

# Synovial Osteochondromatosis: Clinical Characteristics Unique to the Shoulder

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**Background:** Synovial osteochondromatosis (SOC) of the shoulder is a rare condition with unclear characteristics. This study evaluated the clinical features and postoperative functional outcomes of SOC of the shoulder that are distinct from SOC of other joints.

**Methods:** The characteristics of 28 shoulders with SOC that underwent arthroscopy were retrospectively assessed. Ten shoulders (35.7%) had rotator cuff tears (RCTs) and underwent concomitant arthroscopic rotator cuff repair. The mean follow-up period was 83.6 months (range, 24–154 months). Demographic characteristics and loose bodies localized under arthroscopy were compared between cases with and without concomitant RCTs. Radiography, ultrasonography, or magnetic resonance imaging were performed preoperatively and postoperatively. Visual analog scale (VAS) scores for pain and satisfaction were evaluated for all cases, and functional scores were assessed in shoulders with concomitant RCTs.

**Results:** The average age was  $36.2 \pm 15.6$  years among patients without RCTs and  $58.3 \pm 7.2$  years among patients with RCTs. Seven shoulders (7%) had osteoarthritis. Arthroscopy revealed loose bodies in multiple spaces, including the glenohumeral joint, subacromial (SA) space, and biceps tendon sheath. Overall, loose bodies were found in multiple spaces in 12 shoulders (42.9%). Loose bodies were found in the SA space only in 4 shoulders (22.2%) without RCTs and in 7 shoulders (70.0%) with RCTs. VAS for pain decreased significantly from  $3.9 \pm 2.3$  to  $1.1 \pm 1.3$  (p < 0.001). The functional scores increased significantly after arthroscopic management for patients with concurrent RCTs (all p < 0.05). Recurrence of SOC occurred in 3 of the 22 shoulders (13.6%) who underwent postoperative imaging, but no patient had a recurrent RCT.

**Conclusions:** Pain relief and patient satisfaction were achieved via arthroscopic management. Unlike in other joints, loose bodies can occur simultaneously in several spaces in the shoulder, including the glenohumeral joint, SA space, and biceps tendon sheath. Early diagnosis of SOC of the SA space can help prevent osteoarthritis and RCT progression.

Keywords: Osteochondromatosis, Synovium, Shoulder joint, Arthroscopy

Received March 2, 2022; Revised April 15, 2022; Accepted May 8, 2022 Correspondence to: Joo Han Oh, MD Department of Orthopedic Surgery, Seoul National University Bundang Hospital, Seoul National University College of Medicine, 82 Gumi-ro 173beon-gil, Bundang-gu, Seongnam 13620, Korea Tel: +82-31-787-7197, Fax: +82-31-787-405 E-mail: ohjh1@snu.ac.kr Synovial osteochondromatosis (SOC) is a rare condition that may affect any cavity lined with synovium.<sup>1)</sup> The definitive etiology remains unknown, yet it can be classified as primary or secondary. Secondary causes of SOC include trauma, osteochondral dissecans, osteoarthritis (OA), and osteochondral fractures. SOC is characterized by osteocartilaginous loose bodies in diarthrodial joints and occurs most commonly in the knee, followed by the hip, elbow,

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wrist, and ankle, and least commonly in the shoulder.<sup>2-4)</sup> SOC is more common in men than women. The worldwide incidence has been reported to be  $1 : 100,000,^{5}$  and SOC most frequently affects patients in the third to fifth decade of life. Patients with SOC of the shoulder often present with pain, swelling, and a slowly-progressing limited range of motion (ROM).<sup>6)</sup> Due to the insidious onset of clinical features, the diagnosis of SOC of the shoulder is typically delayed for months to years. The classic treatment for SOC of the shoulder is open arthrotomy, synovectomy, and complete removal of loose bodies.<sup>4,7)</sup> Recently, due to improved arthroscopic techniques, enhanced visualization of the surgical field, and shorter rehabilitation periods, arthroscopic treatment for SOC of the shoulder has been reported to be a good treatment option.<sup>1)</sup> The rate of postoperative recurrence after open or arthroscopic therapy ranges from 3% to 23%.89 Malignant transformation may also occur postoperatively.

