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Osteonecrosis of the lateral cuneiform: A singular case in the literature

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Abstract
Introduction: Osteonecrosis is a condition that rarely affects the bones of the foot and, when present, is more common in the talus and navicular. Osteonecrosis of the cuneiforms is extremely rare, with only a few case reports described in the literature, most of which were in pediatric patients. There are no case reports of this disease in the lateral cuneiform, let alone in the adult population.
Objective: Presenting this unique case of an adult patient with osteonecrosis of the lateral cuneiform who underwent surgical treatment.
Methods: A 42-year-old female patient with progressive pain in the left midfoot that rendered full weight bearing gait impossible. There are no known comorbidities and no history of trauma or intense sports activity in the previous months. Apart from tenderness and swelling, there are no exuberant clinical signs.
Magnetic resonance imaging showed dorsal hypodensity of the lateral cuneiform, suggestive of avascular necrosis, with no signs of bone collapse.
Due to failure of conservative treatment, the patient underwent surgical treatment with subchondral drilling.
Results: No postoperative complications were recorded. After 3 months of surgery, the patient was walking pain-free with full weight-bearing and showed signs of revascularization in magnetic resonance imaging.
Currently with excellent clinical progress, with significant improvement in both the AOFAS score (from 35/100 to 90/100) and the visual analogue pain scale (from 8/10 to 1/10). The patient classified the result as excellent and would recommend it to anyone.
Conclusion: Osteonecrosis of the lateral cuneiform is an exceptionally rare condition, this report being the first of its kind. Like in other anatomical areas, it can be successfully treated with surgical decompression with subchondral drilling in the early stages.

Keywords: Osteonecrosis, avascular necrosis, lateral cuneiform, subchondral drilling

Introduction
Osteonecrosis (also known as avascular necrosis) refers to bone cellular death caused by disruption of blood supply, depriving the bone tissue of essential nutrients and oxygen. If not diagnosed and correctly treated, it can evolve to collapse, fragmentation and joint degeneration [1]. In the foot, while uncommon, it typically affects the talus and navicular [1, 2]. Affection of the cuneiforms is extremely rare, with very few cases described in the literature, almost all in children. As far as we are aware, up to this date, this is the only case reported of osteonecrosis of the lateral cuneiform, l*et al*one in the adult population.
The vascular compromise occurring in this disease is not yet entirely known. Nevertheless, it is thought to be caused by a mechanical disruption of the vessels or from occlusion of either arterial inflow or venous outflow. Embolism, thrombosis and abnormally shaped cells (sickle cells, for example) are among the causes of this event. Other factors, like alcohol intake, Gaucher's disease and use of corticosteroids, contribute to the physiopathology by increasing intramedullary pressure, which decreases perfusion [1]. Trauma remains the most common mechanical cause of osteonecrosis, particularly in the foot [3]. This pathology usually affects athletes and/or military personnel subject to repetitive microtrauma and stress fractures [4].
Regardless of the cause of osteonecrosis, the final radiographic finding remains the same: a relative increase in the radiodensity of the bone. This change is caused by bony collapse, impaction and a relative density differential between the avascular and surrounding vascularized bone.

Without the ability to repair itself, such avascular bone eventually collapses, appearing sclerotic and fragmented [3].
Conventional radiographs are only useful for diagnosis after the development of sclerosis, fragmentation or articular collapse. MRI (Magnetic Resonance Imaging) can detect early signs of osteonecrosis before they are seen on x-ray or even before the disease is clinically perceived. Diffuse marrow edema is seen in early osteonecrosis and produces a low signal intensity on T1-weighted images and a high signal intensity on T2-weighted images. In advanced stages of the disease, both T1 and T2-weighted images demonstrate low signal intensity [1]. CT (Computed Tomography) scan is mostly used to determine the extent of bone/articular damage.
There may be no symptoms of osteonecrosis in the early stages of the disease, but, as it progresses, insidious midfoot pain that is activity-related and occurs predominantly at night is expected [3]. Eventually the joint will be painful even when resting, it will stiffen and lose its range of motion, ending in osteoarthritis. The time period between the start of symptoms and loss of function of the joint varies from person to person and ranges from a few weeks to more than a year.
In the initial stages, conservative treatment (immobilization, modified activity, physical therapy and anti-inflammatory medication) can be effective. However, as this pathology tends to be diagnosed later, surgery is often the only course of action in the majority of patients. Core decompression drilling, augmentation with bone grafts, osteotomies and arthrodesis are all possible surgical treatment options.
The aim of the present article is to report this unique case of osteonecrosis of the lateral cuneiform in an adult patient and its surgical treatment.

