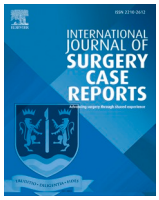
International Journal of Surgery Case Reports 95 (2022) 107261

Contents lists available at ScienceDirect

International Journal of Surgery Case Reports

journal homepage: www.elsevier.com/locate/ijscr



Case report

About a misleading calcified mass of the elbow: Soft tissue osteochondroma: A case report

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ARTICLE INFO

Keywords:
Osteochondroma
Soft tissue
Exostosis
Surgery
Elbow

ABSTRACT

Introduction: Soft tissue osteochondromas are rare lesions, with a misleading radiology, that can present diagnostic dilemma.

Case presentation: We report the case of 16-year-old girl who presented an anterior swelling in his left elbow, slightly painful on palpation, with no signs of inflammation. Elbow mobility was normal. Standard radiographs showed a globular calcified mass. CT scan revealed a well-defined lobulated soft tissue mass with extensive flaky calcification. Magnetic resonance imaging (MRI) showed lobulated and intermuscular non-enhancing mass. The lesion had a low heterogeneous signal on T1 images, and a high heterogeneous signal on T2 FAT-SAT images, with low signal areas suggesting calcifications. There was no continuity to the adjacent ulna and radius. The patient first underwent image-guided core needle biopsy and histological examination concluded to chondroma. The tumour was then resected by anterior approach and the final histological diagnosis was osteochondroma of soft tissue.

Discussion: A diagnosis of extraskeletal osteochondroma should be considered when an ossified mass is localized in the soft tissue particularly on the elbow. Excision is the treatment of choice when the function is reduced and the nature of the tumour remain uncertain.

Conclusion: Orthopedic surgeons should know about elbow localization of osteochondromas to help in accurate diagnosis and management of the tumour, to avoid overtreatment.

1. Introduction

Osteochondromas are one of the most common bone tumours, making up 10–15% of all bone tumours and 20–50% of benign bone tumours [1]. It is a bone exostosis in which a continuous cortical layer projects from the underlying bone [2]. Approximately 50% occur in the long bones of the lower extremity, especially around the knee, in the distal femur, and proximal tibia. In the upper extremity they may be encountered in the proximal humerus [1]. In rare cases, osteochondromas can be discontinuous from underlying bone with a misleading radiological feature. Then, these soft tissue osteochondromas or extraskeletal osteochondromas can present diagnostic dilemma. It has been reported in several tissues of the limbs and in particular localizations such as kidney, liver and tongue [3]. To the best of our knowledge, there are only two case reports of a soft tissue osteochondroma around the elbow, occurring away from the joints [3,4]. We, hereby report third such case.

The work has been reported in line with the SCARE 2020 criteria [5].

2. Presentation of case

A 16-year-old girl presented with a history of a progressive swelling in his left elbow for the previous two years. There was no history of any trauma, fever or joint effusions. On examination, there was an anterior hard swelling of the upper third of the left forearm, slightly painful on palpation, with no signs of local inflammation. Elbow flexion-extension mobility was normal but range of motion of the prono-supination was restricted at the extremes by fifteen degrees. The neurovascular exam was normal.

Standard radiographs showed an anterior calcified and heterogeneous round mass in front to the proximal ends of the two bones of the forearm with no apparent bone continuity (Fig. 1).

CT scan revealed a well-defined lobulated mass with flaky calcifications, measuring 45 mm × 25 mm (Fig. 2a). MRI showed lobulated and intermuscular, non-enhancing mass, without any continuity to the adjacent ulna and radius. The mass creeps in between the biceps brachii and the brachialis muscles, and it was also in contact with the supinator

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https://doi.org/10.1016/j.ijscr.2022.107261
Received 20 March 2022; Received in revised form 24 May 2022; Accepted 28 May 2022
Available online 1 June 2022

