

Primary synovial chondromatosis of the hip joint (PrSC of the hip): A retrospective cohort analysis and review of the literature

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ABSTRACT

Introduction: Primary synovial osteochondromatosis (SOCM) or Synovial chondromatosis (SC) of the hip is a benign metaplastic condition of the synovium that is rare and may present with a spectrum of clinical features and radiological findings.

Patients and methods: A retrospective search using the keyword 'Synovial chondromatosis' (SC) of the hip was performed at a tertiary care orthopaedic referral centre in the UK and a hospital in India. The radiology images were collected from our Picture Archiving and Communication System (PACS) and Radiology Information System (RIS), over 15 years. The patient's data was collated with Electronic Patient Records (EPR), RIS, and correlated with histo-pathology laboratory records where available. The demographic details of the patients, their clinical symptoms, imaging details, and management outcomes were collected.

Results: We found 15 cases, with a mean age of 36.53 years (range: 14–50 years). There were 9 male and 6 female patients. The follow-up ranged from 1 year to 6 years. Predominantly unilateral presentation with insidious onset of symptoms was found. A spectrum of radiological imaging was undertaken. Management strategies included supervised observation, arthroscopic or open synovectomy, and hip arthroplasty. No malignant transformation was found in the analysed cohort.

Conclusion: Primary 'Synovial chondromatosis' of the hip had a male preponderance in our cohort, presenting with a range of clinical features. Radiologically, Magnetic Resonance Imaging (MRI) was the commonest modality of cross-sectional imaging utilised and crucial for the diagnosis, evaluating underlying articular involvement including guiding appropriate patient management presenting with Primary 'Synovial chondromatosis' of the hip.

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1. Introduction

Synovial chondromatosis (SC) is a rare, benign proliferative condition of the synovium associated with cartilage metaplasia resulting in the development of multiple loose bodies.^{1,2} SC is usually a mono-articular condition, predominantly affecting the knee joint (>50%) followed by the hip, elbow, ankle, shoulder, and wrist joints.^{3,4} SC characteristically is seen in the third to fifth decades of life, with men reported being affected up to two to four times more commonly than females.⁵

Primary synovial chondromatosis (also known as Reichel syndrome or Reichel-Jones-Henderson syndrome) is characterised by synovial metaplasia of unknown aetiology. Various theories such as raised bone morphogenic protein (BMP), interleukin-6 (IL-6), and vascular endothelial growth factor-A (VEGF) and trauma leading to the synovial metaplasia and formation of loose bodies in Primary synovial chondromatosis have been suggested but yet unproven.⁶

Microscopically, SC is characterised by the formation of multiple cartilaginous proliferating nodules. These nodules detach and form nodules within the joints or surrounding soft tissue.⁷ Erosion of articular surfaces and mechanical wear of joint surfaces leads to the development of features of osteoarthritis (OA).⁸ These changes lead to the development of typical articular symptoms of pain, locking, and limitations of joint movements.

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Primary 'Synovial chondromatosis' of the hip (PrSC of the hip) is an uncommon condition, and its prevalence is not exactly known.⁹ Diagnosis of SC of any joint is facilitated by imaging studies like plain Radiographs, Magnetic Resonance Imaging (MRI), and Computed Tomography (CT).¹⁰ Radiological features depend on the stage of disease and the extent of calcification or ossification of the cartilaginous nodules. Plain radiographs may reveal soft tissue swelling, joint erosion, and a characteristic appearance of multiple small, juxta-articular radio-opaque nodules.¹¹

MRI and CT scans are valuable in distinguishing primary from secondary SC.¹² The need to differentiate primary and secondary SC is essential to address underlying arthritis processes and allow for an appropriate patient management plan.^{13,14} PrSC of the hip needs to be differentiated from other inflammatory joint diseases like synovial proliferative disorders, crystal arthropathy, tuberculosis, rheumatoid arthritis (RA), malignancies like chondrosarcoma, and isolated acetabular osteochondroma.¹⁵

Since the PrSC of the hip is a rare condition, there is a paucity of knowledge about this condition and its management protocols. An optimal surgical approach and treatment options remain controversial.¹⁶ Several procedures have been described to manage it including joint debridement and synovectomy using open arthrotomy with or without hip dislocation, minimally invasive surgery, or by arthroscopic surgery. In the advanced stage of the disease, hip arthroplasty is advised.^{17–21}

The current article emphasizes the uncommon nature of the PrSC of the hip. We provide and highlight our experience in the management of this condition, the radiological evaluation, and MRI findings at a Tertiary Referral centre. This retrospective analysis is discussed in light of available, published literature.

