

Motor Severity in Children With Cerebral Palsy Studied in a High-Resource and Low-Resource Country



WHAT'S KNOWN ON THIS SUBJECT: There is variability in cerebral palsy prevalence estimates in low-resource countries, related to definitions, detection of milder cases, diagnosis age, and adequate training for clinicians. Thus, differences in prevalence and motor patterns between high- and low-resource countries remain unclear.



WHAT THIS STUDY ADDS: There were more children with dystonia and less with spasticity in Bangladesh compared with Australia (cerebral palsy diagnosis/motor classifications were consistent between settings). Differences in motor patterns between high- and low-resource countries have profound implications for early detection and appropriate interventions.

abstract



OBJECTIVES: To compare the patterns of motor type and gross motor functional severity in preschool-aged children with cerebral palsy (CP) in Bangladesh and Australia.

METHODS: We used comparison of 2 prospective studies. A total of 300 children with CP were aged 18 to 36 months, 219 Australian children (mean age, 26.6 months; 141 males) recruited through tertiary and community services, and 81 clinic-attendees born in Bangladesh (mean age, 27.5 months; 50 males). All children had diagnosis confirmed by an Australian physician, and birth and developmental history collected on the Physician Checklist. All children were classified by the same raters between countries using the Gross Motor Function Classification System (GMFCS), and motor type and distribution.

RESULTS: There were more children from GMFCS I–II in the Australian sample (GMFCS I, $P < .01$; III, $P < .01$; V, $P = .03$). The patterns of motor type also differed significantly with more spasticity and less dyskinetic types in the Australian sample (spasticity, $P < .01$; dystonia, $P < .01$; athetosis, $P < .01$). Birth risk factors were more common in the Bangladesh sample, with risk factors of low Apgar scores (Australia, $P < .01$), lethargy/seizures (Australia, $P = .01$), and term birth (Bangladesh, $P = .03$) associated with poorer gross motor function. Cognitive impairments were significantly more common in the Bangladesh children ($P < .01$), and visual impairments more common in Australia ($P < .01$).

CONCLUSIONS: Patterns of functional severity, motor type, comorbidities, etiology, and environmental risk factors differed markedly between settings. Our results contribute to understanding the patterns of CP in low-resource settings, and may assist in optimizing service delivery and prioritizing appropriate early interventions for children with CP in these settings. *Pediatrics* 2014;134:e1594–e1602

AUTHORS: Katherine A. Benfer, MPH, BSpPath,^{a,b} Rachel Jordan, BPT,^a Sasaka Bandaranayake, MBBS,^c Christine Finn, BPT,^a Robert S. Ware, PhD,^{d,e} and Roslyn N. Boyd, PhD, MSc, P(Biomech), BSc (Anatomy), BAppSc (Physio)^a

^aQueensland Cerebral Palsy and Rehabilitation Research Centre, The School of Medicine, ^dSchool of Population Health, and ^eQueensland Children's Medical Research Institute, The University of Queensland, Brisbane, Australia; ^bCentre for the Rehabilitation of the Paralyzed, Dhaka, Bangladesh; and ^cQueensland Paediatric Rehabilitation Service, Royal Children's Hospital, Brisbane, Australia

KEY WORDS

gross motor severity, motor type, cerebral palsy, high-resource country, low-resource country

ABBREVIATIONS

CP—cerebral palsy

CRP—Centre for the Rehabilitation of the Paralyzed

GMFCS—Gross Motor Function Classification System

OR—odds ratio

Ms Benfer designed the modifications to the Australian study for the Bangladesh context, collected the primary data in country, analyzed and interpreted the data, and drafted the manuscript; Ms Jordan completed the gross motor ratings, analyzed and interpreted the data, and drafted the manuscript; Dr Bandaranayake assisted in the modification of the protocol to the Bangladesh context, confirmed children's diagnosis and motor type, interpreted the data, and provided critical review of the manuscript; Ms Finn completed the gross motor ratings, interpreted the data, and provided critical review of the manuscript; Dr Ware advised on statistical design of the studies and the statistical analysis of the manuscript and provided critical review of the manuscript; Prof Boyd conceptualized the Australian study, assisted in the modification of the protocol to the Bangladesh context, secured funding for the Australian study, assisted in the interpretation of data, provided editorial support for the drafting of the manuscript, and provided study supervision; and all authors approved the final manuscript as submitted. All authors agree to be accountable for all aspects of the work to ensure its accuracy and integrity.

This trial has been registered with the ANZTR Register (identifier 1261200169820).

www.pediatrics.org/cgi/doi/10.1542/peds.2014-1926

doi:10.1542/peds.2014-1926

Accepted for publication Sep 8, 2014

Address correspondence to Katherine A. Benfer, MPH, BSpPath, Queensland Cerebral Palsy and Rehabilitation Research Centre, Department of Paediatrics and Child Health, Level 7, Block 6, Royal Brisbane & Women's Hospital, Herston Queensland, 4029. E-mail: katherine.benfer@uqconnect.edu.au

(Continued on last page)

Cerebral palsy (CP) is the most commonly occurring childhood physical disability,¹ with an overwhelming majority of its global burden in low-resource countries.² It has been estimated that 80% of the global prevalence of CP is in low-resource countries, having larger populations and potentially greater incidence rates.^{2,3} Children who have a disability and their families living in low-resource countries are among the most disadvantaged in their community, with a bidirectional link between disability and poverty.² Bangladesh is a small but densely populated country in the Indian subcontinent (~150 million people, 150 000 km² land area). Almost a third live in extreme poverty (GDP per capita = US\$ 752)⁴ and ~45% of children aged <5 years have chronic malnutrition.⁵ Australia, in direct contrast, is a large but sparsely populated country (22 million people, land mass of 7.7 million km²)⁶ and a major global economy (GDP per capita = US\$ 67 442).^{4,7}

Over the past decade, there have been a number of efforts to standardize the diagnosis of CP and motor type classification among western high-resource countries.^{8–10} The prevalence of CP from various high-resource countries has been estimated at 2.0/1000 live births, and this has remained relatively stable throughout recent decades despite advances in medical practices.¹¹ Spasticity is typically cited as the predominant motor type, occurring in 77% to 93% of CP cases identified by a recent review, dyskinesia in 2% to 15%, and ataxia in 2% to 8%.¹¹ Of the 86.5% of individuals classified with spasticity in the Australian CP Register Report, 38.8% had hemiplegia, 37.5% diplegia, and 23.7% tri/quadruplegia.¹²

