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CASE REPORT - PEDIATRICS

Results of nerve reconstructions in treatment of obstetrical brachial plexus injuries

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Abstract
Background The aim of this study was to evaluate the results achieved using various surgical techniques in patients with partial and total obstetrical brachial plexus palsy.
Methods From 2000 to 2013, 33 patients with obstetrical brachial plexus injury underwent surgery. Twenty had follow-up periods greater than 24 months and met the criteria for inclusion in the study. All patients were evaluated using the Active Movement Scale.
Results The outcomes of different nerve reconstructive procedures including nerve transfers, nerve grafting after neuroma resection and end-to-side neurorrhaphy are presented. The overall success rate in upper plexus birth injury was 80 % in shoulder abduction, 50 % in external rotation and 81.8 % in elbow flexion with median follow-ups of 36 months. Success rate in complete paralysis was 87 % in finger and thumb flexion, 87 % in shoulder abduction and 75 % in elbow flexion; the median follow-up was 46 months. Useful reanimation of the hand was obtained in both patients who underwent end-to-side neurotization.
Conclusion Improved function can be obtained in infants with obstetrical brachial plexus injury with early surgical reconstruction.

Keywords End-to-side neurorrhaphy · Nerve surgery · Neurotization · Obstetrical brachial plexus palsy · Root avulsion

Introduction

Obstetrical brachial plexus palsy (OBPP) displays a stable incidence of 0.15–3 per 1,000 live births [10]. Most children show good spontaneous recovery, but a recent literature review showed that a residual deficit remains in 20 % to 30 % of children [26]. Shoulder dystocia, macrosomia and instrument delivery, forceps or vacuum extraction, present the greatest risk for brachial plexus injury. Caesarean section and having a twin or multiple birth mates seem to offer some protection against injury [10, 12]. Obstetrical brachial plexus injury is caused by excessive lateral traction to the infants' head during delivery, although cases of OBPP injury after nontraumatic caesarean sections have also been described [1]. The resulting nerve injury may vary from neurapraxia or axonotmesis to neurotmesis and root avulsion from the spinal cord. Axonotmetic lesions with intact basal lamina tubes allow for axons to grow from the lesion site down into the basal lamina tube to the original end organ. Complete recovery will usually occur over the course of weeks or months. A more severe traction lesion results in a neurotmetic injury with a rupture of the basal lamina tubes; outgrowth of axons becomes blocked and does not end directly in an endoneural tube. The outcome is a neuroma in continuity formation, a mass of scar tissue and outgrowing axons. However, even in the most severe cases, some axons will pass through and reach some tubes distal to the lesion site [21]. However, axons that successfully pass the neuroma in continuity are likely to end up in a different tube from the original. The process is called

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misrouting and may cause the phenomenon of co-contraction [27]. Root avulsion, the most severe type of lesion, is a total disconnect between the spinal cord and peripheral nervous system. When roots are avulsed, the outgrowth of axons, neuroma formation or misrouting cannot take place. Most often, brachial plexus injuries are associated with the upper trunk (roots C5–C6±C7, 73–86 %) or complete plexus injury (15–20 %). Isolated lower brachial plexus injuries (Klumpke's paralysis) involving the roots of C8–Th1 do occur, but are very rare, with a frequency of only 0.16 %, and are suggested to originate from complete plexus palsy with recovery of the upper plexus component [13, 21, 26, 31]. The aim of this study was to evaluate the results achieved using various surgical techniques in patients with partial and total obstetrical brachial plexus palsy.

