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CASE SERIES

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Intraosseous epidermoid cyst of distal phalanx—Report of two cases

Dong Hwan Kim[1] | **Jung Ho Noh**[2]

[1]Department of Orthopaedic Surgery, Kangwon National University Hospital, Chuncheon-si, South Korea

[2]Department of Orthopaedic Surgery, Kangwon National University School of Medicine, Chuncheon-si, South Korea

Correspondence
Jung Ho Noh, Department of Orthopaedic Surgery, Kangwon National University School of Medicine, 1 Gangwondaehak-gil, Chuncheon-si, Gangwon-do 24341, South Korea.
Email: bestknee@gmail.com

Key Clinical Message

Intraosseous epidermoid cysts in this report were related to a previous injury. A careful history taking is necessary to make a correct impression on the lesion. Definite diagnosis is made by histologic study.

Abstract

This report presents two cases of intraosseous epidermoid cyst of the finger treated with curettage and autogenous bone graft. Radiographs showed a radiolucent lesion with a sclerotic margin. Histologic examination revealed a cystic wall consisting of stratified squamous epithelial cells and keratinized material in the cyst, consistent with an epidermoid cyst.

KEYWORDS

intraosseous epidermoid cyst, phalanx

1 | INTRODUCTION

Epidermoid cyst often occurs in subcutaneous soft tissues. However, it rarely occurs within the bone, presenting as a radiolucent lytic bone lesion or a pseudotumor. The lesion may occur anywhere on the body. It is known to affect the skull and phalanx most frequently.[1] It can also involve the maxilla, mandible, temporomandibular joint, vertebra, tibia, and femur. Intraosseous epidermoid cyst is not easy to be diagnosed radiographically.[2] It requires biopsy and histopathologic evaluation.[2,3] Once diagnosed, this benign tumor can be treated by curettage and sometimes bone graft.[4] Adequate curettage and removal of the capsule are required to prevent recurrence.[1] The origin of this disease has not yet been definitively established, and various theories have been proposed, including previous traumatic origin, congenital occurrence, and soft tissue penetration.[5–7] In this paper, we report two cases of intraosseous epidermal cysts in the phalanx of the finger, both of which had a documented history of trauma in the affected area.

2 | CASE REPORT

2.1 | Case 1

A 34-year-old woman presented with pain on the right ring finger after striking against the corner of a table 4 days before. She had a history of a blunt trauma on the same finger about 30 years ago. However, there was no open wound. She had no symptom until she got impulse 4 days before. She had no other past medical history. Clinical examination revealed a mildly swollen distal phalanx of the right ring finger with clubbing-like bulging of the nail without an external wound (Figure 1). Her sensation, circulation, and motor power were normal.

There was tenderness on distal phalanx without erythema. Radiographs on the visiting day showed a radiolucent lesion sized 10*7*5 mm in the distal phalanx without septum. It had a sclerotic margin. Radiographs also showed bone expansion and cortical defect on the dorsal side (Figure 2). Initial differential diagnoses included giant cell tumor, enchondroma, bone cyst, and chronic osteomyelitis. Curettage, excisional biopsy, and bone graft were considered. During surgery, the approach was done dorsally where the cortical defect existed. Nail was removed and a longitudinal incision was done on the nail bed. Dorsal cortex was defective and cystic wall was just below the nail bed. When the cyst was incised, about 0.5 cc of creamy yellowish white material was released and a thin wall of the cyst was easily peeled from the cavity (Figure 3). Curettage of the lesion was performed. Autogenous iliac cancellous bone was grafted and aluminum splint was applied (Figure 4). The patient was discharged without complications. Histopathologic

