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Diagnostic Challenges of Osteoid Osteoma of the Femoral Neck in an Adolescent: A Case Report

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Case Report Abstract

Non-traumatic hip pain in adolescents presents a significant diagnostic challenge. We report the case of a 19-year-old female patient treated for idiopathic juvenile osteoporosis with zoledronic acid, presenting with chronic inflammatory right hip pain that responded well to NSAIDs, associated with back and buttock pain. The inflammatory markers, including CRP and ESR, were negative. The pelvic radiography was normal, though thoracolumbar spine radiography revealed scoliosis. Consequently, MRI of the sacroiliac joints was performed to search for sacroiliitis but revealed a 5 mm nidus with reactive sclerosis, suggestive of an osteoid osteoma. The natural progression of the patient's pain showed spontaneous regression within one year.

Keywords: Osteoid Osteoma, Case Report, Femoral Neck, Bisphosphonate, Nidus.

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Introduction

Hip pain in children and adolescents presents a diagnostic challenge of varying difficulty, sometimes with rare but treatable causes requiring rigorous diagnostic management. We report the case of an adolescent patient presenting with chronic back pain, buttock pain, and inflammatory right hip pain, where investigations revealed a rare osteoid osteoma.

CASE REPORT

We report the case of J.E., a 19-year-old asthmatic female on inhaled corticosteroids, followed for idiopathic juvenile osteoporosis since 2019, treated with four cycles of zoledronic acid, showing good clinical and densitometric improvement.

The patient was followed for chronic back pain, with standard radiography revealing thoracolumbar scoliosis, diagnosed as idiopathic scoliosis. One year ago, she presented with the onset of inflammatory right buttock pain, exacerbation of her low back pain, and inflammatory right hip pain responsive to NSAIDs. Clinical examination noted pain with sacroiliac maneuvers without spinal syndrome or hip joint limitation.

The inflammatory markers, including CRP and ESR, were negative and pelvic radiography showed no abnormalities. Hip ultrasound revealed a minimal effusion in the right coxofemoral (CF) joint. Given the patient's clinical presentation and history, chronic inflammatory rheumatic disease such spondyloarthritis was first considered. Pelvic MRI to assess the SI joints and CF joint showed no signs of inflammatory sacroiliitis but revealed a condensation image in the anterior femoral neck, suggestive of an osteoid osteoma (OO) with a 5 mm nidus (Figures 1a and 1b). Surgical indication for radiofrequency ablation was made, but the natural progression in our patient showed spontaneous pain regression after one year.





Figure 1: A 5 mm nidus located on the anterior aspect of the femoral neck surrounded by cortical periosteal thickening

DISCUSSION

Osteoid osteoma (OO) was first reported by Jaffe in 1935 in a series of five cases [1]. It is a benign osteogenic tumor characterized by persistent pain, worsening at night, and relieved by NSAIDs and salicylates [2, 3]. It is particularly common in adolescents and young adult males; moreover, 50% of these tumors occur in the second decade of life, being rare before age 5 and after age 35[4].

Positive diagnosis relies on clinical and imaging findings, detecting the typical OO nidus, generally small, measuring up to 1-2 cm, usually surrounded by cortical periosteal thickening [4, 5]. Edeiken's radiographic classification categorizes OO as cortical, medullary, or subperiosteal [4]. Histologically, the nidus comprises an osteoid matrix and trabecular bone surrounded by active osteoblasts, osteoclasts, and vascular structures. The sclerosis surrounding the lesion is a reaction to pressure caused by the highly vascularized nidus, which may be absent or minimal in intra-articular lesions [6].

OO is often located in the diaphysis and metaphysis of long bones, with the femur and tibia affected in over 50% of cases [4]. Femoral neck OO is notable, but due to its proximity to the hip joint, clinical manifestations can be misleading [7], increasing diagnostic error likelihood, with a delay of up to 14 months for diagnosis [3].

