# Enchondroma of the proximal phalanx of the foot: A case report and mini-review of the literature

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**Abstract.** An enchondroma is a benign cartilaginous tumor of the bone. The present study describes a case of a symptomatic enchondroma in the proximal phalanx of the left second toe. A 20-year-old male patient presented with a painless, progressively enlarging mass on the left second toe, which had resulted in gradual deformity over the past 6 months. Over the past 2 weeks, the lump has caused him pain while wearing shoes, hindering his daily activities. An X-ray revealed a radiolucent, eccentric lesion with loss of the medial bone cortex. Magnetic resonance imaging revealed an expansile osteolytic lesion breaching three cortices with no soft tissue involvement, consistent with an enchondroma. The surgical removal of the lesion and autologous iliac bone grafting were performed, and the histopathological analysis of the tumor revealed hypercellular sheets of chondrocytes encased by mature bone trabeculae without cortical destruction, pre-existing lamellar bone entrapment, or soft tissue invasion. Following a 6-month follow-up period, the patient was in a good condition, without any recurrence or complications. In addition, the present study performed a review of 19 cases of enchondroma in different foot locations. The management of these cases included excision with bone grafting, curettage with or without grafting, total phalangectomy and amputation. There were no cases of recurrence. On the whole, in symptomatic patients with enchondroma, the surgical removal of the lesion and autologous iliac bone grafting may result in a good outcome.

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*Key words:* enchondromas, foot, toe, chondrosarcoma, autologous bone grafting, hyaline cartilage

## Introduction

Enchondromas are benign tumors composed of mature hyaline cartilage found within the medullary cavity of bones. They originate from cartilage cell nests that separate from the central growth plate during the development process. This leads to the abnormal accumulation of mature hypertrophic hyaline cartilage that fails to undergo normal resorption or ossification (1-3).

Enchondromas vary in prevalence depending on their location within the human body. They constitute ~90% of all bone tumors in the hand. By contrast, foot enchondromas are much less frequent and primarily affect the phalanges and metatarsal bones. While these tumors can occur at any age, they typically manifest between the first and fourth decades of life, affecting both sexes equally (1,4,5).

Enchondromas in the small bones of the feet are typically asymptomatic and are often discovered incidentally during routine X-ray examinations. When they become symptomatic, patients may present with pain primarily due to increased pressure from the growth of the lesion, which can deform the cortex of the affected bone or from fractures within the lesion, including pathological or stress fractures. Patients may also report a gradual enlargement of the affected digit (1,2,6).

Enchondromas in different locations of the foot have been documented in the literature (1-5,7-12). The presented study reports a case of symptomatic enchondroma in the proximal phalanx of the left second toe. The case report has been prepared in accordance with the CaReL guidelines, and referenced studies were reviewed to ensure the exclusion of non-peer-reviewed data (13,14).

## Case report

*Patient information.* A 20-year-old male patient presented to Smart Health Tower (Sulaymaniyah, Iraq) with a painless, slow-growing lump on his left second toe, which had gradually caused deformity over the past 6 months. Over the past 2 weeks, he began experiencing pain while wearing shoes, hindering his

daily activities. He reported no history of foot trauma, chronic medical conditions, or prior surgical interventions.

*Clinical findings*. Upon a physical examination, a hard mass on the medial border of the proximal phalanx of the left second toe was found, causing a lateral deviation of the toe (Fig. 1). The mass was firmly fixed to the underlying bone, with no tenderness upon palpation or signs of local inflammation. The metatarsophalangeal and interphalangeal joints exhibited a good range of motion and no sensory deficits in the toe.

*Diagnostic approach*. An X-ray revealed a radiolucent, eccentric lesion within the proximal phalanx of the left second toe, with the loss of the medial bone cortex. Magnetic resonance imaging (MRI) revealed an expansile osteolytic lesion breaching three cortices of the proximal phalanx of the left second toe, with no involvement of soft tissue, consistent with enchondroma (Fig. 2).

