CASE REPORT

A recurring osteoblastoma that initially presents as a typical osteoid osteoma: A case report

Un ostéoblastome récurrent se présentant initialement comme un ostéoïde ostéome typique: A propos d'un cas

Mohamed Achraf Ferjani[1], Mohamed Taghouti[1], Leila Abid[2], Mourad Jenzri[1], Khaled Kamoun[1]

1. Elmanar University, Faculty of Medicine of Tunis, 1007, Kassab Orthopedic Institute, Pediatric Orthopedic Department, 2010, Tunis, Tunisia.
2. Elmanar University, Faculty of Medicine of Tunis, 1007, Kassab Orthopedic Institute, Anatomopathology Department, 2010, Tunis, Tunisia.

Abstract

Introduction: Osteoid osteoma and osteoblastoma are benign bone tumors with similar histologic features, often distinguished by size and clinical behavior. Their relationship remains a topic of debate.

Observation: An 8-year-old boy presented with a femoral diaphyseal lesion initially diagnosed as osteoid osteoma based on resection biopsy. However, within six months, the boy experienced increased pain and rapid growth, with subsequent biopsy revealing aggressive osteoblastoma. This suggests the initial lesion may have been an early-stage osteoblastoma.

Conclusion: This case challenges the concept of osteoid osteoma transforming into osteoblastoma. While histologically similar, these tumors should be considered distinct entities, and size alone may not be a reliable differentiating factor. Careful clinical and pathological correlation, with attention to growth rate, is crucial for accurate diagnosis and management.

Key words: Biopsy, Bone Neoplasms, Child, Diagnosis, Differential, Recurrence

Résumé

Introduction: L'ostéome ostéoïde et l'ostéoblastome sont des tumeurs osseuses bénignes présentant des caractéristiques histologiques similaires, souvent distinguées par leur taille et leur comportement clinique. Leur relation reste un sujet de débat.

Observation: Un garçon de 8 ans a présenté une lésion diaphysaire fémorale initialement diagnostiquée comme un ostéome ostéoïde sur la base d'une biopsie-exérèse. Cependant, en l'espace de six mois, le garçon a présenté une douleur accrue et une croissance rapide de la tumeur. Une biopsie ultérieure révélant un ostéoblastome agressif. Cela suggère que la lésion initiale était peut-être un ostéoblastome à un stade précoce de développement.

Conclusion: Ce cas remet en question le concept de transformation d'un ostéome ostéoïde en ostéoblastome. Bien qu'histologiquement similaires, ces tumeurs doivent être considérées comme des entités distinctes, et la taille seule peut ne pas être un facteur de différenciation fiable. Une corrélation clinico-pathologique attentive, en tenant compte du taux de croissance, est essentielle pour un diagnostic et une prise en charge précis.

Mots clés: Biopsie, Diagnostic Différentiel, Enfant, Récidive, Tumeurs osseuses

Correspondance
Mohamed Achraf Ferjani
Elmanar University, Faculty of Medicine of Tunis, 1007, Kassab Orthopedic Institute, Pediatric Orthopedic Department, 2010, Tunis, Tunisia.
Email: achraffjn@gmail.com

LA TUNISIE MEDICALE-2025; Vol 103 (04): 519-522 DOI: 10.62438/tunismed.v103i4.5558

Introduction

Osteoid osteoma and osteoblastoma are rare, benign bone lesions of osteoblastic origin [1, 2]. While historically considered variants of the same process, distinguished primarily by size, till late 2024, they are recognized as distinct entities [3, 4].
Osteoid osteoma typically exhibits minimal growth, rarely exceeding 1-2 cm in diameter, and is often characterized by nocturnal pain relieved by salicylates [1, 2]. Conversely, osteoblastoma can grow larger, often presenting with more aggressive behavior, including bone destruction, soft tissue infiltration, and even extension into the epidural space [5, 6, 7]. While both tumors share histological similarities, distinguishing them remains challenging, particularly in cases with borderline features [3, 4].
This case report highlighted a unique clinical presentation in which a recurring osteoblastoma initially mimicked the presentation of osteoid osteoma, raising questions about the accuracy of initial diagnosis and the potential for misclassification.

Observations

Patient Information

An 8-year-old boy presented with a six-month history of left thigh pain without any prior trauma. There was no relevant past medical or family history.

