

## CASE REPORT

# Primary Synovial Osteochondromatosis of the Shoulder in Pediatric Patient: Case Report and Review of the Literature

Ali Etemad-Rezaie, MD, MS<sup>1</sup>; Magdalena Tarchala, MD, MSc, FRCSC<sup>1</sup>; Andrew Howard, MD, MSc, FRCSC<sup>1</sup>

*Research performed at the Hospital for Sick Children, Division of Orthopaedic Surgery, University of Toronto, Toronto, Canada*

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### Abstract

We report a primary synovial osteochondromatosis in a 6-year-old male with a 1-year history of shoulder pain and stiffness. The patient underwent surgical treatment, resulting in significant improvement in range of motion and functional status. Primary synovial osteochondromatosis of the shoulder is a rare benign condition that can result in shoulder pain, stiffness, and locking. MRI helps identify these lesions within the affected joint. Removal of loose bodies often alleviates symptoms and allows for improved range of motion with satisfactory results.

**Level of evidence:** IV

**Keywords:** Pediatric, Shoulder, Synovial osteochondromatosis

### Introduction

Synovial osteochondromatosis is a rare cause of joint pain and stiffness. Synovial osteochondromatosis is a benign neoplastic condition in which multiple osteocartilaginous nodules develop within a joint, bursa, or tendon sheath. This is thought to arise from metaplasia and synovial membrane proliferation.<sup>1</sup> Clinical presentation includes pain, stiffness, and functional limitations in the affected joint.

Synovial osteochondromatosis can affect any synovial cavity. This uncommon condition typically affects men in the third and fourth decade of life and is usually present in the knee and hip joints.<sup>2-4</sup> There have been around 100 cases of synovial osteochondromatosis reported in the literature, with few reported to affect the shoulder. To date, there have been even fewer reported in the paediatric population.<sup>5-7</sup>

In this case report, we highlight the diagnosis and management of a 6-year-old male who presented with a 1-yr history of shoulder stiffness and inability to do

basic activities of daily living secondary to synovial osteochondromatosis of the right shoulder. We also review the current literature on the presence of synovial Osteochondromatosis in the pediatric shoulder.

### Case presentation

A 6-year-old male was referred to our Orthopaedic clinic with a one-year history of right shoulder stiffness. The patient's mother reported that the patient had had some pain initially, but the pain disappeared shortly after the first month. The patient had an uncomplicated birth history via C-section with no history of complications surrounding the birth, such as brachial plexus palsy. No previous history of trauma, infection, or an autoimmune disease was noted. The mother stated that her son had difficulty dressing or reaching for objects above his head. He was subsequently evaluated by the local pediatrician, who recommended observation after plain films did not reveal any abnormalities. When the

**Corresponding Author:** Ali Etemad-Rezaie, Hospital for Sick Children, Division of Orthopaedic Surgery, University of Toronto, Toronto, Canada  
Email: ali.etemad@mail.utoronto.ca



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stiffness persisted after months of observation, an MRI was ordered, demonstrating numerous nodules within the glenohumeral joint.

Physical examination revealed increased fullness of the right shoulder relative to the contralateral side. On palpation, a few nodules were palpated on the medial aspect of the long head of the biceps origin. The patient's range of motion was limited, with 0-10 degrees of internal and external rotation and up to 10 degrees of forward elevation and abduction when stabilizing scapulothoracic motion. The most apparent shoulder movement was through scapulothoracic motion.

AP X-ray of the right shoulder revealed subchondral irregularity predominantly along the medial contour of the humeral head without apparent nodules or calcifications [Figure 1]. There was noted fullness of the soft tissues in the axillary pouch compared to the contralateral shoulder. MRI of the right shoulder showed numerous well-rounded calcified loose bodies of varying size with peripheral enhancement around the lesions as well as along the synovium surface [Figure 2-4]. Overall, the imaging was suggestive of synovial osteochondromatosis with reactive synovitis.

Conservative treatment was initiated with physiotherapy for three months to assess if the patient's range of motion could be improved through non-operative intervention. However, the patient's stiffness persisted. We then offered the patient an open anterior shoulder arthrotomy and loose body removal to help improve his range of motion.



Figure 1. AP X-ray of the Right shoulder showing subtle cortical irregularity over the medial humeral head.

