

Contents lists available at ScienceDirect

JSES International

journal homepage: www.jsesinternational.org

An extra-osseous intra-articular osteochondroma of the elbow



Abdullah Nouri, MB, BCh, BAO (NUI, RCSI)^{a,*}, Ali Lari, BCh, BAO (NUI, RCSI)^a, Nusaiba Almuhaisen, MD^a, Islam Eldesouky ^a, Hatem Shaker, MD, PhD^a, Rola Ali, MD, FRCPC^b, Magdy Abdel-Mota'al, MD, PhD^a

^aAlRazi Orthopedic Hospital, AlSabah Medical region, Kuwait City, Kuwait

ARTICLE INFO

Keywords: Elbow Osteochondroma Extra-osseous Intra-articular Tumor Excision

Osteochondromas are among the most common benign osseous tumors, accounting for 20%-50% of all benign bone tumors. These tumors often occur before the second decade of life, with a predisposition toward male patients. However, the true incidence is likely underreported due to the potential asymptomatic nature of the tumor, especially in the older subset of affected individuals. Classically, these are surface osseous lesions projecting from the underlying bone and are composed of cortical and medullary components accompanied by a hyaline cartilaginous cap. ^{1,11} The radiographic features of cortical and medullary continuity with the underlying parent bone are often pathognomonic. ¹¹

Occurrence sites commonly include the metaphysis and the diametaphysis of long bones including the femur and the tibia, often growing away from the articular surfaces. Symptoms are often derived from secondary fractures, bursa formation and irritation, neurological compromise, joint level involvement, and malignant transformation. The extra-osseous chondromas are separate entities that classically form in the absence of bony attachments. These exist in three variants: synovial chondromatosis, intra-articular chondromas, and soft tissue chondromas. The intra-articular connective tissue within joints that possess a sizable capsular space. The initial growth is a consequence of cartilaginous metaplasia, ultimately leading to ossification, at which point they are termed extra-osseous intra-articular osteochondromas.

E-mail address: Abdullahfanouri@gmail.com (A. Nouri).

In this study, we report a case of a 23-year-old male presenting with a symptomatic intra-articular elbow lesion. Although radiographic features of the lesion were consistent with an osteochondroma, the extra-osseous characteristics make this presentation unique. To the best of the author's knowledge, this is the first report of an entirely extra-osseous, intra-articular osteochondroma arising in the olecranon fossa.

Case report

A previously healthy, 23-year-old male patient was referred to our tertiary orthopedic center complaining of an insidious onset of dull pain in the right elbow associated with progressive range of motion limitation for the past 3 months. Initial analgesics were attempted by the patient before presenting to health care services. The patient noticed a hard swelling overlying the posterior aspect of the elbow joint. There was no history of trauma, constitutional symptoms, or infection.

Physical examination revealed a hard, mildly tender mass deep to the posterior elbow joint. The range of motion was restricted by 5 to 10 degrees at the extremes of flexion and extension. There were no abnormalities relating to skin condition, temperature, and deformities. Neurovascular examination was unremarkable. A general whole-body examination was unremarkable for any pathology.

Hematological and biochemical investigations performed did not reveal any abnormalities. Plain radiographs of the right elbow revealed an osseous mass located in the olecranon fossa with features of a sclerotic and calcified cortical margin (Fig. 1). The rest of the ipsilateral limb was otherwise normal. A nonenhanced magnetic resonance image showed a well-defined intra-articular

^bDepartment of Pathology, Faculty of Medicine, Kuwait University, Kuwait City, Kuwait

Institutional review board approval was not required for this case report. The authors confirm they have obtained patient informed consent form(s) for this case report.

^{*}Corresponding author: Abdullah Nouri, MB, BCh, BAO (NUI, RCSI), Al-Razi Hospital, Jamal Abdulnaser Street, P.O Box 4235, Safat 13043, Kuwait.

A. Nouri, A. Lari, N. Almuhaisen et al. [SES International 7 (2023) 167–170



Figure 1 Lateral plain radiograph of the right elbow obtained preoperatively showing an osseous mass in the posterior aspect of the distal humerus.

nodule $(2.8 \times 2.6 \times 1.7 \text{ cm})$ in the right elbow's posterior joint space, with internal mineralization areas (Figs. 2 and 3).

Next, a computed tomography—guided core needle biopsy was performed. Samples were subsequently analyzed by a clinical histopathologist. The sections showed two cores of hyaline cartilage, one of which was focally covered with synovial lining. In addition, the hyaline cartilage showed a focal increase in cellularity, with few cells showing mild to moderate nuclear atypia. The histopathological differential diagnosis of well-differentiated cartilage on core biopsy included several bone lesions, such as enchondroma, atypical cartilaginous lesions/well-differentiated chondrosarcoma, cartilaginous cap osteochondroma, and synovial chondromatosis. A definite histopathological diagnosis of osteochondroma could not be made at this point due to the unusual location, atypical radiological features, and inability to appreciate the characteristic architectural features on limited biopsy material.

