

## Case Report

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# CRPS after unicompartmental knee arthroplasty: A case report

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### Abstract

A 59-year-old female presented with pain, warmth, erythema, and swelling over the medial aspect of her left knee that began after medial unicompartmental arthroplasty. Exam fulfilled the Budapest criteria for diagnosis of Complex Regional Pain Syndrome (CRPS) type 2. Topical compound cream and physical therapy were prescribed, and symptoms were improved at follow up visit. CRPS localized to the knee after unicompartmental arthroplasty has not been reported in the literature to date, therefore we report a case of CRPS after unicompartmental knee arthroplasty.

**Keywords:** CRPS; Knee; Arthroplasty; Therapy.

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### Introduction

Complex Regional Pain Syndrome (CRPS) is a poorly understood, debilitating, and painful disorder characterized by dysfunction of the autonomic nervous system. Clinical symptoms may include vasomotor instability, sudomotor symptoms, edema and neuropathic pain that is disproportionate to any inciting event. Often a precipitating event such as trauma, fracture, or surgery has been linked to CRPS. Diagnosis can be made with the Budapest criteria, which has been deemed superior to previous stringent criteria used for diagnosis [1].

Complex regional pain syndrome is more common in women, smokers, older individuals, individuals with reduced bone mineralization, and those with premorbid mood disorders [2]. The definitive etiology is unknown, but is likely multifactorial. Speculation regarding the pathophysiology includes, but is not limited to, microvascular changes at the deep tissue level as a result of inflammatory cytokines and subsequent metabolic change [3], peripheral and central sensitization, neuroplasticity [4] and increased sympathetic tone [5].

Patients often have prolonged post-operative edema and skin temperature changes that are a part of a normal healing course rather than pathology. A recent prospective study by

Kosy et al. discussed that CRPS following Total Knee Arthroplasty (TKA) is likely less than 1% based on the newer and more stringent Budapest criteria [6]. There is a case report of foot and ankle CRPS after TKA [7], and all known studies reporting CRPS after TKA to the authors knowledge do not describe the specific location of symptoms. There are no documented cases of CRPS limited to the knee after a unicompartmental knee arthroplasty, especially symptoms limited to the laterality of the unicompartmental arthroplasty. In addition, specific therapies for treatment of CRPS in this subset of patients has not been reported. This manuscript adheres to the applicable EQUATOR guidelines. Written Health Insurance Portability and Accountability Act (HIPAA) consent has been obtained from the patient for publication of this case report.

### Case description

A 59-year-old female with a remote history of erythromelalgia presented to the outpatient clinic with a 7-month history of pain, warmth, erythema, and intermittent swelling over the medial aspect of her left knee that began after medial unicompartmental knee arthroplasty. Prior to the knee arthroplasty, pain was reported to be sharp, localized to the medial joint line, and associated with catching and locking. Post-operative diagnostic work-up included labs and imaging to rule out sep-

tic arthritis, inflammatory arthritis, and hardware failure. Basic metabolic panel, CBC, CRP, ESR and joint fluid analysis were within normal limits. Post-operative imaging of the knee included plain radiographs at 3 months post-op, which demonstrated intact hardware but with signs of joint effusion and soft tissue swelling. Upon presentation to our clinic, she reported a pain severity score of 6/10 on the Visual Analogue Scale (VAS), which was characterized as sharp and throbbing. She reported that symptoms had become progressively worse since surgery. Exam demonstrated erythema, warmth, swelling, and hyperesthesia along the medial knee, extending approximately 4 centimeters proximal and distal to the knee incision, fulfilling the Budapest criteria for diagnosis of CRPS (Table 1). Range of motion was intact to flexion and extension, despite subjective symptoms of stiffness. Strength and reflexes in the lower extremities were preserved. Gait was normal. Diagnosis was challenging due to her history of erythromelalgia, but rheumatology deemed that symptoms were not due to an erythromelalgia flare based on focal symptoms rather than presentation of symptoms bilaterally and diffusely in the lower extremities, and history without precipitation of symptoms with heat. Orthopedic surgery felt this was less likely an allergy to the surgical hardware, and hardware would need to be removed in order to rule out an allergic etiology. She was prescribed topical compound cream containing ketamine HCl 10%, baclofen 2%, amitriptyline HCl 2%, gabapentin 6%, diclofenac sodium 3%, and lidocaine 5%, 1-2 grams applied to the medial knee 4 times daily, along with physical therapy 1-2 times weekly for 10 weeks for strengthening, desensitization and mirror therapy. Symptoms were improved at follow up visit 6 weeks later, and she elected to continue conservative therapy, noting that the substantial improvement was due to both topical compound cream, desensitization with manual therapy and cold exposure, as well as mirror therapy. She reported her pain to be a 3/10 in severity based on the VAS at that time. Our patient was seen 8 weeks after initial follow up visit, then at 12 weeks, and reported continued improvement in symptoms. At the most recent follow-up visit, she reported that she was using topical compound only as needed due to the substantial improvement of symptoms. She completed 14 sessions of physical therapy, with resulting decrease in pain, erythema, and swelling, decreased stiffness, and improved activity tolerance. Informed consent for publication of this case report was obtained from the patient. This case report conforms to all CARE guidelines and reports the required information accordingly (see Supplementary Checklist).

### Discussion

Complex regional pain syndrome can be difficult to manage, with treatment algorithm geared towards more conservative therapies initially. There are multiple physical therapy approaches to treating CRPS.

