Child's Nervous System (2019) 35:349–354
https://doi.org/10.1007/s00381-018-04036-5

ORIGINAL ARTICLE

Outcomes from primary surgical reconstruction of neonatal brachial plexus palsy in 104 children

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Received: 5 November 2018 / Accepted: 17 December 2018 / Published online: 4 January 2019

Abstract

Purpose The outcome from microsurgical reconstruction of neonatal brachial plexus palsy (NBPP) varies, and comparison between different series is difficult, given the differences in preoperative evaluation, surgical strategies, and outcome analysis. To evaluate our results, we reviewed a series of children who underwent surgical treatment in a period of 14 years.

Methods We made a retrospective review of 104 cases in which microsurgical repair of the brachial plexus was performed. Strength was graded using the Active Movement Scale. Whenever possible, upper palsies underwent surgery 4 to 6 months after birth and total lesions around 3 months. The lesions were repaired, according to the type of injury: neurolysis, nerve grafting, nerve transfer, or a combination of techniques. The children were followed for at least 24 months.

Results The majority of cases were complete lesions (56/53.8%). Erb's palsy was present in 10 cases (9.6%), and 39 infants (37.5%) presented an extended Erb's palsy. The surgical techniques applied were neurolysis (10.5%), nerve grafts (25.9%), nerve transfers (34.6%), and a combination of grafts and transfers (30.7%). The final outcome was considered poor in 41.3% of the cases, good in 34.3%, and excellent in 24%. A functional result (good plus excellent) was achieved in 58.3% of the cases.

Conclusions There is no consensus regarding strategies for treatment of NBPP. Our surgical outcomes indicated a good general result comparing with the literature. However, our results were lower than the best results reported. Maybe the explanation is our much higher number of total palsy cases (53.8% vs. 25% in the literature).

Keywords Neonatal brachial plexus palsy · Brachial plexus reconstruction · Nerve grafts · Nerve transfers

Introduction

Neonatal brachial plexus palsy (NBPP) is an unpredictable complication of childbirth. Despite full awareness of the problem and advances in obstetrics, the incidence of NBPP remains around 1.5 cases for every 1000 live births, with a range from 0.5 to 5 [8, 14, 19, 27]. These differences in incidence may depend on the type of obstetric care and the average birth weight of neonates in different geographical regions [57]. Half of the patients present one or more perinatal risk factors, which include macrosomia (birth weight over 4 kg), shoulder dystocia, previous deliveries resulting in NBPP, multiparous pregnancies, assisted delivery (vacuum or forceps), and breech presentation [27, 55]. The extent of brachial plexus injury and consequently its prognosis differ greatly. The previous belief that the majority of children affected (up to 92% [34]) have mild injury, and spontaneous recovery has been replaced by a more realistic view in which the recovery rate is now acknowledged to be much lower, such that only 66% of the patients achieve complete recovery, while 10 to 15% present some permanent weakness [27, 45]. Although a large number of these patients recover spontaneously to reach a normal or near-normal degree of functioning, surgical treatment may be necessary when the lesion is more severe, to avoid persistent neurological symptoms and deficiencies in upper limb development.

The first surgical repair for NBPP was recorded early in the twentieth century [30], but the unfavorable outcomes from this and in other early reports made most surgeons skeptical about the long-term results from this type of treatment. It was only in 1981 that Narakas [38] published the first good results from the

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surgical treatment of NBPP, performed under magnification, thus encouraging many surgeons to follow his example.

The purpose of this current report was to review the management of and outcomes from a series of children with NBPP who underwent surgical treatment at the Peripheral Nerve Surgery Unit of the Department of Neurosurgery, University of São Paulo Medical School.

Patients and methods

After obtaining approval from the Research Ethics Committee, we evaluated the records of 507 infants with NBPP who had been seen between July 2003 and June 2017 and we selected 104 cases in which surgical repair was performed. The patients comprised 54 girls and 50 boys. In 70 cases (67.3%), the lesion was in the right upper limb and in 34 (32.6%), in the left.

The patients' strength was graded using the Active Movement Scale [11], and they were categorized into four groups in accordance with the system described by Gilbert and Tassin [17] and refined by Narakas [39, 40]. This provides an overall view of the expected prognosis (Table 1): in group 1, C5 and C6 nerve roots are injured (Erb's palsy), thus compromising shoulder abduction and external rotation, along with elbow flexion; in group 2, besides C5 and C6, the C7 nerve root is also injured (extended Erb's palsy) and wrist drop is added to the weakness/paralysis caused by C5/C6 injury; in group 3, all nerve roots are compromised (C5-T1), and in group 4, besides the C5-T1 injury with complete flaccid paralysis, there is an associated Horner syndrome. The likely rates of good spontaneous recovery outcomes provided through classification are 90% in group 1, 65% in group 2, less than 50% in group 3, and no recovery in group 4 [17].