CT is the imaging modality of choice, more precise than MRI in detecting the nidus in 63% of cases [8]. However, gadolinium-enhanced dynamic MRI offers slightly higher diagnostic accuracy than CT, though not significantly [9]. MRI remains useful for assessing joint effusion, synovitis, and bone marrow [6]. Diagnosis is challenging in cases with atypical clinical and radiographic features, particularly with minimal or absent reactive sclerosis in intra-articular and subperiosteal OOs [10].

A study by Hao Zeng et al. reported 35 patients with proximal femur OO, with 15 patients (42.9%) initially misdiagnosed with conditions such as synovitis, Perthes disease, osteomyelitis, intra-articular infection, joint tuberculosis, and hip impingement syndrome, with an average delay of 6.3 months from symptom onset to diagnosis [3].

Natural progression may include spontaneous regression over 6-15 years, though NSAIDs use can reduce this period to 2-3 years [4]. Curative treatment primarily involves surgical intervention with complete tumor excision, leading to healing and an exceptional recurrence risk [11]. Biopsy is recommended for percutaneous treatment, especially for atypical lesions, though it may not be diagnostic in about one-third of cases [4, 12].

OO treatment of the femur varies based on anatomical location, local expertise, and available resources [13]. Femoral OO can be treated medically with NSAIDs or salicylates [14]. However, interventional treatments offer definitive cure and overcome long-term NSAID and salicylate side effects. Traditionally, surgical resection, curettage, and scraping of the OO nidus have been described. New minimally invasive techniques with less morbidity and faster percutaneous recoverv include CT-guided radiofrequency ablation, thermal destruction, laser CT-guided percutaneous ablation, drilling, arthroscopic nidus removal [14].

All minimally invasive treatment options have yielded satisfactory results, exceeding 90% success rates, with CT-guided radiofrequency and arthroscopy being potentially the most effective options [13].

Alternative therapeutic options for inaccessible OOs include bisphosphonates, particularly zoledronic acid, which can inhibit in vitro prostaglandin E2 (PGE2) synthesis by osteoblasts, known to increase in OOs.

Besides their anti-prostaglandin effect, bisphosphonates demonstrate anti-angiogenic properties, affecting endothelial and stromal vascular cell proliferation, adhesion, and migration within the lesion [15].

A study involving three patients treated with monthly zoledronic acid for 3-6 months showed efficacy, with OO regression and no recurrence over a 5-year follow-up [15]. In our case, pain reduction might be due to the natural evolution of the OO or zoledronic acid administration for juvenile osteoporosis in our patient.

CONCLUSION

OO is a benign tumor that can sometimes be confused with chronic inflammatory rheumatism. Today, diagnosis is facilitated by advances in medical imaging. Minimally invasive and non-invasive approaches offer increased efficacy with better tolerance. While bisphosphonates' efficacy is not fully demonstrated, future studies with larger samples are desirable.

REFERENCE

- 1. Jaffe, H. L. (1935). "Osteoid-osteoma": a benign osteoblastic tumor composed of osteoid and atypical bone. *Archives of Surgery*, *31*(5), 709–728.
- de Berg, J. C., Pattynama, P. M., Obermann, W. R., Bode, P. J., Vielvoye, G. J., & Taminiau, A. H. (1995). Percutaneous computed-tomographyguided thermocoagulation for osteoid osteomas. *Lancet*, 346(8971), 350-351. doi: 10.1016/s0140-6736(95)92228-8
- 3. Zeng, H., He, H., Tong, X., Wang, Z., Luo, R., & Liu, Q. (2022). Osteoid Osteoma of the Proximal Femur: Pitfalls in Diagnosis and Performance of Open Surgical Resection. *Frontiers in Surgery*, 9, 922317. doi: 10.3389/fsurg.2022.922317
- Carneiro, B. C., Da Cruz, I. A. N., Ormond Filho, A. G., Silva, I. P., Guimarães, J. B., Silva, F. D., Nico, M. A. C., & Stump, X. M. G. R. G. (2021). Osteoid osteoma: the great mimicker. *Insights into Imaging*, 12(1), 32. doi: 10.1186/s13244-021-00978-8
- Kransdorf, M. J., Stull, M. A., Gilkey, F. W., & Moser, R. P. Jr. (1991). Osteoid osteoma. Radiographics, 11(4), 671-696. doi: 10.1148/radiographics.11.4.1887121
- 6. Garg, G., & Malot, R. (2017). Intra-articular Osteoid Osteoma of Femoral Neck Region: A Simplified Treatment Strategy and Review of Literature. *Journal of Orthopaedic Case Reports*, 7(6), 36-40. doi: 10.13107/jocr.2250-0685.940
- 7. Ly, J. A., Coleman, E. M., Cohen, G. S., & Kropf, E. J. (2016). Unrecognized osteoid osteoma of the

