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Continuous Peripheral Nerve Blocks at Home for Treatment of Recurrent Complex Regional Pain Syndrome I in Children

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Background: **Recurrent complex regional pain syndrome I is not rare in the pediatric population. The authors conducted this study to evaluate the efficacy of continuous peripheral nerve blocks with elastomeric disposable pumps associated with initial Bier blocks for the treatment of recurrent complex regional pain syndrome I in children.**

Methods: **After parental informed consent, 13 children who did not respond to conventional complex regional pain syndrome treatment were included (mean age, 13 yr; range, 9–16 yr). After general anesthesia, peripheral nerve block was performed using 0.5 ml/kg lidocaine, 1%, with epinephrine and 0.5% ropivacaine injected in the peripheral nerve block catheter. Then, a 20-min Bier block was performed using a tourniquet and 0.2 ml/kg lidocaine, 1%; 3 ml/kg hydroxyethyl starch 130/06; and 5 mg/kg buflomedil injected intravenously. A solution of 0.1 ml · kg^{-1} · h^{-1} continuous ropivacaine, 0.2%, was infused through the catheter using an elastomeric pump for 96 h. Need for rescue analgesia, occurrence of side effects, and status of motor and sensory block were recorded at hours 1, 6, 12, 24, 48, 72, and 96. Children and parents completed a satisfaction assessment. All of the children had follow-up visits after 2 months.**

Results: **Postoperative analgesia was excellent. The median pain score was 0 for each period studied. Motor blockade was minimal before 12 h (median, 1) and absent thereafter. One child needed rescue analgesia. All children were able to walk easily after the initial 24-h period (walking score, > 4). Children and parents were all satisfied. Children returned home under parental surveillance beginning in the 24th hour. Neither peripheral nerve block nor Bier block caused side effects. After 2 months, none of the children exhibited any clinical symptom of recurrent complex regional pain syndrome.**

Conclusion: **Ambulatory continuous peripheral nerve block associated with an initial Bier block seems to be a significant and novel contribution to treat recurrent pediatric complex regional pain syndrome I. It allows complete pain relief, early mobilization, and rapid return home, representing a psychological advantage for these children.**

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COMPLEX regional pain syndrome (CRPS) type I is defined as a painful, disabling syndrome accompanied by edema and sudomotor and vasomotor dysfunction. Various treatments in adults have been reported, but no technique has shown superiority for treating or preventing CRPS I.[1] The reported methods include physical therapy, behavioral measures, and use of antidepressants, anticonvulsants, or transcutaneous electrical nerve stimulation. Other authors have proposed regional anesthesia with intravenous local anesthetics, guanethidine, reserpine, bretylium, nonsteroidal antiinflammatory drugs, droperidol or calcium-regulating drugs,[1] or blockade of sympathetic activity by epidural analgesia[2] or peripheral nerve blocks.[3–5]

Complex regional pain syndrome type I has also been described in a pediatric population.[6] The pathophysiology of CRPS I is not clear. A treatment using Bier block with local anesthetics,[7] physical therapy, and cognitive–behavioral treatment should be recommended. Unfortunately, recurrent episodes have been reported in 50% of children.[8] The median time to recurrence was 2 months with physical therapy alone.[8] One of the key elements of CRPS I treatment is intense physiotherapy. Intense exercise therapy has been proposed for initial treatment of childhood CRPS I,[8] but this treatment is very painful. The use of ambulatory continuous peripheral nerve blocks (CPNBs) to alleviate pain, allow intense physiotherapy, and treat CRPS I has not been prospectively studied in children with this syndrome.

Ambulatory elastomeric or electronic pumps have been often used at home for the postoperative period after orthopedic surgery in adults.[9–12] The use of such devices to restore optimal autonomy in children has recently been demonstrated in the postoperative period.[13]

The aim of the current study was to evaluate the feasibility and efficacy of CPNB at home by means of disposable elastomeric pumps associated with an initial Bier block in the treatment of recurrent CRPS I in children.

Materials and Methods

After we received institutional review board approval (Montpellier, France) and consent of each patient or family, 13 consecutive children who had upper or lower

limb CRPS I and who met the inclusion criteria were included in the study. All had either failed to respond to a 6-month regimen of physical therapy, cognitive–behavioral treatment, oral drugs (10 mg/night nortriptyline, 300 mg gabapentin three times a day), and transcutaneous electrical nerve stimulation or had subsequently experienced recurrence.

