

# An Update on the Management of Neonatal Brachial Plexus Palsy—Replacing Old Paradigms

## A Review

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**IMPORTANCE** Neonatal brachial plexus palsy (NBPP) can result in persistent deficits for those who develop it. Advances in surgical technique have resulted in the availability of safe, reliable options for treatment. Prevailing paradigms include, “all neonatal brachial plexus palsy recovers,” “wait a year to see if recovery occurs,” and “don’t move the arm.” Practicing by these principles places these patients at a disadvantage. Thus, the importance of this review is to provide an update on the management of NBPP to replace old beliefs with new paradigms.

**OBSERVATIONS** Changes within denervated muscle begin at the moment of injury, but without reinnervation become irreversible 18 to 24 months following denervation. These time-sensitive, irreversible changes are the scientific basis for the recommendations herein for the early management of NBPP and put into question the old paradigms. Early referral has become increasingly important because improved outcomes can be achieved using new management algorithms that allow surgery to be offered to patients unlikely to recover sufficiently with conservative management. Mounting evidence supports improved outcomes for appropriately selected patients with surgical management compared with natural history. Primary nerve surgery options now include nerve graft repair and nerve transfer. Specific indications continue to be elucidated, but both techniques offer a significant chance of restoration of function.

**CONCLUSIONS AND RELEVANCE** Mounting data support both the safety and effectiveness of surgery for patients with persistent NBPP. Despite this support, primary nerve surgery for NBPP continues to be underused. Surgery is but one part of the multidisciplinary care of NBPP. Early referral and implementation of multidisciplinary strategies give these children the best chance of functional recovery. Primary care physicians, nerve surgeons, physiatrists, and occupational and physical therapists must partner to continue to modify current treatment paradigms to provide improved quality care to neonates and children affected by NBPP.

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Occurring secondary to stretching of the nerves of the brachial plexus, neonatal brachial plexus palsy (NBPP) occurs in 0.5 to 3 per 1000 live births.<sup>1-4</sup> Most studies report a persistent deficit in 20% to 30% of patients.<sup>5,6</sup> In the early 20th century, surgery for NBPP was associated with high morbidity and mortality. However, in 1991, Gilbert et al<sup>7</sup> showed that primary nerve surgery for patients with persistent NBPP was safe. The establishment of safety set the stage for 25 years of progress. Today, with advancements in microsurgical technique, development of new techniques (eg, nerve transfers), and increased understanding of the secondary sequelae of NBPP, we can achieve good, safe, and reliable outcomes for patients with persistent NBPP. Despite this improvement, several paradigms still prevail, including “all neonatal brachial plexus palsy recovers” and “wait a year to see if recovery occurs.” These beliefs put the patient with NBPP at a disadvantage. There is a limited time during which primary nerve surgery can

provide benefit. Thus, the goal of this review is to provide an update on the management of NBPP.

### Pathogenesis

The fundamental injury in NBPP involves stretch to the nerves that comprise the brachial plexus, C5 to T1, during childbirth. Nerve injuries can be classified on the basis of the degree of anatomic disruption of axons and the surrounding endoneurium, perineurium, and epineurium.<sup>8,9</sup> A neurapraxic injury is the least severe and involves a disruption in the myelin sheath around spared axons, causing a conduction block. An axonotmetic injury involves both disruption of the myelin sheath as well as the axon, but preservation of the perineurium and epineurium. Finally, a neurotmetic lesion is a complete rupture of the nerve involving the axon, myelin sheath, and

Table. Active Movement Scale<sup>a</sup>

Finding	Associated Score
With gravity eliminated	
No contraction	0
Contraction without movement	1
Movement <½ range of motion	2
Movement >½ range of motion	3
Full range of motion	4
Against gravity	
Movement <½ range of motion	5
Movement >½ range of motion	6
Full range of motion	7

<sup>a</sup> Scores are assigned on the basis of observed movement compared with achievable passive range of motion, with gravity eliminated and against gravity. A score of 4 must be achieved (full range of motion with gravity eliminated) before a score of 5 to 7 can be assigned.<sup>22</sup>

connective tissues, including the perineurium and epineurium, representing the most severe type of peripheral nerve injury. Neurapraxic and axonotmetic lesions can spontaneously recover, whereas neurotmetic lesions and avulsions have no ability to spontaneously recover.

