International Journal of Case Reports in Orthopaedics

E-ISSN: 2707-8353
P-ISSN: 2707-8345
IJCRO 2024; 6(1): 04-08
www.orthocasereports.com
Received: 14-11-2023
Accepted: 17-12-2023

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A long-standing large enchondroma of 5th metacarpal associated with deformity of the adjacent metacarpal bone: A rare case report

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DOI: https://doi.org/10.22271/27078345.2024.v6.i1a.182

Abstract
Background: Enchondroma is the most common benign hand tumor. Long standing lesion on the skeletally immature group is relatively uncommon and can create an atypical finding.
Methods We present a case of symptomatic large-sized enchondroma in the 5th metacarpal region without pathological fracture despite the 15 years of tumor growth. Moreover, the adjacent 4th metacarpal bone is deformed without causing apparent or functional deformity. The patient was treated with extended intralesional curettage which was augmented with cortical bone graft from iliac crest.
Results The pathological result showed a low-grade cartilaginous neoplasm suggestive of an enchondroma. Post operatively, the patient regained full range of motion and normal grip strength at the three months follow-up. Successful graft integration was evident and no recurrence was observed at the 1-year follow-up.
Conclusions A long-standing enchondroma in skeletally immature bone can cause series of adaptation both in affected and adjacent structure. The intralesional curettage with void filling.

Keywords: Metacarpal, neoplasms, enchondroma, curettage, bone graft

Introduction
Enchondroma is the most common benign tumor of the hand in adults which often presents in the third to fourth decades of life. Pediatric enchondroma is uncommon, as the primary bone tumor itself is relatively uncommon in children. Pediatric enchondroma is reported for up to 24% of all benign bone tumors found in children or adolescents (usually in the second decades) [1–3].
Developed in a benign fashion, enchondroma is usually asymptomatic. This lesion is usually discovered as an incidental finding. Sudden pain associated with pathological fracture or discomfort or pain associated with mass effect is the most common reason for outpatient visits. While small and asymptomatic lesions are generally managed conservatively with regular observations, larger lesions associated with pain, pathological fracture, or recurrence are better treated surgically with adjunctive void-filling graft [4].
We reported a case of a large enchondroma of metacarpal bone which has been developed over 15 years. It remained asymptomatic until it grew large enough to compress its surrounding structure and cause angulation deformity of the adjacent metacarpal.

Case Presentation
A 27-year-old male was presented to the outpatient clinic with mild pain over the 5th right metacarpal region following activities on the past 1 year. No night symptoms, fever, or unexplained weight loss. A slowly progressive diffuse lump with no preceding injury has been developed 15 years back without any pain or discomfort. No other lumps were reported. He had no complaint regarding function. On the physical examination, there was an ill-defined bony lump over the dorso-ulnar aspect of the metacarpal region which was also palpable volarly. No venectations, sinuses, local tenderness, or warmth were noted at the physical examination. He has a full range of motion on MCP and IP joints of all fingers and distal neurovascular status was intact.
The plain radiograph reveals an eccentric lytic lesion with a geographic pattern, punctate calcification, well-defined border, and cortical thinning at the metaphyseal region of the 5th metacarpal neck extended to the proximal third of the diaphysis.

No cortical break, periosteal reaction, or soft tissue involvement were found (Figure 1). Magnetic resonance imaging (MRI) findings showed a large (30x14x18 mm) expansile osteolytic lesion in the meta-diaphysis of the 5th metacarpal with a marked cortical thinning, central area of calcification, and postero-lateral exophytic component. In the T_2 weighted image, the lesion showed hyperintensity with punctate calcification. Both clinical and radiological suggested a benign lesion. Hence, the patient underwent an excisional biopsy and void-filling with cortical strut graft from the iliac crest by an experienced specialist.

A longitudinal dorsal incision between the 4th and 5th metacarpal was made under the brachial block. After the subperiosteal elevation was performed, we trimmed the radial part of the lesion which was compressing the 4th metacarpal as well as the dorsal doom which appeared as a bump over the dorso-ulnar aspect of the hand. Initially, we found a hematoma-filled lesion with normal cortical and cancellous bone. In fact, the bone was hardened and the glistening grey-white tissue was finally discovered on the metaphyseal region just before we finished the excavation (Figure 2). The intralesional curettage was performed carefully with respect to the thinned cortical shell. Finally, thermal and chemical adjuncts to locally control the lesion was performed with high-speed burring and hydrogen peroxide irrigation sequentially. An approximately 3 x 1.5 cm void that was created after the extensive curettage was filled by a cortical bone graft from the iliac crest. The graft included the outer cortical surface to provide a smooth gliding for extensors. After re-shaping the graft to match the void's size and shape, it was impacted into the remaining cortical shell and wrapped with the periosteum

