Synovial chondromatosis associated with osteopoikilosis—a case report

Hasan Havltçloglu, Izge Günal and Sedat Göçen

Dokuz Eylül University Hospital, Department of Orthopedics, Izmir, Turkey. Correspondence: Dr. I. Günal, Korutürk Mah. Rüzgar Sok. Çankaya Apt. No. 51/13, TR-35330 Balçova, Izmir, Turkey. Tel +90 232-2790329. Fax -2772277
Submitted 98-01-13. Accepted 98-03-23

A 42-year-old man was admitted to our clinic with pain and limitation of motion in his left hip for 10 months. There was no history of injury. Radiography of the pelvis revealed multiple loose bodies in the hip joint and small spherical areas of increased density in both proximal femora, resembling osteopoikilosis (Figure). A skeletal survey confirmed the presence of generalized osteopoikilosis. Scintigraphy revealed increased uptake in the left hip. A biopsy of the synovium of the left hip was performed and histologic studies showed synovial chondromatosis. During the operation, the synovial membrane was excised and the loose bodies were removed. The patient became pain-free, 2 months after the operation.

None of the patient's relatives had either osteopoikilosis or synovial chondromatosis.



Synovial chondromatosis in the left hip and osteopoikilosis in both femora.

Discussion

Osteopoikilosis is a kind of chondrodysplasia with discrete spherical spots of increased radiographic density, 2–10 mm in diameter, and is present mostly in the metaphyses and epiphyses of long bones, carpus, tarsus and membranous bones, such as the pelvis and scapulae (Green et al. 1962, Weisz 1982, Benli et al. 1992, Günal et al. 1993). Although the etiology of osteopoikilosis is obscure, autosomal dominant transmission with various penetrance has been reported (Schorr et al. 1972, Benli et al. 1992, Günal et al. 1993). The disorder is usually symptomless but, because of coexisting developmental variations and pathologic conditions, medical attention is required (Mindell et al. 1978, Ayling and Evans 1988, Grimer et al. 1989, Günal et al. 1993).

Synovial chondromatosis is a rare condition characterized by the formation of chondro-osseous bodies in the synovium and subsynovial joints (Lichtenstein 1977). The condensation of cancellous bone in osteopoikilosis consists of a peripheral area of trabeculae in which osteocysts are scant and there are no osteoblasts and osteoclasts, together with a central core of irregular trabeculae, in which both osteoblasts and osteoclasts are present. The lesions appear to be metabolically active, and they become denser with time, but later their size may change or they may even disappear. The precise origin of these abnormal areas remains debatable, but they appear to represent foci of deranged differentiation in cancellous bone (Verbov 1977, Ayling and Evans 1988).

Copyright © Scandinavian University Press *1998. ISSN 0001–6470. Printed in Sweden – all rights reserved.*

The progression of lesions in synovial chondromatosis has some similarities. In early phases, there is a proliferation of stem cells in the stratum synoviale. Later, cells resembling fibroblasts form a primitive chondroid matrix and they are metabolically active. Then they remain free as a loose body and this condition may be followed by resolution or full resorption. Histological examination shows fibroblasts in the subintimal layer of synovium that undergo cartilaginous metaplasia, giving rise to multiple nodules of hyaline cartilage (Murphy et al. 1962, Milgram 1977, Ono et al. 1994).

If cellular activity exists in the foci of osteopoikilosis, it is conceivable that a malignant lesion could develop (Mindell et al. 1978, Van de Stadt et al. 1984, Grimer et al. 1989). This is also true of synovial chondromatosis (Murphy et al. 1962, Lichtenstein 1977, Milgram 1977, Ono et al. 1994).

In 1977, Verbov described dermatofibrosis lenticularis disseminata as the skin manifestation of osteopoikilosis. By taking into account the similarities, we can speculate that chondromatosis is the synovial manifestation of osteopoikilosis (synosteopoikilosis) and increasing evidence suggests that, when osteopokilosis is detected in a case, other associated disorders, especially lesions of fibroproliferative origin, should be sought.

