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ORIGINAL ARTICLE

Mothers' perception of cerebral palsy in a low-income country of West Africa: a cross-sectional study

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ABSTRACT

Purpose: Investigating mothers' perceptions of Cerebral Palsy (CP) and the impact of caregiving on family life in Benin, a west-African low to middle-income country (LMIC).

Methods: In this cross-sectional study we interviewed at their homes 88 mothers of children with CP about their perception of the cause of CP and stigmatization issues. Additionally, they completed the Impact on Family Scale and the Family Support Scale questionnaires. Multinomial and ordinal regressions analyses were used ($p < 0.05$).

Results: Most mothers (78%) referred to non-biomedical causes of CP (God's will or curse). Caregivers with a limited education level were more likely to report non-biomedical beliefs. Only 28% reported having received from physicians an explanation of the causes of CP. Around 38% endorsed neighbors considered their children as cursed, which is a source of stigmatization. Mothers perceived a high impact of caregiving on their family life that correlated with non-biomedical beliefs, low education level, and the severity of CP.

Conclusion: There is a need in Benin, and probably in West-African LMICs generally, to shift to a family-centered care system and to inform the public about the root causes of CP to improve social integration of children with CP and decrease the negative impact on their families.

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Cerebral palsy; caregivers; quality of life; social behavior; developing country

➤ IMPLICATION FOR REHABILITATION

- Mothers with a limited education level are more likely to report non-biomedical causes of CP in this LMIC;
- mothers' level of education and non-biomedical beliefs of causes of CP are associated to a high impact of caregiving on family life in this LMIC;
- mothers of children with CP in this LMIC do not perceive health-workers helpful.

Introduction

Cerebral palsy (CP) is the most common cause of pediatric motor disability worldwide, affecting 2.4–3.6 per 1000 children in Sub-Saharan Africa [1,2]. The motor disability of CP can be accompanied by intellectual, communicative, sensory, and musculoskeletal impairments in various degrees of severity and complexity. Although children are directly affected by the pathology, their parents suffer a secondary impact. Studies in High Income Countries (HICs) and Low- and Middle-Income Countries (LMICs) concur in reporting that parenting children with a developmental disability (such as CP) has a substantial negative impact on caregivers' quality of life. Parents may experience parenting stress, physical burden, financial difficulties, stigmatization, and discrimination [3–7]. Regarding the framework of the International Classification of Functioning (ICF-CY), the living environment is a contextual factor that can contribute to the outcomes of children with CP [8]. The family setting is the main living environment for children with a developmental disability, especially in typical LMICs such of Sub-Saharan Africa, where most children with a developmental disability cannot attend school [9,10]. As such, family is a predominant factor in delivery of care and education to children with developmental disability. What the family brings to the child's environment will influence his or her developmental trajectory, and depends on the family's perception of their child's developmental disability [6,11,12]. Parents holding traditional non-biomedical beliefs about their children's health issue are more likely to seek non-biomedical care (such as from traditional healers), but enhancing parents' knowledge of CP and broadening their skills in caregiving improves parents' quality of life and their children's outcomes [12,13]. Moreover, as argued by Rosenbaum (2011), it seems self-evident that "a child's well-being will be influenced powerfully by the well-being of their parents and families" [14]. The important influence of the family setting on the child's health outcomes is supported by evidence that shifting from a child-centered to family-centered care system in HICs offers a better child and family-support system [14,15]. Unfortunately, the integration of families in a care system for children with developmental disability faces significant obstacles in LMICs. Few studies conducted in Sub-Saharan Africa have addressed parents'

