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Traumatic Myositis Ossificans in a Newborn: A Case Report

Miositis osificante traumática en recién nacido: reporte de caso

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Abstract

Traumatic myositis ossificans (TMO) is a disorder in which heterotopic ossification occurs two to four weeks after one or multiple traumas. The goal of the present article is to describe the clinical and radiological characteristics of a case of TMO in a newborn (NB) after a peripheral intravenous cannulation, a rare procedure in the clinical practice of neonatology. The patient is a premature 33-week-old NB who, 20 days after birth, presented with a 3 cm x 2 cm lump in the distal third of the left forearm that did not seem to cause pain or to limit movement, and with no evidence of infection. The rest of the physical exam was within normal limits. Three weeks before the lesion, a peripheral intravenous catheter (PIVC) was placed in that area. A radiograph of the left forearm showed soft-tissue calcification without disruption of adjacent bone structures. Ultrasound revealed a focal, oval soft tissue lesion with partially-defined borders and posterior acoustic shadow; the rest of study showed normal long bones. The serum levels of alkaline phosphatase, calcium, and phosphorus were all normal. In view of the tumor lesion on the physical examination and the calcified image in soft-tissue on plain X-ray and a recent history of PIVC microtrauma, we reached to the diagnoses of TMO. During the follow-up, the lesion decreased in size until it completely disappeared four months after the diagnosis. No radiological control was needed. Uncommon in NBs, TMO is generally self-limited and with a good prognosis.

Keywords
- myositis ossificans
- neonatology
- orthopedics
- prematurity
- newborn
- trauma

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Resumen

La miositis osificante traumática (MOT) es una enfermedad en la que ocurre osificación heterotópica en dos a cuatro semanas tras uno o múltiples traumatismos. El objetivo de este artículo es describir las características clínicas y radiológicas de un caso de MOT en un recién nacido (RN) después de la canulación intravenosa de vía periférica, poco frecuente en la práctica clínica en neonatología. Presentamos a un RN pretérmino de 33 semanas en que, a los 20 días de vida, se evidenció lesión tumoral en el tercio distal del antebrazo izquierdo de 3 cm por 2 cm, que no impresionaba dolor, ni limitación a la movilización, y en la que no había signos infecciosos. El resto del examen físico osteomuscular era normal. En la zona de lesión, tres semanas antes, se había instalado un catéter intravenoso periférico (CIVP). Una radiografía del antebrazo izquierdo demostró lesión calcificada al nivel de las partes blandas, sin disrupción de las estructuras óseas adyacentes; la ecografía del antebrazo reveló una imagen focal ovalada, de contornos parcialmente definidos, con sombra acústica posterior; el resto de los estudios de huesos largos era normal. Los niveles séricos de fosfatasa alcalina, calcio, fósforo también eran normales. En vista de la lesión tumoral al examen físico y la imagen calcificada en partes blandas a través de radiografía simple, con antecedente de microtraumas de VVP, se concluyó MOT. Se hizo seguimiento, con disminución del tamaño hasta que la lesión desapareció a los cuatro meses. No requirió control radiológico. La MOT es infrecuente en el RN, y, en general, la resolución es autolimitada y tiene buen pronóstico.

Nivel de evidencia IV

Palabras clave

- miositis osificante
- neonatología
- ortopedia
- prematuridad
- recién nacido
- traumatismo

Introduction

The formation of non-neoplastic lamellar bone in soft tissue, where it normally does not exist, is called myositis ossificans (MO). However, it would be more accurate to describe MO as the formation of bone tissue within skeletal muscle, and heterotopic ossification (HO) when it affects soft tissue in general.[1] Samuelson and Coleman[2] divide MO into four different groups: 1) myositis ossificans progressiva or fibrodysplasia of autosomal dominant inheritance; 2) myositis ossificans associated with chronic disease; 3) traumatic myositis ossificans (MOT); and 4) pseudomalignant myositis ossificans (nontraumatic).

