



Synovial chondromatosis: a report of four cases with three diverse localizations

Dört olguda üç farklı yerleşim gösteren sinovyal kondromatozis

Nurzat ELMALI, Irfan ESENKAYA, Alpay ALKAN

Inonu University Turgut Ozal Medical Center Orthopaedics and Traumatology Department

Bu çalışmada üç farklı eklemden yerleşim gösteren sinovyal kondromatozisli dört olgu sunuldu. Üçü erkek, biri kadın olan (ort yaş. 32; dağılım 21-55) olgularımızın ikisinde omuz, birinde ayak bileği, bir diğerinde kalça eklemi tutulumu saptandı. Ağrı ve hareket kısıtlılığı tüm olgularda ortak klinik şikayet idi. Diğer klinik şikayetler, tutulan ekleme göre değişmek üzere kilitlenme, krepitasyon, adale güç kaybı, instabilite ve kitle hissi olarak saptandı. Direkt radyolojik incelemede eklem içinde çok sayıda kalsifiye/radyopak cisimler; kalça eklemi tutulumlu kadın olguda ise serbest cisimlere ek olarak bölgesel osteoporoz ve destrüksiyon saptandı. Tüm olgularda (artroskopiyle başlanan olgu dahil) artrotomi sonrası sinoviyektomiyle beraber serbest cisimler çıkartıldı. Tanı histopatolojik incelemeyle doğrulandı. Olguların takiplerinde semptomların iyileştiği görüldü. Nüks oluşmadı.

Anahtar sözcükler: Ayak bileği eklemi; artroskopi; kartilaj, artiküler/patoloji; kondromatozis, sinovyal/tanı/etioloji/cerrahi; kalça eklemi; eklem serbest cisimciği/etioloji/cerrahi/radyografi; omuz eklemi; sinovyal membran; sinovitis.

Four patients with three diverse localizations of synovial chondromatosis are presented. Three were men and one was a woman (mean age 32 years; range 21 to 55 years). The site of involvement was the shoulder joint in two patients, and the ankle and hip joints in the remaining two. Pain and restricted joint movement were common complaints. Other clinical complaints included locking, crepitus, loss of muscle strength, instability, and feeling of a mass lesion. Plain roentgenograms showed multiple radiopaque nodules/loose bodies, with the female patient additionally having osteoporosis and bone erosion. The patients underwent arthrotomy followed by synovectomy and the loose bodies were removed. Diagnoses were confirmed by histologic examination. All the patients became asymptomatic following surgical treatment and no evidence of recurrent disease was detected.

Key words: Ankle joint; arthroscopy; cartilage, articular/pathology; chondromatosis, synovial/diagnosis/etiology/surgery; hip joint; joint loose bodies/etiology/surgery/radiography; shoulder joint; synovial membrane; synovitis.

Synovial chondromatosis may occur in any diarthrodial joint and is usually monoarticular. The knee, hip or elbow joints are most commonly affected. However, it's quite rare in shoulder joint.^[1-7]

Synovial chondromatosis occurs usually in between the ages of 30 and 50 years, is seen rare before puberty and it is twice as frequent in men as in women. If the complaints starting with monoarticular pain and stiffness are not treated, symptoms may cause secondary osteoarthritis by slow progression to decrease in range of motion, effusion, crepitus and repetitive locking, in some cases.^[1-3]

Four cases of synovial chondromatosis, detected in different regions, are presented in this article.

Case reports

Case 1- A 30-year-old male patient referred to hospital with the complaints of pain in left shoulder and limited range of motion. Patient stated that he felt a severe pain in his shoulder while striking the ball when he was playing volley-ball four years ago; he left playing volley-ball as an amateur, as a result of gradually increasing pain by the time and he also stated that he had repetitive locking in his shoulder.

