Bone marrow edema syndrome: an unusual cause for spontaneous unrelentless bilateral knee pain

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ABSTRACT

Bone marrow edema syndrome is a rare disease with an unknown etiology, self-limited and usually associated with an indolent course, which can also generate severe pain with tremendous functional impairment. The authors present a case of a 19-year-old female patient with a progressive, non-traumatic and unrelentless pain involving both knees, requiring persistently walking aids and analgesic drugs. The imaging studies showed a bilateral distal femur and proximal tibia bone marrow edema in the magnetic resonance imaging. Finally, and after an extensive investigation without any abnormal findings, a bone marrow edema syndrome diagnosis was established, with a spontaneous regression of the clinical and imaging presentation. One year after the initial complaints the patient is fully recovered, without pain or medication, presenting an MRI showing complete regression of the initial findings.

Despite the rarity of this entity, being aware of its existance and clinical manifestations is crutial to allow a proper diagnosis. The case herein presented is, to our understanding, pragmatic regarding bone marrow edema syndrome presentation and clinical course.

Keywords: Bone marrow edema syndrome; Idiopathic bone marrow edema; Clinical symptoms; Differential diagnosis; Patient management.

INTRODUCTION

Bone marrow edema syndrome (BMES) is a rare disease with unknown etiology, characterized by a transient course of pain with unspecific imaging findings of bone marrow edema[1,2]. Characteristically, BMES presents an increased water content translated into intermediate signal on magnetic resonance imaging (MRI) T1-weight images and high signal on T2-weight images, in particular with fat supression. Despite other imaging evaluations can be performed to aid differential diagnosis, BMES is still an exclusion diagnosis[3,4]. Herein, the authors intended to highlight a rare cause for knee pain, with an unusual exacerbated presentation, which is often misdiagnosed in clinical practice.

CASE REPORT

A 19-year old melanodermic female-patient was sent to our practice due to a spontaneous, non-mechanic and non-traumatic, bilateral knee pain, which started months ago requiring pain management with analgesic drugs. The patient had no relevant medical or family history. Clinical examination revealed bilateral intense pain during tibial metaphysis and femoral condyle palpation, without any other positive finding. Despite an MRI presenting bilateral distal femur and proximal tibia bone marrow edema (Figure 1), the remaining imaging studies (Figure 2), bone scintigraphy, blood tests (including investigation for hematologic disorders) were negative. Also, the bone biopsy result reported normal bone without any abnormalities.

A BMES diagnosis was made and a conservative management was adopted, which consisted in optimized pain control management and avoidance of weight bearing. The symptoms persisted for a few more months, after which the patient described a spontaneous gradual improvement to fully asymptomatic after eight months of follow up. Control MRI showed remission of the initial bilateral knee bone marrow edema (Figure 3).

DISCUSSION

This case illustrates a typical presentation for a difficult and not often misdiagnosed clinical condition with unspecific features. BMES has an unknown etiology and

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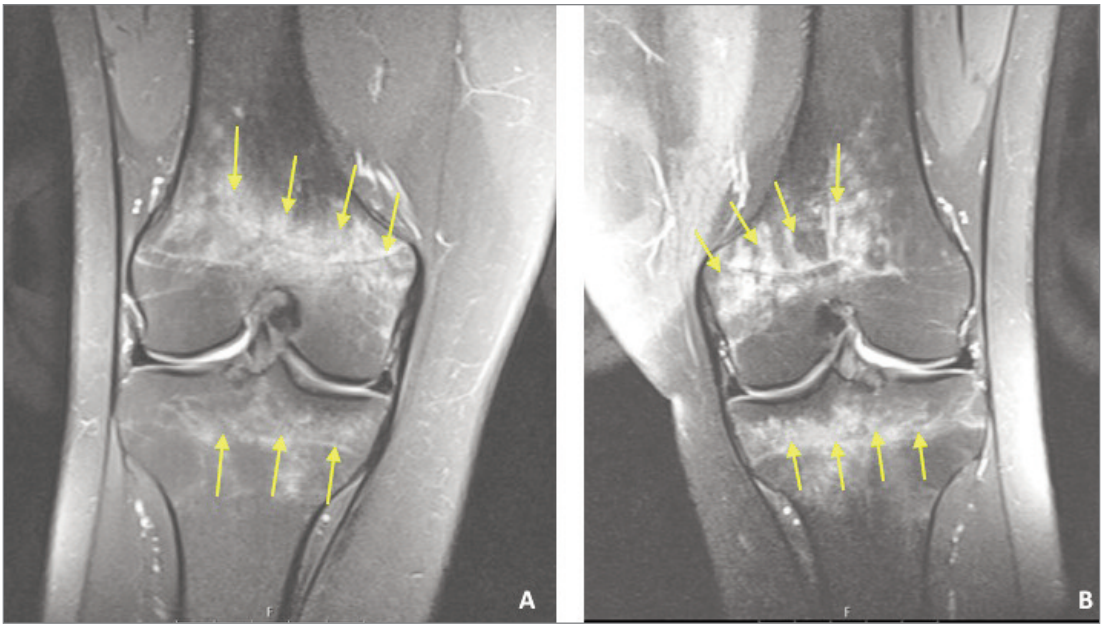