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experiences with caring for children with CP or other developmental disabilities. Such information as is available shows that caregivers of children with developmental disability in LMICs often lack medical explanations and support from health-workers enabling them to understand what ails and how best to help their child, and what they can expect as a prognosis [5–7,16,17]. This might explain in part the epidemiological findings in LMICs showing a huge decrease in the prevalence of severe cases of CP among children aged 4–6 years, implying poor survival in their family environment [2,9]. This study aims to elaborate upon the existing literature from Sub-Saharan Africa by assessing mothers' experiences of caring for children with CP in Benin, a representative LMIC in West-Africa with a highly religious socio-cultural context. We hope to contribute to sensitizing health-workers and health policy makers in LMICs about the great importance of building a family and child support system in pediatric health services for children with CP and other disabilities. More specifically, we aim in this study to investigate mothers' perceptions of the cause of CP, stigmatization issues evident from social behaviors, and their perception of the helpfulness of health-workers, external sources of support, and the impact of caregiving on family life. Furthermore, we investigate how maternal perception of the causes of CP contribute together with other factors to the burden of caregiving on the family. We hypothesized that in Benin, many parents would attribute their children's developmental disability to non-biomedical beliefs, and that these beliefs would negatively influence the impact of caregiving on the family's life. Moreover, we hypothesized that mothers would perceive health-workers as unhelpful, and that a high proportion would report stigmatization issues.

Methods

Study design

This cross-sectional study was part of a larger study describing the clinical spectrum of CP in Benin, for which the participants' recruitment is well-described [9]. The study includes mothers as caregivers of their children with CP, recruited from community-based rehabilitation centers and the rehabilitation departments of teaching hospitals in the north (Parakou) and south (Cotonou) of Benin. From November 2017 to February 2018, the first (ESS) and second (DH) authors screened patients' registers in each setting for children diagnosed with CP aged between 2 and 17 years and accessed their telephone contacts. After explaining the purpose of the study by telephone, we invited the mothers of children to take part in the study. The mothers who agreed to participate were visited at home for data collection.

The ethic committee of the rehabilitation department of the National University Hospital (Cotonou, Republic of Benin) approved the study. All participants provided their informed consent.

Data collection

All data were collected by the first (ESS) and second (DH) authors, who are both experienced pediatric neurorehabilitation physiotherapists. All questions (assessment questionnaires included) were asked through the participant interviews, which were conducted in French (the main language of instruction in Benin) or Fon, the most widely spoken local native language in Benin, and the first language of ESS and DH.

Table 1. Questionnaire about the perception of cerebral palsy.

N	Items
1	What do you think caused the health issue of your child?
2	Did the doctor explain you what happened to your child? (Yes/No) If yes, what did the doctor say?
3.	What do people say about your child in negative way that hurts you?
4.	Do you think that people avoid you because of your child? (Yes/No)
5	Are you sometimes tempted to hide your child (e.g., when someone visits you)? (Yes/No)
6	Are you sometimes tempted not to bring your child in public setting (like market or church or in town) because of the negative look of people? (Yes/No)
7	Are you sometimes tempted to get rid of your child? (yes/No)
8	What specific difficulties do you face with caring for your child?

Perception of cerebral palsy

We interviewed children's mothers with a semi-structured questionnaire of eight items (Table 1), including the mothers' knowledge of causes of CP (items 1 and 2), social behaviors toward children with CP and their mothers (stigmatization issues) (items 3–6), and the burden/difficulties associated with caring for their child (item 7 and 8).

Impact on family

Mothers expressed the impact of their children's developmental disability on the family's life with the French version of the Impact on Family Scale (IOFS) [18,19]. The IOFS is a reliable 15-item ordinal scale measuring the perception of the social and familial impact of a childhood chronic health issue, with a higher score corresponding to a lower impact [18]. For mothers unable to speak French, the questionnaire was directly translated into Fon language during the interview, following a consensus agreement on its translation (ESS and DH).

