Acta Orthopaedica et Traumatologica Turcica 53 (2019) 233–237

Contents lists available at ScienceDirect

Acta Orthopaedica et Traumatologica Turcica

journal homepage: https://www.elsevier.com/locate/aott

Successful percutaneous treatment of osteoid osteoma in a 13 month-old boy with radiofrequency ablation under CT guidance

Cennet Sahin [a, *], Yunus Oc [b], Naim Ediz [c], Aylin Hasanefendioglu Bayrak [d]

[a] University of Health Sciences, Istanbul Sisli Hamidiye Etfal Training and Research Hospital, Radiology Department, Istanbul, Turkey
[b] Tekirdag Corlu Vega Hospital, Orthopedics Department, Turkey
[c] University of Health Sciences, Istanbul Sisli Hamidiye Etfal Training and Research Hospital, Anesthesiology Department, Turkey
[d] University of Health Sciences, Gaziosmanpasa Training and Research Hospital, Radiology Department, Turkey

ARTICLE INFO

Article history:
Received 15 April 2018
Received in revised form
17 September 2018
Accepted 8 October 2018
Available online 13 November 2018

Keywords:
Osteoid osteoma
Radiofrequency ablation
Computer tomography
Pediatric
Treatment
Percutaneous
CT guidance
Efficacy
Safety

ABSTRACT

We present a 13-month-old boy who had a successful Computed Tomography (CT) guided percutaneous radiofrequency ablation (RFA) treatment for the osteoid osteoma (OO) on proximal part of the tibial diaphysis.

The complaints of the patient were being restless due to pain and refusing to bear any weight on his left leg for 6 months. An asymmetrical cortical thickening and a focal sclerosis was detected on medial proximal diaphysis of the left tibia on radiographs and axial T2-weighted STIR-MR image showed bone marrow and soft-tissue edema with low-signal-intensity nidus due to central calcification with a high-signal-intensified unmineralized periphery. CT findings (the nidus on the cortex of tibia with well circumscribed lucent region around a central sclerotic dot and cortical thickening around the nidus) confirmed the diagnosis of OO.

After CT guided percutaneous RFA treatment, the patient had an immediate pain relief in 24 h after and could bear weight on the leg. 12 and 16 months after RFA respectively, CT images and radiographs revealed sclerotic healing of the nidus and a slow regression of the adjacent cortical thickness without any recurrence.

Introduction

Osteoid Osteoma (OO) is one of the benign osteogenic bone tumors that predominantly affects males.[1–5] OO can be seen in a range of 8 months and 70 years of age.[6] The prevalence of OO under 5 years old is shown as 3–8% in the literature.[6] However, the prevalence under 1 year of age is very rare.[6–8] Diagnosis of OO can be challenging under the age of 5 and diagnosis is made according to specific radiological and clinical findings.[9–12] In treatment, radiofrequency ablation (RFA) has a high technical and clinical success and it has been standard treatment method in general population.[13–17] However, there is limited data about its use in pediatric population, especially under 5 years of age.[6,7] We aimed to present a 13-month-old boy who was treated successfully with Computed Tomography (CT) guided percutaneous RFA in our clinic.

Case report

A 13-month-old boy with OO on the proximal diaphysis of his left tibia was referred to our clinic for CT guided RFA treatment. The complaints of the patient were being restless due to pain and refusing to bear any weight on his left leg since he was 7-month-old. The boy's mother told that he woke up with crying in midnights and he had rest after having non-steroidal anti-inflammatory (NSAID) drugs. An asymmetrical cortical thickening and a focal sclerosis was firstly detected on medial proximal diaphysis of the left tibia on radiograph (Fig. 1). Magnetic resonance imaging (MRI) was performed in continuation for further evaluation in the peripheral hospital. Axial T2-weighted STIR-MR image showed the low-signal-intensity nidus due to central calcification with a high-signal-intensified un-mineralized periphery. Bone marrow and

