ORIGINAL

Differences in incidence rate and onset timing of undiagnosed finger symptom among shoulder surgeries related to complex regional pain syndrome

Jun Kawamata^a, Naoki Suenaga^b, Naomi Oizumi^b, Hisashi Matsumoto^c, Akira Kikuchi^d, and Masataka Inoue^e

^aDepartment of Orthopaedic Surgery, Kaisei Hospital, Hokkaido, Japan, ^bUpper Extremity Center of Joint Replacement and Endoscopic Surgery, Hokushin Orthopaedic Hospital, Hokkaido, Japan, ^cDepartment of Rehabilitation, Hokushin Orthopaedic Hospital, Hokkaido, Japan, ^dDepartment of Rehabilitation, Kochi, Japan

Abstract: The purpose of this study was to clarify the difference in onset timing and incidence of undiagnosed finger symptom (UDFS) between various shoulder surgical procedures. In this study, UDFS symptoms included the following four symptoms in the fingers; edema, limited range-of-motion, skin color changes, and abnormal sensations. UDFS cases were defined as those presenting with at least one UDFS. In result, the incidence rate of UDFS cases was 7.1% overall (58/816 shoulders), 7.4% (32/432) in arthroscopic rotator cuff repair (ARCR), 9.0% (11/122) in open rotator cuff repair (ORCR), 1.4% (2/145) in arthroscopic subacromial decompression (ASD), 13.2% (5/38) in open reduction and internal fixation (ORIF), 11.1% (3/27) in humeral head replacement, 4.8% (1/21) in anatomical total shoulder arthroplasty, and 12.9% (4/31) in reverse total shoulder arthroplasty cases. The Rate was significantly higher with ARCR compared to ASD (p < .01). About onset timing in weeks postoperatively, the ORIF group had a statistically earlier symptom onset than the Rotator cuff repair (ARCR + ORCR) group (2.4 weeks vs. 6.0 weeks, p < .01). When classifying the onset timing into before and after the removal of the abduction pillow, the ORIF group showed a statistically higher rate of onset before brace removal than the Rotator cuff repair groups (p < .01). Differences in UDFS among shoulder surgeries were demonstrated in this study. J. Med. Invest. 70 : 415-422, August, 2023

Keywords: Complex regional pain syndrome (CRPS), Perioperative complications, Shoulder surgery, edema

INTRODUCTION

Complex regional pain syndrome (CRPS) is a serious complication of orthopedic surgeries that could lead to permanent disability in the extremities. However, even now, details about the pathophysiology, methods of prevention, and other aspects of CRPS remain unknown. Several reports have shown high incidence rates of CRPS following shoulder surgeries (1-3).

However, only a few in depth studies have so far been conducted on CRPS after shoulder surgeries, and the target of these studies was limited to a single surgical procedure, such as arthroscopic rotator cuff repair (ARCR) (3, 4) or subacromial surgery (5).

Several factors are reportedly related to the onset of CRPS, such as fracture (6), immobilization (7), joint contracture (3), and endothelial damage caused by the surgical incision (8). These factors vary considerably among the different shoulder surgical procedures.

Therefore, we hypothesized that the onset timing and incidence rate might be different among different shoulder surgeries. To the best of our knowledge, however, no report has indicated differences in incidence rates and onset timings of CRPS among different surgical procedures.

Early diagnosis and treatment are important in the clinical course of CRPS. However, to improve diagnostic specificity, the

Received for publication December 8, 2022; accepted June 16, 2023.

Address correspondence and reprint requests to Jun Kawamata, MD, Department of Orthopaedic Surgery, Kaisei Hospital, 16-27 Minami2chome, Nishi23jo, Obihiro City, Hokkaido 080-2473, Japan and Fax:+81-155-37-7202. E-mail:junkawamata@hotmail.com

current major diagnostic criteria for CRPS are based on the presence of two or more suggestive symptoms (9, 10). In addition, the edema of the fingers also contributes to insufficiency of activities of daily living in patients, and some cases result to develop finger contracture. To detect CRPS at an early phase, we defined undiagnosed finger symptom (UDFS) cases by the presence of ≥ 1 of four UDFS (include edema, limited ROM, skin color changes, and abnormal sensations) that are present at high rates in CRPS (11). The purpose of this study was to investigate differences in onset timing and incidence rate of UDFS among the various shoulder surgeries to verify our hypothesis.

MATERIALS AND METHODS

This research was approved by the institutional review boards of the authors' affiliated institutions. A prospective cohort study was conducted at four private orthopedic hospitals. The included surgeries were ARCR, open rotator cuff repair (ORCR), arthroscopic subacromial decompression (ASD), open reduction and internal fixation (ORIF) for proximal humeral fracture with intramedullary nailing, humeral head replacement (HHR), anatomical total shoulder arthroplasty (TSA), and reverse total shoulder arthroplasty (RSA). In consideration of the similarity of the surgical procedures, surgery for nonunion was treated as ORIF in this study.

