Open Journal of
Clinical & Medical
Case Reports

Volume 2 (2016)
Issue 14

ISSN 2379-1039

Neck of Femur Fracture Secondary to Synovial Chondromatosis: A Case Report and Literature Review

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Abstract

A 28 years old lady presented with complaint of chronic left hip pain for 4 years, which increased with exertion and relieved with rest and medication and referred to the left knee. There was no history of fever, pain at any other site or difficulty in walking. On local examination there was mild tenderness over left anterior hip associated with slight restriction of terminal movements. Plain radiograph of pelvis with bilateral hip joints revealed erosion on the inferomedial aspect of the femoral neck. MRI revealed a synovial lesion around neck. Needle biopsy was done from this site and soft tissue mass under C-arm guidance. Report of the biopsy was inconclusive. Post biopsy patient sustained trivial trauma while ambulation followed by severe pain in the affected hip. Plain radiograph revealed fracture neck femur. An excision biopsy with uncemented modular bipolar hemiarthroplasty was planned. Intraoperatively multiple rice bodies were seen in the left hip joint. Synovial lesion located in the inferior aspect had eroded the head and neck of the femur. Frozen section revealed synovial chondromatosis. After excision biopsy of the lesion, uncemented modular bipolar hemiarthroplasty was done. Postoperative course and follow up was uneventful.

The purpose of this case report is to bring out a very rare presentation of synovial chondromatosis of hip.

Keywords

synovial chondromatosis; hip joint; pathological fracture

Introduction

Synovial chondromatosis is a rare disorder in which multiple cartilaginous nodules appear in the synovium and sub synovial connective tissue. The disease is believed to be a benign chondral metaplasia of the synovial membrane with multiple foci [1-4]. The growing nodules in the synovial membrane may become detached and enlarged, due to nourishment by synovial fluid. It can occur in any joint but usually involves large joints, such as the knee. Extra-articular involvement is rare. These nodules may calcify or ossify and occasionally appear as radiopaque loose bodies around the joint on plain radiography.

Primary synovial chondromatosis of the hip joint is rare, and the optimal treatment is still controversial [4]. Removal of the loose bodies only, a radical synovectomy, an open synovectomy with removal of the loose bodies, and an arthroscopic synovectomy with removal of the loose bodies have all been reported. Open hip surgery with joint dislocation is an accepted treatment; however, the recovery periods may be long, with a high risk of complications like osteonecrosis of the femoral head (if a

meticulous surgical dislocation is not followed), neurovascular injury, deep vein thrombosis and wound infection. Arthroscopic approach to this condition has shown good results. A truly total synovectomy is not possible with this technique, but it is possible to remove loose bodies, improve symptoms and possibly delay the development of complications [1-4,6-8].

If this disorder is not recognized early or left untreated, late complications such as secondary degenerative osteoarthritis, capsular contracture, subluxation of the hip, or pathologic femoral neck fracture may follow which may necessitate other procedures like hiparthroplasty [9,10,20,21]. We describe here, one such case, in which it led to erosion and pathological fracture of femoral neck.

Case Report

A 28 years old female presented with complaint of chronic left hip pain for 4 years. The pain used to increase with exertion and relieved with rest and medication. There was also referred pain to left knee. There was no history of fever, pain at any other site or difficulty in walking. General and systemic examinations were normal. On local examination there was mild tenderness over left anterior hip associated with slight restriction of terminal movements. Active SLR (straight leg raising) was present bilaterally. However, the passive range of motion was slightly increased. There was no distal neurovascular deficit.

Plain radiograph of pelvis with bilateral hip joint revealed erosion on the inferomedial aspect of the femoral neck (fig 1a and 1b). Magnetic resonance imaging revealed a synovial lesion/hypertrophy encircling the femoral neck with thinning and scalloping of the neck especially on the inferomedial aspect. Blood investigations were normal. Needle biopsy was done from this site and the underlying soft tissue under C-arm guidance. Report of biopsy was inconclusive. Post biopsy patient sustained trivial trauma while ambulation. Plain radiograph revealed left fracture neck femur (removal of a core of bone from the inferomedial aspect of femoral neck might have further weakened the already scalloped neck and created a stress riser effect which also might have contributed to fracture occurrence)(fig 2).

An excision biopsy with uncemented modular bipolar hemiarthroplasty was planned. The hip was exposed through posterior approach. Intraoperatively multiple rice bodies were seen in the hip joint. Synovial lesion located in the inferior aspect had eroded the head and neck of the femur. A large number of chondral loose bodies (rice bodies) were removed from the hip joint. Partial synovectomy was performed.