Family social support

We modified the Family Support Scale (FSS) to document the sources of support for mothers in caring for their children and how helpful mothers perceived health-workers to be. The scale originally contained eighteen sources of support, with five categories of responses from (1) "Not at all helpful" to (5) "Extremely helpful" [20,21]. Any unavailable source of help is scored as "not available". We combined the last two ratings "very helpful" and "extremely helpful" into one, "very helpful", to make the categories more discriminative to the mothers. Moreover, we modified three items that we found less relevant in our socio-cultural context. The item "Family or child's physician" became "Family physician or Medical doctors who examined the child". We changed the item "Early childhood intervention program" to "Gynecologist or Midwife who followed you during pregnancy and/or delivery". The item "Professional helpers (social workers, therapist, etc.)" became "Social workers, Physiotherapists, nurses, etc." Finally, we added "community-based rehabilitation centers (CBR)" to the list of "professional agencies" and a 19th item "Non-Governmental Organizations (NGO) for health issues". Hence the FSS version submitted to mothers consisted of 19 items with four categories of responses ranging from (1) not at all helpful" to (4) "Very helpful". Additionally, we asked mothers to add any other sources of support not listed in the questionnaire, if necessary.

The severity of the CP was classified during interviews and by observation with the expanded and revised version of the Gross Motor Function Classification system (GMFCS), a recommended tool grouping children on a five-level Likert scale according to gross motor disability, ranging from level I (walks without limitations) to level V (transported in a manual wheelchair) [22]. The clinical subtype of CP was defined according to the Surveillance of CP in Europe instrument, which classifies CP in four clinical types according to the particular movement disorders as unilateral and bilateral spastic, dyskinesia, and ataxia [23]. The first and second authors, who are familiar with the classification systems, performed the GMFCS and the SCPE classification during the interviews.

Statistics analyses

We described the sociodemographic and clinical characteristics of our informants. The open-ended responses were analyzed with a theme coding approach by searching for words and phrases commonly used by respondents [24]. The first and second authors conducted the content analysis until reaching a consensus. We then performed stepwise backward logistic multinomial and ordinal regressions, with a backward variables selection procedure and estimation of odds ratio (OR) to investigate sociodemographic and clinical characteristics associated with the mothers' beliefs of causes of CP and the impact on the family measured with IOFS. For the multinomial regression analysis, "biomedical belief" (including mothers who attributed responsibility to health-workers) was set as the reference modality, comparing to it the God-related and folk-related beliefs. The ordinal regression was computed with four levels of the IOFS (level $1 = \text{IOFS} \leq \text{P25}$; level $2 = \text{P25} < \text{IOFS} \leq \text{P50}$; level $3 = \text{P50} < \text{IOFS} \leq \text{P75}$, and level $4 = \text{IOFS} > \text{P75}$), with the level 4 set as reference. For both analyses, we grouped some variables as follows, with the categories used as references indicated in bold text: mothers' age (age < 40 or $\geq$*40*), their level of education ("not-educated/primary level" or "secondary level" or **"**university level**"**), mothers' professions ("housewife/artisans" or **"**Trader/formally employed**"**), Children's age (age $\leq$ 7 or **>***7 years*), the GMFCS levels ("Levels III_V" or "Levels I_II"), and the FSS ("FSS scores $\leq$ median score or **>***median score*). All statistical analyses were performed with SPSS software IBM 25 with alpha = 0.05 as the threshold for statistical significance.

Results

Participants

We screened and contacted 108 mothers of children with CP. Fourteen declined to participate and six children were no longer alive (aging 3.3–8.9 years). The final sample included 88 mothers. Table 2 displays characteristics of mothers and children. Mothers' mean age (SD) was 36.4 (7.64) years and around 28% were not educated. Children's mean age (SD) was 8.4 (4.14) years. Most of the children (70.5%) were classified as bilateral spastic, 73% were non-ambulatory (GMFCS_III_IV_V) and only 26% attended school.

Table 2. Sociodemographic and clinical characteristics of participants.

Characteristics	Value
Mothers	
Age, mean (SD), years	36.42 (7.64)
Education	
Non-educated/primary school	25/31
Secondary school	23
University	9
Profession	
Housewife/retailer	20/32
Artisans	18
Traders	13
Formally employed	5
Marital status	
Widows	3
Married	85
Children	
Age, mean (SD), years	8.42 (4.14)
Sex, male	62
Siblings (no/yes)	16/72
$ Schooled	21/82
Clinical type	
Unilateral spastic	16
Bilateral spastic	62
Dyskinetic/Ataxic	4/1
Non-classified	5
GMFCS	
I_II	24
III	15
IV_V	49

Note:($)=Children aged at least three years (preschool age) were included.