Materials and methods

The study was approved by the hospital institutional review board, and the necessary informed consent was obtained from the parents of each patient. The senior author, P.H., performed nerve reconstructions on 441 adult patients with brachial plexus injury. Between 2000 and 2013, 125 patients with obstetrical brachial plexus injury were examined at our department, and 33 underwent nerve surgery. The same surgeon carried out all interventions. Twenty of the 33 obstetric patients had follow-up periods greater than 24 months, which was considered to be sufficient for a final analysis; those with shorter follow-up periods were excluded. The patient cohort consisted of 14 males and 6 females, the median birth weight was 3, 885 g (range 2,240–4,880 g), and the right side was involved in 13 cases (65 %). The delivery was aided with forceps or a vacuum extractor in four cases. Horner's syndrome was present in six cases. The patients were divided into two groups for better analysis. Group 1 involved 12 patients with upper brachial plexus birth injuries. In six patients, the limb was typically held in internal rotation and pronation, with the elbow extended. In three other patients, an additional lack of elbow, wrist and finger extension was present. An isolated impairment of shoulder abduction was present in one patient and elbow flexion in two patients because of good spontaneous reinnervation of remaining parts of the plexus. Group 2 included eight patients with complete paralysis; the upper limb was flailing, without any tonus and without hand function. All patients involved in the study were evaluated using the Active Movement Scale (AMS) [7], which grades 15 upper extremity movements from 0 to 7 (Table 1). Electromyographic and imaging studies were performed in all cases. Standard radiological examination revealed five clavicle fractures and two humeral fractures on the side of the injured brachial plexus. Computed tomography myelography was considered to be the technique of choice for diagnostic root avulsions. It was a safe

Table 1 Active movement scale

Tested movements	
Shoulder-flexion	
Shoulder-abduction/adduction	
Shoulder-internal/external rotation	
Elbow-flexion/extension	
Forearm-supination/pronation	
Wrist-flexion/extension	
Finger-flexion/extension	
Thumb-flexion/extension	
Gravity eliminated	
No contraction	0
Contraction, no motion	1
Motion≤0.5 range	2
Motion>0.5 range	3
Full motion	4
Against gravity	
Motion≤0.5 range	5
Motion>0.5 range	6
Full motion	7

although invasive procedure, completed without complications in all patients. Detection of root avulsion was important not only for making the decision for surgery, but it also helped to make our operative strategy more accurate. All patients were hospitalized for 1 week following surgery; the average stay in the intensive care unit was 1.8 days. The extremity was kept in a fixed position in shoulder adduction and elbow flexion for 3 weeks. After the surgical procedure, daily rehabilitation of the denervated muscles using electrostimulation was recommended.

Operative technique

Surgery was performed under general anesthesia without the use of muscle blocking agents. In most cases, a supraclavicular incision proved sufficient for good access to all roots and trunks of the brachial plexus. Once the plexus had been exposed, visual assessment of the location and extent of the neuroma took place; further operation strategies were based on preoperative knowledge of the presence of avulsions and selective electrical stimulation of all involved nerve portions. If a strong muscle response after intraoperative electrical stimulation proximal to the neuroma was present, then only neurolysis was performed. If there was no useful response to electrical stimulation, the neuroma tissue was resected in a stepwise manner in order to minimize the gap between the proximal and distal stumps. The preferred option was direct coaptation between available proximal and distal nerve stumps or the avulsed root. If this was not possible, sural nerves were harvested from both legs and then used as grafts. Removal of these nerves does not produce any severe deficits in the legs. After the nerves have been harvested, there can be a slight loss of sensation in the lateral part of the foot, which tends to diminish progressively with time. When the number of proximal stumps was limited in case of root avulsion, a reconstruction of the entire brachial plexus was performed from a single root stump or from two root stumps (Fig. 1) or using nerve transfers or end-to-side neurorrhaphy (ETSN). Distal target stumps for restoration of the hand were used for the C8, inferior trunk or medial cord. For elbow flexion, we used the C6, superior trunk and lateral cord, and for shoulder movements, we used the C5, superior trunk and posterior cord. In case neurotization from C4 was performed, the root was used after the phrenic nerve branches off to avoid any respiratory compromise. The suprascapular nerve, the crucial nerve in shoulder abduction and external rotation, was targeted from one of the roots or separately with the spinal accessory nerve as donor. We preserved the proximal branches to the trapezius muscle in all cases. Pectoral nerves or transfer of triceps motor branches of the radial nerve were also used for nerve transfers. The ETSN was performed via a perineural suture of the C7 or C8 root onto the side of the C5 or C7 root, respectively, after creation of a perineural window. The precise site on the donor nerve where ETSN was to be performed was chosen by direct bipolar electrical stimulation and registration in the corresponding muscle. We used the site on the surface of the donor nerve from which the maximum amplitude of motor response had been evoked. The type of reconstruction that can actually be performed depends upon the number of available viable proximal spinal nerve stumps for grafting and the availability of donor nerves for transfers.

