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Case Report

Research paper: Two cases of multiple osteoid osteomas in young patients treated with cryoablation: Diagnosis, technical efficacy, and follow-up ☆,☆☆

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ABSTRACT

Osteoid osteomas are benign bone tumors characterized by severe localized pain, often challenging the daily lives of young patients. While these tumors are typically solitary, rare cases of multiple osteoid osteomas have been reported. This research paper presents a case series of 2 young patients with multiple osteoid osteomas, highlighting their clinical presentation, diagnostic workup, treatment with cryoablation, and follow-up. The paper emphasizes the effectiveness of cryoablation as a minimally invasive treatment option for these challenging cases.

Introduction

The first case of osteoid osteoma was first described in 1930 by Bergstrand and first characterized as an entity in 1935 from Jaffe [1]. Osteoid osteoma is the third most common benign bone-forming tumor [2], is a central hypervascular nidus of osteoblastic cells surrounded by sclerotic margins, with diameter less than 2 cm and limited growth potential [2]. The pathophysiology of osteoid osteomas is still controversial. It most commonly develops in the long bones of lower extremities but it can arise in every bone of the body. It also has peak inci-

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dence in the second decade of life and a male predominance [3]. In case of osteoid osteoma suspicion an X-ray is performed and shows a round or oval radiolucent nidus surrounded by a fusiform of bone sclerosis. Computed tomography is the imaging technique of choice. The findings are a well-defined area with low attenuation within surrounding sclerotic reactive bone. In magnetic resonance imaging the nidus has low to intermediate signal on T1-weighted images and variable signal intensity in T2-weighted images, which depends on the degree of mineralization in the center of the nidus. The MRI is inferior to CT for the diagnosis of osteoid osteoma but is useful for recognizing adjacent bone marrow abnormalities and also articular abnormalities [4]. The usefulness of Technetium-99-labeled bonescintigraphy, which reflects the increased osteoblastic activity, is significant in order to clarify the precise localization of the lesion in combination with the results of the CT before the treatment. The most common symptom is localized, deep pain which is worsened during the night. In most cases the pain is rapidly improved with nonsteroid anti-inflammatory drugs (NSAIDs) or salicylates. The surgical excision was the only alternative therapy for osteoid osteoma-associated pain which did not respond to conservative treatment. In the last decades there are many minimal invasive alternative therapies which are considered to be the new gold standards [5], such as Radiofrequency Ablation (RFA), Interstitial Laser Ablation (ILA), Microwave Ablation (MWA), Cryoablation and Magnetic resonance-guided focused ultrasound (MRg-FUS).

Osteoid osteomas are benign bone tumors that predominantly affect adolescents and young adults, causing localized pain that is often more severe at night [6]. Typically, these tumors are solitary, but cases of multiple osteoid osteomas are exceedingly rare [9–12]. The management of multiple osteoid osteomas presents unique challenges in terms of diagnosis and treatment. This case series aims to shed light on the clinical presentation, diagnostic strategies, treatment with cryoablation, and outcomes in 2 young patients with multiple osteoid osteomas.

Case presentations

Case 1

Patient Information: A 17-year-old male patient presented with right shoulder pain persisting for several months. He described severe pain at night, which interfered with her sleep and daily activities.

Clinical findings

Physical examination revealed tenderness over the right shoulder. The range of motion was not affected.

Pain Visual Analogue Score (VAS) score was very high: 10/10.

Imaging findings

Radiographs revealed 3 radiolucent nidus within the surgical neck and diaphysis of the right humerus bone . CT, Spect and MRI confirmed the presence of multiple osteoid osteomas (Figs. 1, 2). On MRI the lesions had a central nidus with low signal intensity on T1-weighted images and surrounding high signal intensity on T2-weighted images (Fig. 3). There was associated periosteal reaction and marrow edema in the adjacent bone (Fig. 4).

Differential diagnoses (DDx)

Some pathologies may mimic osteoid osteomas due to there being similar imaging findings such as:

Osteomyelitis/intraosseous abscess

Fracture/stress reaction

Fig. 1 – (A) CT thick slice showing 3 radiolucent lesions in surgical neck and shaft of right humerus bone. (B) MRI T2W image showing same lesions with intermediate-low signal with sclerotic margins. (C) MRI STIR sequence showing high signal in brachial bone depicting the bone marrow edema.

