

The Bipartite Medial Cuneiform—A Rare Cause of Midfoot Pain: Case Report and Review of Literature

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Introduction

The bipartite medial cuneiform (BMC) is a rare congenital variant of the tarsus. It is described as the separation of the normal medial cuneiform into 2 ossicles by an oblique or a horizontal line with the dorsal ossicle usually being smaller.^{3,4} These 2 parts are forming a pseudoarticulation which is held together by a cartilaginous synchondrosis, a fibrous syndesmosis, or a combination of both.^{6,7} The incidence of the BMC is estimated to be between 0.1% and 2.4% of the population.^{4,11} Normally this variant is asymptomatic but may become clinically relevant when acute trauma or chronic overuse of the foot damages the tissue bridge between the ossicles. Referring to the literature, successful treatment of a symptomatic BMC has been achieved with surgical treatment or injections.

Case Report

A 33-year-old male patient presented with midfoot pain after a twisting trauma of the left midfoot during a kitesurfing accident. Although feeling immediate pain, the patient was still able to finish his kitesurfing session and bear full weight afterwards. However, in the following days, he suffered from increasing medial foot pain during exercise or when walking on uneven ground. The most significant pain trigger was described when the rear foot was brought into pronation during sliding movements in clay court tennis suggesting an activation of the tibialis anterior tendon. Increasing pain after sports as well as pain even during

activities of daily living led to the presentation in a foot surgery department 2 weeks after the accident.

Physical examination revealed no swelling or bruising of the foot. Palpation showed a tenderness over the medial midfoot, and functional testing caused pain in the same region during pronation of the foot against resistance. Weightbearing radiographs (Figure 1) did not reveal an acute fracture, but an inhomogeneous bony structure replacing the normal medial cuneiform. Imaging of the uninjured (and asymptomatic) opposite side showed that it was a bilateral entity. Computed tomographic scan was performed showing a BMC with a thin nonossified pseudoarticulation with no fracture or diastasis of the 2 ossicles, but with increased sclerosis and subchondral cysts in the area of the pseudoarticulation (Figure 2).

Subsequent magnetic resonance imaging (MRI) confirmed the BMC showing clear evidence of bony edema around the synchondrosis that, together with the subchondral cysts and increased sclerosis, suggested an abnormal

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Figure 1. Weightbearing radiographs of the left foot. Bipartition of the bipartite medial cuneiform might be appreciated on (A) the dorsoplantar projection, whereas (B) the lateral radiograph reveals dorsal first metatarsal–medial cuneiform joint exostosis and facet changes in the articulation with the navicular bone.

motion or loading in this formation leading to the above-mentioned stress changes (Figure 3).

With the exception of nonweightbearing and nonsteroidal anti-inflammatory drugs for 2 weeks, the patient refused any kind of further treatment. At the 4-month follow-up, the patient reported no discomfort in activities of daily living, but still had midfoot pain after longer walks, especially on uneven ground. The follow-up MRI showed no significant changes compared with the first imaging. The patient still opted for conservative treatment and was able to return to preinjury-level sports 9 months after the initial injury.

At the 5-year follow-up, the patient reported that he never had any symptoms at the left midfoot again, performing all kind of sports like tennis, running, kitesurfing, skiing, and rock-climbing. A repeat MRI scan showed no significant change in imaging findings (Figure 4). Bone marrow edema of the adjacent first metatarsal had resolved, but still was present in the dorsal ossicle of the BMC.

Discussion

Bipartition of the medial cuneiform is a rare congenital osseous anomaly of the foot.^{3,4} Normally this variant is asymptomatic but can become clinically relevant when a trauma or overuse occurs to the foot, damaging the tissue bridge between the ossicles. In these cases, a fracture has to be differentiated from a BMC. In plain radiographs, the bipartition may be overlooked easily because the 2 segments overlap. Therefore a 30-degree oblique view may be helpful to visualize the bipartition. However, more accurate diagnostic tools like computed tomographic or MRI scan

may be required to confirm the diagnosis and to distinguish between a BMC and a fracture of the medial cuneiform.^{6,8} Such fractures usually show irregular bony contours and a vertical orientation whereas the ossicles of a BMC are well corticated and in a horizontal plane, with the combined volume of the ossicles appearing larger than expected.^{6,8} Bone marrow edema definitely will be seen in a recent fracture, but also can be present in a BMC as reported in our case.