Materials and Methods

An otherwise healthy 42-year-old female patient presented in our outpatient clinic with progressive midfoot pain which rendered full weight bearing gait impossible. The pain had started 2 years before, with significant worsening in the last 6 months. There was no history of trauma or intense sports activity in the previous months. Additionally, there was tenderness and oedema in the dorsal region of the midfoot foot, a mild cavus deformity and shortening of the Achilles tendon.
On the initial radiographic examination, very discrete changes were noted in the dorsal cortex of the cuneiform bone. The magnetic resonance imaging (MRI) showed diffuse areas of hypersignal (bone edema) mingled with other hyposignal areas (bone necrosis) in T1-weighted sequences.
The patient was initially submitted to conservative treatment with only partial weight-bearing allowed using an orthosis, physiotherapy rehabilitation and modifications of her daily activity. Nevertheless, after 6 months of failed conservative treatment, surgical treatment was chosen.
Using a dorsal approach to access the lateral cuneiform bone, we performed subchondral drilling of the bone. Our initial plan was to also include debridement and curettage of the necrotic tissue and autologous spongy bone graft from the distal tibia. However, owing to the advanced sclerosis, the bone was too hard and it would not be possible to proceed as planned. The necrotic and devitalized bone tissue samples removed from the cuneiform bone were sent for anatomopathological examination, which later confirmed the diagnosis of osteonecrosis.

Results

An overall positive response to treatment was obtained. No peri-operative complications were reported. The patient was pain-free and started physical rehabilitation immediately following the surgery and was allowed full weight-bearing after 8 weeks. After 3 months of surgery, the patient was asymptomatic and showed signs of revascularization in magnetic resonance imaging.
Currently with excellent clinical progress, with significant improvement in the scores of both AOFAS (from 35/100 to 90/100) and the visual analogue pain scale (VAS) (from 8/10 to 1/10). The patient classified the result as excellent and would recommend it to anyone.

Discussion

To date, there are very few published cases of osteonecrosis of the cuneiforms, most of them occurring in the pediatric population [5]. This is, to our knowledge, the first reported case of this disease affecting the lateral cuneiform in the literature, either in adults or in children.
Although a relatively uncommon diagnosis, osteonecrosis has been identified in almost every bone in the foot and has been extensively studied in other tarsal bones, such as the talus, the metatarsals and the navicular [1, 4]. Due to the small number of reported cases of this pathology in the cuneiform bones, there is no consensus on the diagnosis and treatment of this entity. In this case, we applied the same principles in the diagnosis and treatment of this disease in other more deeply studied areas of the body.
A progressive dorsal midfoot pain worsening in the last 6 months and not associated to trauma or overuse in an otherwise healthy middle-aged female patient. In the physical examination, tenderness and oedema in the dorsal midfoot, a slight cavus deformity, Achilles tendon shortening and inability to tolerate full weight bearing were noted. In the differential diagnosis, articular (joint degeneration or instability) or soft tissue pathologies (ligamentous or tendinous injury) were the most likely scenarios. Owing to the anatomical location of the pain, avascular necrosis was not our first suspected diagnosis. Due to the lack of exuberant clinical signs and ordinary foot radiographs, an MRI scan was asked which was determinant in the making of the final diagnosis. Early stages of osteonecrosis are hardly recognized on plain radiographs, requiring an MRI for the definitive diagnosis (as was the case with this patient).
Our first choice was the conservative treatment, which included rest, weight-bearing restrictions with orthosis, physical therapy and anti-inflammatory medication. In other anatomical areas, it has been proven to be effective in not only delaying but in many cases reversing the progression of the diseases. Nevertheless, the patient did not improve her condition with this course of action. So, after a period of failed conservative treatment of 6 months, we considered our surgical options.
As the articular surfaces were intact, we opted for a joint-saving procedure: debridement and curettage of the necrotic tissue and subchondral drilling of the bone, as did Chew *et al*. in the medial cuneiform [6]. Initially, we intend to add autologous spongy bone graft from the distal tibia to the cuneiform but we found that the bone was too hard due to

