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Complete List of Authors:	Campos, Taynah; Universidade Federal do Rio Grande do Norte, Schiariti, Veronica; University of Victoria, Gladstone, Melissa; University of Liverpool Institute of Translational Medicine, Women and Children's Health Melo, Adriana; Instituto de Pesquisa Professor Joaquim Amorim Neto Tavares, Jousilene; Instituto de Pesquisa Professor Joaquim Amorim Neto Magalhães, Adriana; Federal University of Rio Grande do Norte – UFRN-FACISA Longo, Egmar; Universidade Federal do Rio Grande do Norte,
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How Congenital Zika Virus impacted my child's functioning and disability: a Brazilian qualitative study guided by the ICF

Taynah Neri Correia Campos, Verónica Schiariti, Melissa Gladstone, Adriana Melo,
Jousilene Sales Tavares, Adriana Gomes Magalhães & Egmar Longo

Correspondence to:

Egmar Longo, PhD
Graduate Program in Collective Health – Federal University of Rio Grande do Norte – UFRN-FACISA
Rua Vila Trairi S/N, Centro, Santa Cruz, RN
59200-000, Brazil, +5584 32912411
egmarlongo@yahoo.es

Taynah Neri Correia Campos, MsC
Graduate Program in Collective Health
Federal University of Rio Grande do Norte – UFRN-FACISA
Rua Vila Trairi S/N, Centro, Santa Cruz, RN
59200-000, Brazil

Verónica Schiariti, PhD
Adjunct Associate Professor – Division of Medical Sciences
University of Victoria
Victoria, BC, Canada

Melissa Gladstone, PhD
Professor of International Child Health and Neurodevelopmental Paediatrics
Department of Women and Children's Health
Institute of Translational Medicine
University of Liverpool
Eaton Road, Liverpool, L12 2AP, UK

Adriana Melo, PhD
Instituto de Pesquisa Professor Joaquim Amorim Neto
Campina Grande – Paraiba Brazil

Jousilene Sales Tavares, PhD
Instituto de Pesquisa Professor Joaquim Amorim Neto
Campina Grande – Paraiba Brazil

Adriana Gomes Magalhães, PhD
Graduate Program in Rehabilitation Sciences – Federal University of Rio Grande do Norte – UFRN-FACISA
Rua Vila Trairi S/N, Centro, Santa Cruz, RN
59200-000, Brazil

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Abstract

Introduction: The Zika Virus outbreak in Brazil had devasting social, medical and financial consequences. Many researchers and clinicians are now following up the children to understand the impact of the Zika on child development, functioning and disability, but outcome measured are heterogenous, and often; it is not clear how meaningful they are to families and children. This study aimed to identify the parents' perspectives on relevant areas of functioning and disability that should be included as outcome measures for children with congenital Zika Syndrome (CZS), guided by the International Classification of Functioning, Disability and Health (ICF).

Methods: Participants were recruited from child rehabilitation centers in two states in northeastern Brazil. Parents of children aged 0 to 6 years with confirmed CZS were included. Interviews were conducted using focus groups. Saturation was achieved with six focus groups. Content mapping followed the WHO's ICF linking rules. Three raters analyzed the content using NVIVO 11.

Results: eighty-eight percent of participants were mothers, average age 30 years. Most of their children were male (59%), all GMFCS level V. Overall, 825 themes were mapped to the 36 ICF categories. Functioning and disability themes were predominantly linked to environmental factors, activities and participation, and body functions. Although parents mentioned areas across all ICF domains, they reported that areas of mobility, eating and recreation were very relevant for them. In addition, environmental factors were highly identified as barriers, specifically services, policies and access to assistive devices. The most predominant facilitators were immediate family support, kind relationships with therapists and extended family.

Conclusions: Although parents emphasized issues related to mobility, their greatest concerns involved environmental factors, such as access and quality of health and social services, systems and policies. These results reinforce the importance of including the parents' perspective when selecting or developing outcome measures for CZS.

Strengths and limitations of this study

- This is the first qualitative study that has explored the perception of parents regarding the aspects considered relevant in children with CZS based on the ICF model.
- Focus groups provided rich qualitative data and in-depth understanding of the perspectives and experiences of parents/caregivers of children with CZS.
- Several ICF categories were identified by parents as very relevant and these need to be considered within a global context for the establishment of a set of main outcomes and for implementing rehabilitation interventions.
- The role of environmental factors in the statements of parents reflects the real need to include the context as an aspect to be assessed in the follow-up of children with CZS.
- The sample was extracted from three Brazilian cities and generated the views of mothers predominantly with very few fathers taking part. In addition, all children had GMFCS level V.

INTRODUCTION

The Zika virus (ZIKV) was identified in Brazil at the beginning of 2015. Concurrently, rates of microcephaly and other congenital brain abnormalities increased, thus suggesting a causal relationship between this congenital infection and the clinical presentation.[1] Since then, clinical reports have documented the teratogenic effect of ZIKV in pregnant women, particularly with infection in the first trimester.[2, 3, 4] The most prominent among the congenital anomalies is microcephaly, but other manifestations such as; spasticity, seizures, eating difficulties, irritability, ocular abnormalities, hearing loss, calcifications, cortical disorders and ventriculomegaly in neuroimaging have been described thus suggesting a new syndrome, called the Congenital Zika Syndrome (CZS).[5, 6, 7] There is still no complete definition of the scope of this new health condition, and adequate outlines are still needed. As a result, surveillance systems are aware of a wider spectrum of congenital malformations which may be associated with ZIKV infection.[8, 9]

A precise estimate of the ZIKV infection incidence is difficult to determine due to the variations between countries regarding the commitment to notifications and the small number of individuals who manifest the health condition caused by ZIKV.[10] There are currently 3,406 confirmed cases of children with microcephaly and/or other neurological signs due to CZS in Brazil, with a further 2,596 cases under investigation.[11]

Given the severity of the syndrome, the scientific community has mobilized efforts to understand the mechanisms of this health condition and to establish outcome measures, which means to define what should be measured and reported in all trials in this area, since creating a standardized core outcome set (COS) validated by specialists in the field will reduce the heterogeneity of the studies and the result bias, and further enable development of meta-analyzes and data sharing between the studies.[12]

The Core Outcomes Measures in Effectiveness Trials (COMET) initiative identifies and disseminates the most relevant outcomes for each clinical health situation through a database and recommends (among other aspects) the involvement of families in developing these outcomes in order to ensure that the research process is more relevant and appropriate to the patient's needs.[13, 14] In this context, parents are encouraged to express their children's strengths and weaknesses in terms of function/structure, activity and participation, environmental factors and personal factors, following the International Classification of Functionality, Disability and Health (ICF) which incorporates a biopsychosocial model of disability.[14] The ICF can be used as a guiding framework which comprises functioning and disability as a dynamic interaction between health and health-related disorders and contextual factors, including personal and environmental factors. In addition, the ICF establishes a scientific basis for studying health determinants and their related conditions, as well as providing a common language for describing health.[15]

This study will contribute to the development of "Zika Cos and congenital infections: a study to develop the Core Outcome Set (COS) for children with CZS and other congenital infections" project which is coordinated by the University of Liverpool in partnership with the University of Victoria (Canada), and integrates the 1st phase of this initiative, consisting of the qualitative stage.[16] Therefore, the objective of this study is to identify the parents' perspectives on relevant areas of functioning and disability which should be included as outcome measures for children with CZS, guided by the ICF.

METHODS

This study implemented a qualitative methodology with an exploratory approach through focus groups. We chose to use a qualitative approach as it enables investigation and in-depth understanding of the perspectives and experiences of parents/caregivers of children with CZS. Focus groups, in particular, provide the potential to explore and clarify points of view and reveal dimensions of understanding which would be less easily accessible in other data collection formats.[17, 18]

This research was approved by the Ethics and Research Committee of the Faculty of Health Sciences of Trairi/Federal University of Rio Grande do Norte, it was also re-approved at Liverpool University Ethics Committee and developed in three Brazilian rehabilitation centers for CZS in the states of Rio Grande do Norte (Physical Therapy School Clinic at UFRN-FACISA, in Santa Cruz) and Paraíba (Center for the Care of Children with Microcephaly at the Professor Joaquim Amorim Neto Research Institute - IPESQ, in Campina Grande, and Physical Therapy School Clinic at UNIPE, in Joao Pessoa). Parents/guardians signed a Consent Form after being provided with an information sheet which was read out to them. Parents also signed an authorization for Voice Recording and use of images, (considering Resolution 466/2012 of the National Health Council which provides for regulatory guidelines and standards for human research).

Participants were purposively selected with members having important common characteristics. These inclusion criteria included being; parents/caregivers of children with confirmed diagnosis of CZS by polymerase chain reaction (PCR) or presumed diagnosis based on obstetric ultrasound, transfontanellar ultrasound, computed tomography (CT) and magnetic resonance imaging (MRI), who were from zero to six years old and who lived in the area covered by the study (states of Rio Grande do Norte and Paraíba) and who attended the respective rehabilitation services. This study did not include parents whose children had congenital syndromes due to other etiologies.