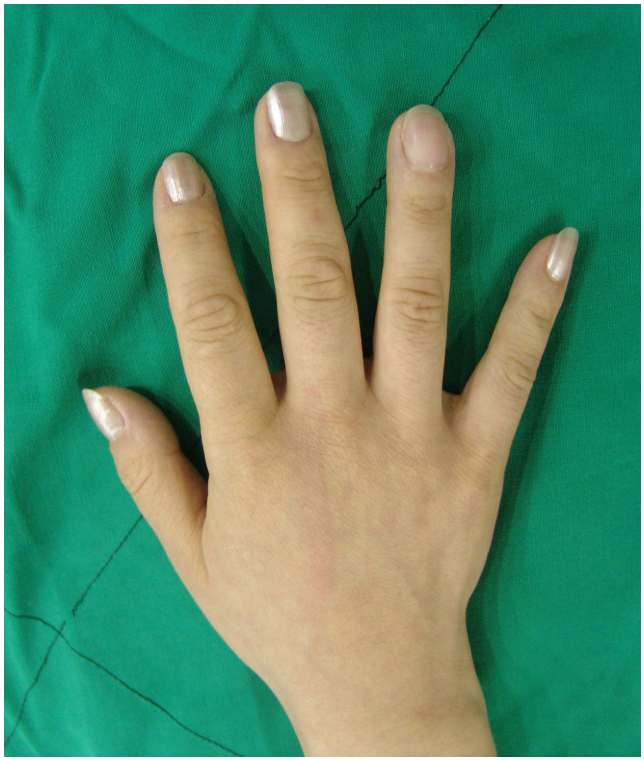

FIGURE 1 Preoperative photograph showing a clubbing-like right ring finger.

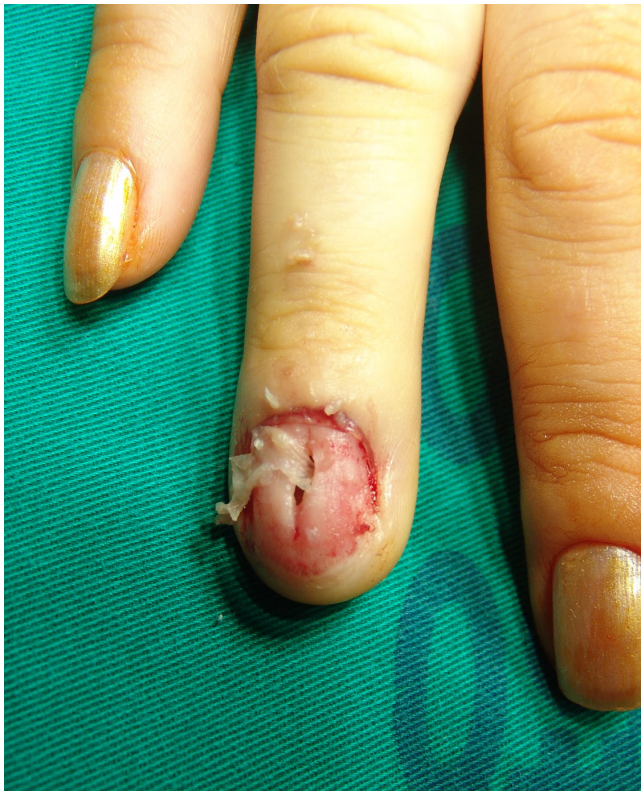

FIGURE 3 Longitudinal incision was done on the nail bed and mass was excised. The cystic wall could be seen.

