Short- and long-term results of an inpatient programme to manage Complex Regional Pain Syndrome in children and adolescents

British Journal of Pain
2017, Vol 11(2) 87–96
© The British Pain Society 2017
Reprints and permissions:
sagepub.co.uk/journalsPermissions.nav
DOI: 10.1177/2049463717695695
journals.sagepub.com/home/bjp

Giovanni Cucchiaro¹, Kevin Craig², Kerri Marks¹, Kristin Cooley²,
Thalitha Kay Black Cox² and Jennifer Schwartz¹

Abstract

The aim of this retrospective study was to determine whether an inpatient approach and the use of regional anaesthesia techniques can accelerate the recovery to normal functions in children with Complex Regional Pain Syndrome (CRPS). This study looked at the data of patients admitted to the rehabilitation unit with a diagnosis of CRPS from January 2010 to April 2015. Variables such as hospital stay, medications administered, regional anaesthesia procedures, changes in functional status prior to treatment and at the time of discharge, psychological evaluation and diagnosis were evaluated. A total of 31 patients (21 females and 10 males) were admitted with a diagnosis of CRPS 1 and 2. In all, 97% of the patients received a peripheral or central nerve catheter for an average of 4 days with pain scores of Verbal Numeric Scale (VNS) score = 1.0 ± 0.7 and an average length of hospital stay of 8.2 ± 2.6 days. The modified Functional Independence Measure for Children (WeeFIM) scores and Canadian Association of Occupational Therapists tests significantly improved at the time of hospital discharge, as well as their pain scores, which decreased from 8.2 ± 2 to 1.6 ± 3 . In conclusion, these data suggest that the use of regional anaesthesia techniques and an intensive inpatient rehabilitation programme could accelerate the recovery of children with CRPS.

Keywords

Children, Complex Regional Pain Syndrome, CRPS, regional anaesthesia, inpatient rehabilitation

Introduction

The existence of Complex Regional Pain Syndrome (CRPS) in children and adolescents is a well-recognized medical problem.^{1,2} There is a paucity of data in the literature on the best approach to manage pain and enable a prompt functional recovery. Physical therapy (PT) and occupational therapy (OT) have been recognized as the foundations of successful treatment of CRPS in children.^{3,4} While previous approaches recommended simple observation with no treatment,⁵ more recent reports have described more aggressive approaches by adding regional anaesthesia techniques (mostly sympathetic nerve blocks)^{6,7} and pharmacological interventions^{8,9} to the PT and OT sessions.

Similarly, programmes managing CRPS in children are increasingly introducing cognitive-behavioural therapy techniques to the list of interventions offered to their patients.^{6,10}

¹Department of Anesthesiology Critical Care Medicine, Children's Hospital Los Angeles, Los Angeles, CA, USA

²Department of Pediatrics, Children's Hospital Los Angeles, Los Angeles, CA, USA

Corresponding author:

Giovanni Cucchiaro, Department of Anesthesiology Critical Care Medicine, Children's Hospital Los Angeles, 4650 Sunset Boulevard, Los Angeles, CA 90027, USA.
Email: gucchiaro@chla.usc.edu

The aims of this retrospective study were to determine how many days were needed for children with CRPS to regain the function of the affected limb, feel minimal pain and regain their daily activity functions once they had been admitted to an intensive inpatient rehabilitation programme, which included an extensive use of regional anaesthesia techniques.

Methods

This study was approved by the paediatric health institution's institutional review board (IRB) committee with a consent waiver. Data for this study were obtained from a database that encompasses the demographics and medical history of patients admitted to the hospital between February 2010 and April 2015 with a diagnosis with CRPS. The diagnosis of CRPS type I, type II and CRPS Not Otherwise Specified (NOS)¹¹ was made based on the Harden/Bruehl clinical criteria for CRPS.¹² They were based on patients' reports of continuous pain which was disproportionate to any triggering event, in conjunction with other symptoms including reports of hyperaesthesia, temperature asymmetry and/or skin colour changes, oedema and/or sweating changes, and decreased motor function and/or trophic changes in the affected extremity.¹¹ The physical examination must have confirmed the patients' reports, as well as the presence of hyperalgesia and/or allodynia.¹² The criteria for admission in the inpatient unit were the presence of chronic pain (greater than 3 months), lack of subjective and objective improvements of symptoms from previous medical interventions and loss of function of the extremity. Demographic (age, sex, type and localization of lesions) and medical history data (duration of symptoms, previous hospitalizations and medications used) were analysed. Also considered within the study were duration of the hospital stay, medications used, regional anaesthesia procedures conducted and changes in functional status prior to treatment and at the time of discharge. The functional status was measured using a modified Functional Independence Measure for Children (WeeFIM)¹³ at the time of admission and discharge from the hospital. The original instrument included 18 items focused on function in three domains: self-care, mobility and cognition. Our study utilized only the first two criteria. The scores for each item ranged from 1 to 7 based on the patient's dependence level (1 = total assistance; 7 = independence). The Canadian Association of Occupational Therapists tests for patients' performance and satisfaction (Canadian Occupational Performance Measure (COPM)) were also conducted at the time of admission and discharge from the hospital.¹⁴ The test is a patient-centred approach that enables children to identify performance

