

Case Report

PERINATAL LEFT FEMORAL SHAFT FRACTURE IN A NEWBORN FEMALE BORN IN EMERGENCY CAESARIAN SECTION: A CASE REPORT AND LITERATURE REVIEW OF THE INCIDENCE RATE

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ARTICLE INFO

Article history:

Received 28 Jan 2023

Accepted 20 Mar 2023

Published 21 Apr 2023

Keywords:

Birth-related fractures, delivery trauma, perinatal femoral fractures, Caesarian section, skin traction, Bryant's traction.

ABSTRACT

Perinatal femoral fractures (PFF) are extremely rare birth-related fractures. These fractures are reported as case reports or small case series. The incidence is 0.024/0.077 of 1000 newborns. Currently the gold standard treatment has not been well codified. This case report gives an adjunctive point of view of diagnosis and treatment about these rare cases. We reported a case of a newborn female with a few hours of life that presented a left shaft fracture of the femur birth-related to cesarean born (AO PCCF 32D/4.1). The patient was treated with Bryant's traction for 21 days. Hard bone callus formation was seen after 9 days from the beginning of the treatment. The fracture showed clinical and radiologic healing with abundant callus formation within 3 weeks after removing the traction; no further immobilization was necessary. No severe complications were observed during hospitalization. At the final follow-up no deformity, shortening, or other significant complications were observed. Given the rarity of this lesion, we believe it is relevant to report these cases to improve the knowledge on the treatment and the clinical outcome of this rare pathology. It should also be noted that this type of trauma can occur in any center and at any time and it is crucial to have clear guidelines on the most accepted therapeutic strategies. An early diagnosis and an optimal treatment allow to obtain better outcomes and optimal newborn recovery.

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1. Introduction

Shaft fractures of the femur during infancy can occur in connection with the birth. In 1922, Ehrenfest et al. (1) described the first perinatal femoral fracture (PFF), induced by the problematic extraction of a large newborn during a cesarean section (CS).

Birth-related fractures are all those fractures diagnosed in the first week of life, without any postnatal trauma (2). These fractures result from trauma during the partum and the most common sites are the clavicle, the humerus and, rarely, the femur.

Perinatal shaft fractures are rare and reported as case reports or small case series. The incidence of neonatal fractures is variable and likely underestimated, ranging between 0.1 and 10.5 per 1000 live births (2, 3, 4).

In a study analyzing the incidence of fractures in infants born in Sweden from 1997 to 2014 (3), the incidence for birth-related femur shaft was 0.024 per 1000 children and that for birth-related humerus shaft fractures was 0.101 per 1000 children. The incidence was 0.154 per 1000 children for later femur shaft fractures and 0.073 per 1000 children for later humerus shaft fractures, according to Tokar et al. in 2009 (5). In a study conducted in the UK in 2019, on 87461 consecutive live births, only one perinatal femoral fracture (PFF) was identified (6).

According to this data, with 404134 newborns in 2020 in Italy, we can estimate an incidence of 9.6 new cases of birth-related femur fractures per year in Italy.

Birth-related femur shaft fracture was associated with shoulder dystocia, cesarean, multiple births, breech, preterm, and small-for-gestational-age.

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DOI: 10.3269/1970-5492.2023.18.9

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A bone fragility diagnosis was recorded in 5% of those with birth-related or later femur shaft fractures (5).

Associated risk factors include some different conditions, such as malpresentation, low birth weight, macrosomia, prematurity, osteogenesis imperfecta, disuse osteoporosis following immobilization, CS (7), harrowing extraction (breech presentation) and impacted foot in the pelvis or any previous uterine surgery leading to a tight uterine incision.

In 2009, Toker et al. (5) observed that the incidence of femur fracture was 0.077 per 1000 deliveries, and the incidence of femur fracture during CS was 0.308 per 1000 CS, concluding that Cesarean section increases the risk of femur fractures ($P < 0.001$) with an odds ratio of 11.26 (confidence interval 3.97-31.97).

