Case Report

Bilateral Complex Regional Pain Syndrome in a Woman with Major Depressive Disorder

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Abstract

Complex regional pain syndrome type 1 (CRPS1) refers to a disorder usually caused by trauma; it is characterized by pain, swelling, limited range of motion, vasomotor instability, skin changes, joint stiffness, and patchy bone demineralization. Most often it occurs after trauma. Other etiologies include myocardial ischemia, cerebrovascular accidents, infection and emotional stress.

We report a case of bilateral CRPS1 of the upper extremities in a 52-year-old woman suffering from major depressive disorder. She was complaining about her hands' stiffness and pain. She also reported swelling of both upper extremities and anhydrosis, thickening of the skin and muscle wasting, finger movement limitation, contracture of the digits and trophic skin changes. The diagnosis of CRPS1 was suspected, according to history, physical examination, radiographic changes and bone scintigraphy.

Keywords: Bilateral, complex regional pain syndrome (CRPS1), depression

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Introduction

Complex Regional Pain Syndrome type 1 (CRPS1) is a complication arising after trauma or surgery, although spontaneous development has been reported. The pathophysiology of CRPS1 is unknown but psychological factors have been suggested to play a role, even though their influence is not clear.[1] Other etiologies include myocardial ischemia, cerebrovascular accident, viral or infectious disorder, medication reaction, tumors, and amputation.[2,3] The overall incidence rate of CRPS1 in the general population based on a retrospective cohort study was 26.2 per 100,000 person years (95% CI: 23 – 29.7). Females were affected more than males (Ratio: 3.4).[4] CRPS1 is characterized by pain, swelling, limited range of motion, vasomotor instability, skin changes, joint stiffness and patchy bone demineralization which is often limited to the extremities.[2,5]

Here we report a case of bilateral CRPS1 of the upper extremities in a woman with major depressive disorder, an unusual underlying cause.

Case Report

On December 2012, a 52-year-old woman was admitted in the Rheumatology ward of Imam Khomeini Hospital Complex with complaint of pain in both hands along with stiffness. There was a history of swelling of both upper extremities and burning pain from 10 months before her admission. After two months of burning pain, she had experienced stiffness, anhydrosis, thickening of the skin and muscle wasting in both hands. One month later, symptoms had progressed and the joint motion was also limited. Finally contracture of the digits occurred and trophic skin changes appeared (Figure 1).

She did not have any history of arthritis, morning stiffness or Raynaud's phenomenon and no history of smoking, alcohol consumption or drug abuse was reported. Significant physical examination was anhydrosis, thickening of the skin, muscle wasting, and complete limitation of motion in both hands. Examination of all joints was normal. Laboratory findings are presented in Table 1.

Mammography, abdomino-pelvic CT scan, transvaginal ultrasonography and chest radiography were normal. No identifiable etiology was found in her past medical history except mood changes after her grandson's death. The tragedy had happened about nine months before beginning of her symptoms.

The psychiatric consultation also confirmed a major depressive and obsessive compulsive disorder; treatment with Lorazepam 1 mg/day and Serteralin 25 mg/day was recommended. Radiography of hands showed soft tissue swelling, severe periarticular osteoporosis and patchy demineralization (Figure 2). Three phase bone scintigraphy showed increased uptake in the late phase in both hands (Figure3).

Regarding her presentation, physical examination, radiographic changes and bone scintigraphy results, CRPS1 was suspected. The patient was treated with Prednisolone (15 mg daily), Propranolol (40 mg daily), and physical therapy. Mood disorder was treated with Lorazepam 1 mg/day and Serteralin 25 mg/day, as recommended.

Significant improvement was observed after six months of treatment. Hand stiffness was ameliorated dramatically and she could make a fist, although not completely.