Perception of cerebral palsy

Mothers' perception about causes of CP

Table 3 presents a summary of frequent statements of mothers regarding the causes of CP. Most of the mothers (65%) referred the cause of CP to God's will (e.g., "God only knows what happened. Anyway, it is my cross and I have accepted it" *[mother of a child with GMFCS_IV])*. Eleven mothers (13%) thought their child's developmental delay was caused by sorcery or evil spell (e.g., "I think it is caused by witchcraft because of jealousy/envy from the family of my co-wife because she did not have a boy child. It is not natural." *[mother of a child with GMFCS_III])*. Twelve mothers (13.6%) mentioned biomedical facts, namely "birth delivery", "infections" or "malformation" (e.g., "I think the delay during delivery caused it". *[mother of a child with GMFCS_V])*. Four mothers attributed responsibility to the health-workers (e.g., "It's the fault of the midwives who had been negligent during childbirth" *[mother of a child with GMFCS_II]*)). Finally, four mothers reported never having seen such a condition and being ignorant of its cause (e.g., "I have never seen it. I don't know what it is or what might cause it" *[mother of a child with GMFCS_IV])*.

Only twenty-five (28%) mothers endorsed having had explanations of the child's developmental disability from their physicians, who had referred to injuries of the child's brain during birth delivery or from an infectious disease or cerebral malaria. Only six mothers (21%) had referred to a biomedical-related explanation when asked for their perception of the cause of their child's CP (Table 3).

Stigmatization

Thirty-three (38%) mothers reported stigmatization from their neighbors who had folk beliefs about CP. Most among these (70%) reported that people considered their children as a malign divinity "vodoun tohossou" (deity of waters). One mother said people blamed her, saying that God had punished her for a wrongdoing. Another mother reported that people blamed her mother in-law for bewitching her child. Few mothers endorsed avoiding public settings ($n = 7$), hiding their child ($n = 2$), or being avoided by others ($n = 7$).

Factors associated with mothers' belief

We removed from the analysis the four mothers who had no biomedical knowledge of CP. The model then indicated a significant association of beliefs with mothers' levels of education. Uneducated mothers and those with primary school level were significantly more likely to ascribe the cause of CP to God's will than were mothers of university level education, with an OR of 6 [95% confidence of interval (95%CI)=1.12; 32.25 ($p = 0.037$)]. Mothers' age and profession, receipt of a medical explanation,

Table 3. Summary of mothers' perception of causes of CP.

Causes reported	Examples of statement	n	Received an explanation from MD, $N = 25$ (28.4%)
Related to God	"God only knows what happened. Anyway, it is my cross and I have accepted it." *[Participant 32, child with GMFCS IV]* "People said it is provoked by enemy. But I believe it is a God's will. He has allowed it to happen. Nothing can happen without God's will." *[Participant 12, child with GMFCS II]*	57	15/25
Related to curse	"This is not a natural disease. It is provoked, a bad spell. We thought about someone in village. He provoked it by jealousy." *[Participant 21, child with GMFCS V]* "I think it is caused by witchcraft because of jealousy/envy from the family of my co-wife because she did not have a boy child. It is not natural."*[Participant 15, child with GMFCS III]*	11	4/25
Health-workers' fault	"It's the fault of the midwives who had been negligent during childbirth" *[Participant 8, child with GMFCS II]*	4	2/25
Biomedical explanations	"I think the delay during delivery caused it" *[Participant 51, child with GMFCS V]* "I think it might be due to the repeated infections he suffered from when he was baby" *[participant 67, child with GMFCS IV]*	12	4/25
Neutral	"I have never seen it. I don't know what it is or what might cause it" *[Participant 70, child with GMFCS IV]*	4	0/25

stigmatization, children's age or sex, the GMFCS levels, and the degree of family-support were not associated with supernatural attribution of CP.

