

Midterm Outcomes of Surgical Reconstruction and Spontaneous Recovery of Upper-Extremity Paralysis Following Acute Flaccid Myelitis

Kazuteru Doi, MD, PhD, Yasunori Hattori, MD, PhD, Sotetsu Sakamoto, MD, Dawn Sinn Yii Chia, MBBS, Vijayendrasingh Gour, MCh(Plastic), and Jun Sasaki, MD

Investigation performed at the Department of Orthopedic Surgery, Ogori Daiichi General Hospital, Yamaguchi, Japan

Background: Acute flaccid myelitis (AFM) is a disabling, poliomyelitis-like illness that mainly affects children. Although various surgical interventions are performed for intractable paralysis due to AFM, the timing of surgery and its long-term outcomes have yet to be established, especially for shoulder reconstruction. This study aimed to analyze the midterm outcomes of nonsurgically and surgically treated upper-extremity AFM and the factors influencing shoulder functional outcomes after surgical reconstruction.

Methods: We retrospectively examined 39 patients with AFM in 50 upper extremities between 2011 and 2019. The degree of spontaneous recovery of completely paralyzed muscles was evaluated at a median of 3, 6, and 37 months after the onset of paralysis. Twenty-seven patients with 29 extremities underwent surgery involving nerve transfer, muscle-tendon transfer, or free muscle transfer for shoulder, elbow, and hand reconstruction.

Results: Patients with complete paralysis of shoulder abduction at 6 months did not show later recovery. Twenty-two patients with 24 extremities underwent shoulder surgery, and all but 1 were followed for at least 24 months after surgery. Although postoperative shoulder abduction recovery was similar between transfer of the spinal accessory nerve and of the contralateral C7 nerve root to the suprascapular nerve, the outcomes obtained with spinal accessory nerve transfer had more variability, likely related to latent spinal accessory nerve paralysis, shoulder instability related to pectoralis major paralysis, and the type of paralysis. Shoulder abduction recovery was also greatly affected by scapulothoracic joint movement. In contrast, the outcomes of the elbow flexion and hand reconstructions were more consistent and acceptable.

Conclusions: All patients had loss of shoulder abduction, and restoration of shoulder function was less predictable and depended on the quality of the donor nerves and recovery of the synergistic muscles. Strict donor nerve selection and additional nerve transfer for shoulder reconstruction are imperative for satisfactory outcomes.

Level of Evidence: Therapeutic Level IV. See Instructions for Authors for a complete description of levels of evidence.

Acute flaccid myelitis (AFM) is a poliomyelitis-like illness that mainly affects children and is characterized by an acute onset of flaccid paralysis in the extremities, and it bears a high epidemiological association with enterovirus D68 (EV-D68)^{1,2}. AFM was first reported in the United States in 2012, and clusters of cases have been observed every 2 to 3 years since then in many countries³.

Incomplete recovery resulting in a persistent motor deficit is expected in >75% of patients⁴⁻⁷. The few published articles on surgical reconstruction, including nerve transfer surgery, in patients with AFM⁷⁻⁹ reported good elbow flexion recovery but variable shoulder function recovery^{8,9}. The reasons postulated for the poorer shoulder functional outcomes included specific targeting of the shoulder girdle muscles by viral replication¹⁰

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TABLE I Patient Demographics and Type of Paralysis*

Case No.	Age at Onset (yr)	Gender	Involved Side	Time from Onset to Exam. (mo)	Lower-Extremity Paralysis	Cranial Nerve Palsy	Type of Paralysis†
1	4	F	L	6	No	No	S-E-H
2	2	M	R	13	No	No	S-E-H
3	4	M	R	5	No	No	S-E
4	9	M	L	8	No	No	N-S-E
5	3	F	R	6	No	No	N-S
6	3	M	L	10	No	No	S-H
7	3	F	L	10	No	No	S-E
8	2	M	L	83	No	No	S
9	4	F	R	3	No	No	S
10	3	F	L	3	Hemi.	No	S
11	1	F	L	4	No	No	S-E
12(R)	4	F	Bil(R)	5	No	No	N-S-E
12(L)	4	F	Bil(L)	5	No	No	N
13	2	F	R	5	No	Facial	Recovered
14	5	M	L	6	No	No	N-S-H-E
15	6	F	R	7	Hemi.	No	S
16	7	M	R	4	No	No	N-S
17(R)	3	F	Bil(R)	6	No	No	S-E-H
17(L)	3	F	Bil(L)	6	No	No	S-E-H
18	2	M	L	8	Hemi.	No	S-E-H
19	3	M	R	6	No	No	S
20(R)	8	F	Bil(R)	4	Quad.	No	N-S-E
20(L)	8	F	Bil(L)	4	Quad.	No	N-S-E
21(R)	4	M	Bil(R)	4	No	No	Recovered
21(L)	4	M	Bil(L)	4	No	No	Recovered
22	3	M	R	6	No	Facial	S
23	4	M	R	6	No	No	N-S-E
24	7	F	L	4	No	No	N-S
25	1	F	R	6	No	Oculo.	S-E-H
26(R)	7	F	Bil(R)	23	No	No	N-S
26(L)	7	F	Bil(L)	23	No	No	N-S-E
27	6	F	L	80	No	No	H
28(R)	3	M	Bil(R)	6	Quad.	Facial	S-E
28(L)	3	M	Bil(L)	6		Facial	S-E
29(R)	4	M	Bil(R)	8	Quad.	No	N-S-E
29(L)	4	M	Bil(L)	8		No	N-S-E
30(R)	3	M	Bil(R)	7	No	No	S
30(L)	3	M	Bil(L)	7	No	No	N-S-E-H
31	3	F	R	6	Tri.	No	S
32	3	F	L	8	No	No	S-E
33	4	F	L	8	No	No	S
34(R)	4	F	Bil(R)	8	No	No	S-H
34(L)	4	F	Bil(L)	8	No	No	S-E-H

continued

TABLE I (continued)

Case No.	Age at Onset (yr)	Gender	Involved Side	Time from Onset to Exam. (mo)	Lower-Extremity Paralysis	Cranial Nerve Palsy	Type of Paralysis†
35(R)	3	M	Bil(R)	8	No	No	N-S
35(L)	3	M	Bil(L)	8	No	No	N-S-E
36	6	M	R	11	No	No	S-E-H
37	1	M	R	11	No	Facial	N-S
38	6	F	L	11	No	No	S-E-H
39(R)	15	M	Bil(R)	30	No	No	N-S-E
39(L)	15	M	Bil(L)	30	No	No	N-S
Median	4			6.5			
IQR	3.0 to 5.5			5.8 to 8.5			
Range	1 to 15			3 to 83			

*Bil = bilateral, Hemi = hemiplegia, Quad. = quadriplegia, Tri. = triplegia, Oculo. = oculomotor nerve palsy, Facial = facial nerve palsy. †S = shoulder, E = elbow, H = hand, N = neck.

and other factors such as the timing of surgery, selection of donor nerves, and complexity of shoulder motion¹¹.