*Therapeutic intervention*. Surgery was decided under spinal anesthesia. The left limb was prepped and draped, and the ipsilateral iliac crest was prepared for harvesting a bone graft. Using a thigh tourniquet following the exsanguination of the leg, a longitudinal dorsal approach incision was made over the center of the toe, extending from the metatarsophalangeal joint to the proximal interphalangeal joint. The extensor digitorum longus tendon was exposed and retracted laterally, revealing the proximal phalanx with its lesion. After separating and protecting the neurovascular structures, the lesion was resected using a no. 15 surgical blade. The tumor bed was subsequently cleaned using Rongeur forceps and a bone curette, followed by shaving the tumor bed with a small, high-speed burr (Fig. 3). Subsequently, after measuring the osseous defect, a tricortical iliac bone autograft was harvested from the ipsilateral site and placed into the defect. A 1.6-mm Kirschner wire was inserted antegrade through the harvested graft into the middle and distal phalanges and then retrograded back into the metatarsal head (Fig. 4). Layered closure was performed for both wounds. A histopathological examination was performed by the laboratory at Smart Health Tower, as follows: The analysis was performed on 5-$\mu$m-thick, paraffin-embedded sections. The sections were fixed in 10% neutral-buffered formalin at room temperature for 24 h, and the sections were then stained with hematoxylin and eosin (H&E; Bio Optica Co.) for 1-2 min at room temperature. The sections were then examined under a light microscope (Leica Microsystems GmbH). The histopathological analysis of the tumor revealed hypercellular sheets of chondrocytes encased by mature bone trabeculae without cortical destruction, pre-existing lamellar bone entrapment, or soft tissue invasion. The tumor had a partly lobular configuration with varying cellularity. Chondrocytes within lacunae in a myxoid and hyaline matrix had elongated and stellate nuclei with fine chromatin. There was no multinucleation, significant pleomorphism, mitotic activity, or necrosis (Fig. 5).

*Follow-up*. Post-operatively, the patient was placed in heel-touch weight bearing. The Kirschner wire was removed after 6 weeks, and the bone showed good signs of healing. At 6 months postoperatively, the patient demonstrated excellent toe range of motion, was pain-free and maintained proper alignment (Fig. 6). An X-ray revealed a complete union of the bone with the graft.

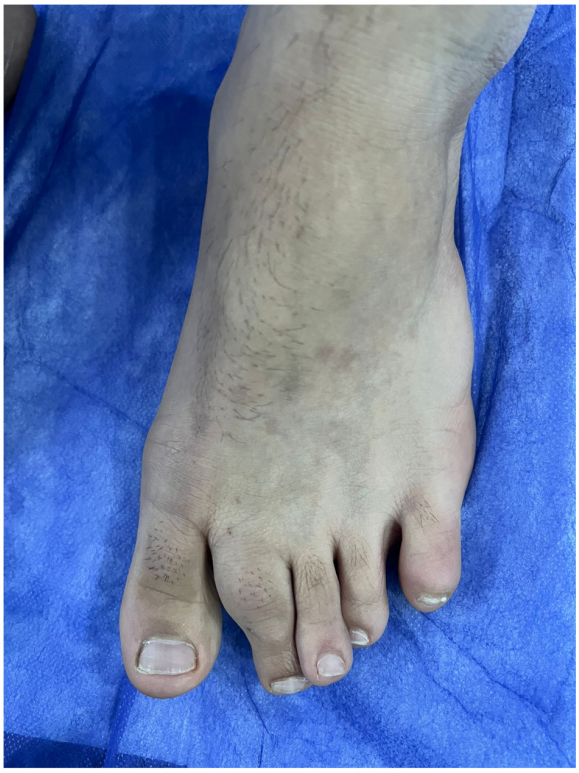

Figure 1. Pre-operative image demonstrating a swelling at the base of the second toe of the left foot with lateral displacement.

## Discussion

Enchondromas grow gradually without infiltrating nearby tissues or spreading to distant body parts (3,15). In reviewing 19 cases of foot enchondroma (Table I), only two instances were found where an enchondroma transformed into chondrosarcoma, leading to amputation (11,12).

Enchondroma primarily manifests in the phalanges of the hand, although it can also occur in the phalanges and metatarsal bones of the foot (1). Among the reviewed cases, the most commonly affected toe was the first toe (38%), followed by the third toe (28.6%), the second toe (19%), the fourth toe (4.8%), the calcaneus (4.8%) and the cuneiform bone (4.8%). Among the 17 cases with 19 lesions located on the toes, the most common sites were the proximal phalanx (57.9%), followed by the distal phalanx (21%), metatarsal (15.8%) and middle phalanx (5.3%). In the present case, the lesion was located on the proximal phalanx of the second toe.