* Corresponding author. Tel.: +90 505 748 57 41.
E-mail addresses: cennetsahin2@hotmail.com (C. Sahin), yunti72@gmail.com (Y. Oc), naimediz@gmail.com (N. Ediz), aylin_has@yahoo.com (A. Hasanefendioglu Bayrak).
Peer review under responsibility of Turkish Association of Orthopaedics and Traumatology.

https://doi.org/10.1016/j.aott.2018.10.002

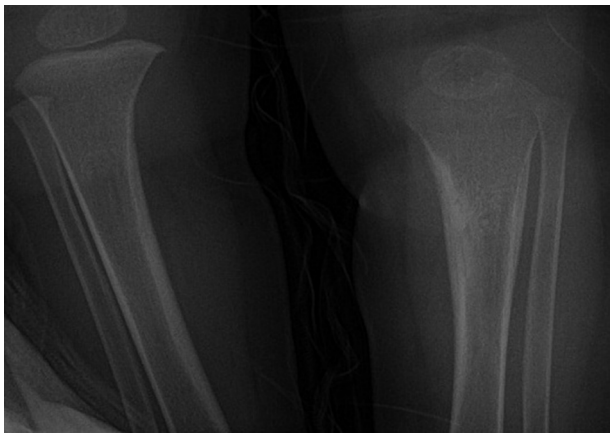

Fig. 1. On pre-treatment antero-posterior radiograph of bilateral lower limb, an asymmetrical (compared to right tibia) cortical thickening with focal sclerosis on proximal diaphysis of left tibia is seen. There is a suspicious oval shaped radiolucency with a central sclerotic dot in this focal sclerosis.

soft-tissue edema was well demonstrated on T2-weighted MR (Fig. 2). Since there was a remarkable bone marrow edema on MRI, osteomyelitis, post-traumatic injury/infection, malignant bone tumors (such as Ewing sarcoma) and osteoid osteoma were thought as differential diagnoses.

The laboratory tests for infection parameters (erythrocyte sedimentation rate, white blood cell count, and C-reactive protein) of the patient were all normal and the patient did not have fever, weakness, extremity swelling or hyperemia. As the clinical or laboratory findings of the patient were not typically compatible with osteomyelitis and the pain was relieved with NSAID, osteomyelitis was not considered to be the correct diagnosis. There was no trauma history either. As the margins of the sclerotic bone area was smooth and there was not a malignant periosteal reaction on MRI or on radiograph, malignancy was excluded. Computed tomography (CT) were performed to make a correct diagnosis and it revealed 8 × 6 x 6 mm nidus on cortex of the tibia with well circumscribed lucent region around a central sclerotic dot and cortical thickening adjacent to the nidus. Finally, the lesion was diagnosed as osteoid osteoma according to these clinical and imaging findings (Fig. 3).

A CT guided percutaneous radiofrequency ablation (RFA) treatment was planned. The study was approved by our institution's ethical committee (Number:1879; 23.01.2018). Informed consent