His shoulder muscles were normal in physical examination; however it's seen that he cannot make anterior flexion over 70 degrees or abduction of the left arm. There was pain in shoulder rotations and sensitivity over bicipital groove. Multiple calcifications were detected in shoulder joint and subcapsular recess in radiographic examination (Figure 1a). Multiple loose bodies were detected inside of glenohumeral joint and the sheath of the long biceps tendon in the MR images (Figure 1b). Multiple chondral loose bodies were seen along with distinct synovitis in shoulder arthroscopy (Figure 1c). There were damages in glenoid cartilage in some regions. Partial synovectomy was done by the arthroscopic procedure. After extracting more than thirty loose bodies, because the arthroscopic extraction of all loose bodies would take time, arthrotomy was applied by the anterior deltopectoral approach. All loose bodies were removed by articular lavage and

opening the sheath of the long biceps tendon (Figure 1d). Nonspecific chronic synovitis and cartilaginous loose bodies of hyaline structure associated with a central ossifications varying between 5-15 mm in size were detected in histological examination (Figure 1e). There was no malignancy finding. Symptoms of the patient were absent a year after the surgery and range of motion of the shoulder was nearly normal.

Case 2— A 21-year-old male patient stated that the pain in his right shoulder, started with throwing stone with his right hand two years ago, became permanent by the time without a discomfort and the pain was gradually increased in last three months. Patient was also feeling weakness in his shoulder during overhead activities. It's seen that the deltoid muscle was slightly atrophic and crepitus was detected in the shoulder joint during motion. There were limitations in abduction over 90 degrees and

