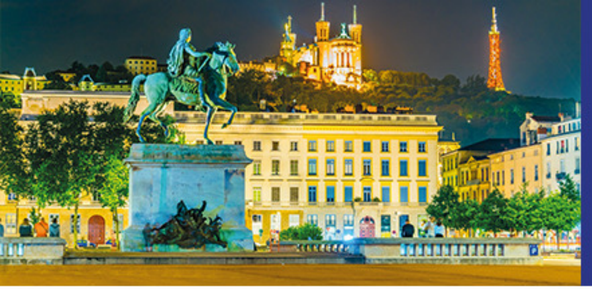

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Bone marrow oedema syndrome of the foot and ankle in a paediatric population: a retrospective case series with serial MRI evaluation

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Background

Bone marrow oedema syndrome (BMOS) refers to a group of conditions characterized by extremity pain and increased interstitial fluid within the bone marrow, with unknown cause. BMOS is a **relatively rare** condition, not often described for the upper limb or in the paediatric population, mostly affecting the hip and knee in adults. Because foot and ankle involvement in BMOS is uncommon and has unspecific symptoms, there is often a **delay in diagnosis**, leading to an extended period of pain and impaired functionality. BMOS is characterized by a **self-limiting course** with a symptom duration between three and 12 months. Treatment of BMOS focusses on **pain relief and shortening of symptom duration**, mostly consisting of non- or partial weight-bearing, nonsteroidal anti-inflammatory drugs and physiotherapy. Pharmacological treatments can include iloprost or bisphosphonates. Surgical treatment can consist of core decompression or drilling. In imaging studies, focused on tarsal bone marrow in children, high-signal T2-weighted changes of the bone marrow on MRI are mostly considered to be a **physiological finding** representing residual hematopoietic marrow. A limitation of many of these studies is the lack of clinical data and the absence of a correlation between MRI findings and the clinical evaluation.
In this study we would like to re-evaluate the underdiagnosed concept of BMOS in the foot and ankle in a paediatric population. By means of a case series we observe the correlation between clinical symptoms and MRI imaging features.

Materials and methods

- Retrospective database search
- 1.5-tesla magnet MRI. Standard MRI protocols included coronal, sagittal and axial T1-weighted sequences, axial T2-weighted sequences and sagittal turbo inversion recovery magnitude (TIRM), T2 3D-double-echo steady state with water excitation and T2-weighted sequences
- Review by first author and specialist musculoskeletal radiology
- Query: multiple MRI scans of the foot and ankle, population born on or after 1 January 2001
- N=6, after exclusion for causes of secondary oedema (e.g. fractures, malignancies or infectious disease), physiological appearance of oedema or lack of clinical data
- Control group N=10

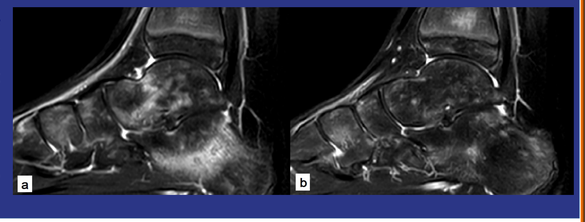
Results

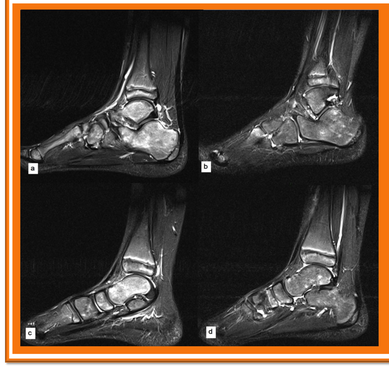
Table 1 Demographic and clinical features

