Neonatal brachial plexus palsy


The authors formulated a decision analytical model to compare 4 treatment strategies (no repair or repair at 3, 6, or 12 months of life) for infants with persistent NBPPs. The model derives data from published studies and projects health-related quality of life and quality-adjusted life years over a lifetime. The authors conclude that their approach supports a delayed approach of primary surgical reconstruction to optimize quality of life, and that early surgery for NBPPs may be an overly aggressive strategy for infants.

We think that the approach taken by the authors is to be commended, but that the methodology contains major flaws. A valid conclusion can, therefore, not be drawn from their utility model.11

Primary Outcome Measure

The authors selected elbow flexion as the primary indicator of outcome, as they considered this the most significant driver of function in Erb’s palsy. In this respect we disagree with the authors. In fact, biceps recovery is not the goal of nerve reconstruction, but biceps function is used by most authors as a proxy to diagnose severity of the upper trunk lesion, and is thus an indicator of nerve reconstruction to improve elbow flexion and—more importantly—shoulder function. In our opinion external rotation is the hallmark function of recovery in NBPP patients with Erb or Erb-plus lesions.2 In clinical practice, elbow flexion recovers practically in all non–surgically treated patients and in more than 90% of surgically treated patients. The most serious deficit affecting daily life is restricted shoulder function.

Outcome Analysis

The authors convert reported outcomes to outcome scores. They use the Mallet subscore of hand-to-mouth function as measurement of biceps recovery, while this Mallet subscore is at least also a measurement of external rotation and supination.7 Additionally, the proposed alignment of the British Medical Research Council (MRC) scale8 with The Hospital for Sick Children Active Movement Scale (AMS)9 seems incorrect. An MRC grade of 3 corresponds to motion against gravity, while an AMS grade of 4 corresponds to full motion with gravity eliminated. In the authors’ version table (Table 1 in their publication) MRC Grade 3 is graded as fair while AMS Grade 4 is graded as good. In our Table 1, the original description from the Mallet score, the MRC grading, and AMS score were inserted to show the discrepancies in their alignment of outcome scores.

Input Data From Surgical Papers

The authors identify in their literature search 17 papers from which they extracted the outcome of elbow flexion after surgery. These are represented in their Table 3. From this table, however, the reader cannot reproduce the data that the authors extracted from these papers. Such data, including ranges (that is, upper and lower bounds), should accompany base-case estimates of all input parameters for transparency of a utility model.11

Three of these papers report the outcome of nerve transfers (the authors’ references to Blaauw and Sloof, Kawabata et al., and Wellons et al.), and 1 paper concerns the outcome of end-to-side transfers (the reference to Pondaag and Gilbert). These surgical methods are usually used in cases of multiple root avulsions. Such serious lesion types do not represent the general population of NBPP patients. These 4 series contribute 76 patients to the total series of surgically treated patients, which might have influenced outcome estimates as input for their model.

The paper the authors cite by Chuang describes the results in adult patients, and not NBPP. In Table 3 the reference Lin and Lin5 should probably be replaced by Lin et al.6 In this specific series8 of 56 patients were treated by neurolysis, while in the methods sections neurolysis patients were said to be omitted from analysis. The paper Ali et al. cite by Kirjavainen et al. reports a mean Mallet score for shoulder function—not the hand-to-mouth subscore—and Gilbert elbow score for elbow flexion and extension. The way the authors estimate the amount of elbow flexion recovery as extracted from this paper is unclear, and in our opinion not possible. The same accounts for their reference to the Ashley et al. study; after re-reading this paper, it seems impossible to us to extract useful input data for the model used in this paper.

One of the cited papers was written by us on the recovery of external rotation.8 In this paper we reported functional external rotation as Mallet subscore, in addition to recovery of biceps force in MRC grade. It is not clear which of these 2 outcome measures was used in the authors’ analysis. The number of patients in our series was calculated as 69, which represents only C5–6 or C5–7 patients in our series; the 17 patients with more extended lesions were not counted. We, however, did not report the outcome of these groups separately. It is therefore unclear
how Ali et al. were able to assess the outcome of our Erb and Erb-plus patients only.