Ayling R M, Evans P E L. Giant cell tumor in a patient with osteopoikilosis. Acta Orthop Scand 1988; 59 (1): 74-6.

Benli I T, Akalin S, Boysan E, Mumcu E F, Kis M, Türkoglu D. Epidemiological, clinical and radiologic aspects of osteopoikilosis. J Bone Joint Surg (Br) 1992; 74 (4): 504-6.

Green A E, Ellswood W H, Collin J R. Melorheostosis and osteopoikilosis; with a review of the literature. Am J Roentgenol 1962; 87 (6): 109 -111.

Grimer R J, Davies A M, Starkie C M, Sneath R S. Chondrosarcoma in a patient with osteopoikilosis. Rev Chir Orthop 1989; 75 (2): 188-90.

Günal I, Seber S, Basaran N, Artan S, Günal K, Göktürk E. Dacryocystitis associated with osteopoikilosis. Clin Genet 1993; 44 (4): 211-3.

Lichtenstein L. Tumors of synovial joints, bursae and tendon sheaths. In: Bone tumors. (Ed. Lichtenstein L). C.V. Mosby, 5th ed, St. Louis, etc: 1977: 428-52.

Milgram J W. Synovial osteochondromatosis. A histopathological study of thirty cases. J Bone Joint Surg (Am) 1977; 59 (6): 792-801.

Mindell E R, Northup C S, Douglas H O Jr. Osteosarcoma associated with osteopoikilosis: case report. J Bone Joint Surg (Am) 1978; 60 (3): 406-8.

Murphy F P, Dahlin D C, Sullivan R. Articular synovial chondromatosis. J Bone Joint Surg (Am) 1962; 44 (1): 77-86.

Ono H, Yojima H, Fukui A, Tamai S. Locking wrist with synovial chondromatosis: report of two cases. J Hand Surg (Am) 1994; 15 (4): 797-9.

Schorr W F, Wis M, Optiz J M, Reyes C N. The connective tissue nevus- osteopoikilosis syndrome. Arch Dermatol 1972; 106: 208-14.

Van de Stadt J, Thova Y, Spiegl G, Rasquin C, Burny F. Synoviosarcome: presentation d'un cas et revue de la litterature. Rev Chir Orthop 1984; 70 (4): 643-8.

Verbov J. Buschke-Ollendorf syndrome (Disseminated dermatofibrosis with osteopoikilosis). Br J Dermatol 1977; 96 (1): 87-90.

Weisz G M. Lumbar spinal canal stenosis in osteopoikilosis. Clin Orthop 1982; 166: 89-92.

Slow-onset subdural hematoma, evolving into paraplegia, after attempted spinal anesthesia—a case report

Paolo Cabitza and Matteo Parrini

5° Orthopaedic Department of the University of Milan Medical School, Ospedale Clinicizzato San Donato, Milan, Italy. Correspondence: Dr. P. Cabitza, Viale Sabotino, 13, IT-20135 Milan, Italy. Tel +39-2-58319234. Fax -55600234
E-mail: paolo.cabitza@unimi.it
Submitted 97-12-30. Accepted 98-09-22

Epidural and subdural hematomas after spinal anesthesia are rare and are reported mainly as a consequence of needle-induced trauma during a difficult procedure and in patients with abnormal coagulation (Dupeyrat et al. 1990, Sanama et al. 1992, Coquet et al. 1993, Taylor and Major 1993). The first signs of this complication are vague and indistinguishable from the frequent symptoms of meningeal irritation and liquoral hypotension that often follow this type of anesthesia (Tarkkila and Kaukinen 1991, Carpenter et al. 1992, Lynch et al. 1994, Dahlgren and Tornebrandt 1995, Lomartire et al. 1995). Since the onset of an epidural or subdural hematoma can have serious neurological consequences, in the worst case irreversible paraplegia, it is important to recognize the first symptoms.

We observed a rare case of delayed subdural hematoma, which resulted in paraplegia after attempted spinal anesthesia.