Neurapraxic injuries begin their repair by rewiring the injured segment with myelin for the nerve to regain its conduction properties. The repair of axonotmetic injuries requires a more involved process. Approximately 2 to 3 days after disruption of the axon, the process of wallerian degeneration begins. The axon then has to grow back down the neurotubule, which occurs at a rate of approximately 2.5 cm per month. Even in a neonate, this equates to months of delay before axons can reinnervate the target.

Ultimately, without reinnervation, irreversible changes occur at the neuromuscular junction, with loss of myofibrils and ultimately death of the muscle cell, such that after these changes have occurred, even if the axons are returned to the proximity of the muscle, functional reinnervation does not occur.<sup>10</sup> In rabbits, these irreversible changes occur approximately 1 year following denervation.<sup>11</sup> In humans, the time course is variable, but most agree that these changes begin to occur between 12 and 24 months after denervation. In addition to muscle and neuromuscular junction changes, prolonged axotomy results in decreased axonal sprouting following nerve repair.<sup>12</sup> These time-sensitive, irreversible changes are the scientific basis for early evaluation and management of NBPP and put into question the mantra of "observe for a year to see if there is recovery."

## Clinical Presentation

The hallmark sign of NBPP is incomplete active range of motion but preserved passive range of motion. While the clinical recognition of NBPP typically comes from asymmetric upper extremity weakness, there are other associated neurologic injuries that can be recognized in some patients. A Horner sign (ptosis, miosis, and anhidrosis) may be present, as the second-order neuron in the sympathetic chain to the eye generally exits the spinal cord between the C8 and T2 levels and can be damaged. Diaphragmatic weakness or paralysis may be present owing to injury of the phrenic nerve, comprising branches from the C3 to

C5 nerve roots, and can be asymptomatic or present as respiratory distress, feeding difficulties, or asymmetric chest rise. Associated orthopedic injuries may also be clinically apparent, including shoulder dislocations or clavicle fractures.

Although there is no way to accurately predict which children will be affected, there are known risk factors associated with the child, the mother, and the delivery. The major infant risk factor is the child's birth weight, with increasing birth weight being associated with increasing risk of NBPP. Maternal risk factors include advanced maternal age, primiparity, maternal obesity, and diabetes. Delivery risk factors include shoulder dystocia, breech delivery, vacuum or forceps assistance, and abnormalities of the second stage of labor.<sup>13-19</sup> Both multiple birth mates and cesarean delivery have been associated with a decreased risk of NBPP.<sup>4</sup> While these risk factors pertain to the incidence of NBPP, we have recently identified risk factors for persistence (ie, incomplete spontaneous recovery). These risk factors include cephalic presentation, induction or augmentation of labor, birth weight more than 4 kg, and having an associated clavicle fracture. Cesarean delivery was associated with a decreased risk of persistence.<sup>20,21</sup>

## Diagnosis

The mainstay of diagnosis is the physical examination. While imaging and electrodiagnostic testing can be helpful in specific circumstances, they should be thought of as extensions of the physical examination and not as replacements. Clinical assessment should include both a thorough history, including gestational and birth history, and a thorough physical examination. The goals of the history and physical examination include localization of the neurologic injury, determining the severity of the injury, identifying risk factors for persistence, identifying any associated nonneurologic injuries, and monitoring for spontaneous recovery on sequential examinations.

A variety of methods and grading scales have been developed to allow appropriate assessment of the neonate and a common language that can be used for the purposes of both research and clinical care. One such scale that is often used is the Active Movement Scale.<sup>22</sup> This scale relies on observation of the degree of movement against gravity and with gravity eliminated (Table). The strengths of this scale include the ability to apply the grading scale in an infant who does not respond to commands or voluntarily participate in the examination and the ability to apply the scale to all movements of the upper extremity. The scale has been shown to have moderate to near perfect interrater reliability for most movements tested.<sup>22</sup> While high interrater reliability of this scale aids in detecting changes on sequential examination, an early referral facilitates sequential examinations by the same person or team over time and aids in the detection of meaningful, spontaneous recovery, even with small changes. The Active Movement Scale has the additional benefit of being able to be converted into a 3-month test score that is predictive of benefit from surgical intervention.<sup>23</sup>