The histopathological examination revealed a cartilaginous lesion with a mild increase in cellularity. The chondrocytes distributed as isolated cells with focal loose clusters and were mildly enlarged with round nuclei and dense chromatin within hyaline cartilage matrix. There were areas with ossification and new bone formation along the periphery of the chondroid fragments. There were focal tiny areas of myxoid changes and necrotic chondrocytes. There were no mitoses, significant cellular atypia, or a destructive growth pattern. These findings confirmed the clinico-radiological diagnosis of enchondroma (Figure 3).

Postoperatively, the patient was splinted for 6 weeks with an ulnar gutter splint with free MCP and IP joints. Active finger motion exercise was initiated immediately. After the splint removal, wrist range of motion exercise and the strengthening program was initiated. On the 3-months follow-up, the patient regained full range of motion with normal grip strength. However, heavy weight lifting was forbidden until the bone achieved a solid union with a complete graft integration which was confirmed on the 6 months follow-up radiograph. At the 12-months follow up, there was no clinical sign of recurrence (Figure 4). No donor site morbidities such as infection or persistent pain were noted during the follow-up. A routine annual follow-up was advised to monitor recurrence.

Figure format

Fig 1: (9 Font size, Times New Roman, Normal, Bold)

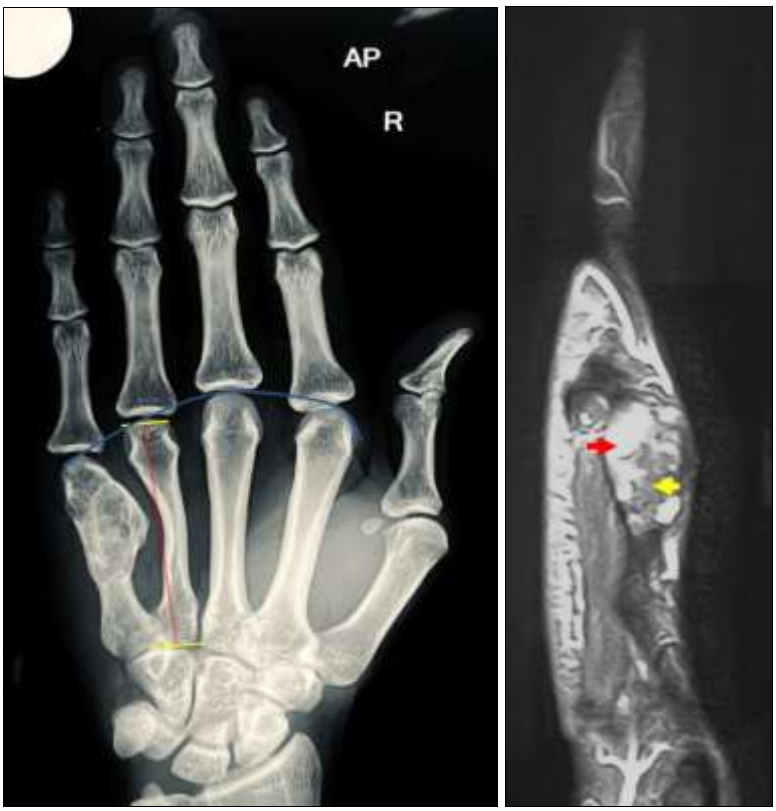

Fig 1: Pre-operative Imaging Studies. A. Plain radiograph of the right hand revealed a solitary lytic lesion on the meta-diaphyseal region of the 5th metacarpal. The 4th metacarpal shaft is angulated with a normal coronal and sagittal alignment (red lines), along with the normal curve made from the neighboring metacarpal heads (blue line). B. The sagittal view of T_2-weighted image on MRI showed hyperintensity area (red arrow) with some calcifications (yellow arrow).

