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Case report 555

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Imaging studies

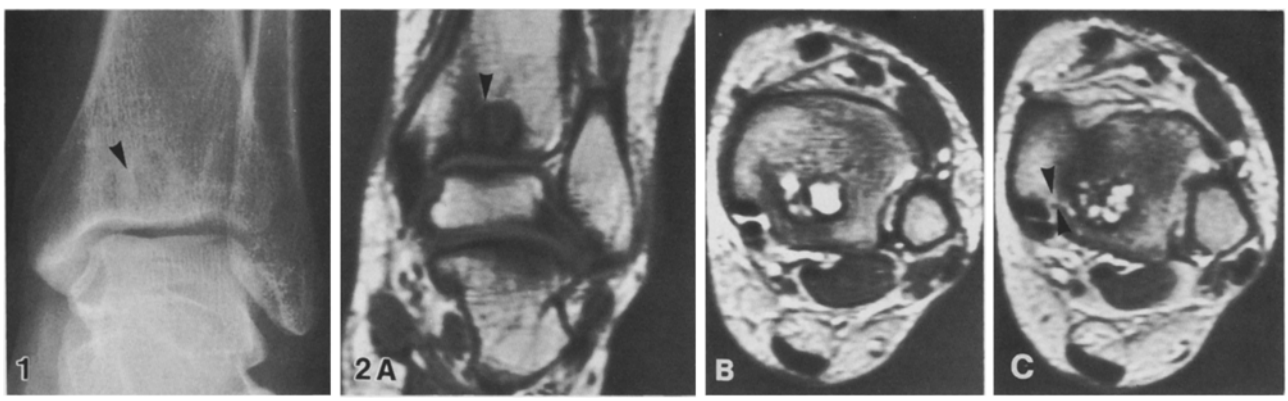

Fig. 1. Anteroposterior radiograph of the left ankle. Lobulated subchondral lucencies are seen at the tibial plafond (*arrows*). The margins are well defined, faintly sclerotic, and the joint space appears normal

Fig. 2. A Coronal spin echo image with relative T1-weighting (TR = 800, TE = 20). Well circumscribed, low signal intensity lesion is seen in the subchondral region. Septation (*arrow*) is imaged as a line of signal void. Adjacent articular cartilage is normal. **B, C** Axial spin echo T2-weighted images (TR = 2000, TE = 80). Lesion shows regions of high signal intensity as well as foci of signal void (**B**). Thin line of high signal intensity appears to connect the lesion with the flexor tendon sheaths (**C**, *arrowheads*)

Clinical information

This 42-year-old woman sprained her left ankle 5 months previously and subsequently experienced persistent pain along the posterior aspect of her ankle. Her medical history was unremarkable except for a parathyroid adenoma which was excised two years earlier. Physical examination revealed moderate tenderness over the posterior aspect of the ankle with no swelling.

Roentgenograms of the ankle demonstrated a lobulated lucency with sclerotic margins in the subchondral region of the distal end of the left tibia (Fig. 1). No evidence of articular or soft tissue involvement was noted. Radionuclide bone scan showed increased uptake in the distal end of the tibia, with no other regions of abnormal uptake.

Magnetic resonance images were obtained, demonstrating a sharply circumscribed lesion in the subchondral region of the distal end of the left tibia (Fig. 2). Except for septations, the lesion appeared as a homogeneous low signal intensity on T1-weighted images. With T2-weighting, foci of very high signal intensity were intermixed with regions of signal void. A serpiginous line of high signal intensity was identified coursing between the lesion and the flexor tendons.

Surgical excision of the lesion was performed.

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Diagnosis: Intraosseus ganglion cyst of the distal end of tibia

The differential diagnosis included brown tumor of hyperparathyroidism (because of history), giant cell tumor, chondroblastoma, chronic infection, and osteoarthritic cyst.