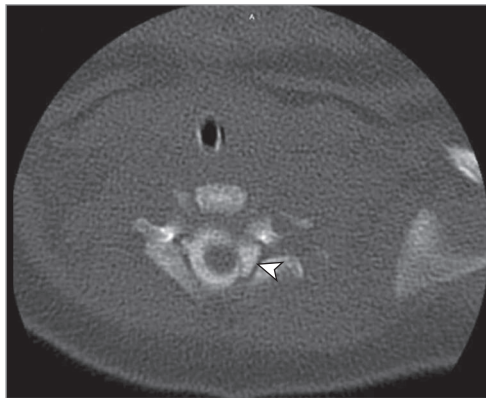
Several additional special tests have been developed and subsequently incorporated into many of the decision algorithms used for determining surgical candidacy. One such test is the cookie test. When the child is aged 9 months, a cookie is placed in his or her hand. The elbow is then held against the child's side. The test is successful if the child can get the cookie to his or her mouth without flexing

Figure 1. Myelography for Patients With Neonatal Brachial Plexus Palsy

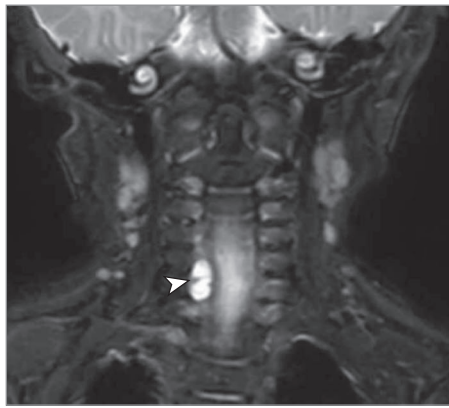
A Coronal CT myelogram



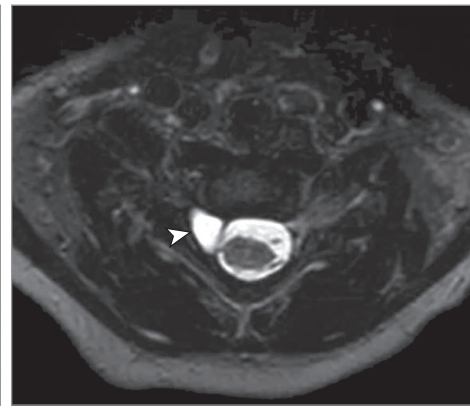
B Axial CT myelogram



C Coronal MR myelogram



D Axial MR myelogram



Coronal (A) and axial (B) computed tomographic (CT) myelogram showing a pseudomeningocele in the neural foramen (arrowheads), suggestive of a nerve root avulsion. Computed tomographic myelography is performed with intrathecal injection of iodinated contrast. Coronal (C) and axial (D) magnetic resonance (MR) myelogram showing a pseudomeningocele (arrowheads) in the neural foramen, suggestive of a nerve root avulsion. Magnetic resonance myelography does not require intrathecal injection, iodinated contrast, or ionizing radiation.

the neck beyond 45°. <sup>24</sup> Similarly, in the towel test, a towel is placed over the infant's eyes and the test is successful if he or she can remove the towel with the affected extremity vs the unaffected extremity. This test has been shown to be useful both for the clinical and electrodiagnostic examinations. <sup>25</sup>

While motor testing and localization are the most important components of the examination of the infant with NBPP, other findings are also significant. The presence of Horner syndrome is associated with a proximal injury (usually a nerve root avulsion) to C8 and/or T1 and correspondingly is predictive of a persistent deficit. <sup>20,21</sup> Asymmetric chest expansion should be noted, as it may suggest injury to the phrenic nerve and hemidiaphragm paralysis, and further investigation with diaphragmatic ultrasonography is warranted. Joint subluxations and contractures are also important clinical findings. Given that both of these abnormalities typically take several months to develop, the early presence of either may indicate an additional musculoskeletal disease process and warrants further investigation. <sup>26,27</sup>

Imaging has not been commonly used in most decision algorithms, although it may be predictive and increasingly used in the future. <sup>21</sup> The most common use for imaging is examining for evidence of nerve root avulsion injuries to aid in surgical planning. The 2 available and most commonly applied are computed tomographic and magnetic resonance myelography. Typical diagnostic criteria for nerve root avulsion, regardless of modality, include either

the presence of a pseudomeningocele or a pseudomeningocele with absent rootlets (Figure 1). Sensitivity for nerve root avulsions by computed tomographic myelography is approximately 70% and the specificity is 85% to 95%. <sup>28-30</sup> Magnetic resonance myelography offers similar sensitivity and specificity, approximately 68% and 96%, respectively, but with the advantages of being noninvasive, lacking the need for intrathecal iodinated contrast, and requiring no exposure to ionizing radiation. <sup>29,31</sup> Magnetic resonance neurography and ultrasonography can be used to image extraforaminal nerves. The relative merits of each test continue to be evaluated. <sup>32-35</sup>

