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High prevalence of early language delay exists among toddlers with neonatal brachial plexus palsy

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Abstract

AIM—Association of language impairment with neonatal brachial plexus palsy (NBPP) has not been reported in the literature. The current treatment paradigm for NBPP focuses on upper extremity motor recovery with little formal assessment of other aspects of development, such as language. We performed a cross-sectional pilot study to investigate early language delay prevalence in toddlers with NBPP and potential NBPP-related factors involved.

METHOD—Twenty toddlers with NBPP were consecutively recruited (12 males, 8 females; mean age 30 mos). Preschool Language Scale Score (4th edition), demographics, and socioeconomic status were collected. NBPP-related factors such as palsy side, treatment type, Narakas grade, muscle MRC score, and Raimondi hand score were reported. Student *t* test, chi-square, or Fisher exact test were applied. Statistical significance level was established at $p < 0.05$.

RESULTS—Of study participants, 30% were diagnosed with language delay, while the prevalence of language delay in the population with normal development in this age range was approximately 5–15%.

INTERPRETATION—We observed high language delay prevalence among toddlers with NBPP. Although our subject sample is small, our findings warrant further study of this phenomenon. Early identification and timely intervention based on type of language impairment may be critical for improving communication outcome in this population.

Keywords

Brachial plexus injury; brachial plexus palsy; language delay; neonate

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Speech-language development during the period from 24 to 36 months of age is an important indicator of a toddler's global development and cognitive function. Early language impairment can lead to other developmental impairments and affect future school performance.¹ The prevalence of language delay among preschool children (2 to 4.5 years old) is approximately 5% to 8%²; for children at age 36 months, the incidence is approximately 14%.³ Early identification of children with language delay is necessary for timely treatment. Similarly, recognizing perinatal conditions that are associated with early language delay may facilitate the identification and treatment of children at risk for language delay. Potential risk factors for language delay include physical impairment, birth deficits, and environmental factors such as family socioeconomic status and parent education level.^{4,5} Of interest with regard to our research study, investigators have reported an association between language delay and motor impairments in school-aged children.⁶⁻⁸

In treating patients with neonatal brachial plexus palsy (NBPP) for over 30 years, our senior clinician has anecdotally observed a higher prevalence of language delay among NBPP patients than in the general population within the 24- to 36-month age group. NBPP manifests as paralysis/paresis and loss of sensation in the affected arm in approximately 0.4-2.6 per 1000 live births^{9,10}; the condition results from stretching of the nerves of the brachial plexus in the perinatal period. Previous studies showed NBPP might be associated with hand preference and language lateralization^{11,12}; we hypothesized that partial or whole arm function loss might interfere with language development and be associated with language delay. No studies have reported a potential association of unilateral motor impairment in NBPP with language delay. Therefore, we assessed the prevalence of early language delay associated with NBPP and attempted to identify any NBPP characteristics that may also be associated with language delay. The contribution of this new knowledge could result in improved clinical treatment paradigms for NBPP in the context of childhood development.

METHODS

Participants

In this observational study, 20 toddlers with NBPP (12 males and 8 females) were consecutively recruited from the University of Michigan Interdisciplinary Brachial Plexus Program from May 2012 to July 2013. All participants had passed hearing screening. Developmental screening was done by physiatrists and those with delays in areas other than speech or related to arm function were excluded from the study population. Inclusion criteria included patients 24 to 36 months of age with a diagnosis of NBPP without other comorbid diagnoses, from monolingual English-speaking only families. In order to control biological or physical risk factors that could possibly lead to language delay, patient medical records were reviewed before recruitment. Infants and toddlers that were preterm, had low birth weight (<1,500 g), low or unknown Apgar score, or that had other physical impairments not associated with NBPP were excluded from recruitment. Once parents of NBPP patients consented, they were asked to complete a questionnaire regarding demographics (child's and main parent's age, gender, ethnicity, and family history of language delay) and socioeconomic status (main parent's marital status, parents' highest education level, and

family's health insurance). Study protocol was approved by the University of Michigan Institutional Review Board.

