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CASE REPORT

A rare mass with atypical localization: Heterotopic ossification associated with flexor hallucis longus

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ABSTRACT

Heterotopic ossification (HO), characterized by the formation of ectopic bone, is a benign mass observed in soft tissues. Depending on its location, it can cause symptoms beyond compression, such as mechanical blockage when associated with joints, leading to limitations in joint movements. In the majority of cases, involvement of the hip and elbow joints is common, while HO can sometimes be observed in atypical locations. Trauma, head injury, and spinal cord injuries are well-recognized risk factors for HO development. However, on rare occasions, in non-traumatic cases are identified without any known risk factors. Herein, we present a rare non-traumatic HO case associated with the flexor hallucis longus (FHL) tendon in a 58-year-old female patient. She complained of pain under the first toe of her right foot while wearing shoes for a year, and a mass was detected on the plantar surface of the foot along with limitation of movement in the first metatarsophalangeal joint. Further examinations revealed that the identified mass was a mature HO lesion. Surgical treatment was performed, and during one-year follow-up, the pain subsided, and joint movements returned to normal, resulting in a satisfactory outcome. In conclusion, although many cases of HO are associated with traumatic injuries, it can sometimes be idiopathic, as in our case, and rarely it is accompanied tendon such as FHL in the foot.

Keywords: Foot, flexor hallucis longus, heterotopic ossification, plantar pain.

Heterotopic ossification (HO) is the formation of ectopic lamellar bone in soft tissues.[1] Although the exact cause of ectopic bone formation has not been fully understood yet, the formation of HO has been observed following total hip arthroplasty, acetabular and elbow fracture surgery, electrocution and burn injuries, and traumatic brain injury or spinal cord injury. It is more frequent in males compared to females.[2] The disability incurred as a result of HO is quite variable. Cases of HO, characterized by clinical§ manifestations such as chronic pain, swelling, and restricted movement associated with periarticular localization, can sometimes present differently, even in healthy individuals without an underlying surgical history or trauma. Hip, elbow, and arms are more frequently affected sites, as they are subjected to high-risk injuries. Although it is a clinically and histologically distinct entity, diagnosis can be very challenging when it occurs in a particularly rare location such as the foot and ankle. Heterotopic ossification can be confused with malignant lesions, such as osteosarcoma and soft tissue sarcoma.[3]

In this article, we present a case of an ectopic bone mass associated with the flexor hallucis longus (FHL) tendon in a female patient without any predisposing factors.

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CASE REPORT

A 58-year-old female patient was admitted to our clinic with a complaint of pain in the plantar region of her right foot. She reported that her pain gradually increased, particularly in the previous year, and that she had difficulty in wearing shoes. The patient had no history of trauma or surgery regarding her right lower extremity and foot. She had no known systemic disease other than a hemangioma in the liver. Apart from hemangioma in the liver, the patient had no systemic diseases. Her pain increased with activity and walking. In the foot examination, there was tenderness and mass on the plantar surface of the foot between the first tarsometatarsal joint and the metatarsophalangeal joint. The mass shifted slightly with big toe flexion and extension. First metatarsophalangeal joint flexion was decreased both actively and passively. However, active or passive extension or flexion of the patient's metatarsophalangeal joint did not cause pain. She was neurovascularly intact in her lower extremity.

In the patient's foot anteroposterior and lateral radiographs, an osseous lesion, which was compatible with HO, was detected in the soft tissue mass area, extending proximally from under the sesamoid bones along the FHL tendon (Figure 1a). Magnetic resonance imaging (MRI) and computed tomography (CT) were recommended to the patient to confirm the diagnosis and plan the treatment. In the CT examination, all images demonstrated amorphous ossification along the long axis of the FHL adjacent to the first metatarsophalangeal joint (Figure 1b-e). The MRI images showed an ossification along the long axis of the medial border of the FHL tendon. These images also demonstrated FHL tendon itself as a hypointense structure (Figure 2a-e). A surgical intervention was recommended to the patient. Subsequently, she underwent primary resection of the heterotopic ossified mass.

