

# Dramatic effect in passive ROM exercise under sedation in a patient with intractable complex regional pain syndrome (type I)

## A case report

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### Abstract

**Rationale:** Complex regional pain syndrome (CRPS) usually occurs after trauma and surgery but can also occur spontaneously. There are various known pathophysiology and treatment protocols for CRPS. However, there is no established treatment guideline. Although physical therapy is known as the first line treatment for CRPS, performing physical therapy is difficult due to severe pain. This case reports the first case with positive effect of physical therapy under sedation that allowed early physical therapy.

**Patient concerns:** A 19-year old female had her right elbow area slightly hit by someone else, after which it began to swell and become more painful. Active and passive range of motion (ROM) of the right upper extremity gradually decreased through 3 months, and pain and edema worsened.

**Diagnoses:** She had allodynia, nonpitting edema, temperature asymmetry, and trophic change in fingernails in the right upper extremity, which met the diagnostic criteria for CRPS. In the 3-phase bone scan, which is a specific tool to diagnose CRPS, there was trace uptake increase in the right elbow and wrist, indicating possibility of CRPS.

**Interventions:** Despite conventional treatments such as pharmacologic and interventional therapies, neither pain nor edema subsided. Pain was so severe that it was impossible to apply physical therapy. Therefore, the patient underwent passive ROM exercise in the right upper extremity under sedation for 30 minutes, which was relatively easier due to decreased pain. After 2 days of passive ROM exercise under sedation, the patient was able to receive passive ROM exercise twice daily without sedation for 18 days.

**Outcomes:** After 20 days of passive ROM exercise, including 2 days of passive ROM exercise under sedation, the circumference of her right hand decreased by 5 cm, wrist 2 cm, and elbow 6 cm compared to the initial measurement. Numeric pain rating scale improved from 9 to 3, and her manual muscle test marked fair plus from trace, Jebsen–Taylor hand function test score 43 from 0.

**Lessons:** This case suggests that passive ROM exercise under sedation may be a successful alternative as a treatment when exercise, currently known as a treatment to CRPS, is impossible.

**Abbreviations:** CRPS = complex regional pain syndrome, JTHFT = Jebsen–Taylor hand function test, MAM = medicine assisted manipulation, MMT = manual muscle test, MUA = manipulation under general anesthesia or conscious/deep sedation, NMDA = N-methyl-D-aspartate, NRS = numeric pain rating scale, ROM = range of motion.

**Keywords:** CRPS, passive ROM, physical therapy, sedation

## 1. Introduction

Complex regional pain syndrome (CRPS) usually occurs after trauma and surgery, but can also occur spontaneously. The

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estimated incidence rate of CRPS is 26.2 per 100,000 people annually. It occurs 3 times more often in females than in males.<sup>[1]</sup> There are 2 types of CRPS. CRPS type I (CRPS-I) is not associated with nerve injury and trauma and CRPS type 2 involves objective nerve injury, usually caused by trauma. CRPS-I has recently been known as a chronic neuropathic pain syndrome generally after tissue trauma.<sup>[2]</sup> The symptoms of CRPS include edema and altered sweating, skin color and temperature which consequently lead to changes in skin, hair, and nails. If symptoms persist or worsen, weakness, decreased range of motion, and tremor would affect activities of daily living.<sup>[1]</sup>

There are various known pathophysiology of CRPS; such as central and peripheral sensitization, altered functions of sympathetic nervous system, and psychologic factors.<sup>[2]</sup> As much as there are multiple mechanisms contributing to CRPS, there are various therapeutic approaches to treat CRPS. However, its management is not necessarily as easy since the response to treatment varies from patient to patient. Common treatments are pharmacotherapies, physical therapy, and interventional therapies. Pharmacotherapies include nonsteroidal anti-inflammatory drugs, corticosteroid, cation channel blockers (eg, gabapentin), anticonvulsants, antidepressants, N-methyl-D-aspartate (NMDA) receptor antagonists

(eg, MK-801, ketamine, amantadine, and dextromethorphan), opioids and IVIG. Interventional therapies include stellate ganglion block, lumbar sympathetic block, brachial plexus block, epidural analgesic infusion, spinal cord stimulation, and peripheral nerve stimulation.<sup>[3]</sup> In the following case, a patient with CRPS-I, who had been tried various treatments which were ineffective, underwent passive range of motion (ROM) exercise under sedation.