All parents responded to a sociodemographic questionnaire with information about their relationship with the child, marital status, age, income, government benefit, education, housing and self-reported health, in addition to information from children about gender, age, mobility, rehabilitation, frequency day care centers or schools, and difficulties with vision, hearing, sleep and epilepsy. The physiotherapist who accompanied the children in the rehabilitation centers also classified their motor abilities using the Gross Motor Function Classification System (GMFCS). This is an age-specific scheme designed for children with CP based on five levels of gross motor function, ranging from level I (most able) to level V (least able)[19] and provided some clear information on the severity of the children whose parents took part in the study. The study flowchart can be viewed in Figure 1 and the COREQ guidelines were considered.

The focus groups were conducted by research assistants, trained both in qualitative studies and in conducting focus groups. We continued to do focus groups until saturation was reached where the testimonies became repetitive, predictable, and were not providing any additional information. Saturation was achieved by the sixth group. Furthermore, we decided not to exceed six parents/caregivers per group in order to enable effective participation of the participants and appropriate discussion of themes.

A questionnaire adapted from a previous ICF-based qualitative study was used to conduct the focus group, covering the different ICF domains (Supplementary material 1).[20] Visual tags were produced with images from the ICF illustrated gallery (Supplementary material 2)[21] to assist in understanding the concepts, as studies indicate that visualization has the potential to support data production during the qualitative interview process.[22] The duration of the focus groups ranged from 60 to 90 minutes. The participants were given a therapeutic toy developed by research team at the end of the groups, called “fun fishing”, and were encouraged by the professional team to use it as one of their tools for stimulation and fun.

All groups were recorded and later transcribed in full. Names and identification characteristics were excluded from the transcripts. The transcribed content was analyzed by three researchers experienced in qualitative research (TC, EL, VS). They split the content into significant units with using NVIVO software, version 11.0 (QSR International, 2019), and associated them with each ICF domain and category using the WHO's linking rules[23] as follows: TC and EL, trained in ICF, performed a blind reading of the content and established the ICF domain and category for each significant unit of the interview to which it was judged to be relevant. VS, a specialist in ICF and with extensive experience in childhood disability, analyzed the disagreements of the two researchers, establishing consensus in relation to the domain/category of the ICF regarding the significant units. Next, the content was analyzed by the researchers who interpreted the data through an inductive and deductive analysis process based on a permanent dialogue throughout the process to ensure consistency and reliability of the interpretations. Any verbatim which did not exceed "five mentions" was discarded as it was felt not to be representative enough of the data.

Aspects of credibility, transferability, reliability and confirmability were considered during all stages in order to guarantee the quality of the qualitative research.[24] Credibility was ensured by thoroughly conducting six focus groups until response saturation was reached, with subsequent blind analysis, review and triangulation, and then a consensus was reached after meetings and debates regarding the interpretation of results. The researchers have general experience and robust training in the area of childhood disabilities and in the use of the ICF for neuro disabilities. Transferability occurred through observation of the rigor to which the research group stopped to describe how the data was obtained through the focus groups, the selection and description of the sample, which in this case involved the participants. Reliability was guaranteed by detailing the data collection processes, presenting the multiple steps followed by the researchers, and showing how the pair analysis and data interpretation took place. A measure of reliability through using the kappa statistic was also used to verify the agreement between the judges, with results indicating almost perfect agreement (0.916) considering the 95% confidence intervals. Finally, verification was achieved through a peer discussion of the data at each stage of the analysis by the research team.

Patient and public involvement: No patient involved.

RESULTS

Six focus groups were conducted with 32 caregivers of children with severe neurodevelopmental disorders associated with CZS. Information related to children can be found in Figure 2 and the characteristics of families are in Figure 3. Most of the children were male, had vision problems and underwent rehabilitation follow-up. The average age was 32.5 (SD= 6.2) months and all presented GMFCS level V Although none of the children walked, only 37.5% of them had a wheelchair.

Thirty-six categories of the ICF were identified (without considering the personal factors), as demonstrated in Figure 4. This figure represents the parents' perspectives on relevant areas of functioning and disability of their children.

The parents mentioned relevant areas that covered all ICF domains, but a greater diversity of categories was observed in the domains of activity and participation and bodily functions. However, the environmental factors domain led the ranking of 12 categories, as can be seen in Table 1.

	CATEGORY	QTY	DOMAIN
1º	e580 Health services, systems and policies	59	Environmental Factors
2º	b760 Control of voluntary movement functions	37	Body Functions
3º	e115 Products and technology for personal use in daily living	33	Environmental Factors
4º	d415 Maintaining a body position	33	Activities and Participation
5º	e110 Products or substances for personal consumption	31	Environmental Factors
6º	b230 Hearing functions	31	Body Functions
7º	b134 Sleep functions	31	Body Functions
8º	d450 Walking	28	Activities and Participation
9º	e120 Products and technology for personal indoor and outdoor mobility and transportation	25	Environmental Factors
10º	b210 Seeing functions	24	Body Functions
11º	d445 Hand and arm use	23	Activities and Participation
12º	s750 Structure of lower extremity	22	Body Structures

Table 1: Table demonstrating the ranking of the 12 most frequent categories of the ICF mentioned by parents

Table 2 shows the distribution of categories related to the environmental factors domain in detail.

Environmental Factors		
e580 Health services, systems and policies	59	33,0%
e115 Products and technology for personal use in daily living	33	18,4%
e110 Products or substances for personal consumption	31	17,3%
e120 Products and technology for personal indoor and outdoor mobility and transportation	25	14,0%
e310 Immediate family	16	8,9%
e540 Transportation services, systems and policies	9	5,0%
e410 Individual attitudes of immediate family members	6	3,4%
TOTAL	179	100,0%

Table 2: Table of areas of the ICF considered important for parents within the area of Environmental factors

Among the categories, **Health services, systems and policies** (e580) was the most expressed by parents and received 59 mentions. This category includes the prevention and treatment of health problems, the provision of rehabilitation services and the promotion of a healthy lifestyle. In the following verbatim, it is possible to observe the father's desire to have access to a rehabilitation service in his own city.

> *"Also that in Alagoas state has been the same possibilities that here in Campina, to don´t need to move from one place to other, it´s so complicated spend 3 months here, searching treatment that there it should have. I don´t know they can´t leave this treatment there, to every states have, for us don´t need to move a lot to do a treatment. For them could at least sit, maintain their trunk, catch and walk."* Mother 20.

Another area which emerged often by the parent/caregivers as a barrier was; the lack of information, of professionals specialized in the management of CZS, the desire for healing and the difficulty in transport to travel to rehabilitation services in other larger cities. Examples of this are evidenced in the statements below.

> *"I guess that it would help a lot if the access is facilitated, on your city, if I have physiotherapy on my city, sure I would go more often."* Mother 03.
>
> *"I would like science produces medicines for this disease, because is a new disease. Other thing, my great difficulty is absent of answers, you go to see a doctor with a child, and doctor is stalling, he never says the reality for you."* Mother 24.
>
> *"If our city would have the treatment, moving is tiring. And if when we arrive would be taken care. We suffer some experiences, in some days I spent many hours."* Mother 04.

Some parents pointed to assistive products and technologies as facilitators. They described how adapted or specially designed equipment improved the functioning of their children and contributed to greater participation;

> *"For E* which facilitates activities it´s on first the glasses, because I take her glasses and the vision been worse, put the glasses and she animates."* Mother 06.

The support of the nuclear family was expressed by participants of all focus groups always as a facilitator of the environment, as described below.

> *"What does facilitate? I have a lot of support from my husband and my sister. I live close to my sister, my parents don´t live in the same city than I, but they always are present, always, until the beginning they are present. My family helps me a lot, and this helps me, when I want to go out and can´t take she together, I leave her with my sister. Then, the family support it´s the main factor that facilitates, because alone, it´s hard."* Mother 14.

Table 3 presents the categories referring to the activity and participation domain.

Activities and Participation		
d415 Maintaining a body position	33	14,5%
d450 Walking	28	12,3%
d445 Hand and arm use	23	10,1%
d550 Eating	21	9,3%
d920 Recreation and leisure	19	8,4%
d430 Lifting and carrying objects	15	6,6%
d110 Watching	15	6,6%
d330 Speaking	12	5,3%
d760 Family relationships	11	4,8%
d160 Focusing attention	11	4,8%
d410 Changing basic body position	9	4,0%
d455 Moving around	9	4,0%
d710 Basic interpersonal interactions	8	3,5%
d730 Relating with strangers	7	3,1%
d440 Fine hand use	6	2,6%
TOTAL	227	100,0%

Table 3: Table demonstrating areas of the ICF within Activity and participation found as important to parents

As shown in Table 3, the most expressive categories were those related to movement, such as **Maintaining a body position - d415**, **Walking - d450**, and **Hand and arm use - d445**. Such aspects were pointed out by the parents as being responsible for limiting activity and restricting participation in daily activities, as evidenced in the statements below.

> *"M* has difficulty to maintain the body position, when I put him stand position supported on the wall he can stay a long time. Sit, he also doesn´t sit alone, but if you put him, he stays, he creeps in, rolls. He doesn´t go to lying down position to sitting, but to sitting to lying he can."* Mother 15.

> *"Eat, she holds but doesn´t know how put it in her mouth."* Mother 01.

> *"His major difficulties are to catch things, he doesn´t catch, we need to put in his hand, than he holds, if he needs to pick up some think, he doesn´t do, holds if I open his hand he catchs, because is difficult for him to open his hand"* Mother 08.
>
> *"I guess it´s too hard these children who doesn´t walk, doesn´t sit yet, if when arrive in some place and it has a wheelchair, if have how to sit him, but he doesn´t sit. My great problem is his weight, it´s my concern."* Mother 17.

