Scholars Journal of Medical Case Reports (SJMCR) **ISSN 2347-6559 (Online)**
Abbreviated Key Title: Sch. J. Med. Case Rep. **ISSN 2347-9507 (Print)**

A United of Scholars Academic and Scientific Society, India

Melorheostosis: About A Case and Reviewed the Literature

Omar Ben Hazim*, Mohamed Kharmaz, Lagdid Abdelkrim, Moncef Boufettal, Reda-Allah Bassir, Molay Omar Lamrani, Mohamed Ouadghiri, Mustapha Mahfoud, Ahmed El Bardouni, Mohamed Saleh Berrada
Department of Orthopedic Surgeryand Traumatology, Ibn Sina Hospital, University Mohamed V, Rabat, Morocco

***Corresponding author**
Omar Ben Hazim

Article History
Received: 23.07.2018
Accepted: 05.08.2018
Published:30.08.2018

DOI:
10.36347/sjmcr.2018.v06i08.005

Abstract: Melorheostosis is defined as progressive bone dysplasia, which can lead to painful complications and deformities. The most common symptoms are pain and limitations of joint mobility due to bone and soft tissue deformities. The diagnosis is based on radiology. Here we report the case of a 39-year-old woman who has been consulting for left ankle pain of increasing intensity for two years, in whom standard radiographs were characteristic of melorheostosis. MRI was performed to better study the behavior and extent of lesions.
Keywords: Melorheostosis, bone dysplasia, osteopathy.

INTRODUCTION

Melorheostosis is defined as progressive bone dysplasia, which can lead to painful complications and deformities [1]. The most common symptoms are pain and limitations of joint mobility due to bone and soft tissue deformities [2]. The diagnosis based on radiology [3,4]. Its appearance is characterized by linear cortical hyper-condensation in "candle casting" [5] on standard radiography. MRI allows specifying the nature, the seat and the extension of the lesions, in particular with the soft parts as well as the evaluation of the bone deformations for a possible surgical correction.

CASE REPORT

A 39-year-old woman who consults for left ankle pain of increasing intensity for two years. Standard radiographs were characteristic of melorheostosis with extended left lateral monomelic involvement on the left lower limb axis. These lesions took the appearance of cortical osteoconductive flows, relatively well limited, parallel to the major axis of the femur and the fibula. In addition to these lesions, there are multiple foci of nodular osteosclerosis in the tarsal bones, particularly the calcaneus and the fibula head. Ossification of the external pararticular soft tissues of the left knee are clearly visible on the left knee X-rays.

MRI shows the presence of multiple nodular hyposignal formations on all sequences at the posterolateral side of the lateral femoral condyle, extending to the pararticular soft parts to the head of the fibula. It associates a perilesional hypersignal on the T2 sequences involving the muscular structures. MRI also shows a thickening of the cortical fibula in irregular nodular signal and responsible for a more or less significant reduction of the mudullary cavity. These signal lesions extend along the soft parts of the lateral surface of the foot with calcaneal and tarsometatarsal involvement. The diagnosis of melorheostosis was already retained on standard radiographs. MRI was performed to better study the behavior and extent of lesions.