the sclerosis that it was impossible to perform. There are no comparative studies that contemplate a superior technique for the treatment of osteonecrosis in the early stages in the cuneiforms due to the singularity of this pathology [7]. According to the literature, in advanced cases with joint degeneration, arthrodesis is the ideal procedure that allows the maintenance of bone length and architecture [5]. However, as mentioned before, there was no joint degeneration, so we chose a joint-preserving technique.
The end result was a satisfied patient, with no clinical signs of the disease. MRI studies performed 3 months and 9 months after the surgery showed a significant recovery of the necrosis.
More studies regarding this subject ought to be made for a better understanding of the disease and its treatment options. Owing to the rarity and scarcity of papers regarding this pathology in the cuneiform bone, the treatment remains controversial and challenging [1, 2].

Fig 1: Preoperative AP foot radiograph

Fig 2: Preoperative oblique foot radiograph

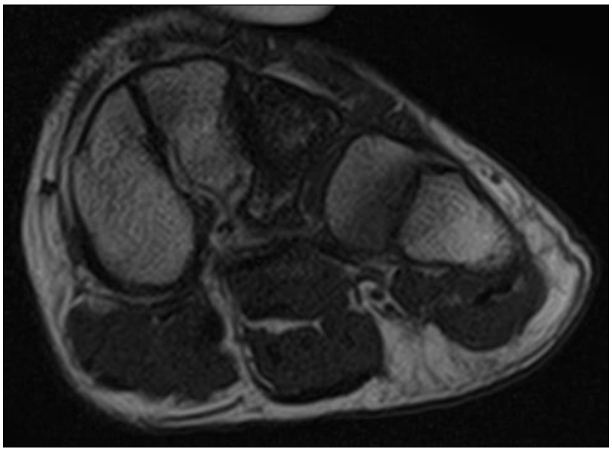

Fig 3: Preoperative MRI scan T2 sequence coronal view

Fig 4: Preoperative MRI scan T2 sequence axial view

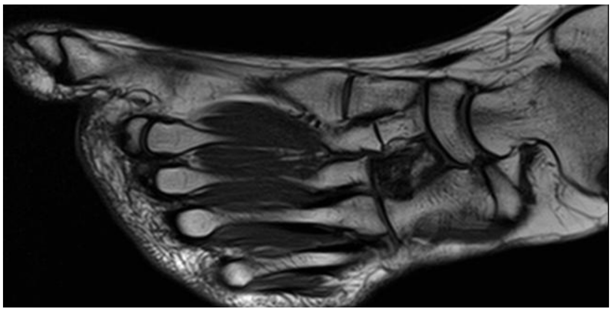

Fig 5: Comparative preoperative MRI scan T2 sequence axial view

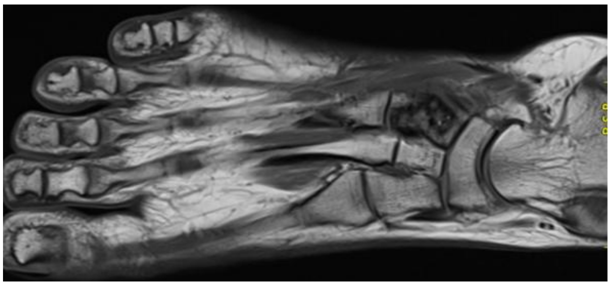

