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ORIGINAL ARTICLE
Hand/Peripheral Nerve

Mid-Term Outcome after Selective Neurotization of the Infraspinatus Muscle in Patients with Brachial Plexus Birth Injury

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Background: Active shoulder external rotation in adduction can be restored by selective neurotization of the infraspinatus muscle with the spinal accessory nerve in select patients with brachial plexus birth injury. Does the improved shoulder external rotation stand the test of time?

Methods: Fourteen consecutive brachial plexus birth injury patients with active shoulder external rotation in adduction of ≤ 0 degrees and active shoulder elevation ≥ 90 degrees underwent selective neurotization of the infraspinatus muscle at mean 2 years of age between 2012 and 2016. All 14 patients had congruent shoulders joints with passive external rotation in adduction of 30 degrees. Pre-and post-operative electromyography was done to seven patients. Shoulder function and the subjective outcome was assessed after a mean follow-up of 3.8 years.

Results: Shoulder external rotation in adduction improved by a mean 57 degrees in the 12 children who did not develop shoulder internal rotation contracture. Shoulder external rotation in abduction and shoulder abduction increased in all 14 patients. Reinnervation of the supraspinatus muscle was evident in all seven children who underwent postoperative EMG. Thirteen patients' parents were satisfied with the outcome.

Conclusions: Functionally significant shoulder external rotation can be restored and maintained by reinnervation of the infraspinatus muscle in brachial plexus birth injury patients with congruent shoulder joints, if internal rotation contracture does not develop. *(Plast Reconstr Surg Glob Open 2020;8:e2605; doi: 10.1097/GOX.0000000000002605; Published online 24 January 2020.)*

INTRODUCTION

Brachial plexus birth injury (BPBI) advances from cranial to caudal roots in normal vaginal deliveries. Shoulder function, especially external rotation (ER), is therefore nearly always compromised in permanent BPBI. At least one-third of all patients with permanent BPBI develop posterior subluxation of the shoulder joint during the first year of life.[1–4]

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Received for publication July 4, 2019; accepted November 4, 2019.

Presented at American Society for Surgery of the Hand Boston 2018, and Narakas 2019, Leiden, The Netherlands.

DOI: 10.1097/GOX.0000000000002605

Absent ER in a congruent shoulder joint is initially treated by passive ER exercises. Internal rotation contracture of the shoulder has been treated with Botulinum toxin-A injections (BTX), splinting, and different soft tissue releases.[5–7] Without recovery of the infraspinatus muscle, normal shoulder development can be compromised.[8] Restoration of active shoulder ER has traditionally been achieved with tendon transfers and more recently by either neurotizing the suprascapular nerve (SSN) or only its infraspinatus branch with the spinal accessory nerve (SAN).[9–12]

We have previously shown that selective neurotization of the infraspinatus muscle does reliably restore active ER in BPBI patients with congruent shoulder joints and passive ER in adduction of more than 45 degrees.[12] The purpose of this study is to report midterm results of this new technique to assess if gain in active ER is permanent.

Disclosure: *The authors have no financial interest to declare in relation to the content of this article.*

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PATIENTS AND METHODS

The study consists of 14 BPBI patients (7 male, 9 left, 1 bilateral) identified between 2012 and 2016. The 3-month Toronto Test Score was mean 4.8 (range, 3.8–5.8) excluding a patient who underwent early brachial plexus reconstruction (patient 13), and three patients referred from elsewhere (Table 1). Indication for neurotization was a congruent shoulder joint with ≤ 0 degrees of active ER in adduction, ≥ 45 degrees of passive ER in adduction and active shoulder elevation of ≥ 90 degrees against gravity. Preoperative electromyography (EMG) performed in seven children showed insufficient muscle activation to produce active shoulder ER. Due to good surgical outcome routine, preoperative EMG was discontinued. Eight of the patients had winging of the scapula preoperatively.

Before the neurotization (mean 18 months; range, 2–50), 11/14 of the patients had received BTX injections to the shoulder internal rotators due to subluxation of the shoulder joint. In addition to BTX, one patient (patient 5) also had a shoulder relocation 42 months before the procedure. At the time of the selective neurotization (mean age, 2 years; range, 1.4–4.7), all 14 shoulders were congruent. However, in two patients, passive shoulder ER in adduction had decreased to 30 degrees during the waiting time to the procedure (patients 2 and 11). In addition to the neurotization, these two patients received 100 international units of BTX to their shoulder internal rotators. The surgical technique has been described earlier[12] (Video 1). (See **Video 1**, [online], which displays surgical technique and early results of the neurotization.)