Electrodiagnostics are fraught with difficulties in neonates and their utility is debated because of a common lack of concordance with clinical findings. While the results are difficult to interpret, interrater reliability is high. <sup>36</sup> Electrodiagnostic studies have been incorporated into some diagnostic and decision-making algorithms, such as the University of Michigan NBPP Treatment Pathway; however, this practice varies from center to center and would not be considered standard of care. <sup>21</sup>

## Management

Management of NBPP has changed significantly over the past 25 years, particularly with the development of safe and effective surgical tech-

niques. As new techniques, such as nerve transfers, continue to be developed, management paradigms are likely to evolve. No longer is NBPP a disease without any viable solutions, which makes the upfront, early management important.

Management begins with astute recognition that NBPP is present and differentiating NBPP from neurologic and musculoskeletal conditions with similar presentations, such as a septic joint, hemiplegic cerebral palsy, or arthrogryposis. Infants and children with NBPP should be referred for occupational therapy as soon as the diagnosis is suspected. Contrary to previous thought and arguing against the old mantra of "don't move the arm," early initiation of range of motion activities does not appear to increase the risk of shoulder subluxation.<sup>37</sup> Furthermore, early immobilization of the affected arm is not indicated except in the case of an orthopedic condition requiring it and may be counterproductive to the patient's recovery. Goals of therapy include maintaining normal passive joint range of motion, promoting functional use of the affected extremity, selectively strengthening affected muscles, and when necessary, providing compensatory techniques to promote independence. Therapeutic exercises should be taught to the caregivers so that they may be continued regularly at home outside of scheduled therapy sessions. Techniques for family education should include direct teaching and demonstration as well as supplemental visual aids, such as video, to ensure that exercises are performed correctly at home.<sup>38</sup>

A variety of therapy techniques may be used and the goals of therapy may change over time. Stretching, targeted strengthening, and splinting are the pillars of a therapy program. Additional therapies, such as desensitization, therapeutic taping, and electrical stimulation, are used at various centers; however, their efficacy is unclear. Preschool-aged children with NBPP may have impairment in self-care skills, necessitating functional task training in addition to the aforementioned techniques.<sup>39</sup>

Patients with NBPP should be screened for associated conditions that occur with increased frequency in this population. Two of these conditions are torticollis and early speech delay, highlighting the importance of multidisciplinary care for children with NBPP.<sup>40,41</sup> Patients should also be assessed for pain, which in preverbal children may present as self-mutilation with excessive mouthing or biting of the affected extremity.<sup>42</sup> Pain may also result from substituted abnormal movement patterns or from overuse of the contralateral arm. Pain during range-of-motion exercises in a young infant should trigger reevaluation for skeletal injury.

While each specialty center may differ in its practice preferences, we and many others agree that a child who has not completely recovered by age 1 month should be referred to a multidisciplinary NBPP specialty program.<sup>43</sup> To avoid missing the window of opportunity in which these children can be significantly helped, early referral from primary care physicians is necessary. Going against the mantras of "all NBPP recover" and "wait a year to see if recovery occurs," the improvement in surgical techniques and the need to intervene before permanent changes in the nerve, muscle, and neuromuscular junction occur mean that early referral should be the new paradigm.

### Indications for Nerve Surgery

Once the child is referred to the specialty center, the ultimate goal is to predict which children will achieve satisfactory spontaneous recovery and which children will not. A variety of decision algorithms have been developed. One such algorithm is the University of Michigan NBPP

Treatment Pathway.<sup>21</sup> This algorithm uses a combination of serial physical examination and electrodiagnostics to arrive at a decision. Ultimately, if the child has not recovered sufficient biceps strength to raise the hand to the mouth by age 6 months against gravity, then he or she is considered a surgical candidate. Overall, this algorithm is largely based on data from Gilbert et al<sup>7,44</sup> suggesting that, if spontaneous recovery of biceps was poor at 3 months, the motor outcomes were correspondingly poor at 5 years with conservative management. Michelow and colleagues<sup>45</sup> challenged these data and pushed for a slightly more delayed decision by demonstrating that utilizing absent biceps function at 3 months to predict long-term biceps recovery is incorrect 12% of the time. This idea ultimately led to the development of the algorithm used by the University of Toronto.

