

## Ultra-Rapid Resolution of Concurrent CRPS with Extrapleural Marcaine Following Bilateral Mastectomy: Case Report

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**Received:** May 14, 2021; **Published:** June 28, 2021

### Abstract

Complex regional pain syndrome (CRPS) is a clinically-defined neuropathic pain syndrome which often can become a permanently disabling painful condition. In this case report, bilateral CRPS was caused by the trauma of venipuncture in the dorsum of each hand and confined to the last three fingers of each hand, right more than left. Clinically evident IASP-Budapest criteria-defined features of burning pain, swelling, sweating, discoloration, temperature and trophic changes affecting the skin and hair were evident. Altered sensory and motor functions in these areas was noted, although ulnar nerve integrity was otherwise intact. Despite vigorous efforts at treatment, including multiple medications, sympathetic nerve blocks and physical therapy, CRPS persisted over the course of four months. It was not until a direct, long-lasting, extrapleural infusion of 0.5% plain Marcaine, utilizing the On-Q Pain Buster® pump system, initiated following bilateral mastectomy, that all symptoms and signs of the CRPS immediately resolved. Occurrence of this sudden dramatic therapeutic improvement deserves further consideration and investigation as a potential method of treatment of this potentially disabling disorder.

**Keywords:** *Complex Regional Pain Syndrome (CRPS); Venipuncture; Extrapleural Marcaine; Bilateral Mastectomy*

### Background

Complex regional pain syndrome (CRPS) is a neuropathic pain disorder which frequently follows a traumatic injury, including venipuncture [1,2]. There are specific IASP diagnostic criteria, identified in the medical literature as the Budapest criteria [3], which are utilized to establish the CRPS diagnosis. Resolution of the disorder is quite variable, and it often progresses to a permanent condition [4]. Although many treatment strategies have been employed over the years [5], CRPS has been difficult to effectively treat.

Movement-based treatments have been most successful [6], including mirror-movement therapy [7], but pain intensity, aggravated by activity, frequently limits effective employment of this strategy. In this context, efforts at early diagnosis and prompt initiation of treatment are imperative [8]. However, despite early diagnosis and reasonable efforts at treatment, including medications, sympathetic nerve blocks and movement-based therapy, even when pain has been reduced to a tolerable level, persistence and progression of CRPS symptoms and signs has remained the predominant response. In this context, I provide a case report of a dramatic, ultra-rapid resolution of the symptoms and signs of CRPS occurring four months after its onset.

### Case Report

The patient was, at the time of treatment, a 52-year old right handed woman. She had a four-month history of CRPS involving the last three fingers of each hand (Figure 1). The onset of CRPS in each hand followed separate venipuncture events, one week apart, following intravenous (IV) needle placement during separate medical procedures. The first onset followed IV access in the ulnar aspect of the dorsum of the right hand as part of a lumbar epidural nerve block procedure for an unrelated condition. Over the course of the following week, burning pain, allodynia (painful sensitivity to light touch), flexed posturing of the last three fingers, swelling, discoloration and sweating between the metacarpal and proximal interphalangeal joint, with increased hair growth, pronounced hair follicles and shiny skin appeared (Figure 2A and 2B). Efforts at flexion or abduction of the fingers were also limited because of increased pain.



**Figure 1:** Swelling and discoloration and of the last three fingers of each hand, right worse than left, prior to the bilateral mastectomy and bilateral extrapleural 0.5% Marcaine infusion.



**Figure 2A:** Swelling of right ring finger (on the left side of this photograph), with evident discoloration, shiny skin and increased hair growth evident over the proximal phalanx of the right ring and long fingers (on the right side of this photograph). This was prior to the bilateral mastectomy and extrapleural 0.5% Marcaine infusion.



**Figure 2B:** Swelling of right ring finger, with evident discoloration, shiny skin and increased hair growth evident over the proximal phalanx of the right ring (on the left side of the photograph) and long (on the right side of the photograph) fingers. This was prior to the bilateral mastectomy and bilateral extrapleural 0.5% Marcaine infusion.

The second onset followed IV access in the ulnar aspect of the dorsum of the left hand, as part of a colonoscopy procedure. As with the right hand, within one week of this procedure there was the onset of burning pain, sensitivity to touch (allodynia), swelling, discoloration, shiny skin and sweating of the last three fingers of the left hand in the same distribution as on the right hand. Increased hair growth, with coarse, dark hairs was noted as well, along with pronounced hair follicles (Figure 3).



**Figure 3:** Swelling of left fifth finger, with evident discoloration, shiny skin and increased hair growth evident over the proximal phalanx. This was prior to the bilateral mastectomy and bilateral extrapleural 0.5% Marcaine infusion.

Over the course of the following four month period, initial treatment with a six-day course of an oral 4mg methylprednisolone tablet “Dosepak®” taper (24 mg/day, 20 mg/day, 16 mg/day, 12 mg/day, 8 mg/day and 4 mg/day), followed by symptomatic treatment with gabapentin, at doses as high as 1800 mg per day (300 mg every three to four hours, six per day) was administered. This produced significant reduction of the burning pain and allodynia, which allowed repetitive movement therapy consisting of simultaneous opening and closing the fingers of both hands and “air piano” movements of the fingers of each hand. However, these efforts did not lead to resolution of the symptoms and signs, and CRPS persisted.