Data Collection

A single board-certified speech-language pathologist administered the Preschool Language Scale, 4th edition (PLS-4), to assess each patient's speech-language development. The PLS-4 is a norm-referenced standardized measurement for identifying children from birth through 6 years 11 months who have a language disorder or delay.¹³⁻¹⁵ The test targets receptive and expressive language skills in the areas of attention, play, gesture, vocal development, social communication, vocabulary, concepts, language structure, integrative language skills, and phonological awareness. Administration and scoring time is 20-45 minutes. The PLS-4 provides age-based standard scores, percentile ranks, and age equivalents for the auditory comprehension and expressive communication subscales, as well as a total language score. Scales are normalized at 100, with a standard deviation of 15. PLS-4 reliability and validity have been established in previous studies for preschool children.¹⁵

NBPP-related characteristics, such as palsy side, treatment type, Narakas grade, Raimondi hand score, and deltoid and biceps power (expressed as British Medical Research Council [MRC] score) were evaluated by 1 of 2 occupational therapists using standard measurement protocol. The Narakas classification is an accepted measure of the extensiveness of NBPP based on the number of affected nerve roots of the brachial plexus, and the Narakas grade expresses the clinical continuum of NBPP.^{16,17} The classification grades range from I to IV, with I as the least extensive palsy (C5-C6) and IV as the most extensive palsy (flail extremity and Horner's syndrome, involving C5-T1). Raimondi hand score is used to evaluate hand function; scores range from 0 to 5, with 0 representing complete paralysis and 5 representing excellent function. MRC score is designed to measure muscle power with motor strength graded from 0 (no movement) to 5 (normal strength).

Statistical analysis

Participant and parent demographics were summarized using descriptive statistics. Numerical variables (age, PLS scores) were compared among delayed and non- delayed groups by Student *t* test and categorical variables by chi-square or Fisher exact test. Logistic regression was applied to analyze factors associated with language impairment. Analyses were performed using SPSS software, version 20 (SPSS Inc., Chicago, IL), with statistical significance level established at $p < 0.05$.

RESULTS

During the study period, 53 toddlers with NBPP were within the 24- to 36-month age range. After initial screening using the exclusion criteria (bilingual families, preterm infants and toddlers, low birth weight, low/unknown Apgar score, or other physical impairments not associated with NBPP), 40 toddlers with NBPP were eligible for the study and a recruitment letter was sent to the families. Twenty families declined study participation because of

commute or no interest in the study. A total of 20 toddlers with NBPP (12 males, and 8 females) were included in the study.

Thirty percent of toddlers with NBPP studied were diagnosed with language delay, approximately twice the incidence of normally developed, age-matched children. Overall, mean age of the toddlers was 30 months (SD, 4 mos), with the majority being white (65%) and male (60%). Mean parent age was 36 years (SD, 11 yrs), with the majority being white (70%), female (85%), and married (50%). There was a trend in parent education level among delayed and non-delayed groups, but with no statistical significance. Two families did not report father's educational level. Essentially, most mothers in the delayed group had a high school diploma (50%), while 43% of mothers in the non-delayed group had a college degree. Only 2 (10%) families reported a family history of language delay, and most families had Medicaid as their primary insurance (55%). As for NBPP-related factors, palsy side was evenly distributed with 1 bilateral participant, and only 15% of patients were treated by surgical intervention. Overall, most children had good arm function with MRC scores of III-IV for deltoid (80%) and biceps (85%), and a Raimondi score of III-V (90%). Approximately half of the children had less-extensive Narakas I-II grade (55%); however, the delayed group tended to have a higher percentage of more extensive Narakas grade (67%). No significant difference exists in demographics, socioeconomic status, or NBPP-related factors between delayed and non-delayed groups (Tables I and II).

Among toddlers with language delay, the PLS-4 age-based total language standard score is 76 (SD = 9), with receptive score of 77 (SD = 7) and expressive score of 79 (SD = 11) (Figure 1). One child had difficulty in receptive language only, 1 in expressive language only, and 4 in both receptive and expressive language skills. The level of impairment was mild to moderate (2 in each category: mild, mild-moderate, moderate). All patients with language delay were referred for speech-language pathology treatment after evaluation.

