Case Report

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Spontaneous fetal femoral fracture: a case report and literature review

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Abstract

Spontaneous fetal femoral fractures are uncommon in the paediatric setting. The major clinical presentations of a spontaneous fetal femoral fracture are femoral angulation, shortness of the femur and even a marked fracture line. This case report describes a spontaneous fetal femoral fracture of the right femur, which was detected by routine ultrasonography during the 19th week of gestation in a 24-year-old woman. On routine follow-up visits, the angulation of the right femur in the fetus gradually improved. A caesarean section was undertaken at 39 weeks +5 days of gestation and an X-ray was taken on the second day after birth, which showed that the fracture had healed and the callus had been absorbed. The lengths of the two femurs of the baby were not equal; the right femur was 84 mm, which was 11 mm shorter than the left femur. In cases like this, postnatal follow-up is essential so that an operation can be carried out in a timely manner when the deformity is apparent.

Keywords

Spontaneous, fetal femoral fracture, femur

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Introduction

Spontaneous fetal femoral fracture is defined as the occurrence of intrauterine fractures in the absence of traumatic factors, bone fragility and other congenital diseases. Few cases have been reported, especially in the femur. A previous report published in 2003 described a case of spontaneous fetal femoral fracture.[1] To date,

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there have been only four reports recorded on the PubMed database.[1–4]

This current report describes the case of a spontaneous fracture of the femoral shaft and presents a review of the published literature on the epidemiology, pathogenesis, diagnosis, treatment and prognosis of this rare condition.

Case report

A 24-year-old woman presented at the Department of Paediatric Surgery, Nanjing Medical University, Affiliated Wuxi People's Hospital, Wuxi, China during the 19th week of gestation in July 2016. During the routine ultrasound examination, the right femur of the baby was observed to be angled and it measured 36 mm long, which was approximately 5 mm shorter than the left femur (Figure 1). All other bones had normal ossification and there were no fetal malformations. The primipara mother was in a healthy condition before and during the pregnancy; and the presence of uterine myoma, type 2 diabetes mellitus and nuchal translucency on antenatal ultrasound scans had been excluded. There was no family history of neurofibromatosis or bone dysplasias. After a period of time, the fracture angulation gradually improved and callus formed at the fracture site. During an ultrasound examination at 27 weeks, the femoral lengths were 58 mm and 62 mm for the right and left femurs, respectively; and at 32 weeks, they were 61 mm and 69 mm, respectively. A caesarean section was undertaken at 39 weeks +5 days of gestation. The newborn was a boy and he weighed 3350 g. The Apgar score and the appearance of the newborn baby were normal. An X-ray taken on the second day after birth showed that the fracture had healed and the callus had been absorbed. The lengths of the two femurs of the baby were not equal; the right femur was 84 mm, which was 11 mm shorter than the left femur (Figure 2).

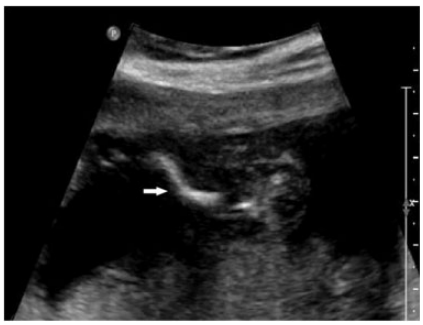

Figure 1. A routine ultrasound image undertaken in a 24-year-old woman during the 19th week of gestation demonstrated that the right femur of the baby was angled (arrow) and it measured 36 mm long, which was approximately 5 mm shorter than the left femur.

The levels of alkaline phosphatase, calcium and vitamin D_3 were all normal. Written informed consent was obtained from the patient to publish this case report.

Discussion

The incidence rate of fetal fracture is low due to the amniotic fluid environment, which can buffer trauma to a certain extent.[5] However, cases of fetal fracture have been reported, mainly affecting the skull,[6,7] spine,[8] forearm[9] and femoral shaft.[10]

The major causes of fetal fracture are as follows. First, trauma, which is often associated with pelvic fractures in the mother, life-threatening haemodynamic changes,[11] and even fetal death or spontaneous abortion, accounts for 65% of fetal fractures.[8,9] Secondly, skeletal diseases such as osteogenesis imperfecta, can be associated with fetal fracture.[12] Other factors include maternal characteristics such as age, primipara, uterine myomas and diabetes mellitus; as well as pregnancy-related characteristics such as high or low birth weight, a difficult delivery, and malpresentation.[13–15] Thirdly, caesarean section, which is suited for situations such as older mothers, primipara status, presence of

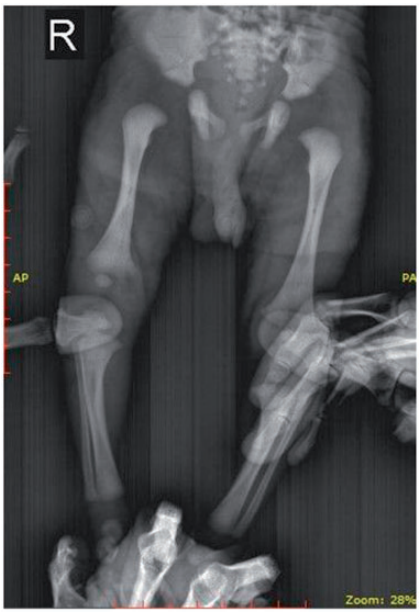

Figure 2. An X-ray was taken on the second day after the birth of a healthy male baby by caesarean section at 39 weeks +5 days of gestation. The X-ray showed that the fracture in the right femur had healed and the callus had been absorbed. The length of the right femur was 84 mm compared with a length of 95 mm for the left femur.