During this time period the diagnosis of intraductal breast cancer of the left breast, following a suspicious mammogram, was identified by MRI and subsequently confirmed by ultrasound-guided biopsy. Because there were three distinct, sub-areolar tumors identified within the left breast, a total mastectomy was scheduled. Bilateral, custom-formed Theraplast® hand braces were made prior to surgery and worn into the operating room, to reduce the risk of further traumatizing the hands during the surgical procedure. The braces were not kept on for the entire duration of the three hour procedure. This surgery also included the removal of seven lymph nodes, initiated with a sentinel node above the left breast, progressing toward the left axilla, node by node, until the seventh node was identified as free of malignant cells. Following removal of the left breast and node dissection, a prophylactic total right mastectomy was performed.

Upon completion of the bilateral mastectomy procedure, breast reconstruction was initiated by placement of tissue expanders in a subserratus/subpectoral pocket created and dissected up to the pectoralis minor muscle. This was followed by placement of an Inamed style 133 LV, 300cc, 13 cm expander, which was filled to 150cc bilaterally. Ketamine, 50 mg, had been administered at the initiation of the operation. In addition to bilateral surgical drains, extrapleural catheters attached to an On-Q Pain Buster (#PM025), 300 ml, with plain 0.5% Marcaine® injected (Lot#542588) were employed [9].

Upon return to the recovery room, the symptoms and signs of CRPS in both hands were completely absent, and they have not returned (Figure 4). When questioned regarding CRPS symptoms in either hand, one hour after returning to the recovery room and now being fully awake, they no longer were present. Immediately following surgery, the only analgesic medication offered was oxycodone/APAP 5/325, two tablets by mouth, as needed, at four-hour intervals [9].



**Figure 4:** Complete resolution of CRPS signs, evident in the recovery room on the day of the bilateral mastectomy and documented two weeks after surgery. There no longer is any swelling, discoloration or trophic changes evident in the skin, hair or nails, and sensation and movements returned to normal.

The On-Q Pain Buster pump remained in place until fully dispersed over a three day period. During this subsequent period, a single dose of OxyContin, 10 mg at bedtime, was the only analgesic medication provided. The symptoms and signs of CRPS did not return.

### Discussion

CRPS meeting the Budapest criteria was established and persisted over four months duration at the time of the bilateral mastectomy. Of note, CRPS symptoms and signs in this individual were confined to the distribution of the last three fingers of each hand, which is comparatively restricted in contrast to the more common distribution throughout the entire distal portion of each upper extremity. Also, unlike the typical clinical presentation, open access to the extrapleural space was available during the bilateral mastectomy. This facilitated bilateral extrapleural catheter placement for the ongoing delivery of 0.5% Marcaine® through the On-Q Pain Buster [9]. Access to this modality remained in effect for the first three days of the post-operative period.

This response, to my knowledge, is the first report of “instantaneous” resolution of CRPS, in the face of this medication (0.5% Marcaine), and is a novel observation. Marcaine has previously been effectively utilized in the symptomatic treatment of CRPS, primarily in the context of sympathetic ganglion nerve blocks [10]. However, the present result of complete resolution of symptoms and signs of CRPS deserves further evaluation as a prospective therapeutic approach to treatment of this potentially intractable condition, although the precise mechanism of action presently remains unclear.

Certainly, this case report does not prove or predict universal efficacy, and alternative, less invasive forms of drug delivery are desirable. However, it does raise the issue of a potential strategy to be explored for providing a long-term benefit, especially when delivered early in the course of CRPS. This is because of the unanticipated occurrence of complete spontaneous remission of all symptoms and signs of CRPS. This result may also be related to the relatively confined CRPS distribution within the sensory ulnar innervated fingers of each hand, and/or treatment early in the course of this condition.

Of note, a dose of ketamine, 50mg, had been delivered in the operating room just prior to the mastectomy. This medication has been utilized in the treatment of CRPS, but never with this degree of efficacy and duration of effect following either administration of a single dose or at the concentration dispensed. Other medications provided during the course of the operation included Versed® (midazolam), fentanyl, Robinul® (glycopyrrolate), Decadron® (dexamethasone), Zofran® (ondansetron), Reglan® (metoclopramide), succinylcholine, propofol, rocuronium and Neostigmine/Robinul. However, none of those medications have a history of having been utilized or suggested as ongoing treatment of CRPS.

While this case report raises an important question regarding a potential treatment strategy for this form of CRPS, there are certain questions that deserve further attention:

1. First, was the dose, direct route accessibility, albeit into an extrapleural rather than intrapleural space, and persistent bathing of critical structures by this long-acting local anesthetic a critical factor in the rapid and persistent resolution of CRPS in this patient? [11].
2. Second, was there a critical pharmacologic combination of medications coincidentally rendered in the treatments administered?
3. Third, was the induction and maintenance of general anesthesia a relevant element in the resolution of symptoms and signs of CRPS in this case report?
4. Fourth, was the relatively short four month duration of symptoms and signs in this individual an important parameter in the rapid resolution of the symptoms and signs of CRPS? It is no longer believed that CRPS progresses through stages in a time-locked progression, as initially suggested by Bonica [12].

5. Fifth, was the relatively restricted distribution of clinical symptoms and signs of CRPS an important factor in the resolution of symptoms and signs of CRPS? Although answers to these questions cannot be provided at the present time, these questions provide valuable parameters to be critically investigated in any future studies.

### Summary

In summary, this case report has documented a rapid resolution of Budapest criteria established CRPS during the course of a bilateral mastectomy after which a long-duration infusion of intrapleural 0.5% Marcaine<sup>®</sup> was administered through an On-Q pump and catheter system over a period of several days.

The CRPS was well-localized, within four months of onset and all symptoms and signs completely resolved during the course of a three hour bilateral total mastectomy. Potential issues for further investigation have been addressed. The last point of which is with respect to the need for early diagnosis of CRPS to be considered in order to have access to patients in whom the CRPS has not progressed beyond a highly localized distribution.

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**Volume 4 Issue 7 July 2021**

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