Logistic regression of the available data regarding demographics, socioeconomic, or NBPP-related factors did not reveal any single factor that was associated with language delay in toddlers with NBPP (Table III).

DISCUSSION

Our study provides the first evidence that a significantly higher prevalence of language delay exists in toddlers with NBPP. Current literature regarding children 2 to 4.5 years old reports the prevalence of language delay as 5-8%; the incidence for children at age 36 months is 14-15.6%.² Although earlier studies¹⁸ suggest preschool children with normal cognitive function who suffer isolated delay in language development usually reach normal language skills later, a more recent study shows that children with isolated language delay without observed cognitive impairments or hearing loss have higher risk of developmental difficulties by age 7 to 8 years.¹⁹ Previous studies have also reported an association between language and motor impairments in school-aged children.⁶⁻⁸ Webster et al.⁸ investigated motor function in children with a preschool diagnosis of language delay and concluded that motor impairment is an important comorbidity in language delay; they hypothesized that

factors common to motor function and to communication (for example, sequencing, motor planning, and timing) might be associated with language impairment.

Even in the context of the seemingly isolated condition of NBPP, global development of a child must be considered. Reported conditions associated with NBPP range from neurologic palsies (phrenic nerve palsy, facial nerve palsy, central developmental disability) to musculoskeletal disorders (torticollis, skeletal fractures), and increasingly, practitioners treating NBPP are considering this disorder as part of the spectrum of perinatal injuries.²⁰⁻²⁴ As our study supports language delay as yet another condition associated with higher prevalence of language delay, early assessment can result in timely treatment via early referrals for formal language evaluation. Children who have difficulties with oral language are at significant risk for later academic and social problems.^{25,26} In general, the period from 24 to 36 months is critical for detection of early language problems, while the most appropriate age and tool for language assessment remains controversial.

No report exists on the relationship of affected side and hand preference with language development in toddlers with NBPP. In most adult and pediatric populations, right limb preference is approximately 90%,²⁷ and studies show right-handedness and language lateralization are mostly controlled by the left hemisphere.^{28,29} In our early investigation on limb preference in children with NBPP, we determined that, unlike the general population with 90% right arm preference, only 17% of children affected by right-sided NBPP expressed right arm preference, whereas children with left-sided NBPP did not significantly differ from the general population in upper limb preference.¹² Our results indicated a potential limb preference alteration because of NBPP, which could potentially affect language lateralization that is also controlled by the left hemisphere. Additionally, Auer et al.¹¹ reported in a small series of 15 NBPP patients that the severity of right arm paresis and hand usage dysfunction correlated with language development and the degree of left-to-right hemisphere shift of language lateralization. The above studies provide supportive evidence of the association of NBPP with hand preference and language development.

This study is the first attempt to prospectively evaluate language function in children with NBPP in an attempt to identify NBPP-specific risk factors that might be associated with language delay. Our study demonstrated a trend toward higher Narakas grades in the delayed group (67%) than in the non-delayed group (36%). Although none of our study patients had concurrent phrenic nerve palsy, toddlers with NBPP can present with diaphragmatic paralysis, and an association between respiratory muscle disturbance and early development has been reported.^{21,30} We also hypothesized that children with more extensive NBPP would use their affected arm/hand less during developmental stages. Our results showed that most toddlers with language delay had good outcomes in triceps and biceps MRC and Raimondi hand scores; however, it was not clear whether these patients utilize their affected hand for communication as they mature. Hand usage and gestures as part of the motor system have proven to be highly correlated with comprehension and production during language development.³¹ By pointing, manipulating objects, or mimicking parents' actions, toddlers learn how to communicate even before they are able to express themselves verbally. Therefore, any interruption in hand/arm usage during stages of language development could possibly influence communicative function. Parents and

physicians should pay close attention to how toddlers with NBPP coordinate their body language with verbal language at early stages. A routine language screening for toddlers with NBPP at 12, 24, and 36 months of age might be beneficial for early identification and timely intervention.