Follow-up was scheduled at 3, 6, 12, and 36 months after the neurotization. Both active and passive shoulder ranges of motion were calculated using a goniometer by two independent observers (physiotherapist and occupational therapist). Parents' satisfaction regarding the functional and cosmetic (scar, scapular winging) outcome was assessed (satisfied versus not satisfied).

EMG was repeated 3–5 years from the neurotization to evaluate the infraspinatus activity with a concentric needle electrode after parents' consent was obtained. Possible spontaneous activity (fibrillations, positive sharp waves as well as discharges) was recorded. Innervation of the infraspinatus muscle was determined by asking the patients to externally rotate the upper arm: the activation pattern and morphology of the motor unit potential (MUP) was recorded. Quantitative multi-MUP analysis was performed after the examination, and the collected MUPs were compared to established normal values[13] and graded (Table 2). As the SAN branch to the upper trapezius is preserved during the procedure, function of the upper part of the trapezius muscle was also evaluated with needle EMG in 6 of the patients (one patient refused).

Statistical analysis regarding age and outcome was done using Spearman's rank correlation analysis.

RESULTS

Mean improvement of active shoulder ER in adduction was at 57 degrees (range, 40–95) and 56 degrees (range, 30–85) in abduction at mean follow-up of 3.8 years (range, 1.6–5.4) excluding patients 2 and 11 who developed shoulder internal rotation contractures. (Table 1) (Fig. 1) (Video 2) (**See Video 2**, [online], which displays midterm result of patient 3. Six years from surgery.)

Active shoulder abduction improved mean 27 degrees (range, 10–60). One patient (patient 11) developed posterior shoulder dislocation due to internal rotation contracture, which was treated by shoulder relocation 2.9 years after the neurotization.

Hypertrophic scars developed in seven patients, all but one subsided with silicone scar treatment. Scapular winging was observed in 11 patients (3 new cases) in the immediate period after the neurotization. At the last follow-up, scapular winging was evident only in five patients, none of whom complained of any pain or functional problems related to their scapular position.

Table 1. Preoperative Findings in Relation to Results

	Preoperative Findings			Results				
Patient	3-Month Test Score	Active ER in Adduction (degrees)	Active ER in Abduction (degrees)	Age at Surgery (years)	Follow-up From Surgery (years)	Active ER in Adduction (degrees)	Active ER in Abduction (degrees)	Subjective Outcome
1	NA*	−20	0	1.6	5.4	45	80	Satisfied
2	5.1*	−20	15	4.1	5.3	−20	50	Not satisfied
3	5.1*	−15	45	3.6	5.2	80	90	Satisfied
4	5.1*	0	30	1.6	4.9	50	90	Satisfied
5	4.5*	0	20	4.7	5	65	80	Satisfied
6	3.8*	0	20	4.3	4.6	45	70	Satisfied
7	NA	0	0	1.6	3.5	45	80	Satisfied
8	4.2	0	10	1.5	3.1	50	70	Satisfied
9	5.2	−20	45	1.4	3	45	75	Satisfied
10	3.8	−20	45	1.9	3.2	40	75	Satisfied
11	5.2	0	60	1.4	3.1	−35	60	Satisfied
12	5.8*	−10	20	4.5	2.7	30	80	Satisfied
13	NA	−20	−10	2.1	2.1	30	75	Satisfied
14	NA	0	45	2.3	1.6	55	75	Satisfied
Sample mean	4.8 ± 0.4	−8.9 ± 5.1	24.6 ± 11			48 ± 7.8	75 ± 5.5	
SD	0.7	9.6	21			13.9	10.6	

Margin of error expressed using 95% confidence level.
*EMG pre- and postoperatively.
ER, external rotation; NA, not available.

