Neurosurgical forum
Letters to the editor

Neurolysis and upper trunk brachial plexus birth palsy

To The Editor: We read the article by Andrisevic et al.1 with interest (Andrisevic E, Taniguchi M, Partington MD, et al: Neurolysis alone as the treatment for neuroma-in-continuity with more than 50% conduction in infants with upper trunk brachial plexus birth palsy. Clinical article. J Neurosurg Pediatr 13:229–237, February 2014). The authors are to be commended for their efforts to improve treatment of patients with this severe birth trauma that has lifelong implications; however, we do have some concerns about the applied treatment and analysis.

The authors evaluated 441 infants, of whom 44 (10%) underwent nerve surgery: nerve grafting in 14 and neurolysis of the upper trunk in 30. Of the 30 patients who underwent neurolysis, 13 did not meet the inclusion criteria. As a result, 17 (39%) of the 44 surgically treated patients were included in the present analysis. Compound motor action potential (CMAP) responses were recorded after direct electrical stimulation was applied proximal and distal to the superior trunk neuroma. The authors used the Laurent and Lee treatment algorithm,6 which employs action potential (CMAP) responses were recorded after direct electrical stimulation was applied proximal and distal to the superior trunk neuroma. The authors used the Laurent and Lee treatment algorithm,6 which employs a 50% conduction threshold as the cutoff for neurolysis alone versus neuroma resection. All patients included in the study had a neuroma-in-continuity with more than 50% conduction across the neuroma, and subsequently C-5 and C-6 neurolysis was performed. Based on their results the authors concluded that their group of patients benefitted clinically from the neurolysis. We would like to address a number of issues concerning the analysis and conclusions that need attention.

Surgical Method in Relation to the Pathophysiology

The most important issue is the authors’ claim that neurolysis had a therapeutic benefit in their series. Neurolysis, as they performed it, was described as “making multiple longitudinal incisions into the thickened epineurium of the neuroma, until axonal tissue could be visualized.” The authors did not include a control group consisting of similar patients in whom they did not make multiple longitudinal incisions. Instead, they made a comparison with cases reported on in the literature. We are not aware of any study supporting the claim that this type of surgical intervention indeed improves axonal regeneration and, consequently, has a beneficial effect on nerve function recovery. In our opinion, the authors’ claim does not hold up in view of the pathophysiological process underlying brachial plexus birth palsy (BPBP). In severe traction lesions of the brachial plexus with axonal rupture, loss of basal lamina tube continuity, fascicular rupture and fibrosis typically result in a neuroma-in-continuity. Neurological recovery is limited because axons cannot successfully cross the lesion site due to an increase in extracellular matrix and expression of growth-inhibiting proteins such as semaphorin 3A by epineural and perineurial fibroblasts.13 Additionally, functional recovery is impaired due to axonal misrouting.7 How neurolysis could improve axonal recovery in such a complex setting is difficult to understand, unless the selected patients had a predominantly axonotmetic lesion.

To support their view, the authors wrote, “Neurolysis is believed to reduce high intraneuronal pressure, improve blood flow, and alleviate the physical barriers to regeneration.”8 The report by Nelson to which they are here referring, however, does not contain any original, experimental, or clinical work showing that these factors play a role in BPBP or are the cause of frustrated regeneration, nor that neurolysis has a beneficial effect on regeneration.

We agree with the authors that absent neurological recovery in a child with BPBP is an indication for surgical exploration. When the brachial plexus elements show fascicular continuity across the lesion site and when the muscles respond to direct electrical stimulation with clear contractions, the surgery is ended. We label this procedure as “neurolysis”—that is, a diagnostic procedure. The recovery that subsequently takes place is the logical result of completing axonal outgrowth through the damaged area, to which neurolysis did not contribute.

Intraoperative Neurophysiology

The authors stated that the protocol they followed at their institution most closely mirrors that of Laurent and Lee.6 It is, in this respect, crucial to stress that the applied methodology has never been validated, and the cutoff point of 50% conduction across the neuroma was arbitrarily set in the original paper. In our opinion, the authors should have highlighted and discussed this more prominently. The authors did not discuss the potential implications of the fact that the infants in their series were twice as old as those in the Laurent and Lee series. The mean age of patients in the Laurent and Lee series was 5 months (range 2–18 months) whereas that in their own series was 10 months (range 6–19 months). The presence of 50% conduction across a neuroma-in-continuity at 5 months cannot be compared to similar findings at 10 months.

