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# A Cross-sectional Study of the Clinical Profile of Children With Cerebral Palsy in Benin, a West African Low-Income Country

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## Abstract

Cerebral palsy is a common cause of pediatric motor disability. Although there are increasing amounts of data on the clinical profile of children with cerebral palsy in high-income countries, corresponding information about low-income countries and developing countries is lacking. Therefore, we aimed to describe the clinical spectrum of cerebral palsy in children in Benin, a representative West African low-income country. Our cross-sectional observational study included 114 children with cerebral palsy recruited from community-based rehabilitation centers and teaching hospitals (median age: 7 years, range 2-17; sex: 66% male). Data were collected through review of medical records and interviews with children's mothers. Assessment included risk factors, clinical subtypes according to the Surveillance of CP in Europe criteria, severity of motor outcome scored by the Gross Motor Function Classification System (GMFCS) and Manual Ability Classification System, comorbidities, and school attendance. We recorded a high prevalence of intrapartum adverse events. Seventeen percent of children had postneonatal cerebral palsy, with cerebral malaria being the most common cause. Most children were severely affected (67.5% as bilateral spastic; 54.4% as GMFCS IV or V), but severity declined substantially with age. Only 23% of the children with cerebral palsy had attended school. Poor motor outcomes and comorbidities were associated with school nonattendance. These results suggest that intrapartum risk factors and postnatal cerebral malaria in infants are opportune targets for prevention of cerebral palsy in Sub-Saharan low-income countries.

## Keywords

cerebral palsy, cerebral malaria, low-income country, Sub-Saharan Africa, epidemiology

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Cerebral palsy is the most common cause of pediatric motor disability.[1] The broad term describes a group of disorders impairing the development of movement and posture due to a nonprogressive injury of the immature brain. Accompanying comorbidities of cerebral palsy can include epilepsy, and impairments of sensation, communication, and musculoskeletal functions.[1]

There are increasing amounts of data on the clinical profiles of children with cerebral palsy in high-income countries obtained through large population-based studies monitoring cerebral palsy cases. However, there remains a relative paucity of corresponding information about cerebral palsy in low- and middle-income countries.[2,3] The prevalence of cerebral palsy is generally estimated at 2 to 2.5 per 1000 of birth survivors in high-income countries, but recent studies have shown a trend toward decreasing incidence.[4-6] This improvement may reflect improved perinatal care protecting against prematurity and low birth weight, which are the most often reported risk factors for cerebral palsy in high-income countries.[5,6] There have hitherto been 3 population-based studies of cerebral palsy in Africa, reporting prevalence of 2.7 per 1000 children in Uganda, 2.04 and 3.6 per 1000 children in 2 different Egyptian districts.[7-9] In West Africa, the clinical profile of cerebral palsy is best described in Nigeria, which is a middle-income

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country.[3,10,11] Most of the Nigerian studies were conducted in tertiary pediatric clinics, and emphasized a description of motor outcomes, comorbidities, and risk factors.[10-12] Cases of severe cerebral palsy with bilateral spasticity and levels IV to V of the Gross Motor Function Classification System (GMFCS) predominated in the Nigerian studies. Risk factors differed from those in high-income countries, notably with respect to greater contribution of factors such as birth asphyxia, kernicterus, and neonatal infections.[3,10-12]

In the present study, we describe the clinical profile of children with cerebral palsy in Benin, a low-income country in West Africa. Despite its low-income country designation, community-based rehabilitation is well developed in Benin. By training parents as therapists, community-based rehabilitation allows free access to rehabilitative care for children of parents unable to afford hospital-based care.[13] This program is organized by the community itself and thereby is intended to promote social integration of children with disability. Thus, implementation of community-based rehabilitation should help to lower the risk of stigmatizing children with disability and their parents.[13,14] Financial impediments for most parents in seeking medical care, together with stigmatization, are factors likely to induce biases in hospital-based studies conducted in low-income countries such as Benin.[15] Indeed, such factors might bring about overrepresentation of high-need children, whose parents possess sufficient financial resources to obtain medical care.[15] Community-based rehabilitation settings, by offering free rehabilitation care, should help to lower this risk of reporting bias.

This study aims to fill in the gaps in understanding cerebral palsy in a West African low-income country by describing the clinical subtypes, risk factors and outcomes through a cross-sectional study of cerebral palsy in community-based rehabilitation and hospital settings in Benin. We hypothesized that term-born children with severe cerebral palsy would predominate in our study population, and predicted that cerebral malaria would be a common postneonatal cause in this endemic area.

# Method

## Study Design

This cross-sectional study included children with cerebral palsy from 5 community-based rehabilitation centers and 2 rehabilitation departments of teaching hospitals in the south (Cotonou) and the north (Parakou) of the Republic of Benin, in West Africa. Cotonou is the economic capital of Benin, whereas Parakou is more rural, albeit being the most economically developed area in the North of Benin. We screened patients' registers in each setting for children aged between 2 and 17 years with diagnosis of cerebral palsy. We attempted phone contacts with discharged children and invited the contactable families to participate in the study. Based on the cerebral palsy definition of Rosenbaum et al,[1] children were included if they experienced (1) motor disorders (muscle weakness or spasticity or movement disorders), (2) due to injury/maldevelopment of the developing brain, (3) with associated difficulties in daily life activities (gross or fine motor activities), (4) without sign of progressive loss of acquired abilities (checked during interview), and (5) with an onset of the disorder observed before 2 years of age.