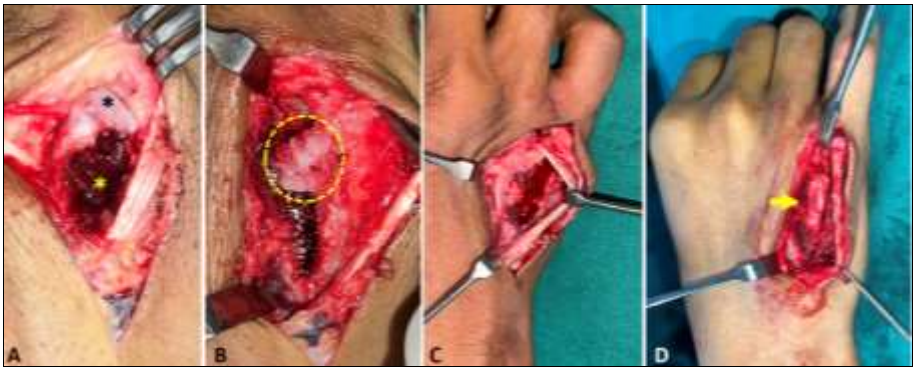

Fig 2: Intraoperative Findings. A. Cortical window has been made and revealed a hematoma-filled cavity (yellow asterix). Note the dorsal cortical bump before trimming (Black asterix). B. Cartilaginous substances on metaphyseal region (yellow circle). C. A huge cavity with thin volar-ulnar cortical shell. D. Shaped-cortical bone graft (yellow arrow) was impacted to fill the void.

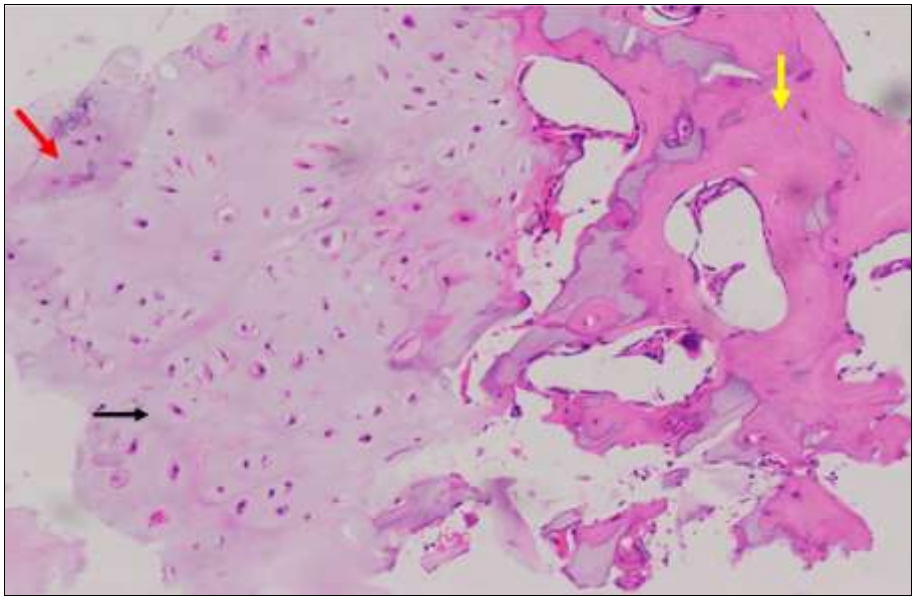

Fig 3: Histopathological findings in hematoxylin and eosin stain represented in 10x magnification. Black Arrow: chondrocytes in lacunae. Red Arrow: Scanty Myxoid Matrix. Yellow Arrow: Endochondral ossification.

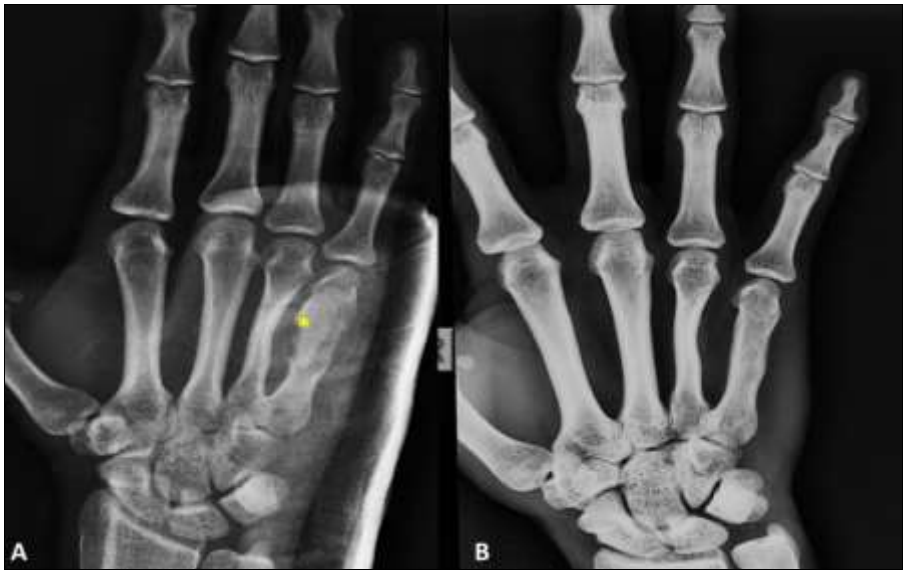

Fig 4: Postoperative Imaging. A. Early post-operative X-Ray shows bone graft (Yellow asterix) is distinguishable from the surrounding cortical shell at meta-diaphyseal region of 5th metacarpal. B. Bone union with successful graft integration has been achieved at the 6-month follow-up.