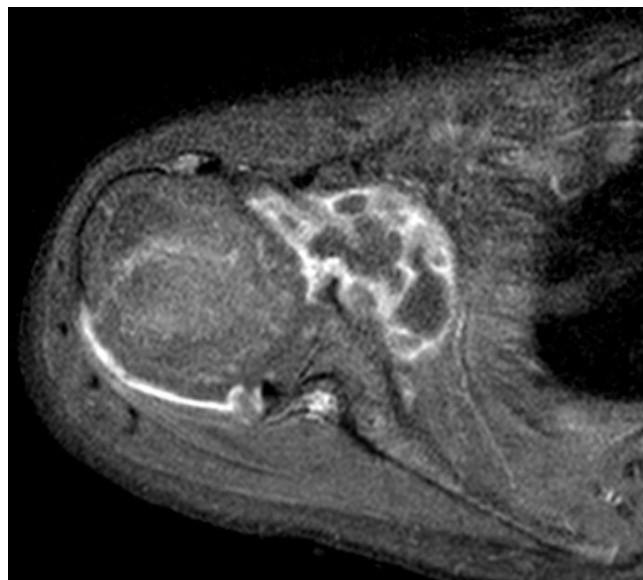


Figure 2. Axial T2 MRI view of the right shoulder. Cartilaginous loose bodies were noted in the axillary pouch and extending anteromedial to the glenohumeral joint.

An exam under anesthesia revealed decreased range of motion of the right shoulder, similar to the exam obtained in the clinic. The patient had an arc of abduction of about 30° or less and an arc of forward flexion of about 30° or less, and essentially no external

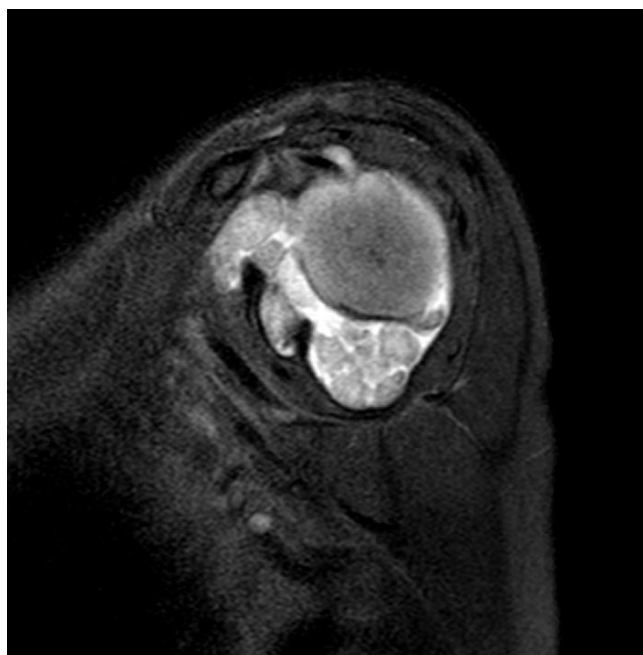


Figure 3. Sagittal T2 MRI view of the right shoulder showing abundant loose bodies within the glenohumeral joint and the axillary recess.



**Figure 4.** AP T2 MRI of the right shoulder shows abundant loose bodies in the glenohumeral joint.

rotation or internal rotation when we stabilized the scapula and tried to move through the glenohumeral joint alone. His resting position was with the arm abducted to the side body, and any elevation beyond 30° was scapulothoracic, and we were unable to bring



**Figure 5.** Intraoperative images of the deltopectoral open approach were utilized to extract the loose bodies.

his hand above shoulder level. A standard deltopectoral approach was used to expose the glenohumeral joint. The deltopectoral fascia was incised. A longitudinal subscapularis tenotomy was performed. Upon exposure, the anterior capsule was significantly thinned. An arthrotomy was performed, and the osteochondral loose bodies were removed [Figure 5]. The loose bodies were localized throughout the glenohumeral joint, in the axillary recess, within the bicep tendon sheath, medial to the coracoid as well as adhered to the humeral head on the superior margin of the articular surface, as well as adhered both to the anterior and posterior surface of the glenoid. We removed over 50 loose bodies measuring approximately 20 cc equivalent [Figure 6]. Notably, it was difficult to assess for complete removal of all loose bodies intra-operatively with direct visualization; therefore, extreme diligence is critical to assess complete removal. After removing all loose bodies, there was an improvement in the right shoulder range of motion, resulting in 50 degrees of external rotation, 20 degrees of internal rotation, 70° of forward flexion, and almost 90° abduction.

The subsequent pathology report confirmed the diagnosis of synovial osteochondromatosis with no inflammatory or malignant features.

Post-operatively, the patient was placed in a sling for comfort for the first two weeks, followed by progressive physiotherapy. At the one-year mark, the patient was pain-free with functional recovery. His shoulder ROM was 135 degrees of forward flexion, abduction of 105 degrees, external rotation of 75 degrees, and internal rotation of 55 degrees [Figure 7].



