

CrossRef DOI of original article:

1           A Patient with Symptomatic Polyostotic Melorheostosis  
2           Successfully Treated with Intravenous Zoledronate Injection-A  
3           Case Report

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5           Received: 1 January 1970 Accepted: 1 January 1970 Published: 1 January 1970

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7           **Abstract**

8           Introduction: Melorheostosis is a rare congenital bone disorder present with insidious onset of  
9           bone pain. Diagnosing this rare condition and managing it is difficult. Knowledge of this rare  
10          entity is important for the physician who is treating patients with musculoskeletal pain. Case  
11          Report: A 25-year-old female with right leg pain for a two-year duration. She has been seen  
12          by a lot of physicians and could not have been managed properly. Her radiographs were  
13          showing hyperostotic left ulna and right tibia. Sinister pathologies were excluded using MRI  
14          and biopsy. Her symptoms settled with intravenous Zoledronate. Pain recurred after two years  
15          and she responded well after repeated Zoledronate injection.

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17           **Index terms—**

18           **1 I. INTRODUCTION**

19          Polyostotic melorheostosis is a rare congenital non-hereditary benign sclerosing mesenchymal dysplasia involving  
20          multiple bones (1)(2)(3)(4). Melorheostosis is presented in the second decade of life. The male and female ratio  
21          is the same. The classical description of appearance in the radiographs is "flowing candle wax" (1). The axial  
22          skeleton is rarely involved. The treatment is mainly non-operative. Surgery is reserved for the lesions complicated  
23          with fractures.

24          We present a case report on a patient with symptomatic polyostotic melorheostosis successfully treated with  
25          intravenous Zoledronate injection.

26          A 25-year-old female presented to the orthopaedic unit with pain over her right leg left forearm and back for  
27          two years duration. She has been seen by a lot of physicians for the same ailment and they could not help her.  
28          The pain gradually worsened over time and made her visit us. On examination, the right leg and the left forearm  
29          were swollen diffusely. There is no history of back pain.

30          Radiographs show hyperostosis of the right tibia and the left ulna (Figure 1). Technetium 99 bone scan  
31          revealed hyperactivity seen on the right tibia, left ulna, left fibula, left femur and the spine (Figure 2).

32           **2 III. DISCUSSION**

33          Melorheostosis is a rare disorder in which sclerosing dysplasia with derangement in endochondral and intramembranous ossification of bones. The summary of demographical details is in Table ???. These lesions can be on  
34          one bone (Monostotic), many bones (Polyostotic), involving one limb (Monomelic) or generalized. Axial skeleton  
35          involvement is rare. According to the anatomical involvement, severity and the deformity caused by the lesions  
36          symptoms and signs will be varied.

37          Commonest symptoms are painful limb swelling, restricted range of motion and contractures  
38          of joints (usually asymmetric). Axial skeleton involvement may present with back, and neck  
39          pain, radicular pain, scoliosis, stiffness, giddiness, symptoms of vertebral insufficiency and even  
40          compression (1).

### **3 IV. CONCLUSION**

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42 Melorheostosis is not a fatal condition. But it impacts significantly the activity of daily living of patients  
43 with this ailment. The management of melorheostosis varies from simple analgesics to surgery. The management  
44 depends on the anatomical site, the extent of lesions, the severity of symptoms and the degree of deformity.  
45 The primary goal of management is to relieve symptoms and achieve mobility. Pain relief, nerve blocks, braces,  
46 and physiotherapy play a vital role. Patients who are not responded to these options will undergo nerve blocks.  
47 Surgery deserves for patients with lesions complicated with fractures.

48 There are few case reports on the usage of intravenous zoledronate for melorheostosis (6)(7)(8). They reported  
49 successful outcomes following treatment after the injection. On our patient single dose of intravenous zoledronic  
50 acid (5mg over 30 minutes) has provided remission for two years and it worked for relapse as well after two  
51 years. Intravenous zoledronate is an effective treatment for symptomatic polyostotic melorheostosis. Zoledronate  
52 is a bisphosphonate which inhibits osteoclasts and reduces bone pain, prevents pathological fracture and reduces  
53 blood supply (6)(7)(8). Bone resorption by osteoclasts, stimulation of pain receptors and increased intraosseous  
54 pressure are major reasons for bone pain among patients with melorheostosis (6-8). Thus, zoledronate is a viable  
55 option for the ones with melorheostosis causing bone pain. Anyway, careful selection of patients and preparation  
56 is necessary before the zoledronate infusion to avoid complications (Avascular necrosis of the jaw, fever, allergic  
57 reactions etc.).

### **58 3 IV. CONCLUSION**

59 Symptomatic polyostotic melorheostosis may be resistant to symptomatic treatment. Intravenous zoledronate is  
a viable option for such patients.



Figure 1: 6 A





Figure 3: Figure 2 :



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Figure 4: Figure 3 :



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61 [ London Journal of Medical and Health Research REFERENCES] , *London Journal of Medical and Health*  
62 *Research REFERENCES*

63 [Sureka et al.] , B Sureka , Kumar Mittal , M Udhaya , K Sinha , M Mittal , A , Bhushan Thukral , B  
64 Melorheostosis . *Two atypical cases* 24.

65 [ Indian Journal of Radiology and Imaging ()] , *Indian Journal of Radiology and Imaging* 2014.

66 [Kumar et al. ()] , R Kumar , S S Sankhala , I Bijarnia , Melorheostosis-Case . [www.jocr.co.in](http://www.jocr.co.in) 2014.

67 [Case report of a rare disease: Melorheostosis Journal of Medical Case Reports and Case Series (2021)] ‘Case  
68 report of a rare disease: Melorheostosis’. *Journal of Medical Case Reports and Case Series* 2021 Dec 10.

69 [Kherfani et al. ()] A Kherfani , H Mahjoub , Melorheostosis . *A rare entity: A case report*, 2014. 18 p. 251.

70 [Hollick et al. ()] ‘Melorheostosis and its treatment with intravenous zoledronic acid’. R J Hollick , A Black , D  
71 Reid . *Case Reports* 2010 Apr 5. 2010. apr05 1. p. .

72 [Drr et al. (2018)] ‘Melorheostosis: Case report of rare disease’. Mehrotra Drr , Kumar Drp , Chaudhary Drd ,  
73 Patel Drp , Singh Dra . *International Journal of Orthopaedics Sciences* 2018 Jan 1. 4 (1g) p. .

74 [Slimani et al. ()] *Successful treatment of pain in melorheostosis with zoledronate, with improvement on bone*  
75 *scintigraphy. Case Reports*, S Slimani , A Nezzar , H Makhloufi . 2013 Jun 21. 2013. jun21 2. 2013009820. p.  
76 2013009820.

77 [Slimani et al. (2013)] *Successful treatment of pain in melorheostosis with zoledronate, with improvement on bone*  
78 *scintigraphy. Case Reports*, S Slimani , A Nezzar , H Makhloufi . 2013 Jun 21. 2013. jun212. 2013009820. p.  
79 2013009820.