The tumor is typically found as a solitary lesion, known as a solitary enchondroma (8). However, they can also appear as multiple lesions, as seen in conditions such as multiple enchondromatosis (Ollier disease) and multiple enchondromatosis associated with hemangiomas (Maffucci syndrome) (3). Enchondromatosis is linked to somatic mutations in the isocitrate dehydrogenase (IDH)1 and IDH2 genes. These mutations produce defective IDH, an enzyme in the tricarboxylic acid cycle that converts isocitrate to α-ketoglutarate. The mutated enzyme facilitates the reduction of α-ketoglutarate to

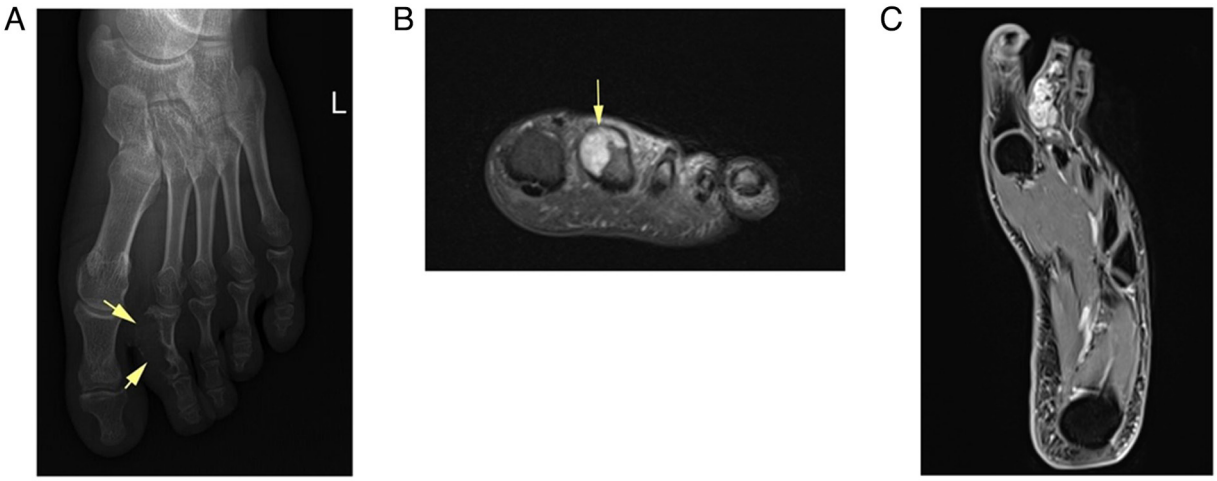


Figure 2. Pre-operative radiological imaging of the bone lesion: (A) Anteroposterior plain radiograph reveals an eccentric, expansible radiolucent lesion within the proximal phalanx of the left second toe, with cortical bone destruction (arrow). (B) Coronal T2-weighted image (T2WI) of the foot illustrating an eccentric phalangeal lesion with hyperintense signal intensity (arrow) and no associated soft tissue component. (C) Axial post-contrast fat-suppressed T1-weighted image (T1WI) exhibits strong enhancement with no apparent invasion of surrounding structures.

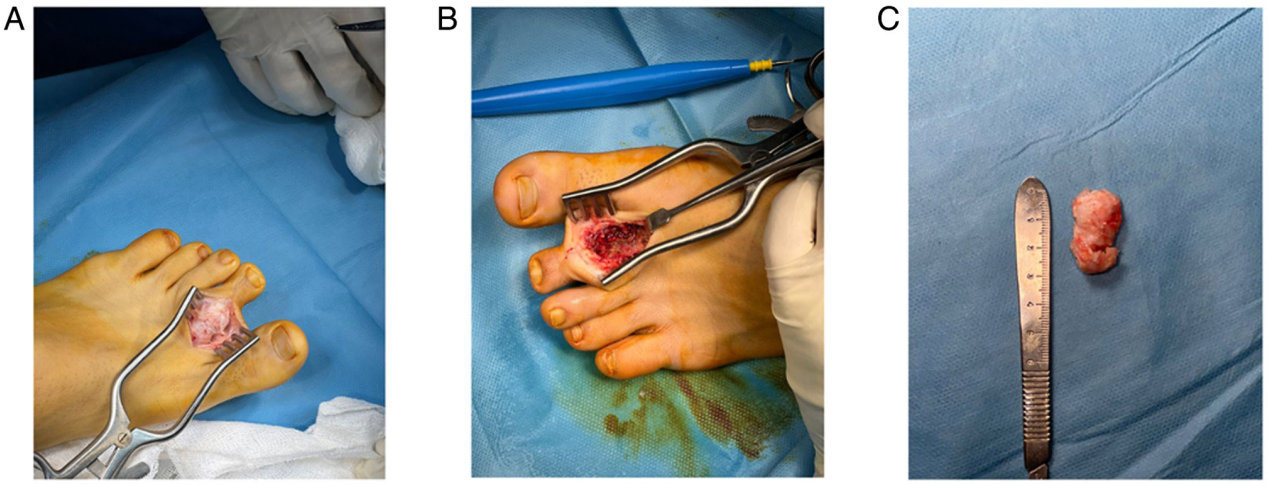


Figure 3. Intraoperative photographs of (A) the proximal phalanx before resection of the enchondroma, (B) after resection of the enchondroma, (C) and the resected enchondroma.

