A Case of Congenital Subcutaneous Fat Necrosis in a Premature Baby and Review of the Literature

Imane E Chtioui, Suzi Mansour, Suzanne Borrhomée*

NICU of hospital of Pontoise, France

*Corresponding author: Suzanne Borrhomée, NICU of hospital of Pontoise, France

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Abstract

Subcutaneous fat necrosis (SCFN) is a form of panniculitis classically affecting healthy full-term infant during the first month of life [1,2]. The lesions usually develop after a free interval on the cheeks, buttocks, trunk and limbs [3]. The evolution is generally benign although it can be associated with severe hypercalcemia, leading to possibly life-threatening complications [4-6]. Here we present a rare case of congenital subcutaneous fat necrosis and a literature review regarding the clinical features, diagnostic and therapeutic aspects and prognosis of this rare entity.

Case Report

The patient was an African preterm boy (36 weeks gestation) weighing 3498g, born by caesarean section for abnormal fetal heart rate. His mother suffered gestational diabetes treated with insulin. At birth, clinical examination showed a painless superficial ulceration, exposing the underlying fascia on the ventral surface of the elbow (Figure 1a, b). He also had a right radial paralysis: right hand, fingers, and wrist were in a flex posture. Main diagnosis initially considered were osteosarcoma, hamartoma, metastasis of neuroblastoma or SCFN. Blood test results found mild thrombocytopenia at 134,000 / mm3. Urea, creatinine, liver panel, calcemia, magnesium and triglyceridemia were normal. Calcemia was and remained normal. It was monitored up to 6 months after healing. Blood cultures and TORCH screen were negative. Arm MRI showed no extension to underlying structures. The histology (Figure 2) confirmed the diagnosis of SCFN.

The lesion was a diffuse panniculitis interesting mostly the adipose lobules, which are the main location of adipocytic necrosis. There was a crystallization in partially necrotic adipocytes. Dermis and epidermis were spared. At the dermo-hypodermal junction and in the hypodermis, there was an inflammatory cell infiltration rich in neutrophils and eosinophils. Local care consisted in a curettage of fibrinous areas with application of hemostatic and healing plasters made with brown algae fiber, until fibrin disappeared. Then hydrocolloid dressings were applied (Figure 1c). We used a splint to keep the elbow in extension in order to avoid limb retraction during healing. Physiotherapy allowed us to avoid amyotrophy and to preserve correct motricity of the right arm, as observed during the two-month follow up (Figure 1d). We did not supplement him with vitamin D.

Figure 1: Outcomes of congenital ulcer of the child’s forearm.
was prescribed physiotherapy to prevent limb retraction. He could not attend his sessions because of the pandemic lockdown, and a mild difference in mobility between both elbows was noted at the 6 months follow up, with less strength and mild amyotrophy in the right arm, and less flexibility for spontaneous extension. There was no difference in passive extension. This difference disappeared at the 12 months follow up, after he attended weekly sessions of physiotherapy. Complications include local tissue breakdown, hypoglycemia, anemia, thrombocytopenia, hypertiglyceridemia and hypercalcemia [17]. The risk of hypercalcemia is directly correlated to the extension of skin lesions [4]. Major hypercalcemia may be responsible for nephrocalcinosis [4,12,18], heart valve and venous calcifications [3]. In our case, only hypoglycemia was noted and was probably related to gestational diabetes. Localized skin lesions and avoiding to take vitamin D are associated with less hypercalcemia [12,15,18,19]. Several mechanisms of hypercalcemia can be entangled [4,5]: necrosis of fat cells leading to an increase in prostaglandins with activation of osteoclasts; release of calcium by necrotic adipocytes and/or abnormal production of 1,25-ihydroxyvitamine D by macrophages, increasing bone turnover. Newborns with SCFN should have long-term follow-up to detect late hypercalcemia and avoid these complications [18].

Conclusion

Congenital neonatal cytosteatonecrosis is a very rare condition often overlooked by practitioners. Functional prognosis can be initiated. Conservative or surgical treatment (skin grafting) depends on the extent of the skin lesions. The main complication is hypercalcemia, which can be life threatening, justifying prolonged monitoring of the calcium level until the disappearance of the skin lesions. Routine intake of vitamin D should be avoided.

References