The children were premedicated with 0.4 mg/kg midazolam. Intraoperative general anesthesia was induced in all children using 3–5 mg/kg propofol and 1 μg/kg intravenous fentanyl.

Mechanical ventilation was used after tracheal intubation or insertion of a laryngeal mask airway. Anesthesia was maintained with 50% nitrous oxide in oxygen and a 2% inspired fraction of sevoflurane.

Peripheral nerve blocks were performed before the Bier block procedure using a nerve stimulator for precise nerve location (Stimuplex®; Braun, Melsungen, Germany). The landmarks described by Singelyn *et al.*[14] were used for popliteal blocks, and the landmarks described by Dalens *et al.*[15] were used for axillary blocks. With a starting output of 1.5 mA (frequency, 1 Hz; time, 50 ms), needles were advanced until they triggered the required muscle contraction. The position was judged adequate when muscle contractions were still elicited at 0.5 mA. A 20-gauge multiperforated catheter (Plexolong®; Pajunk, Geisingen, Germany) was inserted along the guide from 3 to 5 cm. Contrast medium (0.1 ml/kg Iopamidol 200; Shering Pharmaceutical, Lys-Lez-Lannoy, France) was injected into the catheter, and an anteroposterior radiograph of the region was obtained within 5 min. Children with unreliable catheter location were excluded. After catheter placement, 0.5 ml/kg of an equal-volume mixture of 0.5% ropivacaine and 1% lidocaine with 1:200,000 epinephrine was injected *via* the catheter over a 5-min period.

Then, a 20-min Bier block was performed. After exsanguination of the limb extremity and inflation of the tourniquet, 0.2 ml/kg lidocaine, 1%; 5 mg/kg buflomedil; and 3 ml/kg hydroxyethyl starch 130/06 were injected intravenously. Buflomedil is a peripheral vascular dilator and an anti-ischemic agent. This α1 and α2 adrenolytic drug has a relaxant effect on vascular smooth muscle increasing the arterial and venous blood flow with functional restoration of the microcirculation. Hydroxyethyl starch is used to expand the volume of lidocaine and buflomedil after limb exsanguination. The tourniquet was deflated after 20 min without complication, and general anesthesia was discontinued.

In the postoperative period, a continuous infusion of 0.2% ropivacaine was administered at 0.1 ml $\cdot$ kg^{-1} $\cdot$ h^{-1} using disposable elastomeric pumps (Infusor LV; Baxter Laboratories, Maurepas, France). Disposable multirate infusion pumps were used at a flow of 2, 3, 5 or 7 ml/h. A nonsteroidal antiinflammatory drug (40 mg/kg niflumic acid twice daily the first 48 h) was administered intravenously over a 15-min period to all children. In the postoperative period, sensory blockade was evaluated using cold sensation (ether on a ball of cotton wool) tested at hours 1, 6, 12, 24, 48, 72, and 96. At the same time, motor blockade was evaluated for the selected muscles of each nerve territory using a Bromage scale.[16] Nurses evaluated postoperative pain in the surgical ward at hours 1, 6, 12, and 24 and at home at hours 48, 72, and 96 with a visual analog scale ranging from 0 (no pain) to 10 cm (most imaginable pain), as well as amounts of rescue analgesia. If pain control was considered insufficient (visual analog scale score $>$ 3) in the postoperative period, an intravenous injection of 0.2 mg/kg nalbuphine in the first 24 h and a codeine–acetaminophen tablet at home were administered as rescue analgesia. At 24 h, a walking score was noted in all children ranging from 0 (rest in bed) to 5 (complete free walk). If there was no complication, the patient returned home under parental surveillance after 24 h. Intense exercises (elbow flexion and extension [1 patient], ankle movements [12 patients] against resistance: plantar flexion, eversion, dorsal flexion, proprioceptive physiotherapy, and massages) conducted by a physiotherapist were started 1 h after the continuous peripheral nerve block induction and at 6, 12, and 24 h at the hospital and twice daily at home. At the end of the study period, satisfaction levels of children and parents were recorded (unsatisfied, satisfied, or very satisfied). Adverse effects (pruritus, nausea, vomiting, dysesthesia, hematoma, urinary retention, local infection, or local anesthetic toxicity) were noted. All of the children had a follow-up visit after 2 months, at which time absence or severity of pain, walking disorders, and disabling symptoms were recorded.