At surgery a $20 \times 16 \times 15$ mm cystic lesion was discovered with a hard, bony shell, containing a clear, viscous, slimy fluid. No connection to articular surface or bony cortex could be identified. The structure was well encapsulated and removed in its entirety. The resulting defect was then packed with bone chips from the ilium. Histological evaluation revealed a multicompartmentalized cyst-like, fibrous tissue structure (Fig. 3). Focal myxoid degeneration was also evident (Fig. 4).

The patient was doing well 7 months after the procedure.

Discussion

Intraosseous ganglion cyst is a rare lesion, with about 150 cases in the literature. The median age at the time of diagnosis is 44 years with a range from 14 to 86 years. The majority of cysts involve the proximal or distal ends of the tibia, proximal end of the femur, or distal end of the ulna, although any articulated bone may be involved [11].

Clinical features include mild to moderate pain, exacerbated by use of the affected region. There may be tenderness and mild swelling. The lesion may be entirely asymptomatic and found incidentally on a roentgenogram obtained for unrelated reasons.

Radiographic features include an eccentric, subchondral location in the end of a long bone. The lesion may be less obviously eccentric in a small bone such as a carpal bone. The cyst is usually a few mm to 3 cm in diameter, although lesions up to 7 cm have been described [7]. Typically, the cyst appears as a well circumscribed, single or multiloculated lucency, with a rim of sclerosis. The surrounding cortex is usually described as intact, although some connection to surrounding soft tissues may be noted [4, 6]. In this case, the MR images show a defect in the overlying cortical bone. This may have some significance in terms of explaining the etiology of intraosseous ganglion (see below). Due to the infrequent occurrence of these lesions and the fact that MRI is relatively new, no previous MR images of intraosseous ganglion cysts could be found in the literature.

Pathological findings include a smooth, oval cyst with a compartmentalized, white fibrous lining. Microscopic evaluation of the walls and septae reveals benign fibrous to myxoid tissue. The cyst contains a tenacious, stringy white mucinous material. Note that these lesions are not true cysts in that generally no epithelial lining is present.

The etiology of intraosseous ganglion cysts is not definitely established. Based on our review of the literature and pathological experience with numerous soft tissue and occasional osseous ganglion cysts we offer the following pathogenetic scheme:

1. The first event is a herniation or "trapping" of soft tissue or tenosynovial soft tissue between ligamentous and/or tendinous structures within tight compartments adjacent to bone.
2. The entrapped tissue suffers an aberration of its normal homeostasis, perhaps due to partial vascular compromise.
3. Fibroblasts respond to the injury by undergoing metaplasia to myxofibroblasts, stellate cells rich in proteoglycan and hyaluronic acid.
4. Pools of mucinous material accumulate and eventually the pools coalesce, forming a cyst-like mucoid mass. Bands of fibrous tissue may persist between the collections of mucin, giving the cyst a multilocular structure.
5. Occasionally a mass adjacent to bone will be in a soft tissue compartment under sufficient pressure that it erodes into bone. Communication with the soft tissues may persist, but cortical remodelling may also take place, leading to complete separation from the soft tissue element [6].

The above pathogenetic view is consistent with the pathology of ganglion cysts, as well as the location of soft tissue ganglia adjacent to tendon, ligament, or tenosynovial structures. This theory also explains why intraosseous ganglia are always found in the epi-metaphyseal region, where the involved soft tissue structures lie adjacent to bone. Finally, as suggested by Kambolis et al., a soft tissue ganglion which erodes into bone may have an overlying cortical defect, and will most likely remain eccentrically located within the bone.

Several entities are difficult to distinguish from intraosseous ganglion by clinical and radiological means. *Brown tumors* associated with hyperparathyroidism can undergo cystic degeneration and persist even after the parathyroid abnormality is corrected. Such cysts are usually found in the jaws, face, pelvis and ribs, but could potentially involve the distal end of the tibia. The cysts are usually meta-diaphyseal in location and not subchondral, which made this diagnosis unlikely for our case.