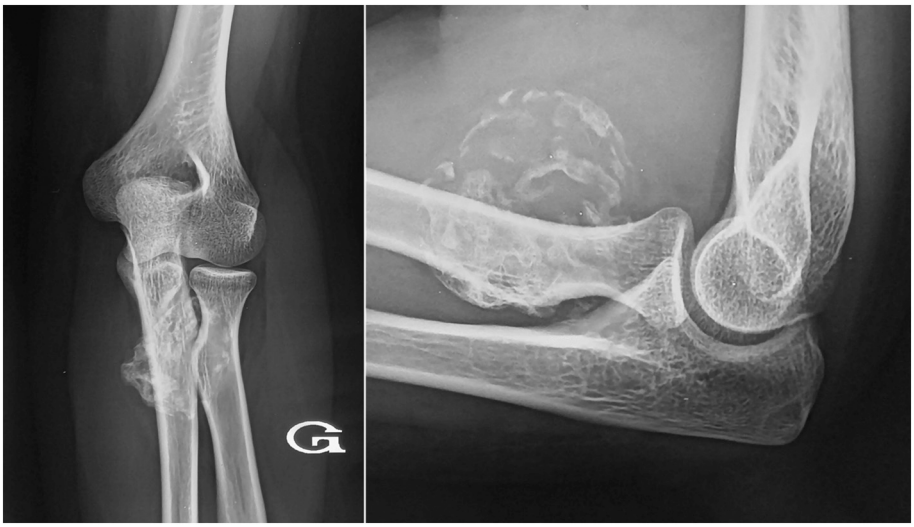


Fig. 1. Standard radiographs showing a globular calcified mass.

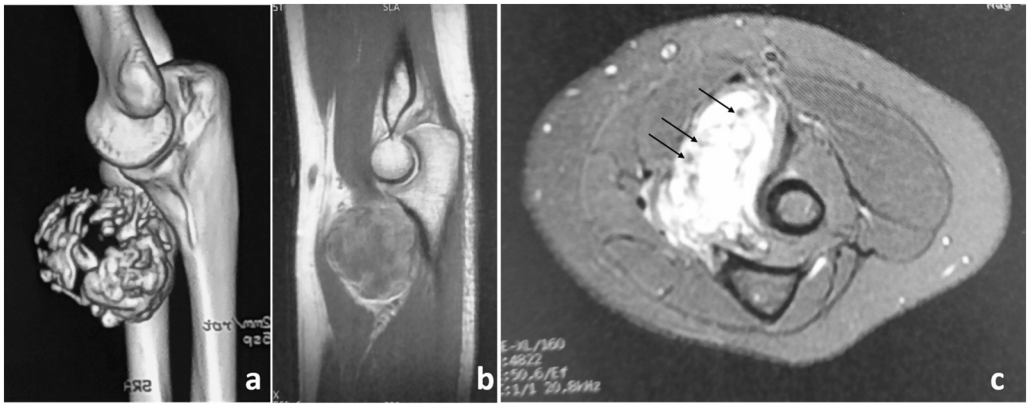


Fig. 2. CT scan showing a well-defined lobulated mass with extensive flaky calcification (2a). In magnetic resonance imaging (MRI), the lesion had a low heterogeneous signal on T1 images (2b), and a high heterogeneous signal on T2 FAT-SAT images (2c). There were low signal areas, predominantly in the periphery, suggesting calcifications (2c).