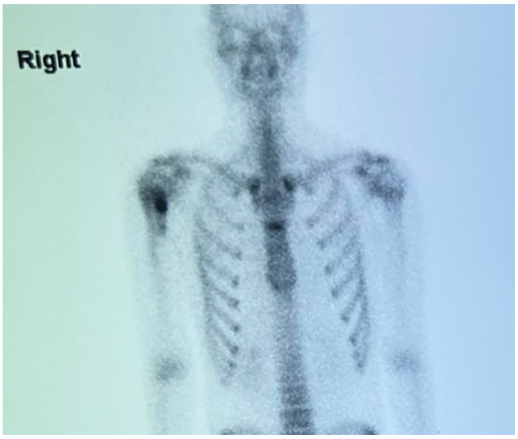

Fig. 2 – A Spect showing increased blood pool and increased 99mTc-Methylene diphosphonate.

Osteoblastoma
Glomus tumor
Crystal deposition disease
Chondroblastoma

In our case the imaging findings according to Diagnostic criteria according to the WHO classification of soft tissue and bone tumors (fifth edition) [7] were typical for one of the following pathologies:

1. **Osteoid Osteoma:** The characteristic imaging findings with a central nidus and surrounding edema are highly suggestive of osteoid osteoma [8].
2. **Osteoblastoma:** Similar to the first case, osteoblastoma can present with imaging findings that resemble osteoid osteoma but is typically larger. than 2.0 cm; less painful; fewer inflammatory changes and reactive sclerosis; smaller response to salicylates; grow progressively; malignant potential and may be associated with other tumors.

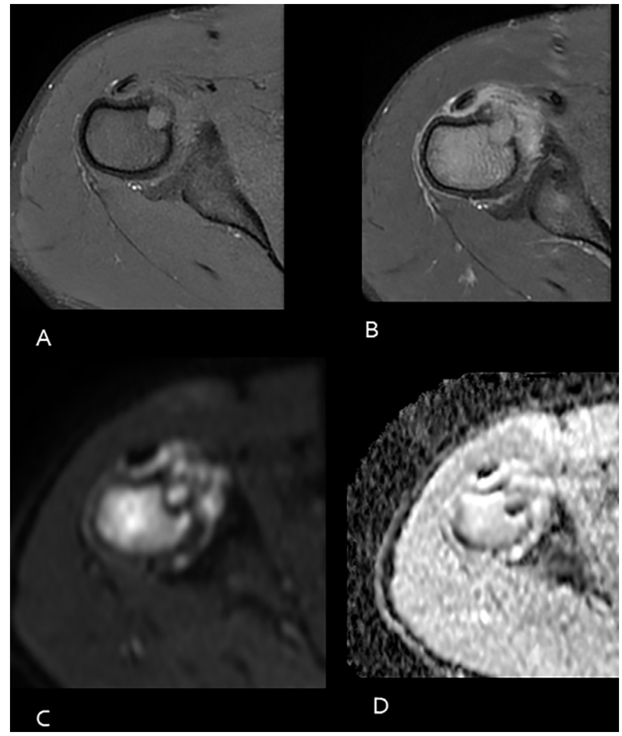

Fig. 4 – (A) MRI axial T1 fatsat weighted image showing a small mass in the surgical neck of the right humerous bone, (B) MRI axial T1w fatsat image with in contrast showing the periosteal reaction with no enchantment of the mass, (C) DWI sequence, and (D) ADC map didn't reveal any restriction of water molecules in mass.

Osteoblastosis (OBLT) is a recently described unusual diagnosis, which must be considered when multiple well-defined lytic lesions are found on imaging studies, especially if the lesions are intracortical, multifocal, and involve the diaphysis/

Fig. 3 – (A and B) CT image in soft (A) and bone (B) window (C). MRI T2 weighted axial image showing the mass in the surgical neck of the humerous bone.

