CASE REPORT

Complex Regional Pain Syndrome after Transradial Cardiac Catheterization

Chih-Jou Lai[1], Chen-Liang Chou[1,2]*, Tcho-Jen Liu[1,2], Rai-Chi Chan[1,2]

[1]Department of Physical Medicine and Rehabilitation, Taipei Veterans General Hospital, and [2]National Yang-Ming University School of Medicine, Taipei, Taiwan, R.O.C.

Complex regional pain syndrome (CRPS) is a disease with unclear pathophysiology. The condition is characterized by pain, soft tissue change, vasomotor change, and even psychosocial disturbance. It may affect the upper more than the lower extremities, and the distal more than the proximal. The trigger factors include carpal tunnel release, Dupuytren's repair, tendon release procedures, knee surgery, crush injury, ankle arthrodesis, amputation, and hip arthroplasty. Rarely, it has been associated with stroke, mastectomy, pregnancy, and osteogenesis imperfecta. Herein, we present a rare case of a patient who was diagnosed with CRPS after transradial cardiac catheterization. CRPS was first diagnosed due to hand swelling, allodynia, paresthesia, and the limited range of motion of interphalangeal, metacarpophalangeal, and wrist joints, with the preceding factor of transradial cardiac catheterization, and was then confirmed by a three-phase bone scan. After intensive physical therapy with hydrotherapy, manual soft tissue release, and occupational therapy for the hand function, there was much improvement in range of motion and hand function. There was no allodynia or painful sensation in the follow-up. After training, the functional status of this patient was adequate for daily activity. [*J Chin Med Assoc* 2006;69(4):179–183]

Key Words: cardiac catheterization, complex regional pain syndrome, reflex sympathetic dystrophy

Introduction

Reflex sympathetic syndrome, a complex regional pain syndrome, is a disease with unclear pathophysiology. Several mechanisms have been proposed: an exaggerated inflammatory process, sympathetic dysfunction, and both central and peripheral nervous system dysfunction. Early in 1985, Roberts[1] proposed that the trauma response produces a long-term sensitization of a widely dynamic range of neurons and results in painful sensations. He called this disease "reflex sympathetic dystrophy (RSD)". In recent years, the concepts and taxonomy of the disease have changed. Now, an umbrella term "complex regional pain syndrome" (CRPS) is used for these syndromes. The term "complex" is used to denote the varied clinical phenomena; the "regional" distribution of the findings is a hallmark of these disorders; and "pain" is the cardinal symptom for this grouping.[2] This disease typically occurs following physical trauma with continuous pain. The symptoms and signs include allodynia, numbness, paresthesia, hyperalgesia, vasomotor and pseudomotor dysfunction, dystonic posture, myoclonic jerks, trophic changes such as nail and skin atrophy, edema, skin color change, temperature change, and contractures and limited range of motion. The disease more frequently affects the upper rather than the lower extremities and is more distal than proximal.[3,4] Changes similar to CRPS may appear in patients with myocardial infarction, after local cold injury, and after revascularization of ischemic extremities.[3–5]

There are 2 types of CRPS. The sole differentiating criterion between CRPS type I and CRPS type II is the presence of a known nerve injury. Herein, we present a rare case that was diagnosed as CRPS after transradial cardiac catheterization.

*Correspondence to: Dr. Chen-Liang Chou, Department of Physical and Rehabilitation Medicine, Taipei Veterans General Hospital, 201, Section 2, Shih-Pai Road, Taipei 112, Taiwan, R.O.C.
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Case Report