This study aimed to report the midterm outcomes of nonsurgically and surgically treated upper-extremity AFM and to analyze the factors influencing shoulder functional outcomes after surgical reconstruction.

Materials and Methods

This was a retrospective study of patients with upper-extremity paralysis due to AFM who were referred to our hospital for potential surgical reconstruction. The study was approved by the hospital Institutional Ethics Committee, and written informed consent was obtained from all patients.

Patient Demographics

Thirty-nine patients, 35 with confirmed and 4 with probable AFM (see Appendix Supplemental Table 1)¹⁻³, were referred to our clinic for surgical reconstruction from 2011 to 2019 after initial care by their pediatricians. Patient demographics are presented in Table I. The sex distribution was approximately even, and the median age at disease onset was 4 years (interquartile range [IQR], 3 to 5.5 years). There were 11 cases of bilateral paralysis (diplegia) and 14 cases each of unilateral paralysis on the left and the right side. The median period from

onset of paralysis to our consultation was 6.5 months (IQR, 5.8 to 8.5 months). Seven patients had lower-extremity paralysis, consisting of 3 cases of quadriplegia, 3 of hemiplegia, and 1 of triplegia. Five patients had cranial nerve palsy, consisting of 4 facial nerve palsies and 1 oculomotor nerve palsy.

Preoperative Evaluations for Spontaneous Recovery

Initial neurological evaluation was performed at a median of 3 months (IQR, 3 to 4 months) after AFM onset. The senior author evaluated the motor function using the British Medical Research Council (MRC) grading scale (see Appendix Supplemental Table 2). Both face-to-face outpatient clinical findings and videos of the child's motor status in daily life taken by the family were used in the evaluation. M0 and M1 grades were differentiated from each other by surface electromyography, while M2 and M3 grades were differentiated by testing under gravity. The differentiation between M4 and M5 grades was accorded less importance during the analysis of the results because of the unreliability of differentiating them clinically in children. We did not use the Active Movement Scale, as the MRC and electrophysiological assessments are more reliable in determining muscle strength¹². We defined M0 and M1 as paralysis and M2 or greater as paresis.

TABLE II Distribution of Paralyzed Muscles (M0 and M1) in 39 Patients with 50 Affected Upper Extremities

	Neck			Shoulder			Elbow		Hand				
	Sternocleidomastoideus	Trapezius	Diaphragm	Abductor	External Rotator	Pectoralis Major	Flexor	Extensor	Wrist		Finger		
									Flexor	Extensor	Flexor	Extensor	Intrinsic Muscle
No. of extremities	10	16	19	38	31	36	22	18	14	13	12	8	10
Fraction of affected extremities	20%	32%	38%	76%	62%	72%	44%	36%	28%	26%	24%	16%	20%

TABLE III Clinical Course of Paralysis Recovery at 3 Stages*†

Patient No.	Age at Onset (yr)	Sex	Involved Side	Shoulder			Elbow			Finger								
				Abduction			Flexion			Extension								
				1st	2nd	3rd	1st	2nd	3rd	1st	2nd	3rd	1st	2nd	3rd			
1	4	F	L	0	0	3‡	0	0	3‡	0	0	0	0	0	3‡	0	0	3‡
2	2	M	R	0	0	2‡	0	2§	3‡	0	0	0	0	0	3‡	0	0	0
3	4	M	R	0	0	4‡	0	0	4‡	3	3	3	4	4	4	4	4	4
4	9	M	L	0	0	3‡	2§	2§	4‡	2	2	3	5	5	5	5	5	5
5	3	F	R	0	0	4‡	2§	2§	3	3	3	3	4	4	4	4	4	4
6	3	M	L	0	0	3‡	2§	2§	4	1	2	2	2	2	3‡	5	5	4‡
7	3	F	L	0	0	3‡	0	0	3	2	2	4	4	4	4	4	4	4
8	2	M	L		0	3‡		2§	3		4	4		4	4		4	4
9	4	F	R	2	2	3	4	4	4	4	4	4	4	4	4	4	4	4
10	3	F	L	2	2	4	3	3	3	3	3	3	4	4	4	4	4	4
11	1	F	L	0	0	2‡	3	3	3‡	0	0	0	3	3	3	0	1	1
12(R)	4	F	R	0	0	3‡	0	0	4‡	0	0	0	3	3	3	3	3	3
12(L)	4	F	L	3	3	3	3	3	3	3	3	3	3	3	3	3	3	3
13	2	F	R	3	3		3	3		4	4		4	4		4	4	
14	5	M	L	0	0		0	0		0	0		0	0		2	2	
15	6	F	R	1	1	4‡	3	3	3	3	3	3	4	4	4	4	4	4
16	7	M	R	0	0		3	3		3	3		4	4		4	4	
17(R)	3	F	R	0	0	3‡	0	0	3‡	0	0	0	0	0	0	0	0	0
17(L)	3	F	L	0	0	2‡	0	0	3‡	0	0	0	0	0	0	0	0	0
18	2	M	L	0	0	3‡	0	0	3‡	0	2	2	3	3	3	3	3	3
19	3	M	R	2	3		3§	3		2	2		4	4		4	4	
20(R)	8	F	R	0	0	0	1	1	1	0	0	0	3	3	3	3	3	3
20(L)	8	F	L	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0
21(R)	4	M	R	3	3	4	3	3	4	3	3	4	3	4	4	3	4	4
21(L)	4	M	L	3	3	4	3	4	4	3	4	4	3	4	4	3	4	4
22	3	M	R	0	0	0	2	3	3	1	2	2	3	3	4	3	3	4
23	4	M	R	0	0	0	0	0	3‡	0	0	0	3	3	4	3	3	4
24	7	F	L	2	3	3	4	4	4	2	3	3	4	4	4	4	4	4
25	1	F	R	0	0	3‡	0	0	4‡	0	0	0	3	3	3	2	2	2
26(R)	7	F	R		4	4		4	4		4	4		4	4		4	4
26(L)	7	F	L	0	0	0	0	0	0	0	0	0	4	4		4	4	
27	6	F	L		4	4		4	4		4	4		3	3		0	0
28(R)	3	M	R	0	0	0	0	0	4‡	3	3	3	3	3	3	3	3	3
28(L)	3	M	L	0	0	0	0	0	0	0	0	3	3	3	3	3	3	3
29(R)	4	M	R	0	0	0	0	0	0	0	0	0	3	3	3	3	3	3
29(L)	4	M	L	0	0	0	3	3	3	3	3	3	3	3	3	3	3	3
30(R)	3	M	R	2	2#	2#	3	3	3	2	2	2	3	3	3	3	3	3
30(L)	3	M	L	0	0	0	0	0	3‡	0	0	0	0	0	0	0	0	0
31	3	F	R	0	0	4‡	3‡	3	4	3	3	3	4	4	4	4	4	4
32	3	F	L	0	0	2‡	0	0	4‡	0	0	0	2	3	3	2	3	3
33	4	F	L	0	1	4	3	3	3	2	2	2	3	4	4	3	4	4
34(R)	4	F	R	0	0	3‡	3	3	3	2	2	2	3	3	3	0	0	0
34(L)	4	F	L	0	0	2‡	0	0	3‡	1	1	1	2	2	2	0	0	0
35(R)	3	M	R		0	3		3	4		2	2		3	3		3	3
35(L)	3	M	L		0	0		0	3‡		0	0		3	3		2	2
36	6	M	R	0	0	3‡	0	0	3‡	0	0	0	0	0	0	0	0	2‡
37	1	M	R		0	0		3	3		4	4		3	3		3	3
38	6	F	L		0	2‡		2§	3‡		2	3		3	3		0	0