uterine fibroids and breech presentation, can increase the risk of femoral fractures.[15,16] Finally, spontaneous fetal femoral fracture, which is an extremely rare condition diagnosed on the basis of ruling out all of the factors described above. Several cases have been reported including skull fractures,[17,18] femoral fractures,[1–4] and fractures of the tibia and fibula.[19]

Spontaneous fetal femoral fracture is extremely rare in the paediatric population and to date only four cases have been reported in the English literature.[1–4] Three of the four cases were male (sex of the fourth case remains unknown) and the fractures were located in the middle of the right femur. This present case is also a male with a right femoral fracture, which suggests that this might not just be a coincidence, but that there might be factors related to sex and the anatomical site that increases the risk of a fracture. As the mothers were both primipara and multipara, this suggests that there is little relationship with maternal pregnancy history and fetal femoral fracture.

Currently, the pathogenesis of spontaneous fetal femoral fracture remains unclear. Temporary brittle bone disease is considered as a cause of unexplained fractures in the first year of life.[20] However, this is not a universally accepted idea. Other hypotheses such as developmental deficiency in a segment in the middle of the bone also seem unlikely.[21] Evaluation of the published case reports demonstrated that the fractures all occurred in middle to late pregnancy.[1–4] Except for one case that was not reported, all three fetal femoral fractures occurred in males and were located in the middle of the right femur, which might indicate a relationship with some intrauterine developmental processes.[1–4] With regard to the sex of the fetus, in our opinion, the low levels of some sex-related hormones such as oestradiol, which have been shown to be correlated with femur strengthen in adult males, might lead to a corresponding weakness in the male fetal femur.[22] As to the location along the femur, the shaft of the femur develops faster than both ends, making this site more fragile. In addition, the site of the fracture might be related to the position of the femur *in utero* as the right femur always lays over the left femur, which could result in a lever/fulcrum effect. Unfortunately, none of the four case reports described the position of the fetus *in utero*.[1–4]

The diagnosis of spontaneous fetal femoral fracture is not difficult and is mainly based on routine ultrasonography during pregnancy. The major clinical presentations are femoral angulation and shortness of the femur or even a marked fracture line diagnosed by ultrasonography. It should be noted that the diagnosis of spontaneous

Table 1. Case reports describing spontaneous fetal femoral fractures.[1–4]

Author, year	First visit	Sex	Anatomical location	Treatment	Longest follow-up	Outcome
Senanayake, 2003[1]	Week 26	Male	Right femur	Surgery	6 years	A 3-cm deficit
Arioz, 2008[2]	Week 34	Male	Right femur	Follow-up	ND	A 2-cm deficit
Sahin, 2014[3]	Week 25	Male	Right femur	Follow-up	ND	Healed *in utero*
Hwang, 2009[4]	Week 23	ND	Right femur	Follow-up	9 months	Grew appropriately

ND, not described.

fetal femoral fracture can only be made when a traumatic history and skeletal diseases have been excluded. It should be emphasized that there might be the possibility of a forgotten trauma during pregnancy, so this must be ruled out conclusively, as even mild traumas can cause intrauterine fractures in the skull and other bones.[23] The mothers are usually healthy, as was the case for the mother described in this current report.

The main differential diagnosis of a congenital short femur was first reported in 1983.[24] This condition can present as a small bone with a normal shape through to the whole bone being missing. The differential characteristics between spontaneous fetal femoral fracture and congenital short femur are the femoral angulation, fracture line and the callus development during the course of the fracture healing.[2] In addition, patients with spontaneous fetal femoral fracture are free of decalcification and all bones including the fractured femurs continue to grow approximately parallel to, but below, the 10th centile line.[1]

The most appropriate mode of delivery for fetuses with spontaneous fetal femoral fracture remains controversial and may depend on the extent of mineralization of the bones.[1,2] Among the four cases reported previously, two had a vaginal delivery and two had caesarean sections,[1–4] as did the case reported here. All of the newborns, including the current patient, were in good birth condition with normal levels of serum calcium and alkaline phosphatase.[1–4]

The treatment and prognosis of spontaneous fetal femoral fracture depends on the level of healing that occurs *in utero* (Table 1). Of these four cases,[1–4] the newborn with the best recovery only had mild femoral angulation without obvious callus due to quick regeneration of the fetal tissue.[25] In contrast, the newborn with the worst recovery had a right femur that was 3 cm shorter than the left femur several years later according to the follow-up and underwent corrective surgery as a consequence. Generally, treatment is explicit and straightforward. Long-term follow-up is essential so that corrective surgery can be undertaken in a timely manner when the deformity is severe. If there is no apparent difference between the lengths of the two femurs, then patients can live normally.

In conclusion, spontaneous fetal femoral fracture is an extremely rare condition diagnosed on the basis of ruling out a traumatic history and skeletal diseases. Postnatal follow-up should be taken seriously and corrective surgery undertaken as and when required. Improvements in the diagnosis and understanding of this condition should result in more cases being identified and more research into its pathogenesis.

Declaration of conflicting interests

The all authors declare that there are no conflicts of interest.

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