2. Patients and Methods

2.1. Study design

A retrospective evaluation of our Radiology Information System (RIS) and Picture Archiving and Communication System (PACS) at our tertiary orthopaedic referral centre in the UK and super speciality hospital in India using the keywords 'Synovial chondromatosis' (SC) of the hip for the last 15 years. Local committee approval was obtained for this study.

2.2. Data collection

The demographics of patients, clinical symptoms, imaging findings, and management were documented.

2.3. Inclusion and exclusion criteria

Patients with underlying causes (e.g. OA, Inflammatory joint disease, trauma) leading to the secondary development of 'Synovial chondromatosis' (SC) of the hip were excluded. It has allowed a pooled analysis of patients with the diagnosis of Primary 'Synovial chondromatosis' of the hip (PrSC of the hip).

2.4. Image analysis

The senior author (RB) and an experienced MSK Radiologist reviewed all the radiological images. Radiographs, MRI, and complementary imaging sequences of Computed Tomography (CT) were analysed. MRI involved a combination of T1, PD, and STIR coronal, T1, and PDFS coronal sequences. CT where available involved axial with multiplanar reformats. Radiological analysis was undertaken to assess the laterality of the condition (Unilateral vs Bilateral), presence of underlying articular changes, and

extension into the iliopsoas bursa. Loose bodies were seen as high signal on STIR, PDFS if non calcified/ossified and low signal on all sequences if calcified/ossified. Calcified or ossified loose bodies were seen as ossified lesions on radiographs and CT. Location of 'loose bodies' were noted and extension of the SC in the surrounding soft tissues in particular iliopsoas bursa was recorded. The details of clinical management and surgical intervention were collected over the follow-up period for each patient included in the study.

2.5. Statistical methods | data analysis

The clinical and investigation data was collated using a Microsoft Excel datasheet. Data was analysed quantitatively with descriptive statistics for preparing a Tabular representation of key features in managing patients with PrSC of the hip.

3. Results

We found 15 patients attending with PrSC of the hip. The mean age of 36.53 years (range: 14–50 years). We noted a male predominance (6 females and 9 males). It was unilateral in the majority (13/15) with a roughly equal proportion of right and left hip involvement (6 right and 7 left). There was the insidious onset of symptoms with pain, locking, and restricted range of movements as presenting features. None of the patients had any history of previous radiation to the pelvis.

Radiographs were the initial modality used in all patients to diagnose synovial osteochondromatosis. CT was performed in 5 cases and 11 had MRI. (Figs. 1 and 2). Predominantly unilateral presentation with insidious onset of symptoms was found. On MRI calcified loose bodies demonstrated low signal on all sequences. Nonossified/calcified loose bodies were seen as high signal on the fluid-sensitive sequence (STIR, PDFS) and demonstrated low signal on T1. The number of loose bodies and location were variable.

Management strategies included supervised observation (4), arthroscopic (6), or open excision (1) of the PrSC of the hip and arthroplasty (4) (Fig. 3). The follow-up ranged from 1 year to 6 years. A summary of clinical features, MRI findings, and clinical management strategies has been shown in Table 1. No malignant transformation was found in the analysed cohort.

4. Discussion

Synovial chondromatosis (SC) is a rare, benign condition affecting any synovial joint. PrSC is an even rarer condition with a reported incidence of 1 in 100,000 and mostly occurring in the 3rd to 5th decade of life. Mussey RD *et al.*, in a case series of 105 cases of SC, found only 5 cases involving the hip.²² Since the SC of the hip is extremely rare, and there are no large series of cases of PrSC of the hip published before in literature documenting its management strategies. We believe that the lower incidence of PrSC in the hip could be due to the relative paucity of synovial tissue in this joint and infrequent injuries, compared to the knee joint.