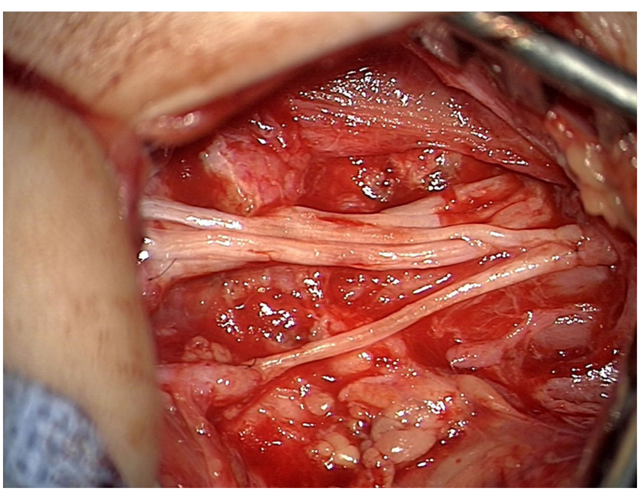

Fig. 1 Neurotization after neuroma resection performed from the C5 and C6 root stumps to the suprascapular nerve and superior, medial and inferior trunk. The C7–Th1 roots are avulsed. Sural nerves were used as grafts. Intraoperative photography

Results

Twenty patients met the criteria for inclusion in the study. The clinical results were assessed before any secondary surgery was performed. In this study, we used Active Movement Scale scores to evaluate obstetric palsy patients, and scores of 6 or 7 were considered successful with regard to demonstrating functionally useful movement; this is a full range of movement if gravity is eliminated and a more than 50 % range against gravity. Surgery was considered unsuccessful when the AMS result was 5 or less. Even though it can be argued that smaller amounts of movement can still be considered useful especially in case of finger movements, our primary objective was nearly complete movement, and this study was to determine the proportion of patients who reached this objective. Computed tomography myelography was performed for diagnostic root avulsions. Thirty roots were found to be avulsed (C5, 6.67 %; C6, 16.67 %; C7, 23.33 %; C8, 30 %; T1, 23.33 %). In most cases the lower roots were affected; avulsion of all five roots was not seen. The summary of different nerve reconstructive procedures and functional outcomes is presented in Tables 2 and 3. In group 1, 12 patients underwent surgery; the median age at the time of surgery was 5.5 months (range 3–15 months). Neuroma resection was followed by root or truncus reconstruction in eight patients. Direct coaptation without a nerve graft was possible in three of the patients. In two patients we transferred triceps motor branches of the radial nerve to the axillary nerve, and in two cases the pectoral nerve was transferred to the musculocutaneous nerve. The suprascapular nerve was targeted from one of the roots or separately with the spinal accessory nerve as donor. The median duration of follow-up was 36 months. During this time we observed that shoulder abduction was successful in 80 %, external rotation in 50 %, elbow flexion 81.8 % and supination 63.6 % of cases. In three cases, the C7 was also injured; in these cases, restitution of elbow extension was successful in 100 % and of the wrist and fingers in 67 %. Preoperative and postoperative AMS scores of selected movements and success rates of operations are summarized in Table 4. In group 2, eight patients underwent surgery. The median age at the time of surgery was 3.5 months (range 2–10 months). Neurolysis alone was performed once, while intraoperative electrical stimulation proximal to the neuroma showed a strong muscle contraction. Reconstruction of the entire brachial plexus from a single root stump took place in two cases and from two root stumps in three cases. ETSN was performed in two patients for the C8 and C7 roots. Nerve grafts were used in five cases. The median total AMS before surgery was 5 (range 3–29). Postoperatively, the total AMS increased to a median of 85.5 (range 51–91), with a median follow-up of 46 months. Shoulder abduction was successful in 87 %, external rotation in 25 %, elbow flexion in 75 % and supination in 25 % of cases. Elbow extension was successful