problems, concerns and issues that are important to them. It is a semi-structured interview designed for use by occupational therapists containing three sections: self-care (personal care, functional mobility and community management), productivity (paid/unpaid work, household management and play/school) and leisure (quiet recreation, active recreation and socialization).¹⁵ For each activity area, there are two self-report scoring domains of performance and satisfaction. These values range between 1 and 10: with 1 indicating poor performance and low satisfaction and 10 indicating very good performance and high satisfaction. The pain scores were measured at the time of the pre-admission visit in the Pain Clinic using the Verbal Numeric Scale (VNS) with values ranging from 0 (no pain) to 10, with 10 being the worst pain imaginable,¹⁶ 12 hours after the peripheral or central neuraxial catheters were removed and at the time of discharge from the hospital. Our treatment protocol contained a provision stating that patients are not asked to rate their pain daily during the hospital admission.

Every patient had a psychological evaluation conducted by a psychologist and a licensed clinical social worker (LCSW) prior to the admission. The LCSW was involved with the patients' care during the hospital stay and follow-up. Every patient was evaluated by a physical and occupational therapist at the time of admission.

Description of rehabilitation programme

Patients who met the criteria for admission to the rehabilitation programme were identified in the Pain Clinic. A detailed explanation of the programme and goals was given to the families and patients. The initial evaluation was conducted by a pain specialist and the LCSW. Patients must have shown signs of neuropathic pain, including numbness, allodynia, hyperalgesia, functional impairment and physical changes in the extremities, including oedema and discolouration. A documented injury was not considered mandatory criteria for admission. Patients must have failed in prior medical and/or rehabilitation interventions.

On the day of hospital admission, the families and patients met the entire team involved with the patient's care during the hospital stay and a detailed description of the programme was given. The physical and occupational therapists set the goals of treatment. The same morning, patients were taken to the operating room for the regional anaesthesia procedure under general anaesthesia.

PT and OT sessions started the first afternoon. During the rest of the hospitalization, patients followed a structured schedule that included single and group

PT, OT, psychotherapy, art and music therapy and school homework with a teacher (Appendix 1). Patients were encouraged to continue with the exercises once they were in their rooms. Parents were only allowed to spend 2 hours a day at dinner time with their children. No cell-phones were allowed during the hospital stay and personal computers were allowed for school work only. Patients were considered eligible for discharge once they had recuperated the function of their affected extremity: walking independently for patients with CRPS of the lower extremities and full use of the hand/arm for patients with CRPS of the upper extremities.

Procedures

Patients received infraclavicular or interscalene catheters for CRPS of the upper extremities, femoral catheters for CRPS of the thigh and knee or sciatic catheters for CRPS of the foot. Epidural catheters were placed for CRPS diffuse to both lower extremities. Peripheral nerve catheters were initially loaded with ropivacaine 0.1% (sciatic catheters) or ropivacaine 0.15% 0.5 mL/kg (max. 20 mL). The nerve catheters were connected to an On-Q® C-bloc pump (I-Flow, Lake Forest, CA) to deliver ropivacaine 0.15% or 0.1% (sciatic catheters) at a rate ranging from 6 to 12 mL/hour depending on catheter location. The epidural catheters were initially loaded with bupivacaine 0.1% + epinephrine 1:200,000 (0.5 mL/kg, max. 15 mL) followed by an infusion of ropivacaine 0.1% and preservative-free morphine (20 mcg/mL) at 0.3–0.4 mL/kg (max. 12 mL). At day 3 of treatment, the catheters were clamped for 6 hours and then removed if patients rated the pain at 2 or less on a numerical scale, otherwise the infusion was restarted for an additional 48 hours. Morphine was added to the epidural infusions to minimize the dose of local anaesthetic needed to provide effective pain relief and prevent motor blockade. The infusion of local anaesthetic was also adjusted during this period of time to prevent motor blocks and allow patients to actively participate in the PT and OT sessions.

Medications

There was an effort to wean patients off most of the medication that they were taking during their hospital stay; in particular, every patient was weaned off opioids. The decision to stop or start antidepressants and anxiolytic medications was done on an individual basis.

PT

PT focused on oedema management, desensitization, range of motion, active use of the extremity, weight bearing on the extremity, strengthening, coordination,

decreased muscle guarding and promotion of typical posture and alignment. Allodynia was treated with desensitization including lotion massage, vibration and textured fabric massage with varying pressure, and fabric and contrast baths with increasing temperature differences. Endurance exercises included stationary bicycling with varying resistance, treadmill walking/jogging/running at various speeds and inclines, mini-trampoline jumping, weight shifting and balance activities, step-ups or jumping jacks. Isolated muscle strengthening, such as heel raises, squats, lunges and general strengthening including push-ups and sit-ups, was also incorporated. Therapy ball exercises were used for weight shifting and rotation, balance, trunk stability and core strengthening. Aquatic therapy was used to encourage weight bearing; improve endurance, balance and coordination; reduce oedema; and decrease hypersensitivity.

