



An Intraosseous Epidermoid Cyst That Originated from the Nail Bed of Great Toe with Concurrent Joint Infection: A Case Report

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We report on a rare case of an intraosseous epidermoid cyst in the distal phalanx of the great toe with concurrent infection in a 71-year-old woman with diabetes mellitus. The lesion was initially considered simple infectious arthritis and concomitant osteomyelitis in a patient with diabetes. However, after surgery, an intraosseous epidermoid cyst originating from the nail bed and involving the articular surface of the distal phalanx was detected. The epidermoid cyst may have contributed to the infectious arthritis in the interphalangeal joint. The lesion was treated via mass excision, arthrotomy, debridement, and intravenous antibiotics.

Key Words: Intraosseous epidermoid cyst, Great toe, Interphalangeal joint infection

Although epidermoid inclusion cysts are common and easy to diagnose in the soft tissues, intraosseous epidermal cysts (IECs) are rare lesions that have been reported to occur in the hand, and skull, and even less frequently in the foot.¹⁾ A congenital etiology with intraosseous inclusion of the embryonic epithelial tissue has been proposed, although post-traumatic or iatrogenic events remain the most prevalent hypotheses.²⁾

It is difficult to diagnose IECs, and histopathological examination is needed to confirm the diagnosis. The differential diagnoses include metastatic tumors, psoriasis, gout, aneurysmal bone cyst, giant cell tumor, and chronic osteomyelitis.¹⁾ The typical radiographic feature is a well-defined radiolucent or osteolytic lesion that causes bone expansion and cortical thinning, and bony sclerosis or reaction is typically absent. However, radiographic diagnosis of this lesion remains challenging, especially when atypical radiographic features are seen due to infection or other concomitant conditions. Well circumscribed predominantly hypoechoic mass

can be seen on the ultrasound. However, if it is small, it can mimic a cyst. Larger masses can be a little heterogeneous. Also, magnetic resonance imaging (MRI) shows variability of signal intensity depending on different cases, and times.³⁾

We report a unique case of IEC of the hallux after informed consent from the patient, which is associated with the nail bed and concomitant septic arthritis in the interphalangeal (IP) joint.

CASE REPORT

A 71-year-old woman was being followed-up at the Department of Rheumatology for Takayasu arteritis, and was referred to our foot and ankle clinic (a tertiary referral center). She complained of having a fever since 10 days along with recently developed pain and swelling of the right great toe. Passive motion of the great toe's IP joint was painful and we observed tenderness in that area. There was no discharge or skin wound, and she did not recollect any surgery or trauma that involved her right great toe.

Blood tests revealed C-reactive protein levels of 6.28 mg/dL, erythrocyte sedimentation rate levels of 55 mm/hr. Foot radiography revealed a localized osteolytic lesion at the base of the right great toe's distal phalanx, although the lesion's margin was neither sclerotic nor identifiable. The medial side of the subchondral

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sclerotic bone in the proximal phalanx was violated, which led to suspicion of joint involvement or pathologic fracture (Fig. 1). Enhanced MRI revealed 1.8-cm irregularly contoured nodular lesion at the base of the great toe's distal phalanx. The lesion involved the distal phalanx with bony erosion and the adjacent flexor tendon, and exhibited low signal intensity without enhancement on T1- and T2-weighted images. We also observed synovial thickening with enhancement and a small amount of joint effusion at first IP joint, suggestive of septic arthritis, and the poorly defined low-intensity T1 signal with enhancement at the first proximal phalanx



Figure 1. Preoperative anteroposterior radiograph reveals a radiolucent and osteolytic lesion at the base of the great toe's distal phalanx, which is more prominent in the medial aspect.

was consistent with associated osteomyelitis (Fig. 2).

The patient subsequently underwent surgical incision and drainage with biopsy of the lesion under regional anesthesia. We performed arthrotomy of the IP joint via a dorsal approach, and observed a yellowish purulent material that was collected for bacterial culture. During the irrigation and debridement, additional dirty creamy-brown material gushed out through the joint in the subeponychial and subungual area. The nail plate was extracted for complete debridement, and we observed that a well-defined whitish cystic mass was attached to the nail plate. The nail bed exhibited a defect due to the mass that extended to the base of the

