

Case Report

## A Neonatal Case of Hemorrhagic Cystic Subcutaneous Fat Necrosis

### *Hipotermi Tedavisi Sonrası Hemorajik, Kistik Cilt Altı Yağ Nekrozu Gelişen Yenidoğan Olgusu*

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#### ABSTRACT

Subcutaneous fat necrosis is rarely seen in the first weeks of life in newborns born with obstetric complications. It often shows a benign course and heals without complications. Metabolic complications, especially hypercalcemia, may accompany the lesions throughout the clinical course. Herein, we report a term newborn with an atypical course of subcutaneous fat necrosis, whose lesions progressed to a hemorrhagic, bullous character and subcutaneous necrosis with an abscess-like appearance and content.

**Keywords:** Newborn, subcutaneous fat necrosis, therapeutic hypothermia

#### ÖZET

Obstetrik komplikasyonlarla doğan yenidoğanlarda yaşamın ilk haftalarında ortaya çıkan cilt altı yağ nekrozu önemli bir morbiditedir. Sıklıkla iyi huylu bir seyir gösterir ve komplikasyonsuz iyileşir. Klinik seyir boyunca lezyonlara başta hiperkalsemi olmak üzere ciddi metabolik komplikasyonlar eşlik edebilir. Burada hemorajik, büllöz karakterde görünümü ve apse benzeri içeriği ile atipik bir seyir gösteren cilt altı yağ doku nekrozlu bir yenidoğan olgu sunulmuştur.

**Keywords:** Yenidoğan, cilt altı yağ doku nekrozu, teröpatik hipotermi

#### INTRODUCTION

Subcutaneous fat necrosis (SCFN) in newborns is a rare panniculitis characterized by purple-red, erythematous, and subcutaneous nodules or plaques (1). It occurs through a self-limiting inflammatory process in the first weeks of life in term and post-term infants, often accompanied by obstetric complications (2). It is also associated with conditions, such as maternal diabetes, hypertension, hypothyroidism, and exposure to smoking (3). Therapeutic hypothermia (TH) applied af-

ter hypoxic delivery is included in the history of most babies who develop SCFN (4).

TH is widely used worldwide as the only proven neuroprotective treatment method for newborns with moderate and severe neonatal encephalopathy. It involves the reduction of the core body temperature to 33.5–35°C for 72 h and is generally well tolerated by newborns. In some cases, however, it may cause complications, such as coagulopathy, thrombocytopenia, sus-

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ceptibility to sepsis, bradycardia, hypotension, and subcutaneous skin necrosis (5).

In this report, we present a case of fullterm female infant with a history of severe hypoxic delivery followed by TH, who developed SCFN on her back and both gluteal regions. The follow-up and management of the case are shared because the lesions in both gluteal regions gradually transformed into large, bullous, hemorrhagic fluid-filled lesions suggestive of abscess formation, which is rare.

## CASE REPORT

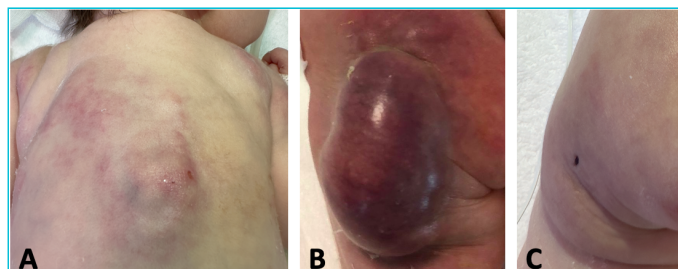
A female infant was delivered from a 23-year-old woman with diabetes mellitus on her second pregnancy after 38 weeks of gestation with emergency cesarean section. The infant weighed 3530 g at birth. Her Apgar scores were 0, 0, and 2 at 1, 5, and 10 min, respectively. Adrenaline was administered once to the resuscitated baby. Her amniotic fluid was meconium stained. The pH of her cord blood gas was 6.8; its base excess, -20; and its lactate, was 25 mmol/L. The baby's Thomson score was 18, and her Sarnat stage was 3.

With the diagnosis of severe hypoxic-ischemic encephalopathy, the baby was subjected to whole-body TH for 72 h. During the follow-up, hypoxic respiratory failure, pulmonary hypertension, systemic hypotension, and renal failure developed. Ventilation support with high-frequency oscillatory ventilation, inhaled nitric oxide treatment, noradrenaline treatment, and peritoneal dialysis were applied. On the eighth day, the baby was extubated. In the second week, the baby was fed enterally fully with an orogastric tube; and in the third week, this was switched to oral feeding. Hypertansaminasemia, cholestasis, and coagulopathy occurred within the first week but normalized in the fourth week.

On the seventh day, red-purple, palpable, and raised skin lesions began to develop in both gluteal regions and on the back (Figure 1-A). This condition was clinically evaluated as SCFN. The lesions in both gluteal regions became larger in the second week and turned into large cystic lesions (12x14x34 mm) with similar colors, which suggested an abscess (Figure 1-B). In the punctures, a hemorrhagic, white, and chalky pus-like material was obtained. No signs of infection were found in the microscopic and culture examinations. The pathological examination of the lesions reported their compatibility with adipose tissue necrosis. Simultaneous with the development of the lesions, the baby's C-reactive protein (CRP) levels increased and reached 145

mg/L at the end of the second week. Although the CRP values started to decline in the third week, the procalcitonin (PCT) value was within normal limits (<0.5 mcg/L) throughout the whole process. In the period when the baby's lesions were prominent, her N-PASS (Neonatal Pain, Agitation and Sedation Scale) score was evaluated as 6 points. She was sedated with morphine. During the follow-up, her calcium and alkaline phosphatase levels remained normal. The 25-hydroxyvitamin D level was 23 mcg/L, so routine vitamin D supplementation (400 IU/day) was continued. The lesions started to regress at the end of the third week.