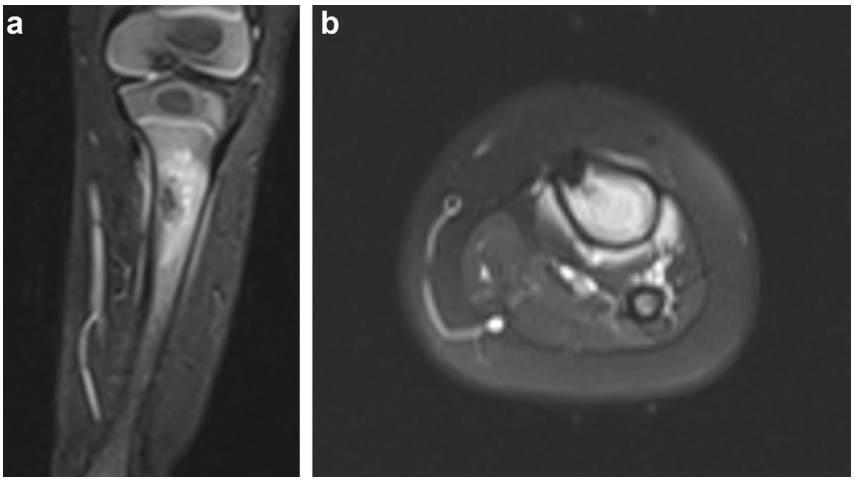

Fig. 2. Pre-procedural coronal (TR: 3520; TE: 32) **(a)** and axial (TR: 3680; TE: 72) **(b)** non-contrast-enhanced T2 weighted STIR-MR images are shown. The nidus is signal void on cortical zone of proximal diaphysis of the left tibia. Adjacent bone marrow and soft tissue has high intensity due to edema. The lesion was firstly misdiagnosed as osteomyelitis.

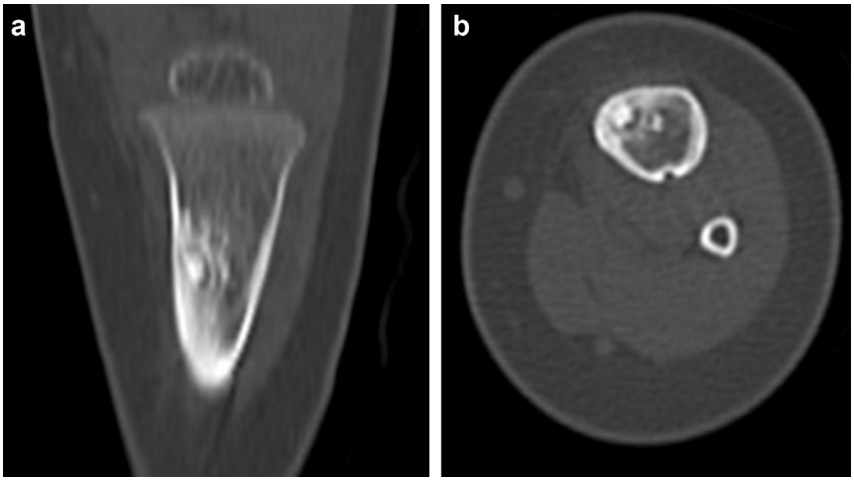

Fig. 3. Coronal **(a)** and axial **(b)** CT images showed an 8 × 6 x 6 mm nidus on cortex of the tibia with well circumscribed lucent region around a central sclerotic dot. There is reactive cortical thickening adjacent to the nidus. Finally, the lesion was diagnosed as osteoid osteoma according to these imaging findings.

for the treatment and anesthesia procedures were obtained from the patient's family before the procedure. RFA (UniBlade, AngioDynamics, USA) was performed under spinal anesthesia with superficial sedation in March 2017 using 0.05 mg/kg of midazolam (Dormicum, Deva) intravenously for sedoanalgesia followed by 0.2 mg/kg of 0.5% heavy marcaine (MarCaine, Astra Zeneca) administration into the subarachnoid space through the L4-L5 intervertebral space in the lateral decubitus position with a 25 Gauge lumbar puncture needle for spinal anesthesia. Since the RFA probe that we used in this procedure was monopolar, grounding pad was placed on the same extremity. After 25 mg/kg intravenous Cephalexin administration in case of prophylaxis, coaxial bone needle (14 G; 6 cm; Apriomed) was inserted into the nidus of OO under CT guidance (Toshiba, Alexion, Japan) by turning the needle with pressure. After reaching to the posterior margin of the nidus with the bone needle, the needle which was used to open the tract was removed. The RFA probe was sent through the bone needle and placed into the middle of the nidus. The outer needle was slightly retracted (till the edge of the cortex) to enable the active end portion of the RFA probe freely interact with the nidus (Fig. 4). Since the length of the tissue area ablated around the active tip of RFA probe was one centimeter, cold application with ice around the needle was performed onto the skin during the process to prevent skin burns. The temperature of the RFA probe was increased to a maximum of 80 °C within 5 min. At the end of the procedure, the intra nidus temperature of the probe was checked whether it was higher than 60 °C or not.