Finally, the areas of Body Function and Structure identified most by parents is shown in Table 4.

Body Functions			
b760 Control of voluntary movement functions		37	16,70%
b230 Hearing functions		31	14,00%
b210 Seeing functions		24	10,80%
b510 Ingestion functions		19	8,60%
b134 Sleep functions		31	14,00%
b710 Mobility of joint functions		18	8,10%
b280 Sensation of pain		16	7,20%
b735 Muscle tone functions		15	6,80%
b320 Articulation functions		10	4,50%
b152 Emotional functions		8	3,60%
b440 Respiration functions		7	3,20%
b770 Gait pattern functions		6	2,70%
	TOTAL	222	100%
Body Structures			
s750 Structure of lower extremity		22	78,60%
s730 Structure of upper extremity		6	21,40%
	TOTAL	28	100%

Table 4: Table demonstrating the categories of d Body Functions and Structures (within the ICF) identified as important by parents

Regarding the body function domain, there was a predominance of the category related to the **Control of voluntary movement - b760**. Many parents expressed concerns about voluntary movements, both simple and more complex, coordination, support functions of the upper and lower limbs and right-left motor coordination, as shown below in this mother's speech.

> *"Movements control, L* has a lot of difficulties, she can´t lift her arm, her hand. About mobility function, she also has difficulties, she can´t lift her arm and hand, she doesn´t have a lot problems with her legs, but can´t move."* Mother 24.

Parents also pointed out problems related to **Hearing functions - b230**, **Seeing functions - b210** and **Sleep functions - b134**, as can be seen below.

> *"The listen function doesn´t work well , only in one side , in the other it doesn´t work , we would make a raffle to earn money and buy a device , because on exam showed that the left one he listens, but on the right he doesn´t anything."* Mother 22.
>
> *"About vision function, he doesn´t see everything, his vision is low, he has difficulties."* Father 01.
>
> *"He has sleep difficulties, but when he falls on sleep (begins to sleep) he sleeps the whole night."* Mother 28.

For "body structure", the category relating to **Structure of lower extremity - s750** most commonly emerged within the parent focus groups. Many parents described issues with foot positioning and how this prevented walking and standing.

> *"The knees, because she could craw and it something that I guess she won´t do. And her feet, to can walk, that will be a great difficulty, because she has a dislocation, we don´t know if she will walk or if she will need a wheelchair for the rest of her life."* Mother 27.
>
> *"About S* is his little foot, when I will put him on stand up position, his foot turn to the side."* Mother 10.

DISCUSSION

This is a pioneering study which revealed the perceptions of parents and caregivers regarding the aspects considered relevant in children with CZS based on the ICF model. Parent/caregiver views, such as these, deserves consideration in constructing core outcome set of measures for this health condition. The use of the ICF as a guide for this study enabled us to provide a holistic model which enables the care perspective to be extended beyond problems with the functions and structures of the body in children with CZS. Despite the number of new cases becoming more stabilized, the complex clinical presentation of CZS and its effect on children's developmental trajectories requires standardized and individualized care during their growth.[25] It also requires researchers to think a bit beyond outcomes which only relate to functioning such as those often measured in child development measures.

Several ICF categories were identified by parents as very relevant for them and their lives and these need to be considered within a global context for the establishment of a set of main outcomes and for implementing rehabilitation interventions.[26] The role of environmental factors in the statements of parents reflects the real need to include the context as an aspect to be assessed in the follow-up of children with CZS.[27, 28] Barriers such as those relating to the lack of specialized professionals who can support the clinical aspects of children with CZS need to be addressed.[29] The CDC has produced a pocket guide summarizing pediatric guidelines on initial assessment and outpatient treatment,[30] but a carefully planned public health approach with contributions and partnerships between specialists in various clinical and public health disciplines, improving the ability to anticipating needs, providing adequate care and ensuring that children reach their full potential is very necessary.[31, 32] This is particularly crucial to really address the environmental issues which so hinder many parents and families.

Many parents/caregivers in our study expressed a desire to receive all treatments and support in a single place or city. Moving from place to place for services and different types of rehabilitation in different places/cities is a real barrier for families. Parents in our study signaled a definite need for reorganizing health services in order to provide more of a "one stop" approach to supporting them. This same aspect has been highlighted in other studies.[33] From our study, it is clear that services for support and rehabilitation in a single location minimize problems such as low attendance and/or avoidance. Lack of attendance of course interferes hugely with creating good outcomes through focused, context-based and evidence-based rehabilitation.[34]

Parents who took part in our study voiced particular concerns around aspects of their children's movement. This permeated the domains of activity and participation, function and body structure and was, of course, expressed as a limiting factor in their participation in daily and leisure activities. It was clear that parents were disappointed and were desperate for a change in mobility of their children and had had little counselling about adaptations to support children in their daily lives through other means. The focus still seems to be on the perspective of "fixing" the disability through therapies.[35]

Alongside this, parents voiced their desires relating to gait acquisition (walking) in the context of their comparison of their children with typical children. This is well documented in previous research with children with cerebral palsy (CP).[36] We know that many parents who have children who will never walk often still resist early introduction of mobility aids such as adapted toy cars despite the fact that studies involving children with CP with severe motor impairment demonstrate positive results using these adaptations in terms of autonomy, self-esteem, social skills and participation.[37] At the same time, a recent study has shown that promoting participation can result in improved body function and structure in children and young people with physical disabilities.[38]

Within the verbatim from parents is a constant viewpoint surrounding the child's incapacity and a desire for healing. There is little acceptance of the child for what they are within their context. This negative view of disability is common for parents with children with similar conditions,[39] with parents tending to focus concerns on the challenges of their children in their daily activities, expressing factors which negatively affected their children's performance.

Our research has demonstrated what areas of the ICF parents of children with congenital infections such as CZS consider important. This is extremely important when attempting to establish a core set of measures which should be used as outcomes for children with CZS in the future. We know that it is essential to also consider the child's own perspective as to what he/she considers to be relevant in outcome measures, however, children with CZS in this case are not yet old enough to express their opinions.

The results obtained in this study must be interpreted taking into account some limitations. The sample was extracted from three Brazilian cities, from public or philanthropic rehabilitation centers, so our findings may not represent the perspectives of all parents of children with CZS. Our study generated the views of mothers predominantly with very few fathers taking part. They may have a very different perspective.

CONCLUSION

This is a unique study which aims to understand the views and perception of parents with regard to the needs of their children with CZS framed within the perspective of the ICF. Although our series of parents did concentrate on issues related to the lack of movement of their children, overall their emphasis of discourse was centred around environmental factors. These factors included issues such as; the context of services, systems and policies for prevention and treatment of health problems through rehabilitation as well as factors supporting a healthy lifestyle to promote the physical and psychological well-being and social status of their children.

Our results reinforce the importance of including the parents' perspective in the development of a core outcome set. Parents are the ones who live and experience the main limitations and potential of their children and it is them who highlight the urgent need for environmental changes to improve the lives of children with CZS and their families in Brazil. In the future, when children with CZS are able to express their own opinions, we should also hear their views as to what they consider relevant for outcome measures.

Figure 1 – Study flowchart

Figure 2 – Sociodemographic data of children

Figure 3 – Sociodemographic data for parent/caregivers

Figure 4: ICF categories representing the parents' perspective

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Contributors: VS, MG and EL designed the study. TNCC recruited the participants and conducted the interviews. VS, MG, EL, TNCC reviewed and refined the data. TNCC wrote the article. VS, MG, EL, AM, JST and AGM revised the article. TNCC received qualitative research training through participation in a qualitative health research methods course at the Federal University of Rio Grande do Norte/Brazil.

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Ethics approval: This research was approved by the Ethics and Research Committee of the Faculty of Health Sciences of Trairi/Federal University of Rio Grande do Norte (75053417.1.0000.5568), it was also re-approved at Liverpool University Ethics Committee (#2083)