SOC in the shoulder joint can occur in the subacromial (SA) space,<sup>9)</sup> sheath of the long head of the biceps tendon (LHBT),<sup>10,11)</sup> and glenohumeral (GH) joint<sup>1,6,12)</sup> and may present with concomitant diseases such as rotator cuff tears (RCTs)<sup>13-15)</sup> or OA.<sup>7,10)</sup> Previous studies addressed SOC condition based primarily on case reports and small series, and only a few reports involved the shoulder.<sup>4,14)</sup> The current study is the largest case series presenting patients with SOC of the shoulder, among which 10 cases were accompanied by concomitant RCTs. Besides, no previous research has been conducted to evaluate the clinical features of SOC in the shoulder joint and the distribution of loose bodies. This study was conducted to evaluate the clinical features and postoperative functional outcomes of SOC of the shoulder that are distinct from SOC of other joints. We hypothesized that SOC of the shoulder would be more likely to present with loose bodies in multiple spaces than SOC of other joints and that arthroscopic treatment could be effective for symptom relief and functional improvement.

### **METHODS**

A multicenter survey was conducted to collect the data of patients diagnosed with SOC of the shoulder for this retrospective study. The inclusion criteria were as follows: patients who had undergone open arthrotomy or arthroscopic treatment with a synovectomy and removal of loose bodies from August 2007 to January 2019 and patients who were diagnosed with SOC of the shoulder with histopathological confirmation after surgical management. This multicenter retrospective study was reviewed and approved by Institutional Review Board of Seoul National University Bundang Hospital (No. B-2102/664-113). The requirement of written informed consent was waived as only preexisting data were used for this study and the confidentiality of the patients was maintained.

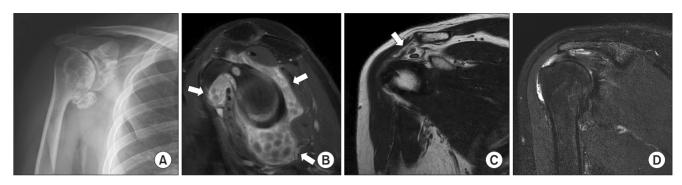
A total of 38 shoulders from five institutions met the inclusion criteria. Upon reviewing the medical records, a total of 10 shoulders were excluded from the study, including 3 shoulders that had undergone total shoulder arthroplasty on the same side and 7 that had less than 24 months of follow-up. The final analysis included 28 shoulders (27 patients) from 4 tertiary hospitals. One shoulder was treated with open synovectomy and loose body removal under arthroscopy, and the others received arthroscopic synovectomy with loose body removal. Of these 27 patients, 1 (3.7%) presented with bilateral SOC in the shoulders. Ten of the total 28 shoulders (35.7%) were found to have SOC associated with RCTs and underwent concomitant arthroscopic rotator cuff repair (RCR).

