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Case Report

Complex Regional Pain Syndrome, an Important Differential Diagnosis in Sports Injuries: a Case Report

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Abstract

Background: Complex regional pain syndrome (CRPS) is a disproportionate and persistent, regional pain related to a minor trauma. Although CRPS is not an infrequent condition its pathophysiology remains unknown and leading to underdiagnosis or late diagnosis. The diagnosis is clinical, according to Budapest criteria of the International Association for the Study of Pain. Bone scintigram is the most effective test to support the diagnosis. The aim of this article is to discuss the importance of clinical suspicion for an early CRPS diagnosis in a sprain's young athlete clinical case. **The Case:** We present the case of a sixteen-year-old male patient with no medical history who suffered two minor ankle injuries in the right foot. The patient developed severe and persistent pain associated with vasomotor, sudomotor and trophic abnormalities. He remained undiagnosed for 10 months until CRPS diagnosis confirmation supported by a bone scintigram. He received multiple treatments until spontaneous remission in the fourth year of evolution. **Discussion:** CRPS poses a diagnostic challenge that requires early suspicion to improve treatment outcomes and prognosis. Maintaining a high index of clinical suspicion is crucial, and CRPS should be considered in the evaluation of any persistent pain sport-related injury. Despite extensive research on CRPS conducted in recent decades, this condition may still be unfamiliar to many healthcare providers. Increasing awareness of CRPS among medical professionals can facilitate timely and accurate diagnosis, which is essential for effective management.

Introduction

Complex regional pain syndrome (CRPS) is defined as an array of painful conditions characterized by a disproportionate continuing regional pain.[1] The incidence varies widely between 5 to 26 per 100,000 inhabitants.[2] It is caused mainly after a local trauma, most frequently a fracture or sprain.[3] In the last two decades, CRPS research has been largely conducted. However, the pathophysiology mechanism is not yet fully understood.[4]

The diagnosis is clinical by the modified diagnosis criteria of the International Association for the Study of Pain (IASP) or Budapest criteria, which established the CRPS as an exclusion diagnosis characterized by a continuing pain disproportionate to any incited event associated with sensory (hyperalgesia and/or allodynia), vasomotor (temperature and/or skin color asymmetry), sudomotor (edema and/or sweating) and motor/trophic changes in a limb region, that doesn't follow any specific nerve territory or dermatome.[5]

CRPS continues to be a diagnosis challenge, with a significant delay in the diagnosis, which worsened the prognosis and treatment response.[6] Considering that the precipitating event in CRPS is often represented by an injury, this condition should be relevant for athletes.[3] The aim of this article is to discuss the importance of clinical suspicion for an early CRPS diagnosis in a sprain's young athlete clinical case.

Highlights:

- CRPS is a disproportionate and persistent, regional pain related to a minor trauma.
- CRPS represents a diagnostic challenge that should always be considered in the context of a persistent pain sport-related injury.
- Despite extensive research on CRPS, it may still be unfamiliar to the medical community. Raising awareness is crucial to aiding healthcare professionals in making timely and accurate diagnoses.

The Case

We present the case of a sixteen-year-old male patient with no medical history, who suffered two consecutives grade one right ankle sprain while playing soccer. He had poor response to 15 kinesiotherapy sessions. Afterwards, he progressed with a burning pain, increased volume and red-purple erythema triggered by the heat. Physical examination ***Figure 1.A*** showed mild edema, erythema, allodynia, higher temperature and hair loss in the right foot, without motor alterations. A right foot ultrasound was performed, revealing a partial anterior talofibular ligament rupture and a mild peroneal tendonitis. Based on these findings, the patient continued with kinesiotherapy.

Figure1. **Patient's Physical Examination at Third and Tenth Month of Evolution.**

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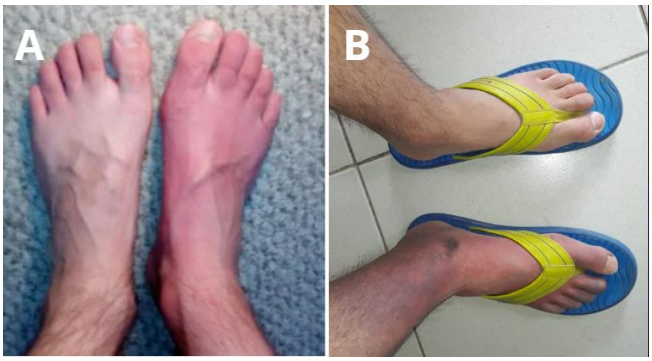
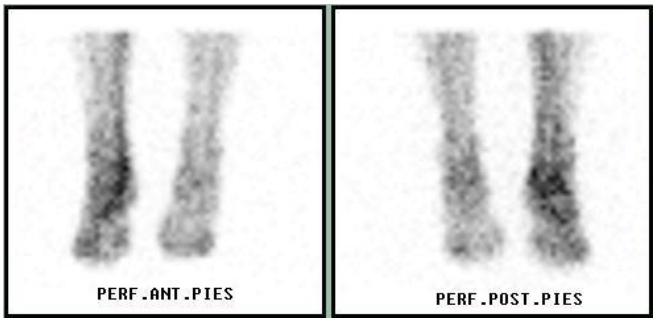


Legend: Physical examination at the third month (Figure 1.A): asymmetrical skin changes, with greater erythema in the right foot. Physical examination at the tenth month (Figure 1.B): increase of asymmetrical erythema, edema and hair loss in the right foot.