The patient stayed in hospital (in orthopedics department) for one day period for pain relief and potential complication management. He was re-assessed for pain relief and complications on the next day and one week after the procedure in interventional radiology department. After RFA therapy, radiographs was recommended for follow-up. On the radiographs, we compared cortical thickness degree and the size of the sclerotic area where the nidus was placed. Clinical and technical success and the safety of the treatment were evaluated by assessing the clinical symptoms and comparing the pre-postprocedural follow-up images. The ablation procedure including sterilization, spinal anesthesia and sedoanalgesia, took only 40 min.

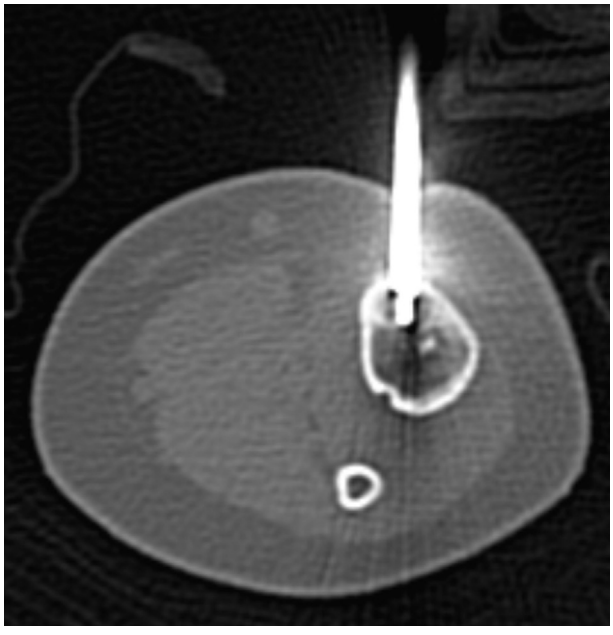

Fig. 4. On axial CT image, coaxial bone needle and the RFA electrode were seen. The tip of the RFA electrode was inserted into the nidus.

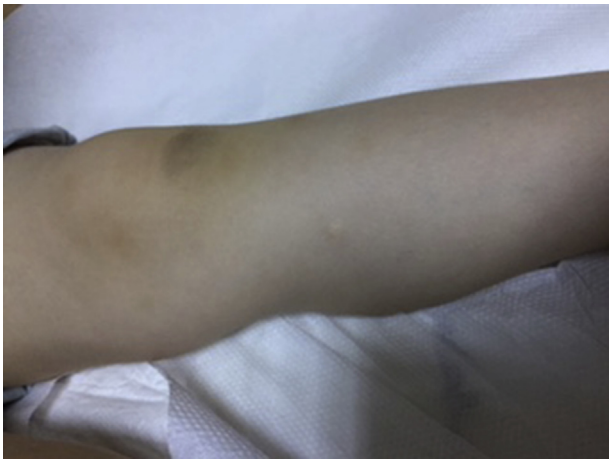

Fig. 5. A photo of the extremity at 16th month follow-up time. After the procedure there was no skin burn on the intervened site. Sixteen months after the ablation, the patient still did not show signs of pain.

No intra or post-procedural complication and no skin burn occurred (Fig. 5). In post-treatment assessments, mother of the patient revealed that, the patient had an immediate pain relief within 24 h that the boy could bear weight on his leg and did not wake up with crying in midnight. Four follow-up radiographs were performed on the 3th, 5th, 12th and 16th months respectively (Fig. 6). There was a slight decrease in the sclerotic area's size on follow-up radiographs. A targeted and a low dose follow-up CT examination was performed under radiologist's supervision on 12th month to check the intervened area more detailed. CT imaging better revealed the healing of the nidus (Fig. 7). MRI examination was not planned due to absence of any clinical or radiological sign for recurrence in the final follow-up examination.

