HAND/PERIPHERAL NERVE

Spinal Accessory Nerve Transfer Outperforms Cervical Root Grafting for Suprascapular Nerve Reconstruction in Neonatal Brachial Plexus Palsy

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Background: The authors evaluated long-term shoulder function in patients with neonatal brachial plexus palsy undergoing suprascapular nerve reconstruction with cervical root grafting or spinal accessory nerve transfer.

Methods: A retrospective review was performed on all infants presenting with neonatal brachial plexus palsy between 1994 and 2010. Functional outcomes were compared by type of suprascapular nerve reconstruction.

Results: Seventy-four patients met the inclusion criteria (46 transfers, 28 grafts). Both groups presented with an active movement scale score of 2.0 for shoulder abduction and 0.0 for external rotation. Postoperative follow-up was 9.0 years for the graft group and 6.7 years for the transfer group. Both groups achieved an active movement scale score of 5.0 for shoulder abduction at 12, 24, and 36 months postoperatively. Active movement scale scores for shoulder external rotation were 1.0, 2.0, and 2.5 for the graft group versus 2.0, 2.0, and 3.0 for the transfer group at 12, 24, and 36 months postoperatively. None of these differences reached statistical significance. Composite Mallet scores were 13.0 for the graft group versus 15.0 for the transfer group at 3 years (p = 0.06) and 13.0 for the graft group versus 16.0 for the transfer group at 5 years postoperatively (p = 0.07). Secondary shoulder surgery was performed on 57.1 percent (16 of 28) of patients with grafts compared with 26.1 percent (12 of 46) of patients with transfers (OR, 3.17; p = 0.02).

Conclusion: Suprascapular nerve reconstruction by cervical root grafting results in poorer shoulder function and a two-fold increase in secondary shoulder surgery compared with spinal accessory nerve transfer. (*Plast. Reconstr. Surg.* 135: 1431, 2015.)

CLINICAL QUESTION/LEVEL OF EVIDENCE: Therapeutic, III.

Restoration of shoulder stability and function and maintenance of normal muscle balance to allow concentric development of the shoulder are a high priority in the treatment approach to infants with neonatal brachial plexus palsy. The suprascapular nerve is important for shoulder abduction and external rotation because it innervates the supraspinatus and infraspinatus muscles, respectively. It most commonly originates at a variable distance from the junction of the C5 and C6 nerve roots.¹ The suprascapular nerve is commonly injured in neonatal brachial plexus palsy and vulnerable to traction injury because it

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is relatively fixed at both ends. The proximal C5 nerve root has soft-tissue attachments to the C5 transverse process, and the distal suprascapular nerve is relatively fixed as it passes through the suprascapular notch.^{2,3}

Given its importance in shoulder function and development, the suprascapular nerve is a priority in reconstruction. Treatment approaches range from neurolysis to reconstruction by nerve grafting from available nerve roots (i.e., C5) or distal nerve transfers (i.e., spinal accessory nerve). The benefits of the former include a donor supply of up to 10,000 myelinated axons and the opportunity for anatomic restoration. Advantages of the latter include an uninjured donor source of 1500 to 1700 purely motor axons and one rather than

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two coaptations closer to the target muscle, thus offering the potential for faster recovery.⁴

The question becomes whether cervical root grafting or nerve transfer provides better shoulder function. The purpose of this study was to evaluate the long-term shoulder outcomes of patients with neonatal brachial plexus palsy undergoing suprascapular nerve reconstruction with either technique.

PATIENTS AND METHODS

A human research ethics committee– approved, retrospective review was performed on all infants treated for neonatal brachial plexus palsy at a single institution between 1994 and 2010. Inclusion criteria consisted of infants who underwent suprascapular nerve reconstruction and with preoperative and at least 36 months of postoperative shoulder outcome data. Exclusion criteria consisted of patients with incomplete operative notes or follow-up data.

Primary brachial plexus reconstructions were performed by one of three plastic surgeons. Patients underwent nerve graft reconstruction of the suprascapular nerve where, after neuroma resection, the proximal nerve roots available were deemed adequate to reconstruct the distal plexus, including the suprascapular nerve. The decision to use either the C5 or C6 proximal nerve root as the donor was made based on the quality and availability of the root. Where possible, grafts were topographically oriented such that they coursed from the rostral (12-o'clock) position of the C5 cross-sectional area to the suprascapular nerve. This was in agreement with the anatomical findings of Siqueira and colleagues.⁵ Reconstruction was performed with usually one or sometimes two sural nerve grafts. The number of grafts used was dependent on the caliber of the cervical root donor and, more important, that of the suprascapular nerve target.