#### **Clinical Assessments**

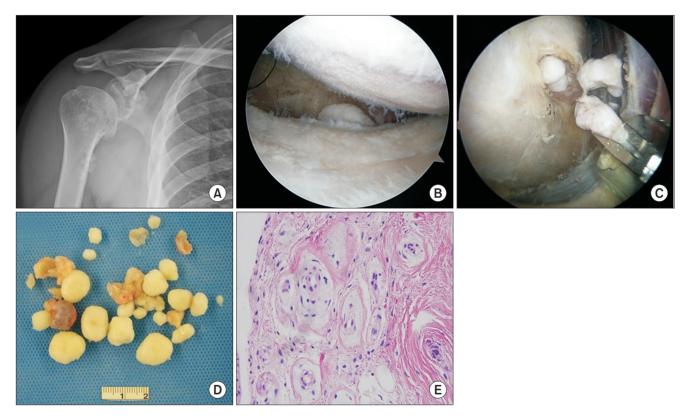
The patients' demographic data including age, sex, hand dominance, operated side, preexisting diseases (including osteonecrosis, OA, and rheumatoid arthritis), and the trauma history were collected from the medical records and evaluated. Ultrasonography of the shoulder was performed in some patients to help with the diagnosis. Plain radiography and magnetic resonance imaging (MRI) were performed in all shoulders preoperatively to determine the presence and locations of radio-opaque loose bodies. SOC was diagnosed through thorough history taking, physical examinations, imaging study, and histological confirmation with metaplastic cartilage nodules. Preoperative degree of OA was identified through radiography using the Samilson and Prieto classification, in which OA was divided into four grades (grade 0, normal; grade 1, mild OA; grade 2, moderate OA; grade 3, severe OA). Concurrent RCT was confirmed on preoperative MRI (Fig. 1). These imaging evaluations were also performed postoperatively to confirm the recurrence of SOC or failed RCR. For all SOC cases, the visual analog scale (VAS) for pain scores, shoulder ROM in forward flexion (FF), external rotator (ER) at the side, and internal rotator (IR) at the back were routinely collected pre- and postoperatively. The VAS was also used to subjectively evaluate patient satisfaction postoperatively. Functional outcome measures for the shoulder, including the simple shoulder test (SST), American Shoulder and Elbow Surgeons (ASES) form, and Constant scores, were assessed preoperatively and at the final followup visit for patients who underwent arthroscopic manage-

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**Fig. 1.** Preoperative radiographic findings of the right shoulder of a 30-year-old male patient. (A) Multiple radiopaque bodies in the glenohumeral (GH) seen on the anteroposterior plain radiograph. (B) Sagittal magnetic resonance image (MRI) revealed multiple loose bodies throughout GH joint, in the subacromial (SA) space, and the sheath of long head of the biceps tendon (arrows). (C) Preoperative T1-weighted, coronal view of MRI showed loose bodies in the SA space (arrow). (D) High-grade partial tear of supraspinatus tendon present on the coronal view (T2).



**Fig. 2.** (A) Preoperative plain radiograph of a 41-year-old male patient showing multiple osseous bodies in the glenohumeral (GH) joint and within the sheath of the long head of the biceps tendon (LHBT). (B) Arthroscopic image showing several loose bodies within the GH joint space. (C) Arthroscopy was performed in the subacromial space and multiple loose bodies were found in the sheath of the LHBT. The loose bodies were removed with an arthroscopic grasper through the lateral cannula. (D) The gross specimen showing several loose bodies that were removed. (E) Histological image of the loose bodies showing fragments of cartilaginous tissue formation was suggestive of synovial osteochondromatosis (H&E, × 40).

ment of SOC with concurrent RCT.

**Surgical Procedures and Postoperative Rehabilitation** All surgical procedures were performed by four experienced senior surgeons (SHK, JCY, YGR, and JHO) with the patient under general anesthesia in the lateral decubitus position. The procedures were performed through standard anterior and posterior portals under a 30° or 70° arthroscope. Accessory portals were also made according to each surgeon's needs. It is a unique characteristic of

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SOC of the shoulder that cartilaginous or osteocartilaginous nodules can be found within the GH joint, in the sheath of LHBT, and in the SA space. Firstly, a posterior portal was placed to assess the GH joint thoroughly. Then, a shoulder arthroscopy was properly introduced into the SA space to detect loose bodies and the involvement of the sheath of the LHBT (Fig. 2).