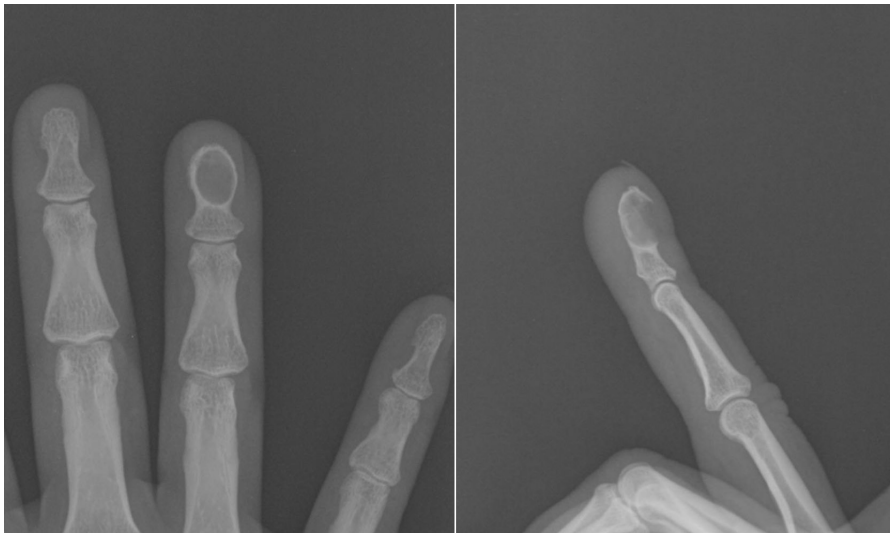

FIGURE 2 Preoperative radiograph showing a radiolucent lesion with cortical defect and sclerotic rim of the right ring finger.

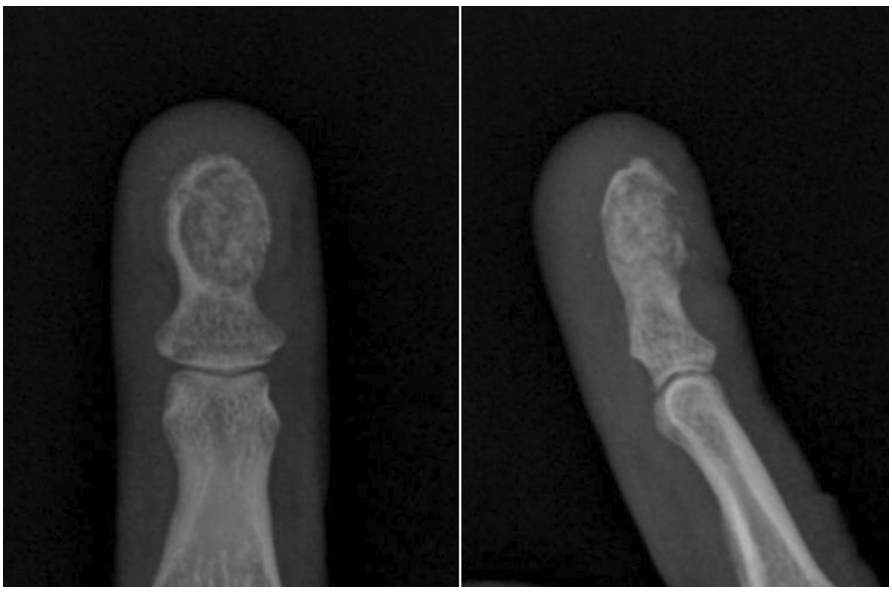

FIGURE 4 Postoperative radiograph. Autogenous iliac cancellous bone was grafted after the mass was completely excised.

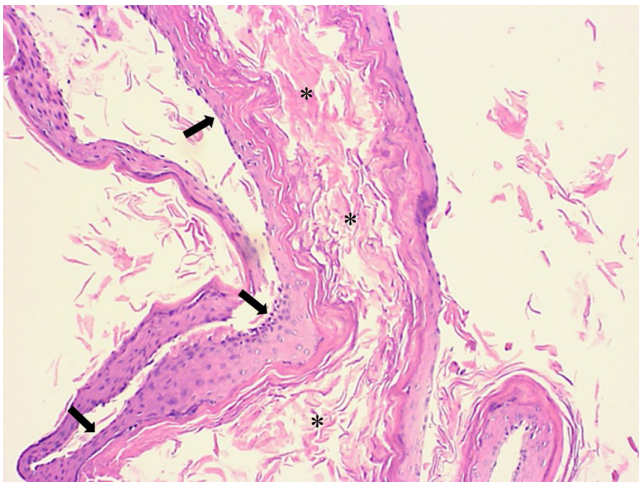

FIGURE 5 Histopathologic appearance of the mass. The cystic wall was composed of stratified squamous cells (arrows) and the cavity was full of keratinized material (asterisks) (H&E, ×100).

examination revealed that the wall of the cyst was composed of stratified squamous cells. The cavity was filled with laminated keratinous material, consistent with an intraosseous epidermoid cyst (Figure 5).

