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Case Study of Neonatal Cytosteatonecrosis: Clinical Findings and Management

Abdessamad Lalaoui[1,2], Sofia Hamdani[1,2], Khalid Abi El Aala[1,2], Ghizlane Kassal[1,2], Fatiha Bennaoui[1,2], Nadia El Idrissi Slitine[1,2], Fatima Ezzahra Hazmiri[3], Hanane Rais[3], Fadl Mrabih Rabou Maoulainine[1,2]

[1]Neonatal Intensive Care Unit, University Hospital Center Mohammed VI, Marrakech, Morocco
[2]Health, Childhood and Development Faculty of Medicine Research Team, Cadi Ayyad University, Marrakech, Morocco
[3]Department of Anatomic Pathology, Med VI University Hospital, Marrakesh, Morocco
Email: lalaoui.abdessamad@gmail.com

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Abstract

Cytosteatonecrosis of the newborn (CSNN), though rare, is a benign panniculitis that causes various skin lesions (hardened, purplish plaques/nodules). This condition can occur in infants of diabetic mothers or those who have experienced birth asphyxia and may be unfamiliar to some healthcare practitioners. CSNN can lead to complications such as life-threatening hypercalcemia and metabolic disorders. This case study reports on a two-week-old newborn with CSNN, admitted to the neonatal intensive care unit at the Pediatric Center of Mohammed VI Hospital in Marrakech. The infant, born to a diabetic mother, was initially treated for neonatal respiratory distress. Four days after discharge, a biopsy-confirmed the diagnosis of CSNN, revealing lesions on the scalp, thighs and neck. At 1.5 months, the patient developed a scalp abscess and hypercalcemia. Hypercalcemia was effectively managed with hyperhydration and diuretics, resulting in the normalization of calcium levels. Abdominopelvic and transthoracic cardiac ultrasounds were normal. By six months, the patient's skin lesions had spontaneously regressed.

Keywords

Newborn, Cytosteatonecrosis, Hypercalcemia

1. Introduction

Cytosteatonecrosis of the newborn (CSNN) is a rare, benign panniculitis that typically manifests within the first few weeks of life [1] [2]. It often affects term and post-term infants following challenging deliveries [3]. The condition is marked by the presence of discolored skin patches, ranging from purplish hues in lighter skin

tones to darker shades in deeper skin tones. These lesions are commonly located on the face, torso, buttocks, and limbs [4]. Histological examination reveals lobular panniculitis characterized by eosinophilic fat cell necrosis beneath otherwise normal-appearing skin [5].

Despite its generally benign course, CSNN can lead to significant complications, including life-threatening hypercalcemia and metabolic disorders [6]. The rarity of CSNN may be attributed to the under-recognition of its cutaneous manifestations. Risk factors associated with CSNN include macrosomia, maternal diabetes, perinatal asphyxia, and obstetric trauma. Severe hypercalcemia remains a critical concern, with the potential for cardiac, neurological, and renal complications, requiring vigilant medical follow-up. Although the condition is rare, it is relatively straightforward to diagnose [1].

Early diagnosis is crucial to prevent perinatal suffering and hypoxia [6]. Research indicates a higher prevalence in female infants [7]. Treatment generally focuses on hyperhydration, diuretics, corticosteroids, avoiding vitamin D supplements, and close monitoring of blood calcium levels [6] [8].

In this paper, we report a case of CSNN, detailing its management and evolution in the neonatal intensive care unit of the pediatric center at Mohammed VI Hospital in Marrakech.

Objective: This study aims to elucidate the clinical and epidemiological characteristics of CSNN.

2. Case Report

The patient is a female newborn delivered by a 38-year-old mother with a history of insulin-managed diabetes. The infant was born at term via vaginal delivery with an episiotomy and forceps, in the context of perinatal asphyxia. Apgar scores were 4/10 at 1 minute and improved to 6/10 at 5 minutes. The amniotic fluid was meconial, and the birth weight was 4300 g.

Initially, the newborn required resuscitation in the delivery room, including drying, suctioning, and ventilation with a resuscitation bag. She was admitted to the neonatal intensive care unit (NICU) due to respiratory distress related to an early-onset neonatal bacterial infection, receiving CPAP and a 7-day course of antibiotics.