Table 2. Spinal Accessory Nerve Connection to the Infraspinatus Muscle as Well as to the Upper Trapezius Muscle Evaluated with Needle Electromyography at Mean 5 Years Postoperative

		Infraspinatus MUP Analysis					
	Patient	Time From Surgery (years)	Interference Pattern	MUP Size	MUP Polyphasia	MUP Duration	Conclusion
MA	1	4.9	Mildly reduced	Normal	Normal	Normal	Very mild abnormality
JJ	2	5.3	Moderately reduced	+++	Normal	+++	Moderate abnormality
ET	3	4.9	Mildly reduced	+++	++	Normal	Mild abnormality
JS	4	5.2	Moderately reduced	++	+	+++	Moderate abnormality
TP	5	5.2	Moderately reduced	+	Normal	NA	Moderate abnormality
OM	6	3.0	NA	Normal	++	++	Mild abnormality
AS	12	4.6	Moderately reduced	+	Normal	+	Moderate abnormality

Moderate abnormality is defined as reduced recruitment of MUPs, abnormally enlarged, polyphasic, and long-duration MUPs. Mild abnormality is defined as mildly reduced recruitment of MUPs, abnormally enlarged, polyphasic, and long-duration MUPs. Very mild abnormality is defined as mildly reduced recruitment of MUPs, no abnormalities in MUP morphology.
+ Slightly increased; ++ moderately increased; +++ severely increased.
NA, not available.

All but one patient's parents were satisfied with the functional and cosmetic outcome of the neurotization. The dissatisfied patient's parents felt that their child did not benefit from the procedure, and saw the scar as a cosmetic problem (patient 2). No correlation between age of surgery and outcome was found (Fig. 2).

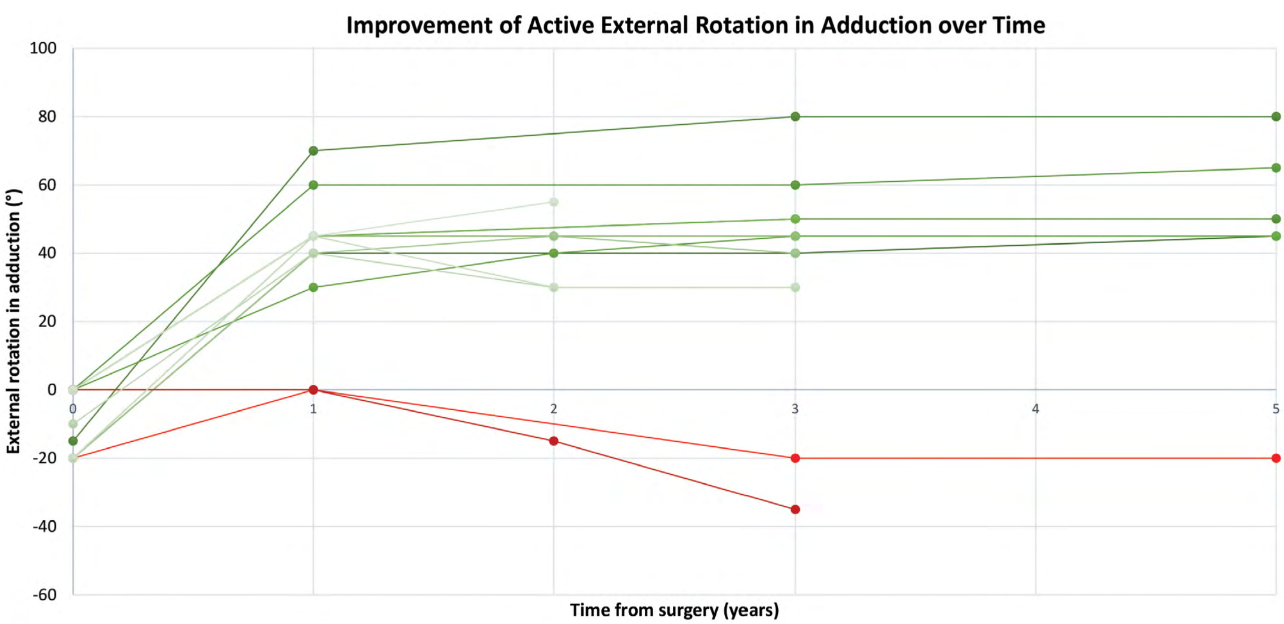

Fig. 1. Active shoulder external rotation improved in 12 patients (green lines). Two patients developed shoulder internal rotation contracture with decreased active shoulder external rotation (red lines).