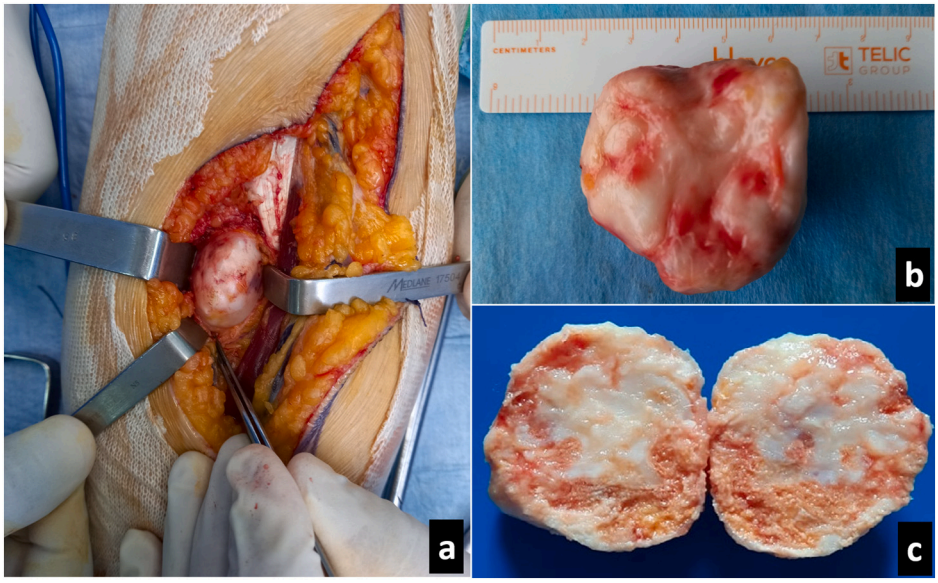
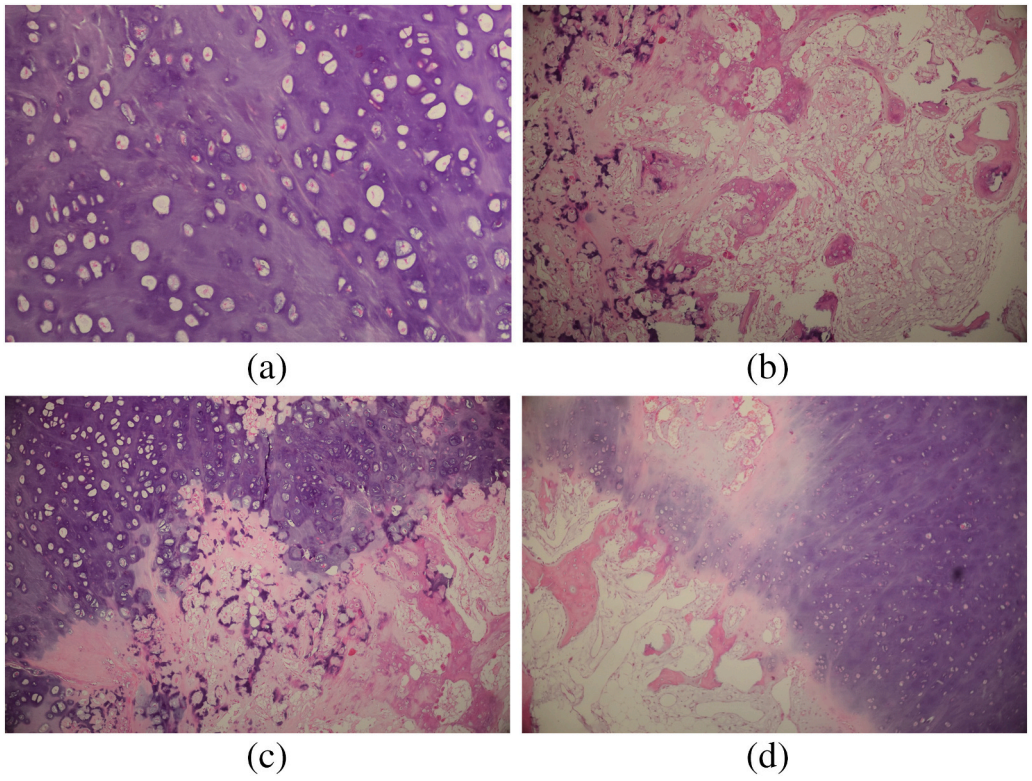


Fig. 3. Intraoperative photo showing a globular tumour in the soft tissues with no accompanying pedicle or connectivity to underlying bones (a). The tumour measured 45 mm × 25 mm (3b). The cut surface showed solid variegated bony hard and soft areas (3c).



