Intraosseous epidermoid inclusion cysts are rare benign bone lesions that occur most commonly in the skull and in the distal phalanges of the fingers. Herein we report a case of an intraosseous epidermoid inclusion cyst occurring in the distal phalanx of the left hallux. Only six occurrences of this lesion have been described in the foot. This patient's presentation, with active drainage (initially appearing as purulent discharge from an acutely tender ingrown hallux nail) and a known inoculation event accompanied by severe peripheral vascular disease, make this case unique. (J Am Podiatr Med Assoc 100(2):133-137, 2010)

**Intraosseous Epidermoid Inclusion Cyst Presenting as a Paronychia of the Hallux**

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In a comprehensive review of 67,000 pedal pathology specimens, Berlin¹ found that subcutaneous epidermoid cysts were the fifth most common soft-tissue tumors in the foot. Of the 2,136 pedal soft-tissue epidermal inclusion cysts, females were affected twice as often as males, with no predilection for either foot. However, as of this reporting, only six intraosseous epidermoid inclusion cysts (IEpCs) of the foot have been documented in the medical literature, with all but one occurring in the distal phalanx of the hallux.²⁻⁷ In the hand, the reported prevalence of IEpCs favors males 2:1, and IEpCs occur most commonly in the left first and third digits.²⁻⁴,⁵,⁸ In the skull, these lesions exhibit no sex preference.⁴

The role of trauma in the development of soft-tissue epidermal inclusion cysts was first established experimentally in 1885 by Masse when he recreated an epidermal inclusion cyst in rats after implantation of epidermis into subcutaneous tissue.⁹ McGraw et al.¹⁰ reported a 45% incidence of antecedent trauma with phalangeal epidermal inclusion cysts, often decades removed from the onset of symptoms. The presumed role of trauma in the development of phalangeal cysts may anecdotally support the observed prevalence of females with foot lesions (caused by footwear)¹¹ and males with nondominant hand lesions (caused by a hammer or other crushing injuries).⁹ In 1930, Harris¹² was the first to suggest that IEpCs of the phalanges were also most likely secondary to traumatic implantation of epidermis into bone, with blunt trauma, amputation, and bony erosion caused by an enlarging subcutaneous cyst extending into bone being subsequently advanced by other authors as likely mechanisms.²⁻⁴,⁵,¹³,¹⁴ Byers et al.⁸ noted that two-thirds of IEpCs were preceded by a recognized trauma. In contrast, intraosseous skull lesions, which are unlike their often tender phalangeal counterparts, are generally attributed to faulty embryogenesis.²⁻⁵,⁸ The following case report describes a combination of traumatic etiologies, namely, a subungual epidermal inclusion cyst iatrogenically converted to an IEpC of the distal phalanx of the hallux caused by a simple probe-to-bone test with a sterile wooden applicator.⁵,⁸,¹⁴

**Case Report**

A 78-year-old man followed closely by the White River Junction Veterans Affairs Podiatry Service for care of mycotic nails and hammer toes in the setting of moderately severe left lower-extremity peripheral vascular disease presented in routine follow-up with increasing pain in his left hallux without precipitating trauma. These symptoms exceeded his baseline mechanical and vascular complaints and were recurring and remitting during the 8 months before the acute presentation. In that time, no overt signs of infection, gangrene, or other objective clinical findings to explain his discomfort were identified. The vascular
surgery service remained active in the patient's care and elected to defer intervention while he remained clinically stable. The patient had several relevant medical issues, which included a 70 pack-year history of smoking, self-reported daily alcohol consumption (three drinks/day), and a well-established history of peripheral vascular disease that required a series of four separate surgical interventions for critical ischemia of his right lower extremity, and subsequently was managed with cilostazol. Despite persistent claudication symptoms, the results of his noninvasive vascular studies remained stable across time, indicating mild right lower-extremity disease and moderate-to-severe disease of the left lower extremity. A 3-month course of oral fluconazole had also been prescribed 5 years earlier, with some improvement of his tender mycotic nails noted.