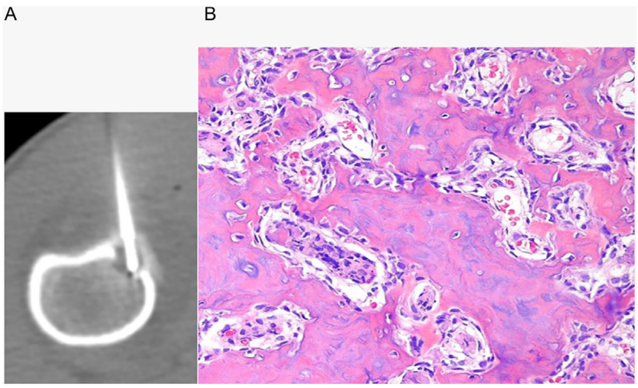

Fig. 5 – (A) Percutaneous bone biopsy from the nidus. (B) Pathology findings supporting the diagnosis of osteoid osteoma: Sclerotic trabeculae of woven bone with osteoblastic rimming, fibrovascular stroma and scattered multinucleated giant cells.

metaphysis of the long bones [6]. Although the term osteoblastomatosis is not yet included in the WHO classification [7], it has been used in the literature since 2007 [13]. This entity is also referred to as multifocal OB, so its radiographic features are the same as insulated OB.

In our case the lesions were <2 cm and multifocal there are not enough cases in the literature and there has never been a large series that suggest that multifocal osteoid osteomas are a separate disease or if there is a correlation with osteoblastosis.

Diagnostic workup

CT-guided core biopsies provided histological confirmation of multiple osteoid osteomas (Fig. 5).

Treatment and Follow-Up

For the treatment of the 3 osteoid osteomas— one larger within the surgical neck of the humerus bone and 2 smaller ones in the shaft—2 distinct cryoablation procedures were conducted under local anesthesia (Fig. 6). First there was an attempt to treat the lager osteoid osteoma with the larger periosteal reaction with the purpose of assessing the clinical response and evaluate the need for treating the smaller lesions. For the larger osteoid osteoma, a single cryoprobe was employed, guided by imaging to accurately target and undergo 2 cryocycles of freezing-thawing. The cryoablation process involved meticulous monitoring of temperatures to ensure precise lesion targeting while minimizing damage to surrounding tissues (Fig. 7).

Immediate and 1 week after the procedure there was a small reduction of the pain level of the patient (VAS score before the procedure was 10 after the procedure it was lower 6-7) and so there was a need for treating the other 2 lesions.

Simultaneously, the 2 smaller osteoid osteomas in the diaphysis were treated with 2 cryoprobes, incorporating hydrodissection to safeguard the brachial nnerve. The cryoablation process involved meticulous monitoring of temperatures to ensure precise lesion targeting while minimizing damage to surrounding tissues.

Postprocedure, the patient was monitored for vital signs, local temperature, and neurological status, with follow-up assessments scheduled to confirm lesion resolution and assess nerve function, providing an effective and minimally invasive therapeutic approach for multiple osteoid osteomas within the humerus bone.

The patient experienced immediate pain relief and complete resolution on follow-up imaging (Fig. 8). Long-term follow-up at 3 years showed no recurrence or residual pain.

Case 2

Patient Information: A 16-year-old male presented with severe and recurrent pain in the right tibia, radiating to the thigh, which had been ongoing for over a year. He was an athlete and had no other medical history. The patient reported that the pain worsened every night and he was relieved by the intake of paracetamol and NSAIDs;, which provided almost complete relief but was only temporary as the pain returned between doses.

Clinical Findings: Upon examination, he had no skin changes, swelling, or effusion of the knee. He had full active range of motion and was tender to palpation on the posterior medial femoral condyle.

Pain Visual Analogue Score (VAS) score was very high: 9/10.

Imaging findings: The radiographs were nondiagnostic, but the patient already had advanced imaging with an MRI prior to referral. MRI of the right hip revealed 2 well-defined lesions with a central nidus of low signal intensity on T1-weighted images and a surrounding rim of high signal intensity on T2-weighted images. There was associated periosteal reaction and marrow edema in the adjacent bone. Two osteoid osteomas (multicentric) were the suspected diagnosis, and a CT without contrast was ordered to confirm the diagnosis. (Fig. 9) presents the preoperative CT scan images of the knee.