Other factors that could also be associated with language development can be categorized into biological and environmental risk factors.⁴ Biological risk factors refer to those associated with patient physical status such as preterm, low birth weight, low Apgar score, or physical impairment. Low socioeconomic status, low maternal education level, single parent, or adolescent mother, and families lacking social/health support are common environmental factors that may place children at increased risk for language problems.³ To reduce bias, we controlled for biological risk factors (preterm, birth weight, Apgar score, or other physical impairment) during recruitment, and we included environmental factors (socioeconomic status, maternal education, maternal status, and insurance) for analysis. Even though none of the environmental risk factors were associated with language impairment in this study, our results were consistent with a larger sample study that found that parental educational levels and socioeconomic status were not associated with late language emergence in 24-month-old toddlers.³² However, other studies report that male gender, family history of language impairment, low maternal education, and socioeconomic status are known indicators for language delay in toddlers.³⁻⁵ Correspondingly, our study reported more college-educated mothers in the non-delayed group (43%) than in the delayed group (33%), supporting evidence suggesting that maternal educational level is highly correlated with environmental factors that may influence a child's language development, including home environment, household income, health, nutrition, and language stimulation.^{33,34} Deprivation in any of these areas could potentially affect the parents' language input to children. Our results suggest a trend that mothers, as the main caretakers, have direct and instant influence on a child's language development, and that lower educational level might suggest lack of resources in the household that could plausibly disrupt the language development process for toddlers with NBPP. While screening NBPP patients for language delay, physicians should also take into account these factors when interpreting results and planning suitable interventions.

Limitations and future studies

Several limitations exist in this study. There might be self-selection bias, in that subjects who volunteer to participate in the study may systematically differ from those who are qualified to participate but do not volunteer. Furthermore, small sample size might limit our ability to detect the overall prevalence of language delay in all qualified NBPP patients. There may be more patients with language delay who were not detected in this study, resulting in reduced significance of environmental risk factors on language impairment. Additionally, there might be other biological and environmental confounders that could also affect the study results. However, we considered the possible risk factors reported in the literature, and minimized potential bias by controlling for biological risk factors during recruitment by including environmental risk factors in the analysis.

In conclusion, the purpose of this study is to inform families and physicians of the higher prevalence of language delay in toddlers with NBPP in the context of child development. As knowledge regarding the general developmental natural history of NBPP is significantly limited by the lack of studies on affected patients followed from birth to maturity, our study contributes a potential novel change in the treatment paradigm for toddlers with NBPP to include language evaluation. Future research using a population-based database to further investigate the physiological/anatomical basis underlying the associative or potentially causative relationship between these two conditions may be warranted.

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WHAT THIS PAPER ADDS

- High prevalence of language delay exists in NBPP population.
- Early language delay identification and timely interventions are crucial.
- Identifying body and verbal language coordination at early stages improves outcomes.
- Biological and environmental language delay risk factors should be considered.
- Contributes improved clinical treatment paradigms for NBPP.

Figure 1.

Language evaluation using the Preschool Language Scale, 4th edition (PLS-4), to assess patient speech-language development. Student *t* test was applied for group comparison; *p*-value less than 0.001 for delay and non-delay comparisons.

Table I

Patient and parent/caretaker demographics

Parameter	All (N = 20)	Delay (n = 6; 30%)	Non-Delay (n = 14; 70%)	p-Value
Patient age, mos (SD) ^a	30 (4)	30 (4)	30 (4)	0.89
Patient gender ^b				0.33
Male	12 (60%)	5 (83%)	7 (50%)	
Female	8 (40%)	1 (17%)	7 (50%)	
Patient race ^b				0.39
White	13 (65%)	4 (67%)	9 (64%)	
Black	5 (25%)	1 (17%)	4 (29%)	
Hispanic	1 (5%)	--	1 (7%)	
Other	1 (5%)	1 (17%)	--	
Caretaker age, yrs (SD) ^a	36 (11)	36 (10)	35 (11)	0.72
Caretaker gender ^b				0.86
Male	3 (15%)	1 (17%)	2 (14%)	
Female	17 (85%)	5 (83%)	12 (86%)	
Caretaker race ^b				0.68
White	14 (70%)	5 (83%)	9 (64%)	
Black	5 (25%)	1 (17%)	4 (29%)	
Hispanic	1 (5%)	--	1 (7%)	
Caretaker marital status ^b				0.59
Single	8 (40%)	3 (50%)	5 (36%)	
Married	10 (50%)	3 (50%)	7 (50%)	
Divorced	2 (10%)	--	2 (14%)	
Mother's education ^b				0.37
Some high school	1 (5%)	--	1 (7%)	
High school graduate	5 (25%)	3 (50%)	2 (14%)	
Tech school or some college	6 (30%)	1 (17%)	5 (36%)	
College graduate	8 (40%)	2 (33%)	6 (43%)	
Father's education ^{b, c}				0.79
High school graduate	7 (39%)	2 (33%)	5 (42%)	
Tech school or some college	4 (22%)	1 (17%)	3 (25%)	