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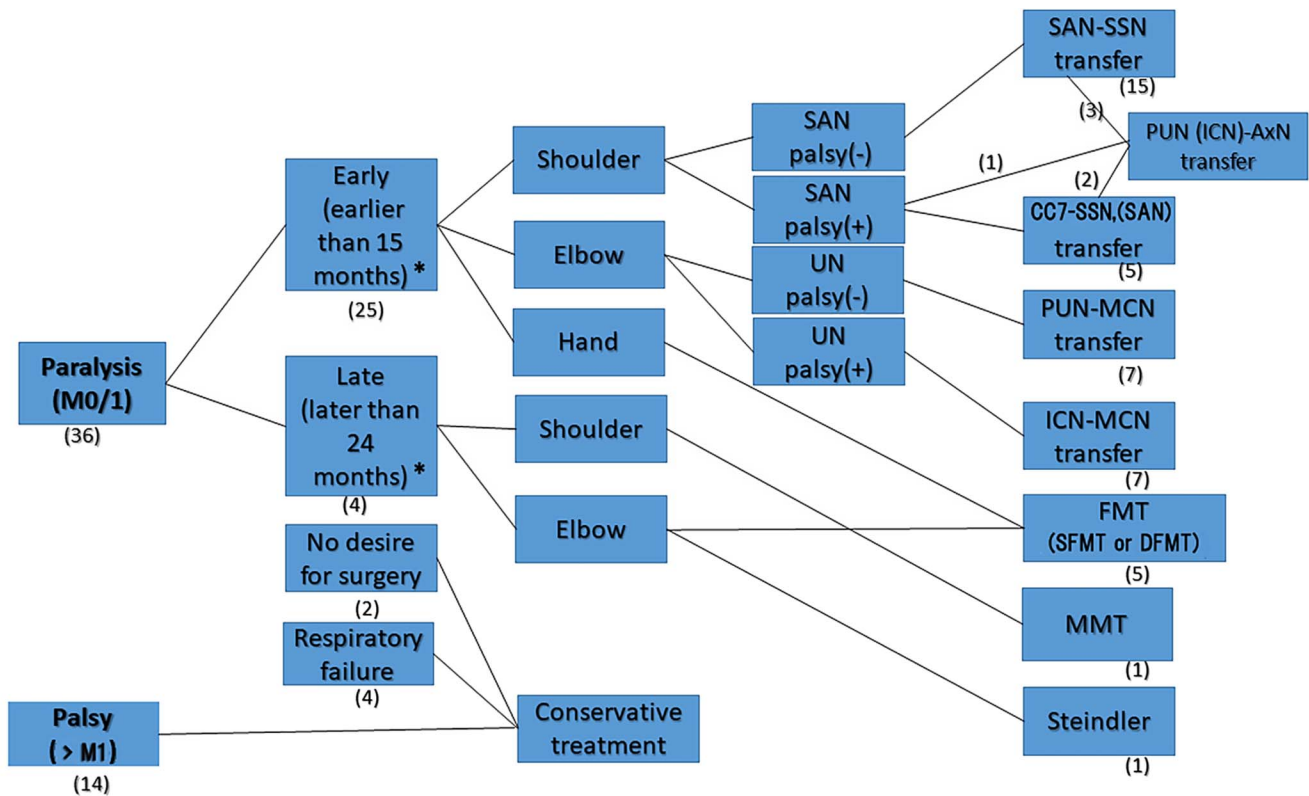
TABLE III (continued)

Patient No.	Age at Onset (yr)	Sex	Involved Side	Shoulder			Elbow			Finger								
				Abduction			Flexion			Extension			Flexion			Extension		
				1st	2nd	3rd	1st	2nd	3rd	1st	2nd	3rd	1st	2nd	3rd	1st	2nd	3rd
39(R)	15	M	R	0	0		0	4†		3	3		3	3		3	3	
39(L)	15	M	L	4	5		4	5		5	5		5	5		5	5	
Median	4																	
IQR	3-5.5																	
Range	1-15																	

*Blank = not recorded. †Stages: 1st stage: median 3 months (IQR: 3 to 4); 2nd stage: median 6 (IQR: 6 to 7); 3rd stage: median 37 (IQR: 28 to 54). ‡After surgical reconstruction. §Biceps was completely paralyzed, but the elbow could be flexed by forearm muscles. #After surgical reconstruction by another surgeon, this extremity was excluded from our analysis of outcomes of surgical reconstruction.

The second neurological evaluation was performed at a median of 6 months (IQR, 6 to 7 months) after paralysis. The shoulder abductors and elbow flexors and extensors were the

most commonly affected muscles, and the finger flexors were less frequently affected (Table II). The trapezius muscle was paralyzed in 32% of affected extremities, and the pectoralis



Paralysis/ Palsy Presentation Paralysis area Accompanied palsy Surgery

Fig. 1
Flowchart for surgical reconstruction. SAN = spinal accessory nerve, UN = ulnar nerve, SAN-SSN = spinal accessory nerve to suprascapular nerve, CC7 = contralateral C7 nerve root, PUN-MCN = partial ulnar nerve to musculocutaneous nerve, ICN-MCN = intercostal nerves to musculocutaneous nerve, FMT = free muscle transfer, SFMT = single free muscle transfer, DFMT = double free muscle transfer, MMT = multiple (non-free) muscle transfer. *The boundary between early and late presentation after the onset of paralysis was not strictly determined; rather, it was defined as an approximate period based on the possible recovery time after nerve transfer surgery.

major muscle was paralyzed in 72%. Paralysis combinations were defined on the basis of the presence of an M0 or M1 grade in the distinct anatomical regions at the time of AFM paralysis onset. A combination including the shoulder and elbow was the most frequent presentation, in 27 extremities, followed by a combination including the shoulder and neck, in 19 extremities. Shoulder involvement was present in 45 extremities (Tables I and II).