At this time, however, the patient presented acutely with unremitting severe pain of 2 days' duration. The severity of his pain prevented him from sleeping, and only mild relief was achieved by placing his leg in a dependent position. The toe was noted to be ruborous, with some fluctuance of the proximal nail fold. Purulent drainage was expressed and cultured (Fig. 1), formal vascular studies were repeated, and a total nail avulsion was performed under local anesthesia. An inconclusive probe-to-bone test was performed, with no hard or gritty end point reached and only a slight increase in resistance to two-finger pressure. Radiographs taken immediately after this examination showed a well-defined tract in the distal phalanx possibly caused by the sterile cotton-tipped applicator used to probe the wound (Fig. 2). No bacterial growth was reported from the cultures, and a toe pressure of 36 mm Hg for the left hallux indicated stable vascular disease in this patient.

At 1-week follow-up, the patient related only temporary relief of his symptoms despite oral antibiosis and local wound care. No tissue loss or gangrene resulting from the nail avulsion was evident at follow-up, so, once again, digital anesthesia was administered, additional drainage was expressed from the hallux, and a smear review was performed that showed copious squamous cells with keratinaceous debris but no visible bacteria or crystals. These findings suggested a tentative diagnosis of an epidermal inclusion cyst of the hallux.

At 2-week follow-up, the severe pain persisted, although no additional drainage was noted from the hallux. Revascularization and amputation of the hallux were discussed as possible options if his pain did not respond to nonoperative care. A third curettage of the digit was performed under anesthetic block, and injectable corticosteroid was infiltrated to address inflammation and pain in this poor operative candidate. At 1-week follow-up, marginal benefit was noted from the injection. Given the severity of his unrelenting pain during the past month, a follow-up radiograph and vascular reconsultation were ordered. The radiograph evidenced significant changes in the distal phalanx (Fig. 3). Given the patient's deteriorating renal function, the vascular surgery service elected to

Figure 1. Initial clinical presentation of the hallux with expressed drainage.

Figure 2. Radiograph with an iatrogenic probe tract seen in the hyperemic soft distal phalanx.
postpone an arteriogram until the patient’s renal function improved or his clinical condition mandated emergent care.

Two months after the onset of his severe persistent pain, the patient once again had expressible drainage from the hallux, and a fourth debridement of the digit was performed. He continued to experience increasing pain, and 2 weeks later he requested revascularization of his left leg as previously outlined. Three months after the onset of acute pain, and 1 year after the initial onset of symptoms, a left lower-extremity multilevel segmental bypass was successfully performed, which greatly ameliorated his toe pain, and 6 weeks postoperatively his hallux pressure was 93 mm Hg. Two months after this procedure, however, the patient once again presented urgently relating a 2-day history of pain and swelling of the left hallux identical to his initial episode. A fifth debridement of the hallux was performed, culture samples were taken, a slide was prepared, and antibiotics were ordered. The slide showed many squamous cells and many crystals from the corticosteroid injected 4 months earlier. Once again, no bacteria grew from the culture. The patient neglected to pick up his antibiotics or to have his blood work done as ordered. Repeated standard radiographs showed no progression of osseous changes in the past 5 months, and the changes remained limited to the distal phalanx of the left hallux. His condition deteriorated quickly, leading to an emergency hospital admission 2 days later for intravenous antibiotic administration and amputation of his left hallux. A transphalangeal amputation of the left hallux with primary closure was performed, and the patient healed uneventfully. Pathologic evaluation confirmed the diagnosis of an intraosseous epidermal cyst (Fig. 4). This procedure offered complete relief of the patient’s pain and provided a functional outcome for the remainder of his life.