Clinical Findings

Physical examination revealed a left limp with muscle wasting and deep tenderness in the upper third of the thigh.

Timeline of current episode

The patient presented with a six-month history of left thigh pain. Initial imaging (X-rays and Computed tomography (CT)) revealed osteocondensation and a central cavity in the upper femur, leading to a presumptive diagnosis of osteoid osteoma. A biopsy and resection were performed, providing two years of pain relief. Recurrence of pain prompted a second resection, again for presumed osteoid osteoma, providing temporary relief for six months. Subsequent recurrence of pain, accompanied by fever and elevated inflammatory markers, led to magnetic resonance imaging (MRI), which revealed juxta-cortical microabscesses. A wide resection with cement spacer placement was performed, with the final diagnosis of osteoblastoma confirmed histologically. Three months later, the spacer was replaced with a bone graft. At two-year follow-up, the patient was asymptomatic with a normal gait and a 3-cm leg length discrepancy, which was addressed with epiphysiodesis (Table 1).

Table 1. Timeline of patient's clinical course.

Timeframe	Symptoms	Exams	Treatments	Findings
6 months prior to presentation	Left thigh pain			
Initial presentation	Limp, muscle wasting, deep tenderness in upper thigh	X-rays, computed tomography scan	Biopsy	Osteocondensation in upper femur, initial diagnosis of osteoid osteoma: resection
2 years post-initial diagnosis	Recurrence of painful symptoms	Radiological examination	Second resection	Cavity-like image, pathological exam confirmed osteoid osteoma
6 months after second resection	Increased pain, fever spikes, feeling of tension in thigh	Laboratory tests, magnetic resonance imaging		Elevated C-reactive protein and erythrocyte sedimentation rate, multiple juxta-cortical microabscesses
Following imaging and lab findings			Wide diaphyseal resection, internal osteosynthesis with cement spacer	Specimen confirmed diagnosis of osteoblastoma
3 months post-resection			Graft using two tibial crests	
2 years post-graft	No painful symptoms, normal gait, 3cm leg length discrepancy	Radiological examination	Epiphysiodesis of distal end of femur	X-rays show graft incorporation and healing, and the patient has a good prognosis.

Diagnostic Assessment

Initial X-rays showed osteocondensation in the upper third of the femur. CT confirmed the osteocondensation and revealed a cavity-like image (Figure 1).
Laboratory tests showed mildly elevated inflammatory markers initially but later revealed C-reactive protein level of 15 mg/L (normal range: < 3 mg/L) and an erythrocyte sedimentation rate of 40 mm/hr (normal range: < 15 mm/hr). MRI demonstrated multiple juxta-cortical microabscesses in addition to the intra-medullary lesion (Figure 2).

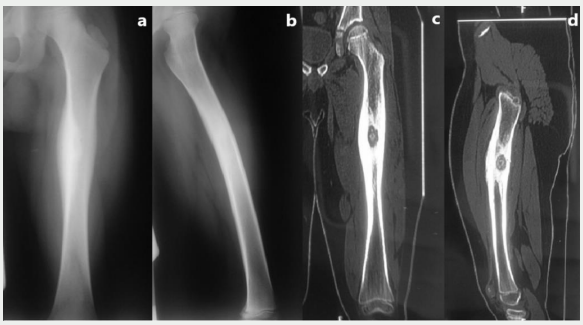

Figure 1. a and b: Initial X-ray showing osteocondensation in the upper third of the femur. **c and d**: Computed tomography scan showing osteocondensation in the upper third of the femur, consistent with a nidus.

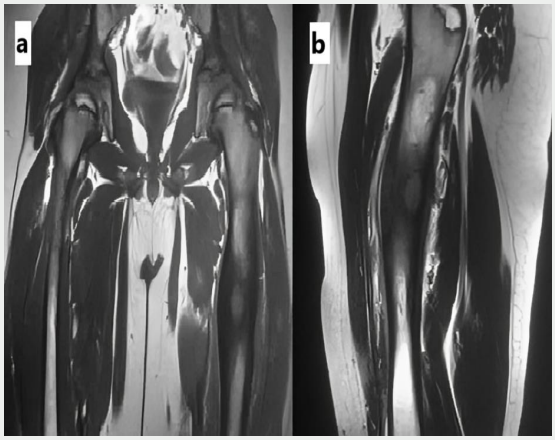

Figure 2. Magnetic resonance imaging demonstrating multiple juxta-cortical microabscesses in addition to the intra-medullary lesion, raising suspicion of infection.