The muscle power of each paralyzed region was evaluated utilizing the MRC grade (Table III) at 3 specified time points to assess the degree of spontaneous recovery and to guide the decision regarding the timing of surgical reconstruction.

Surgical Reconstruction (Fig. 1)

Surgery was indicated in patients who had (1) a persistent motor deficit demonstrated by an inability to abduct the shoulder, flex the elbow, and flex the finger (MRC M0) at a minimum of 6 months after the onset of symptoms; (2) an absence of respiratory insufficiency, as that would have been a contraindication at our hospital because of equipment and staff restrictions postoperatively in the pediatric intensive care unit; and (3) a parent desire for surgical reconstruction.

Two patients with 1 M0 extremity each did not want to undergo surgery, and 2 patients (4 extremities) with respiratory insufficiency were not accepted for surgery. Fourteen

TABLE IV Type of Surgical Reconstruction

Serial Case No.	Side	Type of Palsy*	Reconstructive Procedures†		
			Shoulder	Elbow	Wrist and Fingers
1	L	S-E-H	CC7-SSN + LTN	DFMT	DFMT
2	R	S-E-H	SAN-SSN	SFMT	SFMT
3	R	S-E	SAN-SSN	PUN-MCN	
4	L	N-S-E	CC7-SAN + SSN, ICN-AxN	PUN-MCN	
5	R	N-S	CC7-SAN + SSN, TDN-AxN		
6	L	S-H	SAN-SSN		AIN-UN
7	L	S-E	SAN-SSN, MMT	PUN-MCN	
8	L	S	MMT		
11	L	S-E	SAN-SSN	ICN-MCN	
12	R	N-S-E	ICN-AxN	PUN-MCN	
14	L	S-H-E		ICN-MCN	
15	R	S	PUN-AxN		
16	R	N-S	MPN-SSN, PUN-AxN		
17	R	S-E-H	SAN-SSN	ICN-MCN	
17	L	S-E-H	SAN-SSN	ICN-MCN	
18	L	S-E-H	SAN-SSN	ICN-MCN	
22	R	S	SAN-SSN, PUN-AxN		
23	R	N-S-E	CC7-SAN + SSN	PUN-MCN	
25	R	S-E-H	SAN-SSN	ICN-MCN	
28	R			Steindler	
30	L	N-S-E-H		FMT	
31	R	S	SAN-SSN, PUN-AxN		
32	L	S-E	SAN-SSN	PUN-MCN	
34	R	S-H	SAN-SSN		
34	L	S-E-H	SAN-SSN	ICN-MCN	
35	L	N-S-E		PUN-MCN	
36	R	S-E-H	CC7-C5	SFMT	
38	L	S-E-H	SAN-SSN, ICN-AxN		
39	R	N-S-E		SFMT	

*S = shoulder, E = elbow, H = hand, N = neck, CC7 = contralateral C7 nerve root, LTN = long thoracic nerve, SSN = suprascapular nerve, DFMT = double free muscle transfer, SFMT = single muscle transfer, SAN = spinal accessory nerve, PUN = partial ulnar nerve, MPN = median pectoral nerve, MCN = musculocutaneous nerve, ICN = intercostal nerve, AxN = axillary nerve, TDN = thoracodorsal nerve, AIN = anterior interosseous nerve, UN = ulnar nerve, MMT = multiple muscle transfer, MTN = medial thoracic nerve, C5 = cervical 5th nerve root.

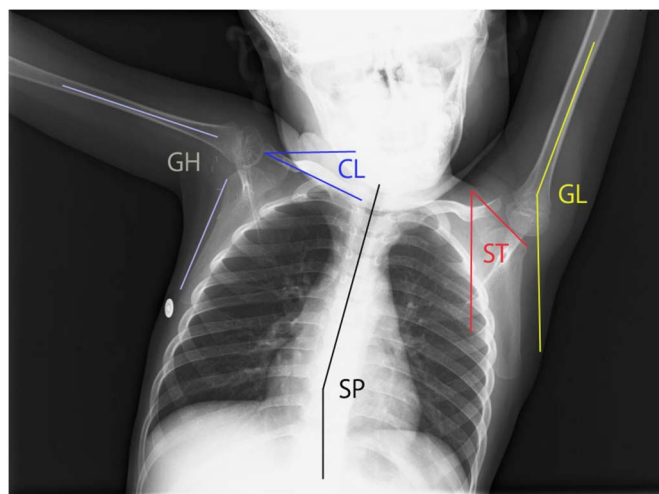


Fig. 2
Patients were examined in a standing position, and anteroposterior radiographs of both shoulder joints in the frontal plane were made in a neutral position and then in maximal active abduction. The arm was held outstretched and internally rotated to eliminate the action of the biceps or other co-working muscles. Global abduction (GL), glenohumeral (GH), scapulothoracic (ST), and clavicular (CL) angles and lateral flexion of the thoracic spine (SP) were measured. GL is the angle between the longitudinal line of the humerus and a vertical line. GH is the angle between the line from the inferior border of the glenoid to the inferior angle of the scapula and the longitudinal humeral axis. ST is the angle between the longitudinal axis of the glenoid and a vertical line. CL is the angle between the line from the midpoint of the sternoclavicular joint to the midpoint of the acromioclavicular joint and a horizontal line. The SP angle measures the lateral flexion of the thoracic spine relative to a vertical line. The difference between the neutral position and abduction was calculated for each of these angles and denoted as Δ . These differences were considered to represent measurements for ranges of motion as follows: Δ GL for global abduction, Δ GH for GH abduction, Δ ST for ST abduction, Δ CL for clavicular abduction, and Δ SP for abduction by lateral flexion of the thoracic spine.

extremities in 12 patients that had recovered to M2 or greater at the sixth month after palsy onset did not undergo surgery and instead continued to undergo rehabilitation.

Twenty-seven patients with 29 extremities underwent surgical reconstruction (Table IV).

Restoration of Shoulder Joint Function

Nerve transfer surgery¹³ was preferred in cases with early presentation (at <15 months). The spinal accessory nerve (SAN) was the preferred donor nerve for suprascapular nerve (SSN) palsy. However, the contralateral C7 nerve root (CC7)¹³ was used if the SAN was unavailable (due to paralysis or its identification as a potential donor for free muscle transfer [FMT]) and was routed subcutaneously through the anterior aspect of the neck to the SSN and SAN. In 6 extremities, the axillary nerve (AxN) was reconstructed utilizing intercostal nerves (ICNs), partial fascicles of the ulnar nerve (PUN), or the thoracodorsal nerve.