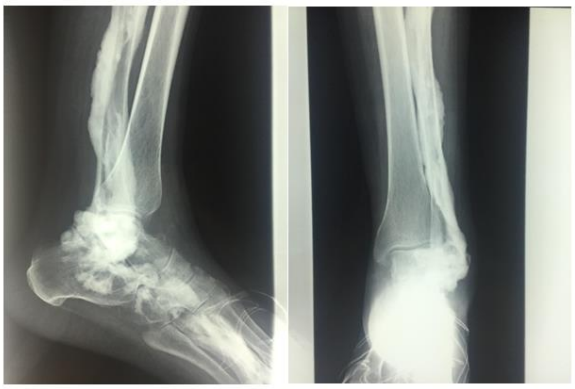

Fig-1 and 2: Ankle x-rays showing melorheostosis

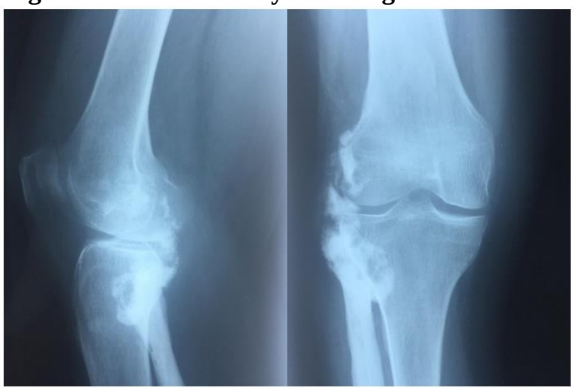

Fig-3 and 4: knee x-rays showing melorheostosis

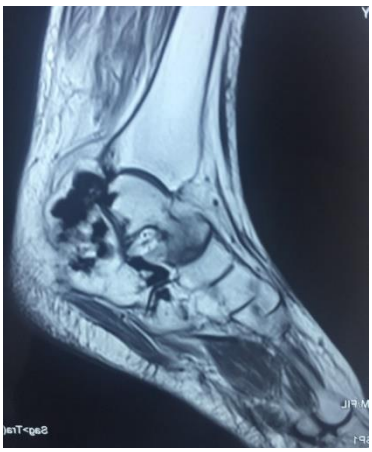

Fig-5: MRI of ankle showing Melorheostosis

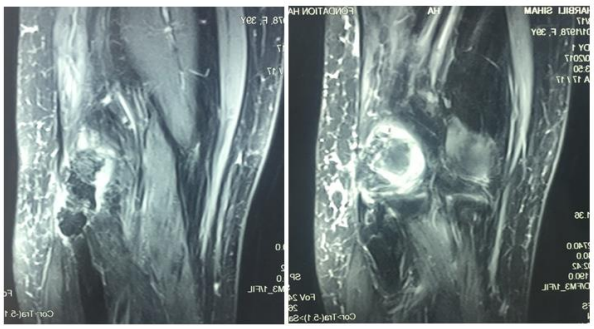

Fig-6 and 7: MRI of the knee showing Melorheostosis

DISCUSSION

Melorheostosis is a rare pathology that affects both sexes. Described for the first time by Leri and Joanny in 1922 [3,4]. This osteopathy begins in childhood with rapid growth during adolescence [5, 6-9]. Its incidence is estimated at 0.9 per million inhabitants [8, 10]. Its etiology remains unknown.

However, some authors suggest that it comes from an attack of one or more sensitive nervous territories (sclerotomes); an attack that may be infectious vascular, degenerative, inflammatory or embryonic [7, 8, 10-12]. Melorheostosis mainly affects membranous ossification and, to a lesser extent, enchondral ossification in a mono, polyostotic or monomelic manner [9].

With an insidious onset, marked by pain such as neuralgia or arthralgia [2], this pathology gradually settles causing joint stiffness and deformity [2, 4, 7, 8, 10-13]. . Soft tissue lesions are frequently associated with osteoarticular structures; cases of band scleroderma have also been reported [2,8]. Associated vascular malformations were reported in 17% of cases [10], an angiodysplasia underlying bone involvement [1, 8, 13].

The diagnosis of melorheostosis is radiological; it is a linear hypercondensation in "candle casting" characteristic along the bones [5, 6]. This

condensation has a metameric distribution [5]. This osteosclerosis often reaches the cortex with a possible propagation in the medulla in the form of a hyperostosis [1, 4, 5, 12].

This pathology affects the limbs following a unilateral, almost vertical extension from the hip to the toes and the shoulder to the fingers [5, 8].

As for soft tissue abnormalities, it is most often fibrosis and edema of muscles and adipose tissue. Ossification of the soft tissues is rare and simulates osteochondromas [7,8]. Large ossifications can be seen at the joints CT is not indicated systematically, it finds its place in the evaluation of the extent of lesions preoperatively or more rarely in some pseudotumoral forms.

MRI too is not systematic. It allows precision soft tissue extension and evaluation of bone deformities for possible surgical correction [9,12]. This is the best imaging technique in the pre-operative checkup. The lesions of melorheostosis typically appear hypo signal on all sequences, without modifying after injection of gadolinium [9, 11, 14]. The foci of para-articular ossification present a signal identical to the cortical lesions.

