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Complex Regional Pain Syndrome and Spontaneous Pain Following Simple Incision With Immobilization: A Case Report

LETTER TO THE EDITOR

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Complex regional pain syndrome (CRPS) is characterized by vasomotor and sudomotor changes. Its etiology generally includes trauma, fracture, stroke, and coronary disease. It is evaluated in two subtypes: CRPS type I (reflex sympathetic dystrophy), presenting with complex regional pain syndromes accompanied by spontaneous pain, hyperalgesia, allodynia, edema, autonomic anomalies, and trophic changes; regional pain occurring after minor injuries and fractures in the extremities; and coexisting vasomotor, sudomotor changes, and sensorial changes, and CRPS type II (coxalgia), including major peripheral nerve injuries, in addition to all of these symptoms (1, 2). Although the pathogenesis of CRPS is not exactly known, peripheral and central sensitization, which causes neurogenic inflammation, is held responsible. The most possible mechanism is considered to be increased sensitivity against catecholamine, depending on sympathetic denervation, in addition to increased numbers or sensitivity of peripheral axonal adrenoreceptors. Moreover, some mechanisms, such as the release of neuropeptides from primary nociceptive afferents and sympathetic efferents [substance P, calcitonin gene-related peptide (CGRP) and neuropeptide Y] and the development of neurogenic inflammation, are suggested. This results clinically in hyperalgesia and central sensitization, in which allodynia develops (3, 4).

CASE REPORT

A 50-year-old female patient had developed a 4-cm transverse cut on the metatarsus profile of her plantar with an edged object 2 months and 10 days ago. She had been able to walk only on her heel for 2 months. She presented with complaints of sudden spontaneous pain and tenderness in the foot lasting for 10 days to the Department of Physical Therapy and Rehabilitation, Kayseri Education and Research Hospital. In the examination performed at the admission, she defined spontaneous pain in the foot. She had an antalgic gait. Moreover, she had too much tenderness around her metatarsus. The inversion and eversion of her ankle were painful, but the movements of the joints in all directions were normal. No swelling, edema, changed skin color, or atrophy was observed. She did not define neuropathic pain. The score on the visual analog scale was 6, and the score on the DN4 scale was 0. Routine laboratory results of the patient were normal. Lateral radiography of the foot revealed spotty osteoporosis covering all tarsal bones, the metatarsus, and phalangeal bones in the right foot (Figure 1, 2). In the 3-phase bone scintigraphy, the right ankle was found to be consistent with increased activity involvement in the regions of the tarsal bones and first metatarsal phalangeal joints (Figure 3). Based on the presence of CRPS findings in the scintigraphy and X-ray images and clinical findings, the patient was diagnosed with CRPS according to the International Association for the Study of Pain (IASP) criteria (Table 1), and then, her treatment was planned. The patient was given physiotherapy in 15 sessions. Her post-treatment pain VAS score decreased to 3, while it was 6 before the treatment. Apparent recovery was observed in the clinical picture of the patient.

Main clinical findings of CRPS include spontaneous or stimulated pain, autonomic changes, motor dysfunction, and trophic changes. It is generally accepted that patients with CRPS pass through 3 phases, including acute, dystrophic, and atrophic phases. However, it has been recently suggested that the disease actually does not progress in three phases, and the time and course of clinical findings can differ, depending on the individual. In a study analyzing 113 cases, Bruehl et al. (5) classified CRPS into three probable subtypes. They defined type 1 as the type in which vasomotor symptoms and signs were observed more often compared to pain and sensorial symptoms; type 2 as the type in which pain and sensorial symptoms were at the forefront; and type 3 as typical reflex sympathetic dystrophy (RSD). In our case, pain was dominant. CRPS type 1 can be confused with various diseases, owing

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Table 1. The 1994 IASP criteria for the diagnosis of CRPS.

CRPS Diagnostic Criteria
1. Presence of a stimulating painful event without (CRPS type I) or with nerve injury (CRPS type II) or immobilization
2. Spontaneous pain that is out of proportion to the original event or allodynia/hyperalgesia (not restricted to only a single peripheral nerve region)
3. Edema, changed blood flow in the skin, or abnormal sudomotor activity in the region of pain in any time
4. Absence of any other condition that can explain these symptoms