In low-resource countries there continues to be large variability in prevalence estimates, related to CP definitions, ability to detect milder cases, age at diagnosis (with CP prevalence influenced by survival rates), and adequate training for

health staff.³ Three population-based studies in low-resource countries have estimated prevalence as low as 1.6/1000 in urban China,¹³ 2.8/1000 in India (4.4/1000 in children aged <4 years),¹⁴ and as high as 4.0/1000 in Bangladesh (29.0/1000 in Dhaka district).¹⁵ Studies of clinic attendees in these settings have reported high rates of spasticity similar to that in high-resource countries, from 70% to 90%.^{14,16–20} Studies have tended to identify higher rates of quadriplegia than those reported in the west (60% to 86%),^{16,17,20} although the only population-based study found a high rate of spastic diplegia (72.9%).¹⁴

Differences in prevalence and motor patterns between high- and low-resource countries reported in the literature remain unclear; however, the etiology has been reported to differ markedly. Owing to improved medical care in high-resource countries, it is now thought that birth asphyxia accounts for only 6% to 8% of CP cases,¹ with an increased proportion of preterm births (45%).¹² In low-resource countries there is poor survival of preterm infants, and home deliveries by unskilled birth attendants continue to dominate.¹⁷ Birth asphyxia and low birth weight are reported as the prevailing causes of CP in low-resource countries,^{16,17} along with kernicterus and postnatal causes such as meningitis and cerebral malaria.^{3,14}

The motor outcomes of children in high-resource countries have been well described based on their level of gross motor function (Gross Motor Function Classification System [GMFCS]). Motor outcomes are impacted by a wide array of factors, including intrinsic child characteristics, family dynamics and functioning, and availability, access, and options for interventions.²¹ Despite these many influences, gross motor functional development in western children who have CP has been shown to follow predictable patterns (along motor curves) based on the child's

overall motor severity.²² Less is known about the role of these environmental factors on motor outcomes in low-resource settings, where children may be in poverty, with less family knowledge and fewer resources to support their child's development, cultural differences in parental interaction style, and lower/delayed access to health services.^{21,23}

Owing to these differences in neonatal risk factors and environmental influences between high- and low-resource countries, the severity and motor patterns of children who have CP in these 2 contexts is thought to differ. This study is the first to our knowledge to explore 2 cohorts of children who have CP in high- and low-resource settings using the same diagnostic and classification methods for each. It also aims to document differences in motor outcomes between settings with reference to gross motor function and motor type. This study will enhance our understanding of risk factors for CP and associated motor outcomes as well as contributing information to understand primary prevention priorities, and providing health ministries with data to plan optimal services.

METHODS

This article compares 2 cross-sectional prospective studies of children who have CP aged 18 to 36 months. The first sample is a cohort of children born in Queensland, Australia, and the second is a sample of clinic attendees residing in Bangladesh. The Australian data represent a subset of children from 2 larger longitudinal studies, Queensland CP Child Motor and Brain Development (National Health and Medical Research Council 465128)²⁴ and Queensland CP child: Growth, Nutrition and Physical Activity (National Health and Medical Research Council 569605).²⁵ It includes only initial assessments of children aged 18 to 36 months seen between January 1, 2009 and March 31, 2013.

Patients

Participants in Queensland were referred to the study through a range of settings from parent referral to community and tertiary care. All children who had a confirmed diagnosis of CP,⁹ aged 18 to 36 months corrected age at initial assessment, and born in Queensland between 2006 and 2009, were invited to participate. Children who had neurodegenerative conditions were excluded.^{24,25}

The Bangladesh sample was recruited through a national rehabilitation center in Bangladesh, the Centre for the Rehabilitation of the Paralyzed (CRP). The center provides services to children who have CP residing in all regions of Bangladesh as outpatients, or through a 2-week inpatient program. The inpatient program provides parent education and training, as well as individual and group therapy. Admission is not associated with illness or medical intervention. All children aged 18 to 36 months who had a confirmed diagnosis of CP attending the center from August to December 2013 were invited to participate. Children who attended as inpatients were prioritized to enable a battery of measurements to be completed (for the larger study).

Procedures

For the Australian cohort, children attended the hospital for a diagnostic appointment with a pediatrician or child neurologist. During this appointment, diagnosis was confirmed based on published guidelines, and a detailed clinical history was taken. Children's motor type/distribution and GMFCS level were classified by 2 independent clinicians (pediatric rehabilitation physician, and an experienced physiotherapist).

In Bangladesh, children attended an initial diagnostic appointment with the primary investigator (KB) and a local pediatric physician, who collected the clinical history from the mother (in Bengali) and provided a preliminary

diagnosis of CP. The written case history and a short video of the child performing functional motor tasks (including lying, rolling, sitting, standing, walking, and transitions between these) were sent to the Australian research team to provide an independent and consistent confirmation of CP diagnosis, motor type/distribution, and GMFCS.

Measures

The child's clinical history was collected by using the Physician Checklist (Supplemental Information A). This was administered by physicians to parents using open-ended questions to gather information on the child's clinical presentation, birth history, comorbidities, and development. This checklist was developed in 2003 for the Australian CP Child Study,²⁴ and was intended as a standardized physician checklist for gathering clinical history, rather than an exhaustive list of causes. Physicians made a judgment from the clinical history regarding factors potentially associated with a diagnosis of CP. Minor modifications were made to this checklist for Bangladesh (Supplemental Information B), which was translated from English into Bengali, and back-translated to confirm accuracy. Gestational age (time between the first day of the last menstrual period and child's date of birth) was recorded, and classified as term (>37 completed weeks of

gestation), preterm (32 to <37 weeks), very preterm birth (28 to <32 weeks), and extremely preterm (<28 weeks).²⁶ The presence of comorbidities was collected from the parent in both contexts, however, using the standardized questions of the *10 Question Screen* in Bangladesh.²⁷ The socioeconomic status of Australian families was classified into tertiles using scores on the Socioeconomic Indexes for Areas Index of Relative Disadvantage.²⁸ The Poverty-Measurement Tool was used to classify the Bangladesh sample into 5 levels from well-off to poor, and has been validated in rural Bangladesh against an asset index and other traditional poverty measures.²⁹ The presumed timing (judged by physicians) of the complicating event was classified as antenatal, intrapartum, postpartum, or post-neonatal, or a combination of these (as reflected in Supplemental Information A). Five-minute Apgar scores <7 or a delayed cry >5 min after birth (in the absence of Apgars) were documented as a marker of neurologic depression.^{17,30} Parents were also asked to report sitting and standing ability, and the age of acquisition of these skills.

