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Case Report

Coxa saltans caused by extraarticular synovial chondromatosis overlying an isolated osteochondroma of the greater trochanter: A rare aetiology

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ABSTRACT

Snapping hip is produced by intraarticular and extraarticular pathology. We describe a rare case of extraarticular snapping hip caused by bursal synovial chondromatosis overlying an osteochondroma. A 32-year-old male presented with swelling in his right gluteal region for 6 years associated with pain and snapping on movements since three months. On examination, he had an ill-defined 10 × 6 cm swelling over the posterolateral aspect of the greater trochanter. Plain radiographs revealed an osteochondroma arising from the greater trochanter. Further imaging showed exostosis with possible haemangioma, lipoma or liquefied haematoma overlying it. We proceeded with excision biopsy and intraoperatively found a bursa with synovial chondromatosis overlying the exostosis. At the end of ten months follow up, patient was asymptomatic and had no snapping. We present this case for its rarity, clinical and radiological diagnostic challenges and for its unique presentation.

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1. Introduction

Snapping hip or coxa saltans is described as the presence of an audible snap with or without associated pain on movements of the hip. It occurs due to either intraarticular or extraarticular pathology. Synovial chondromatosis is a

pathological condition resulting from metaplasia of the synovial membrane producing multiple cartilaginous material. Extraarticular synovial chondromatosis are rare and usually seen in bursa and tenosynovial tissue. We present a rare case of extraarticular snapping hip caused by synovial chondromatosis within a bursa overlying an osteochondroma of the greater trochanter.

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2. Case report

A 32-year-old male presented to our outpatient department with swelling in the right gluteal region since six years associated with pain and snapping since three months. The swelling was insidious in onset and gradually progressive. There was no history of prior trauma, fever. On examination, he had a 10 × 6 cm ill-defined swelling on the posterolateral aspect of the greater trochanter. He had tenderness but no warmth. There was no redness, scars or sinuses. He had snapping on hip movements associated with pain. There was no other swelling in other parts of the body. Blood investigations revealed haemoglobin of 15.2 g/dl and total leucocyte count of 8160/mm³. Erythrocyte sedimentation rate was 15 mm in 1 h. His alkaline phosphatase was 56 IU/L. Our initial diagnosis was trochanteric bursitis. Plain radiograph of the hip revealed an osteochondroma arising from the posterolateral aspect of the greater trochanter with soft tissue shadow along with a loose bony fragment [Figs. 1 and 2]. Ultrasonogram showed collection with low level echoes with floating bone fragments within. Computer tomograph showed exostosis arising from the posterolateral aspect of the greater trochanter, directed away from the joint with cortical and medullary continuity of adjacent bone [Fig. 3]. There was evidence of intramuscular plane ossification immediate posterior to the bony lesion probably haemangioma or lipoma. We decided to investigate further to rule out malignancy secondary to osteochondroma because of recent onset of pain and increase in size of the swelling. Hence magnetic resonance imaging was done which revealed exostosis with

Fig. 1 – Plain radiograph of the right hip anteroposterior view showing a bony prominence extending posterolaterally from the greater trochanter.

Fig. 2 – Plain radiograph of the hip lateral view showing exostosis arising from the posterior aspect of the greater trochanter with soft tissue shadow containing loose body.

evidence of 7 × 3 cm ill-defined focus in the gluteus maximus plane with hypointensity in T1 and hyperintensity in T2 images [Fig. 4]. There were also internal areas of bone signal intensities suggestive of bony fragments. Radiologists reported it could be probably a liquefied haematoma secondary to irritation. There was no evidence of malignancy. Bone scintigraphy showed localised pathology in the proximal femur. We had a dilemma in diagnosing the underlying cause. In view of the benign nature of the lesion and the possibility of

Fig. 3 – Computer tomograph showing bony outgrowth from the posterolateral aspect of greater trochanter.

Fig. 4 – Magnetic resonance imaging axial view T1 fat suppressed image showing central hypointense area close to greater trochanter with thick peripheral rim enhancement.

malignancy was minimal, we decided to proceed for excision biopsy.

Patient was positioned in lateral decubitus position. The lesion was approached through posterior incision. We identified a bursa overlying the exostosis. The bony overgrowth was excised extraperiosteally along with the bursa. The bursa was excised which showed multiple small loose bodies inside along with two large loose bodies suggestive of possible synovial chondromatosis [Fig. 5]. Wound was closed with suction drain. The entire specimen was sent for histopathology. Postoperative period was uneventful.