Giant cell tumor can present as an insidious, dull ache with minimal swelling in a young adult. The classic radiological "soap-bubble" appear-

Pathological studies

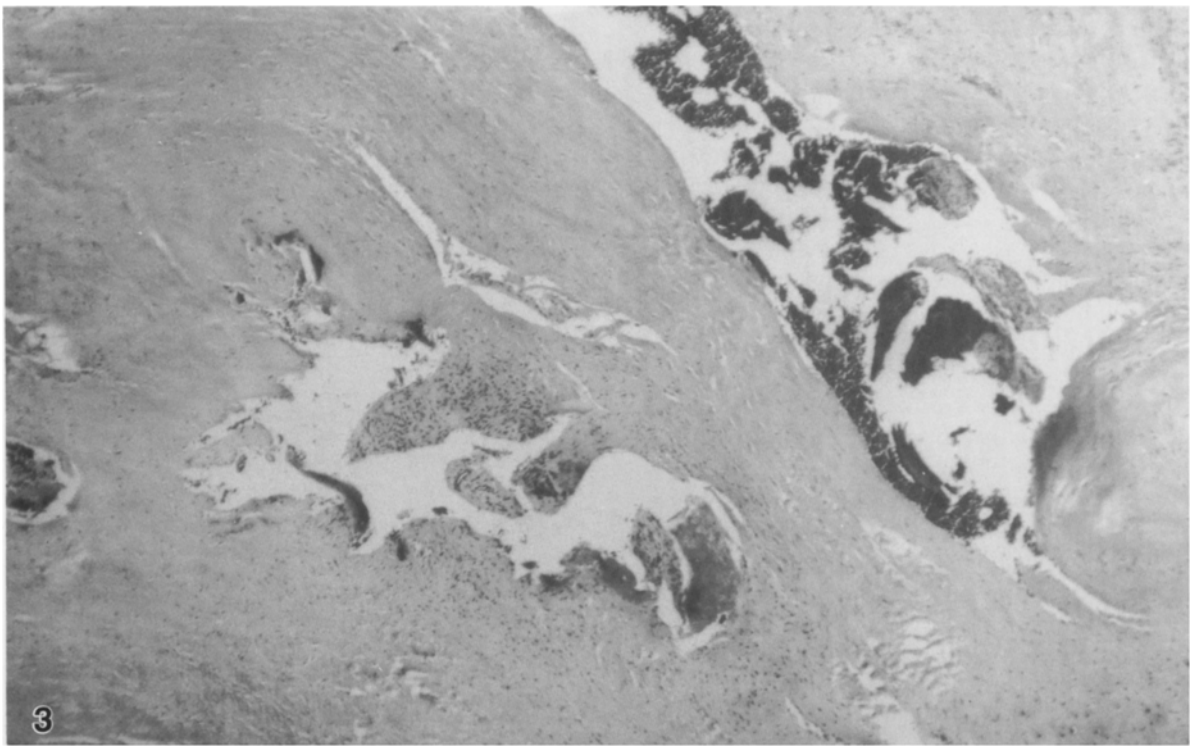

Fig. 3. On pathological examination the cyst-like tissue was predominantly fibroblastic and multicompartmental (HE × 40)