## Data Collection

*Risk factors.* We interviewed each child's mother with a structured questionnaire and reviewed medical records about sociodemographic data, and history of the pregnancy, birth, and postneonatal adverse events (see Table 1). History of pregnancy included maternal alcohol consumption, smoking, fever/infection, malaria, or abnormal bleeding during pregnancy. History of birth involved induced labor, complications during delivery, prematurity, fetal presentation, and other notable events. We could not retrospectively assess the children's APGAR (Atmung, Puls, Grundtonus, Aussehen, und Reflexe, that is, breathing, pulse, tonus, appearance, and reflexes) scores at birth, as this does not appear in the medical records. As an alternate to APGAR scores, we asked the mothers for their recollection of whether the children cried at birth. Postneonatal adverse events potentially leading to cerebral palsy were reviewed in medical records.

*Clinical subtype.* We used the classification tree of the Surveillance of Cerebral Palsy in Europe (http://www.scpenetwork.eu/de/home/) white paper to group the children according to their predominant movement disorders. These were bilateral spastic cerebral palsy, unilateral spastic cerebral palsy, dyskinetic, and ataxic.[16] We further classified children with bilateral spastic cerebral palsy into those with spastic diplegia or quadriplegia.

*Outcomes (motor).* The severity of gross and fine motor function impairments was classified with the Gross Motor Function Classification System (GMFCS)[17] and the Manual Ability Classification System (MACS),[18] respectively. We also recorded the school attendance rate of the children with cerebral palsy.

*Associated comorbidities.* We searched for history of likely seizure by asking mothers during the structured interview whether their child often experienced fits or lost consciousness[19] and by reviewing medical records for any diagnoses of epilepsy/seizure. Positive history of seizure was noted if endorsed by the mother or if mentioned in the medical records. Communication difficulties of nonverbal children and those unable to construct phrases of at least 2 words were recorded when endorsed by the child's mother, or when observed during communication between mother and child. We checked for severe visual impairment presenting with an inability to track visually a colorful and attractive toy or respond with vision-provoked behaviors such as facial expression, blinking eyes, and smiling.[20] Finally, we investigated signs of cognitive impairments by asking the child's mother (1) if the child learns as well as his peers, (2) if he or she needs greater help compared to his peers in learning simple tasks such as the rules of games universally played by age-peers, and (3) if he or she can understand and respond to simple orders from peers. An endorsement of 2 or 3 of these questions was considered a positive sign of cognitive impairments.

## Data Analysis

Statistical analyses of the children's demographic and clinical characteristics were performed with SPSS IBM 25. We used the chi-square test to analyze for associations between categorical variables. We calculated the likelihood ratio where applicable (>20% of expected

**Table 1.** Sociodemographic Characteristics and Risk Factors of 114 Children With Cerebral Palsy in Benin.

| Characteristics | Values |
|---|---|
| Age, y, median [IQR] | 7 [4; 11.1] |
| Gender, n (%) | |
| Female | 39 (34.2) |
| Male | 75 (65.8) |
| Birth weight, kg, median [IQR] (n = 96) [a] | 3 [2.5; 3.5] |
| Siblings, median [IQR] | 3 [2; 4] |
| Firstborn, n (%) | 47 (41.2) |
| Attending school, n (%) | 27 (23.7) |
| Ordinary/specialized school (n) | 25/2 |
| Residence, rural/urban, n | 25/89 |
| Fathers' level of education, n (%) | |
| Not educated | 7 (6.1) |
| Primary school | 38 (33.3) |
| Secondary (college) | 37 (32.5) |
| University | 23 (20.2) |
| Mothers' level of education, n (%) | |
| Not educated | 30 (26.3) |
| Primary school | 44 (38.6) |
| Secondary (college) | 28 (24.6) |
| University | 12 (10.5) |
| Maternal age at delivery, y, median [IQR] | 28 [23; 33] |
| Risk factors in pregnancy, n (%) | |
| Smoking | 0 |
| Alcohol consumption | 0 |
| Received antenatal care | 112 (98.2) |
| Malaria | 17 (14.9) |
| Infection | 10 (8.8) |
| Abnormal bleeding | 4 (3.5) |
| History of birth delivery, n (%) | |
| Birth delivery in hospital | 113 (99.1) |
| Caesarean section | 16 (14) |
| Preterm birth, <37 wk | 8 (7) |
| Induction of labor | 60 (52.6) |
| Complications during delivery, n (%) | 60 (52.6) |
| Breech presentation, n (%) | 1 (0.9) |
| Not crying at birth, n (%) | 61 (53.5) |
| Crying delay, min, median [IQR] (n = 54) [a] | 15 [5; 30] |
| Admitted into NICU, n (%) | 53 (46.5) |
| Days into NICU, median [IQR] [a] | 14 [7; 21] |
| Postneonatal events, n (%)[a] | 19 (16.7) |
| Seizure/convulsions | 5 (4.4) |
| Cerebral malaria | 7 (6.1) |
| Meningitis | 4 (3.5) |
| Jaundice | 2 (1.8) |
| Head traumatism | 1 (0.9) |