SC of the hip being an unusual site, its deeper location along with its varied presenting clinical features of pain, stiffness, locking, and limp may lead it to manifest insidiously.

Radiological Imaging plays a crucial role in the diagnosis of SC and more crucially is extremely useful in the diagnosis of Primary SC. Plain radiographs, CT, and MRI sequences also play a significant role in planning management and 'supervised observation' during follow-up of these patients. Plain radiographs usually tend to be the preliminary investigation in a clinical setting. Plain radiographs show well-developed multiple small, juxta-articular radio-opaque nodules depending on the stage of the disease, and several intra-



Fig. 1. AP radiograph (a) and PDFS coronal (b) and coronal CT(c) of pelvis demonstrating multiple loose bodies in both hip joints. There is secondary osteoarthritis in both hip joints.

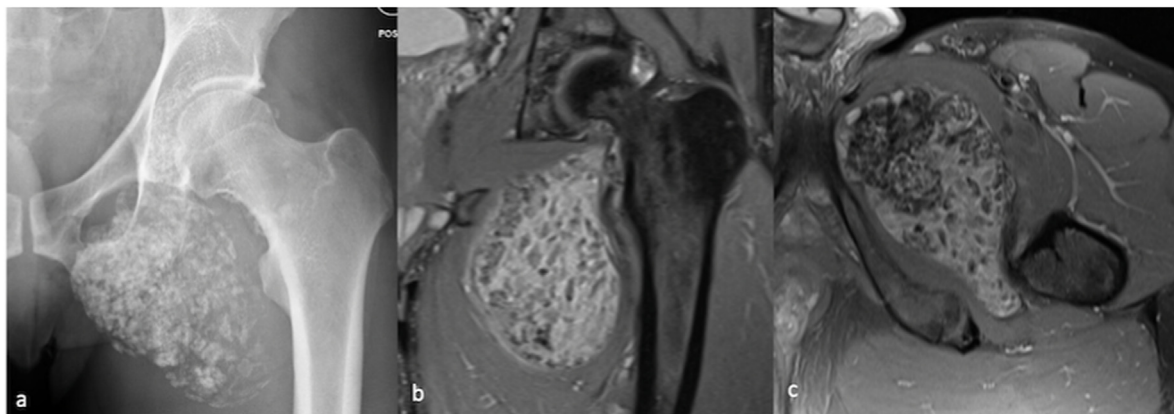


Fig. 2. AP radiograph (a), PDFS coronal (b) and axial (c) showing large left hip joint effusion with multiple loose bodies.

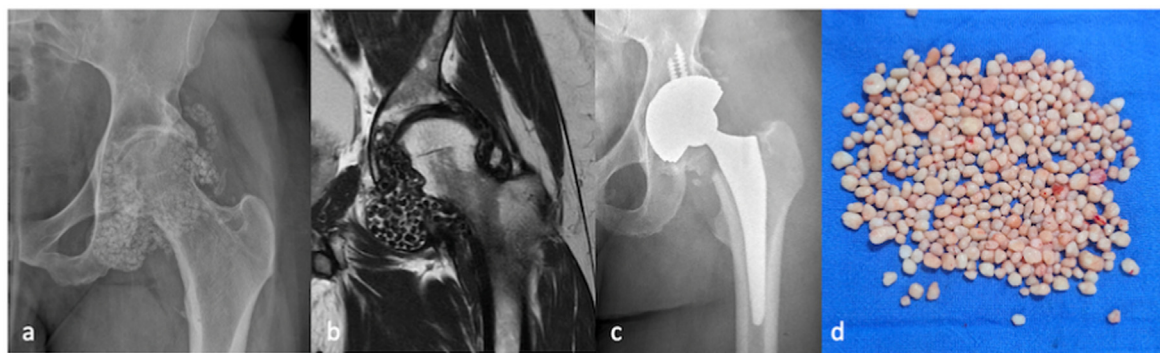


Fig. 3. AP radiograph (a), T2 coronal (b) of left hip showing multiple loose bodies in the left hip. AP radiograph (c) demonstrating Total hip arthroplasty and gross specimen (d) showing multiple loose bodies.