Loose body removal with synovectomy was conducted in all cases. Small loose bodies were extracted with an arthroscopic forceps or grasper, and bigger loose bodies were removed through an outflow cannula with lavage. Extreme care was taken to avoid injury when removing loose bodies in the sheath of LHBT. The biceps tendon was carefully lifted with a probe out of the groove to identify loose bodies in the biceps tendon sheath. Externally rotating the arm was helpful for better visualization to free possible loose bodies in the groove. Intact LHBTs were left in situ, and a biceps tenotomy or tenodesis was performed for LHBTs with lesions. Arthroscopic synovectomy was performed using a motorized shaver or an electrothermal device. Open synovectomy was performed to excise abnormal synovium membrane layer and soft tissues. After loose body removal with synovectomy, immediate passive ROM exercises were encouraged, while active ROM and strengthening exercises began at postoperative 1 week. When tenotomy or tenodesis was performed simultaneously, an abduction brace without a pillow was used for 2 weeks and 4 weeks, respectively. Active ROM exercises began after the removal of the brace.

In shoulders with an RCT, the tear size and retrac-

tion of the rotator cuff were measured intraoperatively after the debridement under direct arthroscopic visualization with a calibrated probe. Arthroscopic RCR was performed after the removal of the loose bodies and synovectomy (Fig. 3). For patients with medium-sized tears, a 60°-abduction brace was used for immobilization for 5 weeks after the surgery. Active-assisted ROM exercises were started after weaning off the brace. Muscle-strengthening exercises were introduced at 12 weeks postoperatively.

#### **Statistical Analysis**

All statistical analyses were performed using IBM SPSS ver. 23.0 (IBM Corp., Armonk, NY, USA). The patients' demographic data were summarized using means and standard deviations for continuous variables and numbers and percentages for categorical variables. Chi-square tests were used to determine the differences of categorical variables between the groups. Shoulder ROM and functional scores obtained preoperatively and at the final follow-up visit were compared using the Wilcoxon signed-rank tests. Statistical significance was set at p < 0.05.

#### RESULTS

#### **Demographic Characteristics**

All 28 shoulders included in this study underwent surgical management for SOC between August 2007 and January 2019 at one of four institutions. Ten shoulders (35.7%) were diagnosed with concomitant RCTs, and 1 patient



**Fig. 3.** (A) Arthroscopic image of a 56year-old female patient showing multiple loose bodies floating in the subacromial space. (B) The loose bodies were removed with a grasper. (C) The gross specimen shows six loose bodies that were removed. (D) A concomitant bursal-side high grade partial thickness supraspinatus tear is shown. (E) The debridement of the supraspinatus tendon is shown. (F) The rotator cuff repair was performed using the modified Mason-Allen suture bridge technique. (3.7%) had bilateral SOC. Ten patients (35.7%) were men, and 18 patients were women. The mean age of the patients without RCT was  $36.2 \pm 15.6$  years, and the mean age of the patients with RCT was  $58.3 \pm 7.2$  years (p < 0.001). All the patients presented with shoulder pain, and the mean time from symptom onset to surgery was 36 months (range, 1–240 months). Nineteen shoulders (67.9%) had SOC on the dominant side. Seven shoulders (25.0%) presented with preoperative OA found on radiographs or MRIs, including 6 shoulders with mild OA and 1 with moderate OA. Seven shoulders (25.0%) had preoperative stiffness with limited ROM. No patients reported previous shoulder injuries or had a history of trauma. The mean follow-up period was 83.6 months (range, 24–154 months) (Table 1).

#### **Intraoperative Findings**

In all shoulders with concomitant RCTs, only the supraspinatus tendon was involved. There was 1 small-sized tear and 9 medium-sized tears with a mean anteroposterior length of  $13.7 \pm 10.8$  mm and a mean retraction of  $13.2 \pm 7.8$  mm. Regarding the distribution and localization of loose bodies (Table 2), 5 of the total 28 shoulders (17.8%) had loose body findings in the GH only. Loose bodies were found in multiple spaces in 12 shoulders (42.9%), including 3 shoulders (16.7%), in which loose bodies were