Abbreviations: IQR, interquartile range [25th percentile; 75th percentile]; NICU, neonatal intensive care unit.
[a]Data extracted from medical records.

frequencies are below 5) and the Fisher exact test for binary variables. $P$ value $\leq .05$ was the threshold for statistical significance.

## Results

*Participants.* We screened 198 children (69 in Parakou and 129 in Cotonou). Thirty-seven could not be contacted because of nonfunctional phone numbers. Among the 161 remaining eligible participants, 34 had moved from the region and could not be interviewed, the parents of 2 children declined to participate, and 10 children were no longer alive. One child was excluded because of an acquired brain injury from sickle cell disease at 3 years of age. The final sample included 114 children, of whom 70% were recruited from community-based rehabilitation centers, with diagnosis of cerebral palsy by pediatricians, neurologists, and physiatrists (specialists in physical medicine and rehabilitation). Table 1 presents the children's demographic characteristics; age ranged from 2 to 17 years, with a median of 7 years. Sixty-six percent were male, 10% were twins, and 41% were firstborn.

*Risk factors.* All mothers but 2 had received antenatal care. Vaccination during pregnancy against tetanus was recorded in all but 9 cases. Twenty-two percent of mothers reported at least 1 adverse antenatal event, that is, malaria, vaginal infections, or abnormal bleeding. No mother smoked or used alcohol during pregnancy. Deliveries were exclusively at hospital, with the exception of 1 home delivery, unassisted by a health professional. Seven percent children were born preterm (less than 37 weeks of gestational age). More than half of the children's mothers reported intrapartum adverse events, that is, induced labor or complications during delivery. Fifty percent of children had not cried until a median of 15 minutes postbirth. Seventeen percent of children had postneonatal events, mainly cerebral malaria and seizure. Demographic data for risk factors are presented in Table 1.

*Clinical subtype.* Bilateral spastic cerebral palsy was the most common subtype (77 children, 67.5%), including 54 (47.4% of the total sample) with quadriplegia and 23 (20.2%) subjects with diplegia. Twenty-two (19.3%) were classified as unilateral spastic cerebral palsy. Five (4.4%) children had dyskinesia, 2 (1.8%) had ataxia, and 8 (7%) were unclassified.

### Outcome

*Motor outcome.* Many of the children in our sample were severely impaired: 54% were classified as Gross Motor Function Classification System IV or V and 45% as Manual Ability Classification System IV or V (Figure 1A). The proportion of children with severe impairment to Manual Ability Classification System increased with higher Gross Motor Function Classification System levels, likelihood ratio (df) = 79.93 (16), $P < .001$ (Figure 1B). Consistently, bilateral spastic cerebral palsy was more common in children with severe gross motor impairment, whereas unilateral spastic cerebral palsy was more common in less impaired children, likelihood ratio (df) = 57.92 (16), $P < .001$ (Figure 1C). We found no association between children's age and Gross Motor Function Classification System levels, likelihood ratio (df) = 17.63 (12), $P = .13$ (Figure 1D). However, when grouping children as "independent walkers" (Gross Motor Function Classification System I and II) versus "non-walkers" (Gross Motor Function Classification System III, IV, and V), the chi-square test was statistically significant, $\chi^2$(df) = 12.27 (3), $P = .01$. The proportion of severely affected