The authors’ choice for the applied electrophysiological method is not clear. The underlying pathophysiological mechanisms were thoroughly investigated with alternative
Neurosurgical forum

Intraoperative testing. Nerve action potential recording, for instance, requires regrowth of axons across the lesion site, which is indicative of commencing successful reinnervation. Comparing the amplitude of CMAPs across a lesion site, however, is usually done to diagnose conduction blocks or demyelination in neuropathies (for instance, compression at the carpal tunnel or elbow). The authors’ choice to compare CMAPs proximal and distal to a nerve traction lesion should be clarified in more detail because it is an unusual method by which to measure axonal loss.

Unfortunately, not all relevant publications on intraoperative neurophysiology were referenced in the article. The available literature contains relevant data that put the authors’ conclusions in a different perspective. König et al. were very unsatisfied with the outcome in their 5 patients after performing neurolysis “only,” when conduction over the neuroma was demonstrated. We have studied the predictive value of nerve action potentials and CMAPs in 95 BPBP patients. Although axonotmesis, neurotmesis, and avulsion could be distinguished on group level, we were unable to identify valid cutoff points for the individual patient to facilitate the decision of whether to cut a neuroma-in-continuity and graft or leave the nerve-in-continuity. We therefore do not share the authors’ opinion that intraoperative electrophysiological testing is of paramount importance to surgical decision making during primary exploration.

Chin et al. used CMAP recordings after direct intraoperative stimulation, as Andrisevic et al. did, but instead of comparing the amplitude resulting from stimulation proximal and distal to the neuroma, they looked at the morphology of the CMAP to help in the decision-making process. Roughly two-thirds of their surgically treated patients (22 of 32) underwent neurolysis only, which resulted in good recovery of abduction and elbow flexion but poor recovery of external rotation.

Functional Recovery and Interpretation

The authors qualified the results of shoulder function and of elbow flexion recovery as good, while functional external rotation recovery occurred in only one-third of their patients. In our opinion, external rotation is the hallmark function of recovery in BPBP patients. We are well aware that the results from nerve grafting or nerve transfer are not spectacularly better as compared to the authors’ results. Improving the outcome of external rotation is one of the most important targets to achieve in treating these children. In that respect, the authors’ treatment paradigm unfortunately did not add to improving external rotation. In fact, biceps function recovery is not the only goal of nerve reconstruction. Biceps function is employed by most authors as a proxy to diagnose the severity of the upper trunk lesion and is thus used as an indicator of nerve reconstruction to improve elbow flexion and shoulder function, including external rotation.

Analysis and Reached Conclusion

The authors claimed to have demonstrated the superiority of neurolysis in the treatment of BPBP over conservative treatment. This claim was based on a selection of 17 patients from a total group of 30 treated with neurolysis. This might have created an inclusion bias. Outcomes in these patients were then compared to those in 7 conservatively treated patients reported on in literature by means of a numerical conversion. This conservatively treated patient group from the literature was already biased by inclusion, as 27 of 66 patients were not included in the analysis in the original paper.

The authors discarded the results from Toronto Sick Kids group (led by Clarke, in the article by Lin and colleagues’), which led them to conclude that neurolysis alone must not be used for treatment of BPBP. The main reason for this condemnation is the limited number of patients in Lin and colleagues’ neurolysis group (n=8), which serves in fact as a control group for grafting (n=48). This number is small indeed but is similar in size as the group that Andrisevic et al. extracted from the literature and used as their control group (n=7) for neurolysis (n=17).

It is, all in all, in our opinion questionable whether the methodology followed by the authors can really support their conclusion, which promotes neurolysis as surgical treatment in BPBP infants.

We would like to conclude with the following quote from Sunderland: “When neurolysis performed under these conditions is followed by recovery it is only natural to attribute the improvement to the surgical procedure which, it is assumed, has removed a barrier to regeneration. The value of neurolysis in these cases is, however, difficult to assess…. It is, therefore, impossible to exclude the possibility that neurolysis in these cases is merely coincidental to delayed, but normal, regenerative processes that would, in any event, have resulted in recovery.”

Disclosure

The authors report no conflict of interest.

References

6. Laurent JP, Lee RT: Birth-related upper brachial plexus inju-