Ampicillin, gentamicin followed by meropenem and vancomycin treatments were administered intravenously. Mupirocin treatment was applied locally. On the 28th day, the baby's CRP values were still high, so the antibiotherapy was discontinued. However, her PCT value was normal, and her other laboratory and clinical findings did not show signs of infection. Follow-up with current diagnoses was recommended in dermatology consultation. At the end of the first month, the baby was discharged from the intensive care unit without respiratory support. At her outpatient follow-up at the end of the second month, her CRP value decreased to normal, her calcium values remained normal, and her skin findings returned completely to normal (Figure 1-C). No calcification was found in the abdominal ultrasonography.



**Figure 1.** A) Erythematous plaques and fluctuant nodules that first appear on the back of the baby. B) A lesion of 2x4 cm in width and 1 cm in height on the left buttock. C) The appearance of the lesion in B at discharge

## DISCUSSION

Here, a case is presented of a newborn baby who developed diffuse SCFN, in which some of the lesions turned into large bullous structures with hemorrhagic and pus-like contents, suggestive of the abscess. SCFN occurs infrequently in newborns and is mostly associated with obstetric complications (4). They appear as erythematous, hard subcutaneous plaques on the ext-

remities, back, hips, and thighs, rarely involving the face, and disappear within a few weeks. More than half of the lesions occur in the first week of life, and it has been reported that 10% can be seen after the neonatal period (1,6).

The plaques formed can be tender and painful. In the case presented, the baby was sedated with morphine when the lesions were active.

SCFN may progress with complications, even without obvious skin findings (7). However, rarely, as in our case, plaques may enlarge and coalesce and acquire a large, hemorrhagic, bullous character. The appearance of the lesions and the chalky pus-like content of the necrotic adipose tissue may be mistaken for abscess (8).

Although the pathogenesis of SCFN has not been fully elucidated, local tissue hypoxia, physical compression, and other conditions have been hypothesized. It is thought that the excess content of saturated adipose tissue, which has a high melting point and tends to crystallize after hypothermia, predisposes someone to SCFN (6). The fact that most SCFN patients have a history of hypoxic delivery and TH also supports these hypotheses. Meconium aspiration, Rh incompatibility, sepsis, obstetric trauma, preeclampsia, and gestational diabetes are also associated with SCFN (9).

Dhanawade et al. reported a case in which similar lesions developed in an infant diagnosed with hypoxic-ischemic encephalopathy but was not treated for hypothermia (10). The lesions occurred in the first week of life, progressed with elevated CRP and thrombocytopenia, and were complicated by hypercalcemia. In the second week, the lesions transformed into fluctuating, abscess-like swellings. Similar clinical findings developed in our case, but hypercalcemia was not observed.

Hypercalcemia observed in approximately half of the cases is associated with increased 1-alpha reductase activity expressed from inflammatory cells (1, 6). It has also been reported that hypercalcemia can persist for months and even cause mortality (6,12). Hypercalcemia may also cause systemic calcified lesions, such as nephrocalcinosis, nephrolithiasis, and hepatic and atrial calcifications (11,12). The follow-up and appropriate management of infants regarding hypercalcemia constitute the most critical step in the follow-up of patients who develop SCFN. Long-term follow-up of infants is vital since signs of hypercalcemia may also occur after the neonatal period. In our patient, hypercalcemia did not develop in the first two months of follow-up,

and systemic calcification findings were not observed in the abdominal ultrasonography.

History assessment and physical examination are usually sufficient for the diagnosis of SCFN. The most critical condition for differential diagnosis with SCFN in newborns is sclerema neonatorum (SN), which occurs secondary to sepsis and is associated with a poor prognosis. The most important difference between SN from SCFN is that SN occurs secondary to sepsis in premature babies. Cellulitis and cold panniculitis are other clinical conditions that should be considered in the differential diagnosis of SCFN. Biopsy and pathological examination or, less invasively, USG and Doppler studies may be helpful for definitive diagnosis in atypical presentations.

Despite SCFN's possible complications, such as hypercalcemia, thrombocytopenia, hypertriglyceridemia, hypoglycemia, anemia, and renal failure, it shows a benign course, and the lesions regress within a few weeks (3,6). However, it should be kept in mind that thrombocytopenia and renal failure, which developed in our patient and required renal replacement therapy, may be associated with hypoxic delivery and TH. In the long term, subcutaneous atrophy may develop in infants following SCFN (3).

## CONCLUSION

SCFN is a panniculitis that may occur not rarely in newborns undergoing TH after hypoxic delivery. These necrotic tissues can sometimes turn into large hemorrhagic bullae with necrotic tissue content, which may suggest abscesses but may heal without complications.

### Patient Consent Form / Hasta Onam Formu

The parents' of this patient consent was obtained for this study.

### Conflict of Interest / Çıkar Çatışması

The authors declared no conflicts of interest with respect to authorship and/or publication of the article.

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