

Arthroscopic Management of Rare case of Synovial Chondromatosis of Hip-A Case Report

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Abstract

Background: We the undersigned declare that this manuscript is original, has not been published before and is not currently being considered for publication elsewhere. We wish to confirm that there are no known conflicts of interest associated with this publication and there has been no significant financial support for this work that could have influenced its outcome. We confirm that the manuscript has been read and approved by all named authors and that there are no other persons who satisfied the criteria for authorship but are not listed. We further confirm that the order of authors listed in the manuscript has been approved by all of us. We confirm that we have given due consideration to the protection of intellectual property associated with this work and that there are no impediments to publication, including the timing of publication, with respect to intellectual property. In so doing we confirm that we have followed the regulations of our institutions concerning intellectual property. We further confirm that any aspect of the work covered in this manuscript that has involved either experimental animals or human patients has been conducted with the ethical approval of all relevant bodies and that such approvals are acknowledged within the manuscript.

Introduction

Synovial Chondromatosis as an independent disease of the synovium was first mentioned in the beginning of the nineteenth century (1813) by Leannac [1]. Primary synovial osteochondromatosis of the hip joint, a rare benign condition, also known as Reichel's disease and was first described by Reichel in 1900, is characterized by multiple intra articular osteochondral loose bodies and synovial hyperplasia which may result in mechanical symptoms and degenerative osteoarthritis. While Secondary synovial chondromatosis is caused by osteoarthritis, trauma or neuropathic arthropathy, it represents an idiopathic benign metaplasia of synovial tissue to cartilage, as there are no cartilage cells in normal synovial membrane [5]. It is described the combined chondromatosis which has both intra-articular and extra articular chondromatosis [6]. It could be different sizes and if it presents a nodule over 1 cm of the diameter it is called giant solitary synovial chondromatosis. [7]

Case Report

A 28-year-old male presented with complain of the left hip pain and pain in left lumbo-sacral region. Pain started approximately 24-30 months earlier, during daytime and during the night. He could not recall any traumatic event. His left hip joint was painful during walking and squatting. During the 2 years, symptoms became more and more severe.

Physical examination of the left hip revealed painful limitation of flexion, abduction and internal rotation compared with

normal range of motions on the contralateral hip. The strength of the quadriceps femoris was slightly reduced, and sensibility was unaffected. Neither thigh muscle atrophy nor leg length discrepancy was found. Radiograph did not show any obvious abnormality. Blood analysis (full blood count, erythrocyte sedimentation rate, C-reactive protein, lactate-dehydrogenase and alkaline-phosphatase) were normal.

Patient was initially managed with NSAIDS and physiotherapy but patient did not have significant relief.

MR imaging of left hip revealed synovial proliferation with multiple nodular lesions and mild synovial effusion causing focal pressure on fovea of femoral head and widening of acetabular fossa. (Figure 1)

The patient underwent arthroscopic debridement and partial resection of hypertrophic synovium and removal of loose bodies in the left hip using standard anterior, anterolateral and posterolateral portal. Patient was placed in the supine position on the fracture table under general anesthesia. Traction of the involved limb at 25° adduction was performed under fluoroscopic assistance. A blunt trocar was placed into the hip joint after sequential dilatation of the tract. An arthroscope with a protecting sheath was inserted, and the hip joint was examined. All visible loose bodies were removed, the synovium was examined for the abnormal area, and these were excised arthroscopically. The appearances of the bodies and the synovium were recorded. Synovial biopsy along with loose bodies was taken and sent for histo-pathological examination.

The histologic examination of the bodies showed lobular masses made up of hyperplastic chondroid tissue. Large cells including some with 2 or more nuclei are seen.

Postoperative period was uneventful, Patient was mobilized with weight bearing as tolerated from day 2 with passive and active hip range of motion exercises were started. Patient was followed up at 1 and 3 months. 3 months later, the patient had achieved total recovery of strength with complete symmetric restoration of motion. Patient had significant decrease in pain and increased range of motion with able to do daily work and