articular loose bodies tend to be more numerous in primary SC than secondary SC.^{6,11} MRI in the assessment of SC allows significant advantages and allows early diagnosis of PrSC of the hip due to

earlier detectable changes and differentiation from secondary causes of SC if plain radiographs are equivocal. Elucidation of an underlying secondary cause of SC can thus direct patient

Table 1
Demographic details, laterality, radiological image and clinical management interventions of patients with Primary Synovial chondromatosis (SC) of the hip joint: A Retrospective Cohort analysis.

Patient/case number	Age (Years)	Sex	Side = R/L/BL	Plain Radiography Findings	CT Scan	MRI Scan	Management	Follow-up in Years
1	14	M	R	Y			Excised-Arthroscopic	7
2	21	M	L	Y	Y	Y	Excised-Arthroscopic	3
3	27	F	R	Y	Y	Y	Excised-Arthroscopic	3
4	31	M	BL	Y	Y		Excised Open Procedure	3
5	32	M	BL	Y	Y		Observation	1
6	34	M	R	Y	Y	Y	Observation	4
7	34	F	R	Y		Y	Excised-Arthroscopic	8
8	37	M	L	Y		Y	THR	8
9	38	M	L	Y		Y	Observation	
10	42	F	L	Y		Y	Excised-Arthroscopic	7
11	44	F	R	Y		Y	THR	6
12	46	F	R	Y		Y	Excised-Arthroscopic	4
13	48	M	L	Y		Y	Observation	11
14	50	M	L	Y			BHR	15
15	50	M	L	Y		Y	THR	1

Abbreviations: R = Right; L = Left; B= Bilateral; CT= Computed Tomography; MRI = Magnetic Resonance Imaging; THR = Total Hip Replacement; BHR= Birmingham Resurfacing Hip; Excised-Arthroscopic = Arthroscopic Excision of Synovial chondromatosis (SC) of the hip joint.

management, whilst a diagnosis of PrSC of the hip is possible if none is found. MRI is also helpful in detecting soft-tissue extension of the disease and patient sequential follow-up.^{6,12-14}

Once the diagnosis of SC is made, 'supervised observation' and regular follow-up can be undertaken if the patient is asymptomatic. Nonoperative management consist of activity modification, and non-steroidal anti-inflammatory medication. However, management of Primary SC tends to be surgical. Considering the uncommon nature of PrSC, management protocols for this condition are only found in some published case reports or a few patients. Several procedures like arthrotomy, arthroscopy, and arthroplasty have been described to manage it.

Arthrotomy of the hip joint has been undertaken for the removal of symptomatic, loose bodies and arthrotomy with and without hip dislocation has been described. Young patients were treated with a mini arthrotomy without hip dislocation. Early surgical intervention with the removal of loose bodies in conjunction with joint distraction and early physiotherapy resulted in better outcomes even in patients with early arthropathy, as reported by Mahajan NP et al.²³ The surgical hip dislocation was suggested by Fang S et al. as an effective approach for managing intra-articular, as well as extra-articular hip lesions with lesser surgical time, fewer complications, good function, and a lower recurrence rate.²⁴ Open synovectomy with the removal of loose bodies may effectively relieve symptoms as reported by Lim SJ et al. at a mean of 4.4 years follow-up. They also deduced that SC may recur in cases with extensive involvement, especially in those who were treated without hip dislocation and had only synovectomy. The complications related to surgery occurred more in cases that were managed with synovectomy done with anterior hip dislocation.²⁵ In a review of all published case reports of hip SC, Gilbert SR et al. found that an open hip synovectomy not only relieves pain but may also delay progression or prevent OA.²⁶

Nakamura Y et al. successfully managed a 44-year-old man with right hip SC, by performing a surgical dislocation, synovectomy, and removal of loose bodies. At three months follow-ups, the patient was reported to have returned to the job and was 'fully' recovered.²⁷