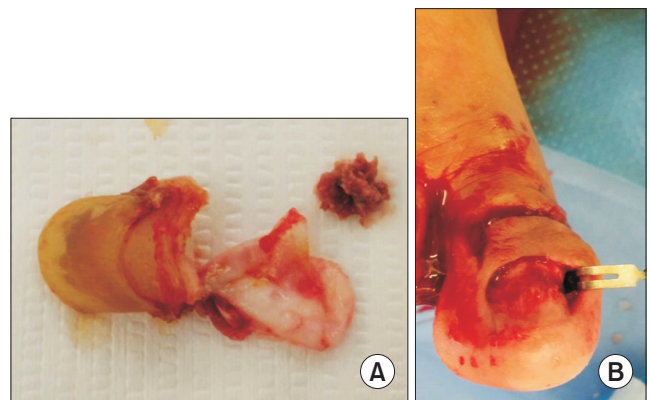


Figure 3. Intraoperative findings. (A) The extracted nail plate and attached intraosseous epidermoid cyst. A dirty creamy-brown material filled the cyst. (B) The defect in the medial side of the nail bed due to the intraosseous epidermoid cyst. Mass excision and curettage was performed throughout the defect, and a silastic drain was implanted.

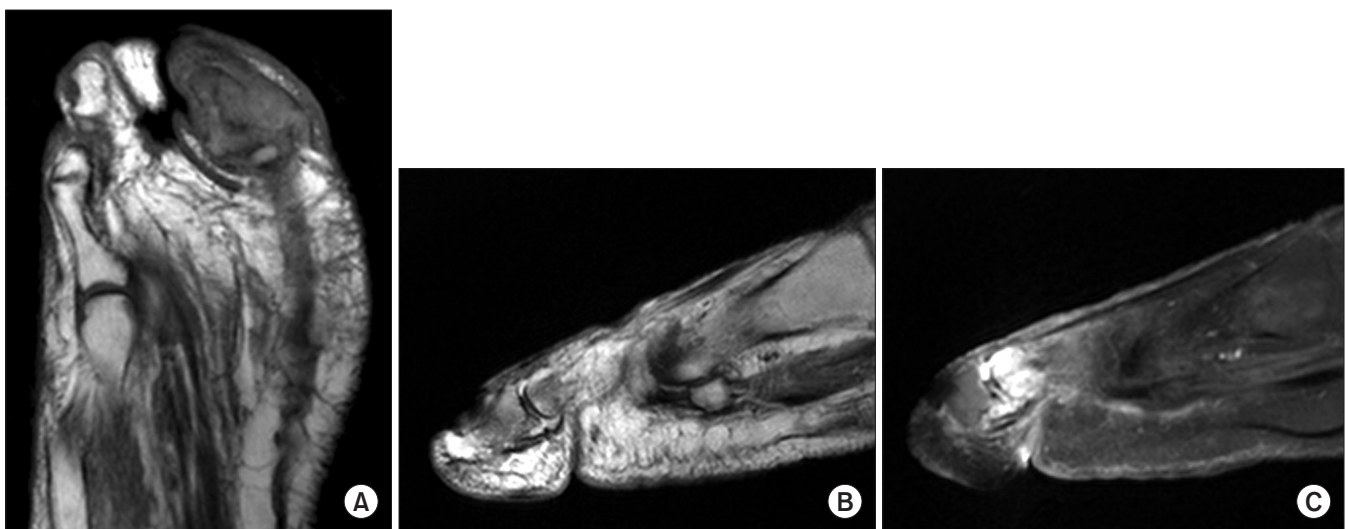


Figure 2. Preoperative magnetic resonance imaging findings. (A) A coronal T1-weighted image reveals an irregularly contoured nodular lesion at the distal phalanx. This lesion involves the great toe's distal phalanx with bony erosion and the adjacent flexor tendon. (B) A sagittal T2-weighted image reveals low signal intensity at the proximal and distal phalanx. (C) An enhanced T1-weighted image reveals low signal intensity at the great toe's proximal phalanx, synovial thickening with enhancement, and a small amount of joint effusion at the interphalangeal joint.

distal phalanx and the medial side of the joint surface. We performed meticulous debridement with complete mass excision.

The excised mass was histopathologically examined, and the biopsy findings were consistent with an IEC (Fig. 3, 4). Bacterial culture also revealed the growth of coagulase-negative staphylococcus which were sensitive to first generation cephalosporin. Intravenous cefazolin was administered for 4 weeks after the operation. The surgical wound was healed well without recurrent infection during the 2 months' follow-up.

