Avascular necrosis of talus simulating juvenile idiopathic arthritis

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Abstract
Avascular necrosis after fracture of the talus is well recognized, but is very rare in children. We report a case of avascular necrosis of the talus simulating juvenile idiopathic arthritis. The diagnosis was confirmed by open biopsy, and non-operative treatment was performed. The patient was asymptomatic with full range of motion postoperatively for two years. We hypothesize that avascular necrosis of the talus without fracture and dislocation might be caused by soft tissue damage around the talus with repeated hyperflexion of the ankle joint and slow arterial occlusion.

Keywords: Talus, Juvenile, Avascular necrosis

Abbreviations: JIA: Juvenile Idiopathic Arthritis; Hb: Hemoglobin; WBC: White Blood Cell; CRP: C-Reactive Protein; ACPA: Anti-Cyclic Citrullinated Peptide Antibody; MRI: Magnetic Resonance Imaging; CT: Computed Tomography; ROM: Range Of Motion.

Level of Evidence: Level V
INTRODUCTION

It is sometimes difficult to differentiate juvenile idiopathic arthritis (JIA) from conditions such as infections, tumors, and reactive arthritis [1,2]. We encountered a case of avascular necrosis of the talus simulating juvenile idiopathic arthritis. Avascular necrosis of the talus after severe ankle injury is well recognized [3]. Fractures of the talus are extremely rare in children, and the reported incidence varies between 0.01 and 0.08% [4-6]. To the best of our knowledge, there have been no reports of avascular necrosis of the talus without fracture and dislocation in children. Here we report a case of avascular necrosis of the talus without fracture and dislocation.

CASE REPORT

A two-year-old child developed swelling in his left ankle and limped while walking, and thus a physician was consulted. The physician could not detect the cause of his condition and referred him to us. His parents stated that he had swelling in his left ankle and was limping. He had no systemic symptoms and no remarkable medical or family history.

Regarding the physical examination, swelling, redness, localized heat, and effusion of the left ankle joint were observed. The range of his ankle motion was 10° dorsiflexion to 40° plantarflexion, and there were no signs of damage to ligaments. No swelling of the other joint was observed.

Laboratory tests revealed that Hb was 11.5 g/dl, WBC was 15300, and CRP was 1.17 mg/dl. Rheumatoid factor, anti-cyclic citrullinated peptide antibody (ACPA), and anti-nuclear antibodies were negative. Aspirated synovial fluid was bloody, and bacterial culture was negative. X-ray showed slight osteosclerosis of the talus of the left ankle (Fig. 1). MRI showed joint effusion and an edematous area in the bone marrow of the left talus, with no obvious abnormalities of articular cartilage (Fig. 2). One week after aspiration of joint fluid, CRP was 0.04 mg/dl, and no symptoms were found. However, X-ray showed bone defects and cavity formation in the talus of the left ankle three weeks later (Fig. 3). CT also demonstrated bone defects and cavity formation in the talus (Fig. 4). MRI showed expansion of a low intensity area in the posterior lesion of the left talus (Fig. 5).

Histological examination revealed osteonecrosis and mild chronic non-specific synovitis, including proliferation of lymphocytes and the presence of granulation tissue. These findings were highly suggestive of idiopathic or posttraumatic avascular necrosis. We then discussed the patient’s medical history with his parents again. They informed us that they recalled that his ankle joint may have once been hyperplantarflexed continuously by sliding over grass in a sleigh, but that he had no symptoms one month after the accident. From this medical history, we diagnosed this case to be posttraumatic avascular necrosis of the talus.

After open biopsy, ankle-foot orthosis was applied to the right leg for two months. ROM exercise and partial weight-bearing were started with a lower leg brace.

Two years after open biopsy, X-ray showed the disappearance of osteosclerosis and remodeling of the posterior portion of the talus (Fig. 6). MRI showed cortical defects of the bone marrow in the posterior lesion of the left talus, with no obvious abnormalities of articular cartilage (Fig. 7).

The patient was asymptomatic with full range of motion postoperatively for two years.
We reported a case of avascular necrosis of the talus without fracture simulating juvenile idiopathic arthritis. The diagnosis was confirmed by open biopsy. We hypothesize that avascular necrosis of the talus without fracture and dislocation might be caused by soft tissue damage around the talus with repeated hyperflexion of the ankle joint and slow arterial occlusion.

CONCLUSION

We reported a case of avascular necrosis of the talus without fracture simulating juvenile idiopathic arthritis. The diagnosis was confirmed by open biopsy. We hypothesize that avascular necrosis of the talus without fracture and dislocation might be caused by soft tissue damage around the talus with repeated hyperflexion of the ankle joint and slow arterial occlusion.

References