In patients with late presentation (at >24 months), muscle-tendon transfers¹³ consisting of the trapezius, biceps, and latissimus dorsi donor muscles were performed.

Restoration of Elbow, Wrist, and Finger Function

The method for restoration of elbow flexion was selected on the basis of the elapsed time since paralysis onset and the presentation type. Seven extremities underwent transfer of the PUN to the musculocutaneous nerve (MCN), 7 extremities underwent transfer of an ICN to the MCN, and 5 extremities underwent an FMT (4 single and 1 double FMTs). A Steindler forearm muscle transfer¹³ was used to reconstruct 1 extremity.

Five extremities underwent FMT to restore elbow flexion (3 extremities) or prehensile finger function (2 extremities)¹³⁻¹⁵.

Secondary Procedures

Four patients required secondary procedures to correct the remaining disabilities. These included Zancolli capsulodesis of the metacarpophalangeal joints for claw-finger correction¹⁵, tendon transfer to permit thumb opposition, tenodesis to permit wrist extension, biceps tendon rerouting to permit forearm pronation, and transfer of the long head of the triceps to the acromion to permit shoulder abduction.

Early Postoperative Assessment and Rehabilitation

Electromyography was used to confirm the reinnervation of muscles following nerve transfer and FMT. It was performed first at 3 months postoperatively and then at regular intervals until recovery of the motor unit potentials was detected. After documentation of electrophysiological evidence of reinnervation, electromyographic feedback was commenced for training of independent shoulder, elbow, and finger function in the patients who could visit the hospital frequently.

TABLE V Spontaneous Recovery of Shoulder Abduction*

No. of Extremities	Time After Onset in Months		
	3	6	37
7	M0	M0	M0
1	M0	M0	NR
21	M0	M0	Surg.
1	M0	M1	M 4
1	M1	M1	Surg.
1	NR	M1	M3
4	NR	M0	M0
2	NR	M0	Surg.
9	M2-3	M2-3	M2-4
3	NR	M4	M4-5
Total, 50			

*NR = no records, Surg. = underwent surgery, M = Medical Research Council (MRC) grade.

Midterm Postoperative Evaluation

The third and final assessment of this study was performed at a median of 37 months (IQR, 28 to 54 months) after AFM paralysis onset to determine the midterm natural recovery and surgical outcomes after upper-extremity AFM.

Upper-extremity function was assessed on the basis of active range of motion in terms of shoulder abduction and external rotation, elbow flexion and extension, and total finger motion.

Dynamic shoulder radiography (DSR) was used to investigate the individual component angles of glenohumeral joint and scapulothoracic joint motion in shoulder abduction^{16,17} (Fig. 2).

The strength of elbow flexion was measured qualitatively with the MRC grading scale and quantitatively with a handheld dynamometer (Micro FET 2; Hogan Scientific).

The last 9 of the 15 patients who underwent SAN-SSN transfer had additional examination of the compound muscle action potential (CMAP) of the trapezius muscle, assessed by intraoperative stimulation of the SAN under general anesthesia prior to surgical reconstruction. The first 6 of the 15 patients did not have CMAP examination because the presence of partial injury of the SAN was not yet suspected.

Laxity tests such as the drawer test assessed the presence of recurrent anterior or posterior glenohumeral insta-

bility. A clunk or snap on subluxation or reduction suggested instability¹⁸.

The Mallet score assessed shoulder abduction and external rotation deficits as a functional score. The Child Health Questionnaire-Parent Form 28 (CHQ-PF28) was used as a patient-reported outcome measure to assess overall improvement in quality of life.

Statistical Analysis

All data are presented as the mean or median with the range or IQR, depending on the normality of the data, and statistical analyses used the Student t test for comparisons of 2 groups and the Kruskal-Wallis test for comparisons of >2 groups. The chi-square independence test was used for comparison of the data in contingency tables. The correlation coefficients between the CHQ-PF28 and functional measurements were calculated using the Pearson test. Correlation coefficient values of >0.5 were classified as strong. The power of the test was calculated post hoc whenever necessary, and the desired power was set at 80% according to conventional recommendations. Tests with <80% power did not undergo further statistical analyses, and the data are merely presented as the mean or median and range or IQR. The level of significance was set at $p < 0.05$.

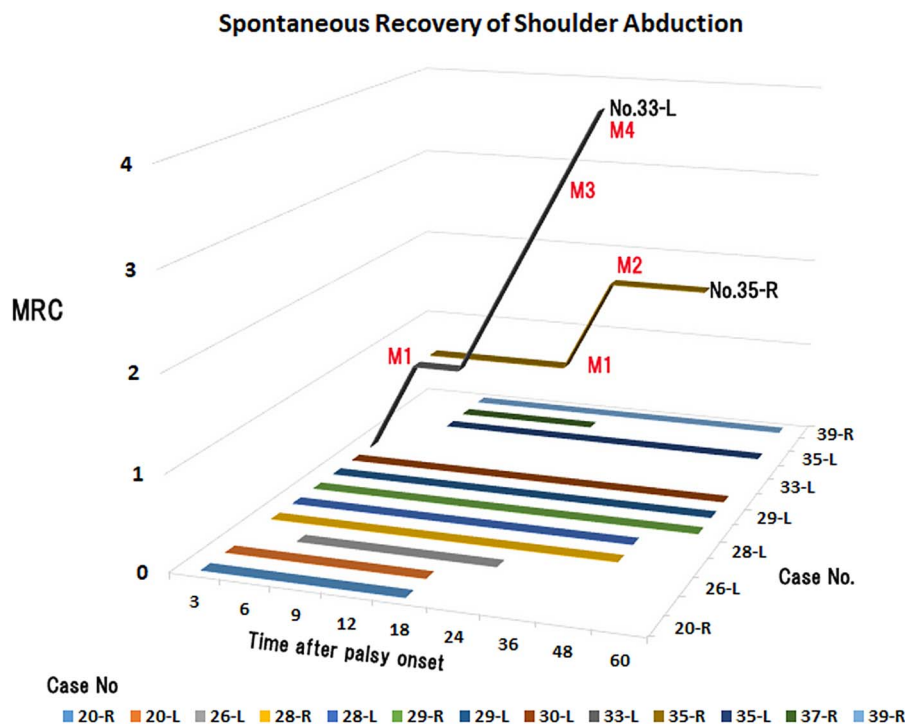


Fig. 3

Clinical course of spontaneous recovery of shoulder abduction. Of the 50 extremities of 39 patients, we investigated the spontaneous recovery of initially M0 or M1 shoulder abduction in 13 extremities of 9 patients at 6 months after onset (excluding 24 extremities that underwent surgical reconstruction, 1 extremity that did not undergo re-examination, and 12 extremities that had M2 or greater shoulder abduction at both the first and second assessments). For example, 20-R indicates the right upper extremity of patient 20. The vertical axis shows the MRC grade, and the horizontal axis shows the time after palsy onset in months. A horizontal bar indicates that the muscle strength was at a plateau.