A 73-year-old male patient was referred to our outpatient department because of persistent right-hand pain and stiffness months after cardiac catheterization. The patient had retired from his job some years prior and denied any trauma before the disease onset. He had a history of hypertension for 2 years. He denied diabetes mellitus or other medical disease. He denied alcohol drinking, but had been a heavy smoker, smoking 1 pack per day for 40 years; he had quit 2 years previously. Three months prior to admission, he had a near syncope attack in the early morning when walking, which was his routine daily exercise. Cold sweating, left anterior chest pain with radiation to the left upper arm, left-side neck, and cheek were noted. The pain persisted, and he was transferred to the emergency room. Under the impression of myocardial infarction, primary percutaneous transluminal coronary angioplasty (PTCA) was arranged. After premedication with heparin and aspirin, the procedure was performed smoothly using a right-side transradial approach with a 6F catheter and a 3.0 × 33 Penta stent was inserted. However, there was poor re-flow (thrombolysis in myocardial infarction grade flow 1, TIMI-1) after stenting and intra-aortic balloon pump (IABP) support was given. After the procedure, the patient underwent heparinization and was transferred to the intensive care unit. A secondary look was arranged 2 days later. This time, a left radial artery approach with a 6F catheter was used. The result was good without re-stenosis at the stenting site (TIMI-3). However, right-hand numbness and pain were noted since the first procedure. The pain was persistent and burning in nondermatomal distribution, from the distal palmar crease to the tips of the fingers, including both dorsal and palmar aspects; both the radial and ulnar sides of the dorsum of the forearm to olecranon process; and finally the pain ascended to the anterior-lateral arm. A limited range of motion of the right distal interphalangeal joint, proximal interphalangeal joint, metacarpophalangeal joint, and wrist were noted. Swelling of the fingers and hand was also reported. The patient asked for medical help again 2 months later because of intractable pain. An X-ray of the hand revealed no evidence of fracture or joint dislocation, but there was soft tissue swelling. Plethysmography led to a suspicion of right forearm deep vein thrombosis. The patient received a low-molecular-weight heparin injection. However, there was no obvious improvement. Electromyography (EMG) and nerve conduction study (NCS) examination diagnosed a right-side median nerve injury (Tables 1 and 2). The clinical symptoms persisted for the following 3 months. Then, muscle atrophy in the thenar eminence intrinsic hand muscle and dorsal interossei was noted. Because of the changes, the patient was limited in his daily activities and needed assistance. Thus, he was transferred to our hospital for further evaluation and management.

Table 1. Needle electromyography: there was active denervation of the right-side abductor pollicis brevis muscle

Muscle	Rest		Motor unit potential			Maximal effort
	Fib	PSW	Amp (mV)	Dur (ms)	Poly	
R APB	+	+	0.2–2	5–15	–	Decreased
R ADQ	–	–	0.2–2	5–15	–	Decreased
R EIP	–	–	0.2–2	5–15	–	Decreased
R PT	–	–	0.2–2	5–15	–	Decreased

APB = abductor pollicis brevis; ADQ = abductor digiti minimi; EIP = extensor indicis proprius; PT = pronator teres; R = right; Fib = fibrillation; PSW = positive sharp wave; Amp = amplitude; Dur = duration; Poly = polyphasic wave.

Table 2. Nerve conduction study: there was prolonged latency in the right median and ulnar nerve

Muscle	Latency (ms)	Conduction velocity (m/s)	Amplitude (V)
Median M	9.3	45.1	1.3m
Ulnar M	7.6	52.1	6.8m
Ulnar F	32.8		170U
Ulnar S	3.9	56.0	13.5m
Radial S	3.8	43.8	3.1U

Median = median motor nerve; Ulnar M = ulnar motor nerve; Ulnar S = ulnar sensory nerve; Radial S = radial sensory nerve; Ulnar F = ulnar F response.

Physical examination at the outpatient clinic revealed a mildly edematous right forearm without evidence of increased sweating. There was no change in nail or hair growth. Stiffness and decreased range of motion of the right wrist and metacarpophalangeal and interphalangeal joints were noted. There was no decrease in the muscle power of the upper right arm, and there was no change in deep tendon reflex. The pulsation of the right radial artery was slightly weak as compared with the left side. Due to the above history, CRPS was highly suspected. An X-ray examination of the hand was done and revealed a mild degree of osteoarthritic change with osteoporosis of the right hand (Figure 1). Three-phase bone scan showed increased blood pooling in the right hand and forearm in the early phase, particularly in the dorsum, wrist, and small joints (Figure 2); the delayed bone scan showed a relatively increased uptake on the right side and in the right wrist, and a mild increase in the small joints of the right hand. These results confirmed CRPS in this patient.

