

# Foot-ankle involvement of complex regional pain syndrome associated with pregnancy

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## ABSTRACT

Complex regional pain syndrome (CRPS) is a condition which develops after a painful incident and characterized by allodynia/hyperalgesia, edema, skin anomalies and anomaly in blood flow and abnormal sudomotor activities independent of the precipitating incident. It is a rare condition in pregnancy and difficult to distinguish from pelvis and lower extremity pains which are inherent in pregnancy. Hips are typically involved, symmetrical involvement of feet and ankles are rarely reported; has a benign course however it is important to diagnose and treat due to fracture risk. Herein, we have presented a 28-year-old patient who came to us in week 32 of her pregnancy and gave birth in week 36 and diagnosed CRPS with bilateral involvement of foot and ankle.

**Keywords:** Pregnancy, complex regional pain syndrome, symmetrical foot-ankle involvement

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As well as complex regional pain syndrome (CRPS) can occur idiopathically it usually develops after a painful event such as trauma, fracture, in addition, central nervous system diseases, medications, hemiplegia can cause the situation. Pathophysiology has not been fully elucidated. Central and peripheral mechanisms are influential. It is associated with sympathetic cutaneous vasoconstrictor activity, allodynia/hyperalgesia, edema, impaired skin blood flow and abnormal sudomotor activity. The complex is a chronic painful condition. The diagnosis is made after the other causes of pain and dysfunctioning are eliminated [1, 2].

CRPS is classified as type 1 and type 2. In CRPS type 1 there is no specific pathology; in type 2 there is nerve damage that is called causalgia, and it is associated with trauma [3].

## CASE PRESENTATION

A 28-year-old patient with 32 weeks of gestation presented to our outpatient clinic for the complaint of gait abnormality with pain, swelling on both ankles and feet. The patient did not have any systemic disease or trauma in the past and had no alcohol use and no smoking in the history. There was no family history of diseases with musculoskeletal involvement. The patient was pregnant for the third time and in previous pregnancies she gave birth to healthy babies after full-term gestations. In this pregnancy there also was no pathology related to the baby.

There was swelling in both ankles especially remarkable in the left ankle, also increased temperature, sweating, and light cyanosis was found during the physical examination, peripheral pulse was



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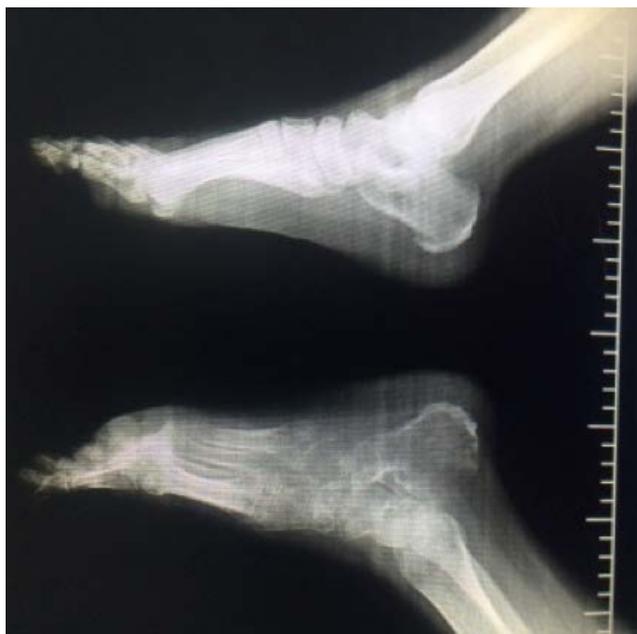
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regular, and there wasn't any neurological deficit. Ankle range of motions (ROMs) were painful during both the active and passive movements and were restricted due to pain. The pain of the foot was increasing when pressure applied and decreased while resting; the patient's ambulation was possible with a wheelchair.

The patient was 32 weeks pregnant when applied to the orthopedic outpatient clinic previously, and ankle strain was considered. For the local treatment, cold application and 5% ibuprofen cream were recommended and Paracetamol 500 mg twice daily were given orally for the pain relief. Routine blood tests were performed at the outpatient clinic to exclude rheumatological diseases. Blood biochemistry, calcium, sedimentation, CRP values were normal; vitamin D was not measured. The patient refused to undergo through the radiological examination. The previous treatment suggested to be continued and rest was recommended. An appointment was set to repeat the examination and checkup. However, the patient did not come to the appointment. During the pregnancy, the patient did not receive any supportive treatment such as supplementary vitamins especially Vitamin D, iron or calcium.

The patient presented in the second week after



**Figure 1.** X-ray study shows speckled osteoporosis in the calcaneus and tarsal bones at both ankles.

delivery. The baby was delivered by cesarean-section under general anesthesia in week 36 and was 3200 kg and healthy. The patient could ambulate with the help of two crutches; edema in her feet was partially regressed however ROMs were found to be painful and tender during the examination; an X-ray and routine blood tests were performed again.