Reviewing symptomatic cases of BMC, most authors describe a surgical intervention to their patients consisting of either excision of the smaller fragment (2 patients), osteosynthesis of the 2 segments (4 patients), or adjunctive fusion to adjacent joints (3 patients).^{1,4,5,10,12} With the exception of 1 patient who still suffered from pain while running, all patients reported complete pain relief. Therefore, authors concluded that surgical treatment is effective when conservative treatment fails. Additionally, presence of bone marrow edema is often interpreted as a sign of pain-inducing interosseous instability indicating surgical intervention.^{4,6,9,11}

However, there are just 3 publications describing the conservative treatment of a symptomatic BMC in a case report. These 3 patients reported full pain relief after steroid injections.^{2,9,12} Authors concluded that local injections should be first-line treatment of a symptomatic BMC. However, all studies lack information on how long patients were treated conservatively before surgical intervention was performed.^{1,4,5,10,12}

There is just 1 publication reporting completely noninvasive treatment (no surgery, no injection) of a symptomatic BMC. Eves et al⁷ describe the case of a skeletally immature 11-year-old patient who was successfully treated with a nonweightbearing cast and pneumatic walker immobilization after a soccer injury to his midfoot.

To our knowledge, this is the first case report recounting the "natural fate" of a posttraumatic symptomatic BMC in an adult, which—despite lasting 9 months and still presenting bone marrow edema on follow-up MRIs—showed complete pain relief without invasive treatment.

Conclusion

The BMC is a rare anomaly of the midfoot. Awareness of its existence is essential to identify this entity and to differentiate it from a fracture or pseudarthrosis in patients with acute or chronic midfoot pain. Our case impressively demonstrates that healing of a symptomatic posttraumatic BMC can be protracted and still be achieved 9 months after the injury. Additionally, this case demonstrates that the presence of bone marrow edema is not synonymous with the presence of pain. Surgeons should take this into account before deciding to perform surgical intervention on a BMC.

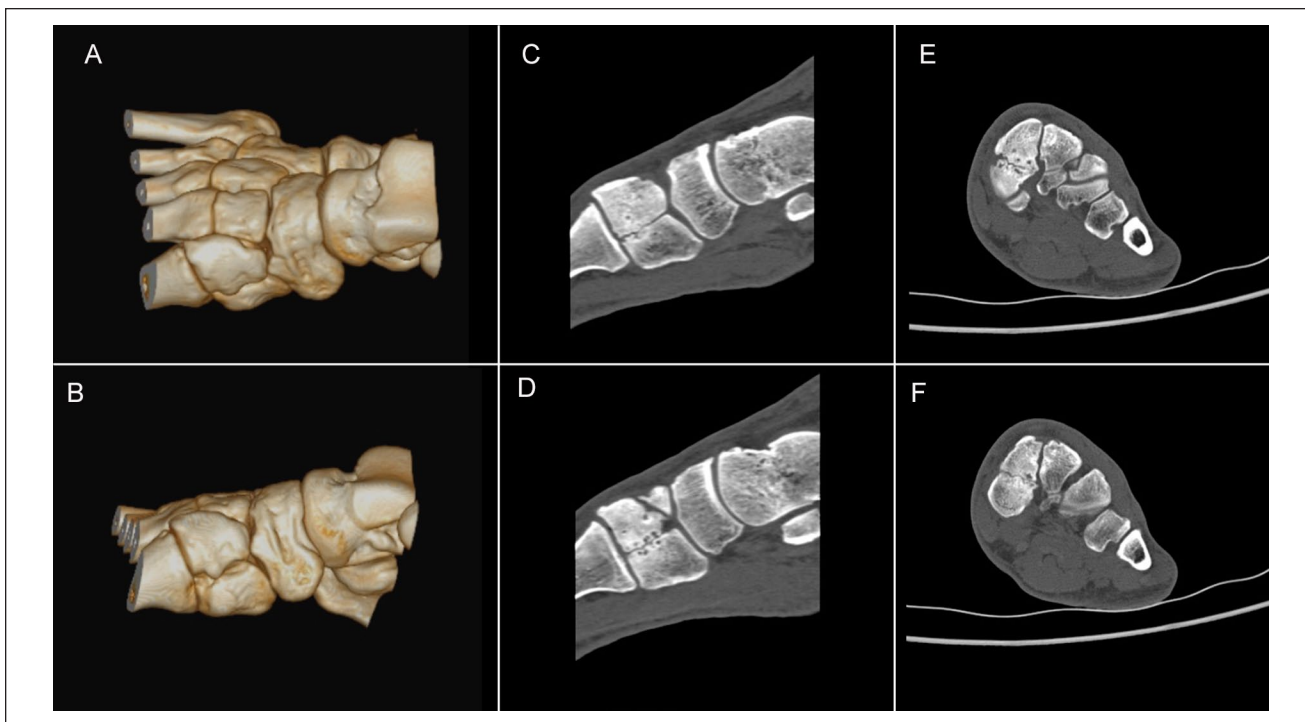


Figure 2. Computed tomography scan showing (A, B) bipartition of the medial cuneiform on 3D reconstruction. (C, D) Increased sclerosis and subchondral cysts in the area of the pseudoarticulation can be seen on sagittal slices. (E, F) Coronal scans reveal complete bipartition of the medial cuneiform.