Figs. 4. (a) Columns of proliferative chondrocytes surrounded by cartilaginous hyaline matrix, (b) Endochondral ossification with normal bone marrow between the bony trabeculae, (c) Endochondral ossification occurs at the bone-cartilage interface, (d) Endochondral ossification of proliferative chondrocytes.

muscle and the medial epicondylar muscles (Fig. 2). The lesion had a low heterogeneous signal on T1 images (Fig. 2b), and a high heterogeneous signal on T2 FAT-SAT images (Fig. 2c). There were low signal areas, predominantly in the periphery, suggesting calcifications (Fig. 2c). Myostis ossificans was the most suspected diagnosis but soft tissue sarcomas diagnosis was not eliminated.

The patient first underwent image-guided core needle biopsy and histological examination concluded to chondroma. Tumour was then resected by modified antero-lateral approach. After incising the skin and deep fascia, the supinator longus and the extensor muscles were retracted externally. Medial traction on the flexor muscles and then on the neurovascular bundle allowed visualization and exposure of the tumour, which has no continuity with the forearm bones (Fig. 3a). The tumour resection was then accomplished easily. The joint capsule was intact and the tumour was extra-articular. The tumour was generally rounded, hard and white in colour, measuring 45 mm × 25 mm. It had a lobulated cartilaginous surface (Fig. 3b). On section, there was a cartilaginous appearance with the presence on the periphery of a bony cap of varying thickness (Fig. 3c).

Microscopic examination revealed endochondral ossification with a hyaline cartilage cap and mature trabecular bone underneath. Normal bone marrow elements were present between the bony trabeculae (Fig. 4). The final histological diagnosis was osteochondroma of soft tissue, with no signs of malignant transformation.

The postoperative phase was uneventful. At six months follow-up, the patient regained normal elbow function without neuro-vascular complications. He had no clinical or radiographic evidence of recurrence.

3. Discussion

Osteochondroma is the most common primary bone tumour. It is usually an extra-articular lesion arising from the metaphysis of long bones, filled with cancellous bone and covered by a cartilaginous tissue [6]. It can occur in two forms: sessile or pedunculated. In some cases, it can be discontinuous from underlying bone, named then extraskeletal osteochondroma or soft tissue osteochondroma. Soft tissues osteochondromas are uncommon and most of them arise from synovial tissues in joints, tendon sheaths, or bursae [7], giving para-articular, soft-tissue, or intra-articular osteochondromas. Approximately 75% of these latter tumours are found in the hands and 25% in the feet [8]. A synovial osteochondromatosis of the elbow was reported by Kamineni et al. [7] in twelve patients. Few para-articular osteochondromas have been reported, with the knee being the most common site (76%), followed by the foot (19%) and ankle (5%) [9,10]. Extra-skeletal osteochondromas arising from soft tissues have been reported but are rarer. These have been reported in buttock, nape of neck and elbow [4,10–13]. To the best of our knowledge, there are only two case reports of a soft tissue osteochondroma around the elbow [3,4]. We, hereby report the third case.

The pathogenesis of such lesions is unknown and multiple theories of origin were postulated. The mesenchymal cells might undergoes metaplasia to form these lesions [6]. Sood et al. [12] concluded that they arise from metaplastic change from adipose tissue, which was lipoma at the beginning. The long list of tissues that are capable of undergoing such metaplasia reveals the complexity of influences that may determine the chondromatous and osseous transformation of connective tissues [6]. Synovium had also been postulated as a possible origin. Kanwat et al. [2] think that a non-union of a fracture of a pedunculated osteochondroma can result in a soft tissue osteochondroma.

Clinically, the symptoms are not specific and may suggest several differential diagnoses. There is no preponderance for gender. The clinical complaints are local discomfort, moderate pain, slow growing mass and some degree of limited motion in the joint [14]. In our case, elbow flexion-extension mobility was normal but prono-supination was slightly limited.

The radioligical features of soft tissue osteochondromas include scattered amorphous calcifications and ossifications within a soft tissue

mass, well-circumscribed and lobulated [15].

The CT scan can show the extra-skeletal location and central dense areas of calcification or ossification of soft tissue osteochondromas [16].

