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The first case report of an intraosseous epidermoid cyst in the distal phalanx of the index finger with infection resulting in single clubbing finger: A case report and review of the literature

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An intraosseous epidermoid cyst at the distal phalanx of the index finger is extremely rare. These cysts are asymptomatic unless ruptured, severely infected, or transformed into malignant squamous cell carcinoma. We present a case of a single clubbing finger in an adult diagnosed with an intraosseous epidermoid cyst in the distal phalanx of the left index finger with no history of pulmonary or cardiovascular diseases. Preoperative MRI showed an expansile lytic lesion with a sclerotic margin. Histopathological examination indicates that there is keratinous cell debris in the cyst with a wall of stratified squamous epithelium, which was the key to the correct diagnosis of an intraosseous epidermoid cyst. Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

KEYWORDS

intraosseous epidermoid cyst, distal phalanx, clubbing, single clubbing finger, enchondroma

Introduction

Intraosseous epidermoid cysts are also known as pseudotumors or keratin cysts. They were first reported in 1923 (1). They are rare benign bone lesions that occur mostly in the skull, face, neck, toe, and finger phalanges, especially the distal phalanges (2). It is quite challenging to diagnose this disease as it has no specificity in clinical and imaging presentations. An expansile lytic lesion with a sclerotic margin is usually shown in a radiological image (3). However, this radiological feature is shared with other diseases, such as enchondroma, giant cell tumors, subungual melanoma, aneurysmal bone cysts, and chondroma (4, 5). Therefore, a correct diagnosis is usually achieved by histopathologic examination (6, 7). A clubbing finger or Hippocratic finger is a finger deformity characterized by enlargement of the fingertips and the curved nails around the fingertips, which often signify poor cardiopulmonary function in patients (8). However, in this article, we report the first case of a single clubbing finger in an adult patient as a result of an infected intraosseous epidermoid cyst in the distal phalanx of the left index finger.

Case report

In 2021, a 50-year-old female patient came to our outpatient department complaining of severe pain in the left index finger. Physical examination showed obvious swelling, redness, and tenderness on the eponychium of the left index finger, with nail root deformity, which looked similar to a clubbing finger, and the remaining fingers appeared normal (**Figure 1**). She denied a history of pulmonary and cardiovascular diseases except for a history of trauma in that she injured her left index finger 3 years ago and then the finger enlarged gradually and painlessly for the first few months after the injury. During this period, the fingernail of the left index finger fell off naturally twice. Six months ago, the patient visited a local hospital complaining of pain and intermittent infection, where the doctor considered the possibility of paronychia and gave her an antibiotic (celixin, the exact dosage, and usage are unknown) as the proper treatment. Unfortunately, the patient's pain was only relieved for a short period and became worse later on. Therefore, she visited our outpatient department



FIGURE 1
Preoperative view showing the swelling, redness, and tenderness on the eponychium of the left index finger.

for further treatment. After finishing the main outpatient check-up, a tumor at a distal phalanx of the left index finger was considered; thereby, the patient was hospitalized, and further examinations were required.

After admission, slightly limited mobility during flexion and circulation of the affected finger was observed and all blood tests were normal. Magnetic resonance imaging (MRI) revealed a lucent lytic lesion involving the distal phalanx that led to cortical thinning and bone expansion (**Figure 2**); thus, a tumor-like lesion was considered. Given her chronic severe pain and well-defined lesions on imaging, surgical excision was performed. After applying brachial plexus block anesthesia, we made a fish mouth flap incision in the distal phalanx of the left index finger, which allowed us to see the thinned phalangeal cortex and a cyst. A window was opened in the thinned cortex, where a creamy material was released, and then we performed curettage. After radical debridement, the skin was adjusted and the nail bed was sutured (**Figure 3**). Postoperative histopathological analysis of the lesion showed a cyst that contained a cavity of keratinized material and a wall of stratified squamous cells. These findings confirmed that the patient had an intraosseous epidermoid cyst (**Figure 4**), which previously had been misdiagnosed. Postoperative recovery was uneventful; the patient was allowed to have full motion of the left index finger 3 days after surgery. After 6 months of surgery, bone regeneration was noted radiographically and the patient was able to do full-weight exercises. After 1 year of clinical and radiological follow-up, the patient shows no sign of recurrence.

Discussion

An intraosseous epidermal cyst or a squamous epithelial cyst is a lesion with keratinous debris surrounded by a wall of squamous epithelium in a bone. The cysts arise more frequently in men than women, and the left middle finger is involved most frequently (9). It is clinically presented as a painless, reddish, tender, and swollen lesion (2, 10, 11).



FIGURE 2
MRI of the distal phalanx of the left index finger. T1 (A) and T2 (B,C) showing a well-defined osteolytic lesion with a clear boundary.