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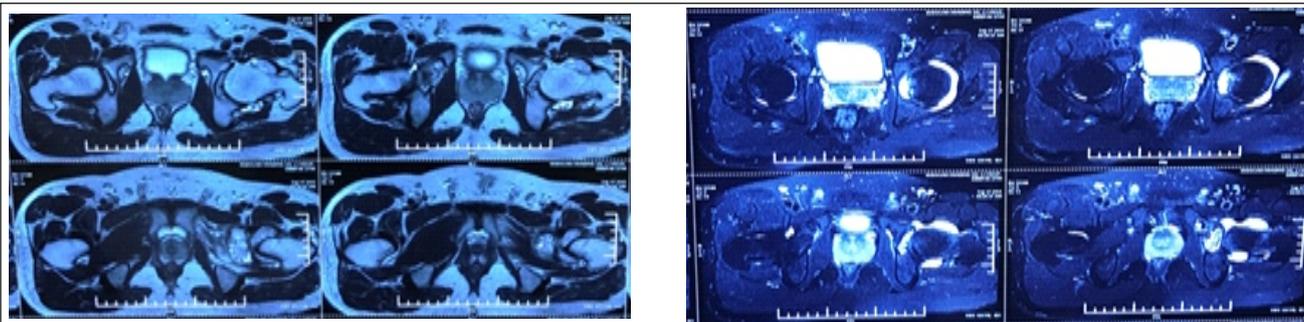


Figure 1: MR imaging of left hip revealed synovial proliferation with multiple nodular lesions and mild synovial effusion causing focal pressure on fovea of femoral head and widening of acetabular fossa.

squat without any pain.

Discussion

PSC of the hip is a very rare disease of patients, mostly aged between 30 and 50 years. It occurs nearly always within the joints, but extra-articular disease has been described (6). There is a predilection in males, with a male-to-female ratio of 1.8:1 (8) and a peak incidence in the fifth decade (9). Most often the presentation is mono-articular and involves the knee, the hip being second most affected. Mussey and Henderson found only 5 cases involving hips in their 105-case series. Maurice et al. reported that 2 of 53 cases of synovial chondromatosis were in the hips. [2,3,4

The aetiology of the disease is still unknown. Many theories have been formulated to explain the origin, but it has so far not been possible to pinpoint an aetiological *primum movens*. There is no family history and usually no convincing history of previous trauma. The pathogenesis of synovial chondromatosis has been assumed to be a reactive, metaplastic process, but the prevalence of well-documented cases of chondrosarcoma originating in synovial chondromatosis gives indirect evidence for a possible neoplastic origin (10). The relative risk of malignant transformation (5%) is quite low at first view, but is much higher than the risk quoted for other well-organised bone diseases predisposing to malignant change (e.g. Paget's disease).

Various theories such as reactivation of residual embryonal cells, traumatic initiation, or benign neoplastic disease have been advocated. The generally accepted pathogenesis of the submesothelial foci of cartilaginous bodies is that they are formed by metaplasia of pluripotential cells in the synovial membrane. (11) These nodules can ossify by endochondral bone formation and attach to the synovium by a thin vascular pedicle. They may break free and become loose bodies in the joint space, and if nourished by synovial fluid, they can continue to proliferate. A recent study showed the presence of transforming growth factor-beta (TGF) and tenascin (TN) in synovial chondromatosis. Another study revealed that fibroblast growth factor receptor 3 (FGFR 3), a specific marker of mesenchymal precartilaginous stem cells, was expressed in primary synovial chondromatosis,

and that elevated levels of fibroblast growth factor 9 (FGF 9), a specific ligand of FGFR 3, had been found in synovial fluids of synovial chondromatosis. (12)

Werner et co-workers in a study on chondromatosis of the elbow, found the disease tends to affect the dominant extremity, which leads to the assumption that biomechanical stress might be a factor in the formation of the SC [13]. Hereditary transmission might be another factor, as several authors increased familial clustering [13,14].

Clinical symptoms are non-specific and a clinical diagnosis of synovial chondromatosis of the hip is difficult and it may be delayed. Pain, stiffness, limited motion, clicking, locking, or limping from the affected hip may be present. Examination may display signs of femoroacetabular impingement (FAI) as PSC is sometimes associated with FAI or disease-specific loose bodies or swelling of the synovium can imitate impingement.

If this disorder is untreated or not recognized early, late complications such as secondary degenerative osteoarthritis, capsular constriction, subluxation of the hip, or pathologic femoral neck fracture may follow.

Plain radiographs in the early stages are usually negative until the osseous bodies become evident in the joint. In 1/3 of cases, no radio-opacity appears, although osseous particles exist. Other imaging modalities, including arthrography, ultrasound, computed tomography, or magnetic resonance imaging may demonstrate this disorder better. Malignant transformation of synovial chondromatosis into a chondrosarcoma is unusual and has been reported only sporadically. (15) A cell proliferative activity study concluded that primary synovial chondromatosis appeared to occupy a position which is intermediate between benign enchondroma and malignant chondrosarcoma, (16) which may explain the occasionally aggressive clinical behavior. Clinical features are not helpful in differentiating these 2 entities.