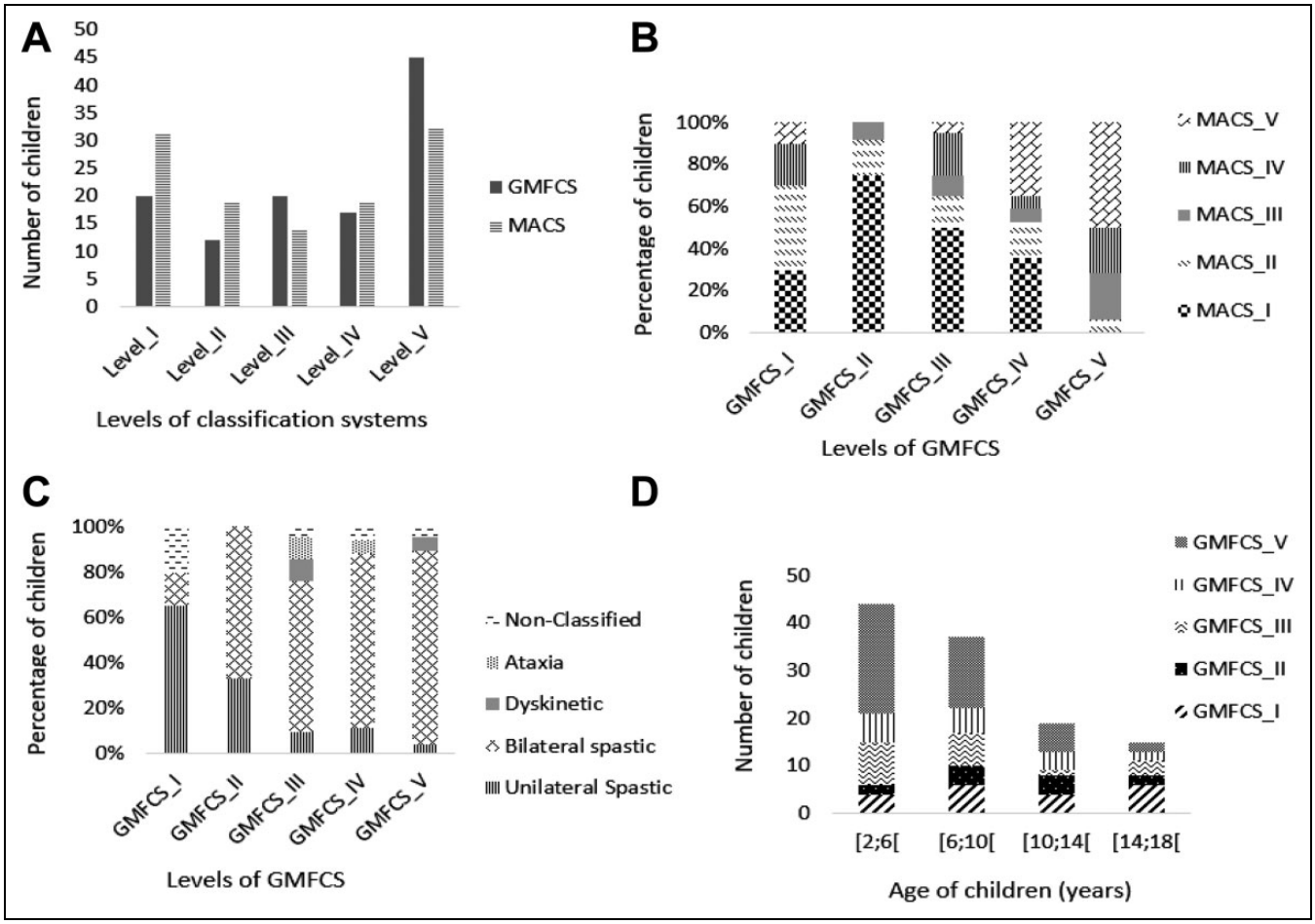


**Figure 1.** Distribution of children in Benin with cerebral palsy (CP) according to their MACS and GMFCS levels (A). Distribution of MACS (B) and CP subtype (C) by GMFCS (percentage of total number of each GMFCS level). Distribution of GMFCS levels by children's age (D). GMFCS, Gross Motor Function Classification System; MACS, Manual Ability Classification System.

children decreased with increasing age, as shown in Figure 2. There was no association between Gross Motor Function Classification System levels and risk factors such as "having an adverse event during pregnancy" ($P = .98$), "induction of labor" ($P = .19$), "crying at birth" ($P = .40$), "crying delay" ($P = .74$), and "admission to NICU" ($P = .19$), and a borderline significant association with "complications during delivery" ($P = .05$). Mothers of a higher proportion of children classified as Gross Motor Function Classification System I had not reported experiencing any complications during delivery (Table 2). Regarding walking aids, 9 of 20 children classified as Gross Motor Function Classification System III used a frame or walker, 4 used crutches, and 7 used a cane. Fourteen of 17 children classified as Gross Motor Function Classification System IV and only 2 of 45 children as Gross Motor Function Classification System V had parentally assisted transport in a manual wheelchair, either at home or at the community-based rehabilitation centers. Severely affected children were most often carried on their mother's back for long-distance travel, or were placed in modified chairs at home. The proportions of children stratified according to Gross Motor Function Classification System level and clinical subtypes did not differ when considering only those children recruited from the community-based rehabilitation centers (Table 3).

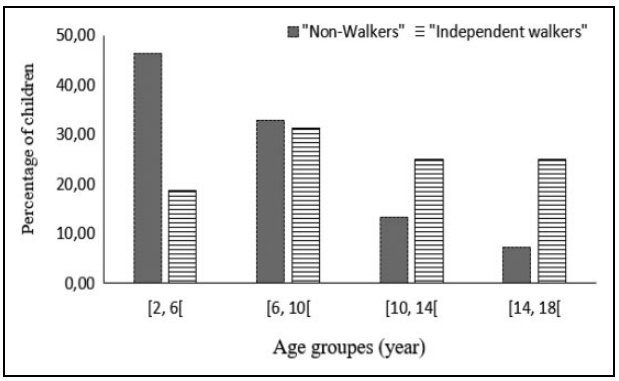


**Figure 2.** Distribution of children in Benin with cerebral palsy (CP) according to their GMFCS levels grouped as "independent walkers" (level I and II) and "Non-Walkers" (level III and V) and their ages. GMFCS, Gross Motor Function Classification System.