TABLE VI Long-Term Functional Outcomes*

Serial Case No.	Follow-up (mo)	Elbow											
		Shoulder				Flexion							Hand
		ROM (deg)				Strength by HHD measurement			Extension		TAM		
		Abd.	Flex.	ER	IR	Deg.	MRC	Involved (Nm)	Uninvolved (Nm)	Ratio	Deg.	MRC	Deg.
1	133	90	80	70	80	145	4	7	28	25%	-30	0	140
2	130	30	30	20	70	145	4	3	13	23%	-40	2	160
3	78	180	180	90	90	150	4	7	17	41%	0	4	270
4	41	100	100	80	90	150	4	9	21	43%	0	3	270
5	77	180	180	90	90	150	4	5	11	45%	0	4	270
6	74	115	120	-10	90	140	4	3	12	25%	0	2	150
7	70	20	20	90	90	100	3	1	9	11%	0	3	270
8	60	90	90	50	90	140	3	5	10	50%	0	3	270
11	39	20	20	90	90	150	4	NR	NR	NR	-40	0	270
12	37	90	60	-60	90	150	4	3	9	33%	-25	0	270
14	NR	NR	NR	NR	NR	NR	NR	NR	NR	NR	NR	NR	NR
15	35	180	180	90	80	150	4	5	13	38%	0	4	270
16	NR	NR	NR	NR	NR	NR	NR	NR	NR	NR	NR	NR	NR
17	38	90	40	90	90	150	4	2	2	100%	0	2	0
17	37	20	20	30	90	140	4	2	2	100%	0	2	0
18	24	60	60	40	60	150	4	4	10	40%	0	0	270
22	33	120	180	30	80	150	4	3	6	50%	0	2	270
23	28	80	30	80	90	150	4	6	19	32%	0	2	270
25	38	90	80	70	80	150	4	NR	NR	NR	-30	4	200
28	24	10	10	10	10	140	3	NR	NR	NR	-30	0	240
30	24	20	20	0	0	90	3	NR	NR	NR	0	0	250
31	47	180	180	90	80	135	4	3	13	23%	0	4	240
32	37	35	25	65	75	150	4	4	10	40%	0	0	270
34	37	90	180	90	80	150	4	1	0	DNT	0	NR	200
34	37	20	0	0	0	150	4	2	0	DNT	-20	NR	180
35	31	45	45	30	90	100	3	NR	NR	NR	0	0	270
36	37	90	90	-50	80	120	3	1	7	14%	-45	0	0
38	28	45	40	60	60	100	3	1	12	8%	0	3	270
39	24	10	0	0	0	130	4	2	38	5%	-30	3	270
Median	37	80	60	60	80	150	4	3	11	33%	0	2	270
Range	24 to 133	10 to 180	0 to 180	-60 to 90	0 to 90	90 to 150	3 to 4	1 to 9	0 to 38	5 to 50%	-45 to 0	0 to 4	0 to 270

*ROM = range of motion, TAM = total active motion, HHD = quantitative measurement by handheld dynamometer, Abd. = abduction, Flex. = flexion, ER = external rotation, IR = internal rotation, MRC = Medical Research Council grade, Ratio = (involved/uninvolved) × 100%, NR = not recorded, DNT = did not test because of bilateral palsy.

Results

Spontaneous Recovery

Table III shows the course of muscle strength recovery according to the MRC scale, recorded at the 3 specified evaluation time points (median of 3, 6, and 37 months after onset) by assessing shoulder abduction, elbow flexion and extension, and finger flexion and extension. Analysis of spontaneous recovery of shoulder abduction function (Table V) focused on patients with an M0 or M1 grade. After excluding the 24 shoulders that underwent surgical reconstruction after the second evaluation, none of the 11

extremities with M0 shoulder abduction at the second evaluation obtained M1 or better recovery by the third evaluation (Fig. 3). Two extremities with M1 shoulder abduction at the second evaluation recovered to M2 and to M4 by the third. Twelve extremities with M2, M3, or M4 shoulder abduction at the first or second evaluation either remained unchanged or improved by 1 to 2 ranks at the third. The same trend was observed in the recovery of elbow flexion and extension and in hand flexion and extension function; 6 months after the onset of paralysis, M0 function did not recover to a level higher than M2 (Table III).



Fig. 4-A



Fig. 4-B

Fig. 4-A Case 3. A 4-year-old boy sustained right shoulder and elbow palsy with complete paralysis of the right deltoid, supraspinatus, and biceps muscles. He underwent right SAN-SSN transfer and PUN-MCN transfer. The CMAP amplitude of the right trapezius by electric stimulation of the SAN was 7.53 mV.

Fig. 4-B Photograph taken 6 years after the operation, demonstrating the patient's ability to abduct the right shoulder fully.

Postoperative Recovery (Table VI)

The median follow-up duration was 37 months (range, 24 to 133 months). The median postoperative shoulder active motion

was 80° (range, 10° to 180°) in shoulder abduction and 60° (range, -60° to 90°) in shoulder external rotation. The postoperative median shoulder abduction after 15 SAN-SSN and 5 CC7-SSN



Fig. 5-A



Fig. 5-B

Fig. 5-A Case 32. A 3-year-old girl sustained right shoulder and elbow palsy with complete paralysis of the right deltoid, supraspinatus, and biceps muscles. She underwent right SAN-SSN transfer and PUN-MCN transfer. The CMAP amplitude of the right trapezius by electric stimulation of the SAN was 2.69 mV.

Fig. 5-B Photographs taken 3 years after the operation, demonstrating the patient's inability to abduct the right shoulder.

TABLE VII Comparative Analysis of Shoulder Abduction Recovery Following SAN-SSN Transfer*

Case No.	Type of Paralysis†	Pectoralis Major (MRC)	SAN-SSN	SAN CMAP (mV)	AxN Repair	Follow-up (mo)	Shoulder Abduction (deg)	GH Instability
3	S-E	M4	Yes	7.53		78	180	No
31	S	M0	Yes	4.32	With PUN	47	180	No
22	S	M3	Yes	7.08	With PUN	33	120	No
6	S-H	M0	Yes	7.73		74	115	No
25	S-E-H	M0	Yes			38	90	No
34(R)	S-H	M3	Yes	5.01		37	90	Yes
17(R)	S-E-H	M0	Yes			38	90	No
18	S-E-H	M0	Yes	3.92		24	60	Yes
38	S-E-H	M0	Yes		With ICN	39	45	Yes
32	S-E	M0	Yes	2.69		37	35	Yes
2	S-E-H	M0	Yes			130	30	Yes
7	S-E	M0	Yes			70	20	Yes
11	S-E	M0	Yes	2.23		39	20	No
17(L)	S-E-H	M0	Yes			37	20	Yes
34(L)	S-E-H	M0	Yes	4.07		37	20	Yes

*Shoulder abduction of >60° was considered good. SAN = spinal accessory nerve, SSN = suprascapular nerve, MRC = Medical Research Council grade, CMAP = compound muscle action potential, AxN = axillary nerve, GH = glenohumeral, PUN = partial ulnar nerve, ICN = intercostal nerve. †S = shoulder, E = elbow, H = hand.

transfers was 60° (range, 20° to 180°) and 90° (range, 80° to 180°), respectively.