Table 2 Summary of patients with upper brachial plexus injury treated with different nerve reconstructive procedures with the pre- and postoperative motor scores for individual movements (group 1)

Patient	Root avulsion	Age	Operation	Follow- up	AMS pre-/postoperative					
					Sh abd	Ext rot	Elb flx	Elb ext	Wrist/fing/th	
									Flx	Ext
1	C6	8	C4→C6	62	5	3	0	7	7	7
					7(x)	6(x)	6	x	x	x
2	0	15	PCT→MCN	49	5	2	1	6	6	6
					x	x	7	x	x	x
3	C5	6	C6→ST; C6→SS	56	1	1	1	6	7	6
					7	6	7	x	x	x
4	0	6	C5→C5	48	2	1	0	7	7	7
					6	6	7	x	x	x
5	C7	6	C4→C5; C5→C5,C6,C7	48	1	0	0	0	7	0
					6	4	6	7	x	1/2/2
6	0	6	C5→C5,C6	45	1	1	1	6	7	7
					6	6	7	x	x	x
7	C7	3	C5→ST,SS; C6→ST,MT	35	0	0	0	6	7	0
					7	7	7	7	x	7/7/7
8	C6	4	C5→ST	32	1	0	0	7	7	7
					7	4	6	x	x	x
9	C6	4	R→AX; MCT→MCN; C4→SS	24	1	0	0	7	7	7
					5	5	5	x	x	x
10	C5	5	R→AX; C6+SS neurolysis	25	0	0	1	7	7	7
					6	6	6	x	x	x
11	C7	5	C5→ST; C6→MT	24	0	0	0	1	6	1
					4	5	4	7	x	6/6/6
12	0	5	XI→SS	24	3	1	7	7	7	7
					6	4	x	x	x	x

Abbreviations: sh abd, shoulder abduction; ext rot, external rotation; elb flx, elbow flexion; wri/fing/th, wrist, finger and thumb, PCT, pectoral nerves; MCN, musculocutaneous nerve; SS, suprascapular nerve; ST, superior trunk; MT, medium trunk; IT, inferior trunk; R, triceps motor branches of the radial nerve; AX, axillary nerve; XI, spinal accessory nerve; MC, medial cord; LC, lateral cord; PC, posterior cord; ETSN, end-to-side neurorrhaphy; x, movement was not the objective of the operation

in 87 %, wrist and fingers, thumb flexion in 50 % and 87 %, respectively, wrist in 50 % and finger, thumb extension in 37 % of cases (Table 5). Findings from the analysis of our surgical results demonstrate that useful reanimation of the hand was obtained in both patients who underwent ETSN. The result can be attributed exclusively to the ETSN method, because roots C8 and Th1 were avulsed in both cases. The result of neurolysis on shoulder movement was not as good as had been expected, although the restoration of the elbow and hand was satisfactory. Changes of the total AMS and each movement during the follow-up period were statistically analyzed and modeled in both groups. The increasing trend of the total AMS is displayed in a predictive model, with the predicted AMS means with 95 % confidential intervals (Fig. 2). Prediction of functional restoration of movements, which was targeted with priority, is shown in Fig. 3. The multilevel mixed-effect model (random intercept and random coefficient model) was used; the unstructured variance-covariance matrix was applied to the correlations between examinations with the estimation method being maximum likelihood; calculations were done using Stata 13.1 software.