Postoperative EMG showed reinnervation of the infraspinatus muscle in all examined patients (Table 2), with mild to moderate signs of old recovered nerve injury. Upper trapezius function was normal in all six patients assessed by EMG.

DISCUSSION

ER of a congruent shoulder in BPBI patients with internal rotation contracture can be improved, at least in short term, with releasing or lengthening tight anterior structures and bracing. However, long-term active ER is not improved without adequate infraspinatus function.[14] Traditionally, lack of adequate infraspinatus function has been treated with latissimus dorsi or teres major tendon transfers[10] and, more recently, by lower trapezius transfer.[9] In long-term follow-up, however, the gained benefit from tendon transfers seems to subside.[15] This is in contrast to our results with neurotizing infraspinatus muscle with SAN, which seem to yield a permanent improvement of shoulder ER in patients who do not develop a progressive internal rotation contracture.

Restoration of active shoulder motion as early as possible is important for normal shoulder joint development.[16–18] Neurotization procedures are generally performed earlier than the above-mentioned tendon transfers are, which should be advantageous. Neurotization of the

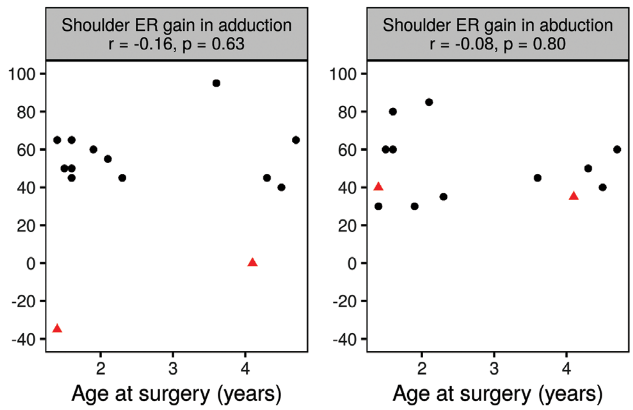

Fig. 2. Patient age at surgery plotted against the gain (•) of shoulder external rotation (ER) in adduction and in abduction at last follow-up. The two patients who did not get improved shoulder ER in adduction are marked in red. Spearman's rank correlation test results are shown below the title.

whole SSN, either by use of the SAN[12,19] or grafting from C5, has been shown to yield variable results. Pondaag et al. compared these two procedures and found no difference between their outcomes: Active shoulder ER in adduction exceeded 20 degrees only in one-fifth of the 86 operated patients.[20] A similar study by O'Grady showed better results at 2-year follow-up with the SAN pro-SSN transfer compared with grafting from C5.[21] Our results support these findings of O'Grady, since shoulder ER could be reliably restored and maintained with reinnervating the infraspinatus muscle by SAN in a subset of children with permanent BPBI.[21]

The likelihood of spontaneous recovery of infraspinatus function in BPBI patients older than 1.5 years is very low.[22] Thus, this serves as a good lower age for the neurotization. The upper age limit is not known, but the oldest patient in our series benefitted from the procedure at 4.7 years of age. A preoperative assessment of the infraspinatus with EMG or MRI, especially in older patients, might be worthwhile, although we found no correlation between age at surgery and outcome, and have ourselves discontinued the use of preoperative EMG.

The downside of the presented technique is that permanent winging of the scapula and a cosmetically displeasing hypertrophic scar might develop after the surgery. In addition to this, lower trapezius tendon transfer cannot be performed to improve shoulder ER if the neurotization was to fail. However, scapular winging does not seem to impair shoulder function and has a tendency to subside over time. These possible adverse effects should nevertheless be discussed with the parents before deciding to proceed with the neurotization.

CONCLUSIONS

It seems obvious that a prerequisite for improved active shoulder ER in adduction after neurotizing the infraspinatus muscle by SAN is maintaining good passive shoulder ER. Both of our patients with no gain in active shoulder ER in adduction after the procedure had only 30 degrees of passive shoulder ER on the day of surgery. However, with free passive (> 45 degrees) shoulder ER, functionally significant active shoulder ER can be restored and maintained in BPBI patients by direct neurotization of the infraspinatus muscle by SAN.

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