The authors describe the case of a 33-year-old man with a solitary cystic lesion of the left distal radius with central radiodensities that radiographically suggested a chondroid matrix. Curettage of the lesion revealed numerous calcospHERites with a lamellar structure. This lamellar pattern is thought to be a result of alternating precipitation and diffusion of supersaturated solutions. This biochemical process has been called the Liesegang’s phenomenon, and the structures have been termed Liesegang’s rings. The presence of these structures in this benign lesion contributed to atypical roentgenographic and pathologic findings. (Key words: Solitary bone cyst; Liesegang’s rings; Calcospherites) Am J Clin Pathol 1989;92:831–833

SOLITARY BONE CYSTS are benign lytic lesions of unknown etiology that usually present with localized pain or pathologic fracture. 1–4,8–10 Histologic material from these cysts has been shown to contain fibrous tissue, chronic inflammatory and giant cells, hemosiderin pigment, and cholesterol clefts. 2 We describe the case of a 33-year-old man with a history of pulmonary histoplasmosis and a cystic lesion of the left distal radius that demonstrated central radiodensities suggesting a chondroid matrix. Curettage of the lesion revealed numerous calcospherites with a lamellar configuration. Such structures are the result of alternating diffusion and precipitation in supersaturated solutions originally described by Liesegang. 5–7 These Liesegang’s rings may be observed in cysts, which usually contain hemorrhagic or proteinaceous material, or in necroinflammatory tissue. They have been described in kidney, synovium, conjunctiva, eyelid, pericardium, omentum, breast, fallopian tube, epididymis, and in endoskeletal calcification in sharks and vertebræ in dogfish. 12 To our knowledge, this is the first case of human solitary bone cyst containing Liesegang’s rings described in the literature.

Report of a Case

A 33-year-old man was admitted to Naval Hospital, Bethesda, Maryland, with a four-month history of increasing pain and tenderness in his left wrist. The patient had a long history of periodic wrist pain following a left Colles’ fracture in November 1967. A lytic lesion at the fracture site was noted in follow-up x-rays in 1968. The patient recently had noted increasing pain associated with activity, in particular cardiopulmonary resuscitation (CPR) training and push-ups.

Past medical history was remarkable for a prolonged illness in 1967 characterized by fever, cough, shortness of breath and myalgia. This occurred following summer travel to the southwest United States. In 1968 he underwent bronchoscopy for prolonged pulmonary complaints; the bronchoscopy was unremarkable. In 1969 a patch test was performed as part of a physical examination; the test showed reactivity to histoplasma antigen. He was evaluated in 1983 for hemoptysis related to a two- to three-month period of flu-like symptoms. Bronchoscopy at that time was negative. Chest x-ray revealed multiple calcified granulomata.

On physical examination the patient was a well-developed, well-nourished white man in no distress. Vital signs were within normal limits. There was mild atrophic change of the left forearm. No edema or tenderness of the extremity was noted, and neurovascular status was intact. Examination was otherwise unremarkable.

Radiographic studies of the left forearm and wrist revealed old healed fractures of the distal shafts of the radius and ulna. There also was an expansile, radiolucent lesion in the region of the radial styloid with a central radiodensity suggesting a chondroid matrix (Fig. 1). Chest x-ray revealed calcified granulomata. Laboratory studies were noncontributory. Based on the clinical findings, neither an infectious nor neoplastic process could be ruled out.

The patient underwent curettage of the lesion following axillary block. On removal of a cortical window, a cyst containing yellow-tan, liquid material was observed. The specimen sent to pathology consisted of multiple fragments of yellow, cheesy material measuring 5 × 4 × 0.5 cm in aggregate dimension. A portion was sent for bacterial, fungal, and mycobacterial cultures. On frozen section, granulomatous inflammation and yeast-like structures were observed, and a tentative diagnosis of fungal infection was made.

Paraffin-embedded sections stained with hematoxylin and eosin revealed lamellar bone and fibrous tissue with areas of necrosis, giant cells and chronic inflammatory cells. No discrete granulomata were identified. Numerous lamellar calcospherites (Liesegang’s rings) were present, some spherical and some elliptical, with varying sizes ranging from 7 to 100 μm (Fig. 2). Alizarin red stain for calcium was positive in these Liesegang’s rings. No microorganisms were identified with routine and modified acid fast stains, Groomori methenamine silver or Brown and Hopps stains.

Culture for fungal, bacterial, and mycobacterial organisms showed no growth, and the patient received no antifungal therapy. He is well and only rarely symptomatic with physical activity 2 years after the procedure.

Discussion

Solitary bone cysts are benign lytic lesions that usually present with localized pain or pathologic fracture and are most often discovered in childhood. 1–4,8–10
FIG. 1 (upper). Radiograph of the left distal radius showing a lytic lesion with central radiodensities suggesting a chondroid matrix.

FIG. 2 (lower). Liesegang’s rings varied in size, from 7 to 100 μm, and in shape, from elliptical to spherical. Brown and Hopps (×100).
The etiology of solitary bone cysts is unknown. Pathogenesis resulting from trauma,9 cystic degeneration of a benign tumor,3 inflammation,10 osteoclast hyperplasia,4 and faulty calcium metabolism8 has been postulated. Grossly, solitary bone cysts are unilocular cysts lined by a thin membrane containing serous or serosanguineous fluid.1,2 Microscopic evaluation of these cysts has revealed a fibrous cyst wall with surface cuboidal lining cells. Following fracture, giant cells, chronic inflammation, fibrous tissue, woven bone, hemosiderin pigment, and cholesterol clefts have been described.2

The solitary bone cyst in our case initially was described as lytic and presented at 16 years of age following a history of trauma. The atypical radiographs 17 years later demonstrated central radiodensities suggesting a chondroid matrix. This correlated with the presence of the lamellar calciospherites on pathologic examination.

The designation Liesegang's rings has been applied by some authors to identify lamellar structures that may be confused with the giant kidney worm, Dioctophyma renale, in tissue sections.5,11,12 These structures are the result of a biochemical process of periodic supersaturation of an insoluble product with alternating diffusion and precipitation around a nidus.6,7 While the calciospherites in our case did not resemble tissue sections of the giant kidney worm, their lamellar appearance conforms to the pattern of precipitation seen in this biochemical process, and they may appropriately be termed Liesegang's rings. Further, an understanding of the pathogenesis of Liesegang's rings makes their presence in the setting of solitary bone cyst reasonable. These cysts contain serous or serosanguineous fluid in which an insoluble product might achieve the supersaturation necessary for the formation of Liesegang's rings.6,7

The microscopic characteristics of Liesegang's rings help to distinguish these bodies from fungal infection or parasitic infestation. In particular, their lamellar configuration, variability in size and shape and positive histochemical staining for calcium help to exclude infectious organisms from the differential diagnosis.

References