Motor type/distribution were classified according to the Surveillance of Cerebral Palsy in Europe guidelines as spasticity (unilateral or bilateral), ataxia, dystonia, athetosis, or hypotonia.¹⁰ The GMFCS classifies children into 5 levels, with the <2-year-old and 2- to 4-year-old scales used in the current study.³¹

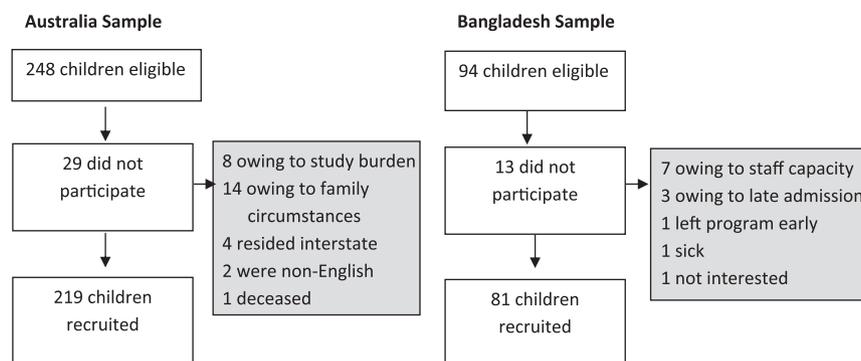


FIGURE 1 Recruitment pathways for Australia and Bangladesh samples.

Ethics

All families gave written informed consent to participate. The Australian study was approved by the Children's Health Services (Royal Children's Hospital Herston HREC07/QRCH/107), Southern Health Ethics (05077C), University of Queensland

(2007001784), Cerebral Palsy League of Queensland (CPLQ2008/2009-1010), and Mater Health Services (1186C). Ethics for the Bangladesh Study were gained through the University of Queensland Medical Research Ethics Committee (2013000625), the Children's Health

Services District Ethics Committee (HREC/13/QRCH/69), Centre for the Rehabilitation of the Paralyzed Ethics Committee (CRP/RE/0401/55), and the International Centre for Diarrheal Disease Research Bangladesh, Ethics Committee (PR-13047).

TABLE 1 Characteristics of Australian and Bangladesh Samples of Preschool-Aged Children Who Have CP

Sample Characteristic	Australia <i>n</i> (%)	Bangladesh <i>n</i> (%)	Crude OR (CI); <i>P</i> value (Bd base)	Adjusted OR (CI); <i>P</i> value
Gender			0.9 (0.5–1.5); .73	1.0 (0.6–1.7); .90
Male	140 (63.9)	50 (61.7)		
Female	79 (36.1)	31 (38.3)		
Preterm or term birth				
Extremely preterm	22 (10.0)	0 (0.0)	17.7 (2.1 to infinity); <.01 ^a	NC
Very preterm	37 (16.9)	3 (3.9)	5.0 (1.5–16.7); .01	5.2 (1.5–18.0); .01
Preterm	35 (16.0)	15 (19.5)	0.8 (0.4–1.5); .48	0.6 (0.3–1.3); .22
Term	125 (57.1)	59 (76.6)	0.4 (0.2–0.7); <.01	0.5 (0.2–0.8); .01
Unknown	0 (0.0)	4 (4.9)		
Motor type				
Spasticity	192 (87.7)	50 (61.7)	4.4 (2.4–8.1); <.01	3.2 (1.7–6.0); <.01 ^b
Unilateral	71 (32.4)	5 (6.2)	7.3 (2.8–18.8); <.01	3.5 (1.2–10.4); .03 ^c
Bilateral (2 limbs)	49 (22.4)	21 (25.9)	0.8 (0.5–1.5); .52	0.4 (0.2–0.8); .01 ^d
Bilateral (3 to 4 limbs)	72 (32.9)	24 (29.6)	1.2 (0.7–2.0); .59	2.8 (1.4–5.7); <.01
Dystonia	6 (2.7)	15 (18.5)	0.1 (0.1–0.3); <.01	0.2 (0.1–0.5); <.01 ^e
Athetosis	3 (1.4)	7 (8.6)	0.2 (0.0–0.6); .01	0.2 (0.1–0.8); .03
Ataxia/hypotonia	18 (8.2)	9 (11.1)	0.7 (0.3–1.7); .44	0.8 (0.3–2.0); .62
GMFCS				
I	88 (40.2)	7 (8.6)	7.1 (3.1–16.0); <.01	7.7 (3.3–17.8); <.01 ^f
II	37 (16.9)	12 (14.8)	1.2 (0.6–2.6); .70	1.1 (0.5–2.3); .81 ^f
III	30 (13.7)	25 (30.9)	0.4 (0.2–0.7); <.01	0.3 (0.2–0.6); <.01
IV	28 (12.8)	14 (17.3)	0.7 (0.4–1.4); .31	0.7 (0.3–1.4); .30
V	36 (16.4)	23 (28.4)	0.5 (0.3–0.9); .02	0.5 (0.3–0.9); .03
Comorbidities				
Epilepsy	51 (23.4)	38 (46.9)	0.3 (0.2–0.6); <.01	0.7 (0.4–1.2); .12 ^g
Vision	75 (34.2)	22 (27.2)	1.4 (0.8–2.5); .25	2.6 (1.4–5.1); <.01 ^h
Hearing	16 (7.3)	2 (2.5)	3.1 (0.7–13.9); .14	4.4 (1.0–20.4); .06 ⁱ
Speech	91 (41.6)	51 (63.0)	0.4 (0.3–0.7); <.01	0.6 (0.3–1.0); .06 ^j
Cognitive	67 (30.6)	70 (82.7)	0.1 (0.1–0.2); <.01	0.1 (0.1–0.2); <.01 ^k
Poverty status				
Well-off		25 (31.6)		
Moderately well-off		27 (34.2)		
Not so well-off		15 (19.0)		
Poor		7 (8.9)		
Very poor		5 (6.3)		
Unknown		2 (2.5)		
Socioeconomic status				
Least disadvantaged	73 (33.3)			
Middle tertile	54 (24.7)			
Most disadvantaged	92 (42.0)			

Adjusted OR models include covariates of GMFCS, age, gender, and preterm status, except when that variable is the main explanatory variable. Bd base, Bangladesh comparison group; CI, confidence interval; NA, not applicable to the context, therefore ORs not calculated; NC, not calculable, as no children in Bangladesh were extremely preterm.