Statistical analysis was performed using SAS software version 8.02 (SAS Institute, Cary, NC). The quantitative anthropometric scores were expressed as median (range), motor blockade was expressed as median (10th–90th percentiles), and pain scores were expressed as median (25th–75th percentiles and range). Repeated-measures analysis of variance was used for continuous variables. Comparisons between values at the times studied were made using the Mann–Whitney U test for nonparametric data, and the chi-square test was used for categorical data. A significance threshold of $P < 0.05$ was retained.

Results

Thirteen patients, four boys and nine girls, were included in this study. The median age was 13 (9–16) yr and the median weight was 57 (26–95) kg. Information on the clinical course of the children before we performed the continuous peripheral nerve blocks is shown in table 1. Twelve popliteal nerve blocks and one axillary nerve block were performed. No block failures were

Table 1. Information on the Clinical Course of the Children before Treatment with Continuous Peripheral Nerve Blocks

Patient No.	Age, yr	VAS Score before PNB	Site of Injury	Type of Injury	Duration of Conservative Treatment	Treatment before Block	Cognitive Behavioral Treatment	Physical Therapy	Disabling Syndrome
1	15	10	Left ankle	Sprain	6 months	Amitriptyline clonazepam, calcitonin	Yes	Yes	Alodynia, hyperesthesia edema, vasomotor disorder
2	11	10	Right ankle	Trauma	6 months	Amitriptyline paracetamol, tramadol	Yes	Yes	Alodynia, hyperesthesia edema
3	13	10	Left ankle	Sprain	6 months	Amitriptyline paracetamol, paroxetine	Yes	Yes	Alodynia, hyperesthesia edema
4	12	10	Wrist right	Trauma	6 months	Calcitonin, amitriptyline	Yes	Yes	Hyperesthesia edema vasomotor disorder
5	13	10	Right ankle	Sprain	7 months	Amitriptyline clonazepam, tramadol, calcitonin	Yes	Yes	Alodynia, hyperesthesia edema
6	12	8	Left ankle	Sprain	6 months	Clonazepam, amitriptyline	Yes	Yes	Hyperesthesia edema
7	9	9	Left ankle	Trauma	7 months	Clonazepam, amitriptyline, calcitonin	Yes	Yes	Alodynia, hyperesthesia edema
8	14	10	Right ankle	Trauma	6 months	Amitriptyline paracetamol, paroxetine, tramadol	Yes	Yes	Alodynia, hyperesthesia vasomotor disorder
9	13	10	Right ankle	Sprain	6 months	Clonazepam, amitriptyline, calcitonin	Yes	Yes	Alodynia, hyperesthesia edema
10	16	10	Left ankle	Sprain	8 months	Clonazepam, amitriptyline, calcitonin, tramadol	Yes	Yes	Alodynia, hyperesthesia edema, vasomotor disorder
11	13	8	Left ankle	Sprain	6 months	Clonazepam, amitriptyline	Yes	Yes	Hyperesthesia edema
12	14	10	Right ankle	Trauma	8 months	Clonazepam, amitriptyline, calcitonin	Yes	Yes	Alodynia, hyperesthesia vasomotor disorder
13	15	9	Right ankle	Sprain	6 months	Clonazepam, amitriptyline, calcitonin	Yes	Yes	Alodynia, hyperesthesia edema

PNB = peripheral nerve block; VAS = visual analog scale.

noted. Confirmation of correct location of all catheters was obtained with contrast medium radiographs.

Postoperative analgesia was excellent. The median pain score was 0 for each period studied. Pain during physiotherapy (visual analog scale values) is reported in figure 1. Rescue analgesia was necessary in only one patient, whose pain scores were outside the 25th–75th percentile range. This child had previously been treated with oral morphine. This patient received two injections of intravenous nalbuphine during the first 24 h of the study.

