site, the patient was put in bilateral below knee slab for 2 months after the surgery. The patient initially presented to the orthopedic surgeon. The subsequent radiological examination revealed callus formation at the site of fracture & no sign of infection. The slab was removed & medications for pain relief were started. Symptoms failed to resolve with conservative management resulting in hospital admission. On initial assessment the patient reported severe, throbbing, and burning pain in both the lower limbs, particularly worse with touch, exposure to wind, and movement. Active and passive range of motion of the hip, knee, and ankle joints were severely restricted bilaterally with evidence of edematous, shiny skin, and warmth over both lower limbs. Hyperalgesia and allodynia was noted. The patient was confined to bed and all the activities of daily living [ADL] were restricted. The Barthel Index for ADL suggested severe restriction.

A provisional diagnosis of CRPS was made based on IASP criteria. (2) Therapeutic regimen of gabapentin and active physiotherapy was started. Physiotherapy session consisted of desensitization followed by motor retraining. Each session was given thrice a day approximately for one hour over a period of 2 weeks, pain was reduced in the intensity, range of motion was improved. Psychiatric reference to rule out commonly found psychological issues in such cases was given; no issue was found on examination. Physiotherapy treatment continued to have a session of relaxation with positive imagery, deep breathing exercises, graded activity pacing, and pleasurable activities to make the patient more actively involved in the treatment. The intensity of therapy was gentle and below the pain threshold, as to not exacerbate the pain, since any painful stimulus may worsen the symptoms. The treatment regimen was continued for two more weeks along with posture correction, lower limb weight bearing exercises, and gait training. At the end of 4 weeks, there was complete pain relief and achievement of total pain free active and passive range of motion in both lower limbs. Barthel Index suggested complete independence, and thus patient was discharged with a home program. There were no signs or symptoms of relapse on the follow ups done at six months and a year.

DISCUSSION
Complex regional pain syndrome causes pathophysiological changes in all parts of the nervous system. This syndrome can present with a wide range of symptoms, but the hallmark is pain out of proportion to the inciting event. A classic feature of CRPS is its tendency to spread proximally to involve the entire extremity or ipsilateral quadrant of the body, the contralateral extremity, or even to other body parts. With early intervention the spread of the disease...
can be controlled. Pain relief is the patient’s primary concern, but improving mobility, function, and a return to daily activities should be our goal as well. Physiotherapy tends to be the primary mode of treatment, with the vast majority recovering and managing relapses with this treatment modality. In a prospective study, Sherry et al. (7) evaluated the effectiveness of short-term, intense physical therapy with complete resolution of all symptoms and full functional recovery. It is thought that exercise works by inhibiting the sympathetic stimulation, increasing circulation, and releasing endorphins. (8) The pain in CRPS is associated with allodynia which can be treated using desensitization. Desensitization involves application of "unpleasant" stimuli to the hypersensitive area. The body is routinely exposed to these stimuli and does not elicit a painful response when presented to non-affected areas. Stimuli may consist of different textures or fabrics, light or deep pressure, vibration, heat or cold. In the present case, cold in the form of ice was used, as it also helps in reducing pain and swelling. The goal of desensitization is to inhibit or interrupt the body's interpretation of routine stimuli as painful. It does not assure that these stimuli will become pleasant or enjoyable, but that they will no longer provoke an extreme pain response. It is of utmost importance that the patient's pain is accepted, not to intensify the psychological sequelae of chronic pain.

Depression and anxiety are common results of chronic pain and must be addressed. Some studies report that psychosocial factors or certain personality traits are frequently found in adolescents with CRPS, suggesting that it may be a stress-related disorder. (9) Hence psychiatric reference has to be sorted out and a psychological treatment as a part of pain management has to be integrated.

Cognitive behavioral therapy, along with exercises, helps in increasing patients’ activity levels through activity pacing and scheduling pleasurable activities. Lee et al. (10) evaluated the combination of physical therapy and cognitive behavioral interventions in a prospective, randomized trial, and found significant improvements for the majority of children. (10) Thus cognitive behavioral therapy in addition to physiotherapy may be helpful in encouraging patients to remain active and, therefore, decrease the potential for the deconditioning associated with activity avoidance.

CONCLUSION

In conclusion, we describe a rare case of CRPS of bilateral lower extremity in order to remind of this entity. The diagnosis and treatment of CRPS undoubtedly depends on the awareness of the condition.

REFERENCES


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