Between January 2018 and December 2019, these surgeries were performed on 922 shoulders in 877 patients by five surgeons who had at least 20 years of experience in shoulder surgery. Unified indications for each surgery were used by the surgeons. We did not include surgeries for shoulder instability because most of these patients were much younger than those

who underwent other surgeries.

The patients were followed for 3 months after surgery to evaluate for the occurrence of UDFS, as described below. The 3-month follow-up period was based on a previous prospective study that reported that the incidence rate of CRPS diminished after the first 3 months postoperatively (12). Among these 922 shoulders, we excluded 106 shoulders from the study for the following reasons: inadequate follow-up duration (<3 months) in 90 shoulders, simultaneous wrist fracture on the same side in two, severe general postoperative complications in four, and postoperative surgical site infection in one shoulder. We also excluded nine cases with the preoperative presence of UDFS. None of the 816 included cases had a previous history of CRPS.

As a result, this study evaluated 816 shoulders in 783 patients. The mean age at surgery was 65.9 years (range, 16–92 years). The patients included 406 men and 410 women. Details about their preoperative diagnoses and surgeries are presented in Table I.

In arthroscopic surgeries, we additionally performed capsular release if contractures were found by evaluation under anesthesia. In prosthetic surgeries for rotator cuff tear arthropathy (CTA), cuff reconstruction with muscle tendon transfers were also performed (13). A systematic postoperative rehabilitation program was conducted in all the patients according to their diagnosis and surgical procedure. In the program, the abduction pillow fixation period was also defined by each surgery, as shown in Table II. In all the cases, finger, elbow, and scapular range-of-motion (ROM) exercises were started on the day after surgery.

We defined UDFS by referencing Veldman's report (11). On the basis of the results of this report, we chose four objective finger symptoms that present high incidence rates in the early phase of CRPS, namely edema, limited ROM, skin color changes, and abnormal sensations. UDFS cases were defined as those presenting at least one UDFS.

Table I. The number of Patients' diseases and surgical procedures

disease	n (shoulders)	Surgical procedure	n (shoulders)
RCT	579	ARCR	432
IM	131	ORCR	122
Fracture	41	ASD	145
CTA	38	ORIF	38
OA	14	HHR	27
LOOSE	5	TSA	21
RA	4	RSA	31
NU	2		
ON	1		
CAD	1		

Data are expressed as the number of shoulders. RCT, rotator cuff tear. IM, impingement syndrome.

Fracture, fracture of proximal humerus. CTA, cuff tear arthritis. OA, osteoarthritis of glenohumeral joint.

LOOSE, component loosening of prosthesis of shoulder joint. RA, rheumatoid arthritis. NU, nonunion of proximal humerus fracture. ON, osteonecrosis of humeral head. CAD, chronic anterior dislocation

ARCR, arthroscopic rotator cuff repair. ORCR, open rotator cuff repair. ASD, arthroscopic subacromial decompression. ORIF, open reduction and internal fixation for proximal humeral fracture with intramedullary nailing. HHR, humeral head replacement. TSA, anatomical total shoulder arthroplasty. RSA, reverse total shoulder arthroplasty.

Table II. Postoperative rehabilitation programs

Surgical procedure	start shoulder ROM	Abduction pillow fixation period
ASD	1 day	2 days
ARCR/ORCR	$1 \mathrm{day}\text{-}2 \mathrm{weeks}$	6-8 weeks
HHR	2-4 weeks	8 weeks
ORIF	1-2 weeks	4-6 weeks
TSA	1 day	2 weeks
RSA	2-4 weeks	4-6 weeks

ARCR, arthroscopic rotator cuff repair. ORCR, open rotator cuff repair. ASD, arthroscopic subacromial decompression. ORIF, open reduction and internal fixation for proximal humeral fracture with intramedullary nailing. HHR, humeral head replacement. TSA, anatomical total shoulder arthroplasty. RSA, reverse total shoulder arthroplasty.

All the cases were observed for the occurrence of UDFS for 3 months postoperatively. In the UDFS cases, the number and onset timing of positive symptoms were recorded. Patient factors (age, sex, history of previous shoulder surgeries, and fracture or non-fracture) were investigated in all the cases. In the fracture cases, the interval between the fracture and surgery was also investigated. The preoperative Constant score for pain (graded, No; 15 pts, Mild pain; 10 pts, Moderate; 5 pts, Severe or permanent; 0.) and ROM of the shoulder (active flexion and external rotation) was also evaluated in all the cases except those with a fracture, chronic anterior dislocation, and traumatic rotator cuff tear, because these patients could not move their shoulder due to the presence of severe pain.