Discussions

Enchondroma is the most common benign hand tumor with prevalence in proximal phalanx, middle phalanx, metacarpal, and distal phalanx in descending order of frequency. Hypothetically, it arises from the proliferation of physeal remnants that are entrapped within the

intramedullary canal (En-: Inside; Chondroma: benign cartilaginous tumor). Hence, it consists of hyaline cartilage (Sometimes with areas of calcification). Most commonly, it is detected in the second decades of life between the ages of 10 to 30 years and presents with pathological fracture [2, 3].

In our patient, there was no pathognomonic symptom such as pathological fracture. The slowly progressing bony lump and the radiological report that shows a geographic lytic lesion suggesting a benign bone tumor. Statistically, any benign-appearing lesion clinically and radiologically in the hand should be considered as enchondroma until it is proven otherwise [2]. Enchondroma itself has varying radiographic morphology – central vs eccentric, non-mineralized (purely lytic) up to markedly mineralized. Hence, the other benign tumor and tumor-like lesions should be kept in mind especially those that shared a similar expansile tendency (GCT, ABC, Brown tumor) [1, 2].

In our case, the lesion tends to grow to a less resistant area radially and volarly. Interestingly, in our case, the progressively growing tumor might lead to plastic deformation of the skeletally immature adjacent metacarpal bone that was subjected to a long-standing compression. However, the overall alignment was well-maintained so that no apparent or functional finger deformity occurred in this patient. This is might be due to a series of bone remodeling especially during growth, that was adapting to the compression force from the tumor and to the tension forces from the surrounding extrinsic and intrinsic tendons and muscles. Moreover, hand enchondromas were usually located in the metaphysis rather than abutting the growth plate. Thus, shortening or growth disturbance is less likely [3].

In our study, the mass has been developed since the patient was 12-years-old and at presentation before the surgery, it has grown to the size of 3x1.4 cm. Although the size of enchondroma is varied, it is usually not more than 5 cm, of which one should rise the possibility of malignant form. Bierry *et al*. [3] study showed that enchondromas in children appear as relatively large lesions. Sollaci *et al*. [5] are reporting their 20 years' experience in their institute and found that the size of the tumors ranged from 0.2 cm^2 to 5.7 cm2, with a mean size of 1.7 cm2 (SD ¼ 1.0 cm^2). [5] Other studies by Pandey [6] and Ambulgekar *et al*. [4] reported large enchondromas presented with and without pathological fracture respectively.

The large and expansile lesions are at risk of pathological fracture due to cortical weakening [1]. Riester *et al*. [7] study in 54 cases showed that the longitudinal percentage of bone occupied by the tumor was found to strongly correlate with the presence of fracture. The ulnar border finger and metacarpal are also highly associated with fracture as they lack mechanical support from the neighboring finger [7]. Hence, surgery is recommended for large lesions, symptomatic patients with or without pathological fractures [4, 5].

A variety of surgery has been described in the literature. Curettage has been the treatment of choice for hand enchondroma, with the endpoint of curettage being complete removal of the tumor under direct visualization. However, debate persists regarding the needs of adjuvant as well as the efficacy of various methods of post-curettage void augmentation [5]. Extended curettage technique for a benign bone tumor is the utilization of various local adjuvants following curettage with or without high-speed burring as a local control to prevent recurrence. The commonly used adjuvant are cryo-ablative agents, polymethylmethacrylate, phenol, alcohol, hydrogen peroxide, zinc chloride, and argon beam coagulation.[8] Mohler *et al*. [9] supported the use of adjuvant particularly for cartilaginous bone tumors of uncertain malignant potential. The recent treatment algorithm of enchondroma proposed by Tang *et al*. [10] opposed the use of adjuvant treatments due to insufficient clinical evidence that supports its necessity. It seemed that despite their benefit in lowering the rate of local recurrence, the meticulous curettage and high-speed burring through a wide exposure of the tumor cavity is the key to successful local control [8].