**Figure 6.** Over 50 loose bodies were extracted during the open anterior shoulder arthrotomy.



Figure 7. Clinical radiographs demonstrate recovery of excellent functional motion. 135 degrees forward flexion-Anterior view (B) Posterior (C) Abduction - 105 degrees (D), internal rotation-55 degrees (E) and external rotation-75 degrees (F).

### Discussion

Primary synovial osteochondromatosis is a benign but rare condition that can affect any joint. It is typically seen in the adult population in their 3rd and 4th decade of life with the hip and knee joints that are usually involved. To date, very few reports have highlighted the presence of synovial osteochondromatosis of the shoulder in a pediatric patient.<sup>5-7</sup> Below [Table 1] is a summary of the three previous case reports.

The etiology of primary synovial osteochondromatosis is unknown. Histological studies have shown that the synovial cells undergo metaplasia to chondrocytes resulting in the deposition of the multiple loose bodies seen within the joint. In secondary synovial osteochondromatosis, there is often a secondary nidus, such as an osteochondral fragment from trauma or loose bodies/osteophytes from osteoarthritis. These lesions will then undergo cartilaginous metaplasia resulting in the characteristic loose bodies. In this case, our patient did not have any trauma suggesting the presence of a primary disease. His delivery history (c-section) suggests that birth trauma to the shoulder is unlikely, but cannot be excluded entirely.

The symptoms associated with primary synovial chondromatosis are often non-specific. Pain, restricted range of motion, catching or locking symptoms, and the presence of a palpable mass are common nonspecific symptoms. The radiographs may show calcifications in the glenohumeral joint as well as the subacromial space. In this case, the initial x-ray findings were subtle, with no osseous 'rice bodies' seen, and the lesions were only detected on MRI. MRI is usually the confirmatory test showing the cartilaginous loose bodies. A biopsy is often not indicated as lesions are pathognomonic of the condition and benign in nature.

Treatment is aimed at alleviating symptoms and increasing the range of motion. A trial of conservative management with physiotherapy can be attempted. In our case, the patient failed this trial, and we opted to perform an open debridement to extract the loose bodies. Debate remains on whether open or arthroscopic technique should be performed. Open debridement offers the added benefit of greater exposure with access to the bicipital groove.<sup>8</sup> Arthroscopic debridement

Table 1. Summary of studies to date outlining synovial Osteochondromatosis in pediatric shoulder

Author (year)	Age of patient	Clinical findings	Intervention	Results
Nashi (1998)	14 yr old boy	-pain with vigorous sporting activity -Asymptomatic with mild pain only at extremes of ROM -loose bodies seen on XR	Observation w/ brief physiotherapy program	2 year radiograph showed increase in size of lesion but patient remained symptom free
Miranda (2004)	10 year old girl	-mild to moderate pain with exercises -clicking in the shoulder	Open deltopectoral approach, biceps tendon sheath	-Symptom-free -Full return to activities -No recurrence at 1-year
Sinikumpu (2020)	14 yr old boy	-pain and stiffness in shoulder for one year -lesions seen both on XR and confirmed on MRI	Open, deltopectoral approach -inferior recess, sub-coracoid bursa, biceps tendon sheath	-Symptom-free -full return to activities -no recurrence @ one-year

provides the added benefit of reduced morbidity and earlier recovery.<sup>9,10</sup> However, exposure and retrieval of larger loose bodies can be difficult through the arthroscopic portals. In our case, we utilized an open approach for three reasons 1) ease of loose body removal, 2) greater exposure to the bicipital groove 3) preference and familiarity. Currently, no consensus exists on which method is superior.

The recurrence rate for this condition is between 15-20%.<sup>11</sup> It is important to follow these patients and obtain a repeat MRI if symptoms recur. Malignant transformation may occur in 5% of patients. It can present as a rapid recurrence, symptom progression, and aggressive features such as muscle infiltration.<sup>11</sup> In our case, the biopsy did not show any malignant characteristic, and the patient has made a good functional recovery at the one-year mark [Figure 7] with no post-operative complications. The patient will have yearly follow-ups to observe for any possible

recurrence.

Primary synovial osteochondromatosis is a very rare benign condition which can result in shoulder pain, stiffness, and locking sensation. Advance imaging studies in the form of MRI are helpful in identifying these lesions within the affected joint. Operative intervention in the form of arthroscopic or open debridement often alleviates symptoms with satisfactory results. Patients should be followed post-operatively for at least two years to screen for recurrence or possible malignant transformation.

Ali Etemad-Rezaie MD MS<sup>1</sup>

Magdalena Tarchala MD MSc FRCSC<sup>1</sup>

Andrew Howard MD MSc FRCSC<sup>1</sup>

<sup>1</sup> Hospital for Sick Children, Division of Orthopaedic Surgery, University of Toronto, Toronto, Canada

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