Among our patients, surgical treatment was indicated when palsies persisted for at least 4 to 6 months in upper lesions and 3 months in total lesions. Neurophysiological studies and MR scans were performed routinely at the beginning of the series.

In our group 1 and 2 cases, the aim of the surgery was restoration of elbow flexion and shoulder function; while in group 3 and 4 cases, the primary objective was restoration of hand function.

The plexus lesions were repaired using different techniques, according to the type of injury: external neurolysis, nerve grafting, nerve transfer, or a combination of techniques.

After surgery, the infants were immobilized, to keep the arm on the operated side flexed at the elbow and on the chest, using a Gilchrist-Velpeau type of bandage [20].

Results

Among the cases selected, 103 infants were born by means of vaginal delivery, and in 29 of them (28.1%), the delivery was assisted (forceps or vacuum). In one case (0.9%), the birth was by means of cesarean section.

One or more risk factors were presented in the majority of these cases. The most common were shoulder dystocia (82 cases/78.8%) and macrosomia (48 cases/46.1%). Diabetes, hypertensive arterial disease, and multiparity were also present at lower incidences: 16.3%, 12.5%, and 10.5%, respectively.

The majority of our cases (56/53.8%) were complete lesions. Erb's palsy (C5, C6) was present in only 10 cases (9.6%), and 39 infants (37.5%) presented a C5, C6, and C7 lesion (extended Erb's palsy). We had only one case (0.9%) of C8-T1 lesion (Klumpke's palsy).

The surgical techniques applied were neurolysis (11/10.5%), nerve grafts (27/25.9%), nerve transfers (36/34.6%), and a combination of grafts and transfers (32/30.7%).

The final general outcome at the last follow-up consultation (at least 24 months after surgery), according to the Active Movement Scale, was considered poor (AMS 4 or less) in 41.3% of the cases, good (AMS 5) in 34.3%, and excellent (AMS 6 or 7) in 24%. A functional result (good plus excellent) was achieved in 58.3% of the cases. Tables 2, 3, and 4 provide a detailed presentation of the surgical results in the different groups of patients.

Table 1 Narakas classification of newborns with brachial plexus palsy

Group	Name	Roots injured	Site of weakness/paralysis	Likely outcome
1	Erb's	C5, C6	Shoulder abduction/external rotation, elbow flexion	Good spontaneous recovery in over 80% of cases
2	Extended Erb's	C5, C6, C7	As above with drop wrist	Good spontaneous recovery in about 60% of cases
3	Total palsy with no Horner syndrome	C5, C6, C7, C8, T1	Complete flaccid paralysis	Good spontaneous recovery of the shoulder and elbow in 30–50% of cases. A functional hand nay be seen in many patients
4	Total palsy with Horner syndrome	C5, C6, C7, C8, T1	Complete flaccid paralysis with Horner syndrome	Expectation of severe defects throughout the limb, without surgery

Table 2 Detailed presentation of the surgical outcome in 9 infants with Erb's palsy (C5, C6)

Movement	Outcome (cases/percentage)		
	Poor (AMS ≤ 4)	Good (AMS = 5)	Excellent (AMS = 6/7)
Shoulder abduction	2 (22.2%)	4 (44.4%)	3(33.3%)
Shoulder external rotation	6 (66.6%)	3 (33.3%)	0
Elbow flexion	3 (33.3%)	1 (22.2%)	4 (44.4%)
Forearm supination	4 (44.4%)	4 (44.4%)	13 (11.1%)
Average	41.6%	36%	22.2%

AMS Active Movement Scale

Discussion

The diagnosis of brachial plexus birth palsy is usually made shortly after birth, consequent to observation of lack of movement in the upper extremities. Taking a detailed history may give information about risk factors. Substantial information regarding the extent of injury can be obtained through simple observation of the child. The physical examination is usually diagnostic and includes tactile stimulation, assessment of the neonatal reflexes, assessment for Horner syndrome, and palpation for bone fractures (e.g., clavicle or humerus). Such fractures may coexist with a brachial plexus birth palsy or cause pseudopalsy secondary to pain. The predominance of lesions on the right side that has been reported (67.3%) probably relates to higher frequency of the anterior left occipital-iliac presentation in vaginal delivery, which places the right shoulder under the maternal pubis.

