

Talus Bipartitus Etiology – Is Neonatal Infection Involved?

Etiologie talus bipartitus – jaký podíl má neonatální infekce?

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SUMMARY

Talus bipartitus is a rare skeletal variation. Several causative factors have been proposed, but none of them seem to be convincing. We hypothesize that talus bipartitus may result from ossification disruption in neonatal period caused possibly by an infection. The observations supporting this link are discussed. The hypothesis is supported by two cases with symptomatic talus bipartitus illustrating the postulated connection.

Key words: ankle, pain etiology, talus abnormalities, talus bipartitus.

INTRODUCTION

Talus bipartitus is a rare skeletal variation, though a surge in the number of reported patients was observed recently. Several authors have proposed causative factors, but none of them seem to be convincing. We hypothesize that talus bipartitus may result from ossification abnormalities related to infection in neonatal period. We present two patients with symptomatic talus bipartitus illustrating the postulated connection.

CASE SERIES

Patient 1

The first patient was a 28-year-old male born preterm (32nd week) from a twin pregnancy with a birthweight of 2100 g. Neonatal sepsis developed in both children and the sibling died. Two years before presentation the patient underwent total hip arthroplasty (THA) for postinfectious deformation (Fig. 1a). After THA, the talus bipartitus of the ipsilateral extremity became symptomatic (Fig. 1b). The talus in this patient was actually multiparted (Fig. 1c). Patient didn't recall any trauma and symptoms onset was gradual. Patient received subtalar arthrodesis with bone grafting due to unremitting symptoms.

Patient 2

The second patient was a 21-year-old female born in the 36th week of gestation with low birth weight (1800 g). The patient was treated for infection of the ankle region during the neonatal period. At this time, a fistula was present leaving a visible scar (Fig. 2a). She became symptomatic in the last 2 years after attending

driving lessons, at which time talus bipartitus was diagnosed in X-ray (Fig. 2b). The patient denied trauma to the region. She was also treated by subtalar fusion (Fig. 2c).

DISCUSSION

The origin of talus bipartitus is not clear (14). Several etiologies have been proposed, but none of them are convincing.

Some anatomists have proposed that talus bipartitus represents a congenital anomaly or atavistic trait, but this has been questioned by others (2). A bipartite talus was also proposed to result from the secondary center of ossification (12, 17). However, a secondary center of ossification is present in the os trigonum, not the talus itself, which ossifies from a single center of ossification (14).

Posttraumatic etiology (16, 19) has been proposed for talus bipartitus. If this was the case the cleft would represent pseudoarthrosis. However, this seems not to be true for several reasons. Firstly many patients (including here presented) denied trauma. Secondly gradual onset of symptoms, observed also in our patients, is more common (14). Finally in our Patient 2 and in one other case (17) the talus was actually multiparted, which strongly contradicts posttraumatic theory.

With all of the described etiologies doubtful the origin of talus bipartitus remains speculative.

We think that, at least in some cases, talus bipartitus represents a disturbance in ossification in neonatal period caused by infection. Our presented two cases of infection add to one already described – a 15-year-old girl had an infection of the tissues overlying the affected ankle 8 days after birth (17).



Fig. 1. Pelvic X-ray of Patient 1 revealing postinfectious proximal femoral deformation (a) and an MR image showing a multiparted talus (b). The 3D-CT reconstruction from above illustrating multipartition of the talar body (c).



Fig. 2. Clinical picture of the fistula scar in Patient 2 (a), X-ray showing a bipartite talus (b) and postoperative X-ray with subtalar fusion (c).

The complications of neonatal infection may present as growth deformity, pathologic fracture, and osteonecrosis (8). The disturbance in ossification pattern is observed, with ossific nuclei irregular, fragmented, delayed or even absent (3). Typical localizations include the hip, knee, and arm joints, though less common sites have also been described (1, 3, 5). Bilateral involvement in neonatal sepsis is not uncommon (3). Similarly, there is at least one case of bilateral talus bipartitus reported (15).

Bacteria or signs of active inflammation have not been observed in talus bipartitus histologic examinations (6, 14). This would seem to refute postinfectious theory. Such observation is, however, typical for other postneonatal sepsis deformations investigated many years after the infection (10, 11).

Ossification of the talus commences after eighth gestational month. In low-birthweight babies the process is even slower and it occurs after delivery (9). Both our patients were born with low weight, therefore we can assume that the talus was not ossified at the time of delivery in either of them. Infection affecting unossified talus was capable of interrupting the process in the manner similar to the one described for humerus (3). Recovery and further development resulted in multiparted talar body.

The postinfectious link of talus bipartitus could also explain the rapid surge in the number of observed patients. Five cases were reported in the whole of the 20th century, but already twenty cases have been reported since 2000 (4, 6, 7, 12, 14, 16, 18). This surge could be explained by improvements in neonatal care leading to the survival of children with neonatal sepsis who would previously have died (as exemplified by Patient 1's sibling).

With the patients described above there are three patients with neonatal infection coinciding with development of talus bipartitus. Two of these patients actually developed multiparted talus, which makes them only two multiparted tali described in literature.

The infection link to talus bipartitus may not be direct. The ossification deformity may be mediated by vascular occlusion and microemboli caused by infection (13).

We are aware that the disadvantage of our theory is its speculative character and that it will be difficult to directly prove. It would require prospective study of substantial cohort to spot the development of such deformation. However, with all other proposed origins doubtful, we think that the ossification disruption caused directly or indirectly by infection is the best available explanation of talus bipartitus in our patients.

CONCLUSIONS

We conclude that at least some cases of talus bipartitus are related to ossification interruption caused by neonatal infection.

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