^a Calculated using episheet, Fisher's exact test.

^b GMFCS significant (OR, 0.7, *P* < .01).

^c Age (OR, 0.9, *P* = .12), GMFCS (OR, 0.3, *P* < .01), and preterm status (OR, 0.4, *P* < .01) significant.

^d GMFCS (OR, 0.6, *P* < .01) and preterm status (OR, 3.1, *P* < .01) significant.

^e GMFCS significant (OR, 2.6, *P* < .01).

^f Age significantly related to GMFCS I (OR, 1.1, *P* < .01), GMFCS II (OR, 1.0, *P* = .05).

^g GMFCS (OR, 2.0, *P* < .01) and preterm status (OR, 0.5, *P* = .05) significant.

^h GMFCS significant (OR, 1.9, *P* < .01).

ⁱ GMFCS significant (OR, 1.7, *P* < .01).

^j GMFCS (OR, 1.3, *P* < .01) and preterm status (OR, 0.6, *P* = .02) significant.

^k Preterm status significant (OR, 0.5, *P* = .03).

Statistical Analysis

Data analyses were performed using Stata 10.0 (Stata Corp, College Station, TX; 2007), with significance at $P < .05$. Sample characteristics were presented descriptively. Differences between countries were compared by using logistic regression (odds ratios [ORs]) for binary outcomes and linear regression for continuous outcomes, using Bangladesh as the comparison group. Presence/absence of each motor type, GMFCS level, and extent of preterm birth were explored by using binomial regression. To account for differences in sample characteristics between Australia and Bangladesh, ORs were adjusted for age, gender, GMFCS level, and preterm status (except when that variable was the main explanatory variable) for the demographics; and age, gender, and GMFCS level for models exploring birth and environmental risk factors and motor outcomes. Multinomial logistic regression analysis was used to explore associations between etiologies and the outcomes of GMFCS and motor type.

RESULTS

A total of 342 children were referred to the studies, of which 300 participated, 219 in the Australian sample and 81 in the Bangladesh sample (recruitment pathways are shown in Fig 1). Children's

ages ranged from 17 to 37 months, with equivalent mean ages between samples (Australia, 26.6 months, SD, 6.5; Bangladesh, 27.5 months, SD, 6.1; $P = .25$). The Australian sample was representative of a population-based sample with regards to gender ($P = .06$), GMFCS ($P = .09$), and motor type ($P = .53$).¹² There were significant differences in participant characteristics between Australia and Bangladesh, as shown in Table 1. Children from GMFCS III–V who had bilateral involvement had significantly higher odds of having visual impairment compared with children in GMFCS I–II who had unilateral/3-limb involvement in Australia (OR, 7.7, $P < .01$), but not Bangladesh (OR, 0.8, $P = .84$). The poverty status of the Bangladesh sample was not associated with GMFCS ($P = .92$) or motor type ($P = .58$).

The prevalence of birth risk factors (according to presumed timing) is presented in Table 2. Home births were more common in Bangladesh, occurring in 37 deliveries (45.6%), compared with only 4 (1.8%) in Australia. The majority of home births in Bangladesh (73.0%) were by an unskilled birth attendant, a further 21.6% by a nurse, and 5.4% by a family member. The influence of birth complications on motor severity and motor type is shown in Table 3.

Children's motor outcomes and associated environmental factors are shown

in Table 4. On average, children from Bangladesh were diagnosed at age 27.5 months, despite mothers reporting concerns from age 8.8 months. Of the 23.5% of Bangladeshi children who had previous access to physiotherapy, all of their treatment was limited to passive stretching. In contrast, 92.2% of Australian children had previous access to physiotherapy, which used motor learning, functional therapy, neurodevelopmental therapy, postural management approaches, or a combination of these. Children from Bangladesh spent on average 71% of their day in passive positions (lying, sitting on mother's lap, being carried), and the amount of passive time was greater for children who had poorer gross motor function (GMFCS I–II, 46.1%; III, 52.0%; IV–V, 94.7%; $r = 0.8$, $P < .01$).

DISCUSSION

The patterns of functional gross motor severity, motor type, comorbidities, birth, and environmental risk factors all differed markedly between the high- and low-resource settings. Our Australian cohort is consistent with previous published data in high-resource countries, where mild CP (GMFCS I–II) constitutes 50% to 60% of any given population.³² This pattern was skewed in the opposite direction in our Bangladesh sample, with only 23% of

TABLE 2 Prevalence of Birth Risk Factors for Australian and Bangladesh Samples of Preschool-Aged Children Who Have CP

	Australia n (%)	Bangladesh n (%)	Crude OR (CI); P value (Bd base)	Adjusted OR (CI); P value
Home delivery	4 (1.8)	37 (45.7)	0.02 (0.01–0.07); <.01	0.02 (0.0–0.07); <.01
Antenatal (only)	42 (19.2)	1 (1.2)	19.0 (2.6–140.3); <.01	19.5 (2.6–146.2); <.01
Intrapartum (only) ^a	34 (15.5)	3 (3.7)	4.8 (1.4–16.0); .01	5.5 (1.6–18.8); <.01
Postpartum (only)	9 (4.1)	49 (60.5)	0.03 (0.01–0.06); <.01	0.03 (0.01–0.06); <.01
Apgar <7/delayed cry	31 (14.1)	62 (76.5)	0.1 (0.0–0.1); <.01	0.1 (0.0–0.1); <.01 ^b
Neonatal jaundice	7 (3.2)	19 (23.8)	0.1 (0.0–0.3); <.01	0.1 (0.0–0.3); <.01
Lethargy/seizures in 72 h	48 (26.7)	32 (41.6)	0.5 (0.3–0.9); .02	0.5 (0.3–0.9); .02
Antenatal plus intrapartum	47 (21.5)	2 (2.5)	10.8 (2.6–45.6); <.01	8.7 (2.0–37.4); <.01
Intrapartum plus postpartum	17 (7.8)	18 (8.2)	0.3 (0.1–0.6); <.01	0.3 (0.1–0.7); <.01
Antenatal plus postpartum	11 (5.0)	3 (3.7)	1.4 (0.4–5.1); .63	1.6 (0.4–6.1); .50
Antenatal, intrapartum, and postpartum complications	21 (9.6)	2 (2.5)	4.2 (1.0–18.3); .06	4.9 (1.1–21.8); .04
Hospital admission	123 (55.9)	51 (86.4)	0.2 (0.1–0.4); <.01	0.2 (0.1–0.4); <.01
Post-neonatal complications	20 (9.1)	6 (7.4)	1.3 (0.5–3.2); .65	1.1 (0.4–2.9); .86

Adjusted OR models include covariates of age, gender, GMFCS. Bd base, Bangladesh comparison group.