Discussion

This case of an IEpC of the hallux presenting as a paronychia in a patient with severe peripheral vascular disease, a known implantation event, a documented timeline, and radiographs demonstrating interval osseous changes offers a unique look at an infrequently recognized condition. In the feet, IEpCs are rare, with all but one of the six previously documented cases involving the terminal phalanx of the hallux. The one reported IEpC of the foot not involving the distal hallux presented 16 months after hammer toe repair of the fourth digit. Histologically, these lesions are similar to those found in the skull, but they are fundamentally different in etiology, sex, age, and pain on presentation. Phalangeal IEpCs typically present in the fourth decade, which is later than skull lesions, and have recognized antecedent trauma. Patients present for medical care more acutely, given the associated pain and disability. Additionally, a presumptive diagnosis was made at the second acute clinical visit by microscopic examination of active drainage from the cyst. This was another unique feature of this case not previously reported with an IEpC, although the cyst may still have been localized to the subungual soft tissue at this time. After a definitive diagnosis by pathologic evaluation, the recommended treatment entails aggressive surgical curettage. Amputation is rarely indicated, although the peculiar circumstances of this case made it the most viable option.

The most common presenting complaint with IEpC of the hands or feet is pain that is often associated with clubbing of the digit. There is a clear consensus in the literature that traumatic implantation is the primary etiology of these cysts, although the inciting event may have occurred decades earlier. Schajowicz et al suggested that the juxtaposition of the proximal nail bed against the periosteum of the proximal phalanx makes this area especially susceptible to IEpCs. Another adjunctive mechanism may be that a subungual cyst in this confined space is prone to erode into the adjacent bone. The cause of the pain so commonly featured in these cases may be multifactorial, including pressure, avascular necrosis,
and a well-circumscribed radiolucent lesion on standard radiography, the diagnosis of an IEpC is rare. The differential diagnosis may include metastatic tumor, psoriasis, sarcoidosis, or gout, but the following radiolucent lesions of the phalanx warrant a higher index of suspicion: chondroma, aneurysmal bone cyst, osteoid osteoma, giant cell tumor, chronic osteomyelitis, and intraosseous ganglion. If cold sensitivity is a dominant feature, a glomus tumor must also be considered.3, 7, 10, 13 In this case, radiographs were initially atypical because the cyst was actively organizing during the course of care.

Histologically, epidermal inclusion cysts and IEpCs are characterized by a wall of squamous epithelium with maturation of the granular cell layer. The cyst contains keratinaceous debris. In contrast to dermoid and pathologic fracture, but the caustic reaction of the soft tissues exposed to the keratinaceous contents of the IEpC was especially remarkable in this case, initially leading to the presumptive diagnosis of hallux paronychia. Previous reports of phalangeal IEpCs demonstrate a highly variable interval between the inciting trauma and the onset of symptoms. There is only one case of foot pain, noted 1 month after trauma, that may have had an onset as precipitous as this case.2

Radiographic features of phalangeal IEpC include expansible radiolucency, absence of trabeculation, thinning cortex, sclerotic margins, and absence of periosteal reactive bone except in response to fracture or rupture of the cyst.2, 4, 5, 8, 10 However, even with clinical presentation of a tender swollen distal phalanx

Figure 4. A, Gross photograph of hallux in a cross section. The white triangle indicates the joint space between the intermediate and destroyed distal phalanges; and black arrow, a keratin-filled cyst, which has destroyed the bony structures of the distal phalanx. B, Intraosseous epidermal inclusion cyst with a squamous wall and keratin flakes adjacent to the joint surface (H&E, ×200). C, The cyst wall with a keratin-filled center adjacent to fibrotic marrow and woven bone (H&E, ×200). D, The nail surface and, deep to the nail, the epidermal inclusion cyst (H&E, ×200).
cysts, epidermal inclusion cysts do not have associated adnexal structures lining the cyst wall. Intraosseous cysts are identical in appearance to soft-tissue epidermal inclusion cysts but are surrounded by bone that may be reactive. This was particularly noted in this case because the cyst had ruptured, and because keratin is very irritating to tissues, it caused marked inflammation with foreign body giant cells.  

Although IEpCs of the feet are rare, patients presenting with pain out of proportion, a remote history of trauma, swelling, radiographic changes as described, and intense localized inflammation of the distal digit should have this lesion included in their differential diagnosis. Timely cultures and microscopic examination of aspirate or biopsy will provide the information needed for an expedient and definitive diagnosis. With this information, the surgeon may proceed with confidence in performing a localized curettage and can anticipate an excellent functional outcome.

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**References**

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