The diagnostic challenge was differentiating between recurrent osteoid osteoma and infection, ultimately leading to the final diagnosis of osteoblastoma after wide resection.

Diagnosis

The final diagnosis was osteoblastoma of the left femur (Figure 3).

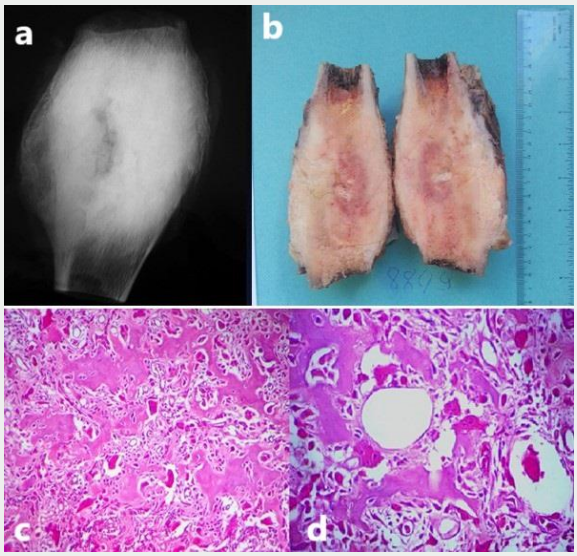

Figure 3. a and b: Entire pathological lesion removed during the final resection, revealing a copious vascular supply. **c and d**: Hematoxylin Eosin stain x 250 and x 400: Histological examination revealing fairly regular trabeculae of osteoid, as well as the presence of osteoblasts and osteoclasts in contact with blood vessels, confirming the diagnosis of osteoblastoma.

Osteoid osteoma was the presumptive diagnosis prior to the final resection. Infection was also considered due to the imaging and laboratory findings. The prognosis is good, based on both the histologically benign nature of the lesion and the two-year follow-up showing resolution of symptoms and a normal gait.

Therapeutic Interventions

Initial treatment consisted of biopsy and resection of the presumed osteoid osteoma. Due to recurrent symptoms and imaging findings, a second resection was performed. Definitive surgical management involved a 12 cm wide diaphyseal resection (Figure 2), internal osteosynthesis with a cement spacer, followed by bone grafting using two tibial crests. Finally, epiphysiodesis of the distal femur addressed the resultant leg length discrepancy.

Follow-up and outcome of interventions

At the two-year follow-up, the patient was asymptomatic with a normal gait. A 3-cm leg length discrepancy, addressed with epiphysiodesis, was noted. Radiological examination confirmed graft incorporation and healing. The patient and his parents expressed satisfaction with the clinical outcome. The patient demonstrated excellent adherence to the treatment plan and follow-up schedule, with normal range of motion in the hip and knee, and excellent functional outcome. No adverse events were reported.

Discussion

The main message of this case report wass that a recurring osteoblastoma can initially mimic an osteoid osteoma, highlighting the challenges in differential diagnosis and the importance of considering factors beyond size when making a diagnosis [3, 8]. The initial presentation, including the response to treatment and the subsequent recurrence, strongly suggested osteoid osteoma. However, the rapid progression of the lesion, the development of more severe pain and systemic symptoms, and the final histological confirmation of osteoblastoma raise important questions about the initial diagnosis.

While size criteria have historically been used to distinguish between these tumors [2], this case highlights the limitations of relying solely on size [9]. The clinical course of this patient underscores the importance of considering the rate of tumor progression, the clinical presentation, and the response to treatment in addition to the histological features.

In 2005, Bruneau et al. [10] reported a case that suggested the possibility of an osteoid osteoma transforming into an osteoblastoma. Similarly, Cappuccio et al. [11] presented a case highlighting the progression of a lesion initially suspected to be an osteoid osteoma into an osteoblastoma. These findings underscore the diagnostic and clinical challenges in distinguishing these two conditions, as well as their potential relationship.