Differential diagnoses (DDx):

Same as case 1 see above*

Diagnostic workup: Computed tomography (CT) guided core biopsy was performed for histological confirmation. A diagnosis of multiple osteoid osteomas was established.

Pathology findings supporting the diagnosis of osteoid osteoma: Sclerotic trabeculae of woven bone with osteoblastic rimming, fibrovascular stroma and scattered multinucleated giant cells (Fig. 10).

Treatment: The patient underwent percutaneous cryoablation targeting both osteoid osteomas simultaneously.

CT guided placement of 2 cryoprobes at the same procedure with extraossseous position (Fig. 11) under local anasthesia. Two cryocycles of freezing-thawing were performed with active warming of the skin for protection.

There were no immediate complications.

Immediate pain relief was observed.

Pain (VAS) score after the procedure was 2 /10 and 1 week after the procedure was at 0/10 with complete resolution of the symptoms.

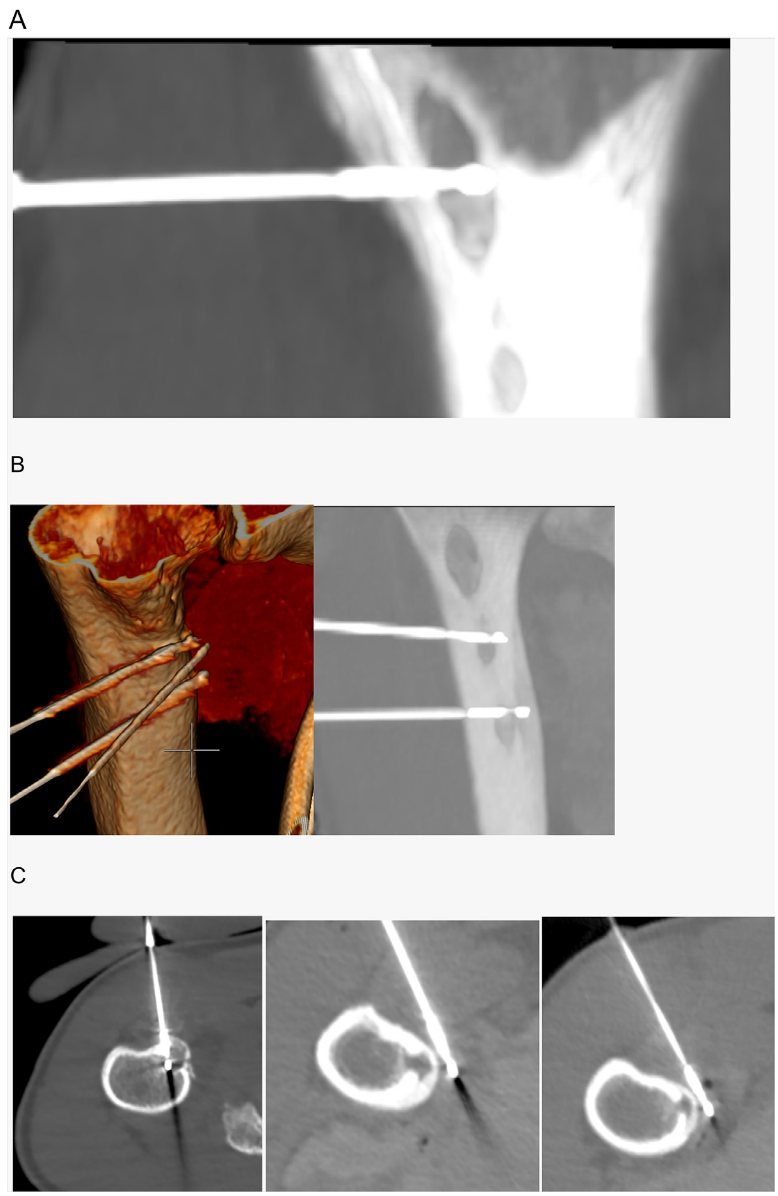

Fig. 6 – (A) CT thick slice showing the cryoprobe placed intraosseous in the lesion in the surgical neck of the right humerus bone. (B) In 3D rendering and in thick slice CT showing the placement of the cryoprobes in exrtaossous position in the diaphyseal masses of the right humerous bone. (C) Showing the 3 cryoprobes two of them placed in extraosseous position and one of them intraosseous.