The patient was placed in the supine position under spinal anesthesia. A 5-cm incision was made in the forefoot starting from the medial of

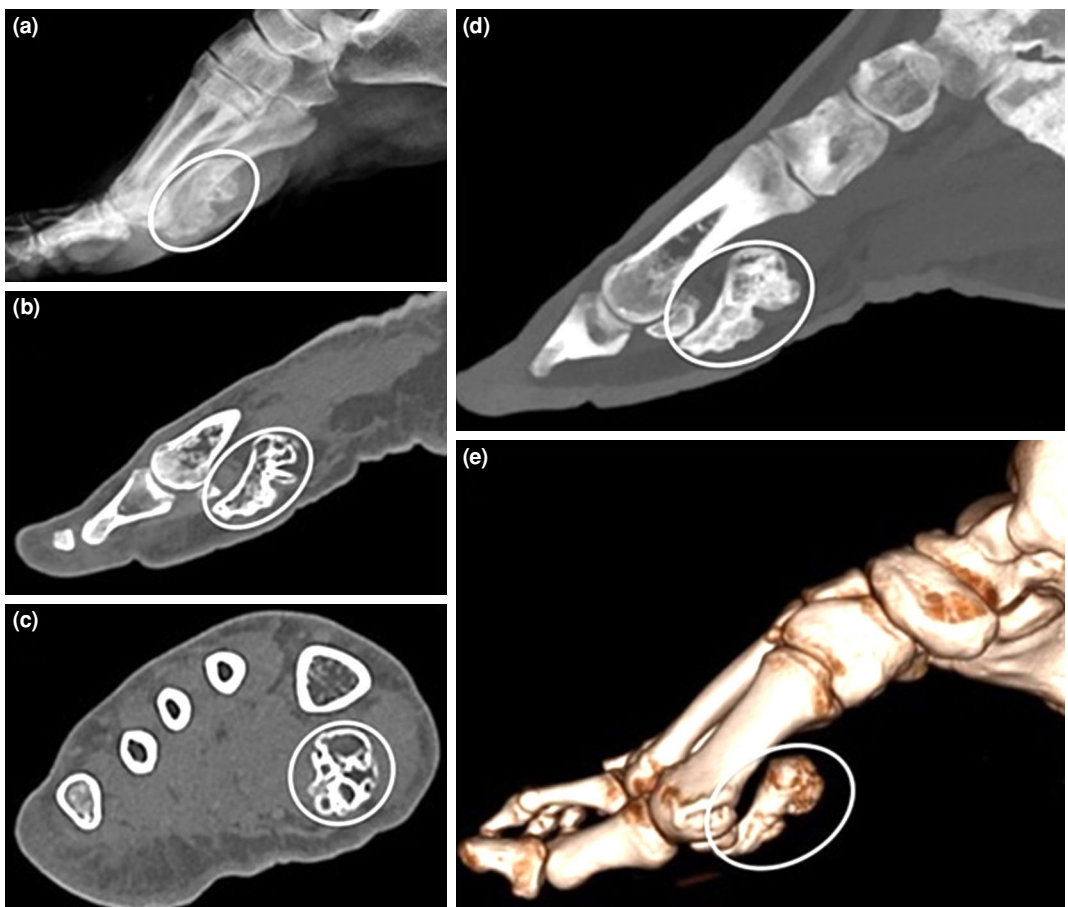


FIGURE 1. Lateral radiograph **(a)** and three-dimensional volume rendering computed tomography image **(b-e)** showing amorphous ossification adjacent to the first metatarsophalangeal joint (circles).