Discussion

OO is very rare in child population who is younger than 5 years-old and diagnosis can be challenging at times. There are a few studies about RFA treatment for OO in children who are younger than 5 years of age.[6,7] In our case, the patient had the symptoms since he was 7 month-old and he had the RFA treatment when he was 13 month-old. Our patient is one of the youngest children to have been treated with RFA.

The diagnosis can be challenging and time-taking because of non-specific symptoms, especially in these age group. Several studies have described the difficulty in diagnosis of OO in young children.[9,10] Pain is the most common symptom and the most important reason for treatment.[6–12] OO can be misdiagnosed as osteomyelitis according to MRI features.[12] CTI is the most preferred imaging method because of its high resolution in demonstrating the nidus.[12,16] In our case, the most prominent symptoms were pain (which is responding to NSAIDs) in nights and refusing to bear weight. Since the clinical symptoms were specific and the CTI revealed a nidus like lesion with peripheral sclerosis in our patient, treatment was planned according to these findings.

For treatment of OO, medication may be inadequate to relief the pain. Although surgical resection of the OO nidus is a curative treatment option, it has disadvantages because of the difficulty in locating the nidus during operation and also long hospitalization. In weight bearing bones, fractures may occur due to large bone resections after surgery.[13] CT guided RFA treatment is a minimally invasive outpatient procedure and has rapid clinical response. This makes it more advantageous.[13,17]