After obtaining consent from the patient, the patient underwent surgical exploration through a direct posterior elbow approach. The lesion was intra-articular, situated in the olecranon fossa, and extra-osseous (Fig. 4). It had no continuity with the underlying bone. Grossly, the external surface was knobby and bluish-white and was consistent with features of an osteochondroma (Fig. 5). Marginal excision of the lesion was performed, and intra-operative fluoroscopic images confirmed adequate resection. Intra-operatively following excision of the lesion, full passive range of elbow flexion and extension was achieved. No elbow immobilization was required, and the elbow and forearm's active range of motion exercises were encouraged immediately postoperatively.

The sample was subsequently sent for a final histopathological assessment. It consisted of a lobulated bony outgrowth with a smooth outer surface measuring $4.0 \times 2.6 \times 1.5$ cm (Fig. 6). The cut surface revealed a cartilaginous cap that is less than 2 cm in maximum thickness with an underlying broad base (Fig. 7). Microscopic sections confirmed the presence of a cartilaginous cap composed of well-differentiated hyaline cartilage and covered by perichondrium. The cartilage displayed a lobular arrangement of chondrocytes with enchondral ossification reminiscent of an epiphyseal plate. No significant nuclear atypia or mitosis was seen, and there was no evidence of permeation of preexisting bone histologically, ruling out well-differentiated chondrosarcoma. Fatty

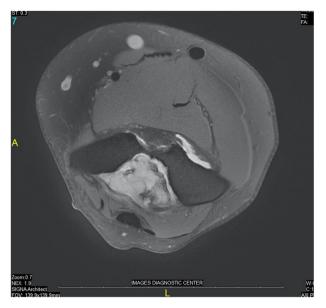


Figure 2 Axial magnetic resonance image (MRI) section of the right elbow showing an intra-articular osseous mass.



Figure 3 Sagittal magnetic resonance image (MRI) section of the right elbow showing an intra-articular osseous mass.

marrow elements were present within the bony stalk. The overall pathological features were those of a sessile osteochondroma.

The patient had an uneventful postoperative phase and performed gradual full range of motion exercises of the elbow joint. The patient regained full range of motion and symptom resolution.

Discussion

Osteochondromas are the most common benign bone tumors occurring around the growth plates of long bones, especially in the skeletally immature population, and tend to extend into the diaphysis. Most common locations include the distal femur, the proximal tibia, and the humerus. Thus, an intra-articular osteochondroma in an adult is quite rare. In 1891, Virchow et al

A. Nouri, A. Lari, N. Almuhaisen et al. [SES International 7 (2023) 167–170



Figure 4 Operative findings: posterior right elbow approach revealing a solid mass consisting of osseocartilaginous nodules situated in the olecranon fossa.



Figure 5 Gross specimen of the osteochondroma after surgical removal.

hypothesized that an osteochondroma is derived from aberrant cartilaginous tissue arising from the physis that separates during growth and gives rise to a separate area of bone formation through endochondral ossification.¹⁴ This hypothesis was further supported by a histopathologic study conducted by Milgram.⁷

Although extra-articular osteochondromas are usually asymptomatic, intra-articular variants cause pain and limitations in the range of motion of the affected joint. Jaffe et al first described intra-articular osteochondromas in 1958 as lesions that occur in the joint capsule or the soft tissue adjacent to the affected joint and thus termed the lesions para-articular chondromas or intracapsular chondromas.⁴ Eventually, the term para-articular osteochondromas was established to discriminate the latter from synovial



 $\textbf{Figure 6} \ \ \text{Macroscopic appearance of the specimen: Bosselated outer surface covered} \ \ \text{by smooth perichondrium.}$



Figure 7 Macroscopic appearance of the specimen: Cut surface showed a cartilaginous cap not exceeding 2 cm in thickness (*arrow*) and a stalk containing marrow elements.

chondromatosis.¹² Extra-skeletal osteochondromas are believed to originate from the differentiation of mesenchymal cells of the soft tissues.⁶ Furthermore, intra-articular osteochondromas are usually described as tumors composed of a single mass with multiple

A. Nouri, A. Lari, N. Almuhaisen et al. [SES International 7 (2023) 167–170

osteochondral nodules in which marginal resection or conventional surgical excision is the definitive treatment of choice, ^{9,12} thus highlighting the importance of appropriate diagnoses to guide management.