Statistical analysis

The individual factors were statistically evaluated between the UDFS cases (group U) and non-UDFS cases (group N). In the fracture cases, the time period between the fracture and surgery was evaluated as a risk factor. In the arthroscopic surgeries (ARCR + ASD), the addition of capsule release was also evaluated as a risk factor.

To clarify the factors affecting the incidence rate of UDFS, incidence rates were compared among several subgroups of the surgical procedures. The ARCR and ASD groups were compared to determine the difference between arthroscopic surgeries, and the ARCR and ORCR groups were compared to determine the difference between the two rotator cuff repair procedures. The TSA, RSA, and HHR groups were compared to determine the difference in terms of the various prosthetic replacements performed; acute fracture cases were excluded from the comparison to eliminate the effects of the fracture, as already reported (14).

For assessment of the onset timing of the UDFS, patients were grouped as the ORIF group, rotator cuff repair (RCR) group (ARCR + ORCR), and prosthesis replacement group (TSA + HHR + RSA) for comparison.

The Mann–Whitney U test was used for analysis of quantitative data and Fisher's exact probability test was used for categorical data. A multi-group comparison and Bonferroni correction were used for comparison of the results in ≥ 3 groups. We set 5% as the level of significance. Logistic regression analysis was used in the comparison between groups C and N, to determine the correlation between individual factors and UDFS.

All the statistical analyses were performed with EZR (Saitama Medical Center, Jichi Medical University, Saitama, Japan), which is a graphical user interface for R (The R Foundation for Statistical Computing, Vienna, Austria).

RESULTS

The overall incidence rate of UDFS cases was 7.1% (58/816 shoulders). Table III shows the incidence rates of each surgery. A high rate of UDFS cases was observed in the ORIF and RSA groups, which included relatively many fracture cases.

The symptoms appeared at a median of 5.5 weeks (range, 1.7–10.0 weeks) after surgery. Edema was present in almost all the cases. In contrast, skin color changes and abnormal sensations were present in only a few patients (Figure 1). Except for the two patients who were lost to follow-up before attaining cure, complete disappearance of the symptoms was confirmed in all

Table III. Incidence rate and onset timing of UDFS

	fracture case (shoulders)	UDFS cases (shoulders)	onset timing (weeks)
ARCR ($n = 432$)	-	32 (7.4%)	6.0 (2.0-10.0)
ORCR ($n = 122$)	-	11 (9.0%)	7.6 (2.0-10.0)
ASD $(n = 145)$	-	2 (1.4%)	2.7 (1.7-3.7)
ORIF $(n = 38)$	36	5 (13.2%)	2.4 (2.0-3.7)
HHR $(n = 27)$	-	3 (11.1%)	8.0 (2.0-8.0)
TSA (n = 21)	-	1 (4.8%)	2.6
RSA $(n = 31)$	5	4 (12.9%)	3.65 (1.7-6.7)
total (n = 816)	41	58 (7.1%)	5.5 (1.7-10.0)

ARCR, arthroscopic rotator cuff repair. ORCR, open rotator cuff repair. ASD, arthroscopic subacromial decompression. ORIF, open reduction and internal fixation for proximal humeral fracture with intramedullary nailing. HHR, humeral head replacement. TSA, anatomical total shoulder arthroplasty. RSA, reverse total shoulder arthroplasty. fracture case and UDFS cases were expressed as the number of shoulders (%). Onset timing were expressed as the number of post operative weeks of median (range).

the patients who presented with UDFS, although the timing of symptom resolution was not clarified in some patients with a long interval between follow-up visits to the hospital.

Regarding individual risk factors, no statistically significant differences were found between groups U and N (Table IV). The mean (\pm SD) preoperative interval between the injury and surgery in fracture cases did not differ significantly between UDFS and non-UDFS cases (22.3 \pm 29.4 days vs. 12.1 \pm 16.9 days, p = .09). In arthroscopic surgeries, addition of capsule release did not prove to be a risk factor for UDFS (odds ratio (OR), 1.51; 95% confidence interval (CI), 0.57–3.57; p = .35). In the comparison of incidence rates between the different procedures, the

(number of presented symptoms in 1 patient)

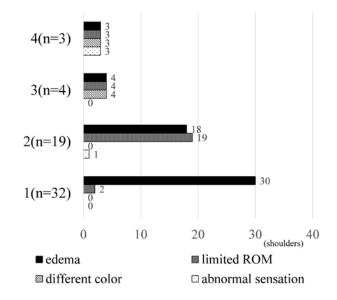


Figure 1. Incidence rates of the four UDFS ROM, range of motion. The data represent the number of shoulders.