Alternatively, suprascapular nerve reconstruction was performed with spinal accessory nerve transfer where there was only limited proximal cervical nerve root cross-sectional area available. The nerve transfer was performed using an anterior approach, where the distal spinal accessory nerve was identified deep to the anterior border of trapezius at the time of supraclavicular plexus exploration and divided as distally as possible to maximize length. This avoided the use of interpositional nerve grafts and preserved the proximal input to the trapezius muscle. From 2010 onward, spinal accessory nerve transfer was performed via the posterior approach using a transverse incision just above the spine of the scapula and exposure by splitting the trapezius muscle in the line of its fibers to expose the descending branch.⁶ Whether by cervical root grafting or spinal accessory nerve transfer, the whole of the caliber of the suprascapular nerve was targeted.

Outcomes of shoulder function were compared between the two groups. For those patients who underwent secondary shoulder surgery, functional outcomes up to the date of surgery were included but not thereafter. Patients were not excluded from the analysis if they underwent shoulder intervention in the form of botulinum toxin A (Botox; Allergan, Inc., Irvine, Calif.) and/ or casting. Shoulder abduction and external rotation were evaluated using the active movement scale⁷ preoperatively as well as 12 months, 24 months, and 36 months postoperatively. At 3 years and 5 years postoperatively, shoulder function was assessed using the composite Mallet score.⁸ For this assessment, five global movements of the shoulder were graded from 1 to 5, for a maximum total score of 25. Composite Mallet scores were compared both with and without exclusion of patients who underwent secondary shoulder surgery.

The requirement for secondary shoulder intervention was also evaluated. These interventions were either administration of Botox, with or without postoperative casting, or surgery in the form of shoulder contracture release and/or tendon transfers. No other procedures, other than those stated addressing shoulder function (abduction and external rotation), were performed on either group of patients. Indications for Botox use included passive range of motion of external rotation less than 30 degrees. Botox treatment of the shoulder contracture usually involved treatment of the subscapularis, latissimus dorsi, teres major, and pectoralis major muscles, with dosage indicated by body weight. The number of treatments with Botox was recorded for each patient in each group. At the time of treatment, the glenohumeral joint was manipulated into maximal passive external rotation, and in the latter part of the series, it was placed in an upper limb spica to maintain external rotation. The incidence of secondary shoulder surgery, involving internal rotation contracture release, glenohumeral joint reduction, and/or external rotation tendon transfer, was also recorded for each group. Indications for shoulder surgery included glenohumeral subluxation, persistent loss of passive range of motion of external rotation despite Botox treatments, and/or the lack of active external rotation beyond

neutral in the setting of preserved distal hand function and adequate donor muscles (i.e., latissimus dorsi). Although there was a defined framework for surgical intervention, ultimately, the decision rested with the parents after careful weighing of the riskbenefit ratio. To account for the possible effects of underlying pathology in the analysis of secondary shoulder surgery, patients in each reconstruction group were then stratified using the Narakas classification as type I/II or III/IV.⁹ In patients with type I/ II injuries, rates of secondary shoulder surgery were then compared for those undergoing nerve grafts versus nerve transfers. A similar form of analysis was performed in patients with type III/IV injuries.

Continuous and ordinal data did not show normal distributions on Shapiro-Wilkes test; therefore, statistical analysis was performed with nonparametric tests. Data were presented as medians, with interquartile ranges for the 25th to 75th percentiles, and comparisons were performed with the Mann-Whitney U test. Contingency data were presented as percentages and analyzed using the 2×2 Fisher exact test. Kaplan-Meier analysis of time to secondary shoulder surgery was performed to account for differential lengths of follow-up. Statistical software was used for analysis (SPSS for Mac, version 19.0; SPSS, Inc., Chicago, Ill.). A p value < 0.05 was considered significant.