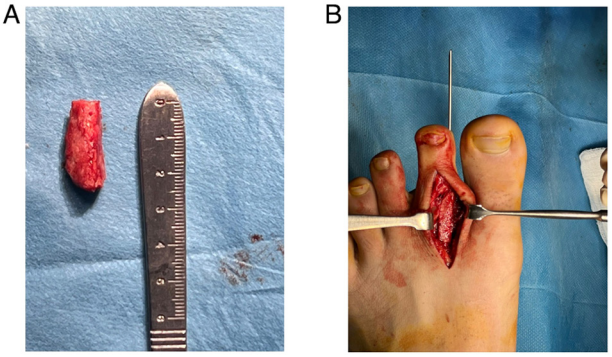


Figure 4. Image of (A) an autologous tricortical bone graft harvested from ipsilateral iliac bone and (B) the inserted autologous bone graft.

the oncometabolite D-2-hydroxyglutarate (D-2-HG), and by competitively inhibiting α-ketoglutarate-dependent enzymes, D-2-HG results in hypermethylation of DNA and modification of histones. These processes encourage the development of cartilaginous tumors and disrupt the normal osteogenic differentiation of mesenchymal stem cells (6). All reviewed cases in the present study involved solitary enchondromas, apart from 1 patient with multiple lesions in the distal phalanx, proximal phalanx, and metatarsal bones of the first toe of the same foot (5).

Foot enchondromas can occur at any age, although they are most commonly observed in patients between the first and fourth decades of life (9). The youngest case reported among the cases reviewed herein involved a 16-year-old female, while the oldest was an 86-year-old female (1,11). In accordance with the study by De Yoe and Rockett (4), in the present study, a review of the literature revealed no sex predilection in the prevalence of foot enchondromas, with 10 males and 9 females. The case in the present study was a 20-year-old male.

Generally, enchondromas remain asymptomatic for extended periods of time. When symptoms do manifest, they may include pain, swelling, or deformity of the affected

