

## Case Report

# Osteoid Osteoma of Elbow Two Case Reports and Review of Literature

A. Zarezadeh MD\*, M. Karimian Marnani MD\*\*, A. Pahlevansabagh MD\*\*

## ABSTRACT

Although osteoid osteoma is a relatively common lesion, it rarely occurs at elbow.

We report two cases of osteoid osteoma of elbow in trochlea. Diagnosis was delayed because of non-specific clinical and radiological features. The two patients suffered from flexion contracture of elbow due to synovitis, while at the same time, pronation and supination remained normal. Only one of the patients complained of specific nocturnal pain. Both patients had latencies between the onset of symptoms and the appearance of radiological signs. Open surgical excision of the nidus resulted in complete relief and motion recovery in both cases. Diagnostic difficulties and treatment options are discussed below.

**Key words:** Osteoid osteoma, Elbow, Trochlea

Osteoid osteoma of long bones was first described as a clinical and pathological entity by jaffe in 1935<sup>1</sup>. It is usually found on diaphysis of the femur and the tibia but the tumor has been reported in almost every long bone<sup>1</sup>. Its incidence is two to three times higher in men, most commonly in 10-30 year age group<sup>8</sup> and is rare over 40 years<sup>9</sup>. Patients complain of increasing pain in the affected area. Typically, the pain is worse at night and is relieved by salicylates especially aspirin<sup>1,4,8,10</sup>. In the radiographic image, the nidus appears as a small radiolucent area within an extensive reactive dense sclerotic bone<sup>1,8</sup>.

In 1947, Sherman<sup>1,11</sup> isolated cases of articular osteoid osteoma and observed that their clinical and radiological presentations differed from cases with lesions on the cortex. In articular lesions, synovitis may be severe and the patient may complain of stiffness, resulting in delay in diagnosis or even misdiagnosis<sup>1</sup>.

The literature contains only a few intra-articular cases of osteoid osteoma and only in a small number of them elbow was involved<sup>1,12-14</sup>.

We report two cases of distal humerus intra-articular osteoid osteoma (in trochlea), which were

wrongly diagnosed as other diseases such as arthritis, infection and trauma. This may be due to unusual presentation in cases with elbow involvement compared to other locations.

## Case History

**Case 1:** A 16-year-old girl who suffered from left elbow pain since 4 years before, referred to our clinic. The patient had suffered constant pain during day with slight exacerbation at night. The pain gradually increased over time. She had a flexion contracture of about 20° and the elbow range of motion decreased to about 40°. Supination-pronation of forearm was normal. The patient experienced partial relief after using NSAIDs. During this period she was managed conservatively for different diagnoses like chronic synovitis, trauma and etc. Applying whole body bone scan and right elbow CT scan, following a normal simple X-Ray, osteoid osteoma of trochlea was discovered. The tumor was excised with a transolecranon approach. Immediately after surgery, pain relieved and elbow range of motion restored to normal several months after surgery (figures 1 & 2).

---

\* Assistant professor, Department of Orthopedics, Isfahan University of Medical Sciences, Isfahan, Iran.

\*\*Resident, Department of Orthopedics, Isfahan University of Medical Sciences, Isfahan, Iran.

Correspondence to: Dr. Abolghasem Zarezadeh, Department of Orthopedics, Al-Zahra University hospital, Isfahan, Iran. E-mail: dr\_drd2000@yahoo.com

**Case 2:** A 19-year-old man who had suffered from right elbow pain since 1.5 years earlier, referred to our clinic. The patient had constant diurnal and nocturnal pain, which had progressively increased over time. He had no local tenderness, and had normal elbow flexion but a lag of extension of about 40° and normal forearm supination and pronation. Since the pain had crippled the patient, he had already undergone elbow surgery in another surgical center. Unfortunately, we were not able to find any information about that procedure and however the patient's pain had persisted even after that operation. His pain relieved only by aspirin. After a whole body bone scan and right elbow CT scan, we found a right trochlear osteoid osteoma which was then completely excised using transolecranon approach. The patient became symptom-free afterwards. Only a 30° lag of right elbow extension remained after the procedure (Figures 3, 4).