RESULTS

Seventy-four patients met the inclusion criteria (46 spinal accessory nerve transfers, 28 cervical root grafts). Comparison of patient demographics can be found in Table 1. Median gestational age and birthweight were equivalent across the two groups. The nerve transfer group had a higher distribution of patients with total brachial plexopathy compared with the nerve graft group; however, this was not statistically significant. Conversely, three quarters of all patients with total palsies had nerve transfers. Median surgical age was equivalent between the nerve graft and transfer groups (9.9 months versus 8.0 months; p = 0.32).

Table 1. Patient Demographics

	Grafts	Transfers	þ
Patients, no.			0.13
Total	28	46	
Upper trunk palsy	22 (78.6%)	28 (60.9%)	
Total palsy	6(21.4%)	18 (39.1%)	
Gestational age, wk	40.0	40.0	0.26
Birth weight, g	4131	4058	0.78
Surgical age, mo	9.9	8.0	0.32
Follow-up period, yr	9.0	6.7	0.07

The median follow-up period was longer for nerve grafts versus transfers, although this difference only reached borderline significance (9.0 years versus 6.7 years; p = 0.07).

Nerve graft and transfer groups initially presented with equivalent active movement scale scores for shoulder abduction and external rotation (Table 2). Postoperative active movement scale scores for shoulder abduction can be found in Table 3. By 12 months postoperatively, both groups had achieved a median score of 5.0 for shoulder abduction. Scores remained stable for both groups at 24 months and 36 months postoperatively. Comparison of scores across the two groups for shoulder abduction at each time point did not reach statistical significance. Table 4 details the postoperative comparison of active movement scale scores for shoulder external rotation. At 12 months postoperatively, the median score for shoulder external rotation was 1.0 for nerve grafts versus 2.0 for transfers. Both groups demonstrated improvement over time in active movement scale scores for shoulder external rotation. Comparison of scores across the two groups for shoulder external rotation at each time point did not yield statistical significance.

Composite Mallet scores were assessed across the two groups, first excluding patients who underwent secondary shoulder surgery (Table 5). At

Table 2. Preoperative Shoulder Function per MedianActive Movement Scale Score*

	Grafts	Transfers	þ
Shoulder abduction Shoulder external	2.0 (0.0-3.0)	2.0 (0.0-3.0)	0.53
rotation	0.0 (0.0-1.0)	0.0 (0.0-0.5)	0.97

*Data are presented as median (interquartile range).

Table 3. Postoperative Shoulder Abduction per Median Active Movement Scale Score*

	Grafts	Transfers	þ
12 Months 24 Months	5.0 (3.0–5.0) 5.0 (3.0–6.0)	5.0 (3.0-5.0) 5.0 (4.0-6.0)	$0.60 \\ 0.68$
36 Months	5.0 (3.5-6.0)	5.0 (5.0-6.0)	0.64

*Data are presented as median (interquartile range).

 Table 4. Postoperative Shoulder External Rotation

 per Median Active Movement Scale Score*

	Grafts	Transfers	þ
12 Months 24 Months 36 Months	$\begin{array}{c} 1.0 \ (0.0 - 3.0) \\ 2.0 \ (1.0 - 4.0) \\ 2.5 \ (2.0 - 5.0) \end{array}$	$\begin{array}{c} 2.0 & (0.0 - 2.3) \\ 2.0 & (1.5 - 4.0) \\ 3.0 & (2.0 - 4.0) \end{array}$	$0.79 \\ 0.99 \\ 0.82$

*Data are presented as median (interquartile range).

Table 5.	Postoperative Composite	Mallet Scores,
Excludin	g Patients with Secondary	y Shoulder Surgery*

	Grafts	Transfers	þ
3 Years†	13.0 (12.0-15.0)	15.0 (13.0-17.0)	0.062
5 Years‡	13.0 (11.5–15.5)	16.0 (13.5–17.0)	0.074
	13.0 (11.5–15.5)	· · · · ·	0

*Data are presented as median (interquartile range). †Six patients in the graft group and five patients in the transfer group were excluded from 3-year analysis.

Fourteen patients in the graft group and 10 patients in the transfer group were excluded from 5-year analysis.