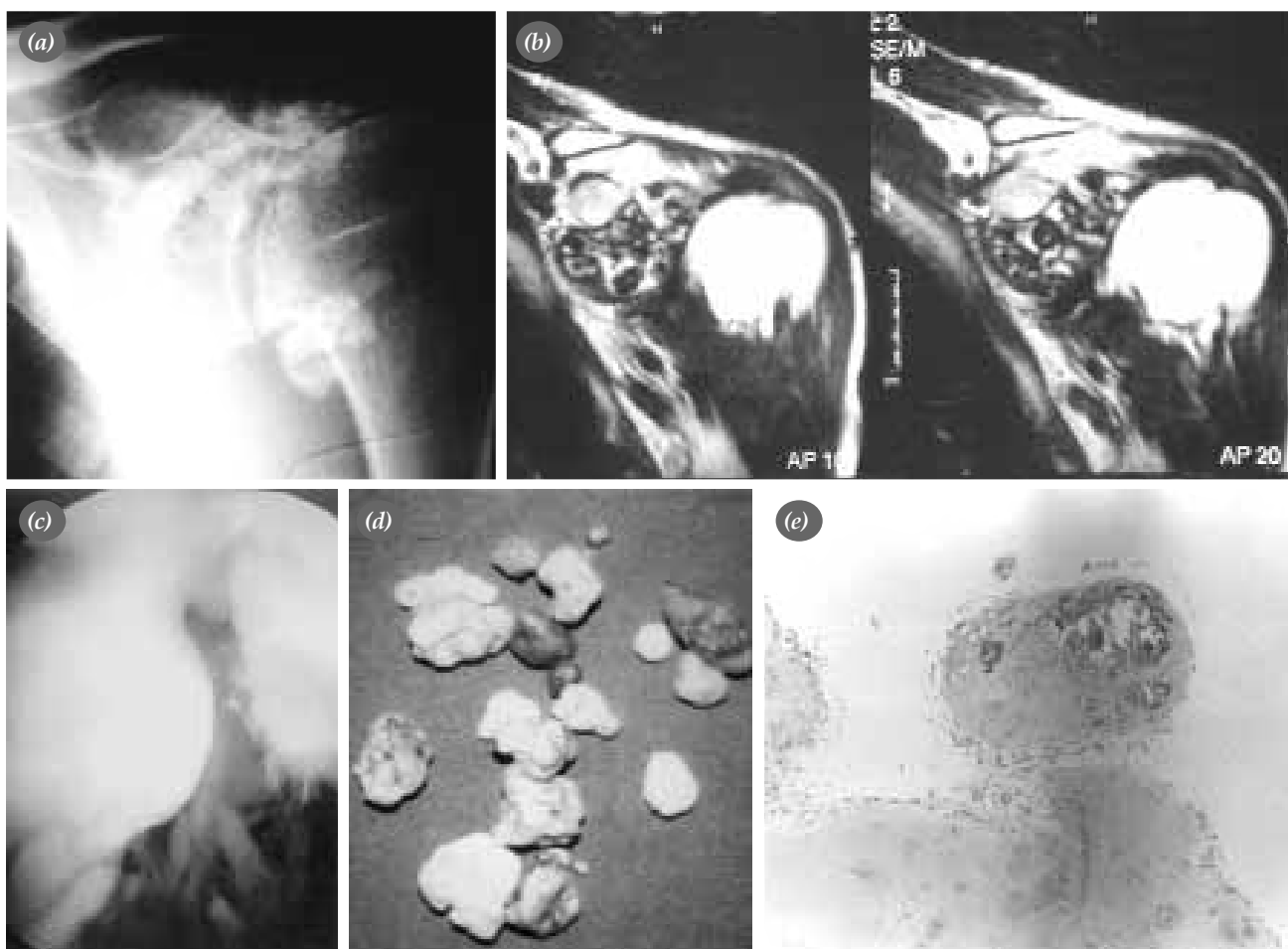


Figure 1. (a) Antero-posterior radiograph, and (b) MR images of the left shoulder of Case 1. (c) Synovial tissue (synovitis) and loose bodies are seen during arthroscopic procedure. (d) Loose bodies extracted from shoulder joint, and (e) their histologic view.

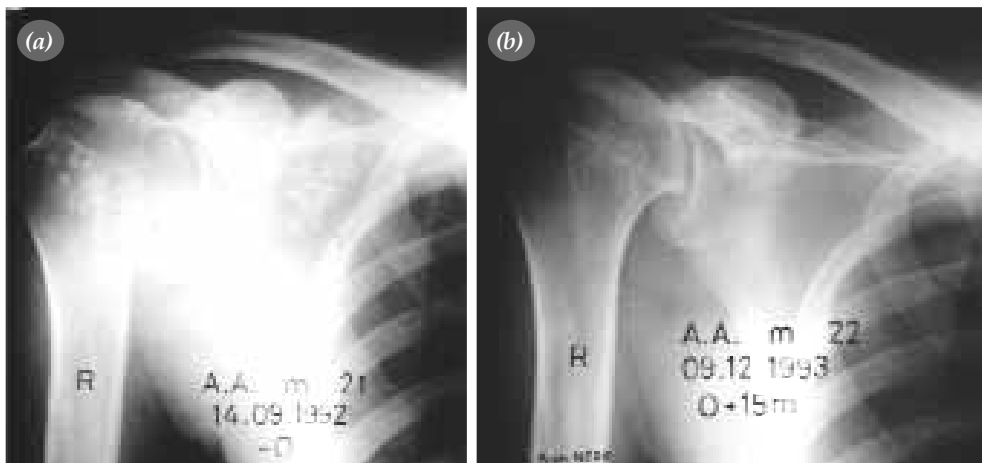


Figure 2. (a) Preoperative, and (b) postoperative antero-posterior radiographs of the right shoulder of Case 2.

internal rotation over 45 degrees. Multiple calcified bodies were detected in plain radiography (Figure 2a). Synovium was excised by anterior deltopectoral (arthrotomy) approach. All loose bodies were removed by articular lavage and opening the sheath of long biceps tendon (Figure 2b). Creamy white colored, multiple typical osteochondral nodules, varying between 0.2-0.6 cm in diameter, were observed. It's seen that the symptoms of the patient were healed in 18 months follow-up after the surgery.