## 2. Case presentation

A 19-year old female without past medical history had her right elbow area slightly hit by someone else in March 2016, after which it began to swell and become more painful. Active and passive ROM of shoulder, elbow, and wrist gradually decreased through 3 months, and pain and edema worsened. Her initial manual muscle test (MMT) was normal but deteriorated as pain and edema worsened. Her quality of life deteriorated accordingly. Trimming right fingernails was impossible due to pain and she had to use wheelchair to move as pain aggravated whenever her right arm wobbled. She gained weight from 73 to 93.8kg. She was preparing for entrance examination at an art college, but could not draw at all. The patient visited the department of vascular surgery, where they ruled out thrombosis through color-doppler ultrasound. For further evaluation, she was referred to the outpatient clinic of department of rehabilitation.

We performed laboratory test including rheumatic factors (CBC, ESR, CRP, BUN/Cr, RF, and Anti CCP Ab) since she had

family history of rheumatoid arthritis, but the results were within normal limits, and no abnormal findings related to clinical symptoms or findings in shoulder magnetic resonance imaging. In 3-phase bone scan which is a specific tool to diagnose CRPS of the upper limb, with a sensitivity of 69% and a specificity of 75%, there was trace uptake increase in the right elbow and wrist, indicating possibility of CRPS.<sup>[4]</sup> However, nerve conduction study to confirm nerve damage was impossible due to severe pain.

On physical examination, we observed allodynia and hyperesthesia, vasomotor symptoms like skin color asymmetry, motor/trophic symptoms like edema, decreased active and passive ROM, tremor, and trophic changes in fingernails (Fig. 1). The thermography test revealed temperature of 28.7° and 29.8° on right and left hand, respectively, showing a temperature difference of more than 1° between the 2 arms. The circumference of her right hand (measured around the hand at the fullest part, or adding the length of her palm and the arch of her hand) was 25 cm, wrist 19 cm, and elbow 35 cm. The circumference of her left hand, or normal side, was 20.5 cm, wrist 18 cm, and elbow 31 cm. Her MMT marked trace on shoulder, elbow, wrist, and hand joints, and Jebsen–Taylor hand function test (JTHFT) score was 0; her right hand was practically nonfunctional; her right hand was practically nonfunctional (Table 1). She was diagnosed CRPS-I according to the IASP criteria and recently proposed clinical criteria for CRPS.<sup>[5,6]</sup>

She took antiepileptic drugs, NSAIDs and antidepressant drugs for 1 month through outpatient clinic but there was no effect. Additionally, she received stellate ganglion block with 5cc of



**Figure 1.** On admission, hand and arm before passive ROM exercise under sedation. ROM=range of motion.

**Table 1**  
**Outcome measurement during various time points in patient's hospital course.**

		Left	Right	Conventional	After 5Ts passive ROM exercise	Discharge
		normal side	admission	treatment (HD 6)	(2Ts under sedation) (HD 11)	(HD 26)
NRS		0	9	9	6	3
Circumference	Elbow	31 cm	35 cm	35 cm	30.8 cm	30 cm
	Wrist	18 cm	19 cm	19 cm	18 cm	17 cm
	Hand	20.5 cm	25 cm	25 cm	21 cm	19 cm
JTHFT		95	0	0	NT	43
MMT	Elbow Flexion	Normal	Trace	Trace	Fair	Fair +
	Wrist Extension	Normal	Trace	Trace	Fair-	Fair +
	Finger Flexion	Normal	Trace	Trace	Fair	Fair +

HD = hospital day, JTHFT = Jebsen-Taylor hand function test, MMT = manual muscle test, NRS = numeric pain rating scale, NT = not tested; ROM = range of motion, T = time.

1.5% lidocaine twice, topical lidocaine cream, oral corticosteroids, oral opioids, intravenous (IV) ketamine, and brachial plexus block for 2 months, which were also ineffective.

She was admitted to the hospital for intensive treatment and started physical therapy along with conventional treatments. However, performing passive ROM was impossible since any light touch on the right upper extremity induced massive pain. Therefore, the patient was given 15 mg IV midazolam 10 minutes before passive ROM exercise in the right upper extremity while monitoring vital signs. She was sedated during 30 minutes of passive ROM exercise, which was relatively easier due to reduced pain. Although pain did not completely disappear in spite of sedation, when the patient woke up from sedation, she could not remember the pain during exercise and thus had far less psychological pressure in the second sedation session on the

next day. In the second trial, the patient received one more passive ROM exercise under sedation for 30 minutes. From the third trial, now without sedation, there was no difficulty in passive ROM exercise. She received 3 more passive ROM exercise without sedation from then. The circumference of her right hand reduced by 4 cm, wrist 1 cm, and elbow 4.2 cm compared to that on admission (Table 1). There was no adverse reaction of using midazolam.