**Table 2.** Risk Factors Associated With Motor Outcome (GMFCS Levels) in Beninese Children With Cerebral Palsy.

| Risk factors | | GMFCS (% in variables) I | II | III | IV | V | Statistic test |
|---|---|---|---|---|---|---|---|
| Pregnancy adverse event | Yes | 4 (16) | 3 (12) | 5 (20) | 3 (12) | 10 (40) | LR(df) = 0.41 (4), $P = .98$ |
| | No | 16 (18) | 9 (10.1) | 15 (16.9) | 14 (15.7) | 35 (39.3) | |
| Induction of labor | Yes | 7 (11.9) | 9 (15.3) | 12 (20.3) | 10 (16.9) | 21 (35.6) | $\chi^2$(df) = 6.2 (4), $P = .19$ |
| | No | 13 (23.6) | 3 (5.5) | 8 (14.5) | 7 (12.7) | 24 (43.6) | |
| Complications in delivery | Yes | 6 (10) | 10 (16.7) | 12 (20) | 8 (13.3) | 24 (40) | **$\chi^2$(df) = 9.3 (4), $P = .05$[a]** |
| | No | 14 (25.9) | 2 (3.7) | 8 (14.8) | 9 (16.7) | 21 (38.9) | |
| Crying at birth | Yes | 13 (25.5) | 5 (9.8) | 7 (13.7) | 7 (13.7) | 19 (37.3) | $\chi^2$(df) = 4.04 (4), $P = .40$ |
| | No | 7 (11.3) | 7 (11.3) | 12 (19.4) | 10 (16.1) | 26 (41.9) | |
| Crying delay | <15 min | 16 (21.9) | 7 (9.6) | 13 (17.8) | 10 (13.7) | 27 (37) | $\chi^2$(df) = 1.97 (4), $P = .74$ |
| | ≥15 min | 4 (12.1) | 3 (9.1) | 5 (15.2) | 6 (18.2) | 15 (45.5) | |
| Admission to NICU | Yes | 6 (11.1) | 7 (13) | 7 (13) | 8 (14.8) | 26 (48.1) | $\chi^2$(df) = 6.18 (4), $P = .19$ |
| | No | 14 (23.2) | 5 (8.3) | 13 (21.7) | 9 (15) | 19 (31.7) | |

Abbreviations: df, degrees of freedom; GMFCS, Gross motor function classification system; LR, likelihood ratio; NICU, neonatal intensive care unit.
[a]Boldface indicates statistical significance.

**Table 3.** Distribution of Children From the Whole Sample and Recruited From CBR According to GMFCS and the Cerebral Palsy Clinical Subtype.

| | GMFCS | | | | | Clinical subtype | | | | |
|---|---|---|---|---|---|---|---|---|---|---|
| | I | II | III | IV | V | US-CP | BS-CP | Ataxia | Dyskinesia | Nonclassifiable |
| Whole sample, n (%) | 20 (17.5) | 12 (10.5) | 20 (17.5) | 17 (14.9) | 45 (39.5) | 22 (19.3) | 77 (67.5) | 2 (1.8) | 5 (4.4) | 8 (7.0) |
| CBR, n (%) (n = 78)[a] | 15 (19.2) | 11 (14.1) | 10 (12.8) | 11 (14.1) | 31 (39.7) | 14 (17.9) | 55 (70.5) | 1 (1.3) | 4 (5.1) | 4 (5.1) |

Abbreviations: BS-CP, Bilateral spastic cerebral palsy; CBR, community-based rehabilitation; GMFCS, Gross Motor Function Classification System; US-CP, unilateral spastic cerebral palsy.
[a]n = 78 is the sample size of children from CBR.

*Schooling.* Twenty-seven of 103 children with cerebral palsy aged at least 3 years (Benin preschool age) were schooled, of whom 2 attended schools for children with special needs (1 unilateral spastic cerebral palsy, Gross Motor Function Classification System II, Manual Ability Classification System I; and 1 dyskinetic, Gross Motor Function Classification System III, Manual Ability Classification System IV). The main factors associated with attendance at regular school were the severity of impairment of ambulation (Gross Motor Function Classification System) and manual ability (Manual Ability Classification System) as well as communication disorders and age (Table 4).

*Comorbidities.* All comorbidities but severe visual impairments are reported in Table 5. We saw only 3 cases of severe visual impairments, who were classified as Bilateral Spastic Cerebral Palsy (2 Gross Motor Function Classification System-V and 1 Gross Motor Function Classification System-III). Signs of seizure were recorded in 18 (15.8%) children. Fifty-nine (51.8%) children had signs of communication disorders and 88 (77.2%) showed signs of cognitive impairments. Every comorbidity except cognitive impairment was significantly associated with the clinical subtype, the Gross Motor Function Classification System level, and children's age. The more severe the cerebral palsy and the younger the child, the higher the likelihood of having a comorbidity.