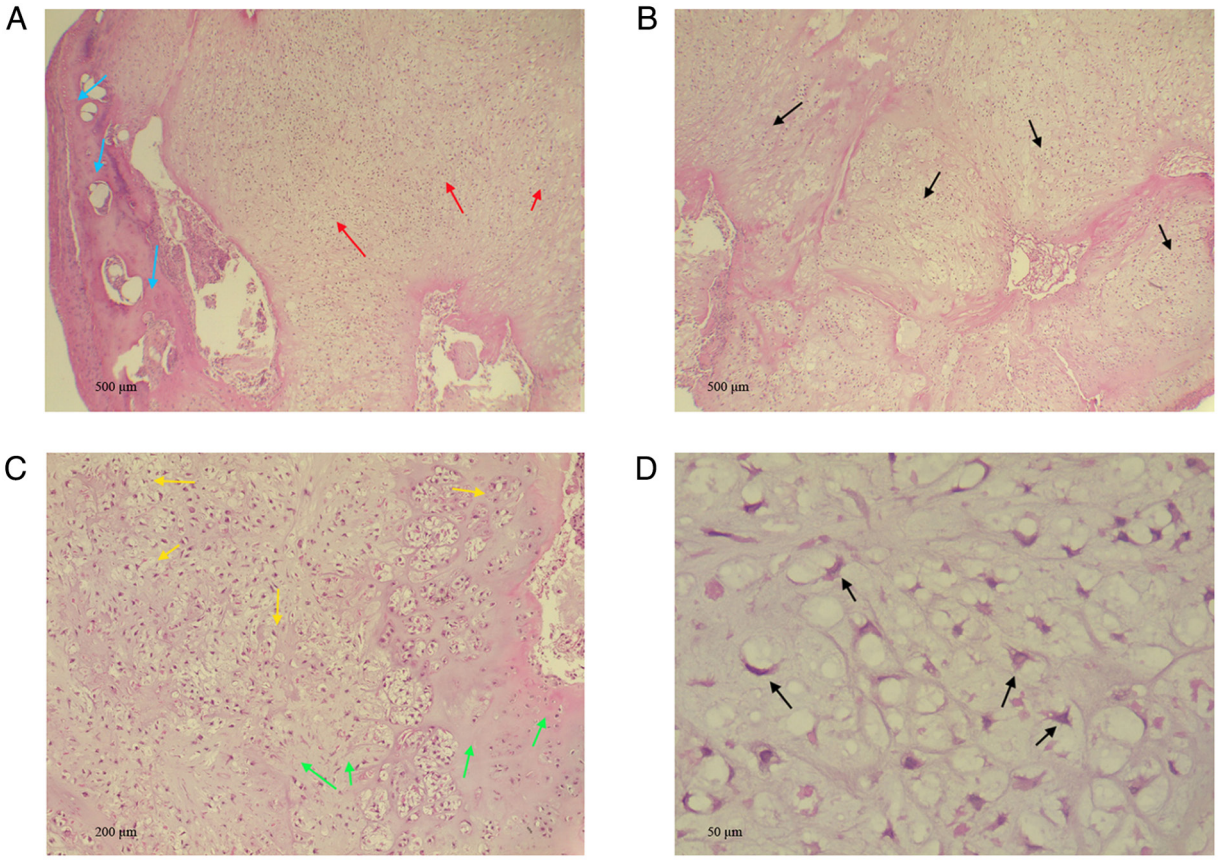


Figure 5. (A) hypercellular sheets of chondrocytes (red arrows) encased by mature bone trabeculae (blue arrows) at the periphery without cortical destruction, entrapment of pre-existing lamellar bone, or soft tissue invasion. (B) The tumor has a partly lobular configuration with hypocellular and hypercellular areas of chondrocytes (black arrows). (C) The chondrocytes are located within lacunae (yellow arrows) lying within a myxoid and hyaline matrix (green arrow). (D) The chondrocytes lie within lacunae and have elongated and stellate nuclei with fine chromatin (black arrows). There is no multinucleation, significant pleomorphism, mitotic activity, or necrosis. The images demonstrate hematoxylin and eosin staining. Original magnification: (A and B) x40, (C) x100, and (D) x400.

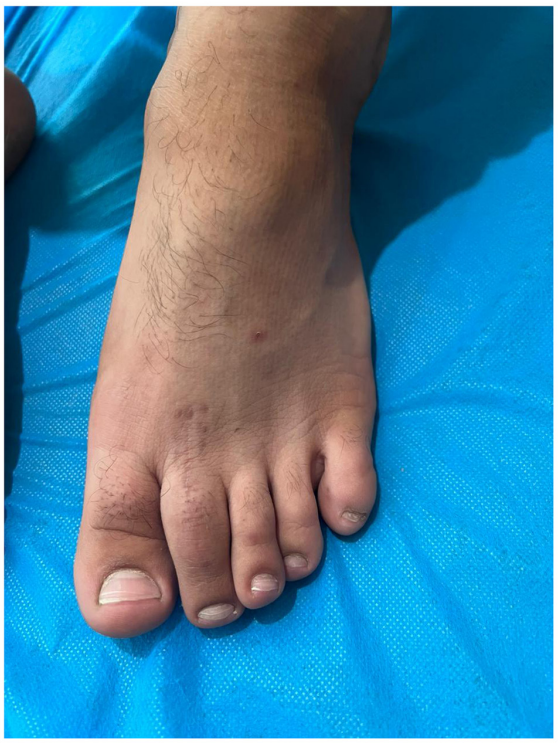

Figure 6. Clinical image of the operated foot 6 months post-operatively.

bone (6). The primary source of pain often stems from elevated pressure caused by the cortical expansion of the lesion, pathological fracture, or malignant conversion of the lesion (1,3,4). The lesion in the majority of the reviewed cases caused pain (76.2%) and food inversion in 1 case (4.8%), while the remaining lesions were diagnosed incidentally on foot radiographs (19%). Additionally, 6 out of the 19 cases had pathological fractures.

Various modalities and methods are available to healthcare professionals for detecting and diagnosing enchondromas. The primary and most crucial method remains a comprehensive clinical history (8). After assessing the clinical presentation, plain radiographs are the preferred initial diagnostic imaging modality (9). Radiographically, enchondromas appear as lytic lesions with clearly defined borders and variable degrees of stippled or punctate calcifications, typically without the involvement of the surrounding soft tissues (3). In general, computed tomography scans and MRIs can provide additional detail about the lesion, particularly when there is rapid growth or suspicion of soft tissue involvement (4). In the identified literature, plain radiography was the most commonly employed diagnostic modality, showing lytic lesions. In the case presented herein, the radiograph revealed a radiolucent, eccentric lesion with loss of the medial bone cortex, and the MRI revealed an expansile osteolytic lesion breaching three cortices of the affected phalanx.