Difficulties associated with caring for children with CP

Two main domains of parenting difficulties emerged. First, there were difficulties related to the children's limitations in ambulation, making travel very hard and forcing mothers to quit their work or to give up commercial activities to take better care of their child with disability. The second difficulty was financial burdens required to continue medical and rehabilitative care for the children. Four mothers reported schooling and professional training of their children as specific challenges due to communication and intellectual difficulties associated with the children's motor disability.

Family support and impact on family life

The median score [P25; P75] on the FSS was 20.5 [18; 23] from a total score of 76 points. The main sources of support were husbands and siblings of the child with CP. Medical staff was mostly perceived as unavailable and unhelpful (Table 4). No other sources of support were indicated by the informants. Mothers reported a relatively high impact of parenting their child with CP on the family's life; the median score [P25; P75] of the IOFS was 33 [28; 40] from a total score of 60.

The ordinal regression revealed three associated factors to IOFS levels: GMFCS, mothers' beliefs and mothers' level of education (Table 5). Mothers of children with severe disability were 4.8 times (OR [95%CI = 1.86; 12.35], $p = 0.001$) more likely to perceive a high impact on the family's life compared to mothers of children with milder disability. Those mothers relating the cause of CP to God's will were 4.3 times (OR [95%CI = 1.41; 13.22], $p = 0.004$) more likely to perceive a high impact on family life compared to mothers referring to biomedical causes of CP. There was as well a borderline significant association of perceiving a high impact on family with curse-related beliefs (OR 3.83 [95%CI = 1.03; 15.09]; $p = 0.055$) compared to biomedical beliefs. Furthermore, uneducated mothers or those with primary school level were [OR 4.07 95%CI = 1.01; 16.31] more likely to perceive a high impact on family compared to mothers with university level of education ($p = 0.048$). The mothers' age, children's age, children's sex, presence of siblings, stigmatization, mothers' profession, and social support were not significantly associated with perceiving a high impact.

Discussion

This cross-sectional study aimed to investigate the perceptions of CP from mothers as (main) caregivers of their child with CP and the impact of caregiving on the family's life in Benin, a representative LMIC of West Africa. The study showed that most caregivers ascribed the causes of CP to non-biomedical beliefs, but that such beliefs were moderated by maternal level of education. Very few mothers had received any explanation from health-workers of the causes and prognosis of their child's disability. Many experienced stigmatization from neighbors who believed that their child was cursed. A higher degree of reported impact on the family's life was associated with mothers' non biomedical beliefs and lower level of education and with the severity of the child's CP. The main sources of family-support were siblings and other close family members.

The general lack of knowledge of caregivers about the developmental disability of their child is well attested in LMICs [5,13,16]. Medical services are still strongly "paternalistic" in Benin and many other LMICs. In such countries, the patient remains "the child" and the parents are mostly not involved in clinical decisions. The parents lack information, attention and support from the health-workers, and are thus left without resources to face the unexpected event of having a child with developmental disability [5,16,26]. In our study, very few mothers referred to biomedical explanations for the cause of CP. Only 28% of the cohort reported having received relevant information from the physicians who examined their child. More surprisingly, having received a medical explanation was not associated with their holding a biomedical belief of the cause of CP. Unfortunately, our structured interview was not informative about what factors might account for the surprisingly poor association between mothers' belief and availability of a biomedical explanation from health-workers. However, in view of the literature, we can suppose that mothers might not have had sufficient time, attention, and support in the process of diagnostic disclosure. Shevell & Shevell (2013)

Table 4. Sources of social support perceived by caregivers using the FSS-scaled modified by the authors.