## Discussion

Intraarticular osteoid osteoma has rarely been described in literature<sup>1</sup>, and there are only a few reports on elbow joint involvement<sup>1,8,12-16</sup>. When this benign tumor is situated in the elbow, the preferential location is the humeral lower epiphysis<sup>1,7,14</sup>, and particularly olecranon fossa as in the two cases presented above. The diagnosis of elbow osteoid osteoma is frequently delayed<sup>8, 14</sup>. Different types of osteoid osteoma namely cancellous, cortical, subperiosteal or intraarticular<sup>7</sup> have by now been described<sup>1</sup>: In our two cases, osteoid osteoma was cancellous beneath the articular surface of trochlea of humerus. The presence of intraarticular osteoid osteoma provokes specific physiopathological and clinical concerns. The most significant consequence is the induced synovitis responsible for osteoarthritis as first suspected by Sherman<sup>1</sup>. This synovitis has already been described as lymphofollicular and pseudorheumatoid<sup>1,17</sup>. It could be due to a mediated reaction of the synovial immune T-cell or to an intraarticular osteoid osteoma<sup>1,19</sup>. Large amount of prostaglandin E<sub>2</sub> and prostacyclin were found directly in the tissue of nidus<sup>7</sup>. Progressive stiffness of the joint will consequently appear. A flexion contracture of elbow is common, especially in cases of delayed diagnosis and in patients who had had a previous diagnostic arthrotomy<sup>8</sup>. The contracture is expected to resolve gradually in most cases follow-

ing excision of osteoid osteoma<sup>8</sup>. This affects the humero-ulnar compartment<sup>1,14</sup> when osteoid osteoma is located in distal humerus, or the proximal radioulnar joint or when the osteoid osteoma is either in the radial head<sup>1</sup> or in the ulnar small sigmoid surface<sup>1,19</sup>. Therefore, the pain can be diurnal and mechanical because of the synovitis and still associated with a typical nocturnal pain<sup>1</sup>. Moreover, synovitis slowly leads to cartilage destruction, which causes a definitive osteoarthritis<sup>1</sup> when the diagnosis is delayed.

Most authors<sup>1,10</sup> have emphasized on the delayed diagnosis of intraarticular humero-ulnar osteoid osteoma (like our two cases). However, clinical and image observation can enable an earlier diagnosis. The clinical presentation is a non-specific joint disease with a variable degree of pain, swelling, muscle atrophy, and inconstant sensitivity to salicylates. The most significant clinical sign is the location of stiffness. When osteoid osteoma is located in the distal part of humerus, pronation and supination remain normal, while flexion and extension progressively decrease. This is especially found in cases of delayed diagnosis. This characteristic feature may be noted in all cases of distal humerus<sup>1,19</sup>. Standard X rays may reveal bone condensation near the nidus on distal humerus, however Cronmeyer et al<sup>1</sup> described subperiosteal bone formation in all adjacent bones. Usually a diffused increase in isotope uptake can be observed in adjacent bones. CT scan is the best imaging modality to show nidus<sup>1</sup> but a subperiosteal nidus can be small and misdiagnosed. MRI may then provide additional signs like bone edema near the nidus<sup>1,2</sup>, effusion and synovitis in adjacent joint. Generally MRI has little role in making the diagnosis<sup>9</sup>.

The indicated treatment is enblock excision of nidus<sup>1</sup>. Medical treatment (NSAID<sub>s</sub> medication) may be successful in controlling the symptoms until spontaneous resolution of lesion occurs<sup>8,20</sup>, though this may take several years. In our two cases, we performed open surgical excision.

Pain relief is usually dramatic and permanent following complete excision of lesion<sup>8</sup>. Nevertheless, other operative techniques such as CT-guided excision or thermal ablation may be considered. If excision of nidus is performed in time, the full range of motion may recover after surgery<sup>1</sup>.

In conclusion, osteoid osteoma of the elbow may easily be missed unless kept in mind.



Figures 1. Axial CT.Scan of elbow (the first Case)



Figure 2. Coronal CT scan of elbow (the first case)



Figure 3. Axial CT scan of elbow (the second case)

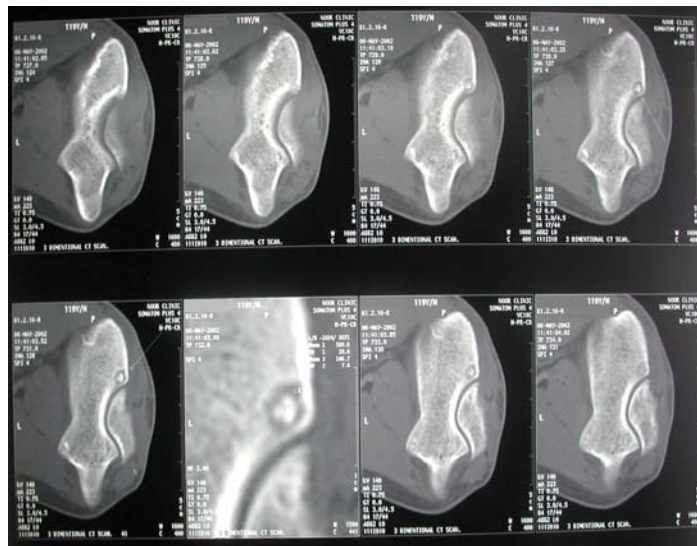


Figure 4. Axial CT.Scan of elbow (the second case)

