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CASE REPORT

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Epidermoid cyst mimicking monoarticular arthritis of the great toe

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Abstract Local swellings of the foot are a common presentation in rheumatological practice. In the following case report, an epidermoid cyst presenting with the typical features of monoarticular arthritis is described in a 27-year-old woman. The differential diagnosis of forefoot swelling is discussed, with particular emphasis on epidermoid cysts and monoarticular arthritis.

Keywords Differential diagnosis · Epidermoid cyst · Monoarticular arthritis

Introduction

Epidermoid cysts are common dermal lesions frequently encountered in the scalp, face, trunk, and scrotum [1, 2]. Descriptions of them in the foot are rare in the literature, although commonly encountered in daily clinical practice [3]. The mode of presentation of epidermoid cysts can vary from circumscribed subcutaneous swelling to a picture similar to arthritis. In the following case report, the diagnostic findings of an epidermoid cyst with an unusual location overlying the first metatarsophalangeal joint (MTPJ) are discussed and compared to those of monoarticular arthritis, which is a rarely mentioned but important differential diagnosis due to its similarity in presentation and relative frequency.

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Case report

A 27-year-old woman presented with swelling of her great left toe, which was painful on walking. The pain and swelling became successively worse, particularly during load-bearing. The patient's medical and family histories were negative for polyarticular disease and monoarticular arthritis. Conservative therapy with several corticosteroid injections was unsuccessful.

On inspection, a large, livid swelling was found on the plantar side of the first MTPJ which extended medially to the dorsum of the foot. Deformities such as hallux valgus or splayfoot were not present. The range of motion of the MTPJ was significantly decreased. On palpation, the lesion was warm, bulging, and tender. Further joints were not involved. History and clinical examination suggested stress-induced bursitis or synovialitis, although there was no improvement after intra-articular corticosteroid injection.

There were no pathological findings from laboratory tests (rheumatoid factor, human leukocyte antigen B27 [HLA-B27], uric acid, leukocyte count, C-reactive protein, erythrocyte sedimentation rate, and antibodies for borreliosis burgdorferi) indicating seropositive rheumatoid arthritis, HLA-B27-related disease, gout, septic arthritis, or Lyme's disease. Synovial fluid analysis demonstrated orange fluid containing cholesterol crystals (Fig. 1) suggestive of a chronic arthritis. The leukocyte count of the arthrocentesis was increased to 42,000/mm^3, showing mainly granulocytes consistent with an inflammatory process. On examination of the fluid aspirate, the microbiological examination was negative, and urate or calcium pyrophosphate crystals were not seen either.

Radiographs of the MTPJ revealed no bony derangement by osteoarthritis, gout, stress fracture, or chondrocalcinosis (Fig. 2). Ultrasonography demonstrated a well circumscribed, subcutaneous, anechogenic zone resembling either a fluid-filled cyst or an outpouching synovial membrane of the first MTPJ or the adjacent extensor hallucis longus tendon. The MRI findings were consistent with a synovitis filled with haemorrhagic fluid. The additional dorsal subcutaneous lesion was interpreted as secondary bursitis or a soft-tissue ganglion arising from the adjacent extensor hallucis longus tendon (Fig. 3). Combining the results of all investigations, synovialitis of unknown origin was suspected. For diagnostic and therapeutic purposes, revision of the great toe was performed.

Using a dorsomedial approach, across the MTPJ a subcutaneous, extra-articular mass near the adductor tendon was prepared. The lesion was partially elastic and partially solid. On excision, orange fluid was emptied. Further plantarly, a lesion measuring 1.5×1.0×0.8 cm was seen and completely excised. Exploration of the first MTPJ did not reveal synovialitis.

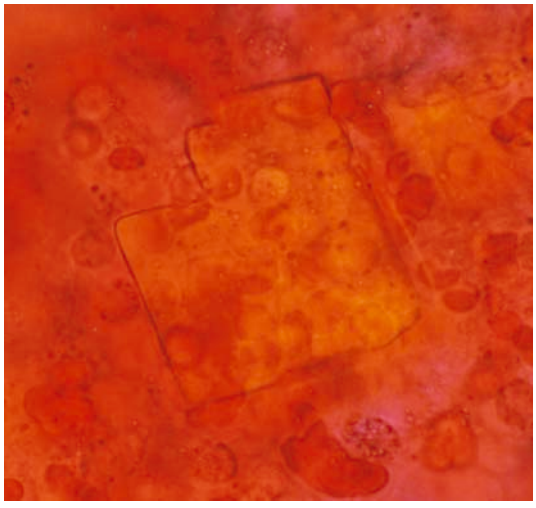

Fig. 1 Synovial fluid analysis. The rectangular crystal with a missing corner is suggestive of a cholesterol crystal. Light microscope, 100× magnification