 $\textbf{Table IV}. \quad \text{Multivariable analysis of preoperative epidemiologic data within each group (Group U vs. Group N) } \\$

Variable	Group U (n = 58)	Group N (n = 763)	OR (95%CI)	P value	
Age (yr)	67.7 ± 9.5	65.8 ± 11.0	0.99 (0.97-1.02)	.64	
sex			1.01 (0.64-2.09)	.62	
Male	29 (50.0%)	377 (49.7%)			
Female	29 (50.0%)	381 (50.3%)			
Constant pain score (points)	4.7 ± 4.2	6.1 ± 4.0	1.04 (0.96-1.11)	.34	
ROM (degrees)					
Active flexion	137.0 ± 28.5	137.2 ± 30.7	1.00 (0.99-1.01)	.64	
External rotation	44.4 ± 18.0	46.5 ± 19.7	1.01 (0.99-1.02)	.52	
History of shoulder surgery	10 (17.2%)	129 (17.0%)	1.02 (0.48-2.18)	.96	
fracture case	7 (12.0%)	34 (4.5%)	120000 (0-inf)	.99	

OR odds ratio CL confidence interval

Data are expressed as the number of shoulders (%) or as the mean \pm standard deviation. (*P < .05) No individual risk factor detected (even in the method of forward-backward stepwise selection method).

incidence rate of UDFS was significantly higher in the ARCR group than the ASD group (ARCR, 32 shoulders (7.4%); ASD, 2(1.4%); OR, 5.7; 95%CI, 1.4-49.8; p = .007) (Table V). No significant differences were found between the ARCR and ORCR groups (Table VI). Further, no significant differences were found between the HHR, TSA, and RSA groups (Table VII).

Regarding the onset timing of UDFS, the ORIF group had

a statistically significantly earlier onset of symptoms than the RCR group (ARCR + ORCR) (Figure 2; Table VIII). When classifying the onset timing into before and after brace removal, the ORIF group showed a statistically higher rate of symptom onset before brace removal than the RCR group. The onset time of UDFS was not significantly different between the prosthetics group as compared to the other two groups (Table VIII).

Table V. Preoperative epidemiologic data and incidence rate within each group (ARCR vs ASD)

Variable	ARCR (n = 432)	ASD (n = 145)		P value
Age (yr)	65.3 ± 9.8	60.6 ± 11.8		<.001*
sex				.56
Male	221 (51.1%)	70 (48.3%)		
Female	211 (48.9%)	75 (51.7%)		
Constant pain score (points)	6.5 ± 3.7	5.1 ± 4.2		.001*
ROM (degrees)				
Active flexion	144.4 ± 25.5	128.9 ± 29.0		<.001*
External rotation	50.6 ± 17.3	39.7 ± 20.2		<.001*
History of shoulder surgery	75 (17.4%)	21 (15.3%)		.60
Variable	ARCR (n = 432)	ASD (n = 145)	OR (95%CI)	P value
UDFS cases (%)	32 (7.4%)	2 (1.4%)	5.7 (1.4-49.8)	.007*

Data are expressed as the number of shoulders (%) or as the mean \pm standard deviation. (*P < .05)

Table VI. Preoperative epidemiologic data and incidence rate within each group (ARCR vs ORCR)

Variable	ARCR (n = 432)	ORCR (n = 122)		P value
Age (yr)	65.3 ± 9.8	67.1 ± 0.9		.07
sex				.01*
Male	221 (51.1%)	84 (68.9%)		
Female	211 (48.9%)	38 (31.1%)		
Constant pain score (points)	6.5 ± 3.7	6.5 ± 0.4		.79
ROM (degrees)				
Active flexion	144.4 ± 25.5	135.2 ± 3.0		.006*
External rotation	50.6 ± 17.3	46.0 ± 1.7		.02*
History of shoulder surgery	75 (17.4%)	19 (15.6%)		.67
Variable	ARCR (n = 432)	ORCR (n = 122)	OR (95%CI)	P value
UDFS cases (%)	32 (7.4%)	11 (9.0%)	0.80 (0.38-1.83)	.56

CI, confidence interval. SMD, standardized mean difference

CI, confidence interval ARCR, arthroscopic rotator cuff repair.

ASD, arthroscopic subacromial decompression.

ARCR, arthroscopic rotator cuff repair.

ORCR, open rotator cuff repair.