Upper extremity hydrotherapy 3 times a week was initiated immediately. After each section of hydrotherapy, the physical therapist used a manual soft tissue release. We also prescribed occupational therapy for hand function and fine motor training, with emphasis on self-care and activities of daily living. At the 5-month follow-up, there was much improvement in the range of motion of the wrist, metacarpophalangeal, and inter-phalangeal joints. There was neither allodynia nor painful sensation. The edema had decreased, but there was still cold on the affected side. The patient could dress by himself and use a spoon in his right hand for eating. He could hold a pen and write well.

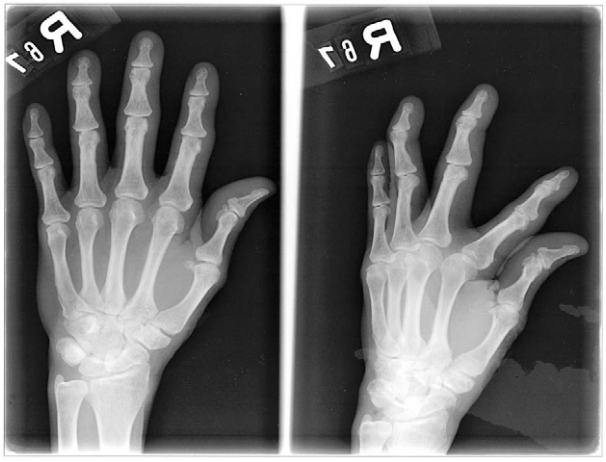

Figure 1. An X-ray of the hand revealed no evidence of fracture or joint dislocation, but there was soft tissue swelling.

Discussion

The diagnosis of CRPS is based on clinical history and symptoms. The International Association for the Study of Pain published the standardized diagnostic criteria of CRPS in 1994.[6] Studies have revealed the high sensitivity but poor specificity of these criteria.[7,8] Bruehl et al[7] and Harden et al[8] in 1999 proposed modifications to the criteria to improve their specificity. Investigations including temperature change by thermography, osteoporosis, cortical erosion in plain radiographs,[9] and increased uptake throughout the blood flow in the three-phase bone scan suggest, but do not diagnose, the disease.[10] Nowadays, the three-phase bone scan is used as the gold standard for diagnosis, and even as a predictor for the risk of developing CRPS.[10–12]

We suspected CRPS in this case based on the clinical presentation, and confirmed the diagnosis by three-phase bone scan. The response to conservative physical therapy in this case was good. Pain subsided weeks after hydrotherapy, thus we did not prescribe more aggressive therapy, such as steroid or calcitonin injection, a sympathetic block procedure, or newer