Table 1. Demographic Data of the Patients					
Variable	SOC without RCT	SOC with RCT	<i>p</i> -value		
No. of patients	18	10	-		
Age (yr)	36.2 ± 15.6	58.3 ± 7.2	< 0.001*		
Sex (male : female)	9:9	1:9	0.048*		
Symptom duration (mo)	38.0 ± 57.9	31.2 ± 33.6	0.737		
Dominance (right : left : both)	18:0:0	9:1:0	0.357		
Operation on dominant side (yes : no)	11 : 7	8:2	0.417		
Osteoarthritis (yes : no)	4:14	3:7	0.674		
Stiffness (yes : no)	7:11	0:10	0.030*		
Intraoperative tear size (mm)					
Anteroposterior	-	13.7 ± 10.8	-		
Mediolateral	-	13.2 ± 7.8	-		

Values are presented as mean ± standard deviation. SOC: synovial osteochondromatosis, RCT: rotator cuff tear. found in the GH joint, SA space, and sheath of the LHBT. Among shoulders without RCTs, loose bodies were found in the SA space only in 4 shoulders (22.2%) and in the GH joint and SA space in 1 shoulder (5.6%). Five shoulders (27.8%) had loose bodies in the GH joint and the sheath of the LHBT. Among shoulders with RCTs, loose bodies were found in the SA space in 9 shoulders (90.0%), including 7 shoulders (70.0%) in the SA space only. One patient (10.0%) had loose bodies in the GH joint and the sheath of the LHBT.

#### **Clinical Outcomes**

All patients had favorable postoperative outcomes. In the total group (n = 28), the mean VAS pain score decreased significantly from  $3.9 \pm 2.3$  preoperatively to  $1.1 \pm 1.3$ postoperatively (p < 0.001). Shoulder ROM increased postoperatively, but the improvement was not significant (FF:  $153.7^{\circ} \pm 18.2^{\circ}$  to  $155.7^{\circ} \pm 15.5^{\circ}$ , p = 0.684; ER at side:  $49.6^{\circ} \pm 23.1^{\circ}$  to  $50.2^{\circ} \pm 24.0^{\circ}$ , p = 0.870; IR at back: T10.5  $\pm$  3.6 to T8.4  $\pm$  3.6; *p* = 0.108). In the SOC without RCT group (n = 18), although pain VAS decreased significantly from  $4.1 \pm 1.8$  to  $1.4 \pm 1.6$  (p = 0.003), shoulder ROM did not increase significantly. Patients in the SOC concomitant with RCT group (n = 10) had significantly lower pain VAS after the treatment  $(3.6 \pm 2.9 \text{ vs. } 0.7 \pm 0.7, p = 0.012)$ . While shoulder ROM did not show significant statistical improvement, all functional scores significantly improved. The mean SST score increased from  $5.6 \pm 3.7$  preoperatively to  $10.0 \pm 1.9$  postoperatively (p = 0.018), the mean ASES score increased from 59.7  $\pm$  19.6 preoperatively to  $93.4 \pm 4.9$  postoperatively (*p* = 0.012), and the mean Con-

Table 2. Frequency and Distribution of Loose Bodies					
Localization of loose bodies	SOC without RCT (n = 18)	SOC with RCT (n = 10)	Total (n = 28)		
GH	5 (27.8)	0	5 (17.8)		
SA	4 (22.2)	7 (70.0)	11 (39.3)		
LHBT	0	0	0		
GH & SA	1 (5.6)	0	1 (3.6)		
GH & LHBT	5 (27.8)	1 (10.0)	6 (21.4)		
SA & LHBT	0	2 (20.0)	2 (7.1)		
GH & SA & LHBT	3 (16.7)	0	3 (10.7)		
Total	18 (100)	10 (100)	28 (100)		

Values are presented as number (%).

SOC: synovial osteochondromatosis, RCT: rotator cuff tear, GH: glenohumeral joint, SA: subacromial space, LHBT: long head of the biceps tendon.

\*Statistically significant.