^a Includes preterm birth.

^b GMFCS (OR, 1.3, $P = .05$) and age (OR, 0.9, $P = .02$) significantly related on logistic regression.

TABLE 3 Association Between Birth Risk Factors and Outcomes of Motor Severity and Motor Type in Preschool-Aged Children Who Have CP in Australia and Bangladesh

Birth Risk Factors	Australia OR (CI); P value I–II, n = 125; III, n = 30; IV–V, n = 64	Bangladesh OR (CI); P value I–II, n = 19; III, n = 25; IV–V, n = 37
Association with motor severity		
Home delivery: GMFCS I–II (base)	Reference	Reference
III	NC ^a	1.0 (0.3–3.4); .97
IV–V	8.1 (0.9–73.7); .06	0.9 (0.3–2.6); .77
Antenatal: GMFCS I–II (base)	Reference	Reference
III	1.5 (0.7–3.5); .30	1.2 (0.2–7.7); .88
IV–V	1.2 (0.7–2.2); .59	0.8 (0.1–4.9); .76
Intrapartum: GMFCS I–II (base)	Reference	Reference
III	1.6 (0.7–3.6); .27	1.0 (0.3–3.7); .98
IV–V	1.2 (0.6–2.1); .66	0.9 (0.3–3.0); .89
Preterm: GMFCS I–II (base)	Reference	Reference
III	1.7 (0.8–3.8); .19	0.4 (0.1–1.6); .19
IV–V	0.8 (0.4–1.4); .41	0.2 (0.1–0.9); .03
Postpartum: GMFCS I–II (base)	Reference	Reference
III	3.1 (1.3–7.1); .01	2.8 (0.2–33.7); .41
IV–V	1.5 (0.9–3.5); .10	0.6 (0.1–3.3); .57
Apgar <7/delayed cry: GMFCS I–II (base)	Reference	Reference
III	3.4 (1.0–11.5); .05	0.6 (0.1–3.8); .60
IV–V	7.5 (3.0–18.9); <.01	0.2 (0.0–1.1); .06
Neonatal jaundice: GMFCS I–II (base)	Reference	Reference
III	2.9 (0.5–18.2); .26	0.5 (0.1–2.2); .35
IV–V	1.3 (0.2–8.2); .76	1.0 (0.3–3.4); .95
Lethargy/seizures in 72 h: GMFCS I–II (base)	Reference	Reference
III	1.0 (0.3–2.9); .97	1.0 (0.3–3.6); .97
IV–V	2.5 (1.2–5.2); .01	1.7 (0.5–5.7); .36
Hospital admission: GMFCS I–II (base)	Reference	NC ^a
III	1.9 (0.8–4.4); .13	
IV–V	1.5 (0.8–2.8); .18	
Postneonatal complications: GMFCS I–II (base)	Reference	NC ^a
III	0.7 (0.2–3.5); .71	
IV–V	1.3 (0.5–3.4); .66	
Association with motor type		
Home delivery: spasticity (base)	Reference	Reference
Dyskinetic	11.9 (1.0–145.0); .05	1.4 (0.5–3.8); .53
Ataxia/hypotonic	11.9 (1.6–90.0); .02	1.7 (0.4–7.2); .45
Antenatal: spasticity (base)	Reference	Reference
Dyskinetic	1.8 (0.4–7.3); .43	0.9 (0.2–5.0); .91
Ataxia/hypotonic	2.5 (0.9–7.1); .09	1.1 (0.1–10.9); .92
Intrapartum: spasticity (base)	Reference	Reference
Dyskinetic	1.7 (0.4–7.1); .45	1.0 (0.3–3.0); .95
Ataxia/hypotonic	1.2 (0.5–3.1); .72	3.2 (0.8–13.7); .12
Preterm: spasticity (base)	Reference	Reference
Dyskinetic	0.4 (0.1–1.8); .22	0.3 (0.1–1.1); .71
Ataxia/hypotonic	1.2 (0.5–3.0); .76	NC ^b
Postpartum: spasticity (base)	Reference	Reference
Dyskinetic	1.5 (0.4–6.2); .58	NC ^b
Ataxia/hypotonic	1.8 (0.7–4.7); .27	0.3 (0.1–1.4); .12
Apgar <7/delayed cry: spasticity (base)	Reference	Reference
Dyskinetic	3.2 (0.8–13.6); .12	0.7 (0.2–2.1); .50
Ataxia/hypotonic	1.2 (0.3–4.4); .79	0.5 (0.1–2.3); .38
Neonatal jaundice: spasticity (base)	Reference	Reference
Dyskinetic	NC ^b	1.2 (0.4–3.6); .80
Ataxia/hypotonic	1.9 (0.2–17.1); .55	0.4 (0.0–3.4); .39
Lethargy/seizures in 72 h: spasticity (base)	Reference	Reference
Dyskinetic	4.1 (0.9–19.3); .07	1.0 (0.4–2.9); .97
Ataxia/hypotonic	1.9 (0.6–6.3); .27	1.5 (0.3–6.6); .61
Hospital admission: spasticity (base)	Reference	Reference
Dyskinetic	0.6 (0.2–2.3); .45	2.6 (0.3–23.1); .41
Ataxia/hypotonic	0.8 (0.3–2.1); .70	0.7 (0.1–7.7); .79

children functioning at GMFCS I–II. Although spasticity was the dominant motor type in the Bangladesh sample, it was a lower proportion than that reported in previous studies in low-resource countries,¹⁷ and significantly lower than the rates identified in our Australian sample. There was a significantly greater number of term births in Bangladesh, consistent with other studies from low-resource settings,^{14,17,20} which would be expected in settings with poorer survival of children born preterm.³ One explanation for the differences in motor severity and type could relate to our use of consistent raters and definitions across both settings, which gives greater certainty when comparing data. Furthermore, in a recent meta-analysis, spasticity was found to be significantly lower (~14%) in term-born children who had CP compared with those born preterm.³² Higher rates of term births with asphyxia, severe jaundice, and post-neonatal complications have also been associated with quadriplegia and dystonia.³ Low Apgar/delayed cry, lethargy/seizures, and term birth were all associated with poorer gross motor function in our study.