Item	Not available	Not at all helpful	Sometimes helpful	Generally helpful	Very helpful
My Parents	0/88	47/88	26/88	9/88	6/88
My spouse's parents	1/88	53/88	18/88	11/88	5/88
My relatives/kin	0/88	54/88	29/88	5/88	0/88
My spouse's relatives	0/88	57/88	21/88	5/88	4/88
My spouse	6/88	7/88	9/88	11/88	55/88
My friends	9/88	66/88	13/88	0/88	0/88
My spouse's friends	4/88	70/88	13/88	1/88	0/88
My own child(ren)	20/88	27/88	8/88	20/88	13/88
Other parents	8/88	67/88	10/88	1/88	2/88
Co-workers	54/88	27/88	6/88	0/88	1/88
Parent groups	81/88	6/88	1/88	0/88	0/88
Social groups/clubs	83/88	5/88	0/88	0/88	0/88
Church members/ minister	23/88	51/88	6/88	2/88	1/88
Family Physician or Physician who examined the child	53/88	32/88	3/88	0/88	0/88
Gynecologist or midwife who followed pregnancy/delivery	59/88	29/88	0/88	0/88	0/88
School/daycare center	74/88	13/88	1/88	0/88	0/88
Social workers, Physiotherapists, Nurses	0/88	56/88	29/88	3/88	0/88
Professional agencies (Health Ministry, Family ministry, **CBR**, etc.)	0/88	43/88	34/88	10/88	1/88
NGO for health issues	4/88	60/88	18/88	5/88	1/88

NGO: non-governmental organization; CBR: community-based rehabilitation. Modified items are in bold.

Table 5. Factors associated to the impact of caring for a child with CP on the family (French version of the impact on family scale) from an ordinal regression analysis.

	OR	OR 95% CI [Lower, upper]	*p*-Value
Threshold			
IOFS score $\leq$ P25	–	–	$<$0.001
P25 $<$ IOFS score $\leq$ P50	–	–	$<$0.001
P50 $<$ IOFS score $\leq$ P75	–	–	0.031
Location			
Biomedical beliefs[a]	–	–	
God-related beliefs	4.32	[1.41;13.22]	0.01
Curse-related beliefs	3.83	[1.03; 15.09]	0.055
GMFCS_I_II[a]			
GMFCS_III_V	4.80	[1.86; 12.35]	0.001
University level[a]			
Not educated/Primary level	4.07	[1.01; 16.31]	0.048
Secondary level	–	–	0.404

[a] Modality considered as reference; CI: confidence of interval; P25, P50, P75 are percentile 25, 50, and 75, respectively; IOFS: Impact on Family Scale. The reference of the IOFS was IOFS score $>$ P75.

summarized the criteria for a successful process of diagnostic disclosure to parents of children with CP [27]. Their criteria included availability (time) and willingness of health-workers to answer questions from parents, as well as compassion, support, honesty, knowledge of CP, and use of terminology adjusted to the parental education level [27]. There are few specialist health-workers such as pediatricians, physiatrists, and neurologists in Benin. Consequently, the few specialists are overloaded with patients and unable to hold thorough and supportive discussions with all patients and/or caregivers. That lack of profession time is well attested in other LMICs such as Malawi and Zimbabwe [16,26]. Moreover, health-workers may themselves lack knowledge about CP. Paget et al., in a qualitative study in Malawi, showed that lack of knowledge and confidence of health-workers were among the main barriers to a successful and comprehensive communication with parents of children with developmental disability, CP included [16]. In a recent report of an international meeting of pediatricians from Sub-Saharan Africa (including Benin), health-workers' training about CP was stressed as a key element to improve the quality of health-care provision, suggesting that health-workers generally lacked sufficient knowledge about CP [25]. Given the markedly religious socio-cultural context of Benin, that lack of a successful, supportive, and comprehensive process of diagnostic disclosure from health-workers might explain the large proportion of mothers still attributing the cause of CP to religious beliefs. Furthermore, our study showed that mothers with a limited level of education were more likely to attribute non-biomedical causes to their child's illness. We suspect that the terminology used by health-workers might not properly fit the comprehension capacity of mothers with a low literacy level, which could be related to language barriers arising from the numerous indigenous languages in Benin; there is no lexicon of biomedical terms across all 50 linguistic groups of Benin. Furthermore, the inclusion of a third party for translation might impede the success of the diagnostic disclosure.