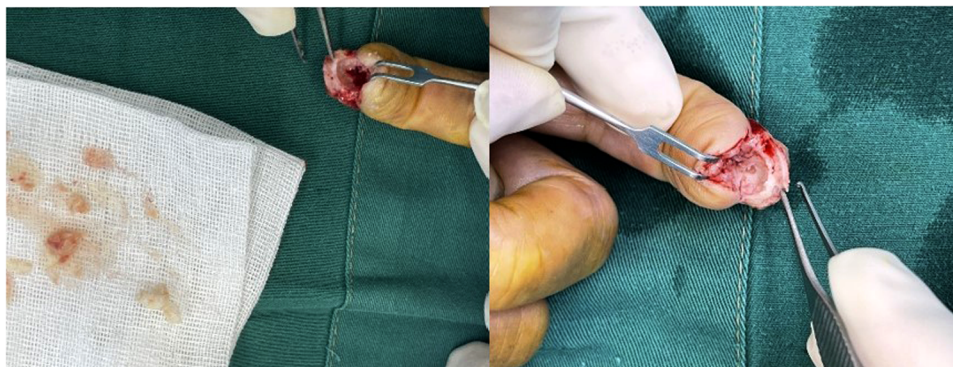


FIGURE 3
Creamy material removed from the cyst of the left index finger during the intraoperative curettage.

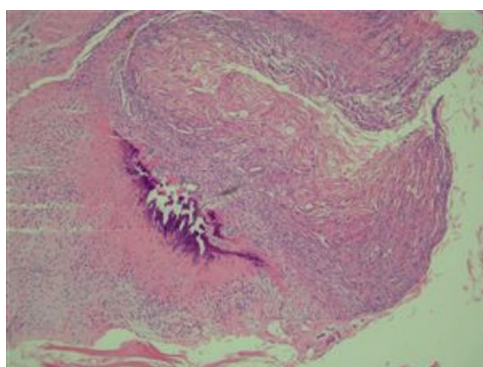


FIGURE 4
Histopathological examination revealing a wall of stratified squamous epithelium and keratinized material inside a cyst.

Many researchers have debated the real etiology of intraosseous epidermal cysts. The majority of previous studies found that trauma, such as crush injuries, is the common cause, and the epithelial cells could be embedded into the bone to form epidermoid cysts (11). Other studies mentioned congenital reasons why the inclusion of ectodermal tissue occurs during fetal development (12). All these etiologies ultimately point to the possibility of iatrogenic alteration of the epithelial cells within the bone, which leads to the formation of epidermoid cysts (11, 13).

Intraosseous epidermoid cysts are generally difficult to get diagnosed, depending on clinical examination and radiological findings, because their appearances and other characteristics are similar to those of other diseases, such as enchondroma, giant cell tumors, synovial cysts, aneurysmal bone cysts, and gout (14). Radiological imaging of an intraosseous epidermoid cyst shows a well-defined lytic lesion with circumscribed margin in a bone (2, 4). This imaging evidence lacks the specificity required for the definitive diagnosis of an intraosseous epidermoid cyst. Therefore, a histopathological test of the lesion tissue is the final resort to confirm the diagnosis of intraosseous epidermoid cysts among the various differential diagnoses. The histopathological analysis presents that an intraosseous epidermoid cyst contains keratinous cell debris surrounded by a wall of the stratified squamous epithelium (2). The most reliable treatments for intraosseous epidermoid cysts are curettage, which avoids recurrence, and bone graft, which reconstructs the wound (3, 15).

The noteworthy point found in our patient is that only her affected finger, a single finger, showed the clubbing finger changes. Clubbing finger changes often appear in multiple fingers and are a sign of multiple severe diseases, such as lung cancer, cystic fibrosis, sarcoidosis, cirrhosis, and cardiovascular diseases, which occur gradually over the years (16–19). Single clubbing finger occurs extremely rarely. A few cases have been reported, and all of them were linked to enchondroma, a type of benign bone tumor (19–21), except one formed as a result of superficial acral fibromyxoma (22) (Table 1).

TABLE 1 Cases with single clubbing fingers.

Gender	Age	Injured site	Case	Treatment	Follow-up period	References
Female	18	Left middle finger	Single clubbing finger caused by enchondroma	N/A	N/A	(21)
Female	23	Left ring finger	Single clubbing finger caused by enchondroma	Curettage and then the cavity filled with synthetic bone substitute	No evidence of recurrence, with a nearly normal-looking finger	(20)
Female	50	Right thumb	Single clubbing finger caused by enchondroma	Curettage and then the cavity filled with synthetic bone substitute	Performed full motion of the thumb, no recurrence after surgery	(19)
Female	58	Right index finger	Single clubbing finger caused by superficial acral fibromyxoma	Complete excision of the lesion	N/A	(22)

The mechanism of the clubbing finger remains ambiguous. A few theories, such as vascular endothelial growth factor (VEGF) (23), platelet-derived growth factor (PDGF) (24), and neurocirculatory reflex (25), are postulated to be participating in the formation of clubbing finger. Clubbing does not occur suddenly, and it has different stages. It starts as periungual erythema with a soft sensation, and then the angle between the proximal nail fold and the nail bed will increase. Clubbing causes the nail to be convex in shape, the skin at the fingertip to be shiny, and the possibility of hyperextension of the interphalangeal joint (26).

Conclusion

We here present the first case of a single clubbing finger associated with intraosseous epidermoid cysts. This case report intends to inspire clinicians that the differential diagnosis of the clubbing finger, especially in the case of a single clubbing finger, should include intraosseous epidermoid cysts.

Data availability statement

The original contributions presented in the study are included in the article/Supplementary Material; further inquiries can be directed to the corresponding author/s.

Ethics statement

Written informed consent was obtained from the individual(s) for the publication of any potentially identifiable images or data included in this article.

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Author contributions

HL, ZW, JL, VK: designed the study, collected data, and analyzed the results. YD, HZ, MA, SE, ZS, and AA drafted the manuscript. All authors contributed to the article and approved the submitted version.

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Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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