Our case, with its relatively rapid progression and the initial misdiagnosis, supports the idea that osteoblastomas may initially mimic the presentation of osteoid osteomas, particularly in cases with aggressive behavior. Asymptomatic osteoid osteomas are relatively rare, accounting for approximately 1.5% to 2% of cases, as reported by Jackson et al. [12] in their literature review identifying 14 cases. Challenges in interpreting the histological features of these lesions may arise from fragmentation or alteration of biopsy specimens.

A notable example is a case described by Dunlop et al. [13] in 1970, involving a metacarpal osteoid osteoma that recurred twice after en-bloc resection. In 1989, following 11 surgical excisions over 21 years, Morton et al. [14] reevaluated their initial diagnosis and proposed that the lesion was not an osteoid osteoma as previously thought but rather a locally aggressive osteoblastoma. This case underscores the significant difficulties in differentiating between osteoid osteoma and osteoblastoma, highlighting the critical role of the tumor's time-course in providing diagnostic clarity.

This case also emphasizes the importance of multidisciplinary collaboration between clinicians and pathologists in evaluating and managing these tumors. A comprehensive assessment of the clinical, radiological, and histological features is crucial to make an accurate diagnosis and guide appropriate treatment strategies.

Conclusion

This case report highlighted a challenging scenario where a recurring osteoblastoma initially mimicked an osteoid osteoma. Our findings suggest that the initial diagnosis may have been a primary osteoblastoma caught in its early developmental stage, presenting as a seemingly typical osteoid osteoma. We emphasize the need for a thorough multidisciplinary evaluation of these tumors to ensure accurate diagnosis and effective management, especially in cases with rapid growth, severe pain, or atypical presentations.

References

1. Frassica FJ, Waltrip RL, Sponseller PD, et al. Clinicopathologic features and treatment of osteoid osteoma and osteoblastoma in children and adolescent. Orthop Clin North Am. 1996;27:559-574.
2. Dahin DC. Bone tumors (Ed. 3). Charles C Thomas, Springfield, Illinois; 1967.
3. Schajowicz F, Lemos C. Osteoid osteoma and osteoblastoma. Closely related entities of osteoblastic derivation. Acta Orthop Scand. 1970;41:272.
4. Gitelis S, Schajowicz F. Osteoid osteoma and osteoblastoma. Orthop Clin North Am. 1989;20:313-325.
5. Jaffe HL. Benign osteoblastoma.Bull Hosp Joint Dis.1956 Oct;17(2):141-51.
6. Dale S, Breidahl WH, Baker D, Robbins PD, Sundaram M. Severe toxic osteoblastoma of the humerus associated with diffuse periostitis of multiple bones. Skeletal Radiol. 2001;30(8):464-8.
7. McLeod RA, Dahin DC, Beabout JW. The spectrum of osteoblastoma. AJR. 1976;126:321-335.
8. Atesok KI, Alman BA, Schemitsch EH, Peyser A, Mankin H. Osteoid osteoma and osteoblastoma. J Am Acad Orthop Surg. 2011;19:678-689.
9. Shields DW, Sohrabi S, Crane EO, Nicholas C, Mahendra A. Radiofrequency ablation for osteoid osteoma - Recurrence rates and predictive factors. Surgeon. 2018;16(3):156-162.
10. Bruneau M, Polivka M, Cornelius JF, George B. Progression of an osteoid osteoma to an osteoblastoma. Case report. J Neurosurg Spine. 2005;3:238-241.
11. Cappuccio M, De Iure F, Amendola L, Corghi A, Gasbarrini A. Cervical osteoid osteoma progression to osteoblastoma. Spine J. 2014;14(6):1070-1.
12. Jackson RP, Reckling FW, Mants FA. Osteoid osteoma and osteoblastoma. Similar histologic lesions with different natural histories. Clin Orthop Relat Res. 1977;(128):303-13
13. Dunlop JA, Morton KS, Eliott GB. Recurrent osteoid osteoma. Report of a case with a review of the literature. J Bone Joint Surg Br. 1970;52(1):128-33.
14. Morton KS, Quenville NF, Beauchamp CP. Aggressive osteoblastoma. A case previously reported as a recurrent osteoid osteoma. J Bone Joint Surg Br. 1989;71(3):428-31.