The 15 cases with SAN-SSN transfer were divided into 2 outcome groups (good and poor) on the basis of their postoperative shoulder abduction relative to the median angle of 60° (Figs. 4-A through 5-B; Video 1) and factors affecting shoulder abduction recovery were assessed (Table VII). The null hypothesis was that none of the factors would be associated with good versus poor shoulder abduction in the 2 groups. There were found to be significant differences in glenohumeral instability ($p = 0.005$, chi-square independence test) and preoperative SAN-CMAP amplitude ($p = 0.009$, t test) between the 2 groups (Table VIII). Tests of the remaining 2 factors, elbow flexion and pectoralis major paralysis, did not satisfy the power requirement.

TABLE VIII Comparison of Factors Affecting Shoulder Abduction Recovery*

Factor		Good	Poor	Test	P Value	Power
Elbow flexion paralysis	(-)	4	0	χ^2	0.013	0.7
	(+)	3	8			
Pectoralis major paralysis	(-)	3	0	χ^2	0.038	0.5
	(+)	4	8			
GH instability	(-)	6	1	χ^2	0.005	0.8
	(+)	1	7			
SAN CMAP (mV)	Mean	6.3	3.2	T test	0.009	1.0
	SD	1.6	0.9			

*GH = glenohumeral joint, SAN = spinal accessory nerve, CMAP = compound muscle action potential, SD = standard deviation.

DSR was used to measure the component angles of shoulder abduction in 15 extremities. The null hypothesis was that the 3 component motions of shoulder abduction would have the same magnitude. The mean postoperative ranges of motion for the 3 shoulder component angles were 18° (range, -38° to 106°) for glenohumeral, 48° (range, 20° to 71°) for scapulothoracic, and 5° (range, -6° to 25°) for abduction by lateral flexion of the thoracic spine. The magnitudes of the 3 component motions differed significantly (Kruskal-Wallis test, $p < 0.001$, power = 1.0). Only 5 shoulders showed a positive range of motion in the glenohumeral joint (Fig. 6).

CC7 transfer was found to provide satisfactory recovery of shoulder abduction, presumably because the CC7 transfers reinnervated not only the SSN to a comparable extent to the SAN or long thoracic nerve but also the SAN (Video 2). There were no postoperative complications of CC7 transfer, such as transient muscle weakness of elbow and shoulder extension or sensory impairment.

Of the 20 extremities that underwent SSN repair, 5 had additional AxN reinnervation by nerve transfer. The mean postoperative shoulder abduction angle was 109° (range, 45° to 180°) in the AxN reinnervation group and 68° (range, 20° to 180°) in the non-AxN reinnervation group (Tables IV and VI).

Elbow Function

The postoperative median elbow flexion was 150° (range, 100° to 150°) following 7 PUN transfers, 150° (range, 140° to 150°) following 6 ICN transfers, and 130° (range, 90° to 145°) following 5 FMTs (Table VI). The null hypothesis was that elbow flexion recovered similarly following the 3 procedures. However, a

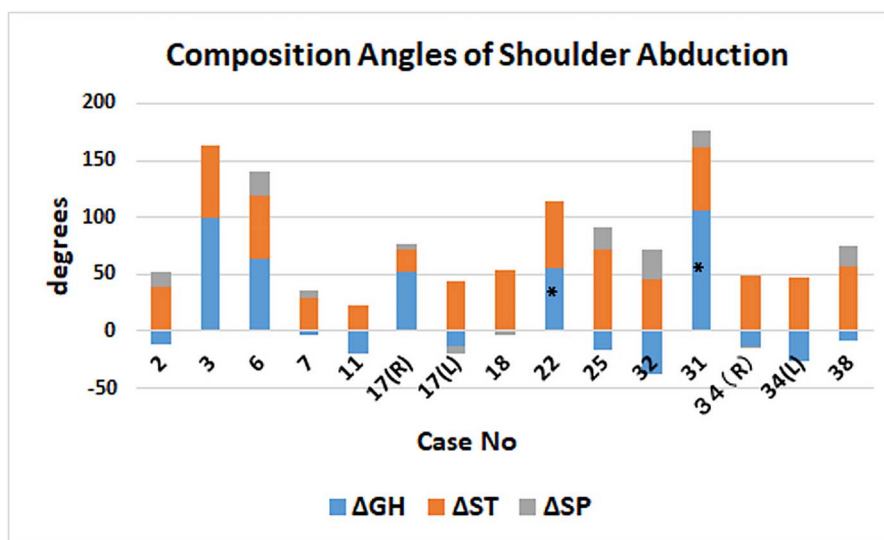


Fig.6
Component angles of shoulder abduction following SAN-SSN transfer, as measured on DSR (Δ GH for glenohumeral [GH] abduction, Δ ST for scapulothoracic [ST] abduction, and Δ SP for abduction by lateral flexion of the thoracic spine. The shoulder abduction angle need not equal the sum of Δ GH, Δ ST, and Δ SP because the starting adduction angle on DSR was not always zero. *A PUN-AxN transfer technique was used in addition to SAN-SSN transfer in this extremity.

significant difference was found among the 3 procedures ($p = 0.040$, power = 1.000).

Hand Function

Two patients with global upper-extremity paralysis underwent FMT to restore hand function, and both obtained satisfactory total active motion of the fingers (Table VI).

Quality of Life

The mean increase in the Mallet score was 8 (range, 0 to 17), from 8 (range, 5 to 18) to 16 (range, 5 to 25) (Kruskal-Wallis test, $p < 0.0001$, power = 1.0) (Table IX).

In terms of quality-of-life improvement for the children, the mean summary scores for both the physical and psychosocial domains of the CHQ-PF28 increased from 19 (range, -3 to 47) to 35 (range, 6 to 54) and 40 (range, 17 to 62) to 49 (range, 34 to 62), respectively. The null hypotheses were that the pre- and postoperative physical and psychosocial scores would be similar. However, both scores were found to have significant postoperative increases (Student t test, $p < 0.0001$ for both, power = 1.0 and 0.8) were obtained (Table IX).

