Neonatal brachial plexus palsy

TO THE EDITOR: We read with interest the article by Ali et al.¹ (Ali ZS, Bakar D, Li YR, et al: Utility of delayed surgical repair of neonatal brachial plexus palsy. Clinical article. *J Neurosurg Pediatr* 13:462–470, April 2014) that describes a utility model on the timing of surgery in neonatal brachial plexus palsy (NBPP).

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 data support 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.⁹ 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 grades good, fair, and poor. 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) scale³ with The Hospital for Sick Children Active Movement Scale (AMS)² 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' con-

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 series⁶ 8 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

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

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

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

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

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.

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Disclosure

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RESPONSE: We thank Drs. Pondaag and Malesy for their careful reading of our utility analysis of NBPP nerve repairs. Their group has meaningfully contributed to this heterogeneous literature, and their interest in our analysis is appreciated.

Drs. Pondaag and Malesy question whether the use of recovery of biceps function is an appropriate indicator of outcome in NBPP. We agree with their point that recovery of shoulder external rotation is a priority in NBPP repair. However, we chose elbow flexion as the primary outcome measure because this function has corresponding measures of utility, which shoulder abduction and external rotation do not, to our knowledge. In addition, current nerve repair strategies are limited in their successful recovery of shoulder abduction and particularly shoulder external rotation. In fact, orthopedic procedures are more commonly used to restore shoulder function^{3–6} and, therefore, shoulder function does not always represent success of nerve repair.

In our outcome analysis, we divided results into 3 categories: good, fair, and poor (Table 1 in our paper), based on the Mallet classification of brachial plexus palsy. Drs. Pondaag and Malesy question our alignment of outcome scores. First, we agree that the hand-to-mouth grade is not exclusively a measure of elbow flexion recovery, but in an attempt to align the variety of scales and scores used to

measure outcome, we labeled thresholds between good, fair, and poor identically in our assessment. This simplification is unfortunately necessary in order to consolidate the variety of outcome scales.

Drs. Pondaag and Malesy criticize our lack of inclusion of data extracted from our literature search, beyond what is already included in our contribution. We elected not to include all the means and ranges for every article (many of which have data for multiple time points), since this would make Table 3 incredibly complex and virtually unreadable. However, if a reader is so inclined to repeat our analyses, variance can be calculated from means and number of observations in each study, using the technique we cited.² The reference to the paper by Chuang in our paper does not accurately reflect the paper from which we extracted surgical results from NBPP surgery. The accurate reference is provided here.¹ We agree that the Lin and Lin reference in Table 3 should be replaced with the paper by Lin et al. Overall, while we recognize the small errors in data extraction that Drs. Pondaag and Malesy note, these do not invalidate our conclusions. In terms of timing of surgery, we included patients in the 3-, 6-, and 12-month time points if they had surgery leading up to those time points. Therefore, having surgery at 5.3 months was included in the 6-month time point.

With respect to the nonsurgical series, we agree with Drs. Pondaag and Malesy that biases and heterogeneity are common in this literature. We refer them to the last 2 paragraphs of our *Discussion*, in which we specifically address these limitations. In addition, we are unable to estimate the quantitative effect of these biases on extracted estimates, again making this a recognized limitation of the study.

Finally, we agree with Drs. Pondaag and Malesy that these results seem counterintuitive. However, even they agree that, “In clinical practice elbow flexion recovers practically in all non-surgically treated patients and in more than 90% of surgically treated patients.” If one accepts biceps recovery as the criterion of success, then, as they suggest, based on clinical practice, later surgery is clearly superior with respect to quality of life, since most infants recover flexion spontaneously. The appropriate timing of NBPP repairs remains a controversial topic, no doubt. However, the only way to answer this question rigorously is to conduct a prospective, multicenter randomized controlled trial comparing outcomes at different surgical time points. This type of trial of “early” versus “late” primary surgical repair is unlikely to occur due to lack of equipoise in management, parental preferences, and recruitment barriers. We feel that our decision analysis model is justified in utilizing quality-adjusted life years as a measure of NBPP burden to assess the value of primary surgical repair of upper trunk birth palsy at different time points. We emphasize that the goal of this contribution is to demonstrate that utility in offering “late” surgery exists. “Early” surgery may be overly aggressive, as we suggest, but may be justified, and, in fact, perhaps it may be better in certain cases. As we state in *Discussion*, “While our study provides a decision framework for the clinician to consider the treatment option that would yield the best QOL [quality of life], it should not substitute for

the individualized clinical decision making required to manage these patients effectively.”

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Challenges in identifying endoscopic third ventriculostomy

TO THE EDITOR: We read with interest the recent paper by Jernigan et al.² (Jernigan SC, Berry JG, Graham DA, et al: The comparative effectiveness of ventricular shunt placement versus endoscopic third ventriculostomy for initial treatment of hydrocephalus in infants. *Clinical article. J Neurosurg Pediatr* **13**:295–300, March 2014).

The authors used the Pediatric Health Information

System (PHIS) administrative database to garner a sizeable retrospective cohort for analysis. They aimed to compare the effectiveness of endoscopic third ventriculostomy (ETV) and shunt placement for hydrocephalus treatment in infants younger than 1 year in a multicenter database. They claim to have identified 4544 patients who underwent shunt placement and 872 patients who underwent ETV treatment. We are not aware of ICD-9-CM codes that specifically identify ETV. We were excited and supportive about the research possibilities of this prospect.