Macroscopically, there was a $3 \times 1.5 \times 1$ cm nodular, irregular bony fragment with cartilage cap measuring 0.2 cm on cut section. There were also multiple small grey white to grey brown nodular bits with two large grey white nodular mass. Microscopically, there were bony trabeculae with cartilage cap suggestive of osteochondroma [Fig. 6]. The multiple bits showed synovial hyperplasia along with

Fig. 5 – Intraoperative photograph of the specimen showing the excised exostosis along with the bursal sac and multiple loose bodies.

Fig. 6 – Photomicrograph showing bony tissue with a cartilage cap suggestive of osteochondroma.

cartilaginous metaplasia suggestive of synovial chondromatosis [Fig. 7].

At the end of ten months follow up, patient did not have snapping or pain. There was no recurrence of the lesion [Fig. 8].

3. Discussion

Coxa saltans or snapping hip is characterised by the presence of an audible snap during hip motion and is occasionally associated with hip pain. It occurs in 5–10% of the general population.¹ Activities which require extreme hip movements usually precipitate painful snapping hip like in athletes,

Fig. 7 – Photomicrograph showing synovial hyperplasia along with cartilage metaplasia (10× magnification).

Fig. 8 – Postoperative plain radiograph of the hip after excision of the osteochondroma.

soccer players, ballet dancers and weight lifters.¹ Diagnosing snapping hip is very challenging because of variety of causes which could be intraarticular or extraarticular. The intra-articular causes are synovial chondromatosis, labral tears, loose bodies, osteochondral fractures and hip instability. Extraarticular causes are further subdivided into external and internal cause. In the external variety, the snapping is due to friction of iliotibial band and gluteus maximus over the greater trochanter. Internal cause is attributed to iliopsoas movement over the iliopectineal eminence.² Snapping hip can also occur secondary to physical trauma, previous surgery to hip or knee and coxa vara.⁴ Extraarticular cause is the most common cause of snapping hip. Venous haemangioma of the gluteus maximus³ and dysmorphic gluteus maximus² have also been described to cause snapping hip.

Osteochondroma of the proximal femur as a cause for hip pain is relatively rare and very few cases are reported till now. Hussain et al⁴ have reported femoral acetabular impingement secondary to osteochondroma. Schaberg et al⁵ have reported two cases of internal snapping hip due to proximal femur exostosis. Sanshiro et al⁶ have reported a case of external snapping hip caused by exostosis of proximal femur.

Extraarticular synovial chondromatosis is rare and it usually arises from the bursa, capsular tissue or tenosynovial sheath.⁷ They are seen in hands and feet. Involvement of other sites is rare. Synovial chondromatosis along with osteochondroma is very rare. Wright et al⁸ have reported a case of bursal osteochondromatosis in the rib. Peh et al⁹ reported a case of secondary synovial chondromatosis within a bursa surrounding osteochondroma of the pubic bone. Errani et al¹⁰ have described a case of synovial chondromatosis within a bursa over osteochondroma arising from lesser trochanter mimicking chondrosarcoma. Brandon et al¹¹ have reported a case of pes anserinus bursal synovial chondromatosis secondary to osteochondroma. To the best of our knowledge, exostosis associated with bursal chondromatosis causing coxa saltans is not described in the English literature.

Our patient presented with pain in the hip on movements associated with an audible snap. Clinically, we thought it

could be trochanteric bursitis. Plain radiograph revealed an exostosis arising from the greater trochanter. Further imaging showed collection in the intramuscular region with few bony fragments. There was a dilemma in diagnosis as the radiologists suggested it could be either lipoma or haematoma. Magnetic resonance imaging was done which revealed a possible intramuscular haematoma probably secondary to irritation with loose bony fragments which again added to confusion. Though bursitis is a possibility when there is an associated osteochondroma, the cause for the presence of bony fragments was not clear. Since the classical picture of synovial chondromatosis is not seen radiologically and the extraarticular origin itself being very rare, we had difficulty in arriving at a diagnosis. Chondrosarcoma secondary to osteochondroma should also be in our mind when there is pain and increase in size of an existing exostosis. Since clinically and radiologically it looked benign, we proceeded with excision biopsy which turned out to be secondary synovial chondromatosis in a bursa overlying an exostosis.

We describe this case to highlight a rare aetiology for hip pain with associated snap and for the diagnostic challenges we faced clinically and radiologically. It is necessary to investigate these patients thoroughly as the differentials vary from trochanteric bursitis to chondrosarcoma. Diagnosing this condition is a real challenge to the treating orthopaedic surgeon and the radiologist. Clinicoradiological and pathological correlation is necessary to differentiate chondrosarcoma from synovial chondromatosis.

To conclude, extraarticular synovial chondromatosis overlying an exostosis in proximal femur is a rare cause for snapping hip. Clinicians should keep in mind about the possible aetiology for patients presenting with hip pain. Early appropriate diagnosis plays a vital role in planning treatment.

Conflicts of interest

All authors have none to declare.

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