CASE REPORT

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Complex Regional Pain Syndrome Following Arterial Thromboembolism: A Rare Case Report

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ABSTRACT Complex regional pain syndrome (CRPS) is a painful clinical condition that usually appears in the distal extremity and is characterized by sensory, motor, autonomic, and trophic symptoms initiated by a trigger stimulus. The disorder is divided into two as Type 1 and Type 2 CRPS. Unlike Type 1 CRPS, peripheral nerve damage is observed in Type 2 CRPS. The etiology of CRPS often includes causes such as trauma, fracture, surgical intervention, immobilization, and stroke. The diagnosis of CRPS is usually made clinically, based on the anamnesis and examination findings of the patient. Success in treatment depends on early diagnosis and the prevention of complications. In this report, a rare case of Type 1 CRPS due to radial and ulnar artery thrombosis is presented.

Keywords: Complex regional pain syndrome Type 1; physical therapy; radial and ulnar artery thrombosis

Complex regional pain syndrome (CRPS) is a clinical disorder that is usually initiated by a trigger stimulus, involves distal extremities, and is characterized by sensory, motor, autonomic, and trophic symptoms that do not show a clear dermatomal and peripheral nerve distribution. The disorder is divided into two types as Type 1 and Type 2 CRPS. Unlike Type 1 CRPS, Type 2 CRPS manifests itself with peripheral nerve damage. The etiology of CRPS often includes causes such as trauma, fracture, surgical intervention, immobilization, and stroke. The diagnosis is made clinically based on the anamnesis and examination findings of the patient. However, if necessary, imaging methods may be used to support the diagnosis. The chance of success in treatment and in the prevention of complications is high with early diagnosis.[1,2] In this report, a rare case of Type 1 CRPS due to radial and ulnar artery thrombosis is presented.

CASE REPORT

A sixty-seven-year-old female patient applied to our outpatient clinic with complaints of severe pain, swelling, and limited movement in the right hand, which have lasted for about three months. The patient had undergone three nasal polyp excisions, in addition to cardioectomy and coronary angiography. The drugs used by the patient were acetylsalicylic acid 100 mg 1*1/day, clopidogrel 75 mg 1*1/day, nifedipine 30 mg 2*1/day, metoprolol 50 mg 1*1/day, and pantoprazole 40 mg 1*1/day. The patient said that the acetylsalicylic acid 100 mg drug, which she had used five days before the nasal polyp operation was discontinued and that she applied to the emergency room with complaints of bruising and pain in her right forearm three days after the operation. The epicrises of the patient were examined and the imaging methods were obtained with the past examination findings. There was no pulse in the right radial artery. As a result of right arterial Doppler ultrasonography (USG) imaging, a thrombus was observed in the radial and ulnar arteries, while the right venous Doppler USG imaging result was normal. We found out that the incision was made from the antecubital region by a cardiovascular surgeon, and that an embolectomy

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was performed first (Figure 1). However, when this intervention was unsuccessful, thrombectomy was performed by angiography, the occluded vessel was opened, and the acetylsalicylic acid was discharged with medication recommendations of 100 mg 1*1/day and clopidogrel 75 mg 1*1/day. The musculoskeletal system examination of the patient revealed restrictions in her right wrist movements and pain at the beginning of the range of motion in all directions while the fingers were flexed. She had severe pain in her right hand, in addition to allodynia, hyperesthesia, edema, mild redness, and radiance (Figure 2). The circulatory system examination of the patient revealed that four extremity peripheral pulses were palpable and rhythmic. Her visual analog scale (VAS) score was 8/10 at rest and 10/10 during activity. Duruoz Hand Index, the total score was 79 before treatment.[3] The lab results were as follows: vitamin D total (25 OH) 7 ng/mL, erythrocyte sedimentation rate 40 mm/h, and C-reactive protein 17.5 g/L. She had no other abnormal findings. On the anteroposterior hand-wrist radiographs, diffuse osteoporosis was observed in the right hand (Figure 3). The follow-up

FIGURE 1: The incision line starting from the antecubital region of the right upper extremity of the patient.

FIGURE 2: Pretreatment of complex regional pain syndrome Type 1 that occurs after a thrombus in the right radial and ulnar arteries. The patient had swelling in the right hand, slight brightness and color change, and restriction of movement in the fingers.

FIGURE 3: Anteroposterior wrist radiographs of the patient (diffuse osteoporosis is observed in the bones of the right hand and wrist).

arterial Doppler USG confirmed the normal blood flow in the radial and ulnar arteries of the right arm. Considering that the patient's complaints had started after radial and ulnar artery thrombosis, the patient was diagnosed as Type 1 CRPS using the Budapest criteria (Table 1).[4,5] Informed consent form was taken from the patient, and she was admitted to the physical therapy and rehabilitation program in our hospital. For the severe pain, pregabalin was started at 75 mg 1*1/day, and the dose was gradually increased to 150 mg 2*1/day. As the total vitamin D (25 OH) value was low, vitamin D_3 replacement therapy was administered at a dose of 50,000 IU/week for eight weeks. A physical therapy and rehabilitation program including active and passive motion exercises, stretching exercises, isometric strengthening exer-

TABLE 1: The Budapest diagnostic criteria for complex regional pain syndrome.[4,5]