The most common form is perhaps TMO, representing between 60% and 75% of cases.[3] In a retrospective study published in 2017, Sferopoulos et al.,[1] analyzed 22 patients (11 girls and 11 boys), aged between 3 and 14 years at the time of diagnosis, 18 (81%) of whom had TMO. It was first described by Reidel in 1883, followed by Dejerne and Ceiller, who observed it in soldiers who suffered spinal cord injuries during World War I.[4] Epidemiological data highlight a prevalence in men between the ages of 30and 40 years, with athletes being the population at the greatest risk.[5] The injury usually occurs in the large muscle groups of the thigh (80% of the cases) and the upper limbs of young men.[1,6] The pathogenesis has not been entirely elucidated to date, but the participation of bone morphogenetic proteins (BMPs) that stimulate the stem cells of the mesenchymal spindle has been postulated, which then migrate to the injured area and become fibroblasts and eventually osteoblasts.[7] Clinically, it is a tumor, which may or may not show signs of local inflammation, preceded by trauma.[6]

The objective of the present work is to describe the clinical and radiological characteristics of a case of TMO in a newborn (NB) after the placement of a peripheral intravenous catheter (PIVC), which is rare in the clinical practice in neonatology.

Clinical case

A 33-week preterm NB, female, product of controlled pregnancy, with prenatal diagnosis of gastroschisis, premature rupture of membranes of 22 hours of evolution, delivered by emergency cesarean section, with weight at birth of 1,900 g, height at birth of 38 cm, and APGAR score of 6 at 1 minute and of 8 at 5 minutes. Abdominal surgery to correct the wall defect was performed, without complications.

At 20 days of life, an indurated tumor lesion in the distal anteroposterior third of the left forearm, measuring 3 cm x 2 cm, emerged. It did not seem to cause pain, or to limit movemtn, and it did not present signs of infection. The PIVC was inserted in the area of the injury described in the first week of life. She did not receive calcium gluconate for this PIVC. There were no findings of dysmorphia in the rest of the physical examination.

At 22 days of age, a radiograph of the left forearm was requested (►**Figure 1**), which revealed a calcified lesion at the level of the soft tissue in the dorsal region, radial in appearance, without disruption of the adjacent bone structures. An ultrasound scan of the left forearm (►**Figure 2**) showed an oval focal image, with partially defined contour,

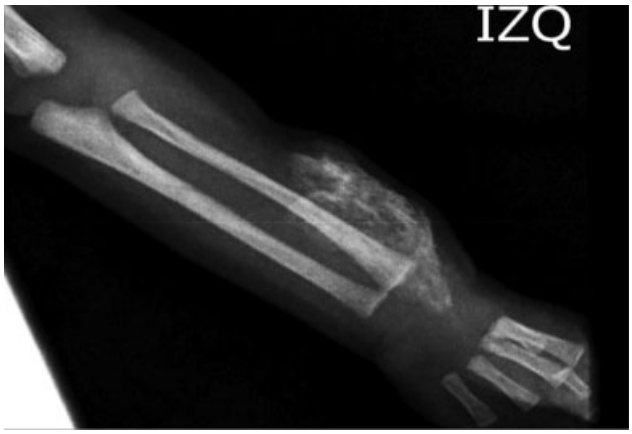

Fig. 1 Radiograph of the left forearm of a newborn: calcified lesion at the level of the soft tissue in the dorsal region, predominantly radial in the periphery, and with a partially radiolucent central area. Between the lesion and the underlying bone, there is a radiolucent area. There is no apparent disruption of the adjacent bone structures.