3-year analysis, six patients in the graft group and five patients in the transfer group were excluded. At 5-year analysis, 14 patients in the graft group and 10 patients in the transfer group were excluded. On comparison, the nerve transfer group had a higher median composite Mallet score compared with the nerve graft group at 3 years postoperatively (15.0 versus 13.0; p = 0.062). At 5 years postoperatively, the median composite Mallet score remained higher in the nerve transfer group in relation to the nerve graft group, although this difference again reached borderline statistical significance (16.0 versus and 13.0; p = 0.07).

The composite Mallet scores were then reassessed after including those patients who underwent secondary shoulder surgery in the analysis (Table 6). At 3 years postoperatively, the nerve graft group had an equivalent median composite Mallet score compared with the nerve transfer group (13.5 versus 15.0; p = 0.20). Similarly, at 5 years postoperatively, the nerve graft group had an equivalent median composite Mallet score compared with the nerve transfer group (15.0 versus 16.0; p = 0.44).

Outcomes of secondary shoulder intervention were compared next, as shown in Table 7. The distribution of Botox use was equivalent across the two groups. The frequency of secondary shoulder surgery was then assessed. Patients in the nerve graft group demonstrated a significantly higher frequency of secondary shoulder surgery compared with those in the nerve transfer group (57.1 percent versus 26.1 percent; OR, 3.17; p = 0.02). Rates of secondary shoulder surgery were then reassessed after stratifying patients by presenting plexopathy. For Narakas type I/II injuries,

Table 6. Postoperative Composite Mallet Scores, Including Patients with Secondary Shoulder Surgery*

	Grafts	Transfers	þ
3 Years	13.5 (12.3–15.0)	15.0 (13.0–17.0)	0.20
5 Years	15.0 (13.0–16.0)	16.0 (13.0–17.0)	0.44

*Data are presented as median (interquartile range).

	Grafts (<i>n</i> = 28)	Transfers $(n = 46)$	þ
Botox One treatment Two treatments Three treatments Shoulder surgery	$\begin{array}{c} 9 \; (32.1\%) \\ 3 \; (10.7\%) \\ 1 \; (3.6\%) \\ 16 \; (57.1\%) \end{array}$	$\begin{array}{c} 17 \ (37.0\%) \\ 6 \ (13.0\%) \\ 2 \ (4.3\%) \\ 12 \ (26.1\%) \end{array}$	$0.80 \\ 1.0 \\ 1.0 \\ 0.02$

patients in the nerve graft group continued to demonstrate a significantly higher frequency of secondary shoulder surgery compared with those in the nerve transfer group (76.2 percent versus 31.0 percent; OR, 7.1; p = 0.004). In type III/IV injuries, the frequency of secondary shoulder surgery was equivalent in the nerve graft group compared with the nerve transfer group (0 percent versus 17.6 percent; p = 0.53). Kaplan-Meier time-to-event analysis further highlighted the discrepancy in secondary shoulder surgery across the entire nerve graft and nerve transfer cohorts (Fig. 1). By approximately 4 years, the trajectory of the two groups markedly diverged, with a diminishing percentage of graft patients being surgery-free compared with the transfer cohort (p = 0.007).

DISCUSSION

Historically, upper trunk neonatal brachial plexus injuries have commonly been reconstructed with nerve grafts.¹⁰⁻¹⁵ More recently,

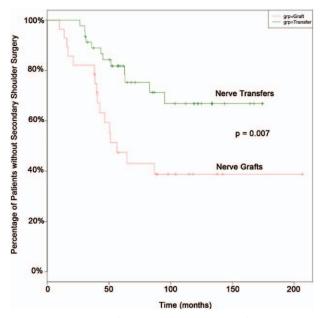


Fig. 1. Time to secondary shoulder surgery with cervical root grafting versus spinal accessory nerve transfers for suprascapular nerve reconstruction.

nerve transfers have been used increasingly in the management of traumatic adult peripheral nerve injuries.^{6,16–18} A recent study by Ladak and colleagues¹⁹ has confirmed that distal nerve transfers are also an option in the management of neonatal brachial plexus injuries. In that report, 10 infants with upper trunk palsies were treated with three distal nerve transfers: spinal accessory nerve to the suprascapular nerve; radial to axillary nerve; and ulnar or median nerve transfer to the musculocutaneous nerve. Active movement scale scores at 24 months were 5.0 ± 0.5 for shoulder abduction, 4.3 ± 0.6 for external rotation, $6.3 \pm$ 0.2 for elbow flexion, and 5.9 ± 0.2 for forearm supination. These scores compared favorably to the functional outcomes reported by Lin and colleagues²⁰ for the conventional technique of brachial plexus reconstruction with nerve grafts. The question then arises, "Should nerve transfers be considered the first-line treatment option?"