Four years after the surgery, the patient remained symptom-free with normal-looking nail (Figure 6). Radiography at one-year follow-up revealed complete healing of the bone defect (Figure 7).

2.2 | Case 2

A 38-year-old woman presented with a 10-day history of tenderness on the nail of left thumb. Physical examination revealed tenderness on lunula of the thumb nail. There was no swelling, redness, or external wound. Her

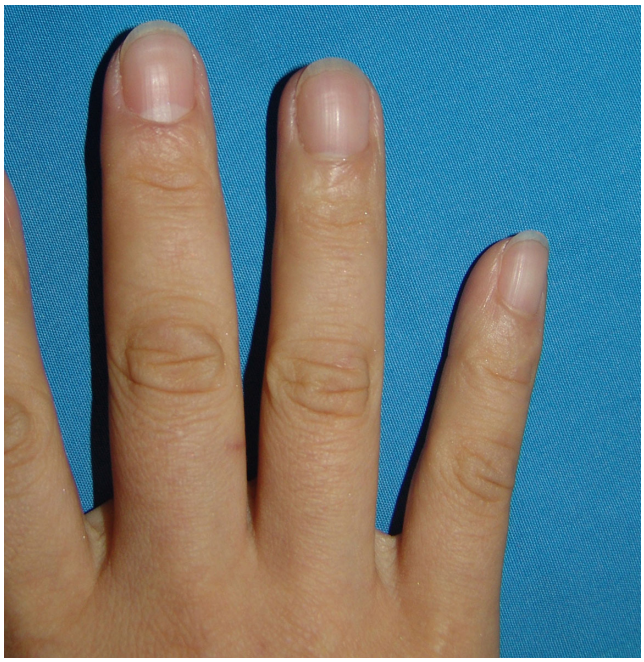

FIGURE 6 Photograph taken at one-year follow-up. The ring finger looks normal.

affected site was pricked with a needle 15 years before. Radiography showed a well-circumscribed osteolytic lesion sized 7*7*5 mm with a sclerotic margin in the distal phalanx of the left thumb. The cyst was completely removed through dorsal longitudinal incision. The nail matrix was involved during incision in this patient because of the lesion site. Curettage and bone graft were done. The result of histopathologic examination was consistent with an epidermoid cyst.

Three years after the surgery, follow-up radiography showed no evidence of recurrence. Her nail was normal-looking despite of an involvement of the nail matrix.

