Case Report

MRI findings of intermediate cuneiform osteochondrosis as a rare cause of foot pain in a child

Ivan R.B. Godoy, M.D., André Fukunishi Yamada, M.D., PhD, Abdalla Skaf, M.D.

Department of Radiology, Hospital do Coração (HCor) and Teleimagem, Rua Desembargador Eliseu Guilherme, 53, 7th floor. CEP 04004-030, São Paulo, SP, Brazil

Department of Diagnostic Imaging, Federal University of São Paulo (UNIFESP), São Paulo, SP, Brazil

ALTA Diagnostic Center (DASA Group), São Paulo, Brazil

Abstract

Few cases of intermediate cuneiform osteochondrosis have been described in the literature. In this report we present a case of a 9-year-old boy with a 2-month history of right foot pain and edema, especially near the third metatarsal, without previous trauma. Also, there were no signs of inflammation, erythema, or fever. Magnetic resonance imaging (MRI) showed bone edema, mild sclerosis, and volumetric reduction of the intermediate cuneiform. Non-steroidal anti-inflammatory medication was prescribed as treatment. Symptoms remitted spontaneously after 2 weeks. The pain did not return, and the patient was asymptomatic after 3 months. Imaging studies such as X-ray, CT, and MRI are important to identify this condition and may avoid unnecessary tests and treatments such as laboratory exams, scintigraphy, and bone biopsies. Due to the benign nature as well as the possibility of it presenting with no symptoms, we believe that it may be considered as a variant of bone maturation or a self-limited condition.

© 2020 The Authors. Published by Elsevier Inc. on behalf of University of Washington. This is an open access article under the CC BY-NC-ND license. (http://creativecommons.org/licenses/by-nc-nd/4.0/)

Introduction

Avascular bone necrosis is a condition that affects usually the endochondral ossification centers in children, therefore osteochondrosis is also a term used for this condition.

The most common types of foot osteochondrosis in children are those affecting the navicular bone (Kohler's bone disease) and the second metatarsal head (Freiberg's infraction or Freiberg's disease), which are identified usually radiologically and frequently are self-limiting. Various etiologies have been proposed that cause bone osteochondrosis including bone

Declaration of competing interests: The authors declare that they have no competing interests.

Funding: No specific funding was received from any funding bodies in the public, commercial, or not-for-profit to carry out the work described in this article.

Corresponding author.

E-mail address: ivanrbgodoy@gmail.com (I.R.B. Godoy).

https://doi.org/10.1016/j.radcr.2020.03.012

1930-0433/© 2020 The Authors. Published by Elsevier Inc. on behalf of University of Washington. This is an open access article under the CC BY-NC-ND license. (http://creativecommons.org/licenses/by-nc-nd/4.0/)
growth, repetitive microtrauma, vascular abnormalities, hormonal factors, and even dietary factors [1–3] although their etiology is unknown.

Seventeen cases of idiopathic primary osteochondrosis involving the cuneiforms have been described: One lateral cuneiform [4], 19 medial cuneiform [4–14], and only 4 in the intermediate cuneiform [16–18]. Of these, 12 had symptoms while the other 4 were incidental findings [4–12]. The radiological characteristics are similar in all described cases, including bone sclerosis and size reduction, but the duration of the symptoms may vary.

This study is the first to describe the magnetic resonance imaging (MRI) findings of a new case of primary idiopathic osteochondrosis of the intermediate cuneiform bone in a child with foot pain.

---

**Case report**

A 9-year-old boy presented with a 2 months history of pain in the right foot. The pain was associated with edema and reduced range of motion. Physical examination showed pain on palpation of the base of the third metatarsal and edema. The patient’s medical history was collected with his parents and did not include neoplastic disease or prior surgery of the lower extremities.

An MRI of the right foot was performed 1 week after presentation. At that time, the patient was presumed to have a tendinous injury or bone contusion. The MRI showed bone edema of the intermediate cuneiform with mild sclerosis and smaller size in comparison with the medial and lateral...
Fig. 2 – Axial CT imaging of the feet (a) showing volumetric reduction of the right intermediate cuneiform (arrow) and of the left navicular bone (arrowhead).

cuneiforms (Fig. 1). The radiology staff suggested that the ordering physician request a comparative radiograph or a CT (computer tomography) of the feet. A CT was performed (Fig. 2) and showed normal left cuneiform bones. Surprisingly, there was asymptomatic left navicular osteonecrosis. The patient was treated with oral ibuprofen for 5 days. After 3 months of follow-up the patient was asymptomatic. No other imaging studies were ordered after symptom relief.