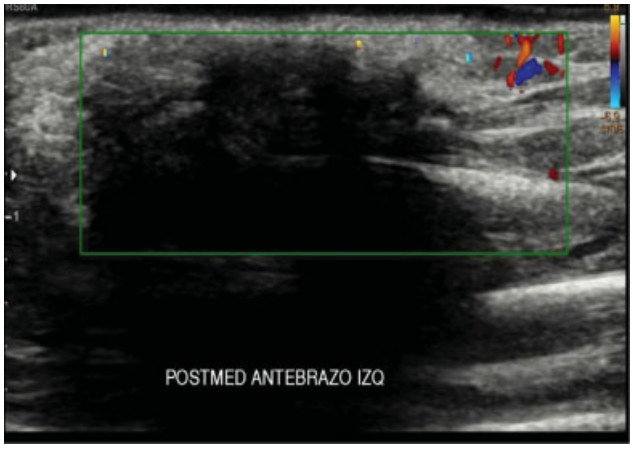

Fig. 2 Ultrasound scan of the left forearm: oval focal image, with partially defined contour, with posterior acoustic shadow, which does not show flow on Doppler. Abbreviation: POSTMED, postmedication.

with posterior acoustic shadow, which did not show flow on Doppler. All other radiological studies of the long bones were normal. The hemogram, erythrocyte sedimentation rate (ESR), and the levels of C-reactive protein, calcium, phosphorus, alkaline phosphatase (ALP), lactate dehydrogenase (LDH), and creatine kinase (CK) were normal. Given these findings and the traumatological and orthopedic evaluation, it was concluded that the diagnosis was TMO.Observation and conservative treatment were chosen. The tumor was checked monthly on an outpatient basis, demonstrating a progressive decrease in the size of the lesion. At the fourth month, the tumor mass was not palpable, and radiological control was not required; the patient was discharged from the orthopedic and trauma clinic.

Discussion

Traumatic myositis ossificans is related to an obvious and direct trauma from a single serious injury or recurrent episodes of minor trauma.[1] In the clinical case herein presented, the trigger was the placement of a PIVC in the injured area. Other risk factors such as severe burns, muscle bleeding due to hemophilia, and orthopedic operations have been described; in infants, battered child syndrome should be suspected.[1] It occurs due to a proliferative mesenchymal response resulting from an initiating soft-tissue lesion. In the first week, proliferative and highly vascular fibroblastic cells predominate. As the lesion matures, a typical zonal pattern with three distinct zones develops: 1) the center consists of rapidly proliferating fibroblasts with areas of hemorrhage and necrotic muscles; 2) the middle zone is characterized by osteoblasts with an immature osteoid formation and islands of cartilage due to endochondral ossification; and 3) the peripheral zone is composed of mature bone, generally well separated from the surrounding tissue by myxoid fibrous tissue. Then, from the third to the fourth weeks, calcifications and ossifications appear within the mass. In the sixth to eighth weeks, a well-organized cortical bone, with cortex and bone marrow space, develops in the periphery. This new bone continues to mature, such that by six months a dense ring of compact bone has developed with a central core of lamellar bone.[8]

The molecular mechanisms of HO have not been fully elucidated; however, one of the most studied molecules are BMPs.[9] which are known to induce the differentiation of proliferative fibroblast cells into chondroblasts and osteoblasts.[1] Overexpression of BMPs inhibits the alignment and fusion of myoblasts that are required for the formation of myotubes. Myoblasts differentiate into osteoblasts after exposure to signaling from these proteins.[10]

Clinically, two to three weeks after the trauma, edema and the formation of a firm, stony, not very painful, expansive mass may be observed; [6] it may also present reduced joint movement, mimicking or coexisting with thrombophlebitis, cellulitis or osteomyelitis. Our patient did not present limitations to flexion or extension of the proximal extremity of the left wrist, neither signs of infection.[4]

With regard to the laboratory tests, the levels of ALP can be as high as 3.5 times the normal levels 4 weeks after the injury, with maximum levels measured at 12 weeks. The increase in the levels of serum ALP is accompanied by an increase in the levels of inorganic phosphate and is preceded by a transient decrease in the levels of serum calcium.[11] An ESR greater than 35 mm/hour may indicate the development of HO. The levels of C-reactive protein can rise early in HO, and the levels of CK can be used to determine the severity of HO.[7] None of these biochemical parameters are specific,[7,11] and, in our case, they were all within normal limits.