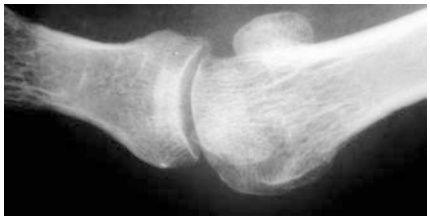

Fig. 2 Anteroposterior radiograph of the first MTPJ. There is no narrowing of the joint space, osteopenia, or bony erosion

On histological examination, the typical features of an epidermoid cyst were seen in both specimens with an accompanying granulomatous reaction. There was no sign of infection on microbiological examination. After 2 weeks of immobilisation in a cast, the patient was pain-free and allowed to bear weight fully. The further postoperative clinical course was uneventful.

Discussion

Epidermoid cyst is a dermal disorder resulting from proliferation of surface epidermal cells within the dermis [4]. Wernher [5] first described epidermoid cysts. In further reports, the terms epidermal cyst, epidermal inclusion cyst, retention cyst, and infundibular cyst have been used synonymously [1].

Typically, such lesions appear on the scalp, face, neck, trunk, or scrotum [1, 2]. Less than 10% occur on the extremities and are most rare on the palm of the hand or sole of the foot [6]. In a review of 67,000 soft-tissue neoplasms collected over a 4-year period, epidermoid cysts were the fifth most common lesion affecting the foot [3]. They are observed throughout all ages but tend to be more frequent in young to middle-aged adults [7], affecting both sexes equally [8].

The cyst develops from the introduction of epidermal fragments into a heterotopic location, where the cyst enlarges through accumulation of epithelial and keratinous debris, resulting in a prominent subcutaneous mass [7]. Until recently, the main pathomechanism of epidermoid cysts of the foot was thought to be either traumatic, e.g. caused by a chronic shoe impingement [7], or iatrogenic, e.g. caused by surgery [9]. Only recently, human papilloma viruses have been identified in plantar epidermoid cysts. Based on these findings, Egawa [10] postulated that penetrating injury does not cause epidermoid cysts directly but introduces the human papilloma virus, which causes the cyst formation. The role of human papilloma virus in this hypothesis explains why epidermoid cysts are not more commonly seen after surgery [9].

Histologically, an epidermoid cyst consists of a cyst wall with stratified squamous epithelium supported by a dense fibrous tissue stroma. The cyst lining may show pseudoepitheliomatous hyperplasia suggestive of epidermoid carcinoma. The cyst contents include moderate amounts of mature keratin, foreign-body giant cells, cholesterol clefts, and chronic inflammatory cells. There is no histological difference among anatomic locations [11]. Epidermoid cysts of the foot typically present as local, bulging swelling associated with redness, pain, and tenderness. Rarely, Morton's metatarsalgia-like symptoms have been observed [12].

The differential diagnosis of plantar epidermoid cysts includes warts, neuromas, ganglion cysts, lipomas, fibromas, and appendageal tumours [1]. Due to similar clinical findings, we and other authors [4, 12] consider monoarticular arthritis a further, important differential diagnosis to plantar epidermoid cysts.

The laboratory findings in epidermoid cysts are negative unless there is superinfection due to recurrent injections [13]. Synovial fluid analysis can demonstrate an increased leukocyte count as well as cholesterol crystal deposits, findings commonly observed in effusions due to chronic rheumatoid arthritis or osteoarthritis. On ultrasonography, epidermoid cysts are subcutaneous, fluid-filled sacs without links to the adjacent joints or tendons, in contrast to monoarticular arthritis, which presents either by an effusion of the affected joint or by a secondary intermetatarsal bursitis. The latter was shown by Koski [14] to be common in patients with early rheumatoid arthritis. Typically, radiographs of epidermoid cysts reveal no abnormalities except in intraosseous ones [11, 15], which demonstrate a characteristic osteolytic lesion outlined by a thin rim of sclerotic bone [11] which might look like a bony erosion of seronegative rheumatoid arthritis. Periarticular calcifications, that are commonly observed in gout or chondrocalcinosis, are not typically observed [16]. The MRI appearance of epidermoid cysts strongly resembles that of simple cysts, except for the cyst contents, which are typically proteinaceous in nature. There is an uptake of contrast agent by the surrounding cyst wall. The cyst itself can be unilocular or, when ruptured, multilocular. In our and others' cases [16], the signal enhancement with contrast agent around the cyst was due to a giant-