1. Continuing pain, which is disproportionate to any inciting event.
2. At least one symptom in ≥3 of the following categories:
Sensory: hyperesthesia and/or allodynia
Vasomotor: temperature asymmetry and/or skin color changes and/or skin color asymmetry
Sudomotor/edema: edema and/or sweating changes and/or sweating asymmetry
Motor/trophic: decreased range of motion and/or motor dysfunction (weakness, tremor, dystonia) and/or trophic changes (hair, nail, skin)
3. At least one finding at time of evaluation in ≥2 of the following categories:
Sensory: hyperalgesia (to pinprick) and/or allodynia (to light touch and/or deep somatic pressure and/or joint movement)
Vasomotor: temperature asymmetry and/or skin color changes and/or asymmetry
Sudomotor/edema: edema and/or sweating changes and/or sweating asymmetry
Motor/trophic: evidence of decreased range of motion and/or motor dysfunction
(weakness, tremor, dystonia) and/or trophic changes (hair, nail, skin)
4. There is no other diagnosis that better explains the signs and symptoms.

cises, the application of discrete ultrasound on the stellar ganglia (3 watts/cm^2, 5 minutes), pneumatic compression hot-cold contrast application, and 15 sessions of mirror therapy was administered. In the follow-up examination, the joint movements of the right wrist were and open in all directions, the first four fingers were in neutral posture, while the fifth finger was in the semi-flexion posture. dema, slight redness, brightness, allodynia and hyperesthesia decreased in the patient's right hand (Figure 4). The VAS score was 2/10 at rest and 4/10 during activity. Duruoz Hand Index, the total score after the treatment was 29.

FIGURE 4: Post-treatment view of complex regional pain syndrome Type 1 that occurs after a thrombus in the right radial and ulnar arteries. Swelling, slight brightness, and discoloration, in addition to restriction of movement, in the right hand of the patient have regressed.

DISCUSSION

CRPS is a disorder characterized by sensory, motor, autonomic, and trophic symptoms that are mostly initiated by a trigger stimulus and generally lack a clear dermatomal and peripheral nerve distribution.[1] Symptoms suggesting CRPS were first described in soldiers who were injured during the war and developed nerve damage in 1864.[6] Many causes can be counted in the etiology such as trauma and fractures, neurological events, postoperative and iatrogenic causes, intrathoracic events, vascular and musculoskeletal system diseases, drugs, neoplasms and paraneoplastic causes.[7] In the literature, a case with radial artery occlusion as a result of compression applied after transradial catheterization was accompanied by Type 1 CRPS.[8] Baillet et al. diagnosed a 77-year-old male patient with Type 1 CRPS with clinical and radiological findings after performing thromboendarterectomy in the right popliteal artery.[9] A case thought to have developed CRPS due to poor compression after a transbrachial catheter administration has also been reported.[10] Following transradial catheter administration, pain, burning, and weakness in the hands, as revealed by electromyography, and nerve damage were detected in two patients and a diagnosis of Type 2 CRPS was made using imaging methods.[11,12] In our case, Doppler USG examination revealed a thrombus in the right radial and ulnar arteries, while related complaints had occurred before surgery of the right forearm. In accordance with the complaints and examination findings of the patient, a diagnosis of Type 1 CRPS due to

thrombus in the right radial and ulnar arteries was made in accordance with the Budapest criteria. We used only direct radiography as the imaging method.

It is very important to diagnose and start a rehabilitation program in early stages of CRPS in terms of success of treatment and prevention of permanent sequels. The aim of treatment is to reduce edema and pain, increase muscle strength and provide joint range of motion, and to ensure that the person is independent in daily life activities. Patients are applied methods such as TENS, interferential flow, such as analgesia, contrast bath, whirlpool bath, external pneumatic compression, mirror therapy acupuncture and massage.[1,13] Occupational therapy promotes the use of the affected limb in daily life activities. The use of customized clothing or dressings can reduce the affected limb edema and sensory overload. As medical treatment, NSAIDs, corticosteroids, bisphosphonates, calcitonin, vitamin C, opioids, free radical scavengers, anticonvulsants such as gabepentin and pregabalin, sympatholytic drugs are recommended.[13-15] In cases where pain does not decrease despite all these applications, various invasive methods can be applied such as sympathetic blockage, surgical sympathectomy, spinal cord stimulation, intrathecal baclofen and even amputation.[1,2] As a result, it should be taken into consideration by the physicians that complaints such as pain, edema, and loss of function occurring in the arteries feeding the extremities may occur due to thrombus, as well as they may occur due to CRPS.

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Conflict of Interest

No conflicts of interest between the authors and / or family members of the scientific and medical committee members or members of the potential conflicts of interest, counseling, expertise, working conditions, share holding and similar situations in any firm.

Authorship Contributions

Idea/Concept: *Ebru Erden, Nebahat Sezer;* ***Design:*** *Ebru Erden, Ender Erden;* ***Control/Supervision:*** *Ebru Erden, Ender Erden, Nebahat Sezer;* ***Data Collection and/or Processing:*** *Ebru Erden, Ender Erden, Nebahat Sezer;* ***Analysis and/or Interpretation:*** *Ebru Erden, Nebahat Sezer;* ***Literature Review:*** *Ebru Erden, Ender Erden;* ***Writing the Article:*** *Ebru Erden;* ***Critical Review:*** *Nebahat Sezer;* ***References and Fundings:*** *Ebru Erden, Ender Erden;* ***Materials:*** *Ebru Erden, Ender Erden.*

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