Parameter	All (N = 20)	Delay (n = 6; 30%)	Non-Delay (n = 14; 70%)	<i>p</i> -Value
College graduate	7 (39%)	3 (50%)	4 (33%)	
Insurance ^b				0.85
Medicaid	11 (55%)	3 (50%)	8 (57%)	
Private insurance	4 (20%)	1 (17%)	3 (21%)	
Other	5 (25%)	2 (33%)	3 (21%)	
Family history of language delay ^b				0.52
Yes, brother	2 (10%)	1 (17%)	1 (7%)	

Delay = language impairment

^aData presented as: mean (SD); Student *t* test was applied for group comparison.

^bChi-square test or Fisher exact test were applied for group comparison.

^cTwo families failed to report father's level of education.

Table II

Results of patient physical evaluation

Parameter	All (N = 20)	Delay (n = 6; 30%)	Non- Delay (n = 14; 70%)	p-Value
Palsy side ^a				0.79
Right	10 (50%)	3 (50%)	7 (50%)	
Left	9 (45%)	3 (50%)	6 (43%)	
Bilateral	1 (5%)	-	1 (7%)	
Treatment type ^a				0.37
Therapy alone	17 (85%)	6 (100%)	11 (79%)	
Nerve surgery	2 (10%)	-	2 (14%)	
Shoulder surgery	1 (5%)	-	1 (7%)	
Narakas grade ^{a, b}				0.34
I-II	11 (55%)	2 (33%)	9 (64%)	
III-IV	9 (45%)	4 (67%)	5 (36%)	
Deltoid MRC score ^{a, c}				0.99
I-II	4 (20%)	1 (17%)	3 (21%)	
III-V	16 (80%)	5 (83%)	11 (79%)	
Biceps MRC score ^{a, c}				0.20
I-II	3 (15%)	2 (33%)	1 (7%)	
III-V	17 (85%)	4 (67%)	13 (93%)	
Raimondi hand score ^{a, d}				0.52
I-II	2 (10%)	1 (17%)	1 (7%)	
III-V	18 (90%)	5 (83%)	13 (93%)	

Delay = language impairment; MRC = Medical Research Council; NBPP = neonatal brachial plexus palsy

^a Chi-square test or Fisher exact test were applied for group comparison.

^b The Narakas classification is an accepted measure of the nerve involvement levels of NBPP based on neurological involvement and clinical examination. Scores range from I to IV with I being the mildest and IV being the most involved.

^c The deltoid and biceps function MRC score indicates muscle strength of NBPP patients. A score less than III is widely accepted as non-functional.

^d Raimondi hand score is used to evaluate hand function; scores range from 0 to 5 with 0 representing complete paralysis and 5 representing excellent function.

Table III

Logistic regression results for patients and caretakers

Parameter	Odds Ratio	95% Confidence Interval	p-Value
Patient gender (ref., female)	10	0.002 to 27	0.09
Caretaker age	1.01	0.88 to 1.15	0.92
Caretaker gender (ref., female)	1.13	0.001 to 64	0.96
Caretaker marital status, married (ref., single)	0.99	0.02 to 26	0.99
Family history, yes (ref., no)	2.70	0.04 to 56	0.64
Injury side, left (ref., right)	1.00	0.13 to 15.08	0.79
Narakas grade (ref., III-IV)	0.28	0.03 to 9.29	0.21
Medical Research Council score (ref., III-V)	0.15	0.01 to 1.42	0.17