The initial infection workup revealed an elevated C-reactive protein (CRP) level of 53.9 mg/L, while the complete blood count and blood cultures were negative. Blood cultures and antibiograms were repeated twice during the patient's hospital stay, with no bacterial growth in either culture.

Four days after discharge, the patient presented with reddish and purplish indurated subcutaneous nodules on the scalp, both thighs, and neck (**Figure 1** and **Figure 2**). Histopathological examination of a skin biopsy confirmed the diagnosis of cytosteatonecrosis, showing lobular panniculitis with characteristic lesions (**Figure 3**).

At this time, the serum calcium level was normal. The lipid profile showed mild elevations, with triglyceride levels at 1.86 g/L, total cholesterol at 0.81 g/L, HDL cholesterol at 0.26 g/L, LDL cholesterol at 0.18 g/L, and vitamin D was low at 3 ng/mL.

The patient has not been tested for genetic disorders or immunodeficiency.

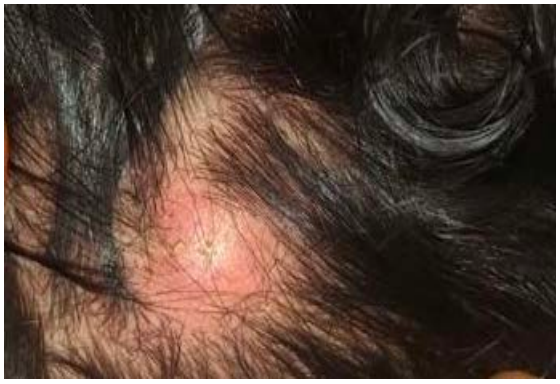

Figure 1. Reddish and purplish indurated subcutaneous nodules on the scalp.

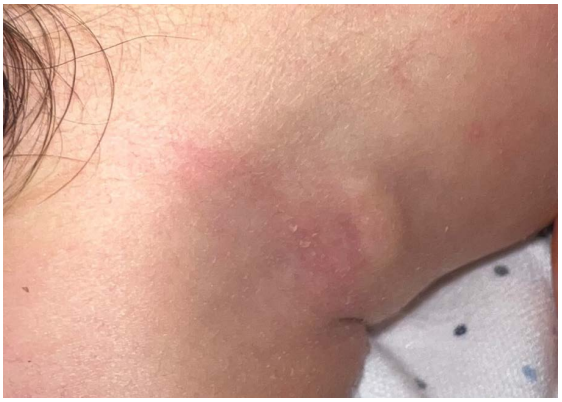

Figure 2. Indurated subcutaneous nodules on the neck.

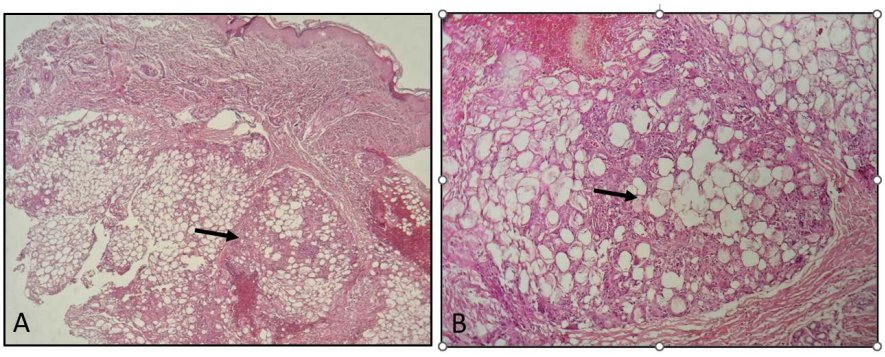

Figure 3. Lobular panniculitis with cytosteatonecrosis lesions ((A): H&E X 10/(B): H&E X 20).

The newborn was placed only under clinical and biological monitoring with no treatment via vitamin D. At 1.5 months of age, an abscess developed in the scalp lesion **(Figure 4),** which was treated with antiseptic cleaning and a local antibiotic solution. The secretion culture of the scalp lesion was not performed. Laboratory tests revealed hypercalcemia with a calcium level of 116 mg/mL. Treatment included hyperhydration and diuretics, which effectively normalized the calcium

levels over time. Both abdominopelvic and transthoracic cardiac ultrasounds were normal, and no calcifications were observed on the skeletal X-ray.