According to the literature, the majority of the NBPP cases (50%) consist of Erb's palsy. Extended Erb's palsy accounts for around 25% of the lesions and panplexus or global lesions account for another 25% of NBPP cases [7, 18, 24, 32, 37, 41, 45, 47, 49]. A true isolated distal root palsy (C8-T1 lesion, Klumpke's palsy) is an exceedingly rare presentation. El-Sayed et al. [13] performed a search in the English-language literature covering a two-decade period and found that only 0.7% of the cases were Klumpke's palsy. Up to 10% of NBPP cases are bilateral, and these are seen almost exclusively in breech presentations [28]. The incidence of brachial plexus birth trauma in children born by means of cesarean section has been reported to be approximately 1% [4]. In disagreement with the literature, we had only a few Erb's palsy cases in our series (10 cases/9.6%) and a very large number of complete lesions (56 cases/53.8%).

There are divergent opinions regarding the degree to which imaging and electrodiagnostic studies are of importance. Nonetheless, these tests certainly do not substitute for physical examination. Computed tomographic myelography and magnetic resonance imaging can provide information about avulsion injuries but, although the presence of pseudomeningocele may give an idea regarding the integrity of the nerve root, false positives and negatives do occur [10, 29]. The drawbacks of imaging studies are the need for general anesthesia or heavy sedation and the invasive nature of CT myelography. Electrodiagnostic studies are difficult to interpret in newborns because of occurrences of early massive collateral sprouting of denervated muscles [54], which leads to overestimation of clinical recovery, especially in relation to the proximal muscles of the shoulder and arm [22]. Incorrect predictions of this nature may provide false hope to the parents and delay referral for surgical intervention. However, neurophysiological studies can still be performed at 1 month of age in selected cases when the prognosis is clinically unclear, including only comparison of

Table 3 Detailed presentation of the surgical outcome in 39 infants with extended Erb's palsy (C5, C6, C7)

Movement	Outcome (cases/percentage)		
	Poor (AMS ≤ 4)	Good (AMS = 5)	Excellent (AMS = 6/7)
Shoulder abduction	6 (15.3%)	22 (56.4%)	11(28.2%)
Shoulder external rotation	27 (69.2%)	10 (25.6%)	2 (5.1%)
Elbow flexion	5 (12.8%)	12 (30.7%)	22 (56.4%)
Forearm supination	19 (48.7%)	17 (43.5%)	3 (7.6%)
Elbow extension	2 (5.1%)	14 (35.8%)	23 (58.9%)
Wrist/fingers extension	11(28.2%)	13 (33.3%)	15 (38.4%)
Average	29.8%	37.5%	32.4%

AMS Active Movement Scale

Table 4 Detailed presentation of the surgical outcome in 56 infants with complete lesions (C5-T1)

Movement	Outcome (cases/percentage)		
	Poor (AMS ≤ 4)	Good (AMS = 5)	Excellent (AMS = 6/7)
Shoulder abduction	16 (28.5%)	31 (55.3%)	9 (16%)
Shoulder external rotation	43 (76.7%)	12 (21.4%)	1(1.7%)
Elbow flexion	22 (39.2%)	20 (35.7%)	14 (25%)
Forearm supination	40 (71.4%)	9 (16%)	7 (12.5%)
Elbow extension	16 (28.5%)	22 (39.2%)	18 (32.1%)
Wrist/fingers extension	32 (57.1%)	15 (26.7%)	9 (16%)
Thumb opponens	34 (60.7%)	11 (19.6%)	11 (19.6%)
Fingers flexion	33 (58.9%)	13 (23.2%)	10 (17.8%)
Average	52.6%	29.4%	17.5%

AMS Active Movement Scale

motor amplitudes during nerve conduction studies (CMAP) on axillary (deltoid) and radial (triceps) nerves [23], and occasionally electromyography of the biceps [53]. Because the impact of these imaging and electrodiagnostic tests on making the decision to undertake primary exploration of the brachial plexus is not high, some investigators [3], including us, have questioned their usefulness in birth palsy cases. We do not perform imaging or EMG studies routinely.

Nonsurgical treatment of NBPP should start early, on the first day of life, with passive range-of-motion exercises and active mobilization of the compromised limb. Its purpose is to ensure that the infant has full passive range of motion, to prevent occurrences of muscle contractures and/or joint deformity. The formal therapy sessions should be complemented with homemade exercises implemented by the parents several times during the day. For parents to avoid forgetting to do these exercises on their child at home, it is worth suggesting to them that these exercises should be done in association with some routine activity, such as changing the infant's diapers. The tactile stimulation that is provided through the exercises is also important for enabling cortical recognition and integration of the affected limb [55].