OT

Patients were taught different yoga sequences, depending on the extremity that was affected by CRPS. These were derived from the Carlson–Watson stress-loading programme,¹⁷ which consists of a specific set of exercises intended to improve the clinical course of patients who suffer from CRPS by using the weight bearing/loading component and making it specific to patients by using yoga postures. Patients received a DVD in their room to help them continue exercising after the one-on-one sessions. The DVD included three sections showing diaphragmatic breathing, guided relaxation, supine and seated postures with gentle weight bearing and sensory/tactile input and exercises for when patients had regained full sensation and the nerve catheters had been removed. They required a higher balance level and focus on single leg stance, full weight bearing and strengthening postures. Therapists also assessed each patient's lifestyle and habits in order to create a healthier lifestyle balance.

Psychotherapy

Psychotherapy, with an emphasis on cognitive-behavioural techniques, played a significant role in the treatment of CRPS and chronic pain.¹⁸ The patients received 1 hour of counselling every day during their hospitalization, focused on teaching relaxation techniques: including diaphragmatic breathing, progressive muscle relaxation, guided imagery and an introduction to biofeedback using heart rate variability (HRV). These modalities assisted the patients in learning ways to internally distract themselves in order to decrease their pain. In addition, cognitive-behavioural therapy was utilized, which focused on teaching patients various

ways to identify and change maladaptive thoughts, feelings and behaviours with a focus on improving their quality of life and decreasing their psychological distress.¹⁹ Diagnosis of mood and/or anxiety disorder was made based on the *Diagnostic and Statistical Manual of Mental Disorders* (4th ed., text rev.; DSM-IV-TR)²⁰ criteria (Appendix 2).

Follow-up

Patients were scheduled for follow-up visits in the pain clinic within 2 weeks of their hospital discharge date. Additional visits were arranged based on the clinical findings. Subsequent outcome data were obtained using phone interviews or by analysing the patient's electronic medical record. Relapse was defined as recurrence of symptoms and signs of neuropathic pain, including numbness, allodynia, hyperalgesia, functional impairment and physical changes in the extremities, including oedema and discolouration.

Statistical analysis

The variables considered in the analysis were tested for distribution using the Skewness and Kurtosis test for normality. Age and WeeFIM scores were the only variables normally distributed. Continuous variables were compared using paired t-tests or Wilcoxon signed-rank tests, as appropriate.

A linear regression was run to determine whether age, sex, duration of symptoms prior to hospitalization and patients' psychological profile would predict a recurrence of CRPS.

A chi-square goodness-of-fit test was used to test whether the observed proportions for gender distribution differed from the hypothesized proportions of 50%.

Results

Population description

A total of 31 patients were admitted during the study period. The patients' demographics and CRPS characteristics are shown in Table 1. The study found a significantly higher number of female patients than male patients ($p = 0.01$) and CRPS affected the lower extremity in the majority of patients (90%, $p = 0.0001$). These results are consistent with those reported in previous studies.^{21,22} The diagnosis at the time of admission was CRPS type 1 in 23 patients, CRPS type 2 in 2 patients, and CRPS NOS in 6 patients.

Patient symptoms were assessed in the study. As a result, allodynia was present in every patient. Other most common symptoms found were skin discolouration, decreased range of motions and oedema in the

Table 1. Patients' demographics.

	Total	Male	Female	p
n	31	10	21	0.01
Age (years)	13 ± 3	14 ± 3	13 ± 3	0.3
CRPS classification				
CRPS 1	23	7	16	
CRPS 2	2	2	0	
CRPS NOS	6	1	5	
CRPS localization				
CRPS upper extremities	2	1	1	
CRPS lower extremities	28	9	19	
CRPS upper/lower extremities	1	0	1	
Mood disorders	14	2	13	0.035

CRPS: Complex Regional Pain Syndrome; CRPS NOS: Complex Regional Pain Syndrome Not Otherwise Specified.

affected extremity. Trophic changes were present in three patients only. Symptoms on admission are illustrated in Table 1.

Furthermore, patients had been complaining of severe pain for an average of 187 ± 131 days prior to the admission into the programme. Of the total patients, 10 (32%) had been hospitalized because of pain before admission into our programme. The number of hospitalizations ranged between 1 and 3 (average = 1.4) and patients spent an average of 14 ± 13 days (range 1–45) in the hospital because of pain symptoms. In all, 13 patients went to local emergency rooms, anywhere between one and six times prior to their admission in the programme because of CRPS-induced pain. Regional anaesthesia techniques, all single injections of local anaesthetics, had been performed in seven patients (23%) by other physicians to manage CRPS-induced pain. These techniques included three lumbar sympathetic blocks, two stellate ganglion blocks, one sciatic nerve block and one ankle block. In all, 16 patients (52%) participated in multiple unsuccessful PT sessions prior to their admission in the programme. No specific interventions to address pain beside medications had been offered to 12 patients (39%).