Apart from the severity of the disability, we found that non-biomedical explanations and low-level of education were associated with a higher impact of caregiving on the family's quality of life. In this study of cross-sectional design, we could not determine the causal relationship between non-biomedical beliefs and high impact on family life. Spirituality, mainly a faith in God, has been reported as a coping strategy for caregivers of children with lifelong disability, which might explain why mothers perceiving a high impact referred to spiritual beliefs [26,28]. However, parents ascribing the cause of CP to metaphysical factors did not actually mention any identifiable cause; in the absence of clear understanding, their default was to refer to God's will or a curse. As such, we suspect that the spiritual beliefs reported might more entail a lack of knowledge than a coping strategy, and that this lack of knowledge led to the high impact perceived by the caregivers. Shevell and Shevell reported in their study in developed counties that successful diagnostic disclosure of cerebral palsy empowers parents to "effectively and directly address their feelings of weakness, vulnerability and loss of control" [27]. The inverse association between level of education with non-biomedical beliefs might explain its association with high impact of caregiving. Moreover, an association between maternal education and the impact on family life has been previously reported [18,29], suggesting that socioeconomically disadvantaged families might perceive a higher impact of caring for children with lifelong disability.

The findings discussed so far should be a wake-up call for health-workers in LMICs to shift from "child-centered" to a "family-centered" approach for pediatric services in relation to developmental disability. Providing families of children with developmental disability with the attention and support they need to understand clearly what has happened to their child and how best to cope would significantly alleviate the burden associated with the child's developmental disability *per se* and improve the child's outcomes [13,14,27]. The centrality of the family's well-being in the child's care system is well attested by evidence supporting the shift from child-centered to family-centered care services in HICs, where families' needs are considered and the families are seen as partners, collaborators, and indeed experts in the care provided to their child [14,15]. This model could be adapted and integrated into pediatric services in LMICs such as Benin through a systematic assessment of the family's quality of life and needs. This is feasible in a model including families (irrespective of their level of education) and at least one involved health professional: either a physician, or more likely a rehabilitation team member (physiatrists, orthotist, physiotherapist, speech and language therapist) could explain carefully and comprehensively the nature of the pathology and present strategies to build an adapted rehabilitation program that could enable the child to gain autonomy and encourage the family to be involved in positive changes [13,14,27]. To achieve this goal training of health-workers is fundamentally important and should include adoption of a family-centered model of pediatric services in LMICs. An example of such an adapted program is the "Getting to Know Cerebral Palsy" program, which has shown promising benefits for parents' quality of life and children' outcomes in a recent study in Ghana, another West-African LMIC [13].

Congruently, health-workers were not perceived as being available or supportive of caregivers. The most supportive people were close family members, who often shared with the family the financial and physical burdens arising from care of children with developmental disabilities in LMICs.

Beyond the physical and financial burdens associated with raising a child with CP, community folk beliefs from communities present another modifiable source of negative impact experienced by caregivers. Folk beliefs about the causes of developmental disability within communities in LMICs usually include supernatural power as punishment for sinful acts, along with bewitchment, leading to discrimination and isolation of families of children with developmental disabilities [5,7,17]. In this study, the children with CP were considered as "divinities of waters". Not only should families of children with disability be informed about the causes of CP, but educational workshops should extend to the whole community. It may be that caregivers' perceptions of CP are sometimes influenced by erroneous beliefs arising from their community, even though we did not find that stigmatization was significantly associated with mothers' perceptions in the present study. Community-based educational workshops should also include the "traditional healers or oracles" who are inevitably consulted by caregivers of chronic, ill-understood health issues in LMICs [12,17,28].