FIGURE 1. T2-weighted (DP SPAIR) MRI coronal plane of the right (A) and left (B) knee, revealing a diffuse area of edema on both femoral and tibial metaphysis.

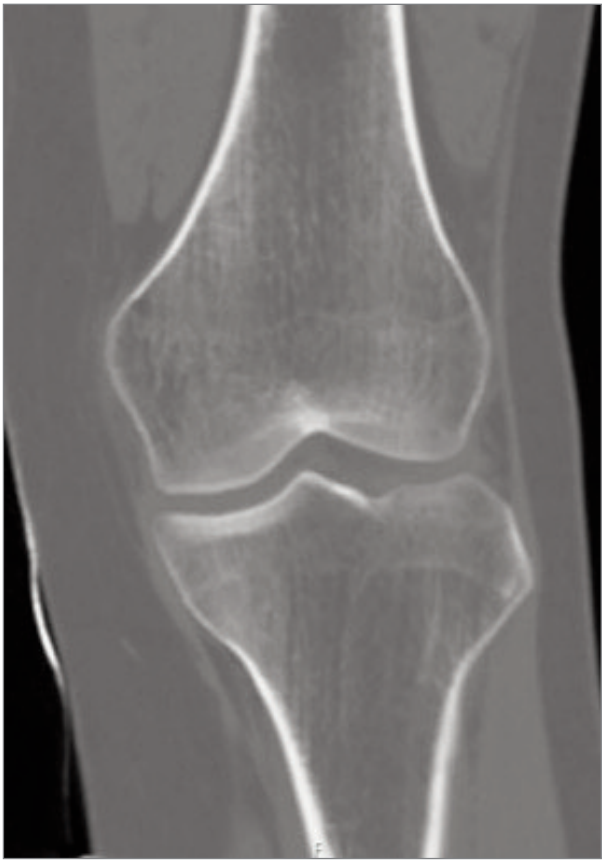

FIGURE 2. CT-scan coronal image of one of the left knee showing no aparent bone abnormalities.

pathogenesis, still being an exclusion diagnosis. Differential diagnosis (Table I) is vital to allow proper management and our patient had no other cause to present a bilateral distal femur and proximal tibia bone edema associated with excruciating pain. This syndrome most often appears in the hip, however, the knee is the second most common anatomical location[1,2,4]. As previously described in literature, BMES is a self-limiting condition, and as such, a recommendation towards supporting treatment was made4. In the past, attempts to hasten recovery with systemic or local drugs have failed to change the natural history of the disease[5]. After eight months of follow up the patient improved greatly, being pain free and presenting a follow up MRI with remission of all bone medullary edema. In the end, these findings also supported the BMES diagnosis.

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REFERENCES

1. Korompilias AV, Karantanas AH, Lykissas MG, Beri AE. Bone marrow edema syndrome. Skeletal Radiol. 2009; 38:425-436
2. Thiryaki WA, Thiryayi SA, Freemont AJ. Histopathological perspective on bone marrow oedema reactive bone change and