Patient and public involvement: No patient involved

Figure 1: Study flowchart

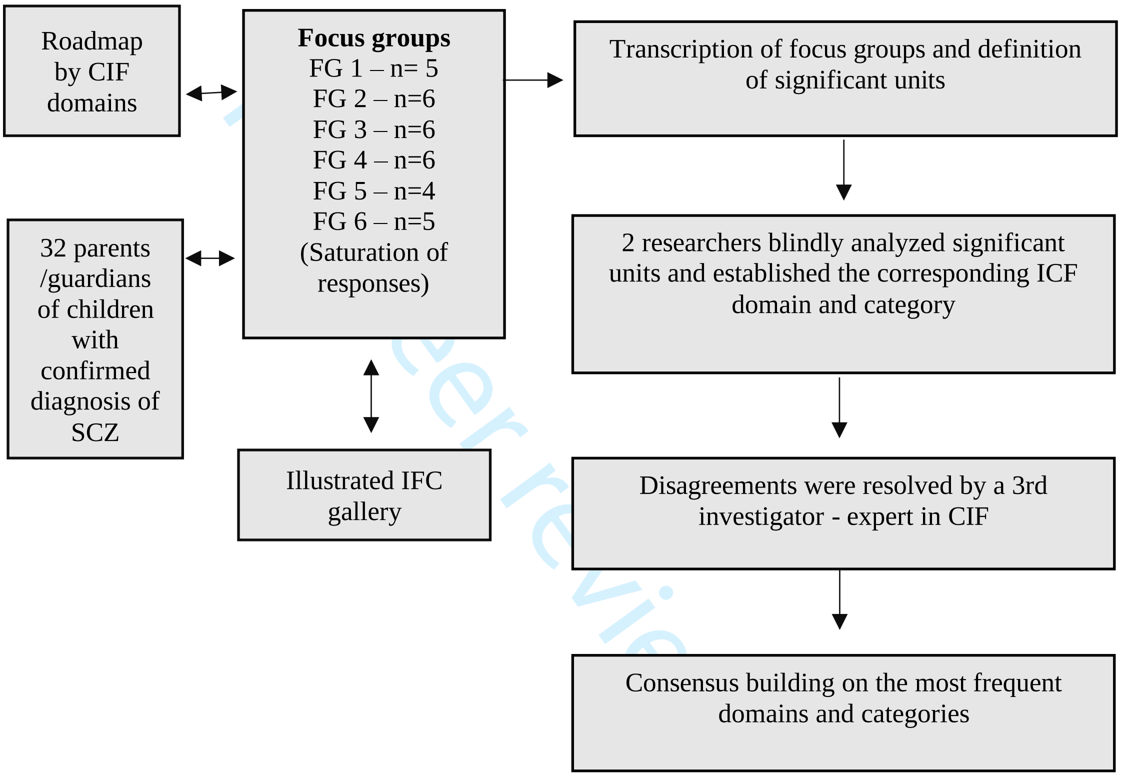


Figure 2 – Sociodemographic data of children

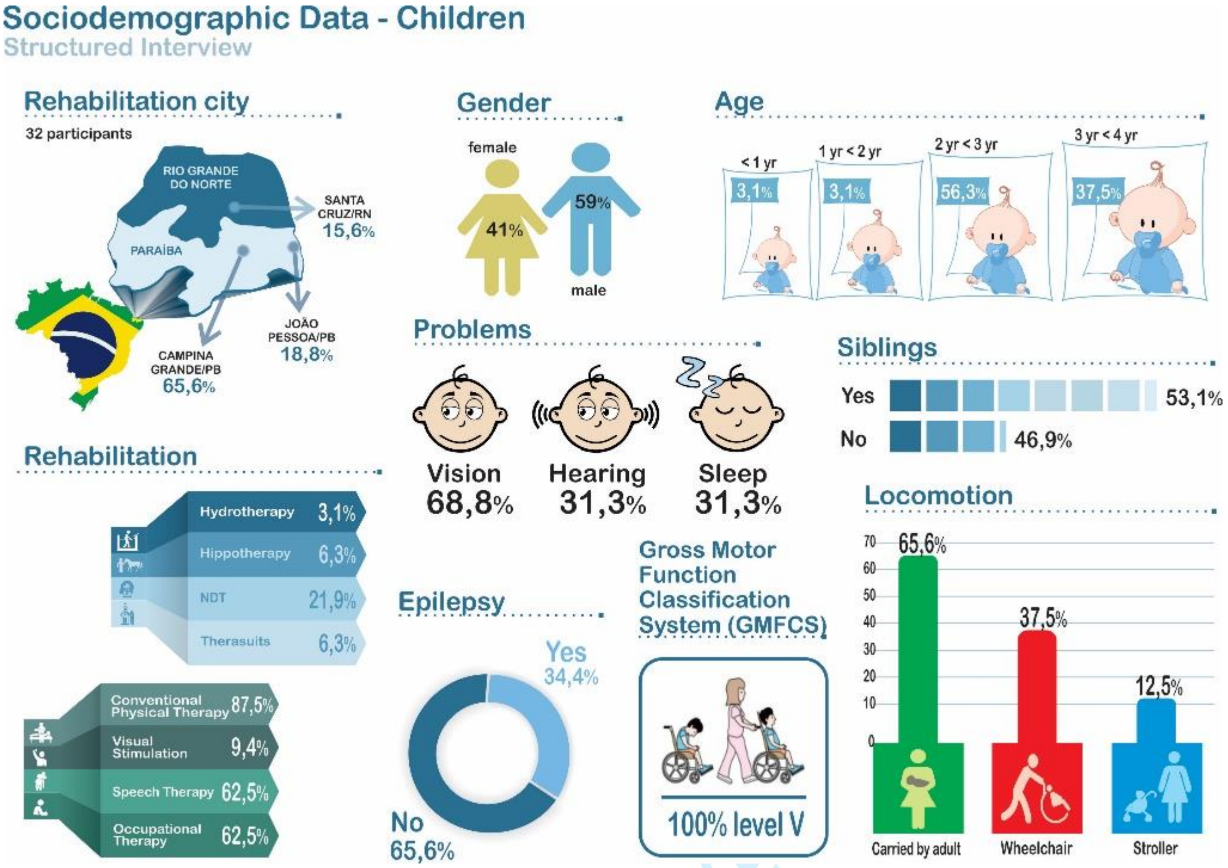


Figure 2 – Sociodemographic data of children

Figure 3 – Sociodemographic data for parent/caregivers

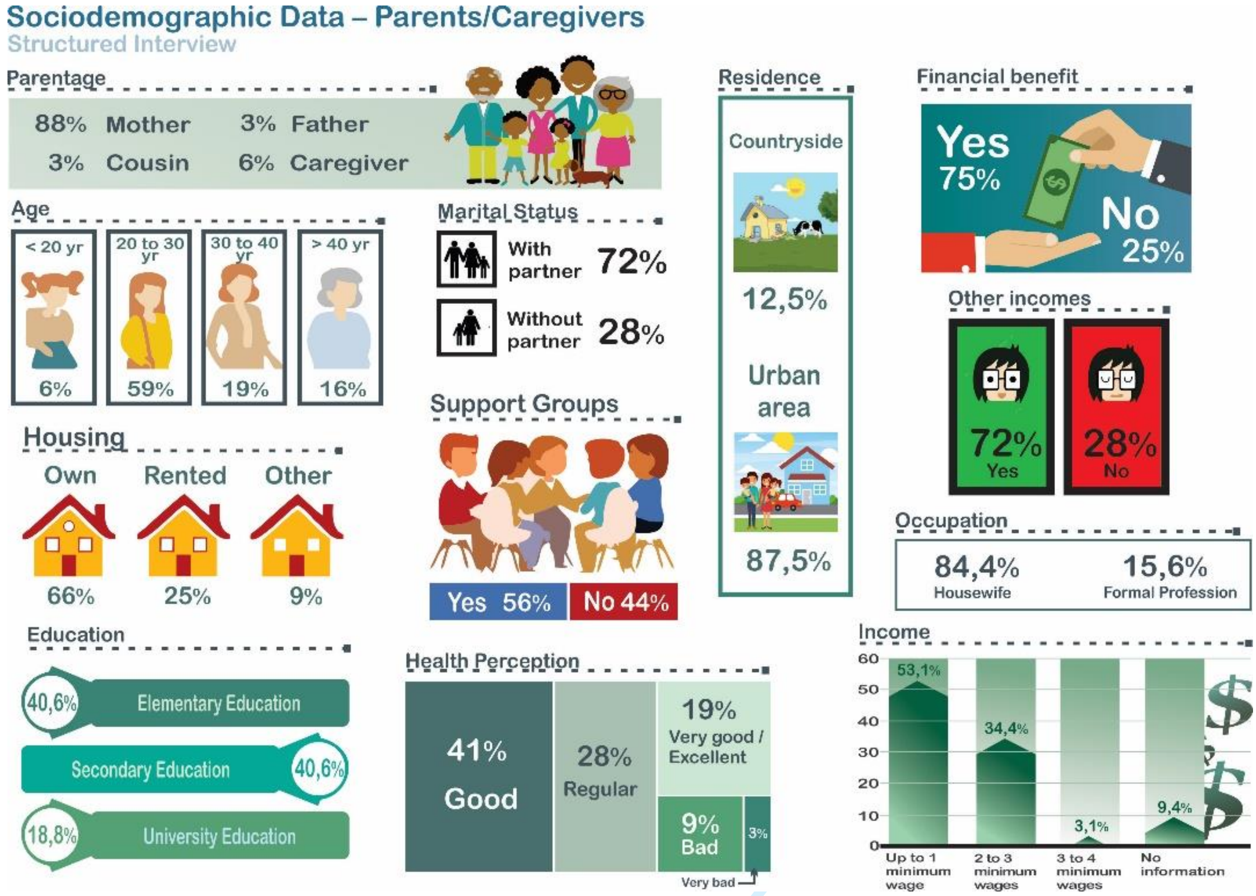


Figure 3 – Sociodemographic data for parent/caregivers

Figure 4: ICF categories representating the parents' perspective

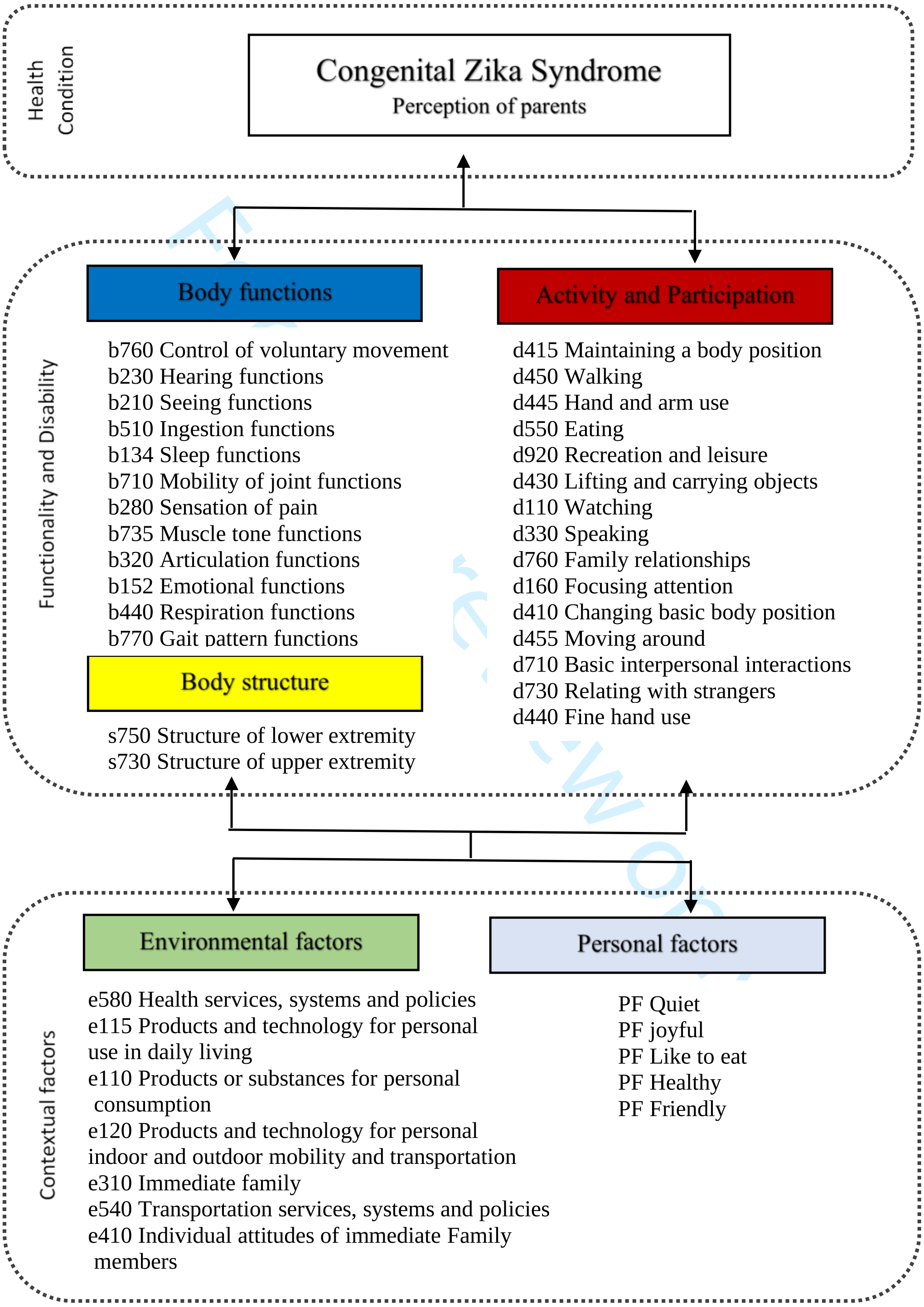


Figure 4: ICF categories representating the parents' perspective

Supplementary material 1

ZIKA FOCUS GROUPS

Aims of focus groups:

To elicit **what outcomes** are considered important to parents and caregivers of children with Zika virus and therefore what should be included in an outcome measure.

Population:

Parents and/or Caregivers of children who have had infants born with congenital infections or similar conditions who have children from 0-10 years and who are willing to take part in a focus group. We will attempt to sample parents of children with a range of severity of disorders and will attempt to sample from both urban and rural settings.

Framework:

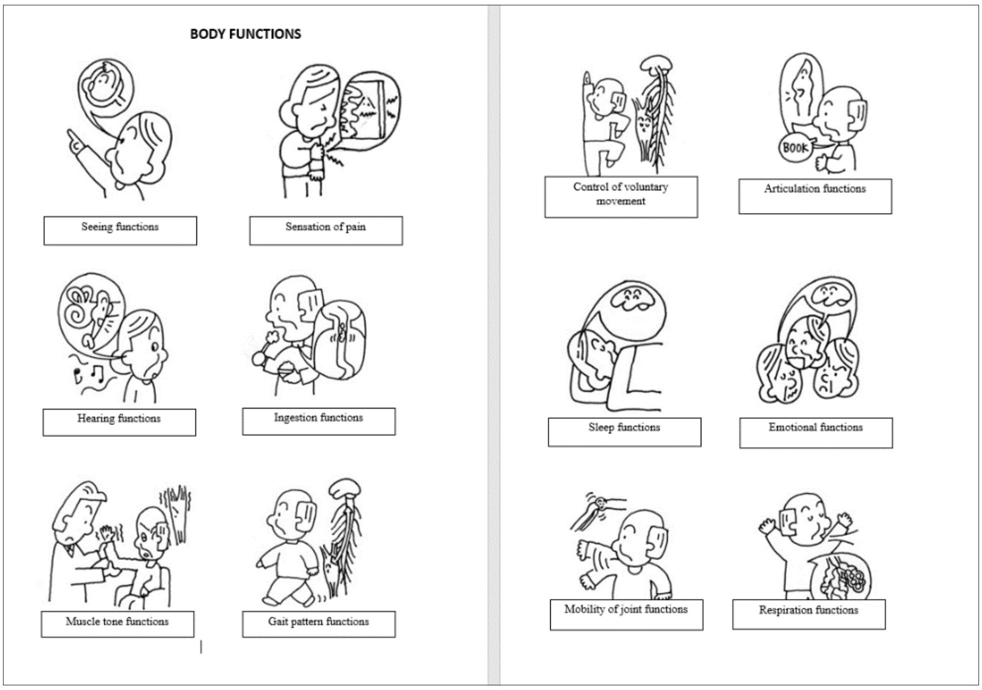
The ICF and ICF Core Sets for children and youth with CP will be used as an overarching framework to ensure a comprehensive representation of outcomes.

Questionnaire:

1. If you think about your child as an individual, what **personal characteristics** are important about him/her? (Moderator provide an example) **PERSONAL FACTORS**
2. If you think about the **daily activities**, (Moderator provide an example, show pictures of activities)
   a. What activities your child is able to do?
   b. What activities does your child find hard/difficult to do?
3. If you think about the **physical and social environment** (**family, school, and neighborhood)** of your child, (Moderator provide an example, show pictures, ask HOME, SCHOOL, COMMUNITY)
   a. What do you find **helpful or supportive** in facilitating participation in different activities?
   b. What **challenges or barriers** does your child experience?
4. If you think about the **body** of your child, which body parts cause your child some difficulties, if any? (Moderator provide an example, may be a picture of the body and they can mark areas with an X)
