

Juvenile Osteochondrosis of the Cuneiform Bones

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ABSTRACT

Osteochondrosis of the tarsal and metatarsal bones is frequent in children. However, involvement of a cuneiform bone is a very rare entity with only scant reports in the world medical literature. The purpose of this paper is to retrospectively estimate the incidence rate of juvenile osteochondrosis of the cuneiform bones at our institution and to review the relevant publications.

Key words: Cuneiform, juvenile, osteochondrosis

REVIEW

Pedal osteochondrosis may cause pain and/or limping in children. The most commonly affected sites include the calcaneal apophysis (Sever's disease), the tarsal navicular (Köhler's disease), the second metatarsal head (Freiberg's infraction or Köhler's disease II), and the apophysis of the fifth metatarsal base (Iselin's disease). Osteochondrosis of the cuneiform bones is apparently a very rare entity.^[1-5]

In 1930, Harbin and Zollinger were the first to report a patient with osteochondrosis of the medial (first or internal) cuneiform.^[6] Buchman, in 1933, described two children with osteochondrosis involving the medial cuneiform.^[7] Haboush, Meilstrup, and O'Donoghue *et al.* reported, respectively, a case with bilateral involvement of the medial cuneiform and abnormal navicular on plain radiographs.^[8-10] Hicks and Smyth were the first to report a unilateral osteochondrosis of the intermediate cuneiform.^[11,12] Breck was able to diagnose 11 patients with osteochondrosis of the medial cuneiform and 9 with osteochondrosis of the intermediate cuneiform among 1000 patients with osteochondroses.^[13] Since then, nine more children with osteochondrosis of the medial and six with osteochondrosis of the intermediate cuneiform bones were reported in the English literature.^[14-29] Mubarak was the only one to document a case with osteochondrosis of the lateral cuneiform.^[30]

Osteochondrosis is defined as an idiopathic syndrome characterized by abnormal endochondral ossification occurring in a previously normal growth mechanism. It is usually due to a vascular insult, leading to secondary osteonecrosis. The precise etiology of osteochondrosis remains unknown since multiple contributing etiologic factors, such as microtrauma, rapid growth, genetic, and/or hormonal factors, may be involved. It may be prudent to consider that the early onset of ossification of the cuneiform bones, before the mechanical stresses of walking on the developing center of ossification are incurred, may be related to the low incidence of cuneiform osteochondrosis in children. The radiographic changes closely resemble those of osteonecrosis (avascular, aseptic, or ischemic bone necrosis). The term osteochondrosis is preferred than that of osteochondritis, which more specifically refers to infection or inflammation of bone and cartilage. Although the term osteochondrosis is the most precise nomenclature to describe the lesion, other definitions, such as osteochondritis, osteonecrosis, and avascular necrosis, are also occasionally used.^[25,26,31-35]

The average age of the patients in the previously published reports ranged from 2.5 to 8 years, although it mainly appears between 4 and 6 years of age, boys were more frequently affected and cases with a bilateral involvement were more commonly reported. Pain and/or limping were the predominant symptoms in the majority of cases, while in the asymptomatic cases, the diagnosis was based on the

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coincidental radiographic examination for foot injuries or abnormalities. The most common finding of the physical examination is tenderness over the related cuneiform bone. However, most commonly the clinical symptoms and the physical findings are nonspecific. Laboratory studies are usually normal. The radiographic changes usually are unremarkable at the beginning, although sites of decreased bone density of the affected cuneiform may occasionally be diagnosed. A decrease in size and marginal irregularity as well as sclerotic areas and structural collapse due to osteonecrosis may be evident on plain films, during the progress of the disease. Magnetic resonance imaging may be useful on the rare occasions that differential diagnosis from symptomatic bipartite medial cuneiform, osteomyelitis, or bone tumors is indicated. Osteochondrosis of the cuneiform bones is occasionally defined as a variant of normality, considering its benign self-limited nature. The disorder necessitates only symptomatic treatment. Medical therapy for pain control and physical activity restriction is the only required treatment modality. The clinical findings as well as the radiographic abnormalities resolve spontaneously, in all patients, irrespective of weight-bearing and immobilization treatment. Aggressive surgical treatment is unnecessary and potentially harmful to the patient. A short leg cast for 6-8 weeks accelerates the resolution of symptoms, although long-term outcomes are favorable regardless of treatment. A biopsy is not recommended for the diagnosis unless there is the need to rule out infection, especially the most common subacute form of osteomyelitis, Brodie's abscess, or malignancy.^[25-30,36-39]