Epilepsy and speech and cognitive impairments were more common in the Bangladesh cohort, and visual and hearing impairments in the Australian cohort. Only visual and cognitive impairments were different once differences in GMFCS and preterm status between samples were accounted for, which were influencing the relationship. These patterns could reflect the sensitivity of the 10-Question Screen used in the Bangladesh sample, which has strong sensitivity to detect motor, cognitive, and seizure disorders, but lower sensitivity for vision and hearing.²⁷ This is particularly significant as universal screening of vision/hearing does not occur in Bangladesh.^{27,33} The prevalence of epilepsy and speech impairments in our Bangladesh sample were comparable

TABLE 3 Continued

Birth Risk Factors	Australia OR (CI); P value I–II, n = 125; III, n = 30; IV–V, n = 64	Bangladesh OR (CI); P value I–II, n = 19; III, n = 25; IV–V, n = 37
Post-neonatal complications: spasticity (base)	Reference	Reference
Dyskinetic	1.3 (0.2–10.9); .82	0.8 (0.1–7.6); .81
Ataxia/hypotonic	1.2 (0.3–5.7); .81	4.5 (0.6–31.7); .13

^a 0 value in base group, therefore not calculable.

^b No children from outcome group had exposure of interest, therefore not calculable.

to previous work in a similar sample from Bangladesh; however, our estimates for visual and cognitive impairments

were much higher, and lower for hearing impairments.³⁴ In the Australian sample, the presence of epilepsy and

visual and hearing impairments was comparable to that reported in our national register report, with speech and cognitive impairments somewhat lower, perhaps owing to the younger age of our sample.¹²

Conducting research in a setting with low resources has unique challenges, particularly when aiming to provide a direct comparison with a high-resource setting. The most significant limitation to this study was the recruitment

TABLE 4 Prevalence of Environmental Factors and Motor Outcomes in the Australian and Bangladesh Samples of Preschool-Aged Children Who Have CP

	Australia	Bangladesh	Crude OR or β (CI); P value (Bd base)	Adjusted OR or β (CI); P value
Mean age of first concern, mo	NA	8.8	NA	NA
I–II		12.7		
III		8.8		
IV–V		6.8		
Mean age of diagnosis, mo	13.3	27.5	–14.2 (–16.4 to 12.0); <.01	–14.6 (–16.6 to –12.6); <.01 ^a
I–II	14.5	26.4		
III	14.8	30.6		
IV–V	9.6	26.0		
Prior contact with physiotherapy, %	92.2	24.1	37.5 (18.4 to 76.7); <.01	102.7 (33.9 to 310.6); <.01 ^b
I–II	88.8	5.6		
III	93.3	28.0		
IV–V	98.4	30.6		
Equipment: chair, %	34.2	6.2	7.9 (3.1 to 20.4); <.01	22.7 (7.8 to 65.8); <.01 ^c
I–II	9.6	5.3		
III	46.7	8.0		
IV–V	76.6	5.4		
Equipment: mobility, %	17.8	1.2	17.3 (2.3 to 127.7); <.01	17.0 (2.3 to 128.4); <.01 ^d
I–II	12.0	0.0		
III	46.7	4.0		
IV–V	15.6	0.0		
Able to sit, %	55.9	48.1	1.4 (0.8 to 2.3); .22	0.8 (0.4 to 1.5); .45 ^e
I–II	69.8	84.2		
III	60.0	80.0		
IV–V	26.6	8.1		
Mean age of sitting, mo	12.5	14.9	–2.4 (–4.8 to –0.0); .05	–1.3 (–3.4 to 0.9); .24 ^f
I–II	10.3	12.4		
III	17.9	17.0		
IV–V	18.4	14.7		
Able to walk, %	35.5	8.6	5.9 (2.6 to 13.3); <.01	3.5 (1.3 to 9.1); .01 ^g
I–II	58.7	36.8		
III	6.7	0.0		
IV–V	3.1	0.0		
Mean age of walking, mo	22.5	20.9	1.6 (–9.3 to 12.4); .77	–1.1 (–7.1 to 4.9); .70 ^h
I–II	20.1	20.9		
III	33.5	NA		
IV–V	NA ⁱ	NA		

Adjusted ORs include covariates of age, gender, and GMFCS (collapsed). Bd base, Bangladesh comparison group.

^a Age ($\beta = 0.7$, $P < .01$) and GMFCS ($\beta = -1.4$, $P = .01$) significant.

^b Age (OR = 0.9, $P = .01$), GMFCS (OR = 2.5, $P < .01$), and preterm status (OR = 2.8, $P = .02$) significant.

^c GMFCS (OR = 5.2, $P < .01$) significant.

^d Preterm status (OR = 3.2, $P < .01$) significant.

^e GMFCS significant (OR = 0.3, $P < .01$).

^f GMFCS ($\beta = 4.4$, $P < .01$) and preterm status ($\beta = 2.9$, $P < .01$) significant.

^g GMFCS (OR = 0.1, $P < .01$) and age (OR = 1.1, $P < .01$) significant.

^h GMFCS significant ($\beta = 34.7$, $P < .01$).

ⁱ No age recorded, but parent reported walking ability.

of a sample of clinic attendees in Bangladesh, which may limit generalizability to the population, although by adjusting the models for differences in gross motor function, we were still able to compare between samples. Over 80% of eligible children attending the center in Bangladesh were recruited to our study, with no systematic bias in their selection, although this recruitment rate was low compared with national prevalence rates. The sample was skewed toward rural families (being the predominant group accessing inpatient services at the center), and included more moderately well-off and well-off families than would be expected for the country.²⁹ Admission as an inpatient at CRP is not associated with illness or medical interventions, and as such is unlikely to skew the sample. There was no association between the poverty status of the Bangladesh sample and motor severity/type, which suggests economic factors were not biasing the motor patterns of those attending for services. Use of parent report for gathering much of the birth history may be biased by recall in both settings. This could be confounded further in Bangladesh, where a greater number of births are unregistered and occur at home.