Discussion

We report a case of bilateral CRPS1 of the upper extremities in a

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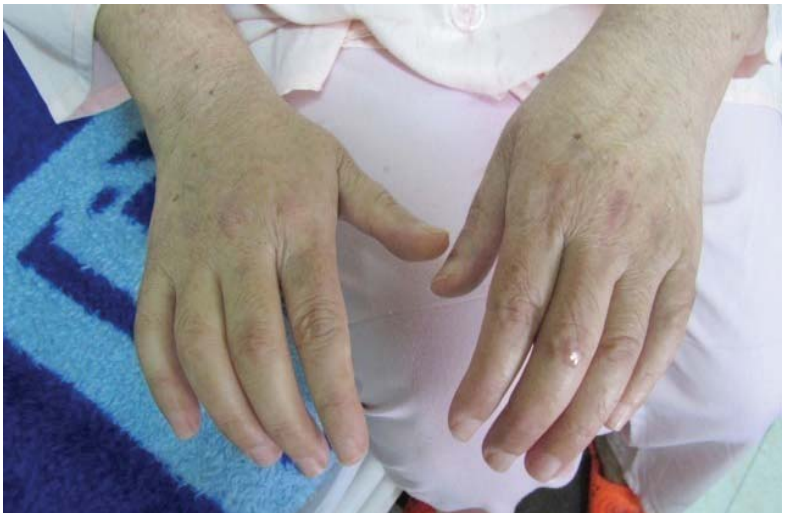

Figure 1. Represents the hands changes of the patient in admission

Table 1. Represents the laboratory findings of the patient after her admission

WBC = 6800 (4000–10000)	Cr = 0.6 mg/dL (0.7–1.3)	Alp = 178 IU/L (44–147)
Hg = 13.5 g/dL (12–16)	Ca = 8.7 mg/dL (8.5–10.2)	LDH = 364 IU/L (105–333)
MCV = 90.5 mg/dL (80-99)	ph = 4.8 mg/dL (2.4–4.1)	RF = 7.6 (-) IU/mL (< 20)
Plt = 224000 (150000–400000)	Na = 143meq/L (135–145)	ANA = 1 (-) IU/ml (< 10)
ESR = 12 mm/h (< 20)	K = 3.7meq/L (3.5–5)	Anti ccp = 0.4 (-) EU (< 20)
CRP = 2 mg/L (< 10)	AST = 15 IU/L (10–34)	Anti centromerAbIgG (-)
CPK = 44 mcg/L (10–120)	ALT = 11 IU/L (10–40)	Anti SCL 70 (-)

WBC: White Blood Cell, Hg: Hemoglobine, MCV: Mean Corpuscular Volume, Plt: platelet, ESR: Erythrocyte Sedimentation rate, CRP: C Reactive protein, CPK: Creatine phosphokinase, Cr: Creatinine, Ca: Calcium, Ph: Phosphorous, AST: Aspartate Aminotransferase, ALT: Alanine Aminotransferase, ALP: Alkaline Phosphatase, LDH: Lactate dehydrogenase, RF: Rheumatoid Factor, ANA: Antinuclear Antibody, Anti CCP: Anti-citrullinated protein antibody, Anti SCL70: Anti-topoisomerase 1 antibody.

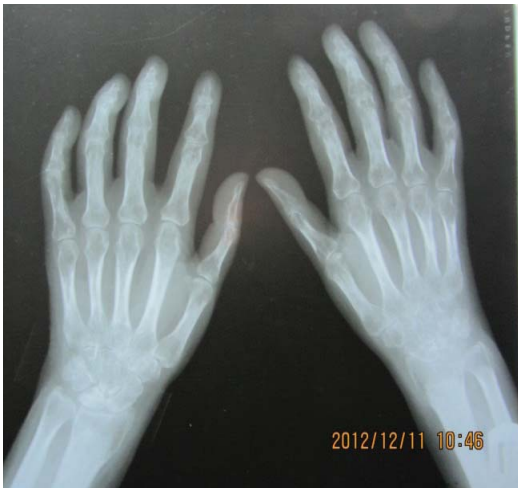

Figure 2. Represents the hands radiography of the patient

52-year-old woman suffering from major depressive disorder. We searched the literature and there was not any similar case report.

Minor trauma, fracture, myocardial infarction, cerebrovascular disease, primary central nervous system disorder, viral or infectious disorder, medication reaction (anti-tuberculosis drugs), tumors, amputation syndromes[2,3] and emotional stress[1] are considered as etiologies for CRPS1.

Some researchers such as Beerthuizen, et al. (2009) did not find any relationship between CRPS1 and psychological factors. Only the association of life events were found;[1] although the conclusions of other reviews were contradictory. However, some studies have shown the relationship between depression and CRPS1.[6,7] The etiology of CRPS1 in the above patient was investigated. A major depressive disorder was the only underlying cause which was found.

Clinical presentations of CRPS1 include hyperalgesia, allodynia, edema, vascular and psudomotor instability, as well as motor and trophic changes.[3,8] Our patient also showed similar symptoms, which were compatible with CRPS1.