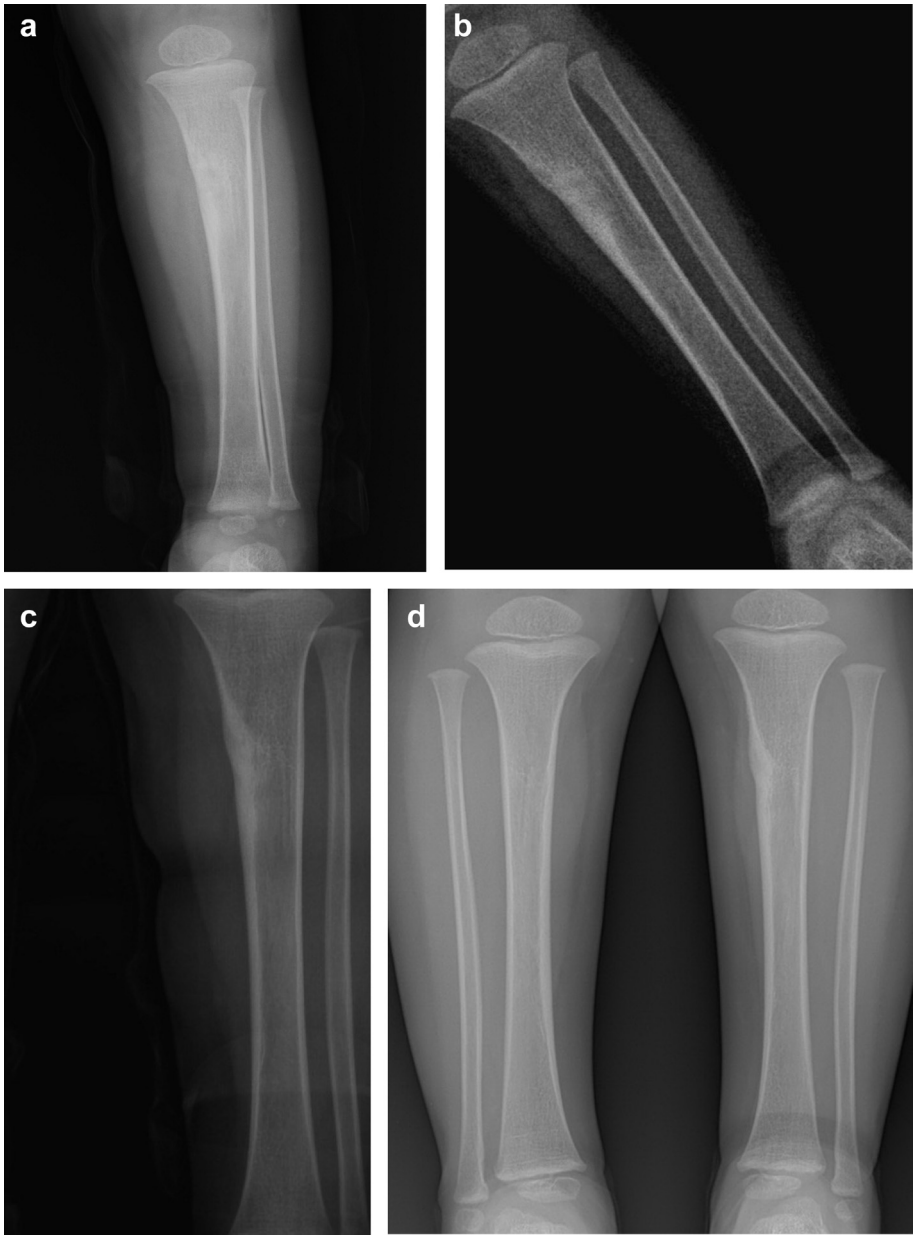

Fig. 6. Healing of the nidus and slow regression of the cortical sclerosis were seen on 3th **(a)**, 5th **(b)** 12th **(c)** and 16th months' **(d)** follow-up radiographs respectively. As the physeal plate was far away enough from nidus for a thermal injury, a comparative radiograph of both tibiae at the time of last follow-up **(d)** proves that there is no limb length discrepancy.

There can be a hesitation about RFA in preschool children because of the small body mass and high complication risks because of close proximity of lesions to neurovascular structures.[15] We can overcome damaging the neurovascular structures through ultrasound/Doppler-ultrasound guidance while inserting the needle and preventing a skin burn through cold application on the skin around the needle during RFA. In our patient, the lesion was 20 mm far away from the growth plate and 14 mm from the skin. The temperature in the growth plate expected to be lower than 39° in Celsius for an 8 mm cortical localized lesion according to Greenberg et al.'s study.[18] The lesion was far enough from the epiphyseal zone and we were not cautious about any bone growth problem in our patient. Although the risk for skin burn was high in our patient, no burn injury occurred owing to cold ice application on the skin around the needle during the procedure.

Time duration of the RFA procedure and total immobilization of the extremity during the treatment is important in our opinion. Patient and extremity motion can interrupt the RFA procedure. Spinal anesthesia or peripheral neural block can shorten the time duration of the treatment procedure and provides using small amounts of sedoanalgesia drugs, decreased complication risks due to prolonged sedoanalgesia. Owing to its early motor recovery and efficiency, spinal anesthesia can be a preferred technique for RFA in the pediatric population.[19] Thus, we performed the RFA procedure under spinal anesthesia and sedoanalgesia; the procedure took approximately 40 min from beginning to the end.

As early response to RFA procedure, pain relief expected to occur in first week after the procedure.[13,14] In our patient pain relief was occurred in 24 h, the boy could bear weight on his leg on the next day and he began to walk one month after the RFA. The clinical success of

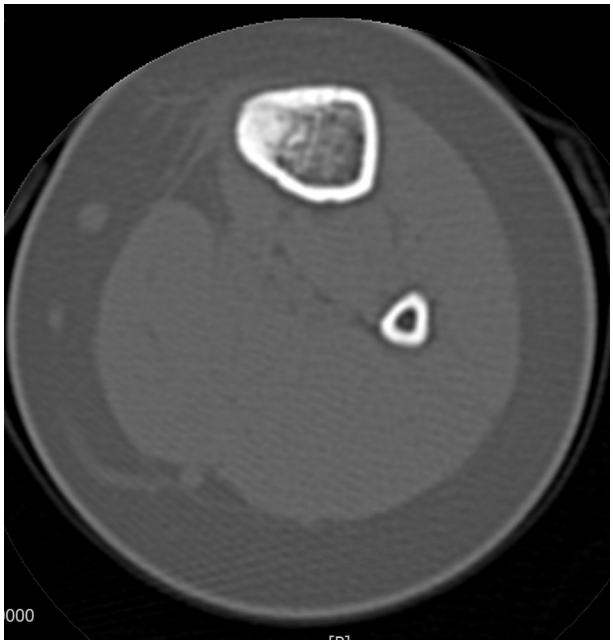

Fig. 7. A targeted and low dose CT examination was performed on 12th month's follow-up. CT imaging revealed sclerotic healing of the nidus and a slow regression of the adjacent cortical thickness.