Case 3 – A 55-year-old female patient referred to our clinic with a pain seriously limiting the functions of her right hip. The pain, which was moderate for three years, was increased in last one year. The patient, having limited motion because of the pain, stated that she had locking a few times in her hip. In

the physical examination of the patient, lacking a trauma history; 70 degrees of flexion and limited internal and external rotations were detected because of the pain. A few opaque shades of round bodies, regional osteoporosis and protrusion of femoral head to acetabulum were observed. The evident destruction, detected in femoral head and neck junction, caused the conical view in femoral neck (Figure 3a). An infiltrative and destructive type soft tissue mass was detected in femoral neck and acetabulum, in T1-weighted (T1W) MR images (Figure 3b). Multiple loose bodies were found inside the joint, in arthroscopy performed by posterolateral approach. Loose bodies were extracted by partial synovectomy. And in the end of one year follow-up, symptoms of the patient were healed.



Figure 3. (a) Synovial chondromatosis view in the radiograph of the right hip joint of Case 3. Conical view of the femoral neck, bone erosion and multiple opaque loose bodies at the junction of the femoral head and neck. (b) MR images of the hip joints.



Figure 4. Preoperative (a) antero-posterior and (b) lateral radiographs of the right ankle of Case 4. Postoperative (c) oblique and (d) lateral radiographs.

Case 4 – A 21-year-old male patient referred to our department with the complaints of pain and mass in the dorsal part of his right ankle. The patient, who encountered a soft tissue trauma in his right ankle one year ago, had episodes of instability for few times. Soft tissue swelling and local sensitivity was detected in anterior and anterolateral parts of the tarsal joint in physical examination. Calcific masses were seen on plain radiographs (Figure 4a-b). It's also seen that the articular cartilage was intact in antero-lateral arthrotomy; subsequently loose bodies were extracted and partial synovectomy was performed. The diagnosis was confirmed by the histopathologic

evaluation. No evidence of recurrence was seen during two years follow-up (Figure 4c-d).

Discussion

Synovial chondromatosis, which has an unknown etiology, is defined as a benign lesion by several authors.^[6,8-11] Despite its progressive character, it may occasionally regress. And transforming into chondrosarcoma is also rare.^[8] Davis et al.^[12] stated that 20 of the cases, reported in the literature, are transformed into chondrosarcoma; and stated that three of the 53 cases (5%) are transformed into malignity. Familial cases are also reported.^[13]

Synovial chondromatosis in shoulder is extremely rare.^[1-7] Small and Jaffe,^[14] in a literature review, established only 25 cases with synovial chondromatosis located in the shoulder. Adiyaman et al.^[15] detected three synovial chondromatosis located in the shoulder, in 21 cases. Buess and Friedrich^[7] established an approximate ratio of 5% shoulder location in 95 synovial chondromatosis cases, observed in three distinct studies.

It's stated that intrasynovial metaplasia was essential for the diagnosis of the lesion.^[1] But Milgram^[2] reported that multiple loose bodies can be found without any synovial location of the disease and divided it into three separate phases. According to this, only active intrasynovial diseases are present and free loose bodies are not seen in stage one. Both active intrasynovial disease and free osteochondral (loose) bodies are present in stage two, which is temporary. Multiple free osteochondral bodies are present, an intrasynovial disease doesn't exist or there can be only nonspecific synovitis in third stage.^[2] Our cases, which had detected lesions in shoulder and ankle, were corresponding to stage two of Milgram;^[2] our patient who had hip located lesion was corresponding to stage three. Along with the intraarticular pathology, loose bodies inside the sheath of long biceps tendon were present extraarticularly in our cases with shoulder located lesions.