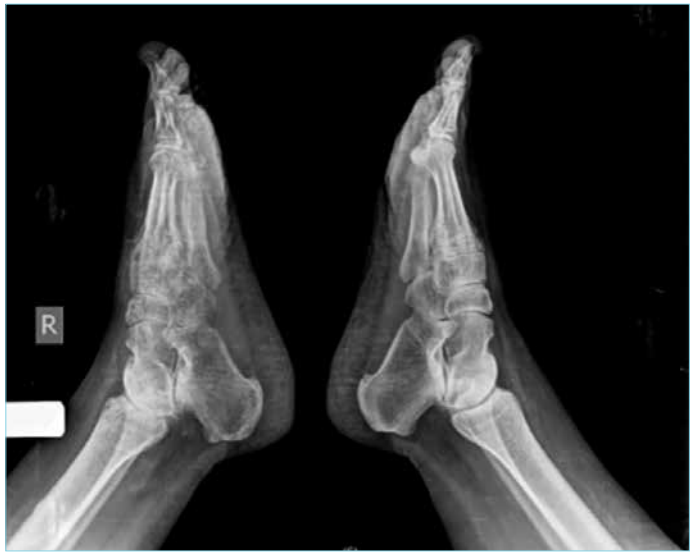

Figure 1. **Lateral radiography of the foot: spotty osteoporosis**

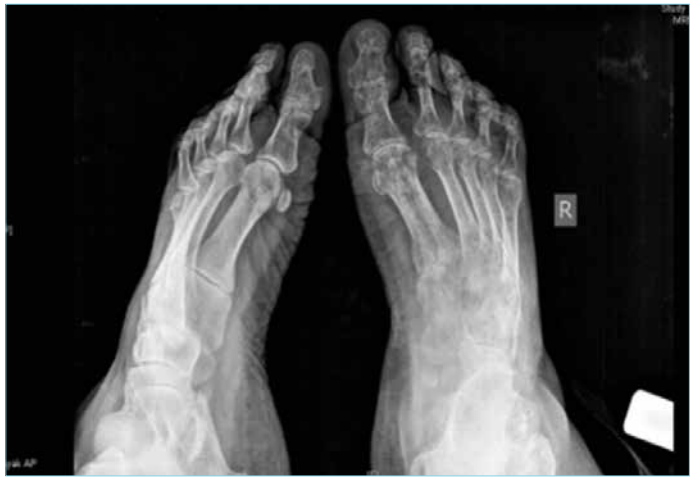

Figure 2. **AP radiography of the foot: spotty osteoporosis**

to the variability of its clinical findings and atypical forms. While performing a differential diagnosis, chronic arterial insufficiency, Raynaud's disease, thromboembolism, infection, cellulitis, osteomyelitis, septic arthritis, erythromelalgia, trauma/fracture, anxiety neurosis, tabes dorsalis, osteonecrosis, cerebrovascular diseases, and psychosomatic disorders should be taken into consideration, as well as inflammatory arthritis (6). In our case, these factors were ruled out, due to a history of immobilization after trauma; normal results of routine laboratory examinations; and the absence of atrophy, swelling, and changed skin color.

Direct radiography, which is used as a diagnosis method for CRPS, may not reveal any radiological findings, but homogeneous or vitreous bone demineralization and spotty osteoporosis may develop in a few weeks (7, 8). Spotty osteoporosis is not specific to CRPS. It is observed in 30%-80% of cases (9). It is encountered in a metaphyseal localization (10). Spotty osteoporosis was also available in our case (Figure 1, 2).

Moreover, three-phase radionuclide bone scintigraphy is another important diagnostic technique for CRPS. Compared to direct radiography, it provides results earlier. The involvement of radionuclide after intravenous injection in seconds (phase 1= arterial phase), minutes (phase II= soft tissue phase), and hours (phase III= mineral phase) is evaluated. The results of increased focal, multifocal, or diffuse activity are obtained. In an examination having a diagnostic value for CRPS, increased activity must be observed in the radiocarpal, intertarsal, and tarsometatarsal joints; metatarsal regions; and juxta-articular regions of the fingers in the late phase. Periarticular thickening is a characteristic finding. The signs of hyperfixation or isofixation and hypofixation can be detected in the area involved. For the method to be effective in diagnosing CRPS, apparent periarticular involvement must be observed in the affected extremity joints in phase III (11). However, it should be remembered that increased activity is not specific to CRPS (12, 13). Scintigraphic findings should be evaluated with clinical criteria. In our case, the right ankle was consistent with increased activity involvement in the regions of the tarsal bones and first metatarsal phalangeal joints in all phases of three-phase bone scintigraphy (Figure 3).

CRPS is seen at a rate of 65% in the distal parts of the extremities exposed to traumas, such as bone fractures, postoperative conditions, contusion, and sprains and strains. It can rarely be encountered in central nervous system damage, including cardiac ischemia, spinal cord injury, and cerebrovascular events. Pain generally occurs in the distal part of the affected extremity spontaneously, and it is described as a burning sensation. It is characterized as extremely intense and out of proportion to the original painful stimulus. The symptoms of mechanical and thermal hyperalgesia and/or allodynia appear. These sensorial abnormalities are usually observed in the distal parts in the early phase, and they are not restricted to the areas of the affected nerve distribution and lesion. Pain typically occurs with movement and pressure over the joints (hyperalgesia from deep somatic tissue) (14). The pain of our case was also out of proportion to the stimulus and not restricted to the areas of nerve distribution or lesion. It was even in the proximal of the lesion in the foot sole.