On gross pathological examination of the synovial tissue and loose bodies, cartilaginous structures of different soft and hard consistencies were noted lying loosely in the pieces of synovium.

Frozen section revealed synovial chondromatosis. After excision biopsy of the lesion, uncemented modular bipolar hemiarthroplasty was done (fig 4). Postoperative course and follow up was uneventful.

Microscopically the structure of soft consistency showed edematous connective tissue covered with synovial cells. In the connective tissue lymphocytic infiltration was noted. The loose bodies consisted of cartilaginous tissue. In the synovium the chondrocytes showed vesicular cytoplasm and hyper chromatic nuclei. In places, the cartilage was covered by a cellular synovial tissue (fig 3). Final histopathology thus confirmed the diagnosis of synovial chondromatosis.

Discussion

Synovial chondromatosis or osteochondromatosis (when ossification is present), also called Reichel's syndrome, was first described by Reichel in 1900 [6]. The etiology of this disorder is still unclear. Various theories such as reactivation of residual embryonal cells, traumatic initiation, or benign neoplastic change have been considered [9]. The generally accepted pathogenesis is that there occurs multifocal cartilaginous meta- plasia of pluripotent cells in the synovial membrane [10]. These nodules can ossify by endochondral bone formation. They may break free and become loose bodies in the joint space, and if nourished by synovial fluid, they can continue to proliferate. Transforming growth factor beta (TGF) and tenascin (TN) has been demonstrated in synovial chondromatosis. [11] TGF beta increases mesenchymal cell differentiation, proteoglycan production, chondroblast replication, and extracellular matrix protein production. TN induces chondrogenesis and osseous transformation in chondral foci. Also, fibroblast growth factor receptor 3 (FGFR 3), a specific marker of mesenchymal precartilaginous stem cells, was expressed in primary synovial chondromatosis, and elevated levels of fibroblast growth factor 9 (FGF 9), a specific ligand of FGFR 3, had been found in synovial fluids of synovial chondromatosis cases [9]. These are absent in normal synovium and cartilage and may explain the pathogenesis of synovial chondromatosis.

Primary synovial chondromatosis should be differentiated from cartilaginous loose bodies that are secondary to other joint diseases such as degenerative arthritis. In an immunohistochemical study of growth potential, [13] loose bodies in primary synovial chondromatosis show plump chondrocytes and irregular calcification, and all have proliferative cell nuclear antigen (PCNA)-positive chondrocytes, whereas loose bodies in secondary synovial chondromatosis show uniform chondrocytes and annular calcification surrounding the core tissue. Only half of them have PCNA-positive chondrocytes peripherally.

Three phases have been described for this disease [14]. In the first active phase, the disease is limited to the synovium without loose body formation; in the second transitional phase, there are both loose bodies and intrasynovial lesions; and in the third quiescent phase, there are only free loose bodies without active intrasynovial process. The disease is most commonly seen in the 3rd to 5th decades, and affects men and women in a ratio of about 2:1 [9]. The most common joint involved is the knee, but the elbow, hip, shoulder, ankle, temporo- mandibular joints, and other small joints have also been described. Extra-articular involvement (bursa or tendon sheath) is extremely rare.

Synovial chondromatosis of the hip is uncommon. Mussey and Henderson found only 5 cases involving hips in their 105-case series [15]. Maurice et al. reported that 2 of 53 cases of synovial chondromatosis were in the hips [16].

Fairbank [17] has described indentation of the neck of the femur by osteochondromata. Erosion of the neck of the femur was also noted by Freund and Friedman [18, 19]. A conical appearance of the femoral neck has also been described.

Pain, stiffness, restricted range of motion, clicking, locking, or limping from the affected hip may be the presenting complaints. If this disorder is not recognized early or left untreated, late complications such as secondary degenerative osteoarthritis, capsular contracture, hip subluxation, or pathologic

femoral neck fracture may be the sequale [9,10,20,21].

To the best of our knowledge only two cases of pathological fracture as a complication of synovial chondromatosis of the hip have been reported in the literature. Szyprt and colleagues reported a case of pathological fracture of the femoral neck caused by weakening due to extensive pressure erosions by synovial chondromatosis for the first time in 1986 and second such case was reported by Sakellariou A et al who managed their case (a 89 years old lady)by cemented arthroplasty [20,21]. We performed an uncemented one, as our patient was a young female. If this disease is recognized early and treated promptly by debridement and synovectomy, such complications may be avoided.