the metatarsophalangeal joint to proximal that centered over the mass. Soft tissue dissection was carefully undertaken between the abductor hallucis and flexor hallucis brevis muscles until exposure of the FHL tendon sheath. The FHL tendon sheath was linearly incised along the length of the lesion (Figure 3). On opening the sheath, a heterotopic bone mass measuring 29×16 mm was found (Figure 4). The bone mass was not embedded within the FHL tendon but adhered. The heterotopic mass was dissected and sharply removed, with clear margins (Figure 3). There was no defect that required tubularization or repair in the tendon. The wound was irrigated and closed. A sterile dressing was applied. The patient was placed in a right lower extremity posterior splint and instructed to remain non-weightbearing for three weeks until the follow-up examination. At the three-week follow-up visit, the right lower extremity splint was removed, and the patient was allowed full weightbearing. The postoperative follow-up visits at 6 and 12 weeks revealed complete pain resolution of her pain with no loss of range of motion in the first metatarsophalangeal joint. The pathology report of the excised tissue was compatible with HO. The pathology report, which included mature bone trabeculae in the excised tissue and a bone marrow area infiltrated by fatty tissue, was consistent with HO (Figure 5). At her one-year follow-up visit, she was completely pain free with a normal first metatarsophalangeal join range of motion and had no pain with wearing shoes.



FIGURE 2. Sagittal T2-weighted fat-saturated **(a)**, axial proton-density (PD) fat-saturated **(b)** and coronal T1-weighted **(c-e)** MRI images showing an ossification along the long axis of the medial border of the FHL tendon (solid arrows). MRI images also showing the FHL tendon itself as a hypo-intense structure (dotted arrows).
MRI: Magnetic resonance imaging; FHL: Flexor hallucis longus.

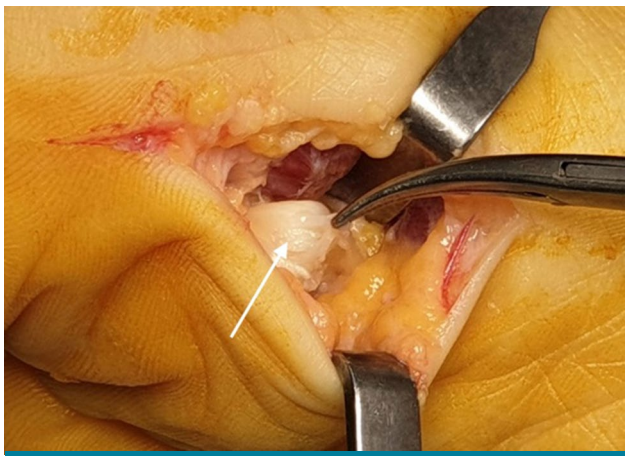


FIGURE 3. Intraoperative image, the flexor hallucis longus tendon sheath held with a clamp under which the heterotopic ossification was excised, and the shiny structure underneath is the flexor hallucis longus tendon (arrow).



FIGURE 4. The excised heterotopic ossified mass (29×16 mm).



FIGURE 5. Photomicrograph of the specimen demonstrating the mature bone trabeculae and the bone marrow space infiltrated by the fatty tissue (H&E, ×100).

DISCUSSION

The fundamental pathology in the formation of HO involves damage to muscle and soft tissues. The activation of osteoprogenitor cells into osteoblasts within the affected area leads to abnormal bone formation through processes such as cartilage formation and endochondral ossification.[4] The origin of HO, extensively studied in terms of its pathogenesis, is typically categorized into two types: acquired and congenital HO. Fibrodysplasia ossificans progressiva is the term used for congenital HO. Trauma is the primary attributed cause in acquired HO.[5] In such cases, the hip and elbow joints prominent out significantly, and conditions such as extended immobilization may potentially elevate the risk of HO.[6] In our case, there were no comorbidities, no hereditary factors, and no predisposing processes for the development of HO. Furthermore, it presented an atypical localization, specifically in the forefoot. Although microtraumas resulting from foot compression could be considered a potential cause, the etiology of this case was essentially idiopathic.

