CASE REPORTS
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Intraosseous epidermal inclusion cyst of the right ring finger for 40 years

Epidermalna inkluziona cista u kosti desnog domalog prsta tokom 40 godina

Meng-Qiang Fan*†, Xiao-Lei Chen‡, Jiang Hua*†, Li-Pei Wang§, Yong Huang‖, Jie-Feng Huang*†

***The First Affiliated Hospital of Zhejiang Chinese Medical University, Department of Orthopaedics, Hangzhou, China; Zhejiang Chinese Medical University, †The First Clinical College, §Basic Medical College, Hangzhou, China; ‡Shanghai University of Traditional Chinese Medicine, Basic Medical College, Shanghai, China; ‖Hospital of Chengdu University of Traditional Chinese Medicine, Department of Orthopaedics, Chengdu, China**

Meng-Qiang Fan and Xiao-Lei Chen made equal contributions, and are co-first author.
Yong-Huang and Jie-Feng Huang made equal contributions, and are co-correspondence author.

Abstract

Introduction. Intraosseous epidermal inclusion cysts (IEpC) are benign bone lesions lined with squamous epithelium. Finger phalanges are the second most common site of predilection after the skull. **Case report.** We presented a case of a typical IEpC at the distal phalanx of the right ring finger following a remote history of a crush injury to the finger (40 years earlier). The patient experienced painful enlargement and progressive swelling of that finger during the previous month. On physical examination, the finger showed typical "clubbing" with local tenderness. X-ray showed bone destruction, and magnetic resonance imaging (MRI) revealed abnormal signals in bone tissue in the distal phalanx of the right ring finger. The patient underwent distal phalanx amputation of the right ring finger. The diagnosis of IEpC was histopathologically confirmed. At follow-up 2 years later, the stump healed well and without recurrence. **Conclusion.** IEpC with a history of up to 40 years is very rare, and although the patient presents with a typical "clubbing" finger, the diagnosis was eventually confirmed by surgery and pathology.

Key words:
amputation; bone cysts; crush injuries; epidermal cyst; fingers; hand.

Apstrakt

Uvod. Intraosealne epidermalne inkluzione ciste (IEpC) su benigne lezije kostiju obložene skvamoznim epitelom. Posle lobanje, falange prstiju su drugo najčešće predilekciono mesto za pojavu ovih lezija. **Prikaz bolesnika.** Prikazan je slučaj tipične IEpC na distalnoj falangi desnog domalog prsta koja je nastala posle povrede prsta (mehanizmom nagnječenja – *crush*), 40 godina ranije. Pacijent se žalio na bolno uvećanje i progresivan otok tog prsta, tokom prethodnih mesec dana. Objektivnim pregledom, prst je imao karakterističan „maljičast" izgled uz lokalnu osetljivost. Radiografskim pregledom viđena je destrukcija kosti, a magnetnom rezonancom abnormalni signali u tkivu kosti u distalnoj falangi desnog domalog prsta. Urađena je amputacija distalne falange desnog domalog prsta. Dijagnoza IEpC potvrđena je histopatološkim ispitivanjem. Praćenjem pacijenta, dve godine kasnije, nađeno je da je patrljak dobro zarastao, bez recidiva. **Zaključak.** Pojava IEpC sa istorijom povređivanja do 40 godina je veoma retka pojava. Bez obzira što prst pacijenta ima tipičan „maljičast" izgled, konačna dijagnoza se potvrđuje hirurškim i patološkim nalazom.

Ključne reči:
amputacija; kost, ciste; povrede, kraš; cista, epidermalna; prsti; ruka.

Correspondence to: Jie-Feng Huang, Department of Orthopaedics, The First Affiliated Hospital of Zhejiang Chinese Medical University, 54 Youdian Road, Shangcheng District, Hangzhou, 310006, China. E-mail: 40983285@qq.com

Introduction

Intraosseous epidermal inclusion cysts (IEpC) are benign bone lesions lined with squamous epithelium. The skull is the most common site, mainly involving the parietal and temporal bones. Finger phalanges are the second most common site of predilection, followed by maxilla, mandible, temporomandibular joint, vertebrae, tibia, and femur [1]. Although the etiology of these tumors is not fully understood, it is believed that they occur due to cortical rupture of the bone leading to the proliferation of epidermal cells in the bone matrix [1–3].

We presented a case of a typical IEpC at the distal phalanx of the right ring finger following a remote history of a crush injury to the finger. The article was approved by the Hospital Ethics Committee. The patient gave informed consent for submitting his pictures for publication.

Case report

A 56-year-old man was presented to the hospital with a 1-month history of painful enlargement and progressive swelling of the distal phalanx of the right ring finger. During this period, he did not receive any treatment, nor did he take relevant drugs, such as anti-inflammatory and analgesic drugs.

