**A rare case of spontaneous intrauterine skull fracture**

**Caso raro de fratura espontânea intrauterina do crânio**

Mariana Dória\*1, Catarina Viveiros\*2, Lia Rodrigues e Rodrigues3, Fátima Soares1

\*Co-primeiros autores

1 Serviço de Ginecologia/Obstetrícia, Unidade Local de Saúde de Matosinhos/Hospital Pedro Hispano, Matosinhos, Portugal

2 Serviço de Pediatria, Unidade Local de Saúde de Matosinhos/Hospital Pedro Hispano, Matosinhos, Portugal

3 Serviço de Neonatologia, Unidade Local de Saúde de Matosinhos/Hospital Pedro Hispano, Matosinhos, Portugal

Hospital Pedro Hispano, Rua Dr. Eduardo Torres, 4464-513 Senhora da Hora.

mariana.doria@ulsm.min-saude.pt

Spontaneous intrauterine skull fracture

**A rare case of spontaneous intrauterine skull fracture**

**Abstract**

Skull fractures are rare in newborns and normally caused by maternal abdominal trauma or complicated deliveries. However, in rare cases, these fractures are found in neonates born after an uneventful pregnancy and delivery. We report a case of a primigravida submitted to cesarean due to failure of descent and malposition of the fetal head. After birth, a right temporoparietal fracture and congenital muscular torticollis were diagnosed. Newborn’s blood tests showed hypocalcemia and relative hypoparathyroidism. Both mother and newborn presented low vitamin D levels. Serial imaging control showed gradual resolution of the lesions, with newborn being discharged at 10th day of life with vitamin D supplementation. This is an interesting case because the combination of three conditions - maternal and fetal hypovitaminosis D, congenital torticollis and malposition of the cephalic pole during labor –may have synergistically contributed to spontaneous intrauterine skull fracture.

**Resumo**

As fraturas do crânio são raras em recém-nascidos, sendo mais comummente causadas por trauma abdominal ou como complicação do parto. Contudo, em casos mais raros, estas fraturas são encontradas isoladamente, sem associação a intercorrências da gravidez ou do parto. Apresentamos o caso de uma primigesta submetida a cesariana por ausência de descida da apresentação e mau posicionamento da mesma no canal de parto. Após o nascimento, foi diagnosticada fratura temporoparietal direita e torcicolo congénito. Analiticamente, o recém-nascido apresentava hipocalcemia e hipoparatiroidismo relativo. Hipovitaminose D da díade mãe-recém nascido. Estudos imagiológicos seriados demonstraram resolução gradual das lesões, possibilitando a alta do recém-nascido ao 10º dia de vida com suplementação de vitamina D. Este caso é interessante porque se conjugam três condições– hipovitaminose D materna e fetal, torcicolo congénito e má orientação do polo cefálico – que, conjuntamente, podem ter contribuído para a ocorrência de fratura craniana intrauterina espontânea.

**Keywords:** skull fractures, newborn, hypocalcemia, vitamin D deficiency, congenital torticollis

**INTRODUCTION**

Skull fractures found in newborn can be linear, usually affecting the parietal bones, or depressed, the so called “ping-pong fractures”. Linear fractures, unlike depressed, generally are not accompanied by underlying brain lesions and do not require specific treatment.1

Cerebral computed tomography (CT) scan, with pediatric low-dose radiation protocol, is the gold standard for the diagnosis.2

Most common causes are maternal abdominal trauma during pregnancy and head injuries caused by instruments or obstetrical maneuvers in complicated deliveries. 3 However, in rare cases, these fractures are found in neonates born after an uneventful pregnancy and delivery and are, in these instances, called “spontaneous”. 4

Probably, these “spontaneous” fractures may result from continuous intrauterine mechanical forces upon the soft fetal skull caused by bony structures.5

Additionally, some vitamin deficits may result in a higher probability of fracture. Vitamin D plays a central role in calcium and phosphate homeostasis and bone metabolism. In the presence of vitamin D deficiency, the intestine is not capable of correct calcium and phosphate absorption, leading to secondary hyperparathyroidism.6

Severe deficits of vitamin D during pregnancy have been associated with compromise to skeletal fetal homeostasis, congenital rickets and fractures in the newborn, as well as neonatal hypocalcemic seizures, since the newborn levels of vitamin D are largely dependent on maternal vitamin D status.6 One of the signs of maternal vitamin D deficiency is craniotabes, a softening of skull bones, which turns the bone more prone to fractures.7 Moreover, maternal vitamin D insufficiency correlates with higher risk of pre-eclampsia, gestational diabetes, preterm birth or low birth weight.6

**CASE REPORT**

We report a case of a 31-year-old primigravid with no significant medical history. She took a multivitamin with 5 µg of vitamin D during pregnancy. The pregnancy was uneventful until 33 weeks, when she presented with threatened preterm labor and did a course of antenatal corticosteroids for fetal maturation. No trauma was reported during pregnancy.