In all, 14 patients (45%) used crutches, six (19%) were wheelchair-bound and three (10%) used a walker at the time of their hospital admission. Two patients with shoulder pain wore a sling and were not able to utilize their arm.

Only three patients were not taking any medication at the time of admission. Every other patient was on multiple medications. Eight patients were on pregabalin and six on neurontin. Six patients were taking opioids, including methadone, oxycodone, codeine or

hydrocodone. Eight patients were on an antidepressant, including sertraline, duloxetine, amitriptyline and citalopram. Five patients were on nonsteroidal anti-inflammatory drugs (NSAIDs).

In all, 14 patients (45%) had been missing school and were receiving poor grades, and 3 patients (9%) had been home-schooled since they had been diagnosed with CRPS.

Of the total patients, 13 (42%) had been diagnosed with a mood disorder by outside providers prior to their admission (depression: 7, anxiety: 4, bipolar: 1). In eight of these patients, the diagnosis of mood disorder preceded the development of CRPS, while in five the diagnosis of depression and anxiety was made after the development of CRPS. Mood disorders were more frequent in girls than in boys (12 and 2, respectively, $p = 0.052$). The diagnoses were confirmed at the time of the initial evaluation by the team psychologist.

Hospital stay

A total of 30 patients had a central or peripheral nerve catheter placed on the day of admission and one refused the procedure. The number and type of procedures performed are listed in Table 2.

The average length of stay was 8.2 ± 2.6 days (range 5–15 days). Catheters were left in place for an average of 4.4 days (range 3–9 days). Catheters were removed after 3 days in 33% of the patients and after 4 days in 32% of

the patients. Pain scores were 1.0 ± 0.7 at the time the catheter was removed. Changes in the VNS, WeeFIM and COPM performance and satisfaction scores are shown in Table 3. Pain scores significantly improved during the hospital stay, and the three indicators of patients function, performance and satisfaction were significantly higher at the time of the hospital discharge compared with those reported at the time of admission ($p = 0.0001$). No patient needed an assistive device at the time of discharge. Most patients were weaned off opioids and other medications during their hospital stay, except for three patients who were discharged on pregabalin and two on Neurontin. Antidepressants were discontinued in four of the nine patients who were taking this class of medications at the time of the admission.

Follow-up

The follow-up was completed in the outpatient clinic on 30 patients (97%) from the study sample. The average follow-up was 13 ± 12 months. Seven patients (23%) continued with PT for 6 weeks after discharge and 10 (31%) with physical and psychotherapy. In all, 10 patients (31%) were referred to a mental health provider. No follow-up treatments were recommended for 5 patients (19%).

In addition, five patients (17%), three girls and two boys, relapsed during the follow-up period. The study defined relapse as a recurrence of symptoms. Each patient who relapsed was evaluated by the medical team. The time between discharge and a new episode of CRPS was 13 ± 8 months. The recurrence involved the same extremity initially affected. In three of the cases, the relapse was due to a documented new trauma. A new admission into our rehabilitation programme was necessary in four of these five patients, while the fifth was admitted to a different hospital. Three of those patients readmitted for recurrent CRPS underwent the same programme and received a central or peripheral nerve block, while the fourth refused a procedure and was managed with psychotherapy and PT and OT sessions. The duration of this second hospitalization was 7.8 ± 4.3 days (range 2–11). Patients

Table 2. List of central and peripheral nerve catheters placed at the time of admission.

Procedure	No. of patients
Epidural catheter	6
Femoral catheter	2
Sciatic catheter	18
Two sciatic catheters	1
Interscalene catheter	2
Epidural + infraclavicular catheter	1
None	1

Table 3. Changes in patients' Verbal Numeric Scale (VNS) scores, functional (WeeFIM), performance (COPM performance) and satisfaction (COPM satisfaction) scores at the time of the patients' initial evaluation and at the time of discharge from the hospital.

Test	Pre	After catheters removed	Post	p
VNS	8.2 ± 2	1.0 ± 0.7	1.6 ± 3	0.0001
WeeFIM	43 ± 10		55 ± 5	0.0001
COPM performance	2.9 ± 1.2		8.3 ± 1.4	0.0001
COPM satisfaction	2.9 ± 1.4		8.3 ± 3.0	0.0001

WeeFIM: modified Functional Independence Measure for Children; COPM: Canadian Occupational Performance Measure.

Table 4. Changes in patients' Verbal Numeric Scale (VNS) scores, functional (WeeFIM), performance (COPM performance) and satisfaction (COPM satisfaction) scores in the four patients who were readmitted in the rehabilitation programme for recurrent CRPS.

Test	Pre	Post	p
VNS	8 ± 1	0	0.0001
WeeFIM	32 ± 5.2	48 ± 2	0.007
COPM performance	3.4 ± 1	8 ± 1.4	0.002
COPM satisfaction	2 ± 0.8	9 ± 1	0.0001

WeeFIM: modified Functional Independence Measure for Children; COPM: Canadian Occupational Performance Measure; CRPS: Complex Regional Pain Syndrome.

were discharged to home after full functional recovery of the affected extremity. Changes in the VNS, WeeFIM and COPM performance and satisfaction scores are shown in Table 4.