Several cases of intra-articular osteochondromas of the elbow joint were reported in the literature. Morin et al reported a case of a 14-year-old girl who presented with a progressive contracture of the left elbow joint. Although initially diagnosed as a posttraumatic flexion contracture, an elbow's computed tomography showed increased bone density arising from the distal humerus with an extension into the coronoid and olecranon fossa. The diagnosis of intra-articular osteochondroma was confirmed by histological examination.⁸

Dittrich et al also reported a case of a 33-year-old gentleman who presented with pain and loss of motion of the right elbow. Imaging revealed small shadows within the proximal radioulnar joint space. Intra-operatively, the capsule was incised and revealed three extra-skeletal bony structures attached by fine cartilage strands and bone. Although extra-osseous, it was situated in the proximal radioulnar joint rather than the olecranon fossa.² Shariatzadeh et al similarly reported a case of a 33-year-old woman who presented with right elbow limitation of motion and pain for 3 years. Radiographical studies showed an osseous mass in the anterior aspect of the distal humerus with a calcified cortical margin. Intra-operatively, they found a completely intra-articular lobulate white mass with a chondral covering consistent with the typical cap of an osteochondroma. In contrast to our case, which was completely extra-osseous, this lesion was continuous with the capitellum and the trochlea. 13 Although there have been reports of arthroscopic excision being performed, open surgical exploration and careful excision remain the gold standard of treatment.⁵

Conclusion

In this study, we describe the first case of an extra-osseous, intra-articular osteochondroma of the elbow joint situated in the olecranon fossa. Appropriate workup abiding by standard principles and subsequent surgical excision results in excellent results and symptom resolution. We advocate for careful diagnoses and a high index of suspicion in these exceedingly rare tumors in order to avoid overtreatment.

Disclaimers:

Funding: No funding was disclosed by the authors.

Conflicts of interest: The authors, their immediate families, and any research foundation with which they are affiliated have not received any financial payments or other benefits from any commercial entity related to the subject of this article.

Patient consent: The authors confirm they have obtained patient informed consent form(s) for this case report.

References

- De Salvo S, Pavone V, Coco S, Dell'Agli E, Blatti C, Testa G. Benign bone tumors: an overview of what we know today. J Clin Med 2022;11:699. https://doi.org/ 10.3390/icm11030699.
- 2. Dittrich RJ. Osteochondromatosis of the elbow. Am J Surg 1946;72:125-7.
- González-Lois C, García-de-la-Torre P, SantosBriz-Terrón A, Vilá J, Manrique-Chico J, Martínez-Tello J. Intracapsular and para-articular chondroma adjacent to large joints: report of three cases and review of the literature. Skeletal Radiol 2001; 30:672-6
- 4. Jaffe H. Tumors and tumorous conditions of the bones and joints. Philadelphia: Lea and Febiger; 1958. p. 558-67.
- Jang S-H, Song H-E. Arthroscopic excision of intra-articular osteochondroma of the elbow: a case report. Clin Shoulder Elb 2016;19:172-5. https://doi.org/ 10.5397/cise.2016.19.3.172.
- Malhotra K, Nunn T, Chandramohan M, Shanker J. Metatarsal stress fractures secondary to soft-tissue osteochondroma in the foot: case report and literature review. Foot Ankle Surg 2011;17:e51-4. https://doi.org/10.1016/j.fas.20 11.05.003.
- Milgram JW. The origins of osteochondromas and enchondromas. A histopathologic study. Clin Orthop Relat Res 1983:264-84.
- 8. Morin B, Pelletier A, Cisa J, Marton D. Intraarticular osteochondroma of the elbow. J Shoulder Elbow Surg 1994;3:270-2.
- Mosher JF Jr, Kettelkamp DB, Campbell CJ. Intracapsular or para-articular chondroma. A report of three cases. J Bone Joint Surg Am 1966;48: 1561-9
- Mulcahy H, Hoch B. An extraskeletal para-articular osteochondroma of Hoffa's fat pad. Radiol Case Rep 2010;5:420. https://doi.org/10.2484/rcr.v5i4.420.
- Murphey MD, Choi JJ, Kransdorf MJ, Flemming DJ, Gannon FH. Imaging of osteochondroma: variants and complications with radiologic-pathologic correlation. Radiographics 2000;20:1407-34.
- Rizzello G, Franceschi F, Meloni MC, Cristi E, Barnaba SA, Rabitti C, et al. Paraarticular osteochondroma of the knee. Arthroscopy 2007;23:910.e1-4. https:// doi.org/10.1016/j.arthro.2006.05.030.
- 13. Shariatzadeh H, Jafari D, Taheri H, Jamshidi K, Pahlevansabagh A. Intra-articular osteochondroma of the elbow: a case report. J Shoulder Elbow Surg 2010;19: e1-4. https://doi.org/10.1016/j.jse.2009.09.013.
- 14. Virchow, R. Ueber multiple Exostosen, mit Vorlegung Von Praparten. Berl. Klin. Wochenschr: Berlin, Germany, 1891;28:1082.