One of the most controversial topics related to infants with brachial plexus birth palsy is the timing of microsurgical intervention. The majority of nerve surgeons agree that infants presenting total plexus palsies with Horner syndrome should undergo microsurgical reconstruction of the plexus at 3 months of age [50, 55]. However, there is no consensus for the remaining 70 to 80% of patients with a typical Erb's palsy or extended Erb's palsy. Some authors have advocated microsurgical intervention if no elbow flexion against gravity is present by the age of 3 months [5, 21, 31, 51]. Equivalent functional outcomes were observed by other authors in infants who regained elbow flexion against gravity between 4 and 6 months of age [1, 56]. On the other hand, some authors have preferred to wait at least 9 months to maximize the possibility of spontaneous recovery before indicating microsurgical treatment [10]. Further delay is not advisable, since poorer results should be expected, especially regarding hand function [9]. In our series, we tried to operate complete lesions 3 months after birth and upper lesions somewhere between 4 and 6 months. However, because of delayed referral, the surgeries were often performed later.

No surgical strategy can be applied in all cases of obstetric palsy. The most widely used technique for restoring function after brachial plexus birth palsy is still neuroma resection and nerve grafting. However, nerve transfers are gaining increased popularity as an addition to nerve grafting or as its substitute.

We performed neurolysis as the only treatment in 11 of our patients, at the beginning of the series, with poor results in all of these cases. Since publication of the important paper by Clarke's group in Toronto [35] demonstrating that the early eventual improvements in function produced by neurolysis were not sustained over time, we have abandoned this technique in favor of grafting or nerve transfer. Interposition of nerve grafting has been the mainstay of surgical treatment in NBPP patients for many years. However, increasing use of nerve transfers in reconstructing traumatic brachial plexus injuries in adults then led to better outcomes in relation to nerve grafting in some lesions [15], and hence, reports relating to nerve transfers in NBPP cases became more frequent [2, 42, 48].

This tendency towards use of nerve transfers subsequently diminished when a report from the Scientific Committee for NBPP of the International Federation of Societies for Surgery of the Hand was published. According to that review, surgeons need to avoid an overreliance on nerve transfers and favor nerve graft reconstruction [52]. Corroborating this committee's findings, a recently published series comparing outcomes after grafting and nerve transfers in patients with extended Erb's palsy demonstrated that distal nerve transfers provide faster recovery, but that there was no difference in outcomes after 24 months after surgery [25]. Nowadays, our strategy is to perform nerve grafting whenever possible. Nerve

transfers do have an important role to play on certain occasions, including in cases of root avulsions, failed primary grafting reconstruction, late presentation, isolated deficits, and as a complement in some grafting cases.

The results from microsurgical reconstruction vary and comparison between different series is almost impossible, given the differences in preoperative evaluation, surgical strategies, and outcome analysis. The recent report by Sarac et al. [46] gives an idea of the problem: in a systematic review, 59 different methods of outcome measurement were found in 217 papers. As a consequence of this difficulty, the results from microsurgical treatment in the literature are quite variable: it has been reported that good return of shoulder abduction occurs in 25 to 92% of the cases and that elbow flexion against gravity occurs in 26 to 86% of the cases. The rate of reinnervation of the hand has ranged from 35 to 64% [6, 9, 12, 16, 21, 26, 34, 36, 43, 47, 51, 56]. In general, our results were lower than the best results the reported in the literature: 75.3% of the cases presented good/excellent results for shoulder abduction; 27.9% for shoulder external rotation; 65.9% for elbow flexion and 41% for finger flexion. One possible explanation may have been our much higher number of total palsy cases (53.8% vs. 25% in the literature).

After surgery, we prefer the Gilchrist-Velpeau type of immobilization, instead of a cast. The access to the chest for physiotherapy is better, the patient breathes more easily, and his or her overall comfort is greater. In a large series, no loss of nerve recovery was noted when using this softer type of immobilization [33]. After 3 weeks of immobilization the child is introduced into a rehabilitation program. The most important aims of rehabilitation are to induce creation of adequate cortical motor patterns and to avoid establishment of wrong motor patterns. As mentioned before, this needs to be started as early as possible.

In many instances, the surgical outcome consists of incomplete recovery of function. If the resultant muscle imbalance is not treated, this will lead to joint deformities. In those cases, secondary orthopedic procedures (most commonly tendon transfers and osteotomies) can be performed to further enhance function.

Conclusions

There is no consensus regarding strategies for treatment of NBPP. Our retrospective analysis on surgical outcomes in our relatively small group of patients with NBPP indicated that good functional results from nerve reconstruction can be expected in a well-selected group of children among whom no substantial functional recovery had been attained through conservative treatment. However, further improvements are hindered, at least in part, by the impossibility of making comparisons among outcome data from different specialized centers. Maybe in the future, the results from consensus surveys (like iPluto [44]) on how to assess these patients, select the best available treatment, and report outcomes will improve our understanding and the way to manage these children.

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