Discussion

Cuneiform bone osteochondrosis is an unusual cause of foot pain in children. The most common types of foot osteochondrosis are calcaneal apophysitis (Sever’s disease), navicular bone (Kohler’s bone disease), and second metatarsal head (Freiberg’s disease). Our case report describes the MRI findings of a case of avascular necrosis of the intermediate cuneiform in a boy with foot pain without trauma.

Buchman et al [4] was the first author to describe 2 cases of osteochondrosis of the bilateral medial cuneiform bone, which also affected the navicular bone, followed by 2 descriptions by Haboush [8] and Meilstrup [9]. The first description of intermediate cuneiform osteochondrosis was done by Hicks et al in 1953, reporting 4 cases [15–18]. None of the previous described cases had MRI findings. The etiology of osteochondrosis of bone in children is unknown. Currently it is believed to be a systemic and multifactorial condition probably due to hereditary factors, coagulopathy, hyperactivity, and passive smoking condition [19–20]. Children with this condition usually have delayed bone age maturation compared with chronological age [21]. In the later stages of bone development, bone age speeds up to reach the corresponding chronological age.

Episodes of microtrauma or mechanical stress on the center of ossification have also been mentioned as one of the most common causes [12,17,18,22], although Stanley and Betts [23] stated that only 5% of the affected bones had documented trauma when studying Freiberg’s disease. Mubarak [4] speculated with regard to the possibility that avascular necrosis may occur in the foot bones in which the start of ossification occurred after the walking age, such as the navicular and cuneiform bones. Therefore, ossification before this age could be a preventive factor. Most cases of cuneiform bones osteochondrosis were reported in boys, such as our case [7,15,18]. Freiberg’s disease reported to be more common in girls, while Kohler’s bone disease is more common in boys.

Osteochondrosis of cuneiform bones is a transient, self-limiting condition that usually does not require treatment. Since it has a benign evolution and short duration of symptoms it is possible underdiagnosed. One of the 2 cases described by Buchman [5] and 3 of the 4 cases described by Vaquer Martin et al [12] were diagnosed by coincidence in radiographs requested for other reasons and were asymptomatic. It is even possible that it does not become evident because of the fact that its symptomatic evolution could be short, just as occurred in the case here presented. It may therefore be treated as an anatomical, as seen in Kohler’s in the navicular bone and Sever’s in the calcaneus bone, which may be present in radiographic signs ordered for other reasons than osteochondrosis. Curiously, it is important to note that in our case, the ‘normal’ asymptomatic left foot shows Kohler’s disorder in the navicular bone (Fig. 2).

There are other conditions that may cause foot pain in children that must be excluded. Leukemia patients may present with diffuse bone pain due to distension of the medullary cavities by proliferation of the hematopoietic tissue. Usually it is present in long bones and the spine. Another entity to differentiate is osteomyelitis that clinically presents with fever, local edema, swelling, and pain. It usually shows limited soft tissue changes in the radiograph in the first 5–7 days. From 7 to 10 days, radiographs depict irregular spotty areas of rarefaction due to the trabecular absorption for local hyperemia. Longer evolution shows areas of radiolucency and sequestered dead bone.

Because of its benign nature, there is no indication for follow-up imaging, bone biopsies, or bone scans such as scintigraphy [4,17]. Clinically, the symptoms include pain, edema, and some local tenderness, which resolve in 2–8 months [4].

Treatment is generally conservative [4,12,17,18] with regression of the pain and restitution of bone shape and size. Rest or reduction in physical exercise as well as symptomatic treatment with non-steroidal anti-inflammatory and/or
analgesic is appropriate. The most important aspect of this publication is to present the MRI findings of a case of osteochondrosis of the intermediate cuneiform bone as a very rare cause of foot pain. A precise diagnosis of this condition is paramount to orientate the correct management of the case, thereby avoiding unnecessary diagnostic tests and resulting in potentially damaging treatment such as casted immobilization, biopsies, or a surgical approach. Due to its benign nature as well as the possibility of it presenting with no symptoms, we believe that it may be considered as an anatomical variant.