Table I. Review of 19 cases of enchondroma of the foot .

| First author, year of publication | No. of cases | Age (years) | Sex | Site of the lesion | Tumor size (cm) | Presenting symptom | Medical history | Physical examination | U/S | X-ray | CT | MRI | Management | Histopathology | Follow-up (months) | Recurrence | (Refs.) |
|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|
| Komurcu, 2015 | 1 | 53 | M | Calcaneus | 2.1 | Pain | Negative | Tenderness and swelling | NA | A lesion with calcification and peripheral sclerosis | Cortical thinning adjacent to the lesion | A lesion with hyperintense signaling on T2-weighted sequences and peripheral heterogeneous enhancement pattern on contrast-enhanced T1-weighted sequences | Curettage of the lesion and grafting | Lobules of different sizes of hyaline cartilage tissue and chondrocytes without atypia inside hyaline cartilage | NA | No | (1) |
| Lui, 2014 | 1 | 51 | F | 1st toe PP | NA | Pain | Negative | Swelling | NA | Fracture with an enchondroma | NA | NA | Endoscopic curettage and bone grafting | NA | 31 | No | (2) |
| Edwards, 2020 | 1 | 27 | M | 4th toe PP | 1 | Pain | Controlled type I diabetes mellitus and depression | Moderate edema and tenderness | Mild soft tissue edema | Lucent lesion | NA | Expansile lesion breaching the inferior and lateral cortical bone | Resection and tibial bone graft | A well circum-scribed tumor composed of lobules of hyaline cartilage encased in bone and covered by fibrous tissue. | 12 | No | (3) |
| De Yoe, 1999 | 2 | 43 | F | 3rd toe MT | 0.7 | Pain | Controlled hyper-tension | Tenderness | NA | A lytic ovoid lesion with disrupted lateral cortex of the metatarsal | NA | NA | Surgical excision with autogenous bone grafting | Confirmed enchondroma (no detail available) | 10 | No | (4) |
| | | 45 | M | Cuneiform | 0.4 | Pain | Negative | Tenderness | NA | Lytic lesion | NA | NA | Curettage with autogenous bone graft | Confirmed enchondroma (no detail available) | 12 | No | (4) |
| Goto, 2004 | 8 | 48 | F | 3rd toe PP | 0.8 | Pain | Negative | NA | NA | Eccentrically located radiolucent area, and pathological fracture | NA | NA | Simple curettage without bone grafting | Confirmed enchondroma (no detail available) | 13 | No | (5) |

Table I. Continued.

| First author, year of publication | No. of cases | Age (years) | Sex | Site of the lesion | Tumor size (cm) | Presenting symptom | Medical history | Physical examination | U/S | X-ray | CT | MRI | Management | Histopathology | Follow-up (months) | Recurrence | (Refs.) |
|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|
| | | 25 | F | 1st toe DP | 2.1 | Incidental finding | Negative | NA | NA | NA | NA | NA | Simple curettage without bone grafting | Confirmed enchondroma (no detail available) | 30 | No | (5) |
| | | 32 | F | 3rd toe PP | 0.6 | Pain | Negative | NA | NA | Pathological fracture | NA | NA | Simple curettage without bone grafting | Confirmed enchondroma (no detail available) | 6 | No | (5) |
| | | 39 | M | 1st toe DP | 0.54 | Pain | Negative | NA | NA | Pathological fracture | NA | NA | Simple curettage without bone grafting | Confirmed enchondroma (no detail available) | 41 | No | (5) |
| | | 23 | F | 2nd toe PP | 0.33 | Pain | Negative | NA | NA | Pathological fracture | NA | NA | Simple curettage without bone grafting | Confirmed enchondroma (no detail available) | 34 | No | (5) |
| | | 50 | M | 3rd toe PP | 0.24 | Incidental finding | Negative | NA | NA | NA | NA | NA | Simple curettage without bone grafting | Confirmed enchondroma (no detail available) | 13 | No | (5) |
| | | 31 | M | 2nd toe MP | 2.1 | Pain | Negative | NA | NA | Radiolucent area with some calcifications and ballooning of the cortex | NA | NA | Simple curettage without bone grafting | Confirmed enchondroma (no detail available) | 18 | No | (5) |
| | | 31 | M | 1st toe DP | 0.2 | Pain | Negative | NA | NA | NA | NA | NA | Simple curettage without bone grafting | Confirmed enchondroma (no detail available) | 12 | No | (5) |
| | | | | 1st toe PP | 4.7 | Incidental finding | Negative | NA | NA | NA | NA | NA | Simple curettage without bone grafting | Confirmed enchondroma (no detail available) | 12 | No | (5) |
| | | | | 1st toe MT | 4.6 | Incidental finding | Negative | NA | NA | NA | NA | NA | Simple curettage without bone grafting | Confirmed enchondroma (no detail available) | 12 | No | (5) |
| Stess, 1995 | 1 | 36 | M | 2nd toe PP | 2.2 | Pain | NA | Swelling and tenderness | NA | An osseous metaphyseal lesion with diminished density | NA | NA | Total phalangectomy of the proximal phalanx with syndactylization of the second and third toes. | NA | 12 | No | (7) |