She presented to our emergency department at 38 weeks and 4 days after premature rupture of membranes in labor. At admission, cervix was 2 cm dilated and 100% effaced with cephalic presentation. Cardiotocography was reassuring.

A cesarean was performed due to failure of descent and fetal malpresentation (no engagement and head hyperextension).No forceps or vacuum extractor were used.

Newborn Apgar score was 9/10 at 1 and 5 minutes, respectively. Weight appropriate for gestational age. Physical exam showed right hemicranium tumefaction of soft consistency at palpation, with crackling in the right temporo-parietal transition and left congenital muscular torticollis (CMT). Neurologic exam proper to gestational age. Skull x-ray suggested a fracture in the right temporoparietal region (image 1).

“Inserir Imagem 1 aqui”

CT scan confirmed the diagnosis showing a solution of continuity in the right parietal convexity, with misalignment of bony tops, compatible with a linear fracture and also 3 focus of haemorrhage: a right temporoparietal haemorrhagic contusion measuring 9.3 x 6.7 x 4.3 mm, a smaller one laterally and linear traces of adjacent sulcal subarachnoid haemorrhage (image 2).

“Inserir imagem 2 aqui”

Both mother and newborn presented hypocalcemia (8,6 mg/dl and 6,9 mg/dl) and vitamin D insufficiency (16 ng/ml and 11 ng/ml), respectively. Parathormone (PTH) values in the newborn were within normal range, although inappropriately low (45.8 pg/mL) in the presence of hypocalcemia. Newborn also presented with hyperphosfatemia (10,4 mg/dl) and hypomagnesemia (1,45 mg/dl).

“Inserir tabela nº1 aqui”

Serial brain CT scans (D3 and D8) were performed, with evidence of partial reabsorption of intra-axial haemorrhagic lesion and resolution of subarachnoid haemorrhage. Skeleton x-ray showed no evidence of additional fractures. Neurological exam was always normal.

Newborn was discharged at D10 with vitamin D and calcium supplementation and maintained multidisciplinary follow-up, with normal staturo-ponderal and psychomotor development. No sequels were identified.

**DISCUSSION**

The prevalence of spontaneous intrauterine skull fractures varies from 1 in 4000 to 1 in 10000 deliveries.8 Establishing etiology can be challenging, especially when there is no evidence of abdominal trauma, no use of instruments or after a cesarean section delivery.

There are few similar cases in literature, with the majority of studies describing depressed skull fractures. To our knowledge, this is the first study reporting a linear fracture in a non-traumatic delivery of a newborn with documented hypovitaminosis D. There are no described cases of fractures attributed to vitamin D deficiency, although it has been cited as a cause of congenital rickets and have been associated with suboptimal fetal growth and reduced fetal skull mineralization. 9

Vitamin D deficiency screening is based on the measurement of serum concentrations of 25-hydroxivitamin D (25(OH)D).6 According to 2016 Global Consensus recommendations, values comprised between 20-100 ng/mL are considered normal, with insufficiency in the range 12-20 ng/ml and deficiency when values are less than 12 ng/ml.10 There is no consensus on an optimal vitamin D level for the pregnancy status and remains unclear whether vitamin D supplementation should be routinely recommended in pregnancy or if it should be reserved for high-risk groups.11

Vitamin D deficiency in neonates secondary to maternal deficiency may lead to hypocalcemia. PTH should increase in response to low serum calcium concentrations, which was not found in our case, with PTH levels abnormally low for the level of hypocalcemia.This suggests a relative hypoparathyroidism, which may also have contributed to newborn hypocalcemia.12

Birth trauma has been pointed as the main etiological factor for congenital muscular torticollis (CMT).13 However, CMT has also been described in cases where no trauma was reported. In our case, congenital muscular torticollis may have been caused by an abnormal intrauterine position during the third trimester which resulted in sternocleidomastoid muscle injury and deformity, as proposed by Stellwagen *et al.*14

The fetal malpresentation, probably due to CMT, may have predisposed the fetus skull to collision with maternal body structures, especially during uterine contractions. The vitamin D deficit and hypocalcemia may have contributed to softening of the fetus skull, predisposing it to fracture.

This is a challenging case in which the combination of three conditions - maternal and fetal hypovitaminosis D, congenital torticollis and fetal malpresentation – may have synergistically contributed to spontaneous intrauterine skull fracture.