Intraarticular loose bodies can be viewed in radiographs regarding to the degree of calcification and ossification. However, because the radioopacity cannot be detected 1/3 of the cases, diagnosis becomes more difficult.^[3] Magnetic resonance (MR) is an important modality in diagnosis by its properties like multi planar scanning and excellent evaluation of the soft tissues.^[9] In cases with chronic disease, compression differences on bones and cartilage, which created by multiple cartilaginous lesions, may cause a more destructive radiologic view. Findings of bone erosion, osteoarthritis and regional osteoporosis in radiograms, make the diagnosis harder to achieve.^[10] Despite the negative radiograms, as a result of persistence of severe pain and limited range of motion, magnetic resonance evaluation was performed in our hip located case. After detecting the loose bodies in the observation, calcified loose bodies were found in re-assessed plain radiographs. It's reported that pathologic fractures may develop in some cases.^[6]

Femoral neck erosion, especially, is accepted as the finding of the hip joint synovial disease.^[16]

Treatment modalities are; arthroscopic or open synovectomy, extraction of the loose bodies and arthroplasty application in the presence of severe osteoarthritis.^[2,4,7,11,15,17]

In conclusion, synovial chondromatosis is a pathology that should be kept in mind, in differential diagnosis of the chronic monoarticular pain.

References

1. Gudmundsen TE, Siewers PB. Synovial chondromatosis of the shoulder. A case report. *Acta Orthop Scand* 1987;58:419-20.
2. Milgram JW. Synovial osteochondromatosis: a histopathological study of thirty cases. *J Bone Joint Surg [Am]* 1977;59:792-801.
3. Christensen JH, Poulsen JO. Synovial chondromatosis. *Acta Orthop Scand* 1975;46:919-25.
4. Taşkıran E, Duran T, Tuğran C, Lök V. Kronik ayak bileği ağrısı nedeni olarak sinovyal kondromatozis: Vaka takdimi. *Acta Orthop Traumatol Turc* 1995;29:440-1.
5. Hjelkrem M, Stanish WD. Synovial chondrometaplasia of the shoulder. A case report of a young athlete presenting with shoulder pain. *Am J Sports Med* 1988;16:84-6.
6. Szypryt P, Twining P, Preston BJ, Howell CJ. Synovial chondromatosis of the hip joint presenting as a pathological fracture. *Br J Radiol* 1986;59:399-401.
7. Buess E, Friedrich B. Synovial chondromatosis of the glenohumeral joint: a rare condition. *Arch Orthop Trauma Surg* 2001;121:109-11.
8. Wuisman PI, Noorda RJ, Jutte PC. Chondrosarcoma secondary to synovial chondromatosis. Report of two cases and a review of the literature. *Arch Orthop Trauma Surg* 1997;116:307-11.
9. Kramer J, Recht M, Deely DM, Schweitzer M, Pathria MN, Gentili A, et al. MR appearance of idiopathic synovial osteochondromatosis. *J Comput Assist Tomogr* 1993;17:772-6.
10. Norman A, Steiner GC. Bone erosion in synovial chondromatosis. *Radiology* 1986;161:749-52.
11. Richman JD, Rose DJ. The role of arthroscopy in the management of synovial chondromatosis of the shoulder. A case report. *Clin Orthop* 1990;(257):91-3.
12. Davis RI, Hamilton A, Biggart JD. Primary synovial chondromatosis: a clinicopathologic review and assessment of malignant potential. *Hum Pathol* 1998;29:683-8.
13. Steinberg GG, Desai SS, Malhotra R, Hickler R. Familial synovial chondromatosis: brief report. *J Bone Joint Surg [Br]* 1989;71:144-5.
14. Small R, Jaffe WL. Tenosynovial chondromatosis of the shoulder. *Bull Hosp Jt Dis Orthop Inst* 1981;41:37-47.
15. Adiyaman S, Demirtaş M, Sağlık Y. Sinovyal kondromatozis. *Artroplastik Artroskopik Cerrahi* 1994;8:55-6.
16. Goldberg RP, Weissman BN, Naimark A, Braunstein E. Femoral neck erosions: sign of hip joint synovial disease. *AJR Am J Roentgenol* 1983;141:107-11.
17. Shpitzer T, Ganel A, Engelberg S. Surgery for synovial chondromatosis. 26 cases followed up for 6 years. *Acta Orthop Scand* 1990;61:567-9.