Usually the pathological study is useless. In histology, cortical hyperostosis is a combination of mature and immature bone tissue separated by thick osteoid trabecula ions filling the Haversian canals [2, 7] and associated with fibrous tissue all around these foci of ossification.

Differential diagnosis is mainly with paraosteal osteosarcoma irrespective of localization and myositis ossificans and calcified hematoma in cases associated with calcification or ossification of the soft tissues [5, 8, 11,14].

According to the authors, medical treatment with bisphosphonates is effective on the symptoms and not on the progression of bone lesions. The use of surgical corrections is justified in severe cases while respecting the maturation of the skeleton [4, 8, 12].

CONCLUSION

Melorheostosis is a rare condensing bone dysplasia, its diagnosis is based on imaging, especially standard radiographs. MRI offers a new semiology to be added to the standard radiology-scintigraphy pair.

REFERENCES

1. Brown RR, Steiner GC, Lehman WB. Melorheostosis: Melorheostosis: case report with radiologic-pathologic correlation. Skeletal Radiol. 2000;29:548-52.
2. Freyschmidt J. Melorheostosis: a review of 23 cases. Eur Radiol. 2001;11:474–9.
3. Ameen S, Nagy L, Gerich U, Anderson SE. Melorheostosis of the hand with complicating bony spur formation and bursal inflammation: diagnostis and treatment. Skeletal Radiol. 2002;31:467-70.
4. Biaou O, Avimadje M, Guira O, Adjagba A, Zannou M, Hauzeur JP. Melorheostosis with bilateral involvement in a black African patient. Joint Bone Spine. 2004 Jan 1;71(1):70-2.
5. Maroteaux P, Fauré C, Fessard C, Rigault P, Le Merrer M. Les maladies osseuses de l'enfant, 3rd ed. Medecine- Sciences : Flammarion. 1995:190-2.
6. Nasu K, Kuroki Y, Nawano S, Murakami K, Kuroki S, Sekiguchi R, Hayashi T, Umeda T. Thoracic cage extent of melorheostosis depicted by multislice CT. Skeletal radiology. 2002 Aug 1;31(8):464-6.
7. Ethunandan M, Khosla N, Tilley E, Webb A. Melorheostosis involving the craniofacial skeleton. Journal of Craniofacial Surgery. 2004 Nov 1;15(6):1062-5.
8. Mariaud-Schmidt RP, Bitar WE, Perez- Lamero F, Barros-Nunez P. Melorheostosis: unusual presentation in a girl. Clin Imaging. 2002;26:58-62.
9. Osgood GM, Lee F, Parisien MV, Ruzal-Shapiro C. Magnetic resonance imaging depiction of tight iliotibial band in melorheostosis associated with severe external rotation deformity, limb shortening and patellar dislocation in planning surgical correction. Skeletal radiology. 2002 Jan 1;31(1):49-52.
10. Ingen-Housz-Oro S, Chigot V, Hamel-Teillac D, Brunelle F, DE PROST Y. Mélorhéostose associée à une malformation artério-veineuse de l'oreille. InAnnales de dermatologie et de vénéréologie 2001 (Vol. 128, No. 8-9, pp. 915-918). Masson.
11. Brennan D, Bruzzi J, Thakore H, O'Keane J, Eustace S. Osteosarcoma arising in a femur with melorheostosis and osteopathia striata. Skeletal radiology. 2002 Aug 1;31(8):471-4.
12. Donáth J, Poór G, Kiss C, Fornet B, Genant H. Atypical form of active melorheostosis and its treatment with bisphosphonate. Skeletal radiology. 2002 Dec 1;31(12):709-13.
13. Greenspan A, Azouz M. Bone dysplasia series: melorheostosis, review and update. Canadian Association of Radiologists Journal. 1999 Oct 1;50(5):324.
14. Joseph SY, Resnick D, Vaughan LM, Haghighi P, Hughes T. Melorheostosis with an ossified soft tissue mass: MR features. Skeletal radiology. 1995 Jul 1;24(5):367-70.