The hip and elbow joints are the most commonly affected areas.[3] Heterotopic ossification commonly develops after surgeries performed for acetabular and elbow fractures. Open fractures, extensive surgical dissection, concomitant head trauma, and prolonged intensive care unit stays may increase the risk of HO development.[7-9] Beyond these well-recognized conditions familiar to orthopedic surgeons, the occurrence of HO in the foot and ankle joints is comparatively infrequent. Ossifications observed in the Achilles tendon are common in the ankle. The most common reported causes of Achilles tendon ossification are previous trauma and surgery; however, diabetes, seronegative arthropathies, metabolic diseases, and a few other conditions have also been reported as predisposing factors.[10] In contemporary times, as ankle arthroplasty and outcomes are disseminated, instances of HO following ankle prosthesis surgeries are being reported and discussed.[11] However, HO associated with tendons in the foot is relatively rare. These few case reports in the literature consist of HO cases involving extensor hallucis longus and peroneus brevis tendons.[12,13] Unlike these two cases, there was no history of trauma in our case. In a case of HO in the peroneus brevis tendon in a pediatric patient, surgery was performed after six weeks of conservative treatment. On the other hand, in case of HO in the extensor hallucis longus tendon, surgical excision was performed without conservative

treatment. The reason for this is to reach a definitive diagnosis, since HO was not mature. Although a differential diagnosis can be easily made in mature lesions, as in our case, mechanical symptoms, pain, and conditions that affect the comfort of life such as wearing shoes, caused us to prefer surgical treatment. In our case, in contrast to these two cases, the mass was located in the plantar region of the foot, which is a factor that must be considered, as it contributes to the increase in symptoms with walking.

Plain radiographs are commonly the first imaging study used to detect HO. In HO, the characteristics of the lesion are characterized by fibroblastic tissue in the early stage and may not show any findings on radiographs, as ossification is not seen.[13] Technetium-99 bone scans are an alternative in this setting. While the advantage of bone scans is the ability to detect HO earlier than radiographs, bone scans are of a limited value in differentiating inflammation from early HO. Although radiography, CT scan, and MRI have low specificity in the early stages, these investigations in later stages can be diagnostic by showing normal lamellar bony architecture within a bony mass 3. In our case, ectopic ossification was evident in the diagnostic radiographs, particularly in the lateral foot radiograph. A CT scan was conducted to assess the boundaries and dimensions of the ectopic ossification tissue, clearly revealing the bony mass. The CT facilitates preoperative planning by improving three-dimensional visualization of HO in relation to important anatomic landmarks. It is worth noting that routine MRI is not advised in cases of HO.[13,14] It may not be required for patients scheduled for conservative treatment or when the diagnosis is unequivocal. However, in areas where mass lesions are infrequent and for patients undergoing planned surgery, MRI becomes valuable to assess the possible relationship between the mass and surrounding soft tissues.[3,14] We examined our case with MRI to evaluate the associate of the mass with neurovascular structures and FHL tendon and whether it would require reconstructive intervention.

The treatment for HO should be personalized based on the specific characteristics of the lesion and the patient's clinical condition. Surgical resection in the early stages of a lesion with incomplete ossification and mineralization may lead to the recurrence of the mass. However, when mature ectopic ossification causes clinical symptoms, surgical treatment becomes necessary, involving the removal of the mass through resection. In our case, a palpable mass was identified on the plantar aspect of the foot during examination.[15] The patient had pain while wearing shoes and limitations in dorsiflexion and plantar flexion of the first toe for a year. Notably, CT and MRI examinations clearly revealed the presence of a HO. Consequently, conservative treatments were not deemed suitable, and we opted for the surgical resection of the mass causing mechanical symptoms.

In conclusion, our case was atypical and non-traumatic, characterized by dimensions, approximately 3 cm, that was considered relatively small. We believe that sharing information about regions where HO is rarely observed, and demonstrating cases similar to ours that are entirely idiopathic, is valuable. The forefoot is a region where classical malignant and benign masses are infrequent. Differential diagnosis should also consider ectopic ossification in such cases.

Patient Consent for Publication: A written informed consent was obtained from the patient.

Data Sharing Statement: The data that support the findings of this study are available from the corresponding author upon reasonable request.

Author Contributions: Conception, design, data collection and/or processing: M.K.; Conception, supervision, analysis: M.K.; Writing, literature review: H.U.Ö.; Materials, design: Z.M.Ö.

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