The use of post-curettage augmentation also brings a lot of controversies. Bachoura *et al*. [1] suggested that a simple curettage without augmentation does not lead to increased complication rates. This recommendation is supported by the algorithm proposed by Tang *et al*. [10] which recommends only the simple curettage without augmentation on an expansile lesion or on symptomatic cases. Cement augmentation can be considered for particularly large tumor cavities to provide mechanical stability. [10] A biomechanical study in fresh cadavers showed that simple curettage weakened the bone by 70% than the intact bone. Moreover, the extended curettage using adjuvant can further erode the cortex. In a real patient, the cortex has already thin due to the tumor expansion. Sollaci *et al*. [5] studies suggested that bone grafting is necessary for a thin cortical shell that has a high risk for fracture and in lesion which has proximity to the articular surface.

Our case had a large cavity following the curettage that lacks mechanical stability so that we support the bone with a cortical bone graft from the iliac crest. Our patient gained a good bone-graft integration on the 6-month follow-up x-ray, despite we didn't fix the graft with hardware fixation. Although the recurrence is uncommon in solitary lesions and the malignant degeneration is exceedingly rare and less likely in patients below the age of 40, we advised the patient to have a regular yearly follow-up.

Conclusion

Enchondroma should be suspected in the benign bony lesion in the hand region until it is proven otherwise. A long-standing lesion since the age of skeletal growth can cause series of adaptation both in affected and adjacent structure. The large-sized enchondroma should increase the awareness of pathological fracture and the possibility of malignant changes. The intralesional curettage with void filling can mechanically support the weakened bone especially in the large lesion. Thus, it is preventing pathological fracture and giving a good functional outcome to the patient.

Acknowledgments

Disclaimer: No patient or author details are included in the figures.

Conflict of interest: None declared

Funding: This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

References

1. Bachoura A, Rice IS, Lubahn AR, Lubahn JD. The surgical management of hand enchondroma without

post-curettage void augmentation: authors' experience and a systematic review. Hand (New York, N.Y.). 2015;10(3):461-471. DOI:10.1007/s11552-015-9738-y
2. McVey MJ, Kettner NW. Pathologic fracture of metacarpal enchondroma: Case study and differential diagnosis. Journal of Manipulative and Physiological Therapeutics. 2002;25(5):340-344. DOI: 10.1067/mmt.2002.124417
3. Bierry G, Kerr DA, Nielsen GP, *et al*. Enchondromas in children: Imaging appearance with pathological correlation. Skeletal Radiology. 2012;41(10):1223-1229. DOI: 10.1007/s00256-012-1377-6
4. Ambulgekar DRK, Bhamare DS, Somani DR. Large enchondroma of metacarpal bone without pathological fracture treated with marginal resection and reconstruction with iliac crest graft: A case report. International Journal of Orthopaedics Sciences. 2020;6(2):354-356. DOI:10.22271/ortho.2020.v6.i2f.2067
5. Sollaci C, Araújo GCS De. Enchondromas of the Hand: A 20-year Experience. Revista Brasileira de Ortopedia. 2019;54(6):714-720. DOI:10.1055/s-0039-1697970
6. Pandey S. Large Enchondroma of Metacarpal Bone with Pathological Fracture Treated with Marginal Resection and Reconstruction with Iliac Crest Graft: A Rare Case Report. Journal of College of Medical Sciences-Nepal. 2019;15(2):150-152. DOI: 10.3126/jcmsn.v15i2.23275
7. Riester S, Ramaesch R, Wenger D, van Wijnen A, Kakar S. Predicting Fracture Risk for Enchondroma of the Hand. Hand. 2016;11(2):206-210. DOI: 10.1177/1558944715626931
8. Bickels JCDA. Local Adjuvant Substances Following Curettage of Bone Tumors. Journal of Bone and Joint Surgery (American Volume). 2019;0(0):1-11. DOI:10.1080/03056244.2018.1546429
9. Mohler DG, Chiu R, McCall DA, Avedian RS. Curettage and cryosurgery for low-grade cartilage tumors is associated with low recurrence and high function. Clinical Orthopaedics and Related Research. 2010;468(10):2765-2773. DOI: 10.1007/s11999-010-1445-y
10. Tang C, Chan M, Fok M, Fung B. Current management of hand enchondroma: A review. Hand Surgery. 2015;20(1):191-195. DOI: 10.1142/S0218810415300028

How to Cite This Article
Sahithya B, Aprilya D. A long-standing large enchondroma of 5th metacarpal associated with deformity of the adjacent metacarpal bone: A rare case report. Int. J Case Rep Orthop. 2024;6(1):04-08.