Due to pain persistence at the sixth month, the case study was complemented with vascular and rheumatological tests, which were negative. Additionally, it was performed a right ankle MRI, which reported the presence of calcaneal bone edema. Therefore, new kinesiotherapy sessions were indicated. At the tenth month, the symptoms worsened, increasing the affected area until middle leg ***Figure 1.B***. Since symptoms could not be explained by the imaging findings, a lower member bone scintigram was requested ***Figure 2***. The exam reported a moderate increase in arterial flow and diffuse hyper uptake in right foot and ankle, confirming the diagnosis of a CRPS.

***Figure 2*. Lower Member Bone Scintigram.**



Legend: An anterior and posterior vision of a lower member bone scintigram evidence a moderate increase in arterial flow and diffuse hyperuptake in right foot and ankle. These findings correlated with the autonomic dysfunction in CRPS, leading to blood flow abnormalities

The patient began treatment with pregabalin and duloxetine in ascending doses until 75 mg every 12 hours and 60 mg per day, respectively. In addition, he received psychotherapy for pain management. Symptoms worsened after one year of treatment. Further therapeutics options were considered. The patient underwent 5 sympathetic blocks sessions, not being able to complete the 10 recommended sessions. Symptoms remained stable and the patient abandoned medical therapies in the third year. He explored alternative medicines such as acupuncture, phytotherapy, mindfulness, among others, without significant relief. Medical follow-up continued and during the fifth year of disease there was a progressive decrease in symptoms until remission. Only mild vegetative symptoms persisted but there was an absence of pain and full recovery of functionality.

Discussion

We presented the case of a sixteen-year-old male with no medical history suffered two minor right ankle injuries. He developed severe, persistent pain with vasomotor, sudomotor and trophic abnormalities. After 10 months, CRPS was confirmed by bone scintigram. He received multiple treatments until spontaneous remission in the fourth year.

CRPS incidence has been more commonly reported in old women between 60 and 70 years and affecting upper limbs.[7] In young population incidence is still greater in women but affecting lower limbs, probably due to the higher incidence of sport-related injuries.[8] In consequence, athletes experiencing worsening conditions after common trauma should be assessed for excluding CRPS.[3] A scoping review searching for CRPS in athletes found only 20 patients reported, 15 females and 5 males between 12 and 20 years, where 50% of cases were after an ankle sprain.[9] CRPS is not a rare condition according to four population-based studies in the last two decades.[2] However, it remains unknown for many physicians and is underdiagnosed or diagnosed late, leading to a worse therapeutic response and prognosis.[10] In some studies, the mean time between the injury and diagnosis is around 2,8 to 3,9 years.[6,11] In our case, the patient pertained to a high-risk group (young athlete with lower limbs injury). Although the time from the ankle sprain until CRPS diagnosis in the presented case was 10 months, less than described in these studies, the diagnosis could have been done earlier under a greater clinical suspicion.

CRPS diagnosis is clinical according to the IASP or Budapest criteria.[5] Laboratory or imaging tests are not needed for CRPS diagnosis, but it could be used to support it or to exclude differential diagnosis.[12] Bone scintigraphy or triple-phase bone scan has shown to be the better complementary diagnosis test over MRI and radiography.[13] In our case, the imaging findings may have acted as confounding factors, since the symptoms and signs probably weren't explained by a tendonitis or bone edema.

There are multiple described treatment options for CRPS, nevertheless the quality of clinical trials remains variable.[14] Immobilization is a well-recognized possible cause and/or perpetuating factor in CRPS,[15] so in this case during the delay diagnosis process the multiple kinesiotherapy may have worked as a therapeutic tool preventing a severe early progress of disease.

In summary, we discussed the case of a late CRPS diagnosis even though it complied with all clinical features of IASP diagnosis criteria and was part of a high-risk group. Physicians should always consider this diagnosis in the context of a sport-related injury with persistent and severe pain. Clinical suspicion is key for an early diagnosis which improves treatment response and prognosis. Even though CRPS research has been largely

conducted in the last decades, it may remain unfamiliar to the medical community. Raising awareness of this entity may help healthcare professionals to make a timely and accurate diagnosis.

Summary – Accelerating Translation

Título: Síndrome de Dolor Regional Complejo, un Caso Clínico que Resalta su Importancia como Diagnóstico Diferencial en Lesiones Deportivas.

Problema principal a resolver: El Síndrome de Dolor Regional Complejo (SDRC) abarca una variedad de condiciones dolorosas caracterizadas por dolor regional persistente y desproporcionado en relación con una lesión menor. Los jóvenes atletas enfrentan un mayor riesgo de desarrollar SDRC en el contexto de lesiones deportivas. Un diagnóstico clínico oportuno es esencial para mejorar los resultados del tratamiento y el pronóstico.

Objetivo del estudio: Este estudio tiene como objetivo discutir la importancia de la sospecha clínica para un diagnóstico temprano de SDRC en el caso clínico de un joven atleta con esguince.

Presentación del caso y conclusión: El caso presentado involucra a un paciente que experimentó un diagnóstico tardío de SDRC, a pesar de que los síntomas coincidían con los criterios diagnósticos y pertenecía a un grupo de alto riesgo (jóvenes atletas). Los médicos deben considerar constantemente el SDRC cuando se enfrentan a lesiones deportivas acompañadas de dolor persistente e intenso. Fomentar la sospecha clínica es fundamental para un diagnóstico temprano, lo que mejora la respuesta al tratamiento y el pronóstico general. A pesar de la investigación considerable sobre el SDRC en las últimas décadas, este aún puede ser desconocido para la comunidad médica. Por lo tanto, aumentar la conciencia sobre esta condición es crucial para ayudar a los profesionales de la salud a realizar diagnósticos oportunos y precisos.

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