The differential diagnosis may be confused in post-traumatic cases, following acute or stress fractures, or in patients with a bipartite cuneiform, which are all extremely rare in childhood.^[37,40,41]

Osteonecrosis of the cuneiform is an unusual pathology to the foot in adults. Risk factors in the case of non-traumatic osteonecrosis include the use of corticosteroids, smoking, alcohol, sickle cell anemia, radiation treatments, as well as rheumatologic, hematologic, and metabolic disorders.^[42-44]

The radiographs of children, younger than 14 years of age, with foot injuries or disorders that were referred to both the emergency and the outpatient department between January 2000 and December 2015, were retrospectively evaluated by the author from the hospital radiographic database. From a total amount of approximately 1500 children, no cases with a diagnosis of osteochondrosis of the cuneiform were identified. From the retrospective review of the radiographs, three cases with radiographic findings consistent with osteochondrosis of the cuneiform bones were identified.

The first patient was a 4-year-old boy who injured his right leg in a road traffic accident. Radiographic examination indicated

a spiral fracture of the right tibia with minimal displacement. Although there was no specific clinical evidence of a foot injury, dorsal-plantar and oblique foot radiographs were performed. A translucent line was evident on the distal pole of the medial cuneiform which had an irregular outline and cystic-like sites of decreased bone density. Sclerotic areas associated with sites of decreased bone density of the intermediate cuneiform were also diagnosed. There was also decreased bone density of the medial aspect of the lateral cuneiform. The radiographic findings were considered to be indicative of osteochondrosis of all cuneiform bones [Figure 1], despite no clinical findings detected from the physical examination or reported in the patient's history from his parents.

Although pedal osteochondrosis occurring at multiple sites has been previously reported,^[8,10,14,16,28,45-48] concomitant ipsilateral osteochondrosis of all the cuneiform bones has not been previously described.

The second patient was a 5-year-old boy who was referred after an in-home injury of his right foot. There was no evidence of a fracture line on the dorsal-plantar foot radiograph. The radiographic findings of marginal irregularity and the sclerotic areas associated with sites of decreased bone density of the medial cuneiform were considered to be indicative of osteochondrosis of the medial cuneiform [Figure 2].

Finally, the third patient was a 6-year-old boy who was referred after a school injury of his left foot. There was no evidence of a



Figure 1: A 4-year-old boy with radiographic findings, consistent with osteochondrosis, involving the full extent of the medial and intermediate cuneiform bones and the medial aspect of the lateral cuneiform bone



Figure 2: A 5-year-old boy with radiographic findings, consistent with osteochondrosis, involving the medial cuneiform bone

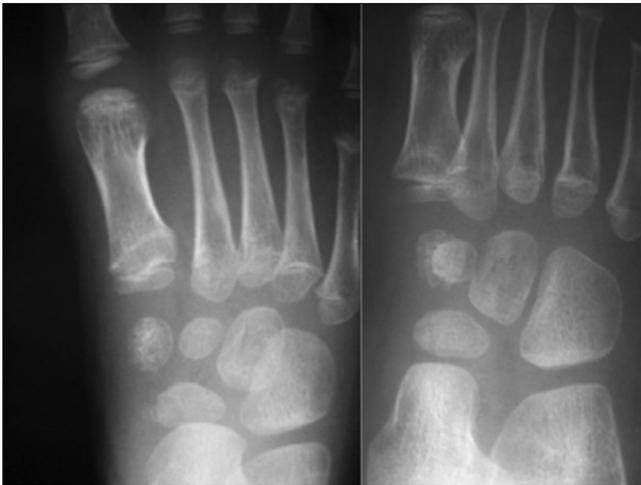


Figure 3: A 6-year-old boy with radiographic findings, consistent with osteochondrosis, involving the medial cuneiform bone

fracture line on the dorsal-plantar and oblique foot radiographs. The radiographic findings of marginal irregularity and the sclerotic areas associated with sites of decreased bone density of the medial cuneiform were considered to be indicative of osteochondrosis of the medial cuneiform [Figure 3].

It is apparent that in all three patients, the diagnosis of osteochondrosis of the cuneiform bones was retrospectively

made from the evaluation of the radiographic examination following an ipsilateral leg or foot injury. It may be reasonable to consider that the patients were asymptomatic or minimally symptomatic before the traumatic insults. It may be prudent to realize that the incidence of osteochondrosis of the cuneiform may be higher than the reported one in the world literature since a great percentage of patients remain undiagnosed due to the mild symptomatic course of the disorder as well as to the age distribution, provided that the disorder usually affects pre-school and early school-age children.

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