Table 3. Preoperative and Post	stoperative RON	l and Function	al Scores
Variable	Preoperative	Final visit	<i>p</i> -value
SOC without RCT (n = 18)			
Pain VAS	4.1 ± 1.8	1.4 ± 1.6	0.003*
Shoulder ROM			
FF (°)	150.0 ± 18.7	153.2 ± 16.9	0.506
ER (°)	42.9 ± 22.6	44.4 ± 26.0	0.832
IR at back	T10.2 ± 3.5	T9.7 ± 2.1	0.489
Satisfaction VAS	-	8.8 ± 1.4	-
SOC with concomitant RCT ( $n = 10$ )			
Pain VAS	3.6 ± 2.9	0.7 ± 0.7	0.012*
Shoulder ROM			
FF (°)	157.1 ± 16.2	160.0 ± 12.5	1.000
ER (°)	60.0 ± 17.0	61.0 ± 20.2	0.811
IR at back	T11.0 ± 3.9	T6.2 ± 4.6	0.073
SST score	5.6 ± 3.7	10.0 ± 1.9	0.001*
ASES score	59.7 ± 19.6	93.4 ± 4.9	0.004*
Constant score	53.3 ± 10.6	73.5 ± 5.7	0.012*
Satisfaction VAS	-	8.6 ± 1.0	-
Total (n = 28)			
Pain VAS	3.9 ± 2.3	1.1 ± 1.3	< 0.001*
Shoulder ROM			
FF (°)	153.7 ± 18.2	155.7 ± 15.5	0.569
ER (°)	49.6 ± 23.1	50.2 ± 24.0	0.903
IR at back	T10.5 ± 3.6	T8.4 ± 3.6	0.108
Satisfaction VAS	-	8.7 ± 1.3	-

Values are presented as mean ± standard deviation.

ROM: range of motion, SOC: synovial osteochondromatosis, RCT: rotator cuff tear, VAS: visual analog scale, FF: forward flexion, ER: external rotation at side, IR: internal rotation measured by the level of vertebral spinous process that could be reached with the patient's thumb, SST: simple shoulder test, ASES: American Shoulder and Elbow Surgeons. \*Statistically significant.

stant score improved from  $53.3 \pm 10.6$  preoperatively to  $73.5 \pm 5.7$  postoperatively (p = 0.018). At the final followup, the mean VAS satisfaction score was  $8.7 \pm 1.3$  (Table 3).

Of the 22 shoulders that underwent examinations (radiography, ultrasonography, or MRI) at the final followup visit to detect the recurrence of SOC with loose bodies or to evaluate healing of the repaired tendon, 3 (13.6%) were found to have recurrence of SOC. As for the 3 patients who presented with SOC recurrence, 1 patient had moderate pain (VAS, 5) and required revision surgery for arthroscopic loose body removal with synovectomy. Another patient had mild shoulder pain (VAS, 4) with 20 degrees of limited external rotation. Revision surgery was recommended but not performed due to patient refusal. All repaired tendons had healed properly.

#### DISCUSSION

The presence of loose bodies in several spaces (including the GH joint, SA space, and sheath of the LHBT) is a unique characteristic of SOC of the shoulder. Significant pain relief, as well as satisfactory clinical outcomes, were obtained after arthroscopic management. An early diagnosis may help prevent OA and RCT progression in patients with SOC in the shoulder.

The pathology of SOC is poorly understood, although it has been described as synovial membrane proliferation and metaplasia and the development of cartilaginous nodules.<sup>8,14)</sup> These nodules become pedunculated and eventually detached from the synovium to form loose bodies<sup>16)</sup> that can continue to grow or undergo ossification. If left untreated, articular erosions may occur and eventually result in secondary OA.<sup>17)</sup> In our study, the patients with SOC of the shoulder with concurrent RCTs were diagnosed at a much older age than patients with SOC without RCT (58.3  $\pm$  7.2 years vs. 36.2  $\pm$  15.6 years, p < 0.001). This is likely due to the fact that the diagnosis is often missed or delayed until patients present with symptoms of RCTs. Thus, based on our results and previous research, we think that an early diagnosis of SOC of the shoulder is important to prevent the progression to OA and an RCT.