**Authors’ contributions**

IG provided the clinical data included in the text. IG wrote the manuscript draft. IG and AFY revised it critically and approved the modified text. IG, AFY, and AS approved the final version of the manuscript. All authors read and approved the final manuscript.

**Availability of data and materials**

The datasets used and/or analyzed during the current study are available from the corresponding author upon reasonable request.

**Consent for publication**

Written informed consent was obtained from the subject for publication.

**REFERENCES**

[1] Siffert RS. Classification of the osteochondroses. Clin Orthop Relat Res 1981;158:10–18.
[2] Douglas G, Rang M. The role of trauma in the pathogenesis of the osteochondroses. Clin Orthop Relat Res 1981;158:28–32.
[3] Herring JA. The limping child. In: Herring JA, editor. Tachdjian's pediatric orthopaedics. Philadelphia, PA: W B Saunders; 2002. p. 83–94.
[4] Mubarak SJ. Osteochondrosis of the lateral cuneiform: another cause of a limp in a child: a case report. J Bone Joint Surg 1992;74-A:285–9.
[5] Buchman J. Osteochondritis of the internal cuneiform. J Bone Joint Surg 1933;15-A:225–32.
[6] Chew JT, Low CK, Mitra AK. A case report on bilateral avascular necrosis of the medial cuneiform. Ann Acad Med Singapore 1995;24:453–4.
[7] Gago Baron M, García Gil H, Chaves Gomez-Orihuela JM, Lebrero Lebrero L. Osteochondritis of the first cuneiform: a case report of a bilateral case. Rev Ortop Traumatol 1988;32:139–40.
[8] Haboush EJ. Bilateral disease of the internal cuneiform bone with an associated disease of the right scaphoid bone (Ko’lier’s). JAMA 1933;100:41–2.
[9] Meilstrup DB. Osteochondritis of the internal cuneiform, bilateral. Case report. Am J Roentgenol 1947;58:329–30.
[10] O’Donoghue AF, O’Donoghue ES, Zimmerman M. Bilateral osteochondritis of the tarsal navicular and the first cuneiform. J Bone Joint Surg 1948;30-A:780–1.
[11] Scarfi G, Landi F. A rare case of bilateral osteochondrosis of the first cuneiform bone. Arch Putti Chir Organi Mov 1983;33:447–9.
[12] Vaquero Martin J, Vicente-Herrera E, Pereiro de Lamo J, Vidal Fernandez C. Osteochondritis of the medial cuneiform. J Pediatr Orthop B 1999;8:69–71.
[13] Zimberg J, Levitt JC, Brahim F. Osteochondrosis of the medial cuneiform: a case report. J Am Podiatr Med Assoc 1985;75:538–9.
[14] Atbasi Z, Ege T, Kose O, Egerci OF, Demiralp B. Osteochondrosis of the medial cuneiform bone in a child: a case report and review of 18 published cases. Foot Ankle Specialist 2013;6(2):154–8.
[15] Hicks BTG. Case report. Osteochondritis of the tarsal second cuneiform bone. Br J Radiol 1953;26:214–15.
[16] Kose O, Demiralp B, Oto M, Sehirioglu A. An unusual cause of foot pain in a child: osteochondrosis of the intermediate cuneiform. J Foot Ankle Surg 2009;48:474–6.
[17] Leeson MC, Weiner DS. Osteochondrosis of the tarsal cuneiforms. Clin Orthop 1985;196:260–4.
[18] Watmough PJ, Tselentakis G, Day JB. Avascular necrosis of the intermediate cuneiform bone. J Pediatr Orthop 2003;13:402–5.
[19] Herring JH. Tachdjian’s pediatric orthopaedics 2008:771–837.
[20] García Mata S, Ardanaz E, Hidalgo A, Martinez Grande M. Legg-Calve’–Perthes disease and passive smoking. J Pediatr Orthop 2000;20:326–30.
[21] Kristmundsdottir F, Burwell RG, Hall DJ, Marshall WA. A longitudinal study of carpal bone development in Perthes’ disease: its significance for both radiologic stand still and bilateral disease. Clin Orthop 1986;209:115–23.
[22] Walsh HP, Dorgan JC. Aetiology of Frieberg’s disease: trauma? J Foot Surg 1988;27:243–4.
[23] Stanley D, Betts RP. Assessment of aetiological factors in the development of Frieberg’s disease. J Foot Surg 1990;29:444–7.