This study aimed to provide some answers by focusing on one important aspect of upper trunk neonatal palsy, suprascapular nerve reconstruction, and comparing cervical root grafting to spinal accessory nerve transfer. Strictly focusing on individual scores for shoulder abduction and external rotation, we observed equivalent outcomes from cervical root grafting and spinal accessory nerve transfer groups at 12 to 36 months postoperatively. Our findings were in agreement with those of published studies. In a study comparing 65 patients with C5 nerve grafting with 21 patients with spinal accessory nerve transfers, Pondaag and colleagues²¹ reported similar degrees of external rotation, Mallet hand-mouth score, and Mallet hand-head scores at a mean follow-up of 3 years. Evaluating 28 grafts versus 25 spinal accessory nerve transfers, Terzis and Kostas²² found equivalent shoulder abduction as per British Medical Research Council grading (3.9 ± 0.7 versus 3.7 ± 0.6) at an average follow-up of 5.6 years. However, interpretation of these findings was confounded by concomitant reconstruction of the axillary nerve in 47 patients by either repair of the posterior cord or direct neurotization. Finally, Tse and colleagues²³ reported on 106 patients with cervical root grafting and 71 patients who underwent spinal accessory nerve transfers and found no difference in external rotation (active movement scale scores 2.2 versus 3.0, respectively) at 3 years postoperatively.

In our study, evaluation of global shoulder movements using composite Mallet scores showed that the spinal accessory nerve transfer group outperformed the cervical root graft group. Such differences were of borderline significance at the 3-year and 5-year evaluations, although this may have stemmed from low study power. Indeed, up to one-half of the patients in the graft group and approximately one-quarter of the patients in the transfer group were excluded from the analyses for undergoing secondary shoulder surgery. Nevertheless, these findings may point to the composite Mallet score being a more sensitive scale for differences in shoulder function, because it captures five complex shoulder movements in contrast to the one-dimensional active movement scale scores for shoulder external rotation or abduction.

In further support of major differences in shoulder function outcomes, the cervical root graft group experienced a two-fold higher frequency of secondary shoulder surgery compared with the spinal accessory transfer group. These findings were in contrast to the report by Tse and colleagues,²³ which highlighted a roughly equal proportion of patients undergoing secondary shoulder surgery before the 3-year follow-up. This discrepancy can be resolved upon taking a closer look at the temporal pattern of secondary shoulder surgery. As shown on Kaplan-Meier time-toevent analysis, the two groups began to markedly diverge at approximately 50 months after primary brachial plexus reconstruction. However, the study by Tse and colleagues captured an earlier snapshot of the two groups during a time when the need for secondary surgery shoulder may not be as readily apparent.

It is also important to point out that the cervical root graft group achieved similar composite Mallet scores if factoring in those patients who underwent secondary shoulder surgery. Because the nerve graft group had a two-fold increase in shoulder surgery compared with the nerve transfer group, it benefited relatively more from the reanalysis. Thus, although the nerve graft group demonstrated poorer shoulder outcomes in relation to the nerve transfer group, these functional differences could be leveled at the cost of a shoulder operation.

One explanation for the poorer shoulder function in patients who underwent cervical root grafting includes the longer time to muscle reinnervation. With axonal regeneration at 1 mm per day,²⁴ an additional 40 to 80 days may be required for functional recovery across sural nerve grafts usually measuring 4 cm each. Prolonged denervation has been associated with reduced capacity for the proximal cervical nerve root and the distal suprascapular nerve stump to support growth of the reconstituting axons and a decline in the

cervical root and/or graft availability. Conversely,

muscular receptivity for the sprouting axons.²⁵ Axonal regeneration is proportionally limited by the slow axonal transport rate of the proximal neuron, which is known to degrade over time.²⁶ Axonal transport declines secondary to a reduction in response to trophic factors at the cell body in an exponential fashion with delay in reinnervation.²⁷ In the distal nerve stump, prolonged denervation has been associated with Schwann cell apoptosis, diminished production of neurotrophic molecules, fragmentation of Schwann cell basal lamina, and collagenization of endoneural tubes.²⁸ These processes compromise the formerly permissive environment for axonal regeneration. In the target muscle, loss of myonuclei and muscle fibers, depletion of satellite cell regeneration, decline in capillary density, and muscular fibrosis reduce muscular receptivity for the sprouting axons.^{28,29}