Fig 6: Comparative postoperative MRI scan T2 sequence axial view

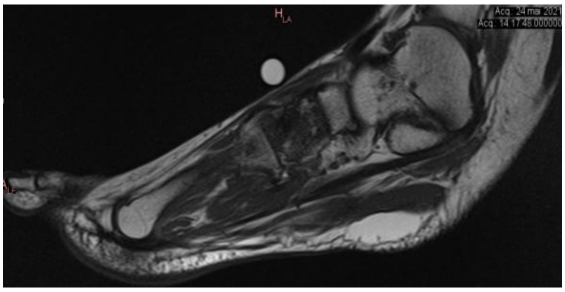

Fig 7: Comparative preoperative MRI scan T2 sequence sagittal view

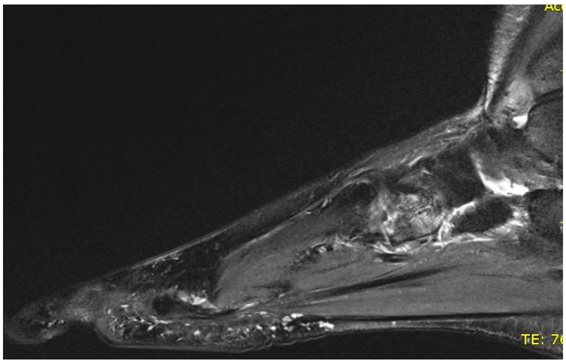

Fig 8: Comparative postoperative MRI scan T1 sequence sagittal view

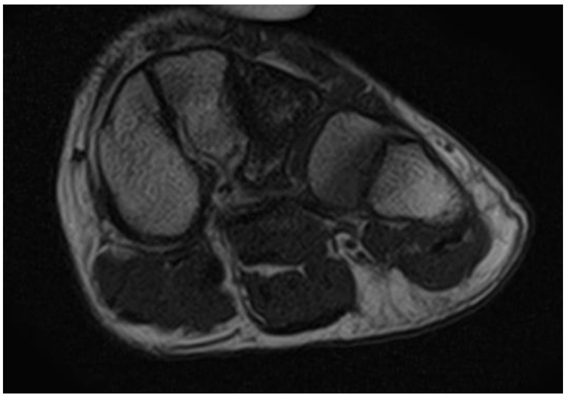

Fig 9: Comparative preoperative MRI scan T2 sequence coronal view

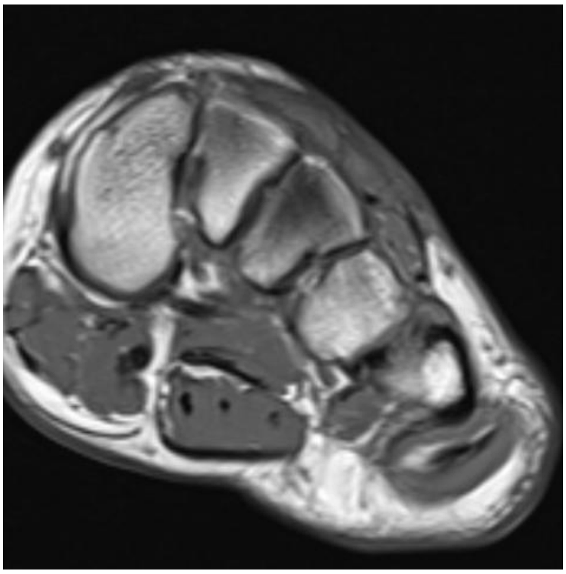

Fig 10: Comparative postoperative MRI scan T2 sequence coronal view

Conclusion

Osteonecrosis of the lateral cuneiform is a particularly rare condition and can be successfully treated with surgical decompression with subchondral drilling. Although there are no records of previously documented similar cases, the standard treatment of the disease in other locations seems to also work in the lateral cuneiform. Since this disease can simulate many other pathologies, physicians should be aware of its presence in this particular location. Even though avascular necrosis of the tarsal bones is infrequent, it should always be considered in the differential diagnosis of both pediatric and adult foot pain.

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Conflict of Interest

Not available.

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Not available.

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