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Case Report

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

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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.

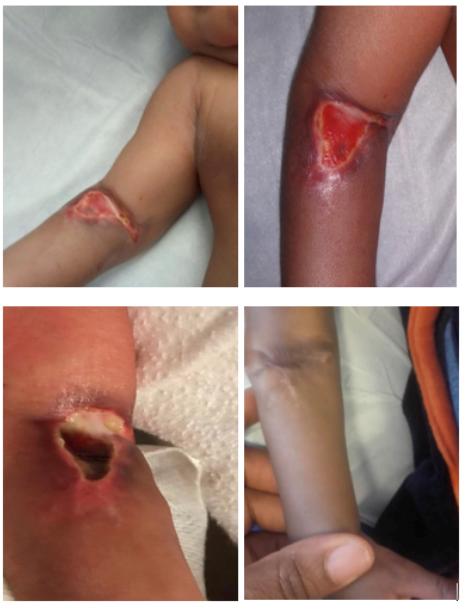

Figure 1: Outcomes of congenital ulcer of the child's forearm.

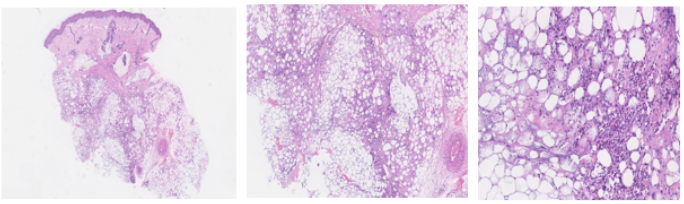

Figure 2: Histological diagnosis of our patient with congenital SCFN.

Discussion

SCFN is a rare type of localized lobular panniculitis usually found in full-post term infants [2] but recently reported more frequently in premature infants [1]. The pathogenesis of SCFN remains unclear. Main hypotheses involve an abnormality of fatty tissues: fat metabolism disorder with excess of saturated fat in the subcutaneous tissue. Hypoxia or hypothermia can also cause the crystallization of saturated fat and fatty necrosis [3,7]. Our patient was born to a mother with gestational diabetes, which was his only risk factor for SCFN. Other risk factors are preeclampsia, placenta previa, perinatal asphyxia, abdominal compressions, hypertension, cocaine or cigarette exposure, calcium blocker use during pregnancy, and familial dyslipidemia [2,3,8-12]. The most common sites of the lesion are cheeks, back, buttocks and extremities [3]. The plaques are usually not ulcerated [1]. To date, only two cases of congenital SCFN with ulcerated lesions of limbs have been reported [13,14]. In our case, the lesion was present at birth without a free interval on the ventral surface of the elbow, and we believe an unidentified amniotic band syndrome is a possible etiology, as it could have caused a local compression on this area. The histology of the SCFN is identical whatever the time of onset (congenital or postnatal) [1,13-16]. There is a normal epidermis and dermis. Underneath dermis is a lobular panniculitis with foci of eosinophilic necrosis of the adipose tissue including optically empty intra-adipocyte radial slits, caused by a dissolution and a lipid crystallization [15], as we found in our patient. Spontaneous resolution of the SCFN in the first year of life is the rule, and management is usually supportive [1]. The treatment of the ulcerated lesions depends on the size and place of the skin defect. If it is small, it can heal spontaneously. Conservative treatment consists in various types of dressings and topical antiseptics, like the treatment in post-natal SCFN [16]. However, in the congenital SCFN, the two patients described in the literature needed repair surgery (skin graft) due to the extensive nature of the lesions. The outcome was positive with complete healing after 6 months in one patient [13,14]. In our case, thanks to the limited nature of the lesion, local care with immobilization of the limb allowed complete skin healing after 2 months. Functionally, he had right radial paralysis, also found in one of the two cases described in the literature [13]. The boy 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, hypertriglyceridemia 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.

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