An additional three patients (10%) were hospitalized during the follow-up period: one due to a new diagnosis of schizophrenia, one for acute episode of back pain and one to undergo a Bankart procedure.

Of the 14 patients who had a history of mood disorder prior to their admission, 11 continued experiencing similar psychological problems during the follow-up period. One patient was lost to the follow-up and two improved during the follow-up period. Eight patients (25%) were newly diagnosed with anxiety and/or depression and one with schizophrenia during the follow-up period. The overall prevalence of psychiatric disorders found during the follow-up was 58%.

Although no significant differences were found in the incidence of psychiatry conditions in patients who relapsed versus those who did not ($p = 0.3$), the linear regression analysis showed that the presence of persistent mood disorders after the discharge from the hospital was a weak predictor of a relapse ($R^2 = 0.15$, $p = 0.033$).

In all, 23 (72%) patients went back to school after being discharged from the hospital. Of these, 10 patients had poor school attendance prior to the hospitalization because of CRPS pain. Three patients who were previously home-schooled continued with their home-school programme. Four patients continued missing classes and one stopped going to school during the follow-up period because of their psychiatric condition.

The hospital charges for patients admitted to the programme, which includes room and board, operating room utilization and medications, were US\$2900 per day on average. The hospital charged US\$270 for 50 minutes outpatient PT or OT sessions.

Discussion

Data from this review show that an inpatient approach which included the prolonged perineural infusion of local anaesthetics combined with intense PT and OT in children with CRPS can lead to pain resolution within 4 days of treatment. Ours is one of the many approaches developed by other pain programmes to manage children with CRPS. They include a day-hospital, outpatient and inpatient approach. In these studies, the time to functional recovery varied between 3 and 7 weeks.^{4,23,24} Similarly, children admitted to the hospital for intensive treatment without the use of regional anaesthesia techniques spent an average of 14 days as inpatients.^{3,25} While PT and OT and cognitive-behavioural therapy remain the mainstay treatments⁹ of patients with CRPS, the role and effectiveness of peripheral or central neuraxial nerve blocks is uncertain. In this review, 23% of our patients received a one-time nerve block by other providers before being admitted to our programme. The blocks were the only treatment offered to these patients, and the pain recurred within 24 hours in all of them. This seems to confirm a recent Cochrane review of the literature on the effects of sympathetic blocks in adult CRPS,²⁶ which suggested a lack of efficacy of single injection lumbar sympathetic blocks. Similarly, previous retrospective studies in paediatric patients have failed to show clear benefits from single injection sympathetic blocks.^{27,28} Conversely, recent studies have reported long-lasting effects on CRPS pain when specific nerves had been targeted (femoral and sciatic nerve), patients had received a prolonged infusion of local anaesthetic and the blocks had been done in the context of a multi-disciplinary approach to CRPS management.^{6,29,30} In the Dadure study, the authors used an approach similar to ours. A total of 13 children received a 4-day prolonged infusion of local anaesthetic, and they were enrolled in PT sessions that began as inpatient and continued at home. This plan resulted in a resolution of pain within 2 months.⁶ A paediatric case report has confirmed the efficacy of prolonged perineural infusion of local anaesthetic with resolution of pain in 3 days.³¹ These data are similar to what has been shown in adults with CRPS.^{32,33} In this study, we also observed a resolution of pain in 68% of our patients after 4 days of local anaesthetic infusion. At this point, it is unclear whether the prolonged infusion of local anaesthetic alone contributes to the resolution of the symptoms,²⁹ or whether it simply facilitates the patients' participation in PT sessions. It should be noted that our patients remained in the hospital an additional 4 days on average after the peripheral and central nerve catheters were removed, even though they reported minimal pain. The additional time was needed for some patients to be able to

ambulate over a variety of surfaces, go up and down two flights of stairs independently and complete their activities of independent daily living. Other patients required a prolonged psychological assessment and adjustments to their psychotropic medications.

The recurrence rate in our population is lower than what has been reported by other authors.^{1,2,6,23} Most of the patients who relapsed suffered from a new injury in the same extremity and were treated accordingly. The relative long-term success may have been due to the close follow-up established for the patients, in combination with the recommendation to continue with psychotherapy and PT after the hospital discharge, and the 100% compliance from patients and families with the recommended treatments.

It was not surprising to find a high percentage of depression and anxiety disorders in our study population. In all, 26% of our patients were treated for a mood disorder prior to developing CRPS, and 19% developed depression and anxiety symptoms after having experienced severe pain for a prolonged period of time. More problematic were the data obtained during the follow-up period: 79% of the patients with a psychiatric disorder diagnosis upon admission presented with the same symptoms during the follow-up period. Furthermore, 23% of our patients were newly diagnosed with anxiety, depression and schizophrenia during the follow-up period, with an overall 58% prevalence of psychiatric disorders in this population. Previous studies have found that Axis I psychiatric disorders, such as major depression, panic disorder, generalized anxiety disorder and post-traumatic stress disorder, are common in CRPS patients.^{27,34–36} The presence of a psychiatric disorder was the only significant predictor of CRPS relapse. This finding strongly shows the need for close psychological and psychiatric follow-up even after the return to normal functions.