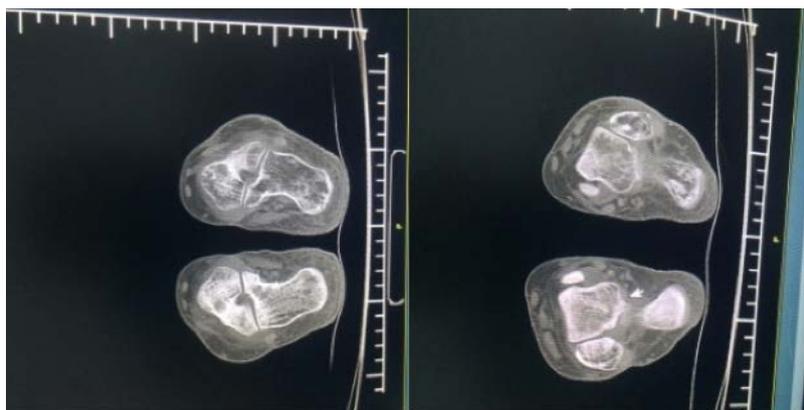
The following results were found in the laboratory tests: Hemoglobin: 11.9 g/dL (12-18), leukocyte: 10.41 mm<sup>3</sup> (4.60-10.2), alkaline phosphatase: 122 U/L (50-136), cholesterol: 416 mg/dl (0-200), triglyceride: 156 (0-150), CRP: 1.27 mg/dL (0.0-0.8), sedimentation rate: 43 mm/hour (0-15 mm/hour), total calcium: 9.2 mg/dL (8.5-10.5), parathormone: 156 pg/ml (15-65), and vitamin D3: 4.4 ng/mL (11-43). In the X-ray examination of both ankles; speckled osteoporosis was present in the calcaneus and tarsal bones (Figure 1).

MRI imaging could not be performed since the patient was claustrophobic. In the computed tomography, widespread speckled osteoporotic deformation in calcanea areas and thickening in the trabecular area on both feet but predominantly on the left foot were seen (Figure 2).

Three-phase bone scintigraphy could not be taken hence the patient wanted to breastfeed her baby and did not want to stay away from the baby because of the radioactivity even for a short time. The patient was diagnosed with CRPS type 1 based on the clinical and radiographic findings.

The patient was recommended to rest at home and not to apply pressure on feet. The medical treatment was given appropriately for breastfeeding. Prednisolone 16 mg/day (to be gradually decreased and stopped in two weeks), calcium 1200 mg and vitamin D 1000 IU/day were initiated. Physiotherapy modalities such as contrast bath, in water ultrasound (5 min, 1.5 w/cm<sup>2</sup>), TENS were applied. Passive ROM, active ROM, active-assisted ROM exercises, targeting the ankle were started for the beginning; these exercises were followed by gradual stretching and strengthening, stress loading. The patient was recommended to repeat these short-term exercises at home twice a day.

After receiving twenty sessions (for 4 weeks every week-day) the patient started to walk without crutches, pain was regressed, and her complaints were almost entirely gone at the end of the first month.



**Figure 1.** Computed tomographic scan shows widespread speckled osteoporotic deformation in calcanea areas and thickening in the trabecular area on both feet.

## DISCUSSION

Nonspecific pain edema and difficulty in walking during in the lower extremity and pelvic region during pregnancy is often confronted due to mechanical and inflammatory causes, and differential diagnosis is difficult and most times a definite diagnosis cannot be made [4]. Among mechanical reasons, ligamentous laxity, sacroiliac joint dysfunction are the frequently encountered pathologies.

Pregnancy induced CRPS is very rare. It was first described by Curtiss and Kincaid [5] in 1959. In 1968 Lequesne [6] described three cases in the same pathology and named them transient osteoporosis or neurotrophic rheumatism, in the further considerations those situations are named as reflex sympathetic dystrophy, algoneurodystrophy and Sudek's atrophy. The most recent terminology change was made in 1993 by the International Association for the Study of Pain (IASP) as 'Complex Regional Pain Syndrome' [7].

In a study structured with disease analysis of nine cases retrospectively and a review of the literature (57 patients and 159 sites of reflex sympathetic dystrophy) by Poncelata *et al.* [8], it was concluded that CRPS in pregnancy was seen in different parts of the body; and the rates were as follows; 88% in hips, 25% in knees and 21% in ankles.

CRPS is frequently seen in the third trimester of pregnancy and the involvement can be bilateral [4, 8, 9]. In our case, there was bilateral involvement of the ankles in the third trimester was accompanied by severe pain, edema, increased temperature, sweating,

tenderness and restriction in ROM. Also there was no underlying reason that can cause CRPS therefore the patient was evaluated as CRPS type 1.

CRPS is an entirely clinical situation and diagnosis is made according to the Veldman diagnostic criteria (Table 1) [10].