Table I. Continued.

| First author, year of publication | No. of cases | Age (years) | Sex | Site of the lesion | Tumor size (cm) | Presenting symptom | Medical history | Physical examination | U/S | X-ray | CT | MRI | Management | Histopathology | Follow-up (months) | Recurrence | (Refs.) |
|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|
| Remba, 2021 | 1 | 30 | M | 1st toe MT | NA | Inversion of the foot | Negative | Edema and pain during mobility and walking | NA | Tumor and soft tissue edema | NA | Heterogeneous intramedullary lesion, hypo-intense in T1 and hyper-intense in T2 | Curettage of the lesion with bone grafting | Confirmed enchondroma (no detail available) | 6 | No | (8) |
| Patel, 2022 | 1 | 17 | M | 1st toe PP | 2.8 | Pain | Negative | Swelling | NA | Lytic lesions, scalloping of the cortex, and whorls of calcification | NA | Hyperintense mass on FS-PD, and soft tissue edema and swelling on T1-weighted sequences | Surgical excision with bone grafting | Confirmed enchondroma (no detail available) | 24 | No | (9) |
| Alhosain, 2020 | 1 | 16 | F | 2nd toe PP | 1.3 | Pain | Negative | Hard, round mass fixed to the bone | NA | Well circumscribed, lucent, a central medullary lesion with cortical expansion and thinning | NA | NA | Curettage and subsequent bone grafting | Lobules of hyaline cartilage encased by normal bone and fibrous tissue | 3 | No | (10) |
| Mahajan, 2009 | 1 | 86 | F | 3rd toe PP | NA | Pain | Negative | Movement restriction of the metatarsophalangeal joint, swelling | NA | Expansile swelling contained within the cortex without malignant change | NA | Cystic change, consistent with benign lesion. Six months later, it turned into malignancy | Ray amputation due to malignant change into chondrosarcoma | Chondrosarcoma grade II, mainly and III focally | NA | NA | (11) |
| Koak, 2000 | 1 | 33 | F | 3rd toe DP | NA | Pain and swelling | Foot trauma | The toe had a drumstick appearance with enlargement | NA | Expanding lytic lesion with a fracture | NA | NA | Partial amputation of the toe | The features indicated low-grade (Grade 1) chondrosarcoma | NA | NA | (12) |

M, male; F, female; PP, proximal phalanx; MP, middle phalanx; DP, distal phalanx; MT, metatarsal; NA, not available; U/S, ultrasound; CT, computed tomography; MRI, magnetic resonance imaging.

Radiological findings suggesting a lesion are not always conclusive for diagnosing an enchondroma; therefore, a histopathological analysis is mandatory (9). Distinguishing between benign and malignant lesions presents a significant challenge. All available tissues need to be thoroughly examined. Enchondromas can be visually identified as bluish, semi-translucent masses of hyaline cartilage arranged in lobular patterns. Microscopically, enchondromas display small chondrocytes within lacunar spaces characterized by round, uniform nuclei resembling those found in hyaline cartilage. Some enchondromas may also exhibit areas of ossification within the cartilage matrix (4,6). The nuclei of these cells are generally regular, showing a few mitotic activities. Enchondromas located near the bone cortex, including those in the hands, may exhibit increased cellularity and atypia while remaining benign (6). Histologically, enchondroma and low-grade (well-differentiated) chondrosarcoma can appear deceptively similar, although they can be distinguished by their tissue architecture and patterns of invasion: enchondromas typically display multiple discrete nodules of hyaline cartilage separated by normal marrow elements and are often surrounded by lamellar host bone conforming to the shape of the cartilage lobules; by contrast, low-grade chondrosarcomas tend to form a single confluent mass of cartilage that permeates the marrow, 'trapping' host lamellar bone, infiltrating the Haversian systems or marrow fat, and often exhibiting fibrous bands between peripheral cartilage lobules. These features reflect its malignant nature. Additional supportive indicators include the presence of lobulation patterns and fibrous tissue formation around the lesion, which have been shown to correlate with malignant recurrence in follow-up studies, whereas enchondromas generally remain benign (16,17). The histopathological analysis of the specimen in the case in the present study revealed hypercellular sheets of chondrocytes encased by mature bone trabeculae at the periphery without cortical destruction, entrapment of pre-existing lamellar bone, or soft tissue invasion. There was no multinucleation, significant pleomorphism, mitotic activity, or necrosis.