RFA was confirmed by follow-up radiographs on 16th month and follow-up CT findings which revealed healing of the lesion on 12th month. Since this 13 month-old patient was the youngest one in our patients, we preferred to have follow-up radiologic images more closely. In our experience, it is not necessary to have close follow-up radiographs or CT examinations if clinical response after RFA is good and if there is no complication or complaints. As it is recommended in previous studies, we think a follow-up imaging (radiograph is preferred) on 3th, 6th and 12th months will be enough for radiologic assessment of response to therapy.[20,21]

Especially when considering the limited patient numbers with OO at this age group, our case is interesting. Comparing the cases reported in the literature, our case had a successful result after first percutaneous RFA procedure, no recurrence occurred in contrast to Virayavanich et al.'s case,[6] and the patient had not a surgical excision before RFA procedure in contrast to Ekström et al.'s case.[7] We wanted to mention both about OO in the differential diagnosis when a patient of this age with these symptoms is presented and also efficiency of RFA treatment in this age group.

Conclusion

OO is very rare in children under 5 years of age and the diagnosis may be challenging. OO must be kept in mind, as a differential diagnosis, even in children who are younger than one year of age. CT guided percutaneous RFA is an effective and a minimally invasive method in the treatment of osteoid osteoma which can also be safely used in an even one-year old child, as in our case. Although it has not been observed during 16-months of follow-up period in our case, close follow-up following RFA is recommended against the possibility of any recurrence.

Conflict of interest disclosure

The authors declared no conflicts of interest.