The authors state that patients undergoing ETV were identified by the “ICD-9-CM procedural code for ventriculostomy (02.2) during hospital admission in the absence of billing charges for implanted ventricular catheters, reservoirs, or shunts suggestive of external ventricular drain placement or ventriculosubgaleal drain placement.”²

We were particularly intrigued about the concept of identifying ETV based on “ventriculostomy” in the absence of the aforementioned hardware billing charges. As our institution is a PHIS site, we explored the validity of this approach. We compared our own institution's surgeon-entered neurosurgery database to cases reported as code 02.2 in the PHIS consortium database. We found low reliability of the 02.2 code in representing ETV surgeries. We used the Texas Children's Hospital (TCH) surgeon-verified neurosurgery database as the standard; comparing TCH internal data to the TCH subset in PHIS, we checked if patients identified with the code 02.2 but not charged for shunts, ventricular catheters, or reservoirs in the PHIS database were truly ETV patients. Below are our findings.

Between October 1, 2010, and September 30, 2013, 64 patients (A in Table 1) underwent ETV and were identified by TCH medical records. Meanwhile, 126 patients (B in Table 1) underwent external ventricular drainage (EVD). Among these patients, 13 patients underwent both ETV and EVD.

In the same time frame, we found 37 patients in the PHIS (TCH subset) with ICD-9-CM procedure codes 02.2, 02.21, and 02.22, and without billing charges for shunts, ventricular catheters, or reservoirs. According to the study by Jernigan et al., these patients would have been assigned as ETV patients. However, using their algorithm, only 32.8% of the 64 patients who had ETV

TABLE 1: Number of cases in each category*

Variable	ETV in TCH Database (A)	EVD in TCH Database (B)	Codes 02.2, 02.21, 02.22 w/o Listed Charges (C)†	Codes 02.2, 02.21, 02.22 in PHIS (D)
no. of cases	64	126	37	133
overlap in A & B	13	13		
overlap in A & D	40			40
overlap in B & D		41		41
overlap in A & C, not B	17		17	
overlap in A & B & C	4	4	4	
overlap in B & C, not A		11	11	
in C, not A or B			5	

* Time period of cases examined: October 1, 2010, to September 30, 2013.

† Listed charges: shunts, ventricular catheters, or reservoirs.

Fig. 1. Composition of the 37 patients assigned as "ETV" by query from PHIS data set.

surgeries at TCH were accurately identified (Fig. 1). Of the 37 patients labeled by the authors' algorithm as ETV patients using the PHIS database, we verified that only 21 of these patients indeed had ETV surgery (56.8%). Eleven of these 37 patients (29.7%) actually underwent external ventricular drain placement without ETV surgeries. In a previous paper by the same group,¹ the authors reasoned that infants are less likely to have external ventricular drain placement, so the ventriculostomy code would be more reliable. The TCH data set does not show this to be the case. We evaluated infants younger than 12 months as a subgroup and found that this age group distinction would not help identify patients with ETV any more precisely: among 17 ETV patients, 7 were younger than 1 year and 10 patients were 1 year or older; among 4 patients who underwent both ETV and EVD, 1 was younger than 1 year and 3 patients were 1 year or older; and among 11 EVD patients, 3 were younger than 1 year and 8 patients were 1 year or older.

Overall, Jernigan's group lays out an algorithm that yields a 32.8% sensitivity, 88.0% specificity, and 56.7%

positive predictive value in correctly identifying ETV surgery in our attempts at subset validation (Table 2). The authors state that they "internally validated the PHIS patients from Boston Children's Hospital by using our internal departmental database to ensure that our patient collection algorithm was both sensitive and specific."² It would be interesting to see how coding practices and clinical practices vary.

We support developing research using administrative data and remain cognizant about the importance of recognizing its limitations. The ability to identify ETV with ICD-9 coding in this way appears to be a notable limitation. Based on review of our own institution's clinical data compared with our institution's contribution to the multi-center PHIS database used by the authors in their study, their outlined algorithm definition of presumed ETV appears to misidentify the patient cohort and would alter the study results. The conclusions must thus be evaluated with caution; the study's ability to address comparative effectiveness as labeled is less convincing.

Further studies into the quality of administrative data

TABLE 2: Sensitivity and specificity analysis*

		Condition Verified by TCH Medical Records		
		True ETV Cases at TCH	Not ETV Cases	
Condition identified through Jernigan et al. algorithm	ETV cases	21 (TP)	16 (FP)	positive predictive value = $TP(21) / (TP(21) + FP(16)) = 56.8\%$
	not ETV cases	43 (FN)	117 (TN)	
		sensitivity = $TP(21) / (TP(21) + FN(43)) = 32.8\%$	specificity = $TN(117) / (FP(16) + TN(117)) = 88.0\%$	

* Time period of cases examined: October 1, 2010, to September 30, 2013. FN = false negative; FP = false positive; TN = true negative; TP = true positive.

and the ability to draw meaningful clinical conclusions from such data sets are warranted. The size of these data is undeniable. We applaud these authors' efforts and also realize there is more work to be done. Together, our field can better understand the strengths and limitations of such data.

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