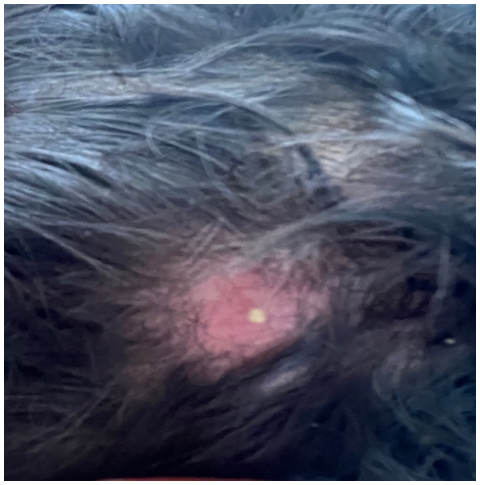

Figure 4. Abscess on the scalp.

By 6 months of age, the patient's condition had improved significantly, with spontaneous regression of the skin lesions.

3. Discussion

Cytosteatonecrosis of the newborn (CSNN) is a rare yet significant form of panniculitis that remains under-recognized despite its diagnostic clarity. The condition predominantly affects macrosomic infants, especially those with a history of perinatal hypoxia and challenging deliveries. Maternal factors such as diabetes and pre-eclampsia are frequently reported, with additional links to familial dyslipidemia and thrombophilias. Clinically, CSNN manifests as erythematous or purplish subcutaneous nodules, typically appearing in the early weeks of life. The condition generally follows a benign course, with lesions resolving over several months, though residual subcutaneous atrophy may occur.

Hypercalcemia is a major complication of Cytosteatonecrosis of the newborn (CSNN), posing significant risks to the infant's health. It can lead to multi-organ damage, particularly in the cardiac system, where calcium deposits may form in the interatrial septum and valves, and in the renal system, where nephrocalcinosis can occur. The exact pathophysiological mechanism behind hypercalcemia in CSNN remains unclear, but it is associated with symptoms such as vomiting and agitation. Effective management of severe hypercalcemia usually involves hyperhydration, diuretics, corticosteroids, or bisphosphonates, particularly during the acute phase [8]-[12].

Our case study underscores a rare presentation of CSNN complicated by hypercalcemia in a neonate. Histopathological analysis reveals lobular panniculitis with fat cell necrosis, an inflammatory infiltrate of lymphocytes, histiocytes, fibroblasts, and giant cells, along with calcium deposits [13]. The low prevalence of

CSNN despite numerous potential risk factors suggests that a complex interplay of multiple factors, including perinatal asphyxia, obstetric trauma, and hypothermia, likely contributes to its development [14]. The lesions typically appear within the first four weeks of life, though some may emerge in the initial days. Hypercalcemia usually develops following the appearance of skin lesions and can lead to a mortality rate of up to 15% [15]. Maternal conditions such as gestational diabetes and dyslipidemia may exacerbate the condition, possibly due to elevated triglycerides and increased fat mobilization in affected infants [9].

Diagnosis of CSNN is generally straightforward, but differentiating it from similar conditions such as neonatal sclerema and erysipelas is critical. Neonatal sclerema involves extensive subcutaneous tissue hardening and fibrosis, and is sometimes considered a severe form of CSNN [9]. Neonatal scleredema, characterized by progressive edema primarily in the lower limbs, also presents diagnostic challenges and should be differentiated, particularly in the context of preceding gastrointestinal or respiratory infections [10] [11].

In our case, the patient with CSNN and hypercalcemia was closely monitored, receiving hyperhydration and diuretics, which led to significant improvement. By six months, the skin lesions resolved spontaneously. This case underscores the importance of early recognition, prompt management, and ongoing follow-up to ensure a favorable outcome and prevent complications like nephrocalcinosis or cardiac calcifications.

4. Conclusions

Recent research has highlighted the need for increased awareness and improved diagnostic protocols for CSNN. Future studies should focus on elucidating the underlying mechanisms of hypercalcemia, evaluating the efficacy of various treatment strategies, and exploring potential preventive measures. Enhanced understanding and early recognition of CSNN will improve management outcomes and reduce associated complications.

Continued awareness and vigilance among healthcare providers are essential for timely identification and management, which can significantly improve outcomes for affected newborns.

Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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