Discussion

Although the indication and timing of surgery remain controversial among authors [2, 9, 13, 21, 32], we believe that the optimal indication and timing of surgery in cases of total palsy with impaired hand function are within the first 3 months of age. In cases of upper brachial plexus injuries, within the first 4 months is acceptable if there is no useful elbow flexion. In partial lesions with questionable movement, we recommend

Table 3 Summary of patients with complete brachial plexus injury treated with different nerve reconstructive procedures with pre- and postoperative motor scores for individual movements (group 2)

Patient	Root avulsion	Age	Operation	Follow- up	AMS pre-/postoperative					
					Sh abd	Ext rot	Elb flx	Elb ext	Wrist /fing/th	
									Flx	Ext
13	C8–Th1	3	C4→C6; C7→C8 ETSN	55	0	0	0	0	0	0
					6	5	7	7	5/7/7	7/7/7
14	C7–Th1	3	C4→C6; C5→C7,C8 ETSN	104	0	0	0	0	0	0
					6	6	1	7	6/7/7	6/5/2
15	C6–Th1	3	C5→MC,PC; C4→LC	90	1	0	0	0	0	0
					6	4	7	6	7/7/7	2/7/2
16	C7–Th1	5	C5→ST, SS; C6→MT, IT	51	2	0	0	0	0	0
					6	5	7	7	7/7/7	2/7/2
17	C8–Th1	5	C5→C5,C6,SS; C6→C7,C8	50	1	1	0	0	0	0
					6	6	4	7	7/7/7	1/7/1
18	C7–Th1	7	C5→ST, MT, IT	33	1	0	0	0	0	0
					6	2	6	1	1/0/0	5/4/2
19	C8	10	C5 -TH1 neurolysis	28	0	0	0	2	1/3/3	7/0/0
					5	4	6	7	6/6/6	7/6/6
20	C6–Th1	2	C4→SS; C5-ST, MT, IT	26	0	0	0	0	0	0
					6	5	6	7	7/7/7	7/5/7

Abbreviations: sh abd, shoulder abduction; ext rot, external rotation; elb flx, elbow flexion; wri/fing/th, wrist, finger and thumb; PCT, pectoral nerves; MCN, musculocutaneous nerve; SS, suprascapular nerve; ST, superior trunk; MT, middle trunk; IT, inferior trunk; R, triceps motor branches of the radial nerve; AX, axillary nerve; XI, spinal accessory nerve; MC, medial cord; LC, lateral cord; PC, posterior cord; ETSN, end-to-side neurorrhaphy; x, movement was not the objective of the operation

surgery until before 6 months of age. Although we perform electromyography and imaging studies, the final decision relies heavily on the clinical examination. Computed tomography myelography was considered to be the technique of choice for diagnostic root avulsions, with a reported overall diagnostic accuracy of 85 % [5]. This examination was able to detect not only the presence of pseudocysts, but also partial root avulsion in the absence of a rupture of the enveloping meninges. So far, no MRI study has proven to be effective in visualizing the roots emerging from the spinal cord better [28]. The surgical objective in cases of upper OBPIs is functional restoration of shoulder abduction with external rotation and biceps function (elbow flexion and forearm supination).

Table 4 Pre- and postoperative Active Movement Scale scores of selected movements and success rates of operations achieved in patients with upper plexus injury (group 1)

	Group 1				
	Preoperative score		Postoperative score		
Movement	Median	Range	Median	Range	Success
Shoulder abd.	1	0–3	6	4–7	80 %
External rot.	0	0–1	5.5	4–7	50 %
Elbow flx.	0	0–3	6	4–7	82 %
Supination	0	0–1	6	2–7	64 %

Abbreviations: abd, abduction; rot, rotation; flx, flexion; ext, extension

Table 5 Pre- and postoperative Active Movement Scale scores of selected movements and success rates of operations achieved in patients with total brachial plexus palsy (group 2)

	Group 2				
	Preoperative score		Postoperative score		
Movement	Median	Range	Median	Range	Success
Shoulder abd.	0.5	0–2	6	5–6	88 %
External rot.	0	0–1	5	2–6	25 %
Elbow flx.	0	0	6	1–7	75 %
Supination	0	0	5	4–6	25 %
Elbow ext.	0	0–2	7	1–7	88 %
Wrist flx.	0	0–1	6.5	1–7	75 %
Wrist ext.	0	0–1	5.5	1–7	50 %
Finger flx.	0	0–3	7	0–7	88 %
Finger ext.	0	0	5.5	1–7	38 %
Thumb flx.	0	0–3	7	0–7	88 %
Thumb ext.	0	0–0	2	0–7	38 %