Data are expressed as the number of shoulders (%) or as the mean \pm standard deviation. (*P < .05)

(95%CI)

1.65

(0.08-103.11)

1.00

1.00

Variable	HHR $(n = 27)$	TSA (n = 21)	RSA ($n = 26$)		P value				
				HHR v	s TSA	HHR v	s RSA	TSA vs	sRSA
Age (yr)	68.9 ± 1.5	73.0 ± 2.4	81.6 ± 6.7	.12	2	<.00	01*	.00	8*
sex				.0′	7	.5	7	.3	3
Male	12 (44.4%)	4 (19.0%)	9 (34.6%)						
Female	15 (55.6%)	17 (81.0%)	17 (65.4%)						
Constant pain score (points)	5.7 ± 0.8	3.1 ± 0.8	4.8 ± 4.7	.02	2	.4	0	.2	3
ROM (degrees)									
Active flexion	135.9 ± 4.7	108.3 ± 7.6	105.8 ± 44.0	.004	4*	.00	8*	.9	4
External rotation	45.0 ± 3.9	33.1 ± 5.3	30.4 ± 21.1	.008*		.5	6		
History of shoulder surgery	5 (18.5%)	9 (42.9%)	12 (46.2%)	.11 .04		1.0	00		
Variable	HHR (n = 27)	TSA (n = 21)	RSA (n = 26)	HHRv	sTSA	HHRv	sRSA	TSAvs	sRSA
				OR (95%CD	P value	OR (95%CD	P value	OR (95%CD	P valu

Table VII. Multiple comparison of preoperative epidemiologic data and incidence rate within each group (HHR vs. TSA vs. RSA)

CI, confidence interval. SMD, standardized mean difference. HHR, humeral head replacement. TSA, anatomical total shoulder arthroplasty. RSA, reverse total shoulder arthroplasty. Data are expressed as the number of shoulders (%) or as the mean \pm standard deviation. (*P<.0167 after Bonferroni correction)

2(7.7%)

(95%CI)

0.41

(<0.01-5.53)

(95%CI)

0.67

(0.05-6.42)

.62

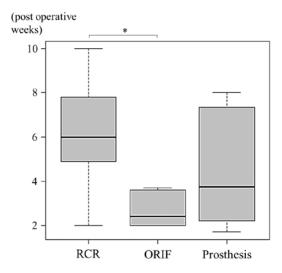


Figure 2. Box-and-whisker plot depicting the onset times of UDFS RCR, rotator cuff repair. ORIF, open reduction and internal fixation. (*P < .0167 after Bonferroni correction).

Table VIII. Onset timing of each group of UDFS

UDFS cases (%)

3 (11.1%)

1 (4.8%)

Variable	RCR ($n = 43$)	ORIF(n = 5)	Prosthesis $(n = 8)$	P value		
				RCRvsORIF	RCRvs Prosthesis	ORIFvs Prosthesis
Postoperative weeks	6.0 (2.0-10.0)	2.4 (2.0-3.7)	3.8 (1.7-8.0)	.002*	.11	.38
Relation to brace removal				<.001*	.43	.11
Before brace removal	14 (32.6%)	5 (100%)	4 (50.0%)			
After brace removal	29 (67.4%)	0	4 (50.0%)			

RCR, rotator cuff repair. ORIF, open reduction and internal fixation.

Data are expressed as the number of weeks of median (range). Incidence rate are expressed the number of shoulders (%). ($^*P < .0167$ after Bonferroni correction)

DISCUSSION

In this study, we clarified the differences in incidence rates and onset timings of UDFS among various shoulder surgeries. The incidence rate of CRPS has been reported in several studies as 0.9%-2.9% in upper extremity surgeries (15-17), 1%-8.2% following ARCR (1, 18), and 13.7% following subacromial shoulder surgery (5). Tanesue reported that the incidence rate of CRPS after ARCR was 0.5-24.2% in Japanese people (4).

Many reports used the criteria for diagnosis of CRPS as the presence of >2 of the four UDFS assessed in our study (2, 9, 10). These criteria have been shown to have a high specificity for making a diagnosis of CRPS. On the other hand, use of these criteria might lead to a delay in diagnosis because patients usually show only one symptom in the early stages of CRPS.

Regarding the clinical course of CRPS, Varitimidis *et al.* reported that the acute phase of the syndrome was reversed (19). De Mos reported a poor outcome in a patient with CRPS who presented a high number of symptoms (20). These reports suggest that early detection and therapeutic intervention are obviously important to improve the clinical course of CRPS.

Therefore, in this study, we defined UDFS cases as the presence of at least one symptom, to facilitate the early detection of UDFS cases. Using the current, more sensitive criteria, the overall incidence rate of UDFS cases in this study was 7.1%, although this would probably have been lower if we had applied other criteria of ≥ 2 positive symptoms.

The reason for the lower incidence rate of UDFS cases in our study is not clear, but our systematic postoperative care could have affected this result. We typically use cryotherapy with an icing machine for pain relief during the early postoperative period (21). We also use interscalene brachial plexus block to control pain, adding dexamethasone to prolong the duration of the nerve block to >24 hours after surgery (22). As part of the rehabilitation program, we start ROM exercises of the scapula, elbow, and finger soon after surgery. These interventions could have contributed to preventing UDFS in the majority of our patients.