The clinical manifestations of SOC in the shoulder are nonspecific and include pain, limited ROM, joint blockade, and palpable masses.<sup>11,16)</sup> While concomitant RCT is infrequent, it can occur<sup>4,14,16)</sup> and is thought to be a distinct clinical presentation of SOC in the shoulder. In this study, all patients presented with shoulder pain, 25% had preoperative shoulder stiffness, and 25% had OA on preoperative shoulder X-rays. Concurrent RCT was found in 10 shoulders (35.7%). Delaying treatment for SOC may lead to the deterioration of the joint cartilage and secondary OA.<sup>1,4,18,19)</sup> Zini et al.<sup>20)</sup> found that 7 of 11 (63.6%) patients with SOC of the hip also had secondary OA. Mueller et al.<sup>21)</sup> reported that 5 of 12 (41.7%) patients with elbow SOC had moderate to severe OA preoperatively. In our current study, 7 of 28 shoulders (25%) had preoperative OA confirmed by plain radiography, including 6 shoulders

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(85.7%) with mild OA and 1 (14.3%) with moderate OA. The severity or presence of secondary OA do not correlate with symptoms such as pain or limited ROM. As the shoulder is not a weight-bearing joint, patient's symptoms are often insidious at disease onset and develop gradually over time, which is in contrast with the development of symptoms in other joints with SOC. Therefore, SOC of the shoulder is often diagnosed at a late stage or when symptoms of RCTs appear. Milgram<sup>22)</sup> analyzed 30 cases of SOC and identified three stages of the disease according to histopathological findings: (1) active intra-synovial disease with no loose bodies, (2) transitional lesions with both active intra-synovial proliferation and free loose bodies, and (3) multiple osteochondral loose bodies with no active intra-synovial disease. The diagnosis of SOC before the appearance of loose bodies is challenging due to its nonspecific presentation. When the loose bodies are identified on imaging studies or result in obvious symptoms, the diagnosis of SOC is less challenging.

Early treatment of SOC of the shoulder is important to relieve pain and prevent further damage to the joint cartilage and tendon, and surgical intervention is necessary in most patients. An open synovectomy with loose body removal is considered the conventional treatment.<sup>23,24)</sup> Arthroscopic treatment of SOC has recently been reported to result in low morbidity and postoperative pain and an early return to function.<sup>23,25,26)</sup> Furthermore, the arthroscopic technique allows for the complete visualization of the GH joint and its associated recesses, including the axillary pouch and the sheath of the LHBT; therefore, the detection and removal of loose bodies can be performed easily using multiple portals. The removal of loose bodies is essential and effective to relieve the symptoms of SOC.<sup>24,27)</sup> Schoeniger et al.<sup>28)</sup> reported that the removal of loose bodies and a synovectomy were necessary to prevent

disease recurrence. The recurrence rate of SOC of the shoulder has been reported as 3%–23% after open surgery or arthroscopy.<sup>8,10,26)</sup> In this study, the removal of loose bodies and a complete synovectomy were performed in all patients with no complications. Although intraoperative fluoroscopy was not performed in all institutions, imaging tests such as plain radiography, MRI, or CT were used within 3 months of the surgery, and no remaining loose bodies were detected. SOC recurred in 3 of 22 shoulders (13.6%) at the final visit. Revision surgery was required in 1 shoulder.