Arthroscopic surgical techniques of the hip have opened an innovative way of dealing with SC of the hip. Marchie A et al. have treated a case with central compartment loose bodies (less than 10 mm), of the hip. They suggested that this procedure is effective for cases with early and mild disease.²⁸ Whereas, more extensive arthroscopic procedures have also been advised by the other

authors. Lee YK et al. found arthroscopic extensive capsulectomy as an effective treatment of hip SC.²⁹ Liu Y et al. reported that the arthroscopic T-shaped capsulotomy provides clinical good results, and lowers recurrence and complication rates.³⁰ Lee JB et al. suggested that treatment of hip SC hip using arthroscopic surgery (synovectomy and loose bodies removal) is effective and allows quicker rehabilitation of the patients'. However, the limitations of arthroscopy include the difficulty in approaching various parts of the hip joint.³¹ Zhang X et al. evaluated patients who underwent arthroscopic management for SC of the hip and concluded that arthroscopic treatment of hip SC provided good clinical and radiological outcomes, with low recurrence and higher patient satisfaction.³² Recurrence after arthroscopic surgical removal can be a concern. In a systemic review, De Sa D et al. suggested that for the hip SC, arthroscopic synovectomy and removal of loose osteochondral fragments/loose bodies are effective and safe. However, there is a mean recurrence rate of 7.1%.³³ Open techniques may be necessary in cases of recurrence. Schoeniger et al. reported no recurrence (at 6.5 years) in eight cases with monoarticular involvement. These cases had extensive synovectomy and joint debridement through a surgical hip dislocation, with a flip trochanteric osteotomy. They found this method of treatment effective, that prevents disease recurrence.³⁴

Management of recurrence remains a challenge. Radiotherapy, Radio synovectomy, and cryotherapy have been described in the management of SC but only a few case reports have been published. A further role of such therapies as individual modes of treatments or adjuvants to surgical procedures needs validation with future, a large cohorts of patients.³⁵⁻³⁷

Total hip arthroplasty (THA) or joint reconstruction remains a cornerstone in the management of patients with extensive disease, recurrence after open or arthroscopic synovectomy, and in patients with underlying arthritis. Prabowo Y et al. reported two cases (55- and 22-year-old men) who presented with SC of the hip. The former presented at a late stage with OA of the hip and underwent a total hip arthroplasty (THA), along with a thorough debridement to evacuate loose bodies and inflamed synovium. The latter had an open arthrotomy, with joint debridement. At 1-year follow-up, both cases demonstrated an improved functional outcome. The Harris Hip Score (HHS) improved from 39 to 91 in the first patient, while in the latter case it improved from 68 to 93. The authors concluded that joint debridement alone or along with THA can be effective options in the treatment of SC of the hip.³⁸

5. Limitations of study

We acknowledge the limitations of this study, being retrospective, and observational. But, it presents our experience in the management of Primary Synovial Chondromatosis (SC) of the hip joint especially the Radiological evaluation, MRI, and follow-up findings at a Tertiary Referral centre.

6. Conclusion

Primary Synovial Chondromatosis of the hip joint is a rare disorder and there is a paucity of knowledge about this condition and its management protocols. This article emphasizes the variety of surgical management strategies undertaken for treating this condition. The review suggests the ideal treatment is still yet to be finalised. Symptomatic patients benefit from an operative intervention like an open arthrotomy or arthroscopic synovectomy with the removal of loose bodies. In patients presenting late with secondary hip arthritis, total hip arthroplasty provides symptomatic and functional benefits. 'Observation' is possible in some patients with appropriate follow-up, depending upon symptoms and Informed shared decision-making process.

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Competing interests

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Disclosure statement and conflict of interest statement

Nothing to disclose. "The authors declare no conflict of interest".

Statement of ethics

Local hospital committee approval was obtained.

Statement of patient consent

All patients underwent investigation and operative management after an Informed Decision-making Process.

Author's contributions

RB, KP and RV involved in Conceptualization, literature search, manuscript writing and editing. AM, KPI, RB, AV, VK and RV Literature search, manuscript writing, references and editing. RV and RB supervised overall submission and approved final draft. All authors read and agreed the final draft submitted.

Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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