Previous studies reported that the individual factors related to CRPS were age (23), female sex (10, 24), preoperative pain (25), preoperative contracture (3), and fracture (6). In our investigation, none of these factors was statistically significantly different between groups U and N. Further, although the odds ratio for developing UDFS was extremely high in patients with fractures, no significant difference was found in logistic regression analysis, probably because the sample size was too small.

Guo reported that tibial fractures with 4 weeks of cast immobilization induced CRPS-like changes in rats (7). This suggests that preoperative immobilization might be a risk factor for CRPS, which could be one of the causes of the high incidence rate of CRPS in fracture cases. Moreover, we hypothesized that the waiting period in fracture cases might relate to the onset of CRPS; however, we found no significant correlation between the duration of the preoperative period and the development of CRPS

In the present study, ARCR cases had a higher incidence rate of UDFS than ASD cases. In a previous study, preoperative contracture was reported to be a risk factor for CRPS (3); however, no significant differences in preoperative ROM and the addition of a capsule release procedure were found between the two surgical procedures. Other factors that would likely be different between the two surgical methods would be operation time, postoperative pain due to rotator cuff traction, etc.; however, the most considerable difference observed between ARCR and ASD cases in our study was the brace fixation duration (Table II), which might explain the high percentage of ARCR cases that showed UDFS after brace removal.

During the brace fixation period, mechanical stimulation of the bone by the skeletal muscle might decrease, resulting in neuropathy of the extremity. Terkelsen $et\,al$. reported that immobilization of healthy upper extremities for 4 weeks induced increases in the levels of inflammatory mediators (IL-1 β , IL-6, and TNF- α) and similar clinical changes (allodynia, temperature changes, and edema) as those elicited by limb fracture with casting (26). Therefore, the longer immobilization period might have induced the UDFS in the ARCR group.

In contrast, the incidence rate of UDFS did not differ significantly between ARCR and ORCR patients. Regarding the pathophysiology of CRPS, Groeneweg et al. reported that endothelial damage caused by surgical incision triggers vasoconstriction that induces CRPS (7, 8). On the other hand, Mertz et al. reported no significant difference in the incidence rate of CRPS between endoscopic and open release procedures for carpal tunnel release (24). In their retrospective cohort study on distal radial fractures, Wang et al. reported that use of a volar locking plate lowered the incidence rate of CRPS as compared with external fixation (27). These reports indicate that skin incision is not necessarily related to the development of CRPS type1. However, cutaneous nerve injury generates CRPS type 2. Therefore, we make a skin incision from the acromion to the outside of the coracoid process when performing shoulder surgeries to reduce the risk of cutaneous neuropathy.

Moreover, we found no significant difference between the three arthroplasties. In other words, no association was found between the process of glenoid component insertion and UDFS. Mortazavi *et al.* reported that compression damage to the main trabecular structure, osteotomy, and hematoma formation associated with the component caused an increase in the level of inflammatory mediators, which might be involved in the development of CRPS (28). However, no previous studies compared the incidence of CRPS between prosthetic surgeries with and without glenoid surfacing. We also could not deny the influence of glenoid component insertion on the occurrence of CRPS due to the small number of cases evaluated.

Amongst the various prosthetic surgical procedures, RSA has a higher potential risk than TSA of causing nerve stretching during surgery, which can induce CRPS (29). However, the present study showed no significant difference among the prosthetic surgeries after exclusion of fracture cases. Additionally, the overall incidence rate of UDFS was high in the ORIF and RSA groups. This might indicate that the higher occurrence of UDFS in these patients was due to the presence of fractures rather than to the surgical procedure itself.

Considering the difference in onset timing between UDFS following RCR and ORIF, the pathophysiology of CRPS might have affected the difference. A longer period of brace fixation was required in the postoperative treatment of RCR than ORIF. As described earlier, postoperative immobilization might be one of the causes of UDFS occurring immediately after brace removal. However, the reason for the onset of symptoms after brace removal is still unclear. It is possible that brace removal with the shoulder in the drooping position leads to traction on the brachial plexus and blood vessels, which in turn leads to poor circulation in the upper extremity, as is seen in thoracic outlet syndrome.

On the other hand, in fracture healing, the hematoma in the early phase contains hematopoietic and immune cells (30), and causes the local release of inflammatory mediators. The consequent local inflammation might induce UDFS in the early stage after surgery. In addition, preoperative fracture immobilization could be related to the onset of CRPS, as already stated (6). As the pathophysiology of CRPS is still unknown, our discussion about the onset timing of CRPS remains a hypothesis, and further studies are necessary to clarify the pathology of each

symptom. Our results might enable earlier diagnosis and therapeutic intervention for UDFS, especially after high-risk shoulder surgeries, which will improve the prognosis and prevent progression to severe CRPS.