5. If you think about the **body** of your child, what parts of his/her body do not work the way it supposes to, if any? (Moderator provide an example, may be a list from the ICF Core Sets then they can mark functions from the list) **BODY FUNCTIONS**
6. Tell me about the **biggest problems** for you at the moment.
7. If people were looking into treatments for your child, what are the things you might **hope to improve:**
   a. In terms of their **daily activities?**
   b. In terms of making it **easier for your child to participate** in daily activities (in the neighbourhood, at home with the family and at school or nursery, if they attend)?

Supplementary material 2

Visual cards with images from the ICF illustrated gallery



Consolidated criteria for reporting qualitative studies (COREQ): 32-item checklist

Domain 1: Research team and reflexivity			
Personal Characteristics			
1	Interviewer/facilitator	Which author/s conducted the interview or focus group?	The first and the last authors conducted the focus groups.
2	Credentials	What were the researcher's credentials? E.g. PhD, MD	Msc Candidate - nurse; PhD- Physical Therapist.
3	Occupation	What was their occupation at the time of the study?	Msc candidate - nurse; Professor.
4	Gender	Was the researcher male or female?	Female.
5	Experience and training	What experience or training did the researcher have?	She was training by the last author, who has a lot of experience conducting focus group in Brazil and Spain. She was following previous qualitative studies as a trainee before start her research.
Relationship with participant			
6	Relationship established	Was a relationship established prior to study commencement?	No.
7	Participant knowledge of the interviewer	What did the participants know about the researcher? e.g. personal goals, reasons for doing the research	Participants were informed that they would be interviewed by a nurse who was conducting research in partnership with the university of liverpool, with the aim of developing a set of outcome measures for children with CZS.
8	Interviewer characteristics	What characteristics were reported about the interviewer/facilitator? e.g. Bias, assumptions, reasons and interests in the research topic	Participants were informed that they would be interviewed by a nurse who was conducting research in partnership with the university of liverpool, with the aim of developing a set of outcome measures for children with CZS.