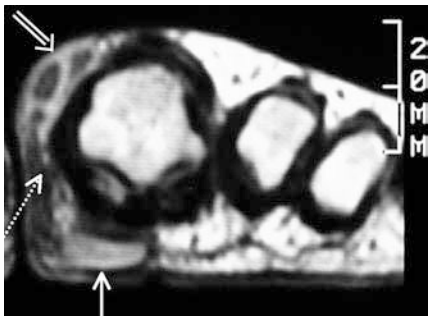

Fig. 3 T1-weighted coronal image of the first MTPJ with i.v. gadolinium pentate. → Plantar epidermoid cyst with an intermediate signal and rim enhancement. ⇒ Dorsal multiloculated cyst with rim enhancement and a decreased signal in the centre. → Granulomatous reaction with an increased signal

cell reaction. In patients with a monoarticular rheumatoid arthritis, there is synovialitis with small bony erosions and accompanying bone marrow edema. In some cases intermetatarsal bursitis may also be present.

Therapeutically, complete excision is the preferred mode of therapy for epidermoid cysts, because recurrence is likely using only incision and drainage or aspiration alone. The excision should involve the entire cyst, including the wall and the covering skin to avoid the malignant transformation reported to occur in 0.33% to 9.2% of cases [17]. Intraosseous epidermoid cysts need to be treated by sorrow curettage of the lytic lesion and, in case of large defects, by bone transplantation [13].

References

1. Fisher BK, MacPherson M (1986) Epidermoid cyst of the sole. J Am Acad Dermatol 15:1127–1129
2. Bartolomei FJ, McCarthy DJ, Gastwirth CM, Peet FJ Jr (1985) Epidermal cyst of the plantar surface. J Am Podiatr Med Assoc 75:207–210
3. Berlin SJA (1984) A laboratory review of 67,000 foot tumors and lesions. J Am Podiatry Assoc 74:341
4. Matsumoto K, Okabe H, Ishizawa M, Egawa M (2001) Intermetatarsophalangeal bursitis induced by a plantar epidermal cyst. Clin Orthop 151–156
5. Wernher A (1934) Das Atherom, eingebalgtes Epitheliom. Virchows Arch F Path Anat 8:221
6. Greer E (1974) Epidermal inclusion cyst of the sole. Arch Dermatol 109:251–252
7. Coulter PD, Bouche RT (1999) Traumatically induced inclusion cyst secondary to shoe impingement: report of three cases. J Foot Ankle Surg 38:271–277
8. Marczak L (1990) Epidermal inclusion cyst of the heel. J Am Podiatr Med Assoc 80:660–661
9. Baran R, Bureau H (1988) Two post-operative epidermoid cysts following realignment of the hallux nail. Br J Dermatol 119:245–247
10. Egawa K, Kitasato H, Honda Y, Kawai S, Mizushima Y, Ono T (1998) Human papillomavirus 57 identified in a plantar epidermoid cyst. Br J Dermatol 138:510–514
11. Samlaska CP, Hansen MF (1992) Intraosseous epidermoid cysts. J Am Acad Dermatol 27:454–455
12. Rahimi H, Barz B (1975) Rare location of an epidermoid cyst in the forefoot region with nerve compression syndrome. Contribution to the differential diagnosis of Morton's metatarsalgia. MMW 117:1125–1126
13. Wu KK (1992) Epidermoid cysts of the foot: with or without bone involvement. J Foot Surg 31:203–206
14. Koski J (1998) Ultrasound detection of plantar bursitis of the forefoot in patients with early rheumatoid arthritis. J Rheumatol 25:229–230
15. Belusa M (1991) Intraosseous epidermoid cyst. Handchir Mikrochir Plast Chir 23:200–201
16. Fisher AR, Mason PH, Wagenhals KS (1998) Ruptured plantar epidermal inclusion cyst. AJR Am J Roentgenol 171:1709–1710
17. Sheff J, Bregman P, Curran T (1998) Bilaterally symmetrical epidermal inclusion cysts with foreign-body giant-cell reaction. J Am Podiatr Med Assoc 88:410–413