Observed motor blockade was minimal before 12 h (median value, 1) and absent thereafter (fig. 2). The 13 children in the study were able to walk easily after a 24-h period (walking score, > 4).

All parents and children reported high satisfaction (both parents and child were satisfied in one case, and in the other cases, both parents and children were very satisfied) with the pain management at rest and during all periods of intense physiotherapy throughout the study.

None of the children had associated hematoma or catheter infection. No dysesthesia, urinary retention, or any other sign of local anesthetic toxicity was noted. No neurologic symptom was noted after withdrawal of the disposable pumps by a nurse in the surgical ward at 96 h. There was no accidental removal of the catheter during the study.

After 2 months, no child presented any clinical symptom of recurrent CRPS, and all of the patients were able

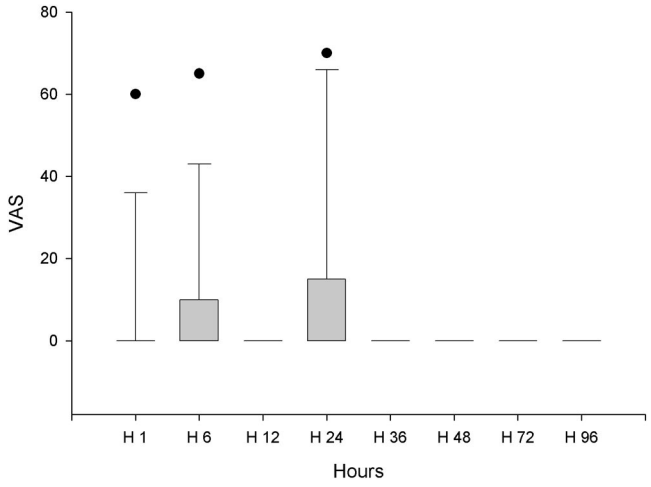

Fig. 1. Visual analog scale (VAS) values during physiotherapy. The *box* represents the 25th–75th percentiles; the *dark line* is the median. The *extended bars* represent the 10th–90th percentiles, and the *dark circles* represent values outside this range.

to resume normal activity. During the period of recruitment of these 13 patients, a total of 37 children were referred to us for CRPS I.

Discussion

This study shows that ambulatory CPNB associated with an initial Bier block is effective against intractable or recurrent CRPS I in children. This treatment provided complete short-term pain relief and allowed early intense mobilization and a rapid return home.

The absence of a comparative control group with saline in the CPNB or without CPNB merits comment. Despite recent case reports,[7] many cases of recurrent CRPS I have been reported in the literature in children treated with serial Bier blocks.[17] In case of recurrent CRPS I, the poor success rate of isolated Bier blocks is already established.[1,17] Furthermore, our ethics committee ruled against a randomized comparative design in children in view of the pain levels anticipated during intensive physiotherapy.

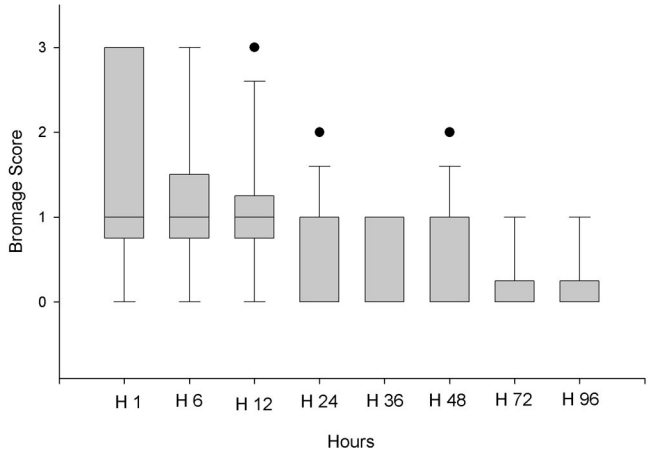

Fig. 2. Course of motor block values during the studied period. At each period, the *vertical lines* represent the 25th–75th percentiles, and the *box* is the median value.

