

## Case Report

# Intraosseous Epidermoid Cyst of Distal Phalanx of Fingers

Chih-Hsin Hsieh,<sup>1</sup> Yin-Chun Tien,<sup>1,2</sup> and Song-Hsiung Chien<sup>1,2,3</sup>

<sup>1</sup> Department of Orthopaedics, Chung-Ho Memorial Hospital, Kaohsiung Medical University, Kaohsiung 807, Taiwan

<sup>2</sup> Department of Orthopaedics, Faculty of Medicine, Kaohsiung Medical University, Kaohsiung 807, Taiwan

<sup>3</sup> Department of Orthopaedics, Kaohsiung Municipal Hsiaokang Hospital, Kaohsiung 807, Taiwan

Correspondence should be addressed to Song-Hsiung Chien, shchien@kmu.edu.tw

Received 13 August 2008; Accepted 5 December 2008

Intraosseous epidermoid cysts of fingers are uncommon lesions. We reported two cases with intraosseous epidermoid cysts of distal phalanx of fingers. Case 1 was a 61-year-old woman with a swelling of the left middle finger without history of trauma. Case 2 was a 47-year-old male with a crushing injury of his right thumb. On radiographic findings, a well-circumscribed osteolytic lesion in distal phalanges was noted. Unexpectedly, histological examinations showed intraosseous epidermoid cyst.

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## 1. Introduction

Epidermoid cysts are commonly found in the subcutaneous tissue of the entire body, but intraosseous epidermoid cysts are unusual. Intraosseous epidermoid cysts arise most frequently in the skull and the distal phalanges of the hand [1–3]. It is very difficult to diagnose them correctly before surgery according to clinical and radiographic findings. We reported two cases with intraosseous epidermoid cysts of fingers and discussed the origins of the tumor.

## 2. Case Presentation

*2.1. Case 1.* A 61-year-old woman came to our clinic due to a swelling of left middle finger tip for 2 months. Tracing her history, no trauma occurred and the swelling developed gradually without tenderness. Erythema of finger tip was found. Roentgenograms demonstrated a well-defined osteolytic lesion over distal phalanx of the left middle finger (Figure 1). Magnetic resonance imaging (MRI) showed a well-defined osteolytic nodule beneath the nail of left middle finger (Figure 2). Under the impression of enchondroma or glomus tumor, surgical intervention was undertaken. Unexpectedly, a well-encapsulated cyst was discovered with grayish stinky material inside. Curettage of the cyst was performed. The pathologic examination demonstrated epidermoid cyst with granulation formation and foreign body reaction in the adjacent tissue (Figure 3).

*2.2. Case 2.* A 47-year-old male got a crushing injury of right thumb 2 years ago. He recovered quickly with normal thumb function; but right thumb pain was noted 6 months later. In physical examination, a reddish subungual patch was noted. Radiographic study showed a radiolucent intraosseous lesion in the distal phalanx of his right thumb (Figure 4). During the operation, a well-encapsulated cyst with white chessy material was discovered. After enucleation of the cyst, filling the defect with autogenous bone graft harvested from ipsilateral distal radius was done (Figure 5). The pathologic examination revealed laminated keratin in the cyst lined by stratified squamous epithelium consistent with epidermoid cyst.

## 3. Discussion

In 1968, Lerner and Southwick [1] reported that the intraosseous epidermoid cysts were twice as common in men as in women whose ages range between 8 and 83 years, and they tended to be concentrated in adults between the ages of 25 and 50 years—most common especially in actively working people. In 1970, Schajowicz et al. [4] cited that men are predominated and that the age of onset of the disease was between 19 and 54 years. The majority were manual workers and the most common location was the end phalanx of the hand. The left hand has a higher incidence of cysts than the right with the left middle finger being the most common site. The thumb, index, and ring finger are the next most



FIGURE 1: Roentgenogram of left middle finger showing a well-defined osteolytic lesion in the distal phalanx.



FIGURE 4: Radiograph of right thumb showing an osteolytic lesion with bony defect and fracture in the distal phalanx.



FIGURE 2: Magnetic resonance imaging of left middle finger showing a well-defined nodule and erosion of bone under the nail.



FIGURE 5: Radiograph of right thumb after curettage of the cyst and filling the defect with autogenous bone graft from ipsilateral distal radius.

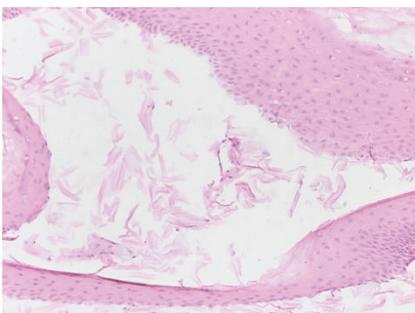


FIGURE 3: Laminated keratin in the cyst lined by stratified squamous epithelium. (Hematoxylin-eosin stain, original magnification, 20 $\times$ ).

common sites [1, 2]. Additional sites of involvement include phalanges of toes and large toe, ulna, femur, tibia, sternum, and amputation stumps [1, 3–5]. The symptoms comprise an increase in finger size with, or without pain and tenderness, redness, and curving of the nail. The interval between the trauma and the appearance of symptoms varies from weeks to years [1, 2, 4].

Epidermoid cysts of the skull are slow growing and are sometimes detected accidentally on radiographic examination of the skull. Most cases arise on the accessory sinus of

the nose and occipital scalp. Headache is the most frequent symptom. The cause of punched-out bony defect of the skull is thought to be long-term continuous pressure by the cyst resulting in the bone defect called scalloping [6].