Interposition sensory nerve grafts also present other unique drawbacks compared with motor nerve transfers, because these conduits do not necessarily provide an optimal environment for nerve regeneration. The slower rate of regeneration has been attributed to diffusion-limited nutrition, architectural issues involved with motor axons crossing the sensory graft, and direct impedance of axonal regeneration by senescent Schwann cells across grafts of increasing length.^{30–33} An interposition nerve graft also involves two separate sites of coaptation, which increases the amount of scar tissue formation and potential impedance to axonal sprouting.^{34,35}

Distal nerve transfers obviate many of these concerns by moving the site of nerve coaptation quite close to the muscular target. This translates into shorter distance and time for the regenerating axons to reach their target, effectively reducing the denervation period and preserving the supportive milieu within the distal nerve stump.¹⁸ Animal models have also demonstrated greater motor neuron regeneration through motor nerve conduits in contrast to sensory nerve grafts.³⁶ Regenerating motor fibers also have an enormous capacity to increase their muscle innervation ratios by approximately five to eight times.^{37,38} Finally, the absence of graft material eliminates the concerns related to impedance of axonal regeneration by senescent Schwann cells.^{32,33}

These findings must be interpreted in light of the study limitations. First, differential criteria were implemented for cervical root grafting versus spinal accessory nerve transfer for suprascapular nerve reconstruction. Spinal accessory nerve transfer was performed in the setting of limited

cervical root grafting was performed in the setting of adequate root and graft availability to satisfy all targets, including the suprascapular nerve. On the one hand, this introduced a potential confounding variable. If anything, one might say that the study design underestimated the functional improvement with nerve transfers, which were performed in the setting of relatively poorer and unfavorable cervical root fascicular architecture. Second, it could be argued that the indications and threshold for secondary shoulder surgery are vague and possibly variable. However, it is likely that in recent years, we have become more enthusiastic and confident about likely outcomes, making shoulder surgery more readily acceptable to families. Patients considered for shoulder surgery had either glenohumeral subluxation or inadequate external rotation active and/or passive range of motion, despite optimal physiotherapy and Botox treatments, as well as a suitable latissimus dorsi for transfer. The impact of shoulder pathology on global upper limb function and cosmesis as well as the attitudes of caregivers and children to any proposed surgery influenced each decision on whether to proceed with any surgery offered. The experience gained with secondary shoulder surgery during the 16 years covered by this report has been mostly positive and will be reported separately. Third, different scales for measuring shoulder outcome were employed by age. Active movement scale scores were recorded preoperatively and during the short term postoperatively; however, composite Mallet scores were reported during the long term. Although a potential disadvantage, the use of multiple grading systems provided additional perspectives on shoulder function. Furthermore, this study may have also experienced low statistical power, possibly explaining the lack of significant differences on particular outcome comparisons, as discussed previously. Finally, the morbidity from spinal accessory nerve transfer currently remains unknown. It may potentially limit use of the lower trapezius muscle for musculotendinous transfer procedures as part of restoration of shoulder stability and function. That being said, Ruchelsman and colleagues³⁹ noted no evidence of trapezius muscle atrophy or weakness on examination at greater than 24 months of follow-up in 25 infants who underwent spinal accessory to suprascapular nerve transfer. As more long-term studies are published on spinal accessory nerve transfers, a clearer understanding of the true donor-site morbidity may be better appreciated. Nevertheless,

this is the first study to comprehensively report on long-term shoulder function after suprascapular nerve reconstruction via cervical root grafting versus spinal accessory nerve transfers in infants with neonatal brachial plexus palsy.

CONCLUSION

Suprascapular nerve reconstruction with cervical root grafting results in poorer long-term functional shoulder outcomes and a two-fold increase in secondary shoulder surgery compared with spinal accessory nerve transfers.

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