Distinct from SOC of the knee, hip, and other joints, loose bodies are often found in several spaces in SOC of the shoulder, including the GH joint, SA space, and sheath of the LHBT. Previous studies have reported SOC in both the GH joint,<sup>1,12)</sup> SA space,<sup>9,13)</sup> and less commonly in the sheath of the LHBT,<sup>6,10,11)</sup> although none of the previous studies fully investigated the clinical characteristics of SOC of the shoulder. In a histopathological study of 30 cases, only 4 of them involved the shoulder joint.<sup>22)</sup> Lunn et al.<sup>10)</sup> investigated the presence of SOC of the shoulder and the outcomes after arthroscopic debridement and synovectomy with or without open biceps tenodesis in 15 patients. There are few reports describing SOC of the shoulder with concurrent RCTs. In a retrospective study, 10 cases of SOC of the shoulder were reviewed and 2 shoulders were found to have concomitant partial-thickness RCTs.<sup>26)</sup> Neumann and Garrigues<sup>14)</sup> presented a case with multiple SA loose bodies resulting in a supraspinatus tear. Ko et al.<sup>16</sup> reported a shoulder with calcified bodies both in the SA bursa and GH joint, causing complete RCTs.

To the best of our knowledge, this is the largest study (Table 4) evaluating the clinical features of SOC of the shoulder: it includes the greatest number of cases of SOC with concomitant RCT. A relatively long follow-up

Table 4. Characteristics of Reported SOC of the Shoulder in the Previous Literature							
Study	Number Mean age (yr)	Mean age	Symptom duration	Preoperative	Localization		_ Rotator cuff tear
		(mo)	ΟA -	GH	SA		
Milgram <sup>22)</sup>	4	39.5	Not listed	Not listed	Not l	sted	Not listed
Lunn et al. <sup>10)</sup>	15	33.3	34.2	8	15	0	Not listed
Utashima et al. <sup>26)</sup>	10	40.7	63	5	6	4	2
Neumann and Garrigues <sup>14)</sup>	1	39	60	Not listed	+	+	+
Ko et al. <sup>16)</sup>	1	46	48	Not listed	+	+	+

SOC: synovial osteochondromatosis, OA: osteoarthritis, GH: loose bodies in the glenohumeral joint, SA: loose bodies in the subacromial space, +: presence of loose bodies or cuff tearing.

(83.6 months; range, 24-154 months) is also a strength of our study. Based on our results, approximately 20% of the patients had loose bodies in the GH joint and approximately 40% of the patients had LHBT involvement. Several case reports have demonstrated that loose bodies in the SA bursa may cause RCTs.<sup>9,15,16)</sup> All RCTs in this study involved the supraspinous tendon only, and 90% of the shoulders with concurrent RCT had multiple loose bodies in the SA space. The majority of RCTs in this study were medium tears. Therefore, we recommend that surgeons carefully evaluate the biceps groove to detect any loose bodies. An evaluation of the integrity of the rotator cuff tendon may be necessary in patients with loose bodies in the SA space. This study is the first case series of patients with SOC of the shoulder with a large number of patients with concomitant RCT. We used several functional scores and assessed the recurrence of RCTs during a 2-year follow-up period. Based on the results, a significant improvement in all postoperative functional scores were observed, and an overall mean patient satisfaction VAS score of 8.6 was achieved.

The present study has several limitations. First, the sample size is relatively small as SOC of the shoulder is a rare condition. Second, our study is limited by its retrospective nature. Third, this study was performed by different surgeons in four institutions, which could have affected the results. Fourth, the follow-up period of 2 years was relatively short (83.6 months), and further research with longer follow-up is needed to detect the recurrence of symptoms. Finally, a comparative analysis of clinical characteristics was not conducted between the SOC of the shoulder and other joints.

This is the largest retrospective cohort study among

patients with SOC of the shoulder. Pain relief and patient satisfaction were achieved via arthroscopic management. Unlike other joints, loose bodies can occur in several spaces in the shoulder simultaneously, including the GH joint, SA space, and sheath of the LHBT. Early diagnosis of SOC of the SA space can help prevent OA and RCT progression.

# **CONFLICT OF INTEREST**

No potential conflict of interest relevant to this article was reported.

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