**CONFLITS OF INTEREST**

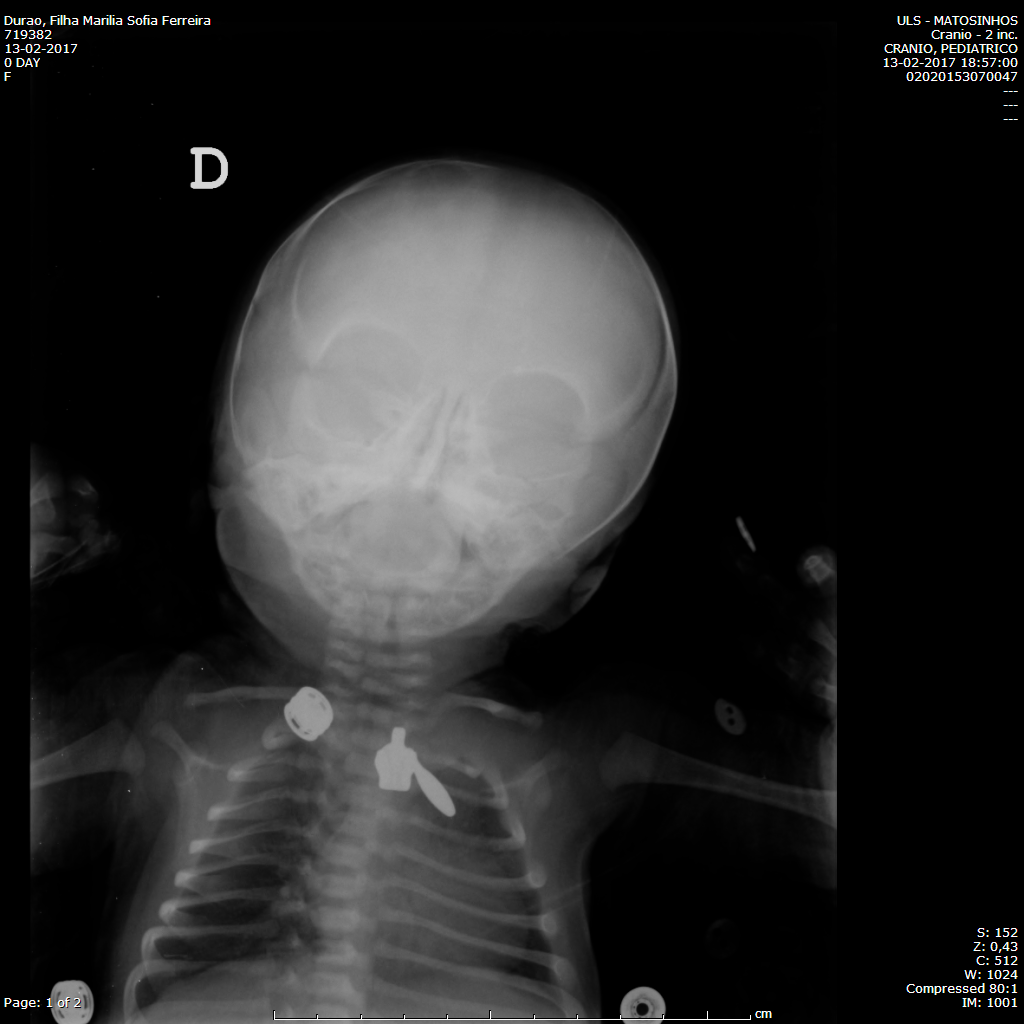
The authors declare that there are no conflicts of interest.

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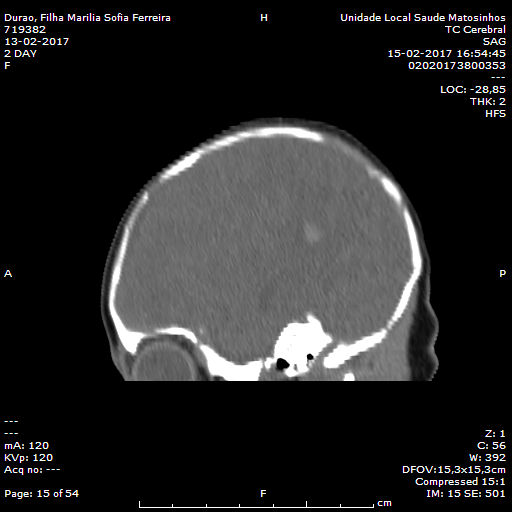
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**Image 1 -** Skull x-ray: Right temporoparietal fracture



**Image 2 –** Brain Computed Tomography scan in D2. Visible 3 focus of hemorrhage.

Table 1: Blood laboratory findings of the newborn and his mother. Reference values are presented inside parentheses.

|  |  |  |
| --- | --- | --- |
| Parameter | Newborn | Mother |
| Calcium (mg/dL) | 6,9 (7,6-10,4) | 8,6 (8,9 -10) |
| Vitamin D (pg/ml) | 11 (16-65) | 16 (30-100) |
| PTH (pg/ml) | 45,8 (15-65) | --- |
| Phosphorus (mg/dl) | 10,4 (4,5-9) | --- |
| Magnesium (mg/dl) | 1,45 (1.6-2,3) | ---- |

PTH: parathyroid hormone.

Adapted from Harriet lane Handbook, 19th edition