



Phalangeal intraosseous epidermoid cyst

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The authors describe two cases of phalangeal intraosseous epidermoid cyst. Two theories try to explain their origin. One theory sees traumatic implantation of epidermal cells as causative. A second theory is based on faulty embryogenesis.

INTRODUCTION

The incidence of intraosseous epidermoid cyst is undetermined but it is considered as a rare benign lesion. It is most frequently located in the skull (predominantly the parietal and temporal bones). The phalanges are the second most commonly affected site, especially the distal phalanx. Other sites include the tibia, ulna, femur or sternum.

CASE REPORT

Case 1

A 39-year-old housewife was referred by her general medical practitioner with pain and swelling of the tip of her right middle finger (non dominant hand). The patient stated that she had trapped the same finger in a door, approximately 17 years previously, resulting in loss of her nail, but this healed uneventfully. She had noticed increasing swelling and tenderness of her fingertip over four weeks but had been otherwise well. On examination there was pseudoclubbing of her fingernail, and tenderness of the nail bed to palpation. The biochemistry was

normal. Radiographic examination (fig 1) demonstrated an ovoid uniform radiolucency in the distal phalanx of her right middle finger with sclerotic margins and loss of the dorsal cortex. Exploration under general anaesthesia revealed an encapsulated lesion containing material of curd-like consistency. Microscopic examination following excisional biopsy confirmed the diagnosis of epidermoid cyst. The postoperative recovery was complicated by a wound infection that responded to a course of oral antibiotics.

Case 2

A 32-year-old male computer networker presented to casualty with a so-called paronychia of his right middle finger (dominant hand) that had failed

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Fig. 1. — (Case 1) AP and lateral views of right middle finger, demonstrating a well defined ovoid radiolucency situated towards the dorsal aspect of the distal phalanx. There is dorsal perforation and soft tissue swelling.

to respond to a nine-day course of antibiotics prescribed by his general medical practitioner. The patient had no recollection of any previous trauma to his hand. He was a type I diabetic but despite the 3-week history of pain and swelling of his distal phalanx he had no systemic signs of illness. Radiographic examination (fig 2) revealed a well-circumscribed lytic lesion in the dorsal aspect of the distal phalanx of his right middle finger. Curettage and biopsy of the lesion were performed under general anaesthesia using a midline incision after avulsion of the nail. The specimen was noted to contain a cheese-like material, and the microscopic examination was in keeping with an epidermoid cyst. Healing was uncomplicated.



Fig. 2. — (Case 2) AP and lateral views of right middle finger, demonstrating an expansile radiolucent lesion in the distal phalanx with sclerotic margins.

DISCUSSION

Harris initially documented intraosseous epidermoid cysts of the digits in 1930 (1). The lesion is most common in adults, especially manual workers; males are more frequently affected than females. Patients are most often in their fourth or fifth decade of life (range 8 to 83 years). The distal phalanx is most commonly affected, but intraosseous epidermoid cysts are also found in the other phalanges. The digits most frequently affected are the thumb, then the middle finger, and least commonly the little finger. Rarely digits of the feet may be affected. In over 45% of the cases, there is a history of minor or major blunt or penetrating trauma (frequently a crush injury), which may have occurred many years prior to presentation. The time from implantation to presentation of the cyst is not known, as the precipitating trauma is often minor. The pathogenesis of phalangeal intra-

osseous epidermoid cyst is not known. It has been proposed that it may result from implantation of epithelial cells into the subcutaneous tissues as a result of trauma. Surviving cells would subsequently proliferate and produce keratin (3). It is thought that phalangeal intraosseous epidermoid cyst, occurring in the absence of trauma, may be due to the proliferation of intraosseous inclusions of epithelial elements during embryogenesis (4).

Clinically the lesions typically occur in the distal phalanx and present with variable degrees of pain, swelling, nail deformity and sometimes erythema. The pain may be of sudden onset if there is a pathological fracture, or more gradual. It is exacerbated by minor trauma and by pressure on the nail bed, and it is throbbing in nature. There may be a pathological fracture of the phalanx. The skin is often normal in appearance, though there may be scars from previous trauma. The nail is usually deformed with pseudo-clubbing and loss of its normal smooth surface.

The radiographic features are typically those of an expansile radiolucency which may be elliptical in the longitudinal axis of the phalanx, without trabeculations. The cortex is usually thinned and may perforate the volar or dorsal aspect but there is no periosteal reaction unless the cyst becomes infected or there are attempts of healing of micro-fractures. The lesion has sclerotic margins and adjacent soft tissue swelling.

Histologically, intraosseous epidermoid cysts have a stratified squamous epithelial capsule surrounding centrally deposited keratin.

The differential diagnosis of phalangeal intraosseous epidermoid cysts includes :

1. *Enchondroma*. Enchondromas are benign tumours of hyaline cartilage within bone; they are usually solitary. Forty to sixty percent of solitary lesions occur in the metacarpal or in the proximal phalanx, though middle and distal phalanges may also be involved, but less commonly. They may present with onset of pain due to pathological fracture or due to malignant degeneration. Radiographically the lesions may contain calcifications but purely lytic lesions may resemble intraosseous epidermoid cysts. Both form lesions without irreg-

ularity of the cortex and without periosteal reaction.

2. *Giant cell tumour*. Giant cell tumours are benign but locally aggressive lesions of bone. They occur most commonly in adults in their third and fourth decades and are among the most common primary bone tumours of the distal phalanx. They frequently present with gradually increasing pain due to irritation of the periosteum. Pain may also herald a pathological fracture as a result of progressive weakening of bone. Radiographically giant cell tumours are most radiolucent in the centre, and are more radiodense peripherally. The lesion appears to expand and thin the cortex and may erode it to invade adjacent soft tissues.

3. *Glomus tumour*. Glomus tumours are benign tumours of the neuromyoarterial apparatus that usually serves to regulate skin circulation. Seventy-five percent of such tumours occur subungually in the hand and they are most common in the fourth and fifth decades. They classically present with a triad of severe pain, localized tenderness and sensitivity to cold. These tumours appear radiographically as well defined osteolytic lesions with sclerotic margins.

4. *Metastatic tumours*. Metastatic tumours are rarely found in the phalanges. They are most frequently secondary deposits from a primary carcinoma of the bronchus or breast.

The most effective treatment of phalangeal intraosseous epidermoid cyst is surgical excision, with curettage of the phalanx. Bone grafting may be necessary for large defects. If the cyst is completely removed, the recurrence rate is low.

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