In adults, few studies have shown short- and long-term benefits of peripheral nerve blocks in the treatment of CRPS I.[2,3] Recently, several case reports have emphasized the importance of continuous or repeated peripheral nerve blocks[4,5,18] for control of acute pain and improvement of CRPS I treatment. Because recurrent CRPS I remains a therapeutic challenge in pediatric patients,[19] we evaluated the use of 4-day continuous peripheral nerve blocks with disposable elastomeric pumps. We chose ambulatory pumps because of their documented efficacy, simplicity, and safety for ambulatory postoperative analgesia in adult outpatients.[9–12] These devices permit optimal pain relief and rapid restoration of a child's autonomy in the postoperative period.[13] Although repeated sympathetic blocks have been shown to be effective against the pain associated with CRPS I in adults, especially in recurrent cases,[20] repeated intravenous guanethidine injections do not improve long-term results regarding the treatment or prevention of CRPS I.[1,17,21] Furthermore, intravenous guanethidine has been associated with many adverse effects, including nausea, vomiting, orthostatic hypotension, dizziness, diarrhea, and weakness.[17] We used a buflomedil–lidocaine mixture to reduce these adverse effects during the initial Bier block and adjuvant 4-day continuous peripheral nerve block, which provided complete pain relief at rest and during exercise. The excellent control of pain achieved in this manner facilitates the other two aspects of CRPS treatment: physiotherapy and psychotherapy.[5,7,22]

Intense exercise therapy is necessary to treat childhood CRPS effectively and to decrease the high incidence of recurrence reported in patients treated using only physical therapy and cognitive–behavioral treatment.[8,19] Continuous nerve blocks are recommended primarily to reduce pain and facilitate intensive physical therapy and functional rehabilitation,[5,22] which are essential goals of CRPS I treatment.[22,23] Because physiotherapy is very painful during CRPS I and CPNB is indicated during postoperative mobilization in children,[13] we used CPNB for physiotherapy in these children. Pain relief was complete in our patients, and intense physiotherapy was fully applied. Continuous epidural analgesia allows the same level of pain management for physical therapy. Nevertheless, continuous peripheral nerve block should be easier at home than is continuous epidural analgesia.

Kotiniemi *et al.*[24] emphasized the usefulness of postoperative pain control and play activities influenced by the hospital environment for behavioral modification in children. The authors recommended encouraging such play to help children cope with the challenging experience of hospitalization. The use of disposable pumps permitting rapid discharge of children would certainly

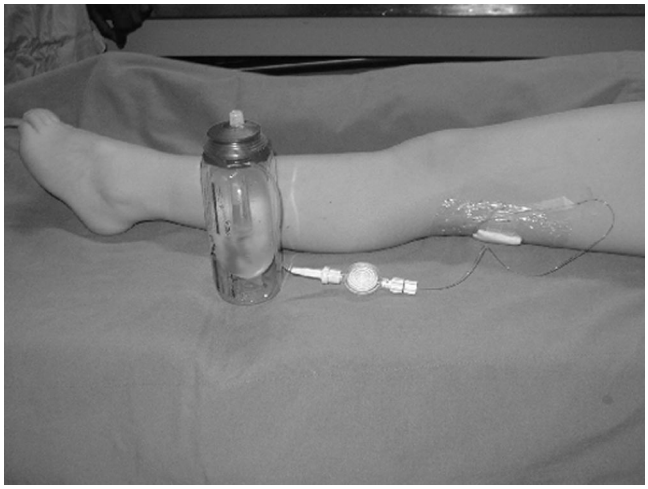

Fig. 3. Continuous infusion of 0.2% ropivacaine *via* a popliteal catheter with a disposable elastomeric pump in a 12-yr-old child.

reduce the need for behavioral modification training designed to improve tolerance of extended hospitalization (fig. 3). Moreover, although cognitive behavioral theory has documented efficacy against chronic pain,[25] the value of cognitive–behavioral psychotherapy has not been fully demonstrated in patients with CRPS I.[22]

In conclusion, this study shows that 4-day CPNB associated with initial Bier block is effective against intractable and recurrent CRPS I in children. A relevant decrease in short-term pain was observed in our population of children. This reduction in pain facilitated physiotherapy and cognitive–behavioral treatment for CRPS I. The use of disposable pumps permitted reduced hospital stay and continuation of this treatment at home. However, if ethically possible, further randomized controlled studies with a greater number of patients may be necessary to establish the usefulness of this double postganglionic sympathetic blockade definitively in children or adults.

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