When seen in pregnancy, CRPS generally follows a benign course and can regress spontaneously so that many cases can heal without being diagnosed however with the diagnosis and treatment regression can occur faster and fracture which is the most common complication can be prevented.

There are various theories about the pathophysiology of CRPS during pregnancy; the pressure on the obturator nerve applied by the baby's head is thought to trigger the autonomic dysfunction, however, this does not provide sufficient evidence for CRPS in the first trimester. The increasing weight of both the mother and the baby during pregnancy and lordosis disrupt the microcirculation of the extremities, hence microtrauma is repetitive, it stimulates autonomic system, spasm occurs in the arterial and precapillary sphincters and the back-flow in capillaries cause passive dilatation; this mechanism is an accepted hypothesis for the occurrence of CRPS; another hypothesis is the unexplained increase in bone turnover and osteoclastic activity [3, 4, 11, 12].

In another theory, the hormonal changes during pregnancy are thought to be the cause, and the postpartum regression of CRPS was shown as the evidence. PTH, 1,25 dihydroxycholecalciferol and calcium balance are claimed to impair. Also in our case, PTH was increased, and serum vitamin D level

**Table 1.** Veldman diagnostic criteria for CRPS

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**Criteria 1:** Positivity of the four symptoms out of the five criteria below supports the diagnosis.

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1- Pain

2- Temperature difference when compared to the other extremity

3- Volume asymmetry when compared to the other extremity

4- Color asymmetry when compared to the other extremity.

5- Limitation in the active joint ROM

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**Criteria 2:** Occurrence or progression of these symptoms during or after exercise

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**Criteria 3:** Symptoms spreading to a wider area after the initiation in the first trauma region.

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was significantly decreased; the calcium levels were in normal range [13].

Hypertriglyceridaemia is a condition that can be seen in pregnancy and increases in the third trimester; in the study of Ponceleta *et al.* [8], triglyceride levels of the patients with CRPS were found to be high and in the study conducted by Acquaviva *et al.* [11] on 765 CRPS patient, especially the involvement of the hip joints were reported with the accompanying hypertriglyceridaemia. Additional inflammatory conditions, cellulitis, arthritis, DVT, vascular disorder, bone tumors and other malignities should be considered for the diagnosis; radiography is helpful however hence the use of radiography is limited during the pregnancy period and the condition can only be detected in 3-6 weeks after the onset, MRI is a preferable method for the diagnosis. Similarly, bone scintigraphy cannot be used during pregnancy [4, 14]. After seeing the postpartum X-ray imaging of our patient, the patient was diagnosed with CRPS based on the clinical findings. Speckled osteoporosis was confirmed and no bone fractures were seen in the computed tomography. MRI scanning and scintigraphy could not be performed hence the patient had claustrophobia and did not give consent for the scintigraphic evaluation. The CRPS treatment guideline published in 1998 is based on three principles; rehabilitation, pain management and physiotherapy. Later, in addition to these basic principles, functional rehabilitation has been updated with self-management techniques and new treatments [15, 16].

When CRPS is detected during pregnancy, symptomatic treatment is applied; physiotherapy modalities, pressure-free cold press elevation of the

feet, passive and active mobilization exercises can be recommended; postpartum use of calcitonin, beta-blockers, antiepileptics, tricyclic antidepressants, NMDA receptor agonists, opioids, tramadol, corticosteroids, griseofulvin, nifedipine, baclofen are in the medical treatment options and if necessary sympathetic blockage can be applied. Calcitonin is not recommended during pregnancy, and the lactation period, IV pamidronate can be given to breastfeeding mothers hence its safe during the breastfeeding period. Pamidronate passes into breast milk in negligible amount, the biological half-life is short and when complexed with calcium it can not be absorbed from the gastrointestinal tract of the baby [12, 14, 17].

For the treatment Prednol, 16 mg/day (to be gradually decreased and stopped in two weeks) and calcium 1200 mg and vitamin D 1000 IU/day were initiated on our patient who wanted to breastfeed her baby. Physiotherapy modalities and exercises were given. One month later, the patient stopped using the crutches and started to walk without aid, and her pain was almost absent.

CRPS should be taken into account for lower extremity pain in pregnancy, although rarely there is a fracture risk, for this reason, it should be treated with early diagnosis. CRPS can also regress with symptomatic treatment during the pregnancy period. Postpartum aggressive treatment can be given but the treatment should be managed cautiously on mothers who want to breastfeed their babies.

## CONCLUSION

Pain in the lower extremity often occurs during

pregnancy. CRPS is a rare cause, and when diagnosed the treatment is easy. Symptomatic treatment can prevent a fracture that can occur as a complication. In pregnancy, this diagnosis should be kept in mind to avoid misdiagnosis and malpractice/wrong treatment.

### *Informed consent*

Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

### *Conflict of interest*

The authors declared that there are no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

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