## Discussion

This cross-sectional study aimed to describe the clinical profile of children with cerebral palsy in the Republic of Benin, a representative West African low-income country. The most frequently reported adverse events related to cerebral palsy were known intrapartum risk factors (such as induced labor and complicated delivery), whereas cerebral malaria and convulsions were the main postneonatal causes of cerebral palsy. Most of the children were severely afflicted with cerebral palsy, with assignment to the bilateral spastic cerebral palsy and Gross Motor Function Classification System IV and V categories. Accompanying comorbidities were mainly present in younger and severely affected children.

Risk factors leading to cerebral palsy differ considerably between high-income countries and low- and middle-income countries. Birth asphyxia is the most frequently reported risk factor in low- and middle-income countries such as Nigeria.[3,21] In contrast, birth asphyxia makes only a minor contribution to the incidence of cerebral palsy in high-income countries, where prematurity is the major risk factor (accounting for about 40% of children with cerebral palsy).[5] Only 7% of the children with

**Table 4.** Distribution of Schooled Children With Cerebral Palsy According to Their Demographic and Clinical Characteristics.

| Variables | Schooled (% in variables) | Statistical test |
|---|---|---|
| Age, y | | LR(df) = 9.52 (3), $P = .02$ |
| 3-6 | 4/33 (12.1) | |
| >6-10 | 8/36 (22.2) | |
| >10-14 | 9/19 (47.4) | |
| >114-18 | 6/15 (40) | |
| Sex | | $P = .82$[a] |
| Female | 8/33 (24.2) | |
| Male | 19/70 (27.1) | |
| MACS regrouped | | $\chi^2$(df) = 13.06 (2), $P = .001$ |
| MACS-I-II | 20/46 (43.5) | |
| MACS-III | 2/11 (18.2) | |
| MACS-IV-V | 5/46 (10.9) | |
| GMFCS, grouped | | $\chi^2$(df) = 38.04 (2), $P < .001$ |
| GMFCS-I-II | 20/30 (66.7) | |
| GMFCS-III | 4/17 (23.5) | |
| GMFCS-IV-V | 3/56 (5.4) | |
| Cognitive impairments | | $P = .06$[a] |
| Yes | 17/80 (21.3) | |
| No | 10/23 (43.5) | |
| Communication disorders | | $P < .001$[a] |
| Yes | 2/55 (3.6) | |
| No | 25/48 (52.1.5) | |
| Residence | | $P = .99$[a] |
| Urban | 22/82 (26.8) | |
| Rural | 5/25 (23.8) | |
| Father's education level | | $\chi^2$(df) = 1.63 (3), $P = .65$ |
| Not educated | 3/7 (42.9) | |
| Primary school | 7/34 (20.6) | |
| Secondary school | 9/32 (28.1) | |
| University | 6/22 (27.3) | |
| Mother's level of education | | $\chi^2$(df) = 0.34 (3), $P = .95$ |
| Not educated | 7/28 (25) | |
| Primary school | 11/40 (27.5) | |
| Secondary school | 7/24 (29.2) | |
| University | 2/10 (20) | |

Abbreviations: GMFCS, Gross Motor Function Classification System; MACS, Manual Ability Classification System.
[a]Fisher exact test.

cerebral palsy in the present study were born preterm. The literature attests to the low rate of preterm survival in low- and middle-income countries, attributed to the limited availability of high-quality neonatal intensive care.[7,21] Unfortunately, determining the leading cause(s) of cerebral palsy in Benin is beyond the scope of the present cross-sectional study. The likely precipitators, which include birth asphyxia, birth defects, genetic factors, TORCH complex fetal infections, and multifactorial causes, may increase the likelihood of an intrapartum adverse event such as induced labor, complications during delivery, or baby not crying at birth,[22,23] as we likewise found in this study. Future longitudinal studies should help understanding the factors underpinning the high proportion of intrapartum risk factors occurring in Benin. We observed that complicated delivery with prolonged labor and instrumented delivery both resulted less frequently in mild disability, and suppose it likely that cerebral gray matter injuries are frequent in term-born infants with perinatal hypoxia-ischemia, and that such injuries contribute importantly to the severe cases of cerebral palsy.[23,24]