Domain 2: study design			
Theoretical framework			
9	Methodological orientation and Theory	What methodological orientation was stated to underpin the study? e.g. grounded theory, discourse analysis, ethnography, phenomenology, content analysis	The methodological orientation used for the study was the current model of the World Health Organization (WHO), the International Classification of Functioning (ICF). The data were analyzed considering the linking rules methodology proposed by Cieza et al. (2019).
Participant selection			
10	Sampling	How were participants selected? e.g. purposive,	Participants were purposively selected.

		convenience, consecutive, snowball	
11	Method of approach	How were participants approached? e.g. face-to-face, telephone, mail, email	Participants were nvited via phone call or in person at the rehabilitation clinics. Participants were interviewed face to face, through focus groups.
12	Sample size	How many participants were in the study?	Six focus groups were conducted with 32 caregivers of children with severe neurodevelopmental disorders associated with CZS.
13	Non-participation	How many people refused to participate or dropped out? Reasons?	No one refused to participate or was absent during the interview.
Setting			
14	Setting of data collection	Where was the data collected? e.g. home, clinic, workplace	Data were collected at the rehabilitation clinic linked to each participant.
15	Presence of non-participants	Was anyone else present besides the participants and researchers?	In the focus groups helded in Campina Grande the psychologist accompanied the interviews.
16	Description of sample	What are the important characteristics of the sample? e.g. demographic data, date	Information on children are in the Figure 4 and the characteristics of the families are in the Figure 5.
Data collection			
17	Interview guide	Were questions, prompts, guides provided by the authors? Was it pilot tested?	A questionnaire adapted from a previous ICF-based qualitative study was used to conduct the focus group, covering the different ICF domains.
18	Repeat interviews	Were repeat interviews carried out? If yes, how many?	In the sixth focus group, the content started to repeat itself, when the saturation criterion was established.
19	Audio/visual recording	Did the research use audio or visual recording to collect the data?	All groups were recorded and later transcribed in full.
20	Field notes	Were field notes made during and/or after the interview or focus group?	Yes, the researcher used a field diary.
21	Duration	What was the duration of the interviews or focus group?	The duration of the focus groups ranged from 60 to 90 minutes
22	Data saturation	Was data saturation discussed?	We continued to do focus groups until saturation was reached where the testimonies became repetitive, predictable, and were not providing any additional information. Saturation was achieved by the sixth group.
23	Transcripts returned	Were transcripts returned to participants for comment and/or correction?	No

Domain 3: analysis and findingsz			
Data analysis			
24	Number of data coders	How many data coders coded the data?	The transcribed content was analyzed by three researchers experienced in qualitative research (TC, EL, VS).
25	Description of the coding tree	Did authors provide a description of the coding tree?	They split the content into significant units with using NVIVO software, version 11.0 (QSR International, 2019), and associated them with each ICF domain and category using the WHO's linking rules (Cieza et al, 2019) as follows: TC and EL, trained in ICF, performed a blind reading of the content and established the ICF domain and category for each significant unit of the interview to which it was judged to be relevant. VS, a specialist in ICF and with extensive experience in childhood disability, analyzed the disagreements of the two researchers, establishing consensus in relation to the domain/category of the ICF regarding the significant units.
26	Derivation of themes	Were themes identified in advance or derived from the data?	The themes were identified from the study data using the ICF model.
27	Software	What software, if applicable, was used to manage the data?	NVIVO software, version 11.0 (QSR International, 2019).
28	Participant checking	Did participants provide feedback on the findings?	No.
Reporting			
29	Quotations presented	Were participant quotations presented to illustrate the themes / findings? Was each quotation identified? e.g. participant number	The participants' quotes were presented to illustrate the themes and identified with the initials of each participant.
30	Data and findings consistent	Was there consistency between the data presented and the findings?	Yes.
31	Clarity of major themes	Were major themes clearly presented in the findings?	Yes.
32	Clarity of minor themes	Is there a description of diverse cases or discussion of minor themes?	Yes.

How Congenital Zika Virus impacted my child’s functioning and disability: a Brazilian qualitative study guided by the ICF

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How Congenital Zika Virus impacted my child's functioning and disability: a Brazilian qualitative study guided by the ICF

Taynah Neri Correia Campos, Verónica Schiariti, Melissa Gladstone, Adriana Melo, Jousilene Sales Tavares, Adriana Gomes Magalhães & Egmar Longo

Correspondence to:

Egmar Longo, PhD
Graduate Program in Collective Health – Federal University of Rio Grande do Norte – UFRN-FACISA
Rua Vila Trairi S/N, Centro, Santa Cruz, RN
59200-000, Brazil, +5584 32912411
egmarlongo@yahoo.es

Taynah Neri Correia Campos, MsC
Graduate Program in Collective Health
Federal University of Rio Grande do Norte – UFRN-FACISA
Rua Vila Trairi S/N, Centro, Santa Cruz, RN
59200-000, Brazil

Verónica Schiariti, PhD
Adjunct Associate Professor – Division of Medical Sciences
University of Victoria
Victoria, BC, Canada

Melissa Gladstone, PhD
Professor of International Child Health and Neurodevelopmental Paediatrics
Department of Women and Children's Health
Institute of Translational Medicine
University of Liverpool
Eaton Road, Liverpool, L12 2AP, UK

Adriana Melo, PhD
Instituto de Pesquisa Professor Joaquim Amorim Neto
Campina Grande – Paraiba Brazil

Jousilene Sales Tavares, PhD
Instituto de Pesquisa Professor Joaquim Amorim Neto
Campina Grande – Paraiba Brazil

Adriana Gomes Magalhães, PhD
Graduate Program in Rehabilitation Sciences – Federal University of Rio Grande do Norte – UFRN-FACISA
Rua Vila Trairi S/N, Centro, Santa Cruz, RN
59200-000, Brazil

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Abstract

Introduction: The Zika Virus outbreak in Brazil had devasting social, medical and financial consequences. Many researchers and clinicians are following up the children to understand the impact of the Zika on child development, functioning and disability, but outcome measured are heterogenous, and it is not clear how meaningful they are to families and children. This study aimed to identify the parents' perspectives on relevant areas of functioning and disability that should be included as outcome measures for children with congenital Zika Syndrome (CZS), guided by the International Classification of Functioning, Disability and Health (ICF).

Methods: This qualitative study included parents or caregivers of children aged 0 to 5 years with confirmed CZS from two states in northeastern Brazil. Interviews were conducted using focus groups. Content mapping followed the WHO's ICF linking rules. Three raters analyzed the content using NVIVO 11.

Results: Thirty-two caregivers participated in six focus groups, 88% were mothers, average age 30 years. Most of their children were male (59%), all level V according to the Gross Motor Function Classification System (GMFCS). Overall, 825 themes were mapped to 36 ICF categories. Functioning and disability themes were predominantly linked to environmental factors, activities and participation, and body functions. Although parents mentioned areas across all ICF domains, they reported that areas of mobility, eating and recreation were very relevant for them. In addition, environmental factors were highly identified as barriers, specifically services, policies and access to assistive devices. The most predominant facilitators were immediate family support, kind relationships with therapists and extended family.

Conclusions: Although parents emphasized issues related to mobility, their greatest concerns involved environmental factors, such as access and quality of health and social services, systems and policies. These results reinforce the importance of including the parents' perspective when selecting or developing outcome measures for CZS.

Strengths and limitations of this study

- This is the first ICF-based qualitative study describing caregivers' perceptions on relevant areas of function in children with CZS in Brazil.
- Several ICF categories were identified by parents as very relevant which contribute to the creation of a set of main outcomes for rehabilitation interventions.
- A limitation is the misrepresentation of fathers' perspectives, as the majority of participants were mothers.

INTRODUCTION

The Zika virus (ZIKV) was identified in Brazil at the beginning of 2015. Concurrently, rates of microcephaly and other congenital brain abnormalities increased, thus suggesting a causal relationship between this congenital infection and the clinical presentation.[1] Since then, clinical reports have documented the teratogenic effect of ZIKV in pregnant women, particularly with infection in the first trimester.[2, 3, 4] The most prominent among the congenital anomalies is microcephaly, but other manifestations such as; spasticity, seizures, eating difficulties, irritability, ocular abnormalities, hearing loss, calcifications, cortical disorders and ventriculomegaly in neuroimaging have been described, suggesting a new syndrome, called the Congenital Zika Syndrome (CZS).[5, 6, 7] There is still no complete definition of the scope of this new health condition, and adequate outlines are still needed. As a result, surveillance systems are aware of a wider spectrum of congenital malformations which may be associated with ZIKV infection.[8, 9]

A precise estimate of the ZIKV infection incidence is difficult to determine due to the variations between countries regarding the commitment to notifications and the small number of individuals who manifest the health condition caused by ZIKV.[10] There are currently 3,406 confirmed cases of children with microcephaly and/or other neurological signs due to CZS in Brazil, with a further 2,596 cases under investigation.[11]

Given the severity of the syndrome, the scientific community has mobilized efforts to understand the mechanisms of this health condition and to establish outcome measures, to define what should be measured and reported in all trials in this area. As such, creating a standardized core outcome set (COS) for CZS will reduce the heterogeneity of the studies and the result bias, and further enable development of meta-analyzes and data sharing between the studies.[12]

The Core Outcomes Measures in Effectiveness Trials (COMET) initiative identifies and disseminates the most relevant outcomes for each clinical health situation through a database and recommends (among other aspects) the involvement of families in developing these outcomes to ensure that the research process is more relevant and appropriate to the patient's needs.[13, 14] As part of the COS developing process, parents are encouraged to describe their children's strengths and weaknesses following the International Classification of Functioning, Disability and Health (ICF) biopsychosocial model.[14] The ICF can be used as a guiding framework providing a common language for describing the dynamic interaction between the person functional abilities and the role of the environment.[15]

Currently, there is no COS for children with CZS. To fill this gap, we are developing the "Zika COS and congenital infections: a study to develop a COS for children with CZS and other congenital infections" project. This project is coordinated by the University of Liverpool in partnership with the University of Victoria (Canada) and Federal University of Rio Grande do Norte - UFRN-FACISA (Brazil). This paper describes a qualitative study that is part of the 1st phase of the COS for CZS project.[16] The specific objective of this qualitative study is to identify the parents' perspectives on relevant areas of functioning and disability which should be included as outcome measures for children with CZS, guided by the ICF.