CONCLUSIONS

This comparative study has implications for understanding the motor severity and patterns associated with CP.

Differences in children's environment, both physical and opportunities provided in the home, may have an effect on children's motor outcomes and GMFCS level. Significantly fewer children from Bangladesh GMFCS IV–V were able to sit, and from GMFCS III able to walk, which may be reflected in their lower access to therapy and supportive equipment, as well as a large amount of time spent in passive activities. This raises questions regarding whether these children “catch up” to children of a similar level from Australia, or whether their poorer gross motor function is likely to persist. Studies using the Gross Motor Function Measure to assess specific gross motor tasks, and longitudinal studies to determine change across time, would help in understanding the applicability of motor curves and whether the prognostic aspect of the GMFCS is valid in this different cultural and economic setting.

This study provides useful information to assist with global perspectives on CP management. The high rates of term-born children who have CP in Bangladesh suggest scope for improved primary prevention, particularly through education and support of unskilled birth attendants.³ The delayed age of diagnosis and access to appropriate treatments in Bangladesh represents an important window of opportunity for secondary prevention through early intervention. The findings from the current study suggest there is likely to be

a significant subgroup of term-born children who have dystonia for whom early motor type diagnosis is more challenging. This group may also require access to different treatments, particularly the use of medications and careful consideration of the appropriateness of surgical interventions. Uptake of classification systems such as the GMFCS has been limited in Bangladesh, so improved training of health staff in such classification systems³⁵ as well as resources to support CP diagnosis and differential diagnosis of motor types would enable fast-tracked screening and appropriate, targeted interventions. Although there are many important factors to prioritize in low-resource countries, initiation of a centralized CP register, initially of clinic attendees, with consistent screening and definitions between centers, may assist in understanding the national picture of the diagnosis, and thereby better targeted management.

ACKNOWLEDGMENTS

We thank Ms Jannatul Ferdous (B Science, SpThy), Dr Sabera Bilkis (MBBS), Ms Hosneara Parveen (B Science Physio), Ms Sharmin Hasnat (B Science, SpThy), Ms Shoma (B Science Physio), and other staff at the Centre for the Rehabilitation of the Paralysed for their support with conducting the research in Bangladesh. We also acknowledge the support of International Centre for Diarrhoeal Disease Research (Dr Baitun Nahar and Dr Tahmeed Ahmed) in collaborating on this research.

REFERENCES

1. Reddihough DS, Collins KJ. The epidemiology and causes of cerebral palsy. *Aust J Physiother*. 2003;49(1):7–12
2. World Health Organization, The World Bank. World report on disability. 2011. Available at: http://whqlibdoc.who.int/publications/2011/9789240685215_eng.pdf. Accessed May 14, 2014
3. Gladstone M. A review of the incidence and prevalence, types and aetiology of childhood cerebral palsy in resource-poor settings. *Ann Trop Paediatr*. 2010;30(3):181–196
4. The World Bank. GDP per capita (current US\$). 2014. Available at: <http://data.worldbank.org/indicator/NY.GDP.PCAP.CD#>. Accessed June 11, 2014
5. Rayhan MI, Khan MSH. Factors causing malnutrition in children under 5 in Bangladesh. *Pakistan J Nutr*. 2006;5:558–562
6. Australian Government. Our country. 2014. Available at: <http://australia.gov.au/about-australia/our-country/>. Accessed May 16, 2014
7. G20. G20 members. 2014. Available at: https://www.g20.org/about_g20/g20_members. Accessed May 16, 2014
8. Badawi N, Watson L, Petterson B, et al. What constitutes cerebral palsy? *Dev Med Child Neurol*. 1998;40(8):520–527

