Rare disease

An unusual complication of subcutaneous fat necrosis of the newborn

Kaltoum Alaoui,¹ Sana Abourazzak,² Abdellah Oulmaati,¹ Moustapha Hida,² Abdelhak Bouharrou¹¹Neonatal Intensive Care Unit, Mère-enfants Hospital, University Hospital Hassan II, Fez, Morocco;²Department of Pediatrics, Mère-enfants Hospital, University Hospital Hassan II, Fez, Morocco**Correspondence to** Dr Sana Abourazzak, abourazzak77@yahoo.fr**Summary**

Subcutaneous fat necrosis (SCFN) is a well-described cause of subcutaneous calcification occurring in a young infant. Calcification of areas of fat necrosis in the neonate is a rare condition. In this case, the radiographic demonstration of soft-tissue calcification revealed the skin calcification in the absence of clinical signs of SCFN and without hypercalcaemia in the first 48 h of life.

BACKGROUND

We report a case with skin and soft-tissue calcifications excluding perivisceral sites without initial clinical signs of subcutaneous fat necrosis (SCNF).

CASE PRESENTATION

A 10-week-old infant girl born at 40 weeks of gestation by urgent caesarean section due to variable fetal decelerations during labour. Apgar scores were 4/6/8 (1'/5'/10'). The birth weight was 4800 g (>97th percentile). There was no history of maternal diabetes during pregnancy or other cause of macrosomia. The baby was transferred to the neonatal intensive care unit. Clinically, perinatal asphyxia was noted with hypotonia and hypoglycaemia 2 mmol/l and normal calcaemia (2 mmol/l). The infant was otherwise well developed and discharged home after 48 h, with an initial physical examination considered normal. Her course over the next 5 days was unremarkable.

The patient was readmitted on the 10th week for with large induration on her lower limbs. A careful exam of the whole cutaneous surface revealed hard plaque-like masses on handling the limbs which were palpable as though the child had a 'coat of armour'. The remainder of the physical examination was normal.

INVESTIGATIONS

All laboratory investigations were normal except for hypercalcaemia in the range of 2.95 mmol/l. A biopsy was not performed because the infant was well and the results of clinical investigations were consistent with SCNF.

Standard radiography showed extensive linear and nodular subcutaneous calcifications in both limbs (figure 1).

DIFFERENTIAL DIAGNOSIS

Differential diagnosis must include neonatal panniculitis and, above all, sclerema neonatorum, a similar condition with severe prognosis. In sclerema, cold livid plaques develop during the first week in newborns with severe diseases, with quick extension to the entire skin surface.

Other diagnoses that can be included from the clinical findings include haemangioma, cellulites, histiocytosis, erythema nodosum, Farber disease, fibromatoses, rhabdomyosarcomas and erysipelas, which are all different histologically.

TREATMENT

Conservative treatment was established with restriction of calcium and vitamin D and follow-up of laboratory parameters in the next months.

OUTCOME AND FOLLOW-UP

Serum calcium levels normalised within a week, and the nodules disappeared without complications after 30 days.

The newborn was a well-appearing infant with a spontaneous regression of the lesions. At age 3 months, no atrophy of the calcifications has been seen. Standard radiography was normal at age of 6 months.

DISCUSSION

SCFN of the newborn is an uncommon disease that appears from the 2nd to the 20th days of life,¹ usually within the first 4 weeks of life. The aetiology is unknown, but there is usually a previous perinatal injury that results in tissue hypoxia, and the newborn subcutis is more susceptible to hypoxia than other tissues.

Newborns who develop SCFN are usually healthy and full term at delivery, but have had some obstetric trauma, meconium aspiration, asphyxia, hypothermia, hypoxaemia or macrosomia.

In our case, the asphyxia, the macrosomia and the metabolic disorders may be implicated.

The characteristic features include painful, firm, indurated, erythematous nodule or plaques, over buttocks, trunk, arms and cheeks. The pathogenesis of SCFN is unknown.

Various mechanisms have been proposed²:

- An underlying defect in neonatal fat metabolism, resulting in an increase in saturated fatty acids within the

Figure 1 Standard radiography showing the extensive linear and nodular subcutaneous calcifications in both limbs.

subcutaneous tissue, which, exacerbated by neonatal stress, may precipitate this condition.

- Fat necrosis caused by local trauma during delivery.
- Hypothermia causing the neonatal fat, which is composed of saturated fatty acids with a relatively high melting point, to crystallise.

The areas of fat necrosis seldom progress to atrophy, scarring or ulceration.

The prognosis of SCFN is good, with self-healing within 4 weeks,¹ but appreciable morbidity and mortality can result from the associated hypercalcaemia, the pathogenesis of which is not known. Various causes have been proposed for including osteoclast activation and increased production of 1,25 dihydroxyvitamin D3 by macrophages resulting in increased bone turnover following increased prostaglandin E activity.³

If the hypercalcaemia is high, systemic calcification,² hypertension and renal lithiasis⁴ are possible. Hypercalcaemia is usually noticed 4–6 weeks after the documentation of skin lesions and may cause metastatic

calcifications in the skin, heart, liver and inferior vena cava.⁵

Children with SCFN commonly present only with skin lesions with or without any other symptoms. Rarely, do they present with symptoms of hypercalcaemia such as irritability, weight loss, apathy, hypotonia and dehydration. Diagnosis is essentially clinical and a skin biopsy is not needed. If done, it will show fat necrosis and granulomatous infiltration with lymphocytes, macrophages, giant cells and calcium crystals with or without fibrosis.

Our child presented at 10 weeks of age with hypercalcaemia and skin calcification, probably the skin lesions were not noticed earlier. Metastatic calcifications of the heart, liver and inferior vena cava and thrombocytopenia were not seen in the present subject.

In our child with history of asphyxia, macrosomia and hypoglycaemia, the skin changes, the soft-tissue calcification in radiography and the hypercalcaemia, the diagnosis of a complication of SCFN was made even if the typical skin lesions were not reported.

Learning points

- ▶ SCNF is a transient disorder of the subcutaneous adipose tissue that develops after trauma or asphyxia during birth and may be associated with hypercalcaemia.
- ▶ Whereas the subcutaneous nodules of SCNF are usually self-limited, the hypercalcaemia, if undetected, may have a fatal outcome and can be life threatening if not treated adequately.
- ▶ This case is of interest as it presents some features of SCNF not previously described. Calcification of the tissues was seen radiologically.
- ▶ SCNF can cause soft-tissue calcification. It is important that radiologists be familiar with the clinical and imaging features of this unusual entity to facilitate a prompt diagnosis.

Competing interests None.

Patient consent Obtained.

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