Follow-up: Follow-up imaging confirmed complete resolution. Long-term follow-up at 2 years revealed no recurrence or residual pain (Fig. 12).

Discussion

The presented cases highlight the diagnostic and therapeutic challenges associated with multiple osteoid osteomas.

In a review of the English language medical literature, we found only 8 patients in whom more than a single focus of OO was present [9–12].

In both cases, the combination of CT and MRI played a crucial role in the diagnostic workup. The characteristic imaging findings of a central nidus with low signal intensity on T1-weighted images and surrounding high signal intensity on T2-weighted images, along with associated periosteal reaction and marrow edema, were highly suggestive of osteoid osteoma.

The main differential diagnosis considered in both cases was osteoblastoma due to the similarities in imaging findings. However, osteoblastomas are typically larger and may exhibit more aggressive features, such as greater extent of bone destruction and soft tissue involvement, which were not observed in these cases.

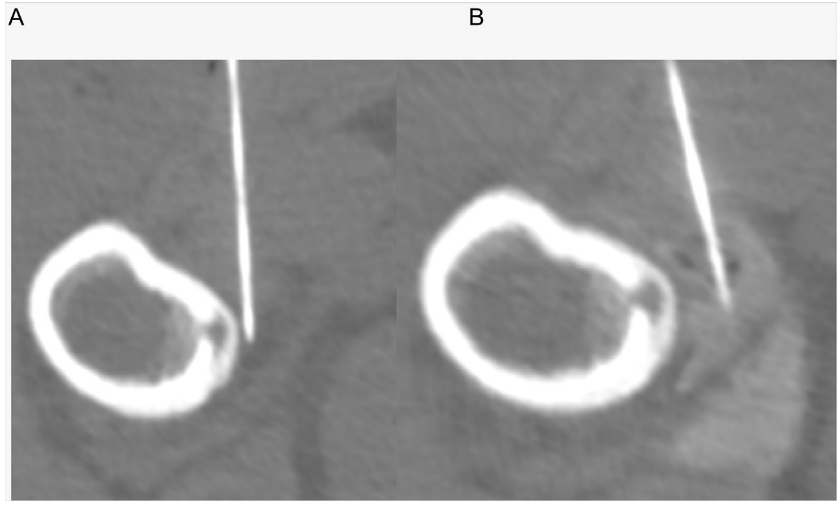

Fig. 7 – (A) Placement of a thermocouple next to the axillary nerve. (B) Hydrodissection of the axillary nerve for temperature monitoring, protection, and active warming.

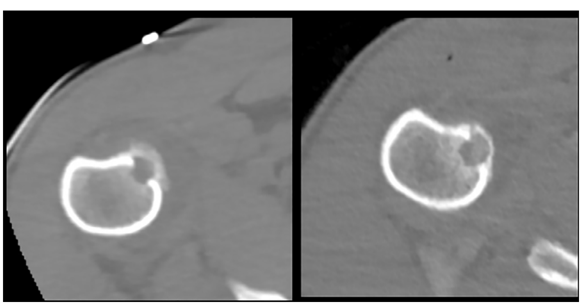

Fig. 8 – One month after cryoablation procedure of osteoid osteoma of humerus bone reduction of the periosteal reaction.

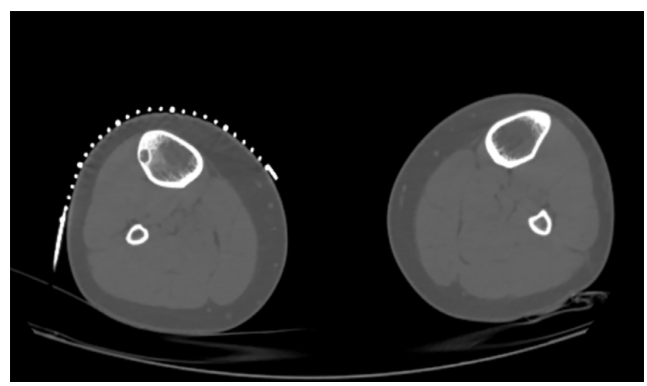

Fig. 9 – Axial CT image of a right tibia osteoid osteoma.