Limitations

The present study has several limitations. First, the number of cases in each surgery group was small, although some of the results were proved to be statistically significant. Second, because the study involved subjective evaluation by each surgeon, the diagnosis of UDFS might have involved some bias. In the future, symptoms should be diagnosed more objectively, for example, by thermographic recording of skin temperature (31) and the pain-to-heat test with an air-pressure-controlled thermode (31). Third, this study included surgeries performed by multiple surgeons. Lastly, there is a possibility that we did not accurately detect the onset timing of UDFS because the patients were examined by the surgeons at certain intervals after surgery.

Future studies with longer follow-up will be useful for providing data on the correlation between the recovery period and factors such as the surgical procedure, number of symptoms, and individual characteristics of the patients.

CONCLUSION

In the present study, UDFS were observed in 7.1% of the cases after various shoulder surgeries. The incidence rate of UDFS was significantly higher following ARCR than ASD. ORIF was associated with a statistically earlier onset of UDFS than RCR. These findings will be useful when considering the start of physiotherapy after shoulder surgeries from the perspective of prevention of CRPS, and could contribute to the early detection of and therapeutic intervention for CRPS.

CONFLICT OF INTEREST

All authors declare no conflicts of interests.

ACKNOWLEDGEMENT

The authors thank Shintaro Yamane, Chika Yoshioka, and Tomoya Matsuhashi for contributing their cases to this study. And authors would like to thank Yukiyoshi Hisada for his technical advices.

REFERENCES

- Arndt J, Clavert P, Mielcarek P, Bouchaib J, Meyer N, Kempf JF, French Society for Shoulder & Elbow (SOFEC): Immediate passive motion versus immobilization after endoscopic supraspinatus tendon repair: a prospective randomized study. Orthop Traumatol Surg Res 98: S131-8, 2012
- Bruehl S, Harden RN, Galer BS, Saltz S, Bertram M, Backonja M, Gayles R, Rubin N, Bhugra MK, Stanton-Hicks M: External validation of IASP diagnostic criteria for Complex Regional Pain Syndrome and proposed research diagnostic criteria. Pain 81: 147-54, 1999
- Hidani K, Nagoshi M, Miyamoto T: The investigation of risk factors for onset and severity of crps type 1 after arthroscopic rotator cuff repair. J Shoulder Elbow Surg 24:e348, 2015
- 4. Tanesue R, Gotoh M, Mitsui Y, Nakamura H, Honda H,

- Ohzono H, Shimokobe H, Tokunaga T, Imai T, Okawa T, Shiba N: Hand lesion after arthroscopic rotator cuff repair: Association with complex regional pain syndrome. J Orthop Sci 23: 70-74, 2018
- Laumonerie P, Martel M, Tibbo ME, Azoulay V, Mansat P, Bonnevialle N: Influence of vitamin C on the incidence of CRPS-I after subacromial shoulder surgery. Eur J Orthop Surg Traumatol 30: 221-226, 2020
- Sandroni P, Benrud-Larson LM, McClelland RL, Low PA: Complex regional pain syndrome type I: incidence and prevalence in Olmsted county, a population-based study. Pain 103: 199-207, 2003
- Guo TZ, Wei T, Li WW, Li XO, Clark JD, Kingery WS: Immobilization contributes to exaggerated neuropeptide signaling, inflammatory changes, and nociceptive sensitization after fracture in rats. J Pain 15: 1033-45, 2014
- Groeneweg JG, Huygen FJ, Heijmans-Antonissen C, Niehof S, Zijlstra FJ: Increased endothelin-1 and diminished nitric oxide levels in blister fluids of patients with intermediate cold type complex regional pain syndrome type 1. BMC Musculoskelet Disord 7:91, 2006
- 9. Amadio PC, Mackinnon SE, Merritt WH, Brody GS, Terzis JK: Reflex sympathetic dystrophy syndrome: consensus report of an ad hoc committee of the American Association for Hand Surgery on the definition of reflex sympathetic dystrophy syndrome. Plast Reconstr Surg 87: 371-5, 1991
- Sumitani M, Shibata M, Sakaue G, Mashimo T, Japanese CRPS Research Group: Development of comprehensive diagnostic criteria for complex regional pain syndrome in the Japanese population. Pain 150: 243-249. 2010
- Veldman PH, Reynen HM, Arntz IE, Goris RJ: Signs and symptoms of reflex sympathetic dystrophy: prospective study of 829 patients. Lancet 342: 1012-6, 1993
- 12. Harden RN, Bruehl S, Stanos S, Brander V, Chung OY, Adams SSA, Stulberg DS: Prospective examination of pain-related and psychological predictors of CRPS-like phenomena following total knee arthroplasty: a preliminary study. Pain 106: 393-400, 2003
- 13. Urita A, Funakoshi T, Suenaga N, Oizumi N, Iwasaki N: A combination of subscapularis tendon transfer and smallhead hemiarthroplasty for cuff tear arthropathy: a pilot study. Bone Joint J 97: 1090-5, 2015
- Guo TZ, Offley SC, Boyd EA, Jacobs CR, Kingery WS: Substance P signaling contributes to the vascular and nociceptive abnormalities observed in a tibial fracture rat model of complex regional pain syndrome type I. Pain 108: 95-107, 2004
- Kleinert HE, Cook FW, Kutz JE: Neurovascular disorders of the upper extremity treated by transaxillary sympathectomy. Arch Surg 90: 612-616,1965
- Kozin F, Genant HK, Bekerman C, McCarty DJ: The reflex sympathetic dystrophy syndrome. Am J Med 60: 321-31, 1976
- Poplawski ZJ, Wiley AM, Murray JF: Posttraumatic dystrophy of the extremeties. J Bone Joint Surg 65: 642-55,1983
- Borgeat A, Ekatodramis G, Kalberer F, Benz C: Acute and nonacute complications associated with interscalene block and shoulder surgery: a prospective study. Anesthesiology 95: 875-80, 2001
- Varitimidis SE, Papatheodorou LK, Dailiana ZH, Poultsides L, Malizos KN: Complex regional pain syndrome type I as a consequence of trauma or surgery to upper extremity: management with intravenous regional anaesthesia, using lidocaine and methyloprednisolone. J Hand Surg Eur 36:771-7, 2011
- 20. De Mos M, De Bruijn AGJ, Huygen FJPM, Dieleman JP,