Conclusion

Here we have reported a very rare case of neck of femur fracture secondary to synovial chondromatosis, treated by uncemented hemiarthroplasty. Early recognition of this disease and prompt treatment by debridement and synovectomy, by either open or arthroscopic means, may prevent such complications.

Acknowledgement and Conflict of Interest

The authors would like to thank faculty members Dr Sharma MK, Dr Gupta AK, and staff members who contributed to the management of this case. No source of support or conflict of interest to be disclosed.

Figures

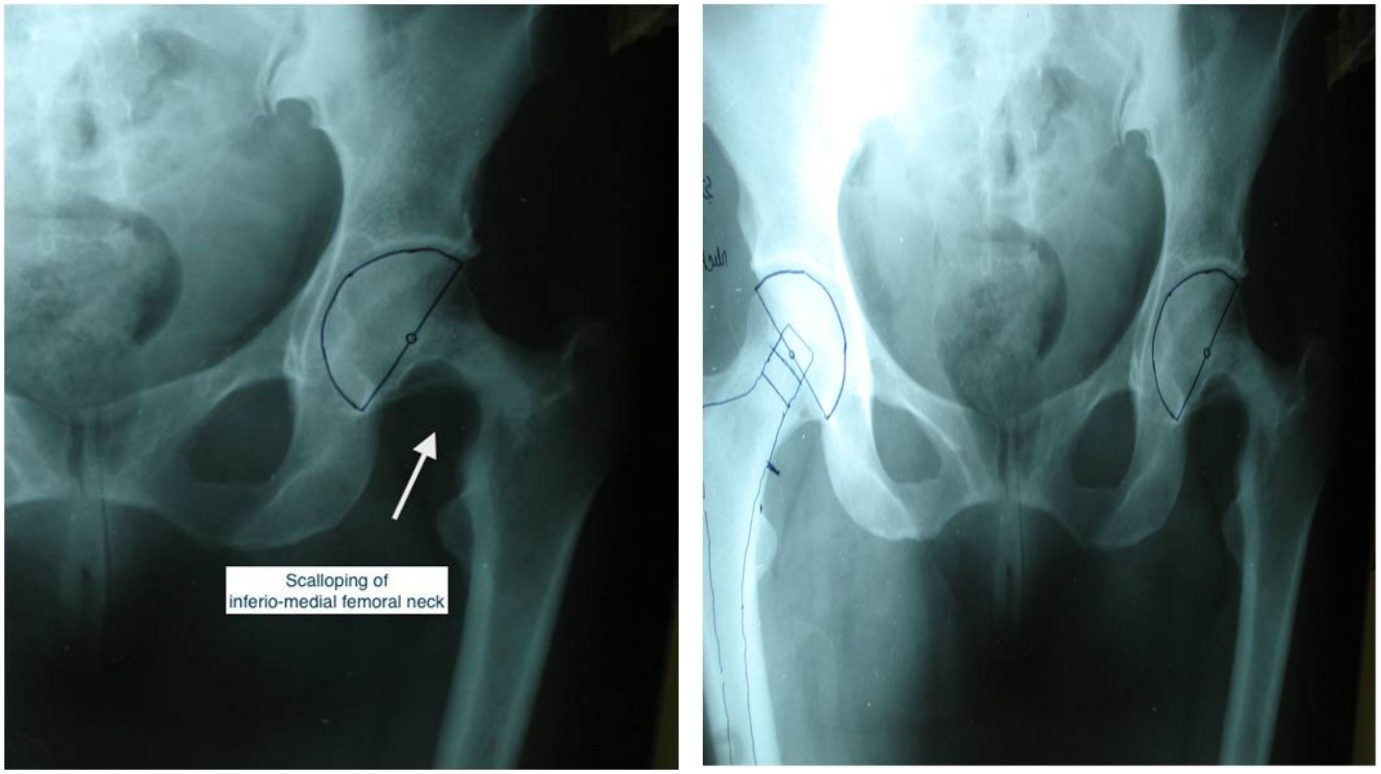

Figure 1(a,b): Neck eroded/scalloped inferiomedially (arrow).Tracing was done for measuring femoral head size (templating) for hemiarthroplasty preoperatively