The University of Toronto Test Score evaluation is applied at 3 months and is based on an evaluation of elbow flexion, elbow extension, and wrist, finger, and thumb extension. The total score is based on conversion of scores for these movements on the Active Movement Scale. A subthreshold score on the Toronto Test Score (<3.5) predicts poor spontaneous recovery with conservative management and is associated with a 5% risk of an inaccurate prediction.<sup>22,24,45</sup> In the University of Toronto algorithm, a failing Test Score at 3 months is considered to be an indication for surgical management; if the score is above that level, the child is reevaluated at 6 and 9 months. The evaluation at 9 months consists of the cookie test.<sup>22</sup> If the child cannot successfully complete the cookie test, primary nerve surgery is pursued. If the child is successful, then primary nerve surgery is no longer considered, but the child is followed up for the development of secondary sequelae in the case of incomplete recovery.<sup>24</sup> Thus, the major decision points in this algorithm are 3 and 9 months, emphasizing the importance of early referral.

The decision to perform primary nerve surgery is a balancing act: on 1 end is the need to allow sufficient time for demonstration of spontaneous recovery, while on the other end are data suggesting that outcomes are improved with earlier intervention.<sup>19</sup> We developed a decision tree that incorporates multiple factors that can be assessed shortly after birth. Factors included in the decision tree were Narakas<sup>46</sup> grade (a system categorizing NBPP by root involvement), presence of a clavicle fracture at birth, Horner syndrome on examination, presence of a pseudomeningocele on imaging (either computed tomographic or magnetic resonance myelography), birth weight, and utilization of induction or augmentation of labor. This decision tree had a high positive predictive value (94%) and moderate negative predictive value (79%) for failure to improve spontaneously beyond surgical candidacy.<sup>21</sup> Based on work from Van Dijk et al<sup>47</sup> and Malesy et al<sup>48</sup> suggesting that early electrodiagnostic and examination findings are predictive, future iterations will focus on improving both predictive values through the incorporation of early clinical examination and electrodiagnostic findings.

Based on generally agreed upon concepts from institution to institution, our suggested management algorithm for the primary care physicians who encounter children with NBPP early in life is demonstrated in **Figure 2**. Regardless of the specific algorithm used for decision making, once surgical candidacy is determined, questions regarding the surgical options and type of surgery need to be addressed.

### Nerve Surgery: Techniques and Results

Currently, primary nerve surgery for NBPP involves nerve grafting, nerve transfer, or a combination of the 2. In nerve grafting, the in-

jured segment is removed and a conduit, typically an autogenous sural nerve graft, is placed between the proximal stump and the distal target. Nerve transfer surgery sacrifices a nerve to a muscle with redundant function or a fascicle with intraneural redundancy and connects it to a denervated target.

Gilbert et al<sup>7</sup> reported a series of 1000 infants with NBPP, 241 of whom had graft repair. They found that, among the surgical group, 81% of infants with C5 to C6 repairs and 76% of those with C5 to C7 repairs achieved shoulder abduction of at least 90° 3 years postoperatively. The success of operative treatment and the lack of reported complications spurred further development of operative techniques. The safety of surgical treatment was further supported in a report from the Hospital for Sick Children in Toronto. The group reported no mortality in a series of 173 consecutive patients undergoing surgical treatment for NBPP and minimal morbidity that was nearly eliminated with simple interventions, such as limiting intraoperative volume of intravenous fluids and suturing in the endotracheal tube intraoperatively.<sup>49</sup>

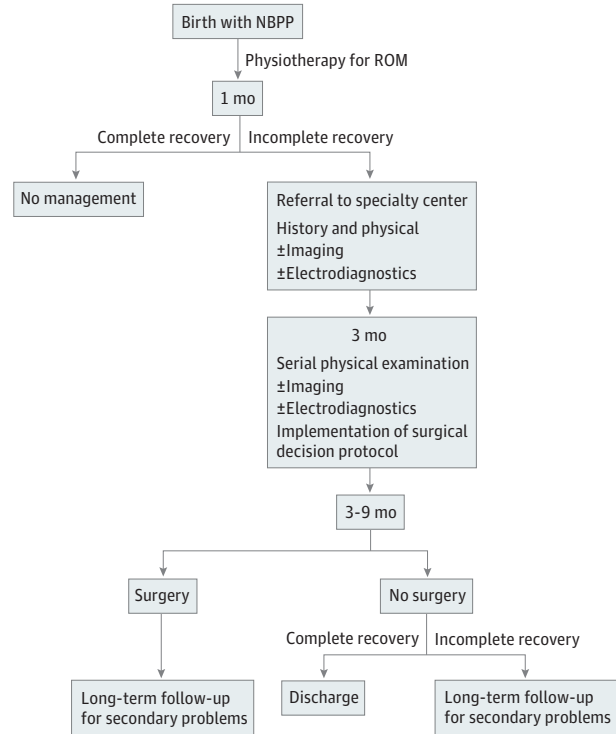
Prior to the initiation of nerve transfers, surgical options were limited to neurolysis and nerve graft repair. Lin and colleagues<sup>50</sup> have shown that both for C5 to C6/7 and total brachial plexus palsy, nerve graft repair results in significant improvement in multiple movements. Graft repair outperformed neurolysis in both groups of patients.