## References

1. Cassard X, Accadbled F, Gauzy JS, Cahuzac JP. Osteoid osteoma of the elbow in children. A case report and review of the literature. *Journal of pediatric orthopaedics*. 2002 Jul; 11(3): 240-244.
2. Assoun J, Richardi G, Railhac JJ, Bsunin C. Osteoid osteoma: MR imaging versus CT. *Radiology* 1994; 191: 217- 23.
3. Weber KL, Morreg BF. Osteoid osteoma of the elbow: a diagnostic challenge. *Journal of Bone and joint surgery*. 1999 Aug; 81(8): 1111-9.
4. Douglas A. Linville. *Other disorders of spine*. in: S. Terry canale. *Campbell's operative orthopaedic*; Philadelphia, Mosby Company, 2003: 2106-7.
5. Schai P, Friederich N, kruger A, Jundt G, Herbe E, Buess P. Discrete synchronous multifocal osteoid osteoma of the humerus. *Skeletal radiology*. 1996 Oct; 25(7): 667-670.
6. Barney L. Freeman. *Scoliosis and kyphosis*. in: S, Terry canale. *Campbell's operative orthopaedic*; Philadelphia, Mosby Company, 2003: 1921-3.
7. Greco F, Tamburrelli F, Ciabattini G. Prostaglandins in osteoid osteoma. *Int orthop*. 1991; 15: 35-37.
8. Higgins T, Curtin J, Kelly M. Osteoid osteoma of the distal humerus mimicking tennis elbow, *I MJ*. 2002 Sept; 95(8): 225-227.
9. Choen MD, Harrington TM, Ginsburg WW. Osteoid osteoma: 95 cases and a review of the literature. *Semin arthritis rheum*. 1983; 12: 265-280.
10. Greenspan, A. Benign bone-forming lesions: osteoma, osteoid osteoma and osteoblastoma: Clinical, imaging, pathologic and differential considerations, *skel. Radiol*. 1993; 22: 485-500.
11. Sherman MS. Osteoid osteoma with change in adjacent joint. A report of 2 cases. *J Bone Surg Am*. 1947; 29: 483-90.
12. Le Huec Jc, Lesprit E, Moinard M, Le Rebeller A. Osteoid osteoma of the elbow. *J shoulder elbow surg*. 1998; 7: 160-3.
13. Moser RP, Kransdorf MJ, Brower AC, Hudson T, Aoki J, Hudson Berrey B, sweet DE. Osteoid osteoma of the elbow. A review of six cases. *Skeletal Radiol*. 1990; 19: 181-6.
14. Otsuka NY, Hasting DE, Fornassier VL. Osteoid osteoma of the elbow case report. *J Hand surg Am*. 1992; 17: 458-61.
15. Brugera JA, Neurman RJ. Primary tumors of the elbow: Review of leeds regional Bone tumor registry. *Orthopaedics*. 1998; 21(5): 551-553.
16. Comparacci M, Ruggieri P, Gasbarrini A, Ferraro A, campanacci L. Osteoid osteoma. Direct visual identification an itralesional excision of nidus with minimal removal of bone. *J Bone joint Surg Br*. 1999; 81(5): 814-20.
17. Sanchis-Alfonso V, Rosello-sastre E, Castellanos J, Esquerdo J. Intra-articular osteoid osteoma of the humerus with synovitis simulating chronic arthritis of the elbow in a recreational tennis player. *Knee surg sports traumatol Arthrosc*. 1994; 2: 45-6.
18. La fforgue P, Senbel E, Boucrat J, Horschowsky N, Golstein MM, chrestian MA, Bernard D, Acquaviva PC. Elbow synovitis related to an intra-articular osteoid osteoma of the humerus with immunologic and histochemical studies. *J Rheumatol*. 1992; 19: 633-6.
19. Pascaryl X, Zipoli B, Honton JL. Osteoid osteoma in the proximal radio ulnar joint, A case report. *Rev chir orthop*. 1993; 79: 306-8.
20. Barbiera F, Bartolotta TV, Lo casto A, Pardo S, Rossello M, D Maria M. Intra-articular osteoid osteoma, diagnostic imaging in three cases. *Radiologia Media*. 2002; 103(5-6): 464-73.