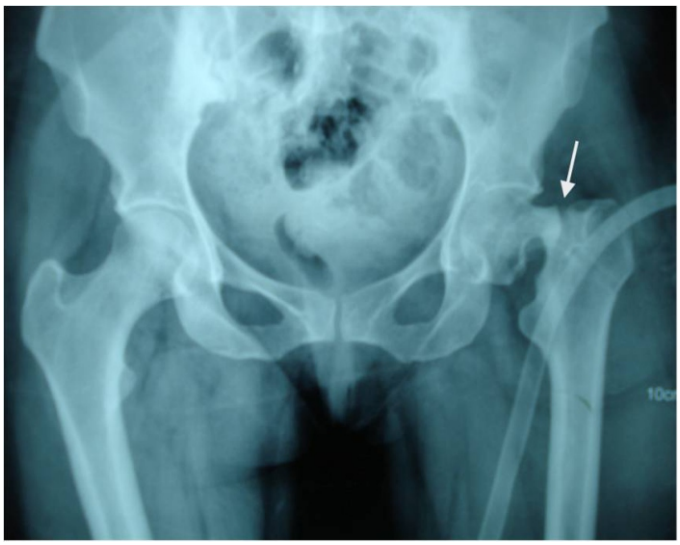

Figure 2: Pathological fracture neck femur (arrow).

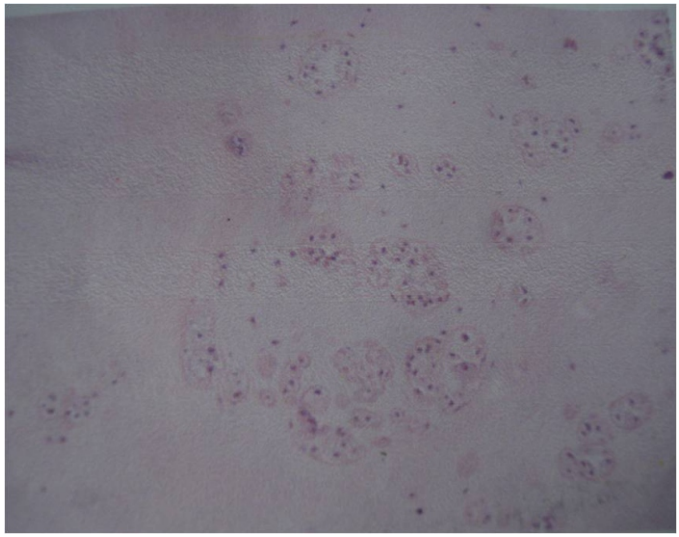

Figure 3: Microscopy-multiple nodules of hyaline cartilage within synovial membrane.

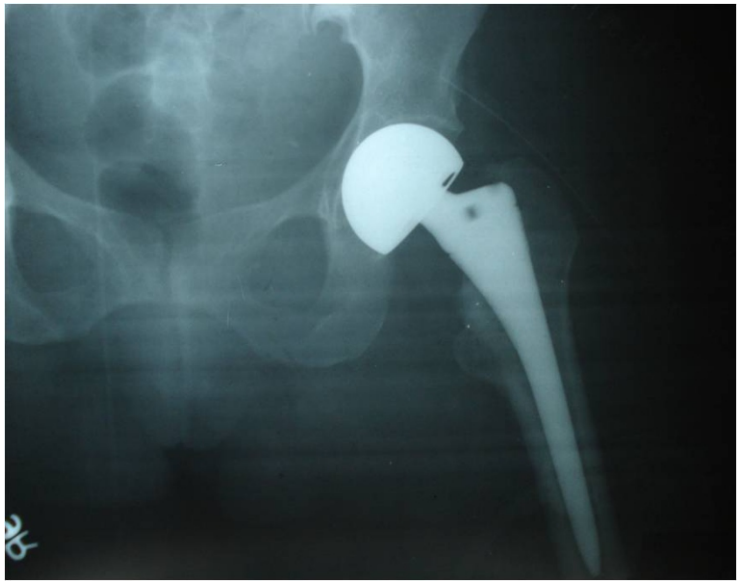

Figure 4: Post operative radiograph after hemiarthroplasty.

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Manuscript Information: Received: March 28, 2016; Accepted: July 26, 2016; Published: July 29, 2016

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Citation: Patralekh M, Yadav VP, Sambharia A, Kumar V. Neck of femur fracture secondary to synovial chondromatosis, a case report and literature review . Open J Clin Med Case Rep. 2016; 1144

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