Kıralp et al. (15) reported a rate of CRPS developing after soft issue injuries of 8.5% in their study. As in our study, it has been proven that immobilization after trauma also plays an important

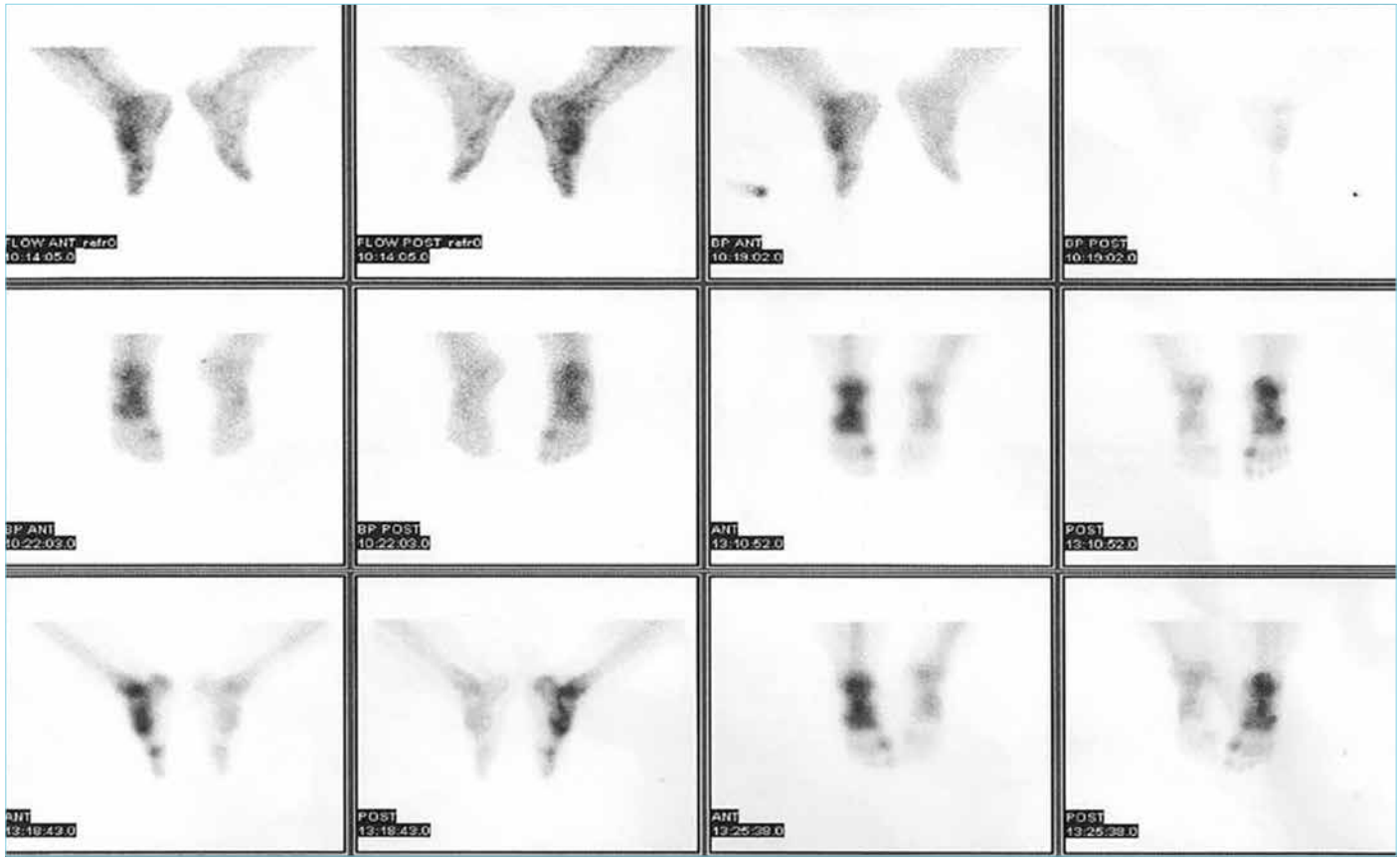

Figure 3. Three-phase bone scintigraphy: increased activity involvement in every phase

role in the formation of CRPS, as well as trauma itself (16). Immobilization having developed after a simple cut injury, which lasted for 2 months, was remarkable in our study.

Complex regional pain syndrome can develop with extremity immobilization after a small and superficial cut injury, which should be considered for patients presenting with the complaint of spontaneous pain.

Informed Consent: Written informed consent was obtained from parents/patient who participated in this study.

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Authors' Contributions: Conceived and designed the experiments or case: HS, HTÇ, STS. Performed the experiments or case: HS, HTÇ, STS. Analyzed the data: HS, HTÇ, STS. Wrote the paper: HS, HTÇ, STS. All authors have read and approved the final manuscript.

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