DISCUSSION

Epidermoid cysts of the soft tissues were first reported by Masse in 1885.⁴⁾ These cysts are common benign soft tissue tumors, with Berlin⁵⁾ reporting that epidermoid cysts were the fifth most common soft-tissue benign tumors in the foot and Mercuri and Casadei⁶⁾ reporting that they were the second most common benign foot tumor. Unlike epidermoid cysts in the subcutaneous tissue, IECs are relatively rare, and only a few cases in the skull and finger have been reported; IECs in the toes are extremely rare. Therefore, the overall incidence or prevalence of IECs is unknown.

Our IEC case is unique among the rare subset of IECs that involve the toes, because this IEC appears to have originated in the nail bed, extended into the bone, and violated the articular surface. Therefore, this IEC might have formed a connection between the IP joint and the nail plate's external surface (where normal flora reside), which may have a possibility to contribute to the joint

infection, even though the medical status of our patient including hypertension, diabetes mellitus, and renal disease can also be a risk factor of infection. Based on the patient's original complaints, we initially suspected septic arthritis with concomitant osteomyelitis in a patient with diabetes. However, during the operation, we unexpectedly observed an IEC with concomitant septic arthritis of the IP joint. This condition is extremely rare, and is difficult to diagnose preoperatively.

The first report of IECs in the digits was provided by Harris⁷⁾ in 1930, although the pathophysiology of IECs is remains unknown. Congenital, traumatic, or iatrogenic etiologies have been suggested, and a congenital etiology is often associated with bony inclusion of the embryonic epithelial tissue during the developmental process.²⁾ A traumatic event is the most popular hypothesis regarding the etiology of IECs, and several studies suggest that the pathophysiology of IECs is directly related to traumatic implantation of epidermal tissue into the bone. Cases of IECs with an iatrogenic origin are usually related to a previous surgical procedure. In one case, a subungual epidermoid inclusion cyst had converted into an IEC in the great toe, owing to bone testing with a probe.⁸⁾

One report has described IEC with concomitant paronychia in the great toe.⁸⁾ In that case, the patient underwent multiple operations and the great toe was ultimately amputated because of multiple recurrences and sudden deterioration in the patient's general condition. In the present case, our patient achieved appropriate healing after mass excision, incision, debridement, and antibiotic treatment. However, the postoperative follow-up was relatively short, and we must carefully monitor the patient for recurrence of the lesion.

Although the preoperative radiographic findings favored infectious arthritis with concomitant osteomyelitis, the MRI findings suggested that the lesion should be histopathologically differentiated from other mass lesions, including enchondroma, giant cell tumor of the tendon sheath, and aneurysmal bone cyst. Also, IECs in the digits are commonly located at the distal aspect of the distal phalanx, but the IEC in the present case was located in the proximal aspect.²⁾ Unfortunately, we were unable to reach a definitive diagnosis of an IEC with concurrent infection until our intraoperative visual examination and histopathological confirmation.

In the present report, we described a unique case of IEC of the hallux, which was associated with the nail bed and concomitant usual septic arthritis in the IP joint. Before surgery, this lesion was presumed to be septic arthritis and concurrent osteomyelitis of the distal phalanx in a patient with diabetes mellitus; however, it was

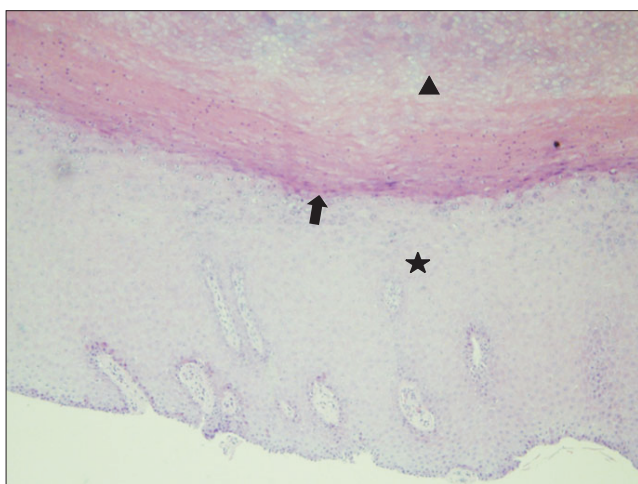


Figure 4. Histopathological findings (H&E stain, ×100). The intraosseous epidermoid cyst consists of the cyst wall (asterisk), granular layer (arrow), and copious keratin material (arrowhead). The cyst wall is lined with squamous cell epithelium and contains some skin appendages.

confirmed to be related with IEC with attached nail bed intraoperatively. Therefore we performed not only arthrotomy and debridement but also including mass excision and nail bed removal. It is important that surgeons should consider unexpected extensive surgical procedures and several complications including wound healing problem particularly in patients with diabetic foot.

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