However, sudden clinical deterioration in long-standing cases, bony destruction by imaging study, or cases with recurrent synovial chondromatosis should alert the clinician to the possibility of malignant transformation. (15) According to PSC's distribution in resected biopsy material, Milgram differentiated between 3 stages of disease [17]. In

the first stage, metaplastic transformation is restricted to the synovium, whereas stage 2 additionally shows intraarticular loose bodies. In stage 3, the synovial tissue may appear normal again with loose bodies as the only sign of PSC.

MR imaging allows for a visualisation of uncalcified lesions or lesions in the initial phase, without the presence of loose bodies.

According to Kramer, there are three different MRI patterns, depending on the presence and distribution of various synovial Chondromatosis components and the stage at which the process is being imaged. Pattern A shows multinucleate cells, isointense or slightly hyperintense to muscle on T1WI and hyperintense on T2WI. Pattern B is similar to pattern A but with additional foci of signal voids due to presence of calcification. Pattern C shows foci of peripheral hypointensity and central fat signal intensity secondary to ossifications. Gadolinium-DTPA may be used for assessment of the synovial activity and to differentiate early synovial chondromatosis from a simple joint effusion [18,19,20].

The histology of synovial chondromatosis shows cellular hyaline cartilage arranged in small clusters separated by fibrous tissue. Chondrocytes may show minor cellular atypia with irregular hyperchromatic nuclei and occasionally binucleate or multi-nucleate cells. These changes may mimic malignancy and can lead to the erroneous diagnosis of chondrosarcoma with subsequent implications of treatment. Bertoni et al. in 1990 described histological criteria to differentiate between benign and malignant transformation of synovial chondromatosis thus: malignant change is characterised by marked cellular atypia; chondrocytes are arranged in sheets with loss of cluster appearance, presence of necrosis, mitoses and myxoid features and invasion into adjacent tissues. Besides standard histological staining, immuno-labeling can be added to the diagnostic investigation.

Because of the rareness of PSC, all studies are retrospective and predominantly include small collectives of patients. In consideration of the analyzed study data, arthroscopic removal of loose bodies and partial synovectomy has been recommended. Minimal-invasive surgery has been proven to shorten the in-hospital stay and postoperative rehabilitation with possible fully-weight bearing. Major complications with significant consequences are seen in connection with surgical hip dislocation. As most patients are young, a necrosis of the femoral head leading to total hip arthroplasty is not acceptable. Total synovectomy as an advantage of open approach may reduce recurrence rate, but does not justify the potential risks related to surgical hip dislocation. Boyer and Dorfmann could illustrate the good clinical outcome of arthroscopic PSC treatment in a numerically powerful study with a remarkable mean follow up of 79 months [21]. We strongly recommend to consider hip arthroscopy as preferred operation technique for arthroscopically experienced surgeons. Because re-operation is needed more

frequently, an accurate follow up is essential, but also sufficient to detect relapse early. Open approaches should be assigned to disease recurrence or disappointing arthroscopic results. Only if total synovectomy is aspired, synovial chondrosarcoma is highly suspected or total hip arthroplasty is considered as an exit strategy [22,23], open surgery is justified as first-line technique. Even in cases of osteoarthritis, arthroscopy is a valuable option to reduce pain and improve mobility and quality of life, as shown in our case report above.

Arthroscopic synovectomy has been shown to be a safe and effective method of synovial ablation, and when feasible, allows faster rehabilitation. Kim et al (24) report excellent results with arthroscopic removal of loose bodies and partial synovectomy in 4 patients with synovial chondromatosis. Hip arthroscopy with thorough removal of loose bodies and subtotal synovectomy, coupled with an aggressive and early rehabilitation program, is an effective way of ameliorating symptoms from hip SC, yielding high levels of patient satisfaction and functional outcomes

Because various recurrence rates from 0% to 15% have been reported, the optimal treatment for primary synovial chondromatosis of the hip is still controversial. Based on the pathogenesis of primary synovial chondromatosis and 1 study which revealed that a synovectomy had a significantly lower recurrence rate, the recommended management is surgical removal of the loose bodies combined with a partial or complete synovectomy in most cases. (24,26,27) Complete removal is only possible with dislocation of the hip joint, but a synovectomy combined with dislocation of the femoral head may result in avascular necrosis of the femoral head. Therefore dislocation of the hip joint is now considered obsolete. With advances in arthroscopic surgeries, an arthroscopic operation of the hip joint with synovial chondromatosis can be a reliable procedure.