- proximal femur with associated cam impingement. Journal of Hip Preservation Surgery, 3(3), 236-237. doi: 10.1093/jhps/hnw002
- Tepelenis, K., Skandalakis, G. P., Papathanakos, G., Kefala, M. A., Kitsouli, A., Barbouti, A., Tepelenis, N., Varvarousis, D., Vlachos, K., Kanavaros, P., & Kitsoulis, P. (2021). Osteoid Osteoma: An Updated Review of Epidemiology, Pathogenesis, Clinical Presentation, Radiological Features, and Treatment Option. *In Vivo*, 35(4), 1929-1938. doi: 10.21873/invivo.12459
- Bourgault, C., Vervoort, T., Szymanski, C., Chastanet, P., & Maynou, C. (2014). Percutaneous CT-guided radiofrequency thermocoagulation in the treatment of osteoid osteoma: an 87 patient series.
 Orthopaedics & Traumatology: Surgery & Research, 100(3), 323-327. doi: 10.1016/j.otsr.2014.02.001
- 10. Ciftdemir, M., Tuncel, S. A., & Usta, U. (2015). Atypical osteoid osteomas. *European Journal of Orthopaedic Surgery & Traumatology*, 25(1), 17-27. doi: 10.1007/s00590-013-1291-1
- 11. Wakrim, S., Siwane, A., Traore, O., Lezar, S., & Essodegui, F. (2016). L'ostéome ostéoïde: à propos d'un cas [Osteoid osteoma: about a case]. *Pan African Medical Journal*, 24, 132. doi: 10.11604/pamj.2016.24.132.9703
- Becce, F., Theumann, N., Rochette, A., Larousserie, F., Campagna, R., Cherix, S., Guillou, L., Mouhsine, E., Anract, P., Drapé, J. L., & Feydy, A. (2010). Osteoid osteoma and osteoid osteoma-mimicking lesions: biopsy findings, distinctive MDCT features and treatment by radiofrequency ablation. *European Radiology*, 20(10), 2439-2446. doi: 10.1007/s00330-010-1811-x
- Efthymiadis, A., Tsikopoulos, K., Uddin, F., Kitridis, D., Edwards, N., Sidiropoulos, K., & Lavalette, D. (2022). Which is the optimal minimally invasive treatment for osteoid osteoma of the hip? A systematic review and proportional meta-analysis. *Journal of Orthopaedic Science*, 27(2), 456-462. doi: 10.1016/j.jos.2020.12.026
- Noordin, S., Allana, S., Hilal, K., Nadeem, N., Lakdawala, R., Sadruddin, A., & Uddin, N. (2018). Osteoid osteoma: Contemporary management. Orthopedic Reviews (Pavia), 10(3), 7496. doi: 10.4081/or.2018.7496
- 15. Larid, G., Valayer, S., Jacquier, C., Lafforgue, P., Laredo, J. D., & Pham, T. (2023). Bisphosphonate treatment in inaccessible osteoid osteomas: An alternative therapeutic approach. *Joint Bone Spine*, *90*(6), 105589. doi: 10.1016/j.jbspin.2023.105589