METHODS

This study used a qualitative methodology with an exploratory approach through focus groups. We chose to use a qualitative approach as it enables investigation and in-depth understanding of the perspectives and experiences of parents/caregivers of children with CZS. Focus groups, in particular, provide the potential to explore and clarify points of view and reveal dimensions of understanding which would be less easily accessible in other data collection formats.[17, 18]

This research was approved by the Ethics and Research Committee of the Faculty of Health Sciences of Trairi/Federal University of Rio Grande do Norte (2.357.552), it was also re-approved at Liverpool University Ethics Committee (#2083) and developed in three Brazilian rehabilitation centers for CZS in the states of Rio Grande do Norte (Physical Therapy School Clinic at UFRN-FACISA, in Santa Cruz) and Paraíba (Center for the Care of Children with Microcephaly at the Professor Joaquim Amorim Neto Research Institute - IPESQ, in Campina Grande, and Physical Therapy School Clinic at UNIPE, in Joao Pessoa). Parents/guardians signed a Consent Form after being provided with an information sheet which was read out to them. Parents also signed an authorization for Voice Recording and use of images (considering Resolution 466/2012 of the National Health Council which provides for regulatory guidelines and standards for human research).

Participants with important common characteristics were purposively selected by members of the research team, through active search by phone call or direct personal approach. The inclusion criteria included: 1) being parents/caregivers of children with confirmed diagnosis of CZS by polymerase chain reaction (PCR) or presumed diagnosis based on obstetric ultrasound, transfontanellar ultrasound, computed tomography (CT) and magnetic resonance imaging (MRI), 2) who were aged zero to five years , and 3) who lived in the area covered by the study (states of Rio Grande do Norte and Paraíba) and attended the respective rehabilitation services.

The habilitation centers are public services linked to research institutions and higher education in the region. This study did not include parents whose children had congenital syndromes due to other etiologies.

All parents responded to a sociodemographic questionnaire, applied by the researchers before the realization of the focus groups, at rehabilitation services, with information about their relationship with the child, marital status, age, income, government benefit, education, housing and self-reported health, in addition to information from children about gender, age, mobility, rehabilitation, frequency day care centers or schools, and difficulties with vision, hearing, sleep and epilepsy. This questionnaire was applied to the first group and observed whether the questions were well understood by the respondents. Two research assistants were involved in the data collection. In each center, the physiotherapist who accompanied the children classified their motor abilities using the GMFCS. This is an age-specific scheme designed for children with cerebral palsy (CP) based on five levels of gross motor function, ranging from level I (most able) to level V (least able)[19] and provided some clear information on the severity of the children whose parents took part in the study. The study flowchart can be viewed in Figure 1. The consolidated criteria for notification of qualitative studies (COREQ) guidelines were considered.

The focus groups were carried out between September/2018 and January/2019, by research assistants (TC, EL), trained both in qualitative studies and in conducting focus groups. We continued to do focus groups until saturation was reached - where the testimonies became repetitive, predictable, and were not providing any additional information. Saturation was achieved by the sixth group. Furthermore, we decided not to exceed six parents/caregivers per group in order to enable effective participation of the participants and appropriate discussion of themes. Of the 36 parents identified and invited to the study, 32 agreed to participate.

A questionnaire adapted from a previous ICF-based qualitative study was used to conduct the focus group, covering the different ICF domains (Supplementary material 1).[20] Visual tags were produced with images from the ICF illustrated gallery (Supplementary material 2)[21] to assist in understanding the concepts, as studies indicate that visualization has the potential to support data production during the qualitative interview process.[22] The duration of the focus groups ranged from 60 to 90 minutes. The participants were given a therapeutic toy developed by research team at the end of the groups, called “fun fishing”, in order to thank the parents for participating in the study. They were encouraged by the professional team to use it as one therapeutic toy for stimulation of their children at home and promote fun. All groups were recorded and later transcribed in full. Names and identification characteristics were excluded from the transcripts. The transcribed content was analyzed by three researchers experienced in qualitative research (TC, EL, VS). They split the content into significant units with using NVIVO software, version 11.0 (QSR International, 2019), and associated them with each ICF domain and category using the WHO’s linking rules[23] as follows: TC and EL, trained in ICF, performed a blind reading of the content and established the ICF domain and category for each significant unit of the interview to which it was judged to be relevant. VS, a specialist in ICF and with extensive experience in childhood disability, analyzed the disagreements of the two researchers, establishing consensus in relation to the domain/category of the ICF regarding the significant units.

Next, the content was analyzed by the researchers who interpreted the data through an inductive and deductive analysis process based on a permanent dialogue throughout the process to ensure consistency and reliability of the interpretations. Any verbatim which did not exceed “five mentions” was discarded as it was felt not to be representative enough of the data. The frequency of each ICF category in the statements of parents / caregivers was used as a parameter to determine the order of importance of ICF domains/categories. After this process, the content was translated from Brazilian Portuguese into English by a bilingual member of the research team and the content checked by two others, to ensure that there was no loss of meaning.

Aspects of credibility, transferability, reliability and confirmability were considered during all stages in order to guarantee the quality of the qualitative research.[24] Credibility was ensured by thoroughly conducting six focus groups until response saturation was reached, with subsequent blind analysis, review and triangulation, and then a consensus was reached after meetings and debates regarding the interpretation of results. The researchers have general experience and robust training in the area of childhood disabilities and in the use of the ICF for neuro disabilities. Transferability occurred through observation of the rigor to which the research group stopped to describe how the data was obtained through the focus groups, the selection and description of the sample, which in this case involved the participants. Reliability was guaranteed by detailing the data collection processes, presenting the multiple steps followed by the researchers, and showing how the pair analysis and data interpretation took place. A measure of reliability through using the kappa statistic was also used to verify the agreement between the judges, with results indicating almost perfect agreement (0.916) considering the 95% confidence intervals. Finally, verification was achieved through a peer discussion of the data at each stage of the analysis by the research team.

Patient and public involvement: Families were not involved in the design, recruitment or conduct of the study. However, the results will be presented to families, professionals and managers at each participating center, with a view to discussing strategies to meet the needs of children with CZS and their families.

RESULTS

Six focus groups were conducted with 32 caregivers of children with severe neurodevelopmental disorders associated with CZS. Information related to children can be found in Figure 2 and the characteristics of families are in Figure 3. The average age of the children was 32.5 (SD = 6.2) months, with a predominance of males and all of them had severe motor impairment, according to the GMFCS. Although none of the children walked, only 37.5% of them had a wheelchair.

Thirty-six categories of the ICF were identified, as demonstrated in Figure 4 (personal factors were expressed only minimally). This figure represents the parents' perspectives on relevant areas of functioning and disability of their children.

The parents mentioned relevant areas that covered all ICF domains, but a greater diversity of categories was observed in the domains of activities and participation and body functions. However, the environmental factors domain led the ranking of 12 categories, as can be seen in Table 1.

CATEGORY		**QTY**	**DOMAIN**
1º	e580 Health services, systems and policies	59	Environmental Factors
2º	b760 Control of voluntary movement functions	37	Body Functions
3º	e115 Products and technology for personal use in daily living	33	Environmental Factors
4º	d415 Maintaining a body position	33	Activities and Participation
5º	e110 Products or substances for personal consumption	31	Environmental Factors
6º	b230 Hearing functions	31	Body Functions
7º	b134 Sleep functions	31	Body Functions
8º	d450 Walking	28	Activities and Participation
9º	e120 Products and technology for personal indoor and outdoor mobility and transportation	25	Environmental Factors
10º	b210 Seeing functions	24	Body Functions
11º	d445 Hand and arm use	23	Activities and Participation
12º	s750 Structure of lower extremity	22	Body Structures

Table 1: Table demonstrating the ranking of the 12 most frequent categories of the ICF mentioned by parents

Table 2 shows the distribution of categories related to the environmental factors domain in detail.

Environmental Factors		
e580 Health services, systems and policies	59	33,0%
e115 Products and technology for personal use in daily living	33	18,4%
e110 Products or substances for personal consumption	31	17,3%
e120 Products and technology for personal indoor and outdoor mobility and transportation	25	14,0%
e310 Immediate Family	16	8,9%
e540 Transportation services, systems and policies	9	5,0%
e410 Individual attitudes of immediate family members	6	3,4%
TOTAL	179	100,0%

Table 2: Table of areas of the ICF considered important for parents within the area of Environmental factors

Among the categories, **Health services, systems and policies** (e580) was the barrier most expressed by parents and received 59 mentions. This category includes the prevention and treatment of health problems, the provision of rehabilitation services and the promotion of a healthy lifestyle. In the following verbatim, it is possible to observe the mother's desire to have access to a rehabilitation service in his own city.

> *"Also that in Alagoas state has been the same possibilities that here in Campina, to don´t need to move from one place to other, it´s so complicated spend 3 months here, searching treatment that there it should have. I don´t know they can´t leave this treatment there, to every states have, for us don´t need to move a lot to do a treatment. For them could at least sit, maintain their trunk, catch and walk."* Mother 20.