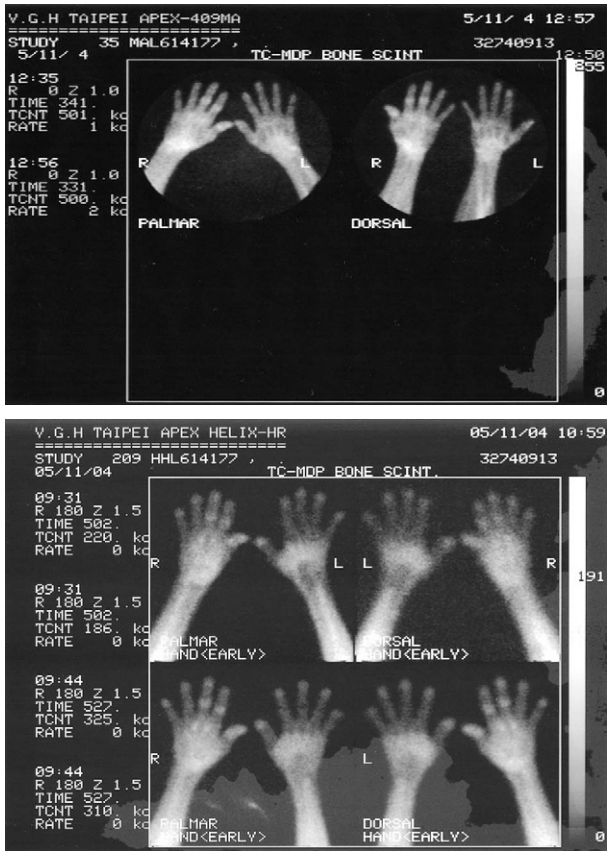

Figure 2. Three-phase bone scan showed increased blood pooling in the right hand and forearm in the early phase, particularly in the dorsum, wrist, and small joint.

techniques such as spinal cord stimulation.[13–19] After training, daily activity was compensated by left hand use. Although the fine motor activity of the right hand was still limited, the functional status of this patient was much improved and was adequate for the activities of daily life.

According to Veldman et al,[4] the most common causes of CRPS were trauma (65%), postsurgery sequela (19%), and no clear precipitant (10%). CRPS after vascular injury such as arterial blood gas sampling,[20] arteriovenous graft placement,[21] and thromboendarterectomy[22] has been reported. The first case of CRPS after a percutaneous vascular procedure was reported by Inoue et al[23] in 2000. In that case, catheterization was done by brachial artery access. In 2002, Papadimos and Hofmann[24] presented another case with CRPS following transradial cardiac catheterization. This case had radial artery thrombosis, without peripheral nerve injury, which was related to the patient's retirement from his job.[24] The Doppler examination in this case was arranged 3 months after symptom onset and the patient had received intravenous heparin initially. The examination of both hands showed no difference in wave-form and pulsation. There was no direct evidence that artery thrombosis had contributed to the disease.

In past studies, although rarely, CRPS has been reported after myocardial infarction,[25] but it occurred on the left side. In our case, the affected side was the right. Therefore, CRPS due to myocardial infarction was less likely.

This case raises a number of questions. First, which type of CRPS was this? As mentioned previously, the sole difference between the 2 types of CRPS is the presence of nerve damage. This case was initially found with median and ulnar nerve injuries at wrist level found in our first investigation. The distribution of pain in this case was out of the area of the median and ulnar nerves, which is compatible with our understanding of CRPS type II. After 7 months of follow-up, there was still numbness in the median nerve area. The NCS/EMG study found active denervation of both the ulnar and median nerves. Since CRPS type II is relatively rare, it is difficult to say whether this was a case of CPRS type I with coincident nerve injury, or CRPS type II presenting with a nerve injury due to compression after cardiac catheterization.

Second, what was the stage of the CRPS in this case? Clinically, the patient had clawing hands and an atrophy stage was suspected. Osteoporosis in the plain radiographs was compatible with the clinical stage. However, an increased uptake in the peri-articular area of the hand was noted on the bone scan, which led to suspicion of an acute stage. This may explain the good prognosis, although a late phase was suspected clinically. A subacute stage, which is the transition between the acute and the atrophy stages, may be a better explanation.

Finally, how could we estimate the effect of this disease on everyday life? Or how to estimate the impairments and disabilities resulting from CRPS? Oerlemans et al[26] used a Rabdoud skills questionnaire, and later reported an impairment level sumscore.[27] As stated in Schasfoort et al's[28] later review, we need to develop objective outcome measures at the level of disabilities and handicaps. Later in 2003, Zyluk[29] proposed another scoring system to evaluate severity of CRPS. However, as Loeser[30] commented, it is difficult to standardize the outcomes of chronic pain. Pain itself is a subjective rather than objective phenomenon. Some studies discuss other objective aspects, such as upper limb activity monitoring, as outcome measures.[31] Further discussion regarding a standard method is needed.

Conclusion

The case presented here alerts us that CRPS is one of the possible complications of cardiac catheterization. Physicians should choose this approach carefully, especially in the dominant hand, because daily activity may be impaired.

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