Term-born children with cerebral palsy generally have more severe impairments than do preterm children, who typically present mainly with white matter injury apt to manifest in milder disability.[24,25] The incidence of severe cases of cerebral palsy was reportedly higher in hospital-based studies in low- and middle-income countries (Nigeria,[10,11] Botswana,[21] and Uganda[26]) compared to high-income countries, where at least two-thirds of children with cerebral palsy are typically ambulatory (Gross Motor Function Classification System I-III) and with a trend toward fewer severe cases.[5,27,28] Accordingly, the proportion of severe cases was higher in our low-income country study, although the proportion of our ambulatory children (Gross Motor Function Classification System I, II, and III) was 45.6%, which exceeds the proportions found in Nigeria (29.3%) and Botswana (40.8%).[11,21] We had expected that our recruitment of children from community-based rehabilitation settings would yield more cases of milder cerebral palsy. However, the unexpectedly higher proportion of severe cases in community-based rehabilitation settings might be attributable to a recruitment bias. Parents with mildly afflicted children (Gross Motor Function Classification System I and II) might not be motivated to bring their children for time-consuming rehabilitation care, given the constraints arising from their working life. Similarly, a recent population-based study in a rural area of Uganda (in the east of Sub-Saharan Africa) found predominance of children with milder disability, in contrast to the earlier Ugandan hospital-based study in which severe cerebral palsy predominanted.[7,26] However, the lower rate of survival of severely affected children might have given rise to a higher proportion of milder cases reported in their rural population-based study. Indeed, the number of children with severe cerebral palsy decreased dramatically with age in our study, as likewise seen in the population-based study from Uganda. It is notable that 10 of 161 eligible participants in our study were no longer alive during the recruitment phase. The apparently low survival rate of severely affected children in Benin and presumably other African low-income countries draws attention to inadequate quality of care provided to these children. Indeed, the management of cerebral palsy does not follow any specific guidelines in the Republic of Benin, as is the case in other low- and middle-income countries in Sub-Saharan Africa.[15] Where there are limited resources for cerebral palsy prevention, the quality of care is also lower for affected children. We contend that low-income countries such as Benin need to promote evidence-based rehabilitation care, including intensive neurorehabilitation approaches such as

**Table 5.** Comorbidities Associated With Cerebral Palsy According to Cerebral Palsy Motor Subtype, GMFCS Level, and Age.

| | Seizure, n (%) (n = 18; 15.8%) | Communication disorders, n (%) (n = 59; 51.7%) | Cognitive impairments, n (%) (n = 88; 77.2%) |
|---|---|---|---|
| Unilateral spastic (n = 22) | 1 (4.5) | 4 (18.2) | 15 (68.2) |
| Bilateral spastic (n = 77) | 13 (16.9) | 46 (59.7) | 61 (79.2) |
| Dyskinesia/ataxia (n = 7) | 4 (57.1) | 6 (85.7) | 5 (71.4) |
| Nonclassified (n = 8) | 0 (0) | 3 (37.5) | 7 (87.5) |
| Statistical test | LR(df) = 11.89 (3), $P = .018$ | LR(df) = 19.87 (3), $P = .001$ | LR(df) = 3.71 (3), $P = .447$ |
| zGMFCS-I (n = 20) | 1 (5) | 3 (15) | 13 (65) |
| GMFCS-II (n = 12) | 0 (0) | 2 (16.7) | 9 (75) |
| GMFCS-III (n = 20) | 3 (15) | 8 (40) | 15 (75) |
| GMFCS-IV (n = 17) | 1 (5.9) | 9 (52.9) | 12 (70.60) |
| GMFCS-V (n = 45) | 13 (28.9) | 37 (82.2) | 39 (86.7) |
| Statistical test | LR(df) = 12.87 (4), $P = .012$ | $\chi^2$(df) = 37.63 (4), $P < .001$ | LR(df) = 5.67 (4), $P = .225$ |
| Age, y | | | |
| 2-6 (n = 44) | 9 (20.5) | 25 (56.8) | 34 (77.3) |
| >6-10 (n = 36) | 8 (22.2) | 21 (58.3) | 29 (80.6) |
| >10-14 (n = 19) | 0 (0) | 10 (52.6) | 13 (68.4) |
| >14-18 (n = 15) | 1 (6.7) | 3 (20) | 12 (80) |
| Statistical test | LR(df) = 9.37 (3), $P = .025$ | LR(df) = 7.52 (3), $P = .057$ | LR(df) = 0.37 (3), $P = .947$ |

Abbreviations: df, degrees of freedom; GMFCS, Gross Motor Function Classification System; LR, likelihood ratio.

Hand-Arm Bimanual Intensive Therapy including Lower Extremity (HABIT-ILE), Constraint-induced Movement Therapy (CIMT) goal-directed training, with adaptation to the circumstances of low-income countries.[29,30] Such efforts would certainly help to improve the quality of care provided to children with cerebral palsy.