The use of CT allowed for a noninvasive evaluation of the lesions' characteristics, aiding in the accurate diagnosis and subsequent treatment planning with cryoablation.

In both cases, cryoablation was chosen as a minimally invasive treatment option due to its precise targeting of multiple lesions and reduced morbidity compared to surgical excision [14,15].

Cryoablation proved highly effective, providing immediate pain relief and complete resolution of the osteoid osteomas

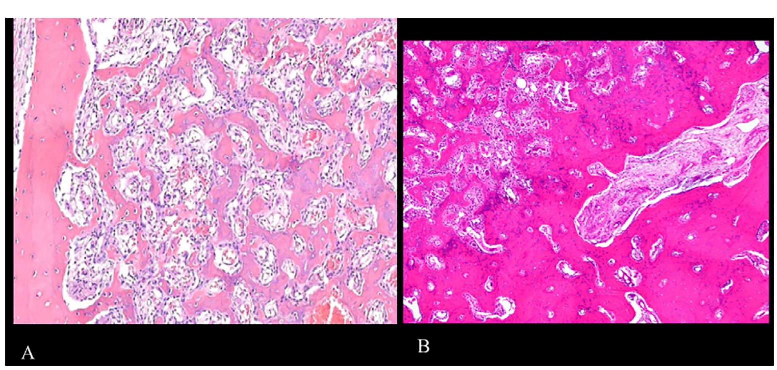

Fig. 10 – (A) Nidus: Haphazard trabeculae of woven bone with prominent osteoblastic rimming well defined borders. (B) Surrounding bone: Thickened trabeculae of bone with adjacent loose fibrovascular stroma.

Fig. 11 – (A) Placement of the cryoprobe at ana extraosseous position parallel to the cortex. (B) The hypodense ice covering the whole lesion.

[15]. Long-term follow-up demonstrated sustained pain relief and the absence of recurrence in both cases, underscoring the durability of this treatment modality.

Conclusion

Multiple osteoid osteomas are rare but challenging clinical entities in young patients. This case series illustrates the successful use of percutaneous cryoablation for the treatment of multiple osteoid osteomas, resulting in immediate pain relief and long-term resolution of the tumors. Cryoablation is a valuable minimally invasive option for patients with multiple osteoid osteomas, offering the potential for excellent outcomes and improved quality of life.

Patient consent

We are confirming that written, informed consent for publication of the cases was obtained from both patients.

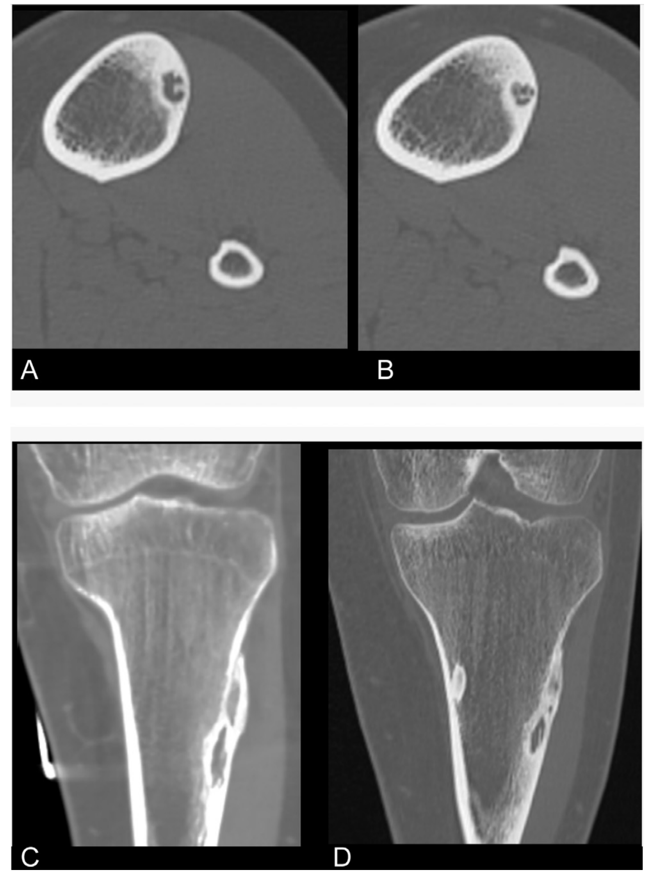

Fig. 12 – One month after cryoablation procedure of osteoid osteoma of tibial bone showing the sclerotic changes (A) before the cryoablation (B) after the cryoablation Coronal images of the same lesions before (C) and after the cryoablation (D).