Although the specific roles for nerve graft repair and nerve transfer continue to be defined, the development and popularization of nerve transfers have significantly advanced the field.<sup>51,52</sup> In adults, transfer of a median or ulnar nerve fascicle to the biceps branch of the musculocutaneous nerve has proven to be an effective nerve transfer. Little and colleagues<sup>53</sup> helped to establish this nerve transfer as an effective option for NBPP. They showed that 87% of patients achieved a score of 6 or more on the Active Movement Scale (joint motion greater than one-half range of motion against gravity) with this nerve transfer, with only 1 patient having transient anterior interosseous nerve palsy as a complication. Other donor options for nerve transfer to the biceps branch include the medial pectoral nerve and intercostal nerves. Pondaag and Malessy<sup>54</sup> showed that over 88% of patients with NBPP achieved Medical Research Council grade of 3 or more biceps functions using these donor nerves. In patients with C5 to C7 injuries, outcomes for distal nerve transfers compared with supraclavicular nerve grafting have been shown to be similar at 24 months, although recovery of movement typically occurs faster for nerve transfers.<sup>55</sup>

Restoration of shoulder function, including abduction and external rotation, has proven somewhat more difficult. Malessy and Pondaag<sup>56</sup> used nerve graft repair, grafting from C5 to the posterior division of the upper trunk. This technique resulted in Mallet IV abduction (>90°) in 65% of patients, whereas only 32% achieved Mallet IV external rotation (>20°). Spinal accessory to suprascapular nerve transfer has shown promise, but in one study of 14 patients, only 29% achieved Mallet IV external rotation.<sup>57</sup> Comparatively, spinal accessory nerve transfer has been shown to outperform nerve graft repair, both for overall postoperative external rotation recovery and the need for secondary shoulder surgery.<sup>58</sup>

Overall, nerve surgery in appropriately selected candidates can significantly improve function for those with NBPP. In a meta-analysis, surgery outperformed nonoperative management with a relative risk of functional impairment of 0.58. Adverse events were

**Figure 2. Proposed Management Algorithm for Neonatal Brachial Plexus Palsy (NBPP)**



This proposed management algorithm for NBPP is based on general concepts that are consistent from institution to institution. This algorithm emphasizes both the need for early referral to a multidisciplinary specialty program and the need for long-term follow-up to evaluate for potential secondary sequelae. ROM indicates range of motion.

rare.<sup>59</sup> With the increasingly established role for surgical management, the trend has been toward more surgical utilization. Nonetheless, surgery likely remains underused. In a large database study, only 3.3% of children born with NBPP underwent primary nerve surgery. With 20% to 30% of patients with NBPP having a persistent deficit, this likely represents an underutilization of surgical management.<sup>60</sup> We believe that improved awareness and early referral by primary care physicians could have a significant benefit on the long-term outcomes in children with NBPP.

## Conclusions

Advances in surgery for NBPP have given us options to help restore function for these children. Mounting data support both the safety profile and effectiveness of surgery for patients with persistent NBPP. Despite this support, surgery is underused.<sup>60</sup> The prevailing paradigms of "all neonatal brachial plexus palsies recover" and "wait a year to see if spontaneous recovery occurs" are harmful and are not supported by current data. These paradigms should be replaced with one of early referral. Referral to a specialty center at age 1 month is desirable if recovery is not progressing, especially if the child is lacking antigravity elbow flexion. Early implementation of multidisciplinary strategies and early recognition of persistent NBPP give these children the best chance of

restoration of function. Primary care physicians are the initiators of care for this pathology and can provide a major benefit on the outcomes. Surgery is but one part of the multidisciplinary care of NBPP. Primary

care physicians, nerve surgeons, physiatrists, and occupational and physical therapists must partner to change the paradigm and provide high-quality care to neonates and children affected by NBPP.

## ARTICLE INFORMATION

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