A review of the literature and our experience leave us with the unresolved question of whether the rehabilitation process should occur in either outpatient or inpatient settings. The lack of randomized studies and differences between treatments protocols make it impossible to formulate a definitive recommendation. Sherry et al.³ found that patients who did not respond completely to his therapy programme were receiving outpatient rather than inpatient therapy. The time to full functional recovery is another element that should be taken into consideration.

The decision on whether to start an in- or outpatient rehabilitation programme for children with CRPS is mostly determined by contracts that hospitals have with the different payers.

The cost of an 8-day admission in our rehabilitation programme is approximately US\$23,300 at the hospital, which is higher in comparison to the cost of 2 weeks

of four daily PT and OT sessions, which would cost approximately US\$15,000 at our institution. However, this estimated amount for outpatient services does not include the costs of placing a peripheral or central nerve catheter and psychotherapy sessions. Furthermore, there are additional factors that play a role in deciding which approach to use in these patients. Private payers in California and the California Department of Health Care Services will not reimburse for 2 weeks of four daily outpatient PT and OT sessions unless specific contracts are negotiated between the payers and the specialty care centre. There are also other costs often overlooked, including lost wages and travelling/housing costs that families incur in order to be with their children during these lengthy interventions.

In conclusion, the combination of regional anaesthesia and an intensive inpatient rehabilitation programme could be a useful, alternative approach to the treatment of children with CRPS because of a fast recovery to normal function. It seems important to consider the psychological profile of these patients and to establish a long-term counselling programme to help with the transition to a normal social life and minimize the rate of symptoms recurrence.

Conflict of interest

The author(s) declared no potential conflicts of interest with respect to the research, authorship and/or publication of this article.

Funding

The author(s) received no financial support for the research, authorship and/or publication of this article.

References

1. Lee BH, Scharff L, Sethna NF, et al. Physical therapy and cognitive-behavioral treatment for complex regional pain syndromes. *J Pediatr* 2002; 141(1): 135–140.
2. Berde CB and Lebel A. Complex regional pain syndromes in children and adolescents. *Anesthesiology* 2005; 102(2): 252–255.
3. Sherry DD, Wallace CA, Kelley C, et al. Short- and long-term outcomes of children with complex regional pain syndrome type I treated with exercise therapy. *Clin J Pain* 1999; 15(3): 218–223.
4. Logan DE, Carpino EA, Chiang G, et al. A day-hospital approach to treatment of pediatric complex regional pain syndrome: initial functional outcomes. *Clin J Pain* 2012; 28(9): 766–774.
5. Betend B, Lebacqz E, Kohler R, et al. Metaphyseal osteolysis. Unusual aspect of reflex algodystrophy in children (author's transl.). *Arch Fr Pediatr* 1981; 38(2): 121–123.
6. Dadure C, Motais F, Ricard C, et al. Continuous peripheral nerve blocks at home for treatment of recurrent complex regional pain syndrome I in children. *Anesthesiology* 2005; 102(2): 387–391.