Follow-up duration and elbow flexion were significantly correlated with the parental impact and family activities domains of the CHQ-PF28 (see Appendix Supplemental Table 3).

Discussion

Spontaneous Recovery

The prognosis of a paralyzed muscle may depend on the involvement of associated muscles innervated by the same spinal segments, as in poliomyelitis¹⁹. However, to our knowledge, there is no evidence of the occurrence of such an association with spinal segments in AFM.

According to some studies, reconstructive surgery is indicated within 6 months of onset^{11,20}, but to our knowledge no studies have investigated the maximum duration that spontaneous recovery might take. In this study, after excluding the 24 shoulders that had undergone surgical reconstruction before the 12-month evaluation, none of the patients with M0 shoulder abduction at the 6-month evaluation recovered M1 or better shoulder function in the subsequent follow-up. Only 2 shoulders with M1 shoulder abduction recovered further, to M2 and M4. Similar observations were seen in the recovery of elbow function and hand and finger function during subsequent follow-up. Therefore, patients with persistent M0 shoulder, elbow, and/or hand paralysis at 6 months after onset should be considered for surgical reconstruction.

Shoulder Function

The outcomes of shoulder joint reconstruction in AFM differ greatly from those in brachial plexus palsy^{9,11}. SAN-SSN transfer is considered the most reliable surgical method for restoration of shoulder abduction in patients with brachial plexus palsies²¹. In our patients with AFM, however, half of the patients treated with SAN-SSN transfer did not regain the expected shoulder abduction. Hence, we evaluated the integrity of the SAN preoperatively by the MRC scale and electromyography, and intraoperatively by trapezius contraction on electrical stimulation. From the analysis of these results in our study, we propose that trapezius muscle paralysis should be assessed with preoperative CMAP, as the outcomes after SAN-SSN transfer showed a significant difference depending on whether the CMAP was above or below a cutoff amplitude of 4.1 mV. When the CMAP is lower than that cutoff, SAN-SSN transfer is not recommended, and other nerve transfers to the SSN or AxN should be considered instead.

TABLE IX Preoperative and Postoperative Quality-of-Life Scores*

Serial Case No.	CHQ-PF28								
	Mallet Score			Physical			Psychosocial		
	Preop.	Postop.	Diff.	Preop.	Postop.	Diff.	Preop.	Postop.	Diff.
1	5	19	14	19	54	35	62	57	-5
2	5	15	10	25	42	17	60	59	-1
3	18	25	7	8	51	43	34	49	15
4	10	16	6	19	23	4	29	49	20
5	11	25	14	15	48	33	25	37	12
6	15	18	3	34	46	12	46	61	15
7	12	16	4	24	36	12	32	59	27
8	8	16	8	26	39	13	50	55	5
11	5	13	8	-3	34	37	50	48	-2
12	5	11	6	7	32	25	17	45	28
14	5	NR	NR	47	NR	NR	48	NR	NR
15	17	21	4	13	35	22	35	55	20
16	5	NR	NR	30	NR	NR	35	NR	NR
17	5	17	12	10	27	17	35	40	5
17	5	9	4	10	27	17	35	40	5
18	5	16	11	NR	NR	NR	NR	NR	NR
22	12	24	12	26	50	24	45	38	-7
23	5	12	7	18	46	28	24	34	10
25	5	19	14	28	31	3	48	49	1
28	5	5	0	-1	12	13	29	50	21
30	5	7	2	29	38		51	46	-5
31	14	21	7	11	9	-2	55	50	-5
32	5	17	12	46	40	-6	47	62	15
34	7	24	17	15	30	15	31	36	5
34	5	13	8	NR	NR	NR	NR	NR	NR
35	5	12	7	15	40	25	30	35	5
36	5	10	5	8	6	-2	43	56	13
38	5	15	10	24	39	15	41	55	14
39	5	8	3	NR	NR	NR	NR	NR	NR
Mean	8	16	8	19	35	16	40	49	9
Range	5-18	5-25	0-17	-3-47	6-54	-6-43	17-62	34-62	-6-28

*CHQ-PF28 = Child Health Questionnaire-Parent Form 28, Diff. = difference, NR = not recorded.

Pectoralis major paralysis, which implies damage to the cells in the medial anterior horn of the extensive C5-T1 spinal segment, was also observed in 72% of the AFM surgical cases in our study, the same as in poliomyelitis¹⁹. The steering group muscles²² of the glenohumeral joint, such as the subscapularis, supraspinatus, and infraspinatus muscles adjacent to the pectoralis major, were also likely injured. Paralysis of these muscles produced instability of the glenohumeral joint and further deterioration of shoulder abduction function.

Although CC7 transfer following posttraumatic brachial plexus injury has not appeared to justify the risk of donor-site morbidity involving permanent motor and sensory loss²², we


recommend CC7 transfer as a safe surgical alternative in AFM to provide additional donor motor nerves with comparable surgical outcomes, especially for pediatric patients with SAN paralysis²³ unless diplegia is present (Video 2). CC7 transfer may also have provided considerable recovery of SAN paralysis, which DSR would be able to demonstrate.

The results of DSR analysis after surgery were surprising. In patients who underwent SAN-SSN transfer, 5 extremities with good shoulder abduction recovery had substantial recovery of glenohumeral joint abduction, whereas shoulder abduction occurred mainly at the scapulothoracic joint in the remaining cases (Fig. 5). The scapulothoracic joint abduction was mainly achieved by the trapezius and serratus anterior muscles, whose

individual contribution could be assessed by further analysis of the DSR, especially for CC7-SAN transfers.

In conclusion, AFM damages the ventral horn cells of the spinal cord, similar to poliomyelitis, but the pathomechanism needs to be better understood to guide surgical reconstruction, especially through nerve transfer. When selecting a donor nerve for nerve transfer, the quality of the transferred nerve and the extent of combined paralysis should be considered. A thorough evaluation might reveal that a nerve from a distant source would be preferable as a donor for transfer—for example, use of CC7 for SSN reinnervation if nearby nerves such as the SAN are unreliable.

Appendix

 Supporting material provided by the authors is posted with the online version of this article as a data supplement at [jbjs.org \(http://links.lww.com/JBJSOA/A632\)](http://links.lww.com/JBJSOA/A632). ■

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Kazuteru Doi, MD, PhD¹
Yasunori Hattori, MD, PhD¹
Sotetsu Sakamoto, MD¹
Dawn Sinn Yii Chia, MBBS²
Vijayendrasingh Gour, MCh(Plastic)¹
Jun Sasaki, MD¹

¹Department of Orthopedic Surgery, Ogori Daiichi General Hospital, Yamaguchi, Japan

²Sengkang General Hospital Singapore, Singapore

Email for corresponding author: doimicro@saikyo.or.jp

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