This kind of diagnosis usually occurs immediately after the delivery but, in some cases, can be delayed with milder symptoms (4). The most common clinical characteristics are a swollen, tender, warm thigh held without motion. Often, clinicians or obstetricians feel a crackle during the partum. Then, a radiographic assessment confirms the diagnosis, while MRI and ultrasound are often functional to make a differential PFF diagnosis (8, 9).

Since initially proposed in 1873, Bryant's traction seems to be a useful method to treat PFF (10)

Bryant wrote in his textbook that in 1870 he was the first to use a form of overhead traction at Guy's Hospital in London; this method was utilized for fractures of the femoral shaft in children. In his series, he reported that the youngest was 8 months and the oldest 5 years of age. In 1882, Kummel (11) reported 40 cases that he treated similarly from 6 hours of age to 4 years. In 1960 Lidge (12) described the complications of Bryant's traction. In his study, most of the complications occurred within 48 to 72 hours after the treatment and were considerable skin irritation, gangrene and peroneal palsy.

If a complication occurs, the traction should be removed immediately and a different type of treatment is to be adopted, such as a hip spica cast wedging it to correct angulation or rotation deformity. Other successful PFF treatments to immobilize the femoral shaft have been described like Pavlik harness, gallow's traction, and spica cast splinted by two tongue depressors.

2. Material and methods

We reported a case of a newborn female, with a few hours of life, that underwent a left femoral shaft fracture after a cesarean section born (AO PCCF 32D/4.1). The clinical course is reported, a literature review is described to establish the real incidence of these rare lesions, the risk factors and the common technique of treatment.

A literature review was conducted in PubMed, EMBASE and Google Scholar with the keywords "Perinatal", "Fracture", "Newborn", "Caesarian section". 30 articles were reviewed by three reviewers. 16 were excluded because they did not have epidemiological data, and 14 were selected. All these articles contained case reports, case series and epidemiological studies or review and are listed in the References (1- 8, 13-18). Epidemiological data were analyzed to establish the most accurate possible current incidence rate.

3. Results

We report the case of a newborn girl, P.G., who was born early in the morning and hospitalized in the neonatal intensive care unit (NICU). The newborn was born at term, the mode of delivery was an emergency Caesarean section (CS), starting in the breech position, length: 47 cm, circumference 34 cm, weight: 3,170 gr , gestational age: 38 weeks + 4 days.

During the CS, at the time of the extraction of the newborn, the gynaecologists had noticed an unusual noise. This alarmed them and ask for Orthopaedic consultation.

The newborn was very agitated and showed preternatural mobility of the left thigh, angular deformity, pain, no nervous or vascular deficit (Figure1).

A radiographic survey was requested immediately. The x-ray examination showed a fracture of the femur (Figure 2).



Figure 1. Clinical Examination.



Figure 2. X-ray of the left femoral shaft fracture At 1 day post CS

According to the AO Pediatric Comprehensive Classification of Long Bone Fractures (PCCF), the fracture was ranked as 32-D/4.1, diaphyseal complete transverse $<30^\circ$. The time from delivery to fracture diagnosis was 6 hours.

Initially, a Pavlik harness was made but subsequently the definitive treatment was a Bryant's traction in the incubator in a specialized center, due to unavailability of Pavlik harness in good time. Hard bone callus formation was seen after 9 days from the beginning of the treatment. (Figure 3).

The fracture showed clinical and radiologic healing with abundant callus formation within 3 weeks after removing the traction (figure 4); no further immobilization was necessary.

No severe complications were observed during hospitalization. At the final follow-up no deformity, shortening, or other significant complications were recorded.