The patient reported a history of finger injury without fracture 40 years ago. It got healed, but the swelling was slowly increasing in size. The patient did not go to the doctor for treatment because the finger showed no symptoms such as pain and febrile except slight swelling over the years. He had no symptoms or personal history of rheumatoid arthritis.

On physical examination, the finger showed a typical "clubbing" (Figures 1–3) with local tenderness. X-ray showed bone destruction, and magnetic resonance imaging (MRI) revealed abnormal signals in bone tissue in the distal phalanx of the right ring finger (Figures 2 and 3). The patient underwent distal phalanx amputation of the right ring finger. During the operation, we found that the swelling part was well wrapped, and it had a milky soft-tissue appearance. Histopathological examination revealed an IEpC (Figure 4). The wound healed well 2 weeks after the operation, and the stitches were removed. Two years later at follow-up, the stump healed well and without recurrence.



Fig. 1 – The right ring finger showed obvious "clubbing" finger.

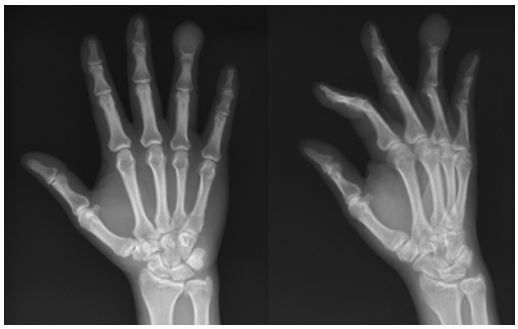


Fig. 2 – X-ray showed bone destruction in distal phalanx of the right ring finger.

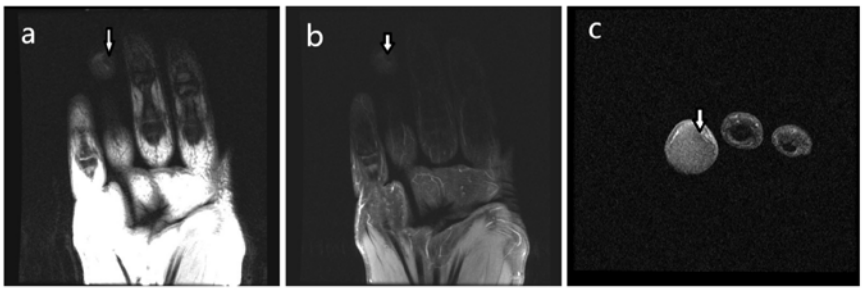


Fig. 3 – Magnetic resonance imaging (MRI) revealed abnormal signals in bone tissue in distal phalanx of the right ring finger [figures b and c are contrast-enhanced MRI; the arrows indicate the typical cystic region].

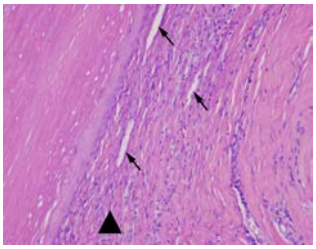


Fig. 4 – Histopathological examination revealed intraosseous epidermal inclusion cyst (hematoxylin and eosin staining, ×200).
("▲" Squamous epithelium; "→" The stratum corneum thickens and separates).

Discussion

IEpC are usually seen in subcutaneous tissue, however, intraosseous IEpC is a rare benign lesion [4]. Finger phalanges are the second most common site of predilection after the skull [1]. The most common age range is 25–50 for men and less for women [1, 3–5]. Phalangeal cysts can result from any type of injury caused by traumatic implantation of epidermal fragments into the bone or from the migration of nail bed fragments into the phalangeal.

The typical manifestation of IEpC in a finger is "clubbing". According to relevant reports of epidermal cyst [4, 6–8], our patient's "clubbed finger" is the most typical, and the patient's 40-year history is very rare. The imaging findings of IEpC are typically well-defined osteolytic lesions with or without soft tissue swelling. This is different from infection or metastasis, which would be a poorly defined osteolytic lesion [1]. The differential diagnoses of phalangeal lesions are broad and include inflammatory (i.e., chronic infection), as well as benign and malignant processes. Distally-based erosive cystic lesions include those secondary to tophaceous gout or a localized giant cell tumor of the tendon sheath, giant cell reparative granuloma, simple bone cyst, aneurysmal bone cyst, osteoid osteoma, and epidermoid inclusion cyst [4]. The usual treatment is curettage and removal of the cyst capsule [3]. Recurrence can occur if the excision was incomplete [1, 3, 4]. Bone grafting is rarely indicated. In our case, with the bone mass of the distal phalanx destroyed by the cyst, the decision was made to perform an amputation.

Conclusion

IEpC with a history of up to 40 years is very rare, and although the patient presents with a typical "clubbed finger", the diagnosis was eventually confirmed by surgery and pathology.

Conflict of interest

The authors declare that they have no conflict of interest. There is no funding source.

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