

Amputation as an Unusual Treatment for Therapy-Resistant Complex Regional Pain Syndrome, Type 1

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Background: Complex regional pain syndrome, type 1 (CRPS-1) causes severe pain that can be resistant to multiple treatment modalities. Amputation as a form of long-term treatment for therapy-resistant CRPS-1 is controversial.

Case Report: We report the case of a 38-year-old man who failed all treatment modalities for CRPS-1, including medication, steroid injections, and spinal cord stimulator implantation. Below-the-knee amputation to relieve intractable foot and ankle pain resulted in a favorable outcome for this patient.

Conclusion: Select patients with severe CRPS-1 who are unresponsive to all forms of treatment for pain may benefit from amputation as a last option for relief of suffering. Larger studies are needed to prove the efficacy of amputation.

Keywords: Amputation, complex regional pain syndrome–type 1, reflex sympathetic dystrophy

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INTRODUCTION

Complex regional pain syndrome, type 1 (CRPS-1) is a chronic neurologic condition that encompasses multiple clinical symptoms, including severe spontaneous pain, allodynia, vasomotor dysfunction, motor impairment, swelling, and autonomic instability.¹ CRPS-1 is difficult to treat, and treatment results are usually unsatisfactory. Treatment modalities include medication, physical therapy, psychological therapy, and neuromodulation including spinal cord stimulator (SCS) implantation.²⁻⁴

Amputation as a form of long-term treatment for therapy-resistant CRPS-1 is controversial.⁵ Limb amputation is rarely done exclusively for pain relief because of concerns that pain and disability will persist after the procedure.⁶

We present the case of a patient with CRPS-1 resistant to all treatment modalities who underwent below-the-knee amputation, resulting in improvement of pain postsurgery.

CASE REPORT

A 38-year-old man with a history of hypertension and obesity had sustained a left ankle injury and fracture in 2005, leading to multiple unsuccessful surgeries. He had a 5-year history of severe pain that extended from the left knee to the left foot. He was diagnosed with CRPS-1 after his initial ankle surgery. The pain affected his daily activities (functional class 3-4), and he had not been able to walk for more than 5 years.

The patient had tried medication, physical therapy, and multiple failed neuraxial interventions, including epidural

steroid injections, sympathetic nerve blocks, SCS (implanted in 2010 and removed in 2012), ketamine infusions (sometimes on a weekly basis), and ketamine coma. All interventions failed to relieve his pain or to improve his functional status. We offered the patient the option of having a below-the-knee amputation as a last resort; because of his long-term incapacitating pain, he agreed to surgery.

The patient underwent ketamine infusion the week before surgery. He was admitted to the hospital for tunneled epidural placement and titration prior to surgery. The below-the-knee amputation was performed with no immediate complications. Continuous femoral and popliteal catheters were placed for postoperative pain control, and the patient was discharged after 9 days.

Two days after discharge, the patient was readmitted to the hospital for debridement. On readmission, continuous femoral and popliteal catheters were again placed, but they did not effectively control his pain and were removed on postoperative day 3. A lumbar epidural catheter was then placed that controlled his pain during the hospital stay. During the hospital course, the patient received adjuvant pain medication, including intravenous Dilaudid (hydromorphone) patient-controlled analgesia, Dilaudid po, Neurontin (gabapentin), and Oxycontin (oxycodone). The patient stayed in the hospital for 13 days during the second admission.

At 6-week follow-up, the patient reported significant improvement in his pain level, 3-4/10 down from 8-10/10 before surgery. He complained of minor phantom limb pain but was satisfied because he did not have the incapacitating

CRPS-1-related pain he had had in the past. The patient received a prescription for a left lower extremity prosthesis.

DISCUSSION

CRPS-1 is associated with functional loss because of dysregulation of the autonomic and central nervous systems. CRPS-1 is a chronic neurologic disorder associated with severe intractable pain that can cause long-term disability.^{2,3} Patients with CRPS-1 usually experience psychological distress.¹ The diagnosis of CRPS-1 is based on clinical signs, symptoms, and physical examination findings. Despite developments in the understanding of CRPS-1, many cases are suboptimally treated, and patients remain disabled.^{1,7} CRPS-1 is often resistant to therapy and has an unpredictable clinical course.^{2,5} Treatment, including physical rehabilitation and pharmacologic pain therapy, needs to be started as early as possible. If no improvement is achieved, interventional pain management should be pursued. All options should be explored for pain control and dysfunctional limb treatment before amputation of the limb is considered. Despite all efforts, if a patient has long-standing and therapy-resistant CRPS-1, amputation can be justified, as it was in this case.

Our patient eventually improved with loss of pain and autonomic instability. Previous studies show a possibility of recurrence of CRPS-1 in stump and other limbs, as well as phantom pains.^{5,7,8} However, the possibility and severity of these complications are low, and overall patient satisfaction with pain relief after amputation is significant.^{7,9}

Patient satisfaction may be influenced by other factors such as coping skills and resilience.^{5,10} Select patients with resistant CRPS-1 have reported better quality of life and less psychological distress after amputation.¹⁰

CONCLUSION

We believe select patients with severe CRPS-1 who are unresponsive to all forms of treatment for pain may benefit from amputation as a last option for relief of suffering. Further research needs to be conducted to help determine which patients may benefit from amputation, level of

amputation, recurrence of CRPS-1, patient satisfaction, and level of functional gains postamputation.

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