Additionally, the timing of surgical repair extracted from the surgical papers is an oversimplification. As we have the data from our own paper, we are able to illustrate this oversimplification. The mean age at surgery in our patient series was 5.3 months, which meant that our results were grouped by the authors within their “6 months” category.

In Fig. 1, we graphically represent the month in which the surgery actually took place. The figure shows that it is imprecise to label our patients’ age at surgery as 6 months; 49 of our 69 Erb/Erb-plus patients (71%) were surgically treated before 6 months of age. Another example is the patient series by Lin et al. (reference 55), which reports a mean age at surgery of 9.4 ± 2.1 months, and this paper was included in the “12 months” category.

Input Data From Nonsurgical Series

The nonsurgical series (42 patients) from the literature are difficult to interpret. The authors do not discuss the different biases these patient series have, which may have influenced the extracted estimates they use for their model.10 These estimates were not shown in their tables.

Experimental Data

The conclusion Ali et al. draw based on their model that surgery at a later age provides better results is intuitively incorrect. This should have evoked the idea that there might be something wrong with their models’ input data.11 Instead, the authors refer to one basic science paper that found that there is not much difference in a rat model between 2 and 6 months’ denervation time (the study by Rönkkö et al. referenced in their paper). Besides the notion that vast differences in nerve regeneration in adult and newborn rats or patients exist, the authors fail to report that the majority of experimental work shows that results of delayed nerve reconstruction decrease significantly over time. Besides the high-quality work that Ali et al. cite from Gordon’s group (the studies by Fu and Gordon referenced in their paper), we would like to point out that the vast majority of experimental models contradict the authors’ conclusion that later surgical repair results in better results.4,12

Although we applaud the intention of Ali et al. to set up a utility model to aid decision making in NBPP infants, we cannot conclude other than that their attempt failed. The patient series from both surgical and nonsurgical papers were seriously subject to bias, the way the input data were extracted from these papers is unclear or incomplete, the conversion of extracted data to outcome grade is inconsistent, and the resulting model cannot be explained at an intuitive level.11

This leaves no other choice than to discard the model and its conclusions. The only conclusion that can be drawn from the paper is that the current literature does not allow for pooling and meta-analysis of outcome data for NBPP patients.

Disclosure

The authors report no conflict of interest.

References


TABLE 1: Conversion table used by Ali et al., supplemented with the description in the original scoring systems

<table>
<thead>
<tr>
<th>Grade</th>
<th>Mallet</th>
<th>MRC</th>
<th>AMS</th>
</tr>
</thead>
<tbody>
<tr>
<td>Score</td>
<td>Definition</td>
<td>Score</td>
<td>Definition</td>
</tr>
<tr>
<td>good</td>
<td>V hand to mouth normal</td>
<td>5 normal</td>
<td>7 full motion against gravity</td>
</tr>
<tr>
<td></td>
<td>IV hand to mouth easy</td>
<td>4 against resistance</td>
<td>6 &gt; half range against gravity</td>
</tr>
<tr>
<td>fair</td>
<td>III hand to mouth w/ difficulty/ trumpet sign</td>
<td>3 against gravity</td>
<td>5 &lt; half range against gravity</td>
</tr>
<tr>
<td></td>
<td>II hand to mouth impossible</td>
<td>2 w/ gravity eliminated</td>
<td>4 full motion gravity eliminated</td>
</tr>
<tr>
<td>poor</td>
<td>I none</td>
<td>1 contraction w/o motion</td>
<td>2 &lt; half motion w/ gravity eliminated</td>
</tr>
<tr>
<td></td>
<td></td>
<td>0 no contraction</td>
<td>1 contraction, no motion</td>
</tr>
</tbody>
</table>

Fig. 1. Number of patients in each age category from our own series.