References

1. Munk PL, Ryan AG. *Teaching Atlas of Musculoskeletal Imaging*. Thieme; 2008: 348–352.
2. Greenspan A. Benign bone-forming lesions: osteoma, osteoid osteoma, and osteoblastoma. Clinical, imaging, pathologic, and differential considerations. *Skeletal Radiol*. 1993 Oct;22(7):485–500. https://doi.org/10.1007/BF00209095. PMID: 8272884.
3. Frassica FJ, Waltrip RL, Sponseller PD, et al. Clinicopathologic features and treatment of osteoid osteoma and osteoblastoma in children and adolescents. *Orthop Clin North Am*. 1996 Jul;27(3):559–574. PMID: 8649737.
4. Chai JW, Hong SH, Choi JY, et al. Radiologic diagnosis of osteoid osteoma: from simple to challenging findings. *Radiographics*. 2010 May;30(3):737–749. https://doi.org/10.1148/rg.303095120. PMID: 20462991.
5. Cohen MD, Harrington TM, Ginsburg WW. Osteoid osteoma: 95 cases and a review of the literature. *Semin Arthritis Rheum*. 1983 Feb;12(3):265–281. https://doi.org/10.1016/0049-0172(83)90010-0. PMID: 6603021.
6. Virayavanich W, Singh R, O'Donnell RJ, et al. Osteoid osteoma of the femur in a 7-month-old infant treated with radiofrequency ablation. *Skeletal Radiol*. 2010 Nov;39(11):1145–1149. https://doi.org/10.1007/s00256-010-1014-1. PMID: 20694724.
7. Ekström W, Söderlund V, Brosjö O. Osteoid osteoma in a 1-year-old boy—a case report. *Acta Orthop*. 2006 Aug;77(4):686–688. https://doi.org/10.1080/17453670610012809. PMID: 16929450.
8. Haberman E, Stern R. Osteoid osteoma of the tibia in an eight-month-old boy. *J Bone Joint Surg (Am)*. 1974 Apr;56(3):633–636. https://doi.org/10.2106/00004623-197456030-00025. PMID: 482252.
9. Kaweblum M, Lehman WB, Bash J, Grant AD, Strongwater A. Diagnosis of osteoid osteoma in the child. *Orthop Rev*. 1993 Dec;22(12):1305–1313. PMID: 8127616.
10. Kaweblum M, Lehman WB, Bash J, Strongwater A, Grant AD. Osteoid osteoma under the age of five years. The difficulty of diagnosis. *Clin Orthop Relat Res*. 1993 Nov;(296):218–224. https://doi.org/10.1097/00003086-199311000-00037. PMID: 8222430.
11. Thiagarajan P, Camina P, Das De S, Bose K. Osteoid osteoma in a three-year-old child—a case report. *Ann Acad Med Singapore*. 1996 Sep;25(5):769–770. PMID: 8924027.
12. Bhat I, Zerin JM, Bloom DA, Mooney 3rd JF. Unusual presentation of osteoid osteoma mimicking osteomyelitis in a 27-month-old child. *Pediatr Radiol*. 2003 July;33(6):425–428. https://doi.org/10.1007/s00247-003-0916-7.
13. Rosenthal DI, Hornicek FJ, Wolfe MW, et al. Percutaneous radiofrequency coagulation of osteoid osteoma compared with operative treatment. *J Bone Joint Surg Am*. 1998 Jun;80(6):815–821. https://doi.org/10.2106/00004623-199806000-00005. PMID: 9655099.
14. Donkol RH, Al-Nammi A, Moghazi K. Efficacy of percutaneous radiofrequency ablation of osteoid osteoma in children. *Pediatr Radiol*. 2008 Feb;38(2): 180–185. https://doi.org/10.1007/s00247-007-0690-z. PMID:18040677.
15. Kuyumcu G, Sundaram M, Schils JP, et al. Osteoid osteoma of the hand and foot in children successfully treated with radiofrequency neurotomy probes. *Skeletal Radiol*. 2017 July;46(11):1561–1565. https://doi.org/10.1007/s00256-017-2702-x. PMID: 28689337.
16. Motamedi D, Learch TJ, Ishimitsu DN, et al. Thermal ablation of osteoid osteoma: overview and step-by-step guide. *Radiographics*. 2009 Nov;29(7): 2127–2141. https://doi.org/10.1148/rg.297095081. PMID: 19926767.
17. Gilliaux O, de Wispelaere JF, Charlier H, Bodart E. Osteoid osteoma in children: 5 cases treated with electrocoagulation. *Arch Pediatr*. 2012 Nov;19(11):1177–1181. https://doi.org/10.1016/j.arcped.2012.08.019. PMID: 23037581, Epub 2012 Oct 1.
18. Greenberg A, Berenstein WT, Sosna J, Applbaum J, Peyser A. The distribution of heat in bone during radiofrequency ablation of an ex vivo bovine model of osteoid osteoma. *Bone Joint J*. 2014 May;96-B(5):677–683. https://doi.org/10.1302/0301-620X.96B5.32822. PMID: 24788505.
19. Verma D, Naithani U, Gokula C, Harsha. Spinal anesthesia in infants and children: a one-year prospective audit. *Anesth Essays Res*. 2014;8(3):324–329. https://doi.org/10.4103/0259-1162.143124. PMID: 25886329.
20. Rehnitz C, Sprengel SD, Lehner B, et al. CT-guided radiofrequency ablation of osteoid osteoma: correlation of clinical outcome and imaging features. *Diagn Interv Radiol*. 2013 Jul-Aug;19(4):330–339. https://doi.org/10.5152/dir.2013.096. PMID: 23491835.
21. Vanderschueren GM, Taminiau AH, Obermann WR, et al. The healing pattern of osteoid osteomas on computed tomography and magnetic resonance imaging after thermocoagulation. *Skeletal Radiol*. 2007 Sep;36(9):813–821. https://doi.org/10.1007/s00256-007-0319-1. PMID: 17492439.