In plain X-rays, calcifications usually appear between two and three weeks, with a clearer pattern between four and six weeks after the trauma.[12] The most important radiographic hallmark is the presence of the so-called zonal phenomenon, which is characterized by a central radiolucent area, which indicates the formation of immature bone, and by a dense peripheral zone of mature ossification. In addition, a thin radiolucent cleft separates the ossified mass from the adjacent cortex.[8] In most cases, the lesion does not adhere to the

underlying bone,[6,8] which was also true in the present clinical case. Ultrasound shows infiltration of the subcutaneous fat and muscle, as well as hyperechoic lines with posterior acoustic shadow, although these do not appear until two to three weeks of evolution.[6] On magnetic resonance imaging (MRI), it appears as an inhomogeneous soft-tissue mass. In its early stages, only an isointense nodule can be distinguished from the surrounding muscles on T1-weighted images, which increase contrast in its periphery on T2-weighted images. Only in the later stages are the MRI findings compatible with MO. Computed tomography (CT) generally shows a border of mineralization around the lesion after four to six weeks, which is much less evident than on MRI.[8] Computed tomography and MRI have higher resolution than ultrasound, but are not routinely requested.[6] Triphasic bone scan is more sensitive for early detection approximately 2.5 weeks after the injury.[4] A biopsy may be requested if the diagnosis is unclear, or in cases that do not show the typical radiographic zonal ossification pattern.[8] Microscopically, two defined regions are observed: a peripheral zone with ossification and a central cellular zone. Mature lamellar bone with active osteoclasts is observed in the outer region. Sometimes, an osteoid intermediate zone is observed, with cartilage or bone tissue formation and active osteoblasts; and a central region is usually composed of fibrovascular tissue containing spindle cells and mesenchymal cells. This presentation is pathognomonic and distinguishes it from osteosarcoma.[12]

In the case herein reported, no MRI scans, CT scans, or biopsy were performed, because there was a history of microtraumas caused by the placement of the PIVC on several occasions in the area where the lesion developed, and because we had a simple X-ray compatible with TMO.

An important form of MO that is worth further discussion is the myositis ossificans progressiva, which is present in the first decade of life and is associated with the progressive formation of extraskeletal bone and disfiguring manifestations. This chronic disorder is easily recognized by the microdactyly or adactyly associated with the first toe and thumb. Our patient did not have any of these deformities. Other differential diagnoses would be calcified hematoma (calcium deposit in the muscle), soft-tissue sarcomas, osteosarcoma, hyperparathyroidism, sarcoidosis, dermatomyositis, and sequelae of subcutaneous fat necrosis after extravasation of calcium gluconate.[4]

Regarding treatment, resolution is spontaneous; in most cases, surgical removal of the calcified lesion is not necessary in children, unless it is painful or interferes with movement.[1] In the case herein presented, observation was chosen because there was no limitation to movement, there was no pain on palpation, and no progression of the size of the lesion. The follow-up was carried out until the tumor mass was not palpable, which was at the fourth month.

Conclusion

In patients in pediatric age and even more so in the NB, it is rare to observe the development of MO. the case herein reported is interesting since it is of the traumatic form resulting from the history of PIVC placement, and the lesion was confirmed with plain radiology. The histological study is reserved for those patients who do not present a typical pattern of traumatic MO on imaging studies. According to the literature and as happened in the present case, TMO follows a benign, self-limited course, and does not require surgery in children. It is important to consider the genetic and malignant differential diagnoses, taking a detailed medical history to take appropriate conduct.

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Conflict of Interests
The authors have no conflict of interests to declare.

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