The proportion of postneonatal cases of cerebral palsy was higher in our study compared to findings in high-income countries, where estimates are less than 10%.[23,27] This same trend was observed in other clinically based studies in African low- and middle-income countries, including one in Nigeria and the population-based study in Uganda.[7,10,21] Post-neonatal cerebral palsy in high-income countries is mainly due to head injury, especially cerebrovascular accident, whereas infections are the second most frequent cause.[31,32] In contrast, infections are the major cause of postneonatal cases of cerebral palsy in low- and middle-income countries. In our study of a low-income country, cerebral malaria and seizures were the main postneonatal causes of cerebral palsy. Seizure can be a consequence of cerebral palsy, irrespective of its cause, and is almost universally occurring in cerebral malaria, as previously reported in Uganda.[7] Intracranial infections and bilirubin encephalopathy secondary to jaundice were the most frequently reported causes of postneonatal cerebral palsy in Nigeria.[10,11] Malaria is highly prevalent in children in Benin, with a reported incidence of 84 of 1000 children per month. Most parents attempt to treat their children at home, only seeking hospitalization in difficult cases.[33,34] Consequently, cerebral malaria is lamentably common in children in Benin, and brings a mortality rate of 47% according to a recent study.[33,35] Moreover, malaria was the most frequently reported maternal health issue during pregnancy in our cohort, and is a known risk factor for fetal growth restrictions.[36,37] Prevention and management of malaria both in pregnant women and in children must be emphasized through active engagement of makers of health decisions in parental education. Our findings highlight the need for provision of long-lasting insecticidal nets and free hospital access for pregnant women early during their first trimester, in a concerted program to reduce the incidence of cerebral palsy in endemic malaria areas such as Benin.[37,38]

Schooling of children with disability present a serious challenge in low- and middle-income countries. In Benin, there are practically no public schools for children with special needs and disability. As a result, most children with significant disability simply do not attend school, as was the case of 74% of our cohort. Considering the milder cases (independent walkers, Gross Motor Function Classification System I to II), two-thirds attended school whereas the national net rate of children's school attendance is estimated at 87%.[39] Factors associated with school nonattendance in our cohort were impaired ability to ambulate and manipulate objects and communication disorders. In a recent study carried out in Benin, Kpadonou et al reported a high rate of school dropout by children with handicap, which was attributed to low academic success.[35] Other than particular aspects of their handicaps, broad factors linked to low school success rate included difficulties of school accessibility, social marginalization, and lack of help.[35] It is noteworthy that the social environment plays a significant role in restraining children with disability from participating in social activities. In Benin, there are practically no accommodations in public settings to facilitate accessibility for nonambulatory children in need of assistive devices such as manual or powered wheelchairs. In contrast, such devices are freely available in high-income countries, which facilitates participation even of children with severe cerebral palsy in schooling, playing, and other social activities.[40] These findings emphasize the crucial

role of public health policies in the fight against disability in low- and middle-income countries.

### *Limitations*

We cannot generalize present findings to the entire population of cerebral palsy children in Benin because this is not a population-based study. However, having drawn our sample in part from community-based rehabilitation settings makes it likely to be representative of all children with cerebral palsy in Benin. Indeed, community-based rehabilitation centers are organized within the communities by community members and are freely accessible for any parent of a child with cerebral palsy or any other developmental disability. As such, they offered us the opportunity to encounter children with mild cerebral palsy symptoms, whose parents could not afford institutional care that is often located far from home. Our data collection was mainly through interviews, which might have resulted in a recall bias in the results. Moreover, by limiting our study group to children diagnosed with cerebral palsy in community-based rehabilitation and hospital centers, we likely missed important factors such as neonatal and infant death rates among this vulnerable population. Asking mothers to recollect whether their children cried at birth is not an ideal proxy measure of breathing, and is certainly not equivalent to the entire APGAR score, which represents another limitation in this study. Furthermore, we missed the assessment of other potentially relevant comorbidities such as hearing impairment, undernutrition, and musculoskeletal disorders; such assessments that were logistically possible only included simple checks for severe visual impairments or blindness, and subjective signs of seizures, without clinical confirmation. This may well have induced bias in the reported proportion of children experiencing seizures. Moreover, we used only simple questions to explore cognitive impairments. Applying a standard, validated tool such as the Wechsler intelligence scales[41] would have been preferable, but there is currently no adaptation of this tool to the sociocultural context of Benin. Furthermore, the low access to assistive equipment might induce a bias in this study, because children with real ability of Gross Motor Function Classification System III or IV might tend to be misclassified as Gross Motor Function Classification System IV or V because of the lack of equipment such as manual or electric wheelchair. Population-based and longitudinal studies are needed to obtain a deeper understanding of cerebral palsy in West Africa.

## Conclusion

Cerebral palsy is a cause of severe cognitive and motor disability in Beninese children, and is a life-threatening health issue in the younger age group. The incidence of cerebral palsy is largely attributable to preventable risk factors such as intrapartum adverse events and cerebral malaria, thus emphasizing the necessity of improved public health policies aiming toward cerebral palsy prevention in Benin, which serves as a representative low-income country of West Africa. Low school attendance of children with cerebral palsy in Benin reflects the lack of dedicated schools for children with special needs, the inadequate social environment, and the poor availability of assistive devices.

### Author Note

Data sharing statement: data set is available from ESS at emmanuel.sogbossi@gmail.com

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### Author Contributions

ESS and YB performed the statistical analysis and interpretation of data and drafted the paper. ESS, DH, and TGK contributed to data collection. All authors contributed to the study design and reviewed and accepted the final manuscript.

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### Ethical Approval

The ethics committee of the rehabilitation department of the National University Hospital (Cotonou, Republic of Benin) approved the study (date of approval: June 13, 2017). Caregivers of all participants provided their informed consent.

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