9. Smithers-Sheedy H, Badawi N, Blair E, et al. What constitutes cerebral palsy in the twenty-first century? *Dev Med Child Neurol*. 2014;56(4):323–328
10. Sanger TD, Delgado MR, Gaebler-Spira D, Hallett M, Mink JW; Task Force on Childhood Motor Disorders. Classification and definition of disorders causing hypertonia in childhood. *Pediatrics*. 2003;111(1). Available at: www.pediatrics.org/cgi/content/full/111/1/e89
11. Blair E, Watson L. Epidemiology of cerebral palsy. *Semin Fetal Neonatal Med*. 2006;11(2):117–125
12. Australian Cerebral Palsy Register Group. The Australian Cerebral Palsy Register Report 2013. 2013. Available at: www.cpresearch.org.au/pdfs/2013_ACPR-Report_Web.pdf. Accessed March 16, 2014
13. Liu JM, Li S, Lin Q, Li Z. Prevalence of cerebral palsy in China. *Int J Epidemiol*. 1999;28(5):949–954
14. Banerjee TK, Hazra A, Biswas A, et al. Neurological disorders in children and adolescents. *Indian J Pediatr*. 2009;76(2):139–146
15. Bangladesh Ministry of Health and Family Welfare. *Survey of Autism and Neurodevelopmental Disorders in Bangladesh*. Dhaka, Bangladesh: Sudipta Printers & Packagers Ltd; 2013
16. Suvanand S, Kapoor SK, Reddaiah VP, Singh U, Sundaram KR. Risk factors for cerebral palsy. *Indian J Pediatr*. 1997;64(5):677–685
17. Singhi PD, Ray M, Suri G. Clinical spectrum of cerebral palsy in north India—an analysis of 1,000 cases. *J Trop Pediatr*. 2002;48(3):162–166
18. Khan NZ, Ferdous S, Munir S, Huq S, McConachie H. Mortality of urban and rural young children with cerebral palsy in Bangladesh. *Dev Med Child Neurol*. 1998;40(11):749–753
19. Karumuna JMS, Mgone CS. Cerebral palsy in Dar Es Salaam. *Cent Afr J Med*. 1990;36(1):8–10
20. Nottidge VA, Okogbo ME. Cerebral palsy in Ibadan, Nigeria. *Dev Med Child Neurol*. 1991;33(3):241–245
21. Bartlett DJ, Palisano RJ. A multivariate model of determinants of motor change for children with cerebral palsy. *Phys Ther*. 2000;80(6):598–614
22. Rosenbaum PL, Walter SD, Hanna SE, et al. Prognosis for gross motor function in cerebral palsy: creation of motor development curves. *JAMA*. 2002;288(11):1357–1363
23. Piper MC. Efficacy of physical therapy: rate of motor development in children with cerebral palsy. *Pediatr Phys Ther*. 1990;2(3):126–130
24. Boyd RN, Jordan R, Pareezer L, et al. Australian Cerebral Palsy Child Study: protocol of a prospective population based study of motor and brain development of preschool aged children with cerebral palsy. *BMC Neurol*. 2013;13(57):e57–e69
25. Bell KL, Boyd RN, Tweedy SM, Weir KA, Stevenson RD, Davies PSW. A prospective, longitudinal study of growth, nutrition and sedentary behaviour in young children with cerebral palsy. *BMC Public Health*. 2012;2010(10):e179–e191
26. World Health Organization. Preterm birth: fact sheet N°363. 2013. Available at: www.who.int/mediacentre/factsheets/fs363/en/. Accessed May 29, 2014
27. Durkin MS, Davidson LL, Desai P, et al. Validity of the ten questions screened for childhood disability: results from population-based studies in Bangladesh, Jamaica, and Pakistan. *Epidemiology*. 1994;5(3):283–289
28. Australian Bureau of Statistics. Census of population and housing: socio-economic indexes for areas (SEIFA), Australia. 2011. Available at: www.abs.gov.au/ausstats/abs@nsf/mf/2033.0.55.001. Accessed June 25, 2014
29. Bhuiya A, Mahmood SS, Rana AK, Wahed T, Ahmed SM, Chowdhury AMR. A multi-dimensional approach to measure poverty in rural Bangladesh. *J Health Popul Nutr*. 2007;25(2):134–145
30. Lie KK, Grøholt E-K, Eskild A. Association of cerebral palsy with Apgar score in low and normal birthweight infants: population based cohort study. *BMJ*. 2010;341:c4990
31. Palisano R, Rosenbaum P, Walter S, Russell D, Wood E, Galuppi B. Development and reliability of a system to classify gross motor function in children with cerebral palsy. *Dev Med Child Neurol*. 1997;39(4):214–223
32. Himpens E, Van den Broeck C, Oostra A, Galders P, Vanhaesebrouck P. Prevalence, type, distribution, and severity of cerebral palsy in relation to gestational age: a meta-analytic review. *Dev Med Child Neurol*. 2008;50(5):334–340
33. Berg AL, Papri H, Ferdous S, Khan NZ, Durkin MS. Screening methods for childhood hearing impairment in rural Bangladesh. *Int J Pediatr Otorhinolaryngol*. 2006;70(1):107–114
34. Khan MSZ, Moyeenuzzaman M, Islam MQ. A study on patients with cerebral palsy. *Bangladesh Med Res Counc Bull*. 2006;32(2):38–42
35. Rosenbaum P, Eliasson AC, Hidecker MJ, Palisano RJ. Classification in childhood disability: focusing on function in the 21st century. *J Child Neurol*. 2014;29(8):1036–1045

(Continued from first page)

PEDIATRICS (ISSN Numbers: Print, 0031-4005; Online, 1098-4275).

Copyright © 2014 by the American Academy of Pediatrics

FINANCIAL DISCLOSURE: The authors have indicated they have no financial relationships relevant to this article to disclose.

FUNDING: Funded by the Australian National Health and Medical Research Council Postgraduate Medical and Dental Scholarship (1018264–KAB), Career Development Fellowship (APP1037220–RNB), and Project Grants (569605 and 465128). Funding was also received from The University of Queensland Graduate School International Travel Award (KAB).

POTENTIAL CONFLICT OF INTEREST: The authors have indicated they have no potential conflicts of interest to disclose.

Motor Severity in Children With Cerebral Palsy Studied in a High-Resource and Low-Resource Country

Katherine A. Benfer, Rachel Jordan, Sasaka Bandaranayake, Christine Finn, Robert S. Ware and Roslyn N. Boyd
Pediatrics 2014;134:e1594

DOI: 10.1542/peds.2014-1926 originally published online November 24, 2014;

Updated Information & Services

including high resolution figures, can be found at:
<http://pediatrics.aappublications.org/content/134/6/e1594>

References

This article cites 24 articles, 2 of which you can access for free at:
<http://pediatrics.aappublications.org/content/134/6/e1594#BIBL>

Subspecialty Collections

This article, along with others on similar topics, appears in the following collection(s):
Children With Special Health Care Needs
http://www.aappublications.org/cgi/collection/disabilities_sub
International Child Health
http://www.aappublications.org/cgi/collection/international_child_health_sub

Permissions & Licensing

Information about reproducing this article in parts (figures, tables) or in its entirety can be found online at:
<http://www.aappublications.org/site/misc/Permissions.xhtml>

Reprints

Information about ordering reprints can be found online:
<http://www.aappublications.org/site/misc/reprints.xhtml>

American Academy of Pediatrics

DEDICATED TO THE HEALTH OF ALL CHILDREN™



PEDIATRICS®

OFFICIAL JOURNAL OF THE AMERICAN ACADEMY OF PEDIATRICS

Motor Severity in Children With Cerebral Palsy Studied in a High-Resource and Low-Resource Country

Katherine A. Benfer, Rachel Jordan, Sasaka Bandaranayake, Christine Finn, Robert S. Ware and Roslyn N. Boyd

Pediatrics 2014;134:e1594

DOI: 10.1542/peds.2014-1926 originally published online November 24, 2014;

The online version of this article, along with updated information and services, is located on the World Wide Web at:

<http://pediatrics.aappublications.org/content/134/6/e1594>

Data Supplement at:

<http://pediatrics.aappublications.org/content/suppl/2014/11/19/peds.2014-1926.DCSupplemental>

Pediatrics is the official journal of the American Academy of Pediatrics. A monthly publication, it has been published continuously since 1948. Pediatrics is owned, published, and trademarked by the American Academy of Pediatrics, 141 Northwest Point Boulevard, Elk Grove Village, Illinois, 60007. Copyright © 2014 by the American Academy of Pediatrics. All rights reserved. Print ISSN: 1073-0397.

American Academy of Pediatrics

DEDICATED TO THE HEALTH OF ALL CHILDREN™