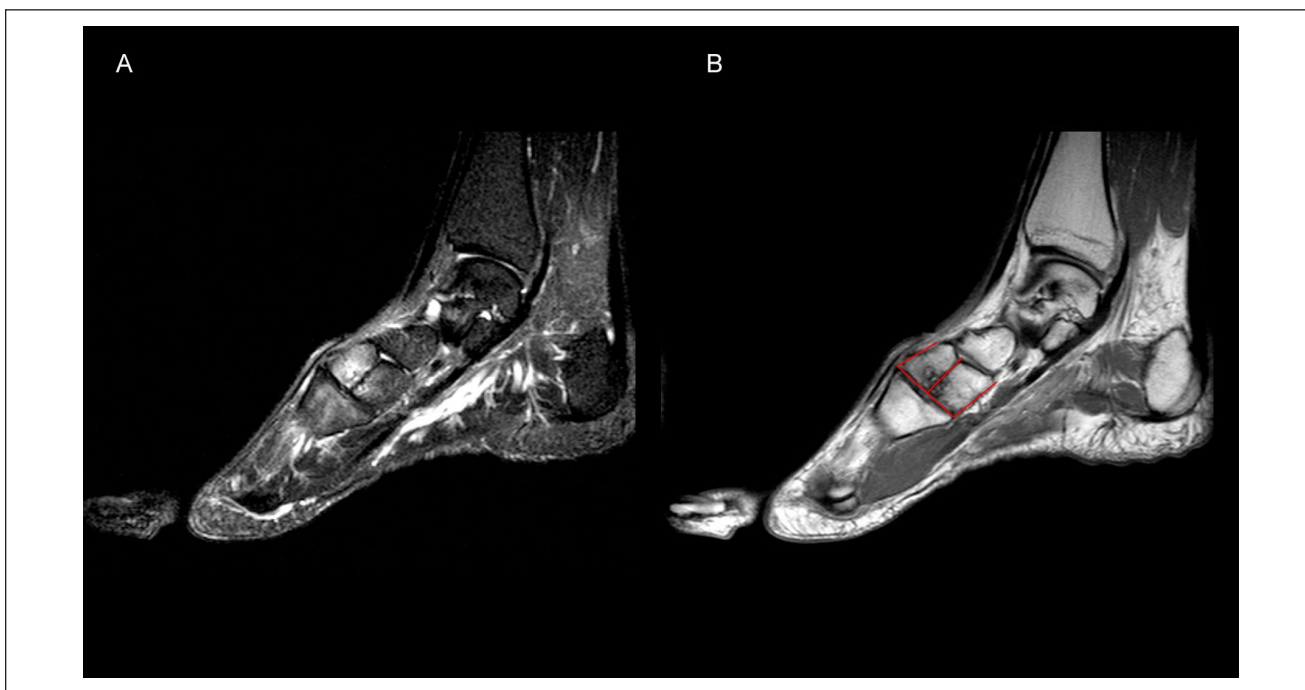


Figure 3. Magnetic resonance imaging showing the presence of bone marrow edema but no diastasis at the synchondrosis of the 2 ossicles in (A) T₂- and (B) T₁-weighted sagittal scans. Joint spaces between the head of the first metatarsal and the distal aspect of the 2 medial bipartite cuneiform bones as well as the horizontal line between the 2 ossicles of the medial cuneiform create a characteristic formation that has previously been described as the “E-sign” (red lines).⁶