Magnetic resonance imaging (MRI) better delineates the border of the mass, thus distinguishing it from a sarcoma. Soft tissue osteochondromas have low-to-intermediate signals on T1-weighted images and a mixture of intermediate-to high signals in the cartilaginous areas on T2-weighted images. An intermediate T2 signal intensity is shown in areas of mature ossification except densely calcified areas that have low-signal intensities [16]. MRI provides accurate measurements of these tumours and their relationship with the adjacent structures. MRI also demonstrates the absence of osseous continuity with underling bone.

Histologically, soft tissue osteochondroma may be confused with a bizarre parosteal osteochondromatous proliferation (BPOP) also known as Nora's lesion. It is a rare benign surface bone lesion most commonly involving the small bones of the hands and feet and more common in adults during the second to third decades of life. Histopathological features show typically a disorganized lesion of a cellular cartilaginous cap with atypical chondrocytes, regions of incomplete endochondral ossification, a densely mineralized cartilaginous tissue referred to as basophilic bone or blue bone and interspersed fibrovascular stroma with spindle cells without marrow component.

However, soft tissue osteochondroma, shows a continuity of the cartilaginous cap with endochondral ossification with fatty intertrabecular spaces, generally without cartilaginous atypia and lacks blue bone [16,17].

The radiographic differential diagnosis of such cases must include myositis ossificans, tumoral calcinosis, synovial chondromatosis, and soft tissue osteosarcoma [18]. The correlation of histological and radiographic features allows the distinction.

Diagnosis is based on histological results [3]. Pathologically, the lesion may be confused with periosteal chondroma, enchondroma, chondrosarcoma, parosteal or periosteal osteosarcoma. Whole tissue should be processed to rule out malignant transformation [18].

The treatment of choice is surgical resection, which may not be easy like in our case because of the neuro-vascular bundles. Although the exact biological nature of these tumours is not well characterised, limited data suggest that they behave in a benign fashion and local recurrence is rare [6].

4. Conclusion

Soft tissue osteochondromas of the elbow are rare benign lesions and must be differentiated from other benign or malignant tumours. Clinically, the symptoms are not specific. Imaging shows a misleading calcified mass. Surgical excision is the procedure of choice when function is reduced and the nature of the tumour remain uncertain. A definitive diagnosis is made through histopathological examination. A knowledge about this rare lesion can help in accurate diagnosis and management, to avoid overtreatment.

Consent

The authors confirm that informed consent was obtained from the patient for publication of this case report.

Sources of funding

No.

Provenance and peer review

Not commissioned, externally peer-reviewed.

CRediT authorship contribution statement

Ameni Ammar: Writing- review end editing. Oussama Abcha: Writing- review and editing. Marouen Berriri: translation. Samia Sassi: Supervision. Mahmoud Smida: Supervision.

All the authors read and approved the final version of the manuscript.

Declaration of competing interest

The authors report no declarations of interest.

References

[1] G.M. Rayan, J. Upton, in: Osteochondromas: Congenital Hand Anomalies and Associated Syndromes, Springer Berlin Heidelberg, 2014, pp. 11–17. https://link.springer.com/chapter/10.1007/978-3-642-54610-5_2.

[2] M. Kanwat, V. Jangira, B. Sharma, D. Chauhan, Can an exostosis be free floating without any continuity to the underlying bone: a diagnostic dilemma, J. Clin. Orthop Trauma 8 (2) (2017) 87–89. https://pubmed.ncbi.nlm.nih.gov/29339849/.

[3] L. De Smet, Posterior interosseous neuropathy due to compression by a soft tissue chondroma of the elbow, Acta Neurol. Belg. 105 (2) (2005) 86–88. https://pubmed.ncbi.nlm.nih.gov/16076062/.

[4] R. Narula, A. Arya, L. Verma, S. Singh, Soft tissue osteochondroma in elbow joint: a case report, Malays. Orthop. J. 6 (1) (2012) 53–56. https://pubmed.ncbi.nlm.nih.gov/25279045/.