Abbreviations: abd, abduction; rot, rotation; flx, flexion; ext, extension

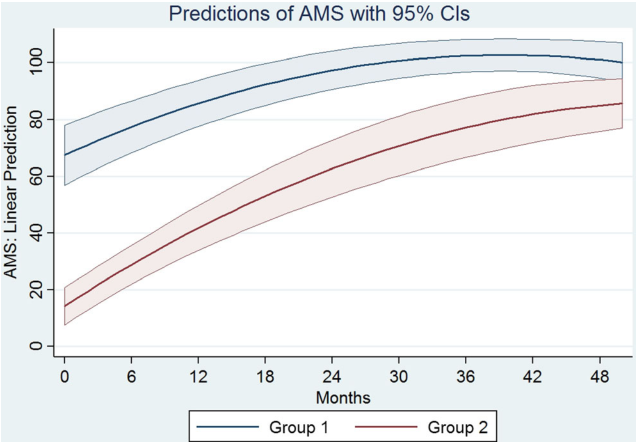

Fig. 2 The increasing trend of total AMS is displayed in a predictive multilevel model; predicted total AMS scores with means with 95 % confidential intervals

The upper trunk generally represents the most vulnerable part of the plexus. If the injury also affects the lower plexus, the upper plexus injury becomes more severe and has a tendency to poorer outcomes for repairs of both C5 and C6 in cases of total palsy [3]. When statistically analyzed, our results for group 1 and 2 seem similar, with the exception of shoulder abduction, but not statistically significantly. The results of global shoulder recovery and elbow flexion in the present study and also in a number of published series are good [3, 9, 13, 21, 32]. In contrast, the results with regard to external rotation were poor; successful results were achieved in 50 % in group 1 and 25 % in group 2. Pondag et al., in a series of 86 patients, found that only 20 % achieved more than 20 degrees of true glenohumeral external rotation [23]. El-Gammal et al. found 34 % and 54 % after tendon transfers [9]. No difference in final external rotation was found between patients who underwent nerve grafting from the proximal stump of C5 and patients who underwent spinal accessory nerve transfer

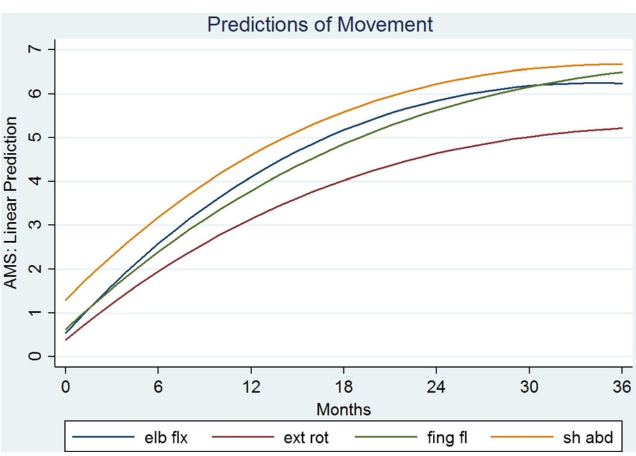

Fig. 3 Prediction from a multilevel model of functional restoration of movements, which was targeted with priority, in the follow-up period. Abbreviations: sh abd, shoulder abduction; ext rot, external rotation; elb flx, elbow flexion; fing flx, finger flexion