The treatment of enchondroma can range from close monitoring and regular follow-up, particularly for small, asymptomatic lesions, to complete surgical removal with bone grafting for larger, symptomatic lesions (2). Surgery is recommended for patients experiencing ongoing symptoms and lesions >2 cm, as they pose a significant risk of pathological fractures. It includes complete tumor removal with or without bone grafting, as well as curettage followed by bone grafting. Goto *et al* (5) reported that the radiographic and functional outcomes of simple curettage without bone grafting are comparable to those of curettage with autologous bone grafting. They also highlighted several advantages of performing curettage without bone grafting for foot enchondromas: i) It eliminates the pain and discomfort associated with the bone donor site; ii) the procedure can be performed on an outpatient basis; and iii) the shorter operation time provides economic benefits and decreases the risk of infection (5). However, Edwards and Kingsford (3) reported that curettage alone is not recommended due to a high rate of non-union (67%). They found that the surgical option involving bone grafting is more suitable, as it offers a recovery period similar to that of curettage alone, despite the additional wound that requires healing (3). Patel *et al* (9) also reported that the latest management option for foot enchondroma involves using an autologous bone graft from the iliac crest, which can be either cortical or cancellous. Chun *et al* (18) reported a series of 20 cases in which all patients underwent tumor curettage followed by bone grafting. No recurrences or post-operative complications were observed during the 24-month follow-up period (18). In addition, Futani *et al* (19) conducted a retrospective cohort study comparing osteoscopic and conventional open surgery for foot enchondromas. A total of 17 patients underwent osteoscopic surgery, and 8 patients underwent open surgery. They reported that functional recovery was significantly improved in the osteoscopic group at 1 and 2 weeks post-operatively, though no differences were noted after 1 month. Additionally, osteoscopic surgery was associated with fewer complications (12% vs. 50%) and no recurrences in either group (19). Among the reviewed cases, 8 (42.1%) cases were managed with excision or curettage combined with bone grafting, another 8 (42.1%) cases were managed with simple curettage without bone grafting, 1 (5.3%) case was managed with total phalangectomy, and 2 cases underwent partial and ray amputation (10.5%). All reported cases had favorable surgical outcomes without any recurrence. In the case in the present study, the tumor was surgically removed, and a tricortical iliac bone autograft from the ipsilateral site was placed into the defect. The surgical outcome was favorable, with no signs of recurrence or complications. The unretrievable last follow-up X-ray image, which revealed the complete union of the bone with the graft, may be a limitation of the present case report.

In conclusion, enchondroma is a benign tumor rarely found in the foot. For symptomatic cases, the surgical removal of the lesion combined with autologous iliac bone grafting may result in favorable outcomes.

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**Availability of data and materials**

The data generated in the present study may be requested from the corresponding author.

**Authors' contributions**

FHK and AKG were major contributors to the conception of the study, as well as to the literature search for related studies. HOA, MGH and SOK were involved in the literature review, in the conception and design of the study and in the writing of the manuscript. RJR, AMA and HAS were involved in the literature review, in the design and conception of the study, the critical revision of the manuscript, and the processing of the figures and table. AAM was the radiologist who performed the assessment of the case. RMA and AMA were the pathologists who performed the diagnosis of the case. FHK and AKG confirm the authenticity of all the raw data. All authors have read and approved the final manuscript.

## Ethics approval and consent to participate

Written informed consent was obtained from the patient for his participation in the present study.

## Patient consent for publication

Written informed consent was obtained from the patient for the publication of the present case report and any accompanying images.

## Competing interests

The authors declare that they have no competing interests.

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