Diagnostic approach to CRPS1 is based on the patient's history and clinical manifestations.[9] Other techniques such as radiography imaging, Magnetic Resonance Imaging (MRI) and the three-phase bone scintigraphy[10] can support the clinical diagnosis. Abnormalities related to CRPS1 in a three-phase bone scintigra-

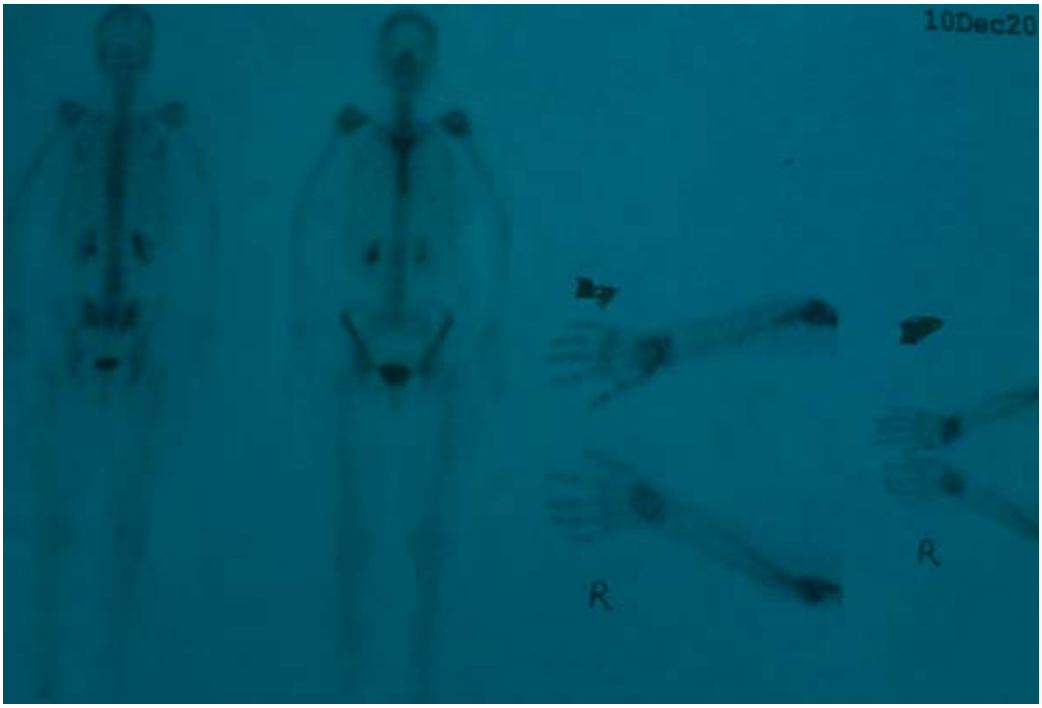

Figure legend 3. Represents the hands scintigraghy of the patient

phy are increased in blood flow and blood pool uptake with delay images showing increased uptake in a periarticular pattern. Even though the bone scan increases diagnostic sensitivity, a normal bone scan does not rule out the diagnosis of CRPS1.[8]

In the presented case, radiography of the hands (soft tissue swelling, severe periarticular osteoporosis, and patchy demineralization) and three-phase bone scintigraphy results (increased uptake in the late phase) along with her symptoms, all were compatible with the diagnosis of CRPS1 (Figures 2 and 3).

The interesting point in the patient was bilateral involvement of both hands and the underlying etiology, which was the mood disorder.

We should emphasize that the most important aspects of CRPS1 management are early diagnosis, investigation for underlying disorders and treatment of the disease. Early diagnosis and treatment of the CRPS1 can prevent irreversible changes, joint contracture, stiffness and osteoporosis. Physical therapy and treatment of the underlying disease (depression in our patient) is the mainstay of the CRPS1 treatment.[11] Propranolol, Corticosteroids, Calcium channel blockers, Antidepressant agents[12] Gabapentin, Alendronate, intravenous Pamidronate, repetitive Transcranial Magnetic Stimulation (rTMS), low dose intravenous ketamine, topical transdermal Isosorbide Dinitrate and physical therapy[13] have been used to treat CRPS1.

Our patient was treated with Prednisolone, Propranolol, physical therapy and mood stabilizers. Considerable improvement was seen after six months of treatment, but she couldn't make a complete fist due to delayed treatment and chronicity.

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