[23, 30]. Reanimation of the shoulder and elbow can also be performed with other nerve transfers, especially if the child is presented late. The use of pectoral nerve to musculocutaneous nerve neurotization has been described with good results [4, 34]. The transfer of triceps motor branches of the radial nerve to the axillary nerve appears to be safe and effective. The functional loss relative to the triceps, with a single nerve transferred, is negligible because of compensation by the remaining heads [22]. Nerve transfers permit faster reinnervation of muscles than nerve grafting because the nerve repair can be performed much closer to the neuromuscular junction. Gilbert and Raimondi [14] argue that if roots are available, it is preferable to do a classical reinnervation and reserve distal transfers for later use. In cases of conducting neuroma, Lin et al. documented the superiority of long-term functional results following excision and grafting compared to neurolysis [20]. In three cases upper brachial plexus injury developed in isolated impairment of deltoid or biceps muscle as the remaining parts recovered spontaneously. An explanation is offered by Vredeveld et al., who described the critical role of the C7 root in spontaneous recovery. In a series of 162 infants with OPBI, evidence of extra innervation of the biceps brachii and deltoid muscles through the C7 root was found in the electrophysiological observations [33]. The surgical objective in cases with complete OBPI is recovery of hand function and establishing the ability to use the affected hand in bimanual activities [9, 25, 32]. Bimanual execution of daily activities requires strong finger flexion in combination with good elbow flexion [19]. Reanimation of the hand is essential; otherwise, the maximal function obtained for the affected limb is that of a hook [25]. Recovery of the hand is dependent on the patient's age at the time of surgery—the younger the age, the better the outcome [29]. Our series achieved useful hand reanimation in 87 % of cases. Hand function outcomes in published reports are difficult to interpret, given the variety of evaluation methods and scales used for assessment. After 8 years and several tendon transfers, Gilbert et al. [13] managed to achieve a 76 % success rate in children, but found that recovery was very slow. Birch et al. achieved a 93 % success rate for similar surgeries [3]. In the series presented by Pondaag and Malessy [25], 69 % of hand reanimations were viewed as successful. In their opinion, more emphasis should be placed on reconstructing the median nerve (predominantly controlled by the C8 root) for innervating extrinsic flexor muscles of the fingers and opposition of thumb rather than the ulnar nerve innervating intrinsic muscles (Th1 root). In addition, due to the long outgrowth trajectory, severe atrophy can occur. The use of the end-to-side technique has been abandoned by most surgeons because of the poor results [24]. We present two cases with surprisingly good results, which can be attributed exclusively to the ETSN technique. In both patients, we placed a perineural suture after the creation of a perineural window. We use this method as our standard technique, since studies with

experimental models have indicated that the success rate of such sutures is high [11]. Successful end-to-side neurotization takes place via collateral sprouting of intact axons and also through direct growth of some injured axons into the recipient nerve. Admittedly, the damage to donor nerves may be subclinical and the procedure useful. Without the contribution of the injured axons, the outcome of ETSN would be very poor since only about 1.4 % of motor neurons with intact axons can send out collateral branches [8]. In experimental models, there were no signs of denervation of the donor nerve after creation of a perineural window [11, 16]. Similarly, none of the patients in our series or our recently reported study on adults had any functional deficit of the donor root [16]. This result, although with only two cases presented, may be explained by the superior nerve regeneration capacity in infants compared with adults. Laboratory reports showed better results after ETSN in young rats than in older animals [18]. Although this is only on the level of a case report, the results bear some value. One advantage of ETSN over end-to-end neurotization is that with ETSN there is no need to sacrifice the surrounding nerves. One possible method of achieving better results in the future may be the use of neurotrophic factors to support reinnervation [8, 17]. In cases of late obstetrical brachial plexus palsy, nerve surgery will likely be ineffective, and thus secondary surgery is called for to improve the hand, forearm, elbow and shoulder function and to prevent muscle contractures and bone shortening. The procedure includes a surgical release of muscle contractures, tendon transfers, free-muscle transplantation and osteotomies. Surgery should be carried out after the age of 4 years when the child has ability to cooperate with rehabilitation and severe contractures are absent [15, 6]. Although the present study contains a limited number of patients, our results demonstrate that improved function can be obtained for infants with obstetrical brachial plexus injury with early surgical reconstruction.

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Conflicts of interest None.

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