Movement Representation Techniques (MRT) includes mirror therapy and graded motor imagery. These therapies involve observing or imagining normal and pain free movements, typically performed simultaneously with sensory stimulation and active motion. The aim of MRT is to facilitate pain free movements of a painful limb and desensitization. A meta-analysis found that MRT were effective in reducing pain and disability in persons with chronic pain syndromes [8]. Graded exposure involves individually tailored practice tasks in which the patient

**Table 1:** Budapest Criteria (2007).

Continuing pain, which is disproportionate to any inciting event
Must report at least one symptom in <i>three of the four</i> following categories:
<ul style="list-style-type: none"> <li>• Sensory: reports of hyperesthesia and/or allodynia</li> <li>• Vasomotor: reports of temperature asymmetry and/or skin color changes and/or skin color asymmetry</li> <li>• Sudomotor/Edema: reports of edema and/or sweating changes and/or sweating asymmetry</li> <li>• Motor/Trophic: reports of decreased range of motion and/or motor dysfunction (weakness, tremor, dystonia) and/or trophic changes (hair, nail, skin)</li> </ul>
Must display at least one sign at time of evaluation in <i>two or more</i> of the following categories:
<ul style="list-style-type: none"> <li>• Sensory: evidence of hyperalgesia (to pinprick) and/or allodynia (to light touch and/or temperature sensation and/or deep somatic pressure and/or joint movement)</li> <li>• Vasomotor: evidence of temperature asymmetry (&gt;1°C) and/or skin color changes and/or asymmetry</li> <li>• Sudomotor/Edema: evidence of edema and/or sweating changes and/or sweating asymmetry</li> <li>• Motor/Trophic: evidence of decreased range of motion and/or motor dysfunction (weakness, tremor, dystonia) and/or trophic changes (hair, nail, skin)</li> </ul>
No other diagnosis better explains the signs and symptoms.

Harden RN, et al. "Proposed new diagnostic criteria for complex regional pain syndrome." *Pain Med.* 2007; 8: 326-331.

agrees to engage in fearful activities, movements or situations that they have been avoiding as much as possible until anxiety levels decrease. Graded exposure therapy in chronic CRPS has been found to be successful in decreasing levels of self-reported pain-related fear, pain intensity, disability, and self-reported peripheral abnormalities [9]. Pain exposure therapy is another approach that is geared towards functional restoration. It involves direct exposure to painful stimuli, with the focus placed on ignoring pain in order to break the fear-avoidance pain pattern. Patients are explained that the pain is a false warning sign due to functional disturbances in the affected limb. Patients also learn how to decrease skin sensitivity for touch and pressure by performing self-massage, and use of the affected extremity with progressive-loading exercises [10]. Our patient was given MRT, which resulted in improvement of her pain and activity tolerance.

Topical compound creams have the potential to alleviate symptoms, since the delivery of therapeutic agents to the subcutaneous tissues may reverse or lessen pathological mechanisms. Various components can be used to target nociceptive, neuropathic, or mixed pain. A case series found that topical cream consisting of ketamine, pentoxifylline, clonidine, and Dimethyl Sulfoxide (DMSO) provided both pain and symptom relief in 69% of patients [11]. The proposed mechanism for relief of neuropathic pain is due to the effect of ketamine on the NMDA receptor. Ketamine lowers the threshold for nerve transduction, reduces the effects of substance P, and targets the opioid, sodium, and potassium channels, thus reducing allodynia and hyperalgesia. Pentoxifylline and clonidine are thought to modulate the associated microvascular dysfunction associated with CRPS, and DMSO acts as a free radical scavenger theorized to decrease inflammation. Other substances that may be included in topical compound creams used for pain include li-

docaine, prilocaine, gabapentin, capsaicin, diclofenac, doxepin, and baclofen.

Based on a Cochrane systematic review, there is low-quality evidence for the effectiveness of many treatment modalities such as the use of bisphosphonates, calcitonin, intravenous ketamine, graded motor imagery, physical or occupational therapy [12]. A recent European pain federation task force was created to establish diagnosis and management standards, and pharmacologic treatments for CRPS are extrapolated from medications used for other neuropathic pain states, since high-quality studies in CRPS are not available. Treatment with bisphosphonates and/or steroids may be considered, although evidence for or against their efficacy and safety is lacking. Patients who have not responded to physical therapy and medications may be considered for invasive neuromodulation such as spinal cord stimulation of dorsal root ganglion stimulation [13]. Which we may have tried with our patient if more conservative therapies had not offered her adequate pain relief.

The differential diagnosis of CRPS in the acute phase includes erythema migrans, early inflammatory lesions of acrodermatitis chronica atrophicans, eosinophilic fasciitis, erythromelalgia and an acute contact dermatitis [14]. However, our patient was seen initially in the chronic phase, approximately 7 months since onset of symptoms. Primary erythromelalgia causes recurrent episodes of painful bilateral neuropathy and edema, typically affecting the limbs symmetrically and occurs commonly in the soles of the feet and hands. While the exact pathophysiology is yet to be fully elucidated, it is thought that there are neural alterations such as altered electric excitability, adrenergic impairment and a microvascular dysfunction. Erythromelalgia is known to have triggering events such as heat exposure or pressure, prolonged standing and physical activity, stress, alcohol and spicy food ingestion [15]. However, our patient reported no exacerbation of symptoms with any known factors.

Our patient reported that she was overall satisfied with her treatment, and felt that both therapy and topical compound cream improved her pain and allowed for improved activity tolerance. A strength of our case report is that our patient was assessed at multiple follow up visits, so we were able to adequately assess her improvement. A limitation is that we did not evaluate the patient in clinic until 7 months after onset of symptoms. Early recognition and treatment of this clinical diagnosis is crucial.

### Conclusion

In conclusion, we report a case of CRPS after unicompartmental knee arthroplasty which has not been documented in the literature to date.

### Declarations

**Conflicts of interest:** None.

**Financial gains:** None.

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