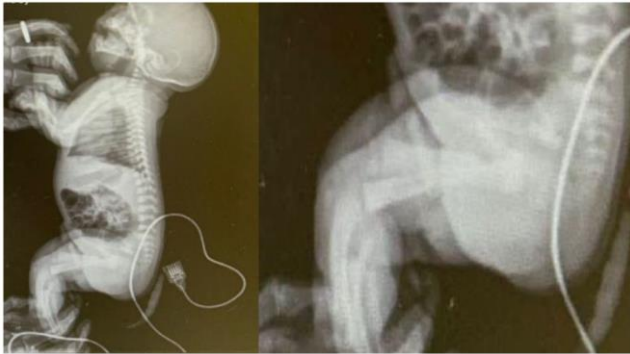


Figure 3. X-ray control in lateral view at 9 days.



Figure 4. Xray control at 3 weeks.

4. Discussion

PFF is a very rare trauma. According to the recent literature, the newborns in Italy in 2020 were 400000; and it is estimated an incidence of 9.6 new cases of birth-related femur fractures per year in Italy. The data analyzed in the current literature confirmed that prematurity, low birth weight, and CS seem to be important predisposing factors (4, 7).

A Portuguese study on 7364 newborns found 76 fractures classified as birth trauma and the most common fractured bones were: the clavicle (79%), followed by parietal (7%), humerus (5%), and femur (4%) (13). Morris et al. (14) reported a PFF incidence of 0.13/1000 live births. Basha et al. (2) reported an incidence of 0.17/1000 live births. In the past literature, CS was considered to reduce the risk of fracture (15). However, according to some studies (4, 5), the considerable number of maneuvers and tractions performed during a CS can result in fractures, indeed the incidence is 0,077 per 1000, and the incidence increases to 0.308 in emergency Caesarean sections (5).

Vasa et al. (4) emphasize that CS does not reduce the traumatic morbidity in average-sized infants to zero. Burnes and Van Geem (15) and Alexander et al. (16) reported midshaft distal metaphyseal fractures after CS.

Many studies report some relationship between femur fractures and CS with a breech presentation (5,14). Linder et al. (2013) reported an increased risk of birth trauma for deliveries during at risk hours (between 4 pm and 8 am) and weekends, related to the iatrogenic nature of the lesion.

Several authors have shown an inverse association between the rate of birth trauma and the level of experience and training of the attending staff. (17).

In a case series at ten-year follow up Pavone et al. in 2020 reported only 8 cases; that support that PFFs are very rare and confirm that factors such as prematurity, low birth weight, and CS are predisposing factors to PFF (18).

Our case had a breech presentation and CS, that in literature are considered the main predisposing factors. The main cause seems to be a mistake in the technique of extracting the newborn during CS and the tendency to perform very small skin incisions. The case of this study was unrelated to prematurity or low birth weight.

The chosen treatment was the use of Bryant's skin traction and our results confirm the effectiveness of this treatment to achieve optimal outcomes. The choice of traction was done for the delay in having a Pavlik harness and seemed appropriate given the placement of the newborn in the incubator. It's important in the case of Bryant's traction careful daily observation of the patient for the possible appearance of skin irritation, gangrene and peroneal palsy. In this case it's necessary to reduce traction immediately and made a spica hip cast. The Bryant's traction has the possibility of complications during the treatment, but careful observation makes it possible to avoid them. Further no complication of any kind has been observed in our case. This underlines the validity of the treatment and its reproducibility.

Given the rarity of this lesion, we believe it is necessary to report each case in the literature to improve the knowledge on the best modality of treatment. It should also be noted that this type of trauma can occur in any center and at any time and it is crucial to have clear ideas about the diagnosis and treatment strategies. Clinical outcome in this type of patients are very good and angular deficits are easily corrected.

An early diagnosis and an optimal management allow to improve the results and newborn recovery and to prevent complications.

5. Conclusions

Our case report describes a very rare neonatal traumatic lesion and the modality of treatment. This case supports the use of Bryant's skin traction to achieve optimal outcomes and describe how to prevent complications. This case report, even if on a single patient, due to the injury's paucity, concur to achieve more data in the literature on this rare lesion that occur only in 9 cases per year in Italy and with an incidence rate around 0.024/0.077 per 1000 born in the world.

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