7. Wettrell G, Hallbook T and Hultquist C. Reflex sympathetic dystrophy in two young females. *Acta Paediatr Scand* 1979; 68(6): 923–924.
8. Perez RS, Zuurmond WW, Bezemer PD, et al. The treatment of complex regional pain syndrome type I with free radical scavengers: a randomized controlled study. *Pain* 2003; 102(3): 297–307.
9. Wilder RT. Management of pediatric patients with complex regional pain syndrome. *Clin J Pain* 2006; 22(5): 443–448.
10. Bruhl S and Chung OY. Psychological and behavioral aspects of complex regional pain syndrome management. *Clin J Pain* 2006; 22(5): 430–437.
11. Borchers AT and Gershwin ME. Complex regional pain syndrome: a comprehensive and critical review. *Autoimmun Rev* 2014; 13: 242–265.
12. Harden RN, Bruhl S, Stanton-Hicks M, et al. Proposed new diagnostic criteria for complex regional pain syndrome. *Pain Med* 2007; 8(4): 326–331.
13. Ziviani J, Ottenbacher KJ, Shephard K, et al. Concurrent validity of the functional independence measure for children (WeeFIM) and the pediatric evaluation of disabilities inventory in children with developmental disabilities and acquired brain injuries. *Phys Occup Ther Pediatr* 2001; 21(2–3): 91–101.
14. Law M, Baptiste S, McColl M, et al. The Canadian occupational performance measure: an outcome measure for occupational therapy. *Can J Occup Ther* 1990; 57(2): 82–87.
15. Cusick A, McIntyre S, Novak I, et al. A comparison of goal attainment scaling and the Canadian occupational performance measure for paediatric rehabilitation research. *Pediatr Rehabil* 2006; 9(2): 149–157.
16. Bailey B, Daoust R, Doyon-Trottier E, et al. Validation and properties of the verbal numeric scale in children with acute pain. *Pain* 2010; 149(2): 216–221.
17. Watson HK and Carlson L. Treatment of reflex sympathetic dystrophy of the hand with an active ‘stress loading’ program. *J Hand Surg Am* 1987; 12(5): 779–785.
18. Logan DE, Williams SE, Carullo VP, et al. Children and adolescents with complex regional pain syndrome: more psychologically distressed than other children in pain? *Pain Res Manag* 2013; 18(2): 87–93.
19. Otis JD, Sanderson K, Hardway C, et al. A randomized controlled pilot study of a cognitive-behavioral therapy approach for painful diabetic peripheral neuropathy. *J Pain* 2013; 14(5): 475–482.
20. American Psychiatric Association. *Diagnostic and statistical manual of mental disorders*. Washington, DC: American Psychiatric Association, 2000.
21. Meier PM, Zurakowski D, Berde CB, et al. Lumbar sympathetic blockade in children with complex regional pain syndromes: a double blind placebo-controlled crossover trial. *Anesthesiology* 2009; 111(2): 372–380.
22. Zernikow B, Dobe M, Hirschfeld G, et al. Please don’t hurt me!: a plea against invasive procedures in children and adolescents with complex regional pain syndrome (CRPS). *Schmerz* 2012; 26(4): 389–395.
23. Murray CS, Cohen A, Perkins T, et al. Morbidity in reflex sympathetic dystrophy. *Arch Dis Child* 2000; 82(3): 231–233.
24. Maynard CS, Amari A, Wiczorek B, et al. Interdisciplinary behavioral rehabilitation of pediatric pain-associated disability: retrospective review of an inpatient treatment protocol. *J Pediatr Psychol* 2010; 35(2): 128–137.
25. Low AK, Ward K and Wines AP. Pediatric complex regional pain syndrome. *J Pediatr Orthop* 2007; 27(5): 567–572.
26. Stanton TR, Wand BM, Carr DB, et al. Local anesthetic sympathetic blockade for complex regional pain syndrome. *Cochrane Database Syst Rev* 2013; 8: CD004598.
27. Wesdock KA, Stanton RP and Singsen BH. Reflex sympathetic dystrophy in children. A physical therapy approach. *Arthritis Care Res* 1991; 4(1): 32–38.
28. Wilder RT, Berde CB, Wolohan M, et al. Reflex sympathetic dystrophy in children. Clinical characteristics and follow-up of seventy patients. *J Bone Joint Surg Am* 1992; 74(6): 910–919.
29. Everett A, Mclean B, Plunkett A, et al. A unique presentation of complex regional pain syndrome type I treated with a continuous sciatic peripheral nerve block and parenteral ketamine infusion: a case report. *Pain Med* 2009; 10(6): 1136–1139.
30. Kato J, Gokan D, Ueda K, et al. Successful pain management of primary and independent spread sites in a child with CRPS type I using regional nerve blocks. *Pain Med* 2011; 12(1): 174.
31. Franklin A and Austin T. The use of a continuous brachial plexus catheter to facilitate inpatient rehabilitation in a pediatric patient with refractory upper extremity complex regional pain syndrome. *Pain Pract* 2013; 13(2): 109–113.
32. Detaille V, Busnel F, Ravary H, et al. Use of continuous interscalene brachial plexus block and rehabilitation to treat complex regional pain syndrome of the shoulder. *Ann Phys Rehabil Med* 2010; 53(6–7): 406–416.
33. Toshniwal G, Sunder R, Thomas R, et al. Management of complex regional pain syndrome type I in upper extremity-evaluation of continuous stellate ganglion block and continuous infraclavicular brachial plexus block: a pilot study. *Pain Med* 2012; 13(1): 96–106.
34. Bernstein BH, Singsen BH, Kent JT, et al. Reflex neurovascular dystrophy in childhood. *J Pediatr* 1978; 93(2): 211–215.
35. Sherry DD and Weisman R. Psychologic aspects of childhood reflex neurovascular dystrophy. *Pediatrics* 1988; 81(4): 572–578.
36. Harden RN, Oaklander AL, Burton AW, et al. Complex regional pain syndrome: practical diagnostic and treatment guidelines. 4th edition. *Pain Med* 2013; 14(2): 180–229.