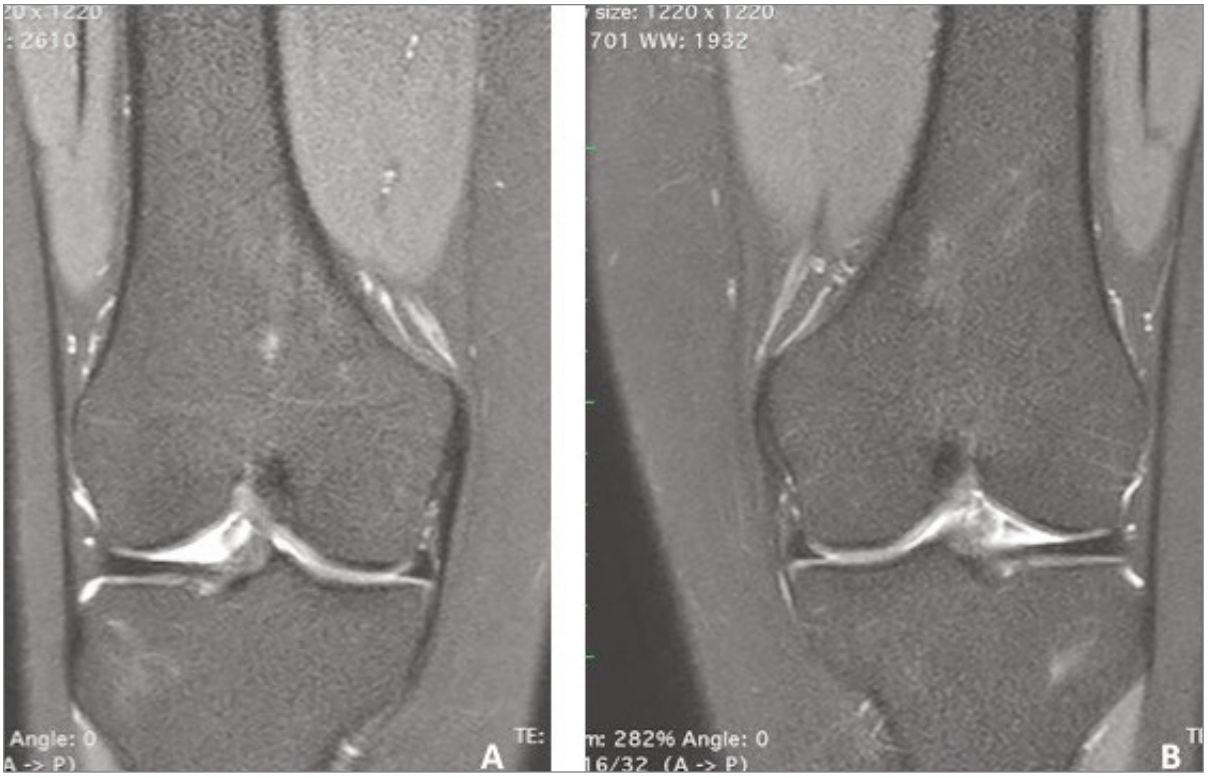

FIGURE 3. T2-weighted (DP SPAIR) MRI coronal plane of the right (A) and left (B) knee, eight months after the initial MRI, showing full regression of the initial findings.

TABLE I. CAUSES FOR BONE MARROW EDEMA

Causes for bone marrow edema	Etiology
Fracture	Traumatic
Post-operative	Traumatic
Complex regional pain syndrome	Traumatic
Infection	Septic
Inflammation	Inflammatory
Osteoarthritis	Degenerative or mechanical
Osteochondral lesions	Degenerative or mechanical
Bone stress injuries	Degenerative or mechanical
Tumors (benign or malignant)	Neoplastic
Avascular osteonecrosis	Ischemic
Neuro-osteoarthropathy	Neurogenic
Osteoporosis (primary or secondary)	Metabolic
Bone Marrow Edema Syndrome	Idiophatic (exclusion diagnosis)

(Adapted from the Ludwig Maximilians University consensus classification for bone marrow edema according to their etiology[6])

haemorrhage. Eur J Radiol. 2008; 67(1): 62-7
3. Starr AM, Wessely MA, Albastaki U, Pierre-Jerome C, Kettner NW. Bone marrow edema: pathophysiology, differential diagnosis, and imaging. Acta Radiol, 2008; 49(7):771-86
4. Patel, S. Primary bone marrow oedema syndrome. Rheumatology. 2014; 53, Issue 5: 785–792
5. Papadopoulos EC, Papagelopoulos PJ, Kaseta M, Themistocleous GS, Korres DS. Bone marrow edema syndrome of the knee: a case report and review of the literature. The Knee. 2003; 10(3): 295–302
6. Baumbach SF, Pfahler V, Pozza BD, et al. How we manage bone marrow edema—an interdisciplinary approach. Journal of clinical medicine. 2020. 9(2), 551