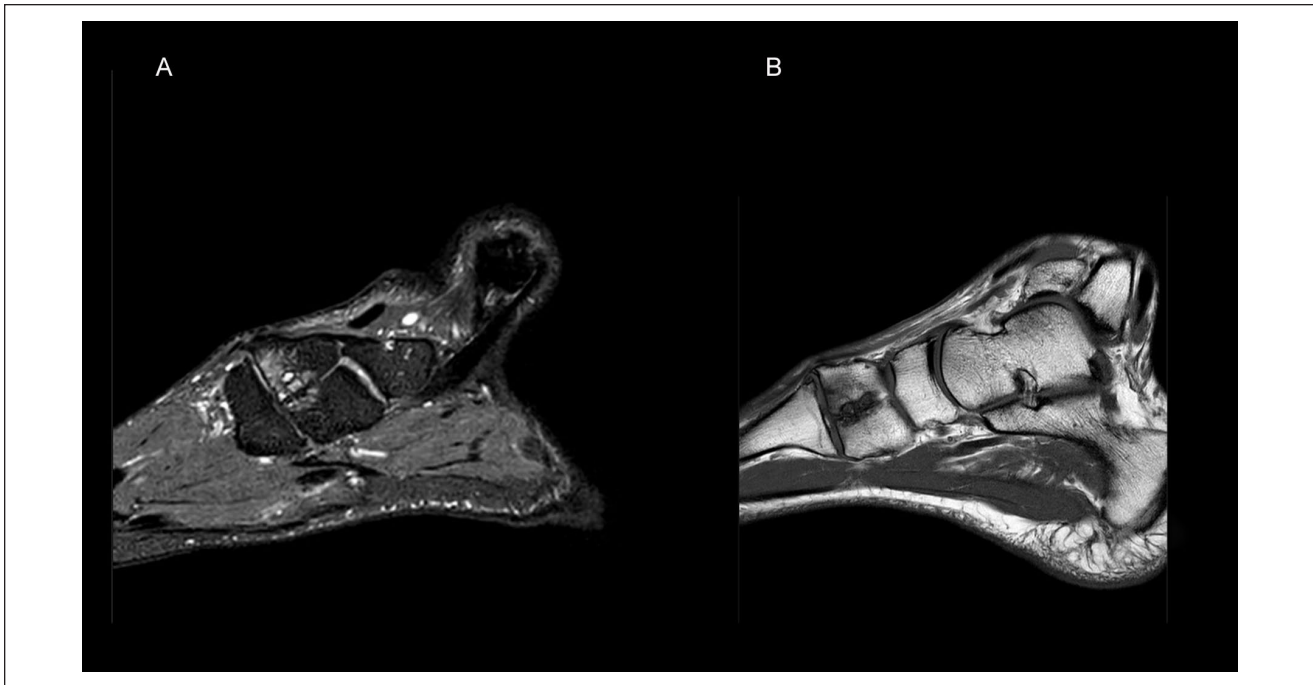


Figure 4. Magnetic resonance imaging 5 years after the incident showing a decreasing but still observable bone marrow edema of the 2 ossicles in T₂- (A) and T₁-weighted (B) sagittal scans, whereas the bone marrow edema in the first metatarsal completely resolved.

Ethical Approval

Ethical approval was not sought for the present study because proposed case study does not constitute a systematic investigation, and is intended as an interesting example for educational purposes. All therapies performed involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki Declaration and its later amendments or comparable ethical standards. The authors confirm that informed consent of the patient is taken for publication of this case report.

Declaration of Conflicting Interests

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References

1. Azurza K, Sakellariou A. 'Ostoesynthesis' of a symptomatic bipartite medial cuneiform. *Foot Ankle Int.* 2001;22(6):499-501.
2. Bismil Q, Foster PAL, Venkateswaran B, Shanker J. Symptomatic bipartite medial cuneiform after injury: a case report. *Foot Ankle Surg.* 2005;11:55-58.
3. Burnett SE, Case DT. Bipartite medial cuneiform: new frequencies from skeletal collections and a meta-analysis of previous cases. *Homo.* 2011;62:109-125.
4. Chang GH, Chang EY, Chung CB, Resnick DL. Bipartite medial cuneiform: case report and retrospective review of 1000 magnetic resonance (MR) imaging studies. *Case Rep Med.* 2014;2014:130979.
5. Chiodo CP, Parentis MA, Myerson MS. Symptomatic bipartite medial cuneiform in an adult athlete: a case report. *Foot Ankle Int.* 2002;23(4):348-351.
6. Elias I, Dheer S, Zoga AC, Raikin SM, Morrison WB. Magnetic resonance imaging findings in bipartite medial cuneiform - a potential pitfall in diagnosis of midfoot injuries: a case series. *J Med Case Rep.* 2008;2:272.
7. Eves TB, Ahmad MA, Oddy MJ. Sports injury to a bipartite medial cuneiform in a child. *J Foot Ankle Surg.* 2014;53:232-234.
8. O'Neal ML, Ganey TM, Ogden JA. Fracture of a bipartite medial cuneiform synchondrosis. *Foot Ankle Int.* 1995;16(1):37-40.
9. Panu A, Konin G, Saboieiro G, Schneider R. Symptomatic bipartite medial cuneiform treated with fluoroscopic and ultrasound-guided injections. *HSS J.* 2014;10:92-97.
10. Pollack D, Diament M, Kotlyarova Y, Gellman Y. The bipartite medial cuneiform. *J Am Podiatr Med Assoc.* 2021;111(6).
11. Serfaty A, Pessoa A, Antunes E, Malheiro E, Canella C, Marchiori E. Bipartite medial cuneiform: magnetic resonance imaging findings and prevalence of this rare anatomical variant. *Skeletal Radiol.* 2020;49:691-698.
12. Steen EF, Brancheau SP, Nguyen T, Jones MD, Schade VL. Symptomatic bipartite medial cuneiform: report of five cases and review of the literature. *Foot Ankle Spec.* 2016;9(1):69-78.