Appendix 1. Daily schedule for inpatient management of children with CRPS

7:00 a.m. – Good morning! Wake up, get ready for the day.
 7:45 a.m. – Breakfast in the dome (common area)
 8:00 a.m.–9:00 a.m. – Psychotherapy
 9:00 a.m.–10:00 a.m. – Occupational Therapy (OT)
 10:00 a.m.–11:00 a.m. – Physical Therapy (PT)
 11:00 a.m.–12:00 p.m. – PT group
 12:00 p.m.–1:00 p.m. – Lunch in the dome
 1:00 p.m.–1:30 p.m. – Patient practices OT exercises in his or her room
 1:30 p.m.–2:00 p.m. – OT
 2:00 p.m.–2:30 p.m. – PT
 2:30 p.m.–3:30 p.m. – Patient practices PT exercises in his or her room
 3:30 p.m.–4:30 p.m. – School Homework or Playroom
 Tuesday – Music therapy
 Thursday – Art therapy
 4:30 p.m.–5:00 p.m. – Patient practices OT exercises in his or her room
 5:00 p.m.–7:00 p.m. – Dinner in the dome
 7:00 p.m.–8:00 p.m. – School Homework
 8:00 p.m.–8:30 p.m. – Activity of your choice, out of bed.
 8:30 p.m. – Shower
 9:00 p.m. – Lights out. Goodnight!

Appendix 2. *Diagnostic and Statistical Manual of Mental Disorders* (4th ed., text rev.; DSM-IV-TR) criteria for diagnosis of anxiety and depression*Generalized Anxiety Disorder*

- A. Excessive anxiety and worry, occurring more days than not for at least 6 months, about a number of events or activities.
- B. Difficult to control the worry.
- C. Anxiety and worry are associated with three or more of the following symptoms:
1. Restlessness or feeling keyed up
 2. Being easily fatigued
 3. Difficulty concentrating or mind going blank
 4. Irritability
 5. Muscle tension
 6. Sleep disturbance
- D. Focus of anxiety and worry is not confined to features of an Axis I disorder, such as fears about having a panic attack, being embarrassed in public, being contaminated, being away from home, gaining weight, having multiple physical complaints or having a serious illness, and the anxiety and worry do not occur exclusively during Post-Traumatic Stress Disorder.
- E. Anxiety, worry or physical symptoms cause clinically significant distress or impairment in social, occupational or other important areas of functioning.
- F. The disturbance is not due to the direct physiological effects of a substance or a general medication condition and does not occur exclusively during a Mood Disorder, Psychotic Disorder or Pervasive Developmental Disorder.

Unspecified Anxiety Disorder

Includes disorder with prominent anxiety or phobic avoidance that do not meet criteria for any specific Anxiety Disorder, Adjustment Disorder with Anxiety or with Mixed Anxiety and Depressed Mood.

1. Mixed anxiety-depressive disorder: clinically significant symptoms of anxiety and depression but the criteria are not met for either a specific Mood Disorder or a specific Anxiety Disorder.
 2. Clinically significant social phobic symptoms that are related to the social impact of having a general medical condition or mental disorder.
 3. Situations in which the disturbance is severe enough to warrant a diagnosis of an Anxiety Disorder but the individual fails to report enough symptoms for the full criteria for any specific Anxiety Disorder to have been met.
 4. Situations in which the clinician has concluded that an Anxiety Disorder is present but is unable to determine whether it is primary, due to general medication condition, or substance induced.
-

(Continued)

Appendix 2. (Continued)*Depressive Disorder Not Otherwise Specified (NOS)*

Includes disorder with depressive features that do not meet criteria for Major Depressive Disorder, Dysthymic Disorder, Adjustment Disorder with Depressed mood or with Mixed Anxiety and Depressed Mood.

1. Premenstrual dysphoric disorder.
2. Minor depressive disorder: episodes of at least 2 weeks of depressive symptoms but with fewer than the five items required for Major Depressive Disorder.
3. Recurrent brief depressive disorder: depressive episodes lasting from 2 days up to 2 weeks, occurring at least once a month for 12 months.
4. Postpsychotic depressive disorder of Schizophrenia.
5. A Major Depressive Episode superimposed on Delusional, Psychotic NOS or the active phase of Schizophrenia.
6. Situations in which the clinician has concluded that a depressive disorder is present but is unable to determine whether it is primary, due to general medication condition, or substance induced.

Dysthymic Disorder

- A. Depressed mood for most of the day, for more days than not, as indicated either by subjective account or observation by others for at least 1 year in children and adolescents, 2 years in adults.
- B. Presence, while depressed, of two or more of the following:
 1. Poor appetite or overeating
 2. Insomnia or hypersomnia
 3. Low energy or fatigue
 4. Low self-esteem
 5. Poor concentration or difficulty making decisions
 6. Feelings of hopelessness
- C. During the 1 year of the disturbance, the person has never been without the symptoms in Criteria A or B for more than 2 months at a time.
- D. No Major Depressive Episode has been present during the first 2 years of the disturbance and is not better accounted for by chronic Major Depressive Disorder.
- E. There has never been a Manic Episode, a Mixed Episode or a Hypomanic Episode, and criteria have never been met for Cyclothymic Disorder.
- F. The disturbance does not occur exclusively during the course of a chronic Psychotic Disorder, such as Schizophrenia or Delusional Disorder.
- G. The symptoms are not due to the direct physiological effects of a substance or a general medical condition.
- H. The symptoms cause clinically significant distress or impairment in social, occupational or other important areas of functioning.