[5] R.A. Agha, T. Franchi, C. Sohrabi, G. Mathew, for the SCARE Group, The SCARE 2020 guideline: updating consensus Surgical CAse REport (SCARE) guidelines, Int. J. Surg. 84 (2020) 226–230.

[6] Y. Gulati, A. Maheshwari, V. Sharma, R. Mattoo, D. Arora, N. Gupta, Extraskeletal osteochondroma of the thigh: a case report, Acta Orthop. Belg. 71 (1) (2005) 115–118. http://www.actaorthopaedica.be/assets/1104/20-Maheshwari_et_al.pdf.

[7] S. Kamineni, S.W. O'Driscoll, B.F. Morrey, Synovial osteochondromatosis of the elbow, J. Bone Joint Surg. 84 (7) (2002) 961–966. https://pubmed.ncbi.nlm.nih.gov/12358386/.

[8] E.L. Gayle, W.B. Morrison, J.A. Carrino, T.W. Parsons, C.Y. Liang, A. Stevenson, Extraskeletal osteochondroma of the foot, Skelet. Radiol. 28 (10) (1999) 594–598. https://jhu.pure.elsevier.com/en/publications/extraskeletal-osteochondroma-of-the-foot-4.

[9] J.D. Reith, T.W. Bauer, M.J. Joyce, Paraarticular osteochondroma of the knee: report of 2 cases and review of the literature, Clin. Orthop. Relat. Res. 334 (1997) 225–232. https://pubmed.ncbi.nlm.nih.gov/9005917/.

[10] H. Sakai, K. Tamai, A. Iwamoto, K. Saotome, Para-articular chondroma and osteochondroma of the infrapatellar fat pad: a report of three cases, Int. Orthop. 23 (2) (1999) 114–117. https://pubmed.ncbi.nlm.nih.gov/10422030/.

[11] S.-C. Lim, Y.-S. Kim, Y.-S. Kim, Y.-R. Moon, Extraskeletal osteochondroma of the buttock, J. Korean Med. Sci. 18 (1) (2003) 127–130. https://pubmed.ncbi.nlm.nih.gov/12589103/.

[12] I.D. Sood, N.S. Mudbhatkal, Extraskeletal osteochondroma, a case report, Indian J. Pathol. Bacteriol. 10 (1) (1967) 72–76. https://pubmed.ncbi.nlm.nih.gov/6036495/.

[13] R. Singh, A.K. Sharma, N.K. Magu, K.P. Kaur, R. Sen, S. Magu, Extraskeletal osteochondroma in the nape of the neck: a case report, J. Orthop. Surg. 14 (2) (2006) 192–195. https://pubmed.ncbi.nlm.nih.gov/16914787/.

[14] N. Neves, M. Gutierres, M.-E. Paiva, A.-T. Cabral, Para-articular osteochondroma: a case report, Rev. Chir. Orthop. Reparatrice Appar. Mot. 90 (1) (2004) 71–74. https://pubmed.ncbi.nlm.nih.gov/14968006/.

[15] Z.J. Liu, Q. Zhao, L.J. Zhang, Extraskeletal osteochondroma near the hip: a pediatric case, J. Pediatr. Orthop B. 19 (6) (2010) 524–528. https://pubmed.ncbi.nlm.nih.gov/20802344/.

[16] E. Washington, L. Menendez, A. Fedenko, A. Tomasian, Bizarre parosteal osteochondromatous proliferation: rare case affecting distal ulna and review of literature, Clin. Imaging 69 (3) (2020) 233–237. https://pubmed.ncbi.nlm.nih.gov/32977195/.

[17] A. Yoshida, E.F. McCarthy, Bizarre parosteal osteochondromatous proliferation, in: WHO Classification of Tumours, 5th EditionSoft Tissue and Bone Tumours, 2020, pp. 348–350.

[18] S. Batra, B. Garg, S. Arora, Large soft tissue osteochondroma of the heel: a case report and literature review, Musculoskelet. Surg. 97 (3) (2013) 255–258. https://pubmed.ncbi.nlm.nih.gov/22081265/.