- Stricker BHC, Sturkenbooma MCJM: Outcome of the Complex Regional Pain Syndrome. Clin J Pain 25: 590-7, 2009
- Barbara CT: Evidence-based impact of cryotherapy on postoperative pain, swelling, drainage and tolerance after orthopaedic surgery. Pflege 20: 258-67, 2007
- 22. Morita S, Oizumi N, Suenaga N, Yoshioka C, Yamane S, Tanaka Y: Dexamethasone added to levobupivacaine prolongs the duration of interscalene brachial plexus block and decreases rebound pain after arthroscopic rotator cuff repair. J Shoulder Elbow Surg 29: 1751-7, 2020
- Kim H, Lee CH, Kim SH, Kim YD: Epidemiology of complex regional pain syndrome in Korea: An electronic population health data study. PLoS One 13: e0198147, 2018
- 24. Mertz K, Trunzter J, Wu E, Barnes J, Eppler SL, Kamal RN: National Trends in the Diagnosis of CRPS after Open and Endoscopic Carpal Tunnel Release. J Wrist Surg 8:209-14, 2019
- 25. Kozin F, Ryan LM, Carerra GF, Soin JS, Wortmann RL: The reflex sympathetic dystrophy syndrome (RSDS). III. Scintigraphic studies, further evidence for the therapeutic efficacy of systemic corticosteroids, and proposed diagnostic criteria. Am J Med 70: 23-30, 1981
- 26. Terkelsen AJ, Bach FW, Jensen TS: Experimental forearm

- immobilization in humans induces cold and mechanical hyperalgesia. Anesthesiology 109: 297-307, 2008
- Wang J, Lu Y, Cui Y, Wei X, Sun J: Is volar locking plate superior to external fixation for distal radius fractures? A comprehensive meta-analysis. Acta Orthop Traumatol Turc 52: 334-342, 2018
- 28. Mortazavi SM, Hansen P, Zmistowski B, Kane PW, Restrepo C, Parvizi J: Hematoma following primary total hip arthroplasty: a grave complication. J Arthroplasty 28:498-503, 2013
- 29. Lowe JT, Lawler SM, Testa EJ, Jawa A: Lateralization of the glenosphere in reverse shoulder arthroplasty decreases arm lengthening and demonstrates comparable risk of nerve injury compared with anatomic arthroplasty: a prospective cohort study. J Shoulder Elbow Surg 27: 1845-51, 2018
- 30. Schindeler A, McDonald MM, Bokko P, Little DG: Bone remodeling during fracture repair: The cellular picture. Semin Cell Dev Biol 19: 459-66, 2008
- 31. Birklein F, KuÈnzel W, Sieweke N: Despite clinical similarities there are significant differences between acute limb trauma and complex regional pain syndrome I (CRPS I). Pain 93: 165-71, 2001