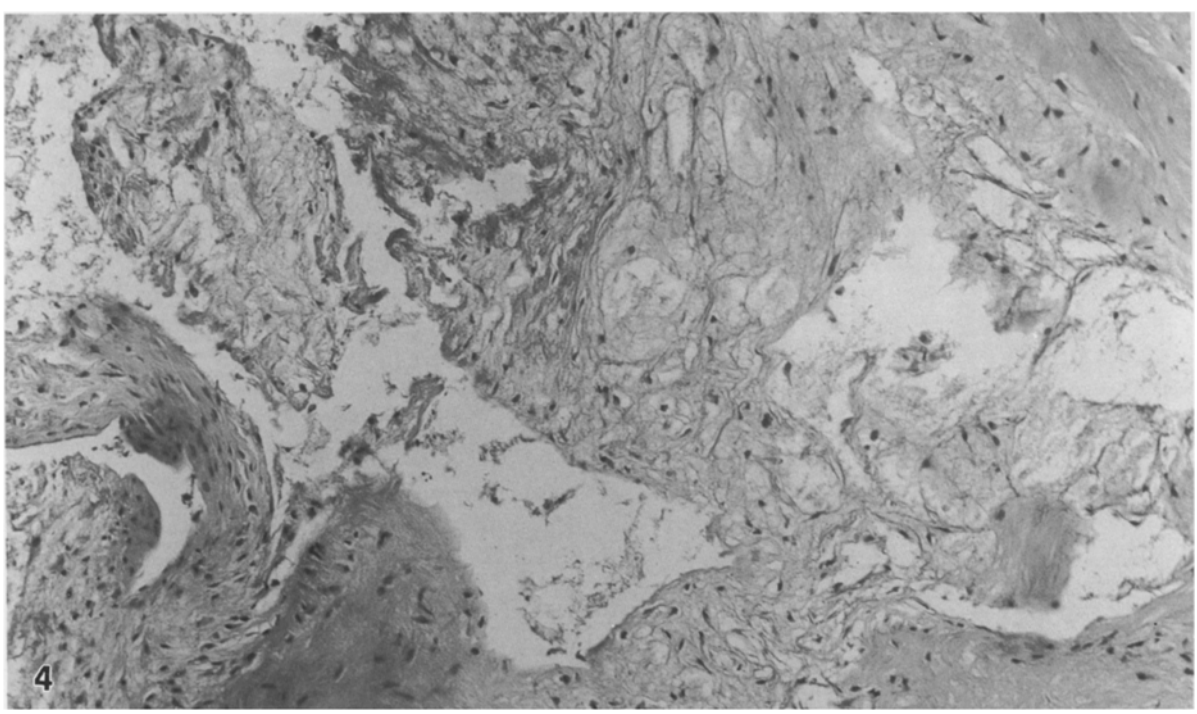

Fig. 4. Some portions of the wall showed myxoid fibroblasts in addition to the fibroelastic tissue. This is characteristic of early to mid-phase ganglion cysts (HE × 125)

ance with heavy septations is often not present, and an eccentrically located subchondral lucency may be all that is appreciated. Although there generally is more cortical thinning than was present in this case, giant cell tumor could not be excluded.

Chondroblastoma is another consideration with an eccentrically located epiphyseal lucency, but it is uncommon in a patient with fused epiphyseal plates.

Simple bone cyst is also a possibility in this sort of lucency, but this lesion is more frequently centrally located, metaphyseal, and associated with a thin and expanded cortex.

Chronic focal infection can lead to a cystic subchondral lesion, a *Brodie's abscess*, but it is more often metaphyseal, elicits a periosteal reaction, and usually is associated with significant pain.

Chronic abnormalities of the synovium, as in *rheumatoid arthritis* or *pigmented villonodular synovitis*, may lead to cyst-like lesions in the epiphyseal bone, but these cysts are associated with a preexisting synovial proliferation and soft tissue mass.

On many occasions, authors have confused *osteoarthritic cysts* with intraosseous ganglia. These "cysts" are the result of cartilage erosion with synovial fluid being squeezed through cartilage into the subchondral bone. The cysts are rarely solitary and are more centrally located along the weight-bearing surface of the joint. Joint space narrowing, eburnation, and osteophyte formation are all usually prominent before subchondral cysts are seen in osteoarthritis. Other joint abnormalities are also found, including a defect in the subchondral plate, which connects the cyst to the articular space.

True synovial cysts are similar to the cysts seen in osteoarthritis but occur independent of any chronic joint abnormalities. They are virtually identical to intraosseous ganglia both radiographically and histologically. The only difference is the presence of a synovial lining and a connection to the articular space.

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