In the first 6 days among the 26 days of admission, the patient received conventional treatment only, and after 2 days of passive ROM exercise with sedation, the patient was able to receive passive ROM exercise and pneumatic compression twice daily without sedation for 18 days. On discharge, the circumference of her right hand decreased by 5 cm, wrist 2 cm, and elbow 6 cm compared to the initial measurement (Fig. 2). There was no



**Figure 2.** Hand and forearm on discharge. After multiple passive ROM exercise including 2 times of sedation session. ROM = range of motion.

circumference difference between the bilateral hands. In fact, the circumference of right elbow was 1.5 cm shorter than that of the left. Numeric pain rating scale (NRS) improved from 9 to 3, and her MMT marked fair plus from trace, JTHFT score 43 from 0 (Table 1). When the performance status reached near normal she was discharged to return to school. After discharge, she received passive ROM exercise and pneumatic compression twice a week, while continuously taking gabapentin and antidepressants. After 3 months, the circumference far shortened and the patient's pain level was between 1 and 3 on NRS, which was tolerable enough for daily activities. The patient entered college of art as it became possible to draw. For 18 months after discharge she lost weight from 93.8 kg to 78 kg, thus the circumference difference of her right elbow (by 5 cm compared to 15 months before) presumably attributed to body weight change rather than pure decrease in edema.

### 3. Discussion

A 19-year-old female, whose symptoms met IASP criteria, had allodynia, non-pitting edema, temperature asymmetry, and trophic change in fingernails in the right upper extremity which also met the new diagnostic criteria for CRPS.<sup>[5,6]</sup> Pain and edema persisted and the hand function deteriorated; MMT marked trace and JTHFT score was 0. Conventional treatments such as pharmacologic, physical and interventional therapies did not help. Due to the severe intractability of the patient represented in the case, ketamine, antagonist of NMDA receptors, was thought to be a good treatment alternative.<sup>[7]</sup> The patient was given 50 mg IV ketamine over a period of 30 minutes, but pain exacerbated regardless.

In severe cases of CRPS, it is necessary to promote improved circulation to the area as well as facilitate more normal tissue length/positioning during the rehabilitation process.<sup>[3]</sup> According to the experience done by Mayo clinic, physical therapy is the first line treatment for CRPS.<sup>[9]</sup> In childhood CRPS-I, aerobic exercise, functionally directed exercise, hydrotherapy, desensitization, occupation therapy including gentle active movement, and manual edema mobilization are known to be effective.<sup>[3,8]</sup> For this patient, unable to undergo any physical therapy, we supposed that performing exercise without patient's full consciousness might be an effective approach. More importantly, the fact that the patient could not remember pain during exercise was a great help in the second sedation session.

Since the 1930's, medicine-assisted manipulation has been used for treatment of musculoskeletal disorders such as frozen shoulder and back pain. It has been modified by various methods such as manipulation under general anesthesia or conscious/deep sedation (MUA), manipulation under joint anesthesia, or manipulation under epidural anesthesia/epidural steroid injection, and has been used to treat of musculoskeletal disorders. A number of studies and therapeutic effects of MUA have been

reported.<sup>[10,11]</sup> However, there was no report of physical therapy under sedation for CRPS patients. We believe this is the first reported case of positive effect in physical therapy under sedation for CRPS. It was also notable that as edema relieved and the ROM improved, pain decreased to such an extent that physical therapy could be performed without sedation. Therefore, it is not necessary to worry about adverse reaction caused by sedation drugs, since sedation is needed only for some initial treatments.

In conclusion, in this case, passive ROM exercise under sedation was effective treatment in improving function in patients with intractable CRPS-I.

### 4. Ethical Review

Ethics committee or institutional review board approval was obtained (approval number: KNUH 2018-06-015). And patient signed informed consent for the publication of this case report.

### Author contributions

**Conceptualization:** Hyun-Min Oh, Ae Ryoung Kim.

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