The radiographic findings of intraosseous epidermoid cysts are not diagnostic. Roentgenograms show a well-defined osteolytic lesion frequently outlined by a thin rim of sclerotic bone. When the lesion reaches a certain size, the cortex becomes expanded and thinned, and the fractures or the bony defects may be observed [1, 4]. The differential diagnosis includes enchondroma, glomus tumor, aneurysmal bone cyst, metastasis, and osteomyelitis. Enchondroma is the most common and destructive primary bone tumor of the hand, and occurs more commonly in the middle and proximal phalanges. Unlike intraosseous epidermoid cysts, glomus tumors cause severe pain, cold sensitivity, and point tenderness. Glomus tumors produce pressure erosion and demonstrate smooth, concave deformity at one side or on the dorsum of the phalanx beneath the fingernail, or as a punched-out defect in the tuft. In the aneurysmal bone cyst, the eccentric bulb-like lesion occurs in the metaphysis of tubular bone, causing pain and limitation of motion. The sharp edge of an epidermoid cyst is distinctly different from the poorly defined osteolytic lesions observed with osteomyelitis and metastatic disease [1, 2, 4]. So, it is difficult to differentiate these tumors according to radiographic findings. Histological examination is the only way for accurate diagnosis.

Macroscopically, the cystic unilocular cavity of intraosseous epidermoid cysts reaches a diameter from 1 to 20 mm. The outer membrane is 1 to 5 mm thick and easily separated from the surrounding bone [4]. Histological examination demonstrates a cyst wall with stratified squamous epithelium with a well-defined granulous layer covered by laminated masses of keratin. While the cyst wall is interrupted and the keratin masses come into direct contact with the pericystic connective tissue, the foreign-body and giant-cell granuloma will germinate. The cyst contents comprise keratin, foreign-body giant cells, cholesterol cleft, and chronic inflammatory cells. There is no histologic difference between anatomic locations [2, 4].

The etiologies of phalangeal epidermoid cysts are thought to be (1) traumatic and being secondary to implantation of epidermal elements into the bone [1–5, 7, 8], (2) blastomatous formations, originating from displaced neural tube epithelial cell remnants during embryonic development [2, 6], or (3) iatrogenic origin related to a previous surgery, such as amputation stump [4]. In 2003, Ambo et al. [6] described a case of epidermoid cyst with perforation of the skull, and hypothesized that long-term continuous pressure by the cyst resulted in a bone defect. In case 2, the cause is believed to be due to traumatic introduction of epidermal tissue into bone. However, no trauma history was noted in Case 1 and there was no prior surgical history. Therefore, the cause of the lesion is hypothesized to be ectodermal cell rests during embryonic development.

In intraosseous epidermoid cysts, curettage with or without bone grafting is adequate. Amputation should not be performed as a primary treatment [1, 2, 4, 5]. The

clinical symptoms and radiological findings of intraosseous epidermoid cysts are nonspecific. It is challenging to differentiate intraosseous epidermoid cysts from other bone tumors of hands. Histological examination is very important for accurate diagnosis of intraosseous epidermoid cyst. In conclusion, the physicians should recognize the entity, and the intraosseous epidermoid cyst should be included in the differential diagnosis of bone tumors of hands. Histological examination should be performed as early as possible for correct diagnosis and appropriate managements.

## References

- [1] M. R. Lerner and W. O. Southwick, "Keratin cysts in phalangeal bones. Report of an unusual case," *The Journal of Bone & Joint Surgery. American Volume*, vol. 50, no. 2, pp. 365–372, 1968.
- [2] C. P. Samlaska and M. F. Hansen, "Intraosseous epidermoid cysts," *Journal of the American Academy of Dermatology*, vol. 27, no. 3, pp. 454–455, 1992.
- [3] B. Y. Wang, J. Eisler, D. Springfield, and M. J. Klein, "Intraosseous epidermoid inclusion cyst in a great toe. A case report and review of the literature," *Archives of Pathology & Laboratory Medicine*, vol. 127, no. 7, pp. e298–e300, 2003.
- [4] F. Schajowicz, C. L. Aiello, and I. Slullitel, "Cystic and pseudocystic lesions of the terminal phalanx with special reference to epidermoid cysts," *Clinical Orthopaedics and Related Research*, vol. 68, pp. 84–92, 1970.
- [5] R. A. B. Mollan, A. R. Wray, and D. Hayes, "Traumatic epidermoid cyst of the ulna. Report of a case," *The Journal of Bone & Joint Surgery. British Volume*, vol. 64, no. 4, pp. 456–457, 1982.
- [6] M. Ambo, M. Kumakiri, and Y. Chohno, "Epidermoid cyst with perforation of the skull," *Journal of the American Academy of Dermatology*, vol. 49, no. 5, pp. 934–936, 2003.
- [7] U. Handa, S. Kumar, and H. Mohan, "Aspiration cytology of epidermoid cyst of terminal phalanx," *Diagnostic Cytopathology*, vol. 26, no. 4, pp. 266–267, 2002.
- [8] R. Yang, M. C. Chang, Y. Liu, and W. H. Lo, "Intraosseous epidermoid cyst in distal phalanx of finger: a case report," *Zhonghua Yi Xue Za Zhi*, vol. 60, no. 2, pp. 109–112, 1997.