References

1. Fanburg-Smith JC. Cartilage and bone-forming tumors and tumor-like lesions. In: Miettinen M, ed. *Diagnostic soft tissue pathology*. Philadelphia, Pa: Churchill-Livingstone, 2003; 403–425.
2. Mussey RD Jr, Henderson MS. Osteochondromatosis. *J Bone Joint Surg Am* 1949; 31A (3):619-627.
3. Maurice H, Crone M, Watt I. Synovial chondromatosis. *J Bone Joint Surg Br* 1988; 70(5):807-811.
4. Shpitzer T, Ganel A, Engelberg S. Surgery for synovial chondromatosis 26 cases followed up for 6 years. *Acta Orthop Scand* 1990; 61(6):567-569.
5. Edeiken J, Edeiken BS, Ayala AG, Raymond AK, Murray JA, et al. (1994) Giant solitary synovial chondromatosis. *Skel Radiol* 23: 23-29.
6. Sviland L, Malcolm AJ (1995) Synovial chondromatosis presenting as painless soft tissue mass: a report of 19 cases. *Histopathology* 27: 275-279.
7. Springer KR (1991) Synovial chondromatosis. *J Foot Surg* 5: 446-449.
8. Davis RI, Hamilton A, Biggart JD. Primary synovial chondromatosis: a clinicopathologic review and assessment of malignant potential. *Hum Pathol* 1998; 29: 683-688.
9. Taconis WK, van der Heul RO, Taminiou AM. Synovial chondrosarcoma: report of a case and review of the literature. *Skeletal Radiol* 1997; 26: 682-685.
10. Hermann G, Klein M, Abdelwahab I et al. Synovial chondrosarcoma arising in synovial chondromatosis of the right hip. *Skeletal Radiol* 1997; 26: 366-369.
11. Hardacker J, Mindell ER. Synovial chondromatosis with secondary subluxation of the hip. *J Bone Joint Surg Am* 1991; 73:1405-7.
12. Robinson D, Hasharoni A, Evron Z, Segal M, Nevo Z. Synovial chondromatosis: the possible role of FGF 9 and FGF receptor 3 in its pathology. *Int J Exp Pathol* 2000; 81:183-9.
13. Werner A, Wild A, Mueller T, Borys A, Gohlke F, et al. (2002) Primary synovial chondromatosis of the shoulder. *Z Orthop Ihre Grenzgeb* 140: 404-408.
14. Steinberg GG, Desai SS, Malhotra R, Hickler R (1989) Familial synovial chondromatosis: brief report. *J Bone Joint Surg Br* 71: 144-145.
15. 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.
16. Davis RI, Foster H, Arthur K, Trewin S, Hamilton PW, Biggart DJ. Cell proliferation studies in primary synovial chondromatosis. *J Pathol* 1998; 184:18-23.
17. Milgram JW. Synovial osteochondromatosis. A histopathological study of thirty cases. *J Bone Joint Surg*. 1977; 59:724–792.
18. Crotty JM, Monu JU, Pope TI Jr (1996) Synovial osteochondromatosis. *Radiol Clin North Am* 34: 327-342.
19. Wittkop B, Davies AM, Mangham DC (2002) Primary synovial chondromatosis and synovial chondrosarcoma: a pictorial review. *Eur Radiol* 12: 2112-2119.
20. Ko E, Mortimer E, Fraire AE (2004) Extraarticular synovial chondromatosis: review of epidemiology, imaging studies, microscopy and pathogenesis, with a report of an additional case in a child. *Int J Surg Pathol* 12: 273-280.
21. Boyer T, Dorfmann H. Arthroscopy in primary synovial chondromatosis of the hip: description and outcome of treatment. *J Bone Joint Surg Br*. 2008; 90(3):314–318.
22. Ligato A, Nelson S, Bengs BC. Hip resurfacing as treatment for synovial chondromatosis. *Orthopedics*. 2010 Mar; 33(3). doi: 10.3928/01477447-20100129-27.
23. Ackerman D, Lett P, Galat DD Jr, Parvizi J, Stuart MJ. Results of total hip and total knee arthroplasties in patients with synovial chondromatosis. *J Arthroplasty*. 2008; 23(3):395–400.
24. Kim SJ, Choi NH, Kim HJ. Operative hip arthroscopy. *Clin Orthop* 1998; 353: 156-165.
25. Ogilvie-Harris DJ, Saleh K. Generalized synovial chondromatosis of the knee: a comparison of removal of the loose bodies alone with arthroscopic synovectomy. *Arthroscopy* 1994; 10:166-70.
26. Gilbert SR, Lachiewicz PF. Primary synovial osteochondromatosis of the hip: report of two cases with long-term follow-up after synovectomy and a review of the literature. *Am J Orthop* 1997; 26:555-60?
27. Knoeller SM. Synovial osteochondromatosis of the hip joint. Etiology, diagnostic investigation and therapy. *Acta Orthop Belg* 2001; 67:201-10.

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