REFERENCES

[1] Jaffe HL. Osteoid-osteoma. Arch Surg 1935;31(5):709. doi:10.1001/ARCHSURG.1935.01180170034003.
[2] Lee EH, Shafi M, Hui JH. Osteoid osteoma: a current review. J Pediatr Orthop 2006;26(5):695–700. doi:10.1097/01.bpo.0000233807.80046.7c.
[3] Resnick D, Greenway GD. Tumors and tumor-like lesions of bone: imaging and pathology of specific lesions. Chapter 74 Bone and Joint Imaging. 2nd Ed. USA: W.B Saunders Company; 1996. 4 Zhang και Rosenberg.
[4] Chang C, Garner H, Ahlawat S, Amini B, Bucknor MD, Flug JA, et al. Society of Skeletal Radiology- White Paper. Guidelines for the diagnostic management of incidental solitary bone lesions on CT and MRI in adults: bone reporting and data system (Bone-RADS). Skeletal Radiol 2022;51(9):1743–64. doi:10.1007/s00256-022-04022.
[5] Parmeggiani A, Martella C, Ceccarelli L, Miceli M, Spinnato P, Facchini G. Osteoid osteoma: which is the best mininvasive treatment option? Eur J Orthop Surg Traumatol 2021;31(8):1611–24. doi:10.1007/S00590-021-02946-W.
[6] Quílez-Caballero E, Martel-Villagran J, Bueno-Horcajadas ÁL, López-Brasal L, Díaz-Candamio MJ. Osteoblastomatosis: an unusual diagnosis and treatment. Skeletal Radiol 2018;47(8):1183–9. doi:10.1007/s00256-018-2891-y.
[7] W. H. O. Classification WHO Classification of Tumours Editorial Board, Who Classification of Tumours Editorial. Soft Tissue Bone Tumours. (2020) ISBN: 9789283245025 - Google Books.

[8] Kransdorf MJ, Stull MA, Gilkey FW, Moser RP. Osteoid osteoma. Radiographics 1991;11(4):671–96.
[9] Sampath Kumar V, Khan SA, Palaniswamy A, Rastogi S. Multifocal osteoid osteoma of tibia. BMJ Case Rep 2013 bcr2013201712. doi:10.1136/bcr-2013-201712.
[10] Sreenivas T, Menon J, Nataraj AR. Synchronous symmetrical atypical osteoid osteoma of tibia: a case report. Eur J Orthop Surg Traumatol 2012;22:251–4.
[11] Arslan A, Sarlak AY, Tosun B. Synchronous multicentric osteoid osteoma with associated fibrous cortical defect. Orthopedics 2008;31:95.
[12] Alcalay M, Clarac JP, Bontoux D. Double osteoid osteoma in adjacent carpal bones. J Bone Jt Surg Am 1982;64:779–80.
[13] Zon Filippi R, Swee RG, Krishnan Unni K. Epithelioid multinodular osteoblastoma: a clinicopathologic analysis of 26 cases. Am J Surg Pathol 2007;31(8):1265–8.
[14] Santiago E, Pauly V, Brun G, Guenoun D, Champsaur P, Le Corroller T. Percutaneous cryoablation for the treatment of osteoid osteoma in the adult population. Eur Radiol 2018;28(6):2336–44. doi:10.1007/s00330-017-5164-6.
[15] Le Corroller T, Vives T, Mattei JC, Pauly V, Guenoun D, Rochwerger A, Champsaur P. Osteoid osteoma: percutaneous CT-guided cryoablation is a safe, effective, and durable treatment option in adults. Radiology 2022;302(2):392–9. doi:10.1148/radiol.2021211100.