Case	1	2	3	4	5	6
Sex	Female	Male	Female	Male	Female	Male
Age at onset	9 yrs 8 mths	8 yrs	14 yrs 8 mths	14 yrs 10 mths	8 yrs 3 mths	10 yrs 4 mths
Medical history	IgA vasculitis, Hashimoto thyroiditis	None	None	None	None	None
History of trauma (before first presentation)	Grade 2 ankle sprain 6 mths prior	None	None	None	Fall from stairs, undisplaced fracture base proximal phalanx hallux 5 mths prior	Undisplaced fracture distal MT3-4 after step on irregular hard object 4 mths prior
Duration of symptoms before diagnosis	6 mths	4 mths	1 yr 3 mths	1 mth	5 mths	5 mths
Subjective complaints	Pain on stance, need for crutches or wheelchair	Pain during soccer, quit sports	Pain during running and jumping, quit volleyball	Pain during soccer, quit sports	Unable to bear weight	Pain during gait and sports
Specific treatment	Soft insoles	Weight-bearing cast 4 wks, insoles	No additional treatment	Short leg walker boot, weight baring cast 4 wks, walker boot, orthotic insoles	Weight-bearing soft cast	No additional treatment
Treatment duration	3 mths	11 mths	6 mths	14 mths	15 mths	4 mths
Time until improvement of initial symptoms	4 mths	1 yr	6 mths	2 mths	6 mths	5 mths
Recurrence/migration	Pain on sporting activities 5 mths after regression of initial symptoms	Migration to TMT1 pain 6 mths after start of treatment	None	Progression of pain at same site after initial improvement, 10 mths after start of treatment	Stagnation of symptoms after 6 mths of initial improvement	None
Final regression of symptoms after start of treatment	16 mths	1 yr	6 mths	14 mths	16 mths	5 mths

after minor trauma
migratory disease
recurrent disease

♀ 9y, pain localized to sinus tarsi
(a) T2-w MRI: patchy areas of increased signal intensity at anterior talar body and bow-shaped at midportion of the calcaneus
(b) Regression of patchy areas after 7m with improvement of symptoms, remaining punctiform red bone marrow islands





♂ 8y, atraumatic pain in the sinus tarsi on weight-bearing initially
(a and c) Sagittal turbo inversion recovery magnitude MRI, patchy areas in talus and calcaneus
(b and d) Pain transfer to first tarsometatarsal joint after seven months of restricted weight-bearing, correlating with diminished signal intensity in talus and calcaneus, and new appearance of marrow oedema at the proximal metatarsal area

Discussion

- Differential diagnosis of complex regional pain syndrome, common pathology in child (ankle instability and Severs disease)
  - Δ Assess for **risk factors** such as lipid metabolism disorder or vitamin C/D deficiency
  - Δ Can have a **unique** localization, mostly in the talus, or in a **diffuse** pattern
  - Δ Can follow a **migratory** or **recurring** disease course

- BMOS defined by **patchy areas** of T2-weighted high intensity signal
  - ⇔ small **punctiform** increased signal intensity foci as physiological residual islands of red bone marrow in three children after symptom resolution
  - ⇔ no well-defined patchy area involvement of increased signal intensity on T2 and TIRM-weighted images in control group

- History of **minor trauma** in 3 children
  - ► part of everyday life in children, should not lead to fulminant reaction
  - ► contusion or altered weight bearing pattern possible

- Treatment
  - ✚ Goal = **minimize pain symptoms**, maintain **functionality**
  - ✚ Conservative
    - Rest and restricted weight-bearing
    - Insoles, cast, walker boot when necessary
    - NSAID
  - ✚ Iloprost? Insufficient data on long term, risk of adverse effects
  - ✚ Decompressive drilling? Moderate results in calcaneum

Conclusion

In this case series both **clinical and MRI data** of six paediatric patients with BMOS of the foot and ankle is reported. As BMOS is **transient and likely self-limiting**, we believe no pharmacological treatment is warranted, and conservative treatment measures should be implemented to **improve patient comfort** during the disease course. The clinical progression of symptoms shows a parallel course to the MRI findings, making **consecutive follow-up MRI imaging unnecessary**. A swift and correct diagnosis of BMOS, based on clinical findings and bone marrow oedema patterns on MRI with no definite underlying causes, could **prevent unnecessary diagnostic investigations and invasive treatments** for patients.

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