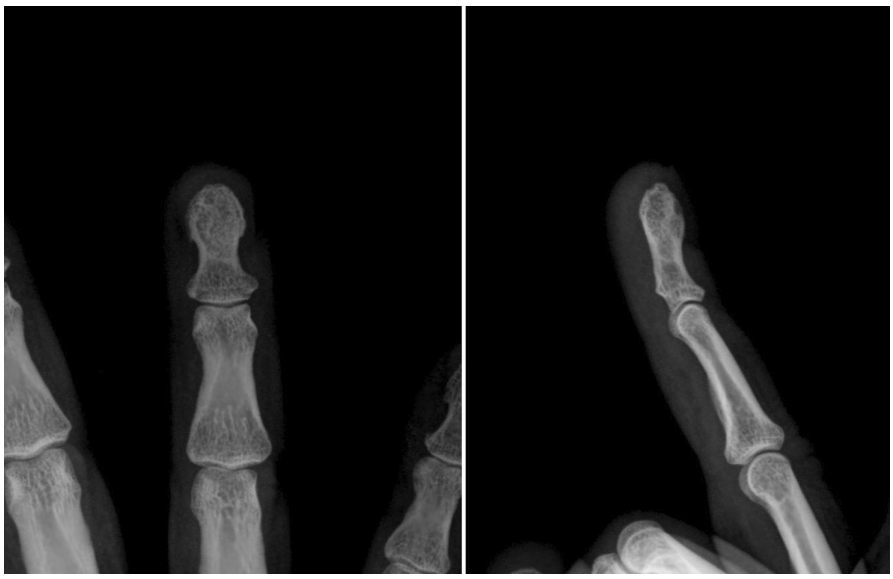

FIGURE 7 Radiographs taken at one-year follow-up showing a well-healed finger.

3 | DISCUSSION

Intraosseous epidermal cyst commonly affects the skull or finger phalanges.[1] The most common site of the phalanx is the distal phalanx, especially the third finger.[8] There have been several theories with regard to the origin of the cyst. First, traumatic theory suggests that trauma or surgical procedures to the bone may result in migration of a fragment of the nail bed into the phalangeal bone.[5] Second, congenital theory suggests a pathogenesis to embryonal misplacement of epithelial cells into the bone.[6] The third theory proposes that an epidermoid cyst from soft tissue can penetrate into the adjacent bone.[7] Both patients presented in the current paper had a history of previous injury to the involved phalanx, which might support the first theory. The first case had a blunt injury without external wound, and the second case had a needle puncture injury. Epidermal cyst generally takes a long time for symptoms to appear after the onset of the lesion. In order to make an accurate diagnosis, meticulous attention should be given when taking the patient's history, considering that even minor injuries that may have been forgotten by the patient over a long period of time could potentially be a contributing cause.

The peak incidence of an epidermal cyst is seen in the age group of 25–50 years. Its occurrence is three times higher in males than in females.[8] This might be because males are prone to be exposed to the danger of injury.[9] Patients usually have no pain. They go to see a doctor for enlargement of the fingertip. It is a solitary lesion in most cases. Radiologically, intraosseous epidermoid cyst presents as a well-defined, unilocular, osteolytic lesion with a sclerotic margin with or without soft tissue swelling. Its differential diagnoses include chronic infection, enchondroma, intraosseous ganglion, osteoid osteoma, simple bone cyst, aneurysmal bone cyst, giant cell tumor, glomus tumor, and metastatic bone lesion.[10] Histologic study is required for a definite diagnosis. Microscopically, the cyst is lined by stratified squamous epithelium and filled with keratinized material. As the lesion grows, cystic wall becomes thinner. Once it is diagnosed, curettage of the cyst and removal of the cystic wall are required to prevent recurrence. Once completely excised, lesions do not recur.

The lesion in the first case was preceded by a blunt trauma without fracture or gross disruption of soft tissues. Considering that trauma history could potentially be a contributing factor to the development of intraosseous epidermoid cysts, the authors believe that the etiology can be more clearly elucidated by carefully taking patients' history of previous trauma which may be overlooked by patients or medical staff.

In summary, intraosseous epidermoid cyst seems to be related to past traumatic history. Although a histological examination is required for an accurate diagnosis, precise assessment of patients' trauma history is also crucial. In general, curettage and, if necessary, bone graft may be considered as sufficient treatment options.

AUTHOR CONTRIBUTIONS

Dong Hwan Kim: Writing – review and editing. **Jung Ho Noh:** Conceptualization; data curation; writing – review and editing.

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CONFLICT OF INTEREST STATEMENT

The authors have no conflicts of interest to disclose.

DATA AVAILABILITY STATEMENT

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

ETHICS STATEMENT

This case report did not need an IRB approval because a case report of three or fewer patients does not need an IRB approval according to the IRB policy.

CONSENT

Written informed consent was obtained from patients to publish this report in accordance with the journal's patient consent policy.

ORCID

Jung Ho Noh https://orcid.org/0000-0002-7898-7349

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