It is noteworthy that, despite the erroneous beliefs arising from the community, very few mothers reported negative attitudes towards their children, for example by avoiding public settings with the children, hiding their children from guests, or being tempted to get rid of their child. This high degree of acceptance suggests that mothers were more involved in positive attitudes and strengthening the family-support system would reinforce these positive attitudes for the well-being of the family and their child.

The mortality rate in our sample (six children out of 94, i.e., 6.4 and 6.9% for children aged less than 15 years) slightly exceeds the general mortality rate of 5.4% in similarly aged children between in Benin [30]. Elevated mortality has been previously observed in other LMICs like Ghana, where children with severe CP aged between 1 and 12 years were nearly six times more likely to die compared to children in the general population [13]. These findings raise questions about the effect that financial burden and community stigmatization may have on life expectancy of these children.

Limitations

We note several potential limitations of our study. We used a cross-sectional design, which does not allow causal inferences about associated factors. Moreover, our interviews were not sufficiently detailed to bring out more contextual factors associated with mothers' knowledge of CP. Our questions were limited to knowledge of causes of CP, and we thus missed perceptions about other aspects, such as treatment approaches and prognostics. Furthermore, we did not test the psychometric properties, and content validity and reliability of our semi-structured questionnaire about the perception of CP. By only investigating the burden of having a child with CP, we confined questions about negative aspects of social behaviors to some leading questions such as, "What do people say about your child in a negative way that hurts you?" Consequently, our study missed potential positive aspects of having a child with CP, such as the emergence of a social support network with other parents of children with developmental disability, as reported by Davis et al. [31]. However, we fear that the general perception of CP in Benin is highly negative, entailing considerable negative pressure on families. Indeed, we are convinced that an extensive program to inform about CP is needed in Benin, which would include the development of family support and encourage more a positive attitude towards CP.

Furthermore, questionnaires such as the IOFS were directly translated into Fon, the predominant dialect of southern Benin. However, obtaining a validated back/forward translation of the questionnaire in local languages was not possible, especially since many languages of Benin lack a written form. While the language of education is French, efforts are ongoing to integrate Fon and other local languages in educational program. Language issues may contribute to the association between mothers' education level and IOFS scores. However, our results in this regard were congruent with previous studies [18,29].

We modified some items of the FSS to make it more adapted to our context, aiming to bring out potential sources of support specific for health-workers in a developing country, without having obtained prior validation of the modifications. While that may have affected psychometric properties of the questionnaire, we note that the FSS mainly served to investigate the sources of support and how helpful mothers found health-workers, which would not likely be impaired by our minor modifications.

In the regression analyses, some subsamples' sizes were small, this limiting the interpretation of the results. We did not record monthly family incomes or information about mothers' psychological stress, which might both have impacted on caregiving [18]. The present findings call for further investigation by studies of longitudinal design to learn how an adapted family-support system might improve caregivers' perception of CP and their well-being, to the benefit of children's outcomes. Fourteen mothers declined to participate (>10%), which might bespeak a particular stigmatization in their communities.

Conclusion

The lack of biomedical knowledge of caregivers of children with CP in Benin, especially among those with a limited level of education, is associated with a highly negative impact on the family's life. Very few caregivers had received information about CP from health-workers. We see a need to shift towards a family-centered approach of child-care in LMICs. Health-workers should in their routine clinical practice be providing families of children with developmental disability with the information, support, and attention they need. Furthermore, apart from the physical and financial burden associated with caring for children with CP, caregivers experience stigmatization due to folk beliefs that children with CP are cursed; we can only hope that such perceptions shall yield to education programs.

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Ethical approval

The ethic committee of the rehabilitation department of the National University Hospital (Cotonou, Republic of Benin) approved the study. Caregivers of all participants provided their informed consent.

Author contributions

ESS and YB performed the data analysis and drafted the paper. ESS, DH (both experienced pediatric physiotherapists) contributed to data collection. All authors contributed to the study design, reviewed and accepted the final manuscript.

Disclosure statement

No potential conflict of interest was reported by the author(s).

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