Another barrier reported by parents/caregivers was the lack of information, professionals specialized in the management of CZS and the difficulty in transport to travel to rehabilitation services in other larger cities. Examples of this are evidenced in the statements below.

> *"I guess that it would help a lot if the access is facilitated, on your city, if I have physiotherapy on my city, sure I would go more often."* Mother 03.

> *"I would like science produces medicines for this disease, because is a new disease. Other thing, my great difficulty is absent of answers, you go to see a doctor with a child, and doctor is stalling, he never says the reality for you."* Mother 24.

> *"If our city would have the treatment, moving is tiring. And if when we arrive would be taken care. We suffer some experiences, in some days I spent many hours."* Mother 04.

Some parents pointed to assistive products and technologies as facilitators. They described how adapted or specially designed equipment improved the functioning of their children and contributed to greater participation;

> *"For E* which facilitates activities it´s on first the glasses, because I take her glasses and the vision been worse, put the glasses and she animates."* Mother 06.

The support of the nuclear family was expressed by participants of all focus groups always as a facilitator of the environment, as described below.

> *"What does facilitate? I have a lot of support from my husband and my sister. I live close to my sister, my parents don´t live in the same city than I, but they always are present, always, until the beginning they are present. My family helps me a lot, and this helps me, when I want to go out and can´t take she together, I leave her with my sister. Then, the family support it´s the main factor that facilitates, because alone, it´s hard."* Mother 14.

Table 3 presents the categories referring to the activities and participation domain.

Activities and Participation		
d415 Maintaining a body position	33	14,5%
d450 Walking	28	12,3%
d445 Hand and arm use	23	10,1%
d550 Eating	21	9,3%
d920 Recreation and leisure	19	8,4%
d430 Lifting and carrying objects	15	6,6%
d110 Watching	15	6,6%
d330 Speaking	12	5,3%
d760 Family relationships	11	4,8%
d160 Focusing attention	11	4,8%
d410 Changing basic body position	9	4,0%
d455 Moving around	9	4,0%
d710 Basic interpersonal interactions	8	3,5%
d730 Relating with strangers	7	3,1%
d440 Fine hand use	6	2,6%
TOTAL	227	100,0%

Table 3: Table demonstrating areas of the ICF within Activity and participation found as important to parents

As shown in Table 3, the most expressive categories were those related to movement, such as **Maintaining a body position - d415**, **Walking - d450**, and **Hand and arm use - d445**. Such aspects were pointed out by the parents as being responsible for limiting activity and restricting participation in daily activities, as evidenced in the statements below.

> *"M* has difficulty to maintain the body position, when I put him stand position supported on the wall he can stay a long time. Sit, he also doesn´t sit alone, but if you put him, he stays, he creeps in, rolls. He doesn´t go to lying down position to sitting, but to sitting to lying he can."* Mother 15.
>
> *"Eat, she holds but doesn´t know how put it in her mouth."* Mother 01.
>
> *"His major difficulties are to catch things, he doesn´t catch, we need to put in his hand, than he holds, if he needs to pick up some think, he doesn´t do, holds if I open his hand he catchs, because is difficult for him to open his hand"* Mother 08.

> *"I guess it´s too hard these children who doesn´t walk, doesn´t sit yet, if when arrive in some place and it has a wheelchair, if have how to sit him, but he doesn´t sit. My great problem is his weight, it´s my concern."* Mother 17.

Finally, the areas of Body Function and Structure identified most by parents is shown in Table 4.

Body Functions			
b760 Control of voluntary movement functions		37	16,70%
b230 Hearing functions		31	14,00%
b210 Seeing functions		24	10,80%
b510 Ingestion functions		19	8,60%
b134 Sleep functions		31	14,00%
b710 Mobility of joint functions		18	8,10%
b280 Sensation of pain		16	7,20%
b735 Muscle tone functions		15	6,80%
b320 Articulation functions		10	4,50%
b152 Emotional functions		8	3,60%
b440 Respiration functions		7	3,20%
b770 Gait pattern functions		6	2,70%
	TOTAL	222	100%
Body Structures			
s750 Structure of lower extremity		22	78,60%
s730 Structure of upper extremity		6	21,40%
	TOTAL	28	100%

Table 4: Table demonstrating the categories of d Body Functions and Structures (within the ICF) identified as important by parents

Regarding the body function domain, there was a predominance of the category related to the **Control of voluntary movement - b760**. Many parents expressed concerns about voluntary movements, both simple and more complex, coordination, support functions of the upper and lower limbs and right-left motor coordination, as shown below in this mother's speech.

> *"Movements control, L* has a lot of difficulties, she can´t lift her arm, her hand. About mobility function, she also has difficulties, she can´t lift her arm and hand, she doesn´t have a lot problems with her legs, but can´t move."* Mother 24.

As shown below, parents also pointed out problems related to **Hearing functions - b230**, **Seeing functions - b210** and **Sleep functions - b134**.

> *"The listen function doesn´t work well , only in one side , in the other it doesn´t work , we would make a raffle to earn money and buy a device , because on exam showed that the left one he listens, but on the right he doesn´t anything."* Mother 22.
>
> *"About vision function, he doesn´t see everything, his vision is low, he has difficulties."* Father 01.
>
> *"He has sleep difficulties, but when he falls on sleep (begins to sleep) he sleeps the whole night."* Mother 28.

For "body structure", the category relating to **Structure of lower extremity - s750** most commonly emerged within the parent focus groups. Many parents described issues with foot positioning and how this prevented walking and standing.

> *"The knees, because she could craw and it something that I guess she won´t do. And her feet, to can walk, that will be a great difficulty, because she has a dislocation, we don´t know if she will walk or if she will need a wheelchair for the rest of her life."* Mother 27.
>
> *"About S* is his little foot, when I will put him on stand up position, his foot turn to the side."* Mother 10.

DISCUSSION

This is a pioneering research which revealed the perceptions of parents and caregivers regarding the aspects considered relevant in children with CZS based on the ICF model. Parents/caregivers highlighted the importance of environmental factors and motor function for the follow-up of children with CZS and it deserves be considerate in constructing core outcome set of measures for this health condition. The use of the ICF as a guide for this study enabled us to provide a holistic model which enables the care perspective to be extended beyond problems with body functions and structures in children with CZS.

Despite the number of new cases becoming more stabilized, the complex clinical presentation of CZS and its effect on children's developmental trajectories requires standardized and individualized care during their growth.[25] It also requires researchers to consider a holistic approach and the complex needs of children with neurodevelopmental disabilities.[26]

Several ICF categories were identified by parents as very relevant for them and their lives and these need to be considered within a global context for the creation of COS for CZS for implementing rehabilitation interventions.[27] The role of environmental factors in the statements of parents reflects the real need to include the context as an aspect to be assessed in the follow-up of children with CZS.[28, 29] Barriers such as those relating to the lack of specialized professionals need to be addressed.[30] Parents expressed a great need for information about the condition of their children.[31] The Centers for Disease Control and Prevention (CDC) has produced a pocket guide summarizing pediatric guidelines on initial assessment and outpatient treatment for CZS,[32] but a carefully planned public health approach, improving the ability to anticipate needs, providing adequate care and ensuring that children reach their full potential is still lacking.[33, 34] This is particularly crucial to really address the environmental issues which hinder many parents and families' every day functioning.

Many parents/caregivers in our study expressed a desire to receive all treatments and support in a single place or city. Moving from place to place for services and different types of rehabilitation in different places/cities is a real barrier for families. Parents in our study signaled a definite need for reorganizing health services in order to provide more of a "one stop" approach to supporting them. This same aspect has been highlighted in other studies.[35] The lack of access to services, mainly due to distance, cost and lack of availability has been a big need expressed by families of children with SCZ as well as families with children with CP.[36] From our study, it is clear that services for support and rehabilitation in a single location minimize problems such as low attendance and/or avoidance. Lack of attendance of course interferes hugely with creating good outcomes through focused, context-based and evidence-based rehabilitation.[37]

Parents who took part in our study voiced particular concerns around aspects of their children's movement. This permeated the domains of activities and participation, functions and body structure and was, of course, expressed as a limiting factor in their participation in daily and leisure activities. It was clear that parents were disappointed and were desperate for a change in mobility of their children and had had little counselling about adaptations to support children in their daily lives through other means. The focus still seems to be on the perspective of "fixing" the disability through therapies.[38]

Alongside this, parents voiced their desires relating to gait acquisition (walking) in the context of their comparison of their children with typical children. This is well documented in previous research with children with CP.[39] We know that many parents who have children who will never walk often still resist early introduction of mobility aids such as adapted toy cars despite the fact that studies involving children with CP with severe motor impairment demonstrate positive results using these adaptations in terms of autonomy, self-esteem, social skills and participation.[40] At the same time, a recent study has shown that promoting participation can result in improved body function and structure in children and young people with physical disabilities.[41]

Our findings also showed that parents concentrate on the child's impairments and parents have high expectations for healing or a cure. Unfortunately, in Brazil, there is little acceptance of disabled children for what they are within their context. This negative view of disability is common for parents with children with similar conditions,[42] with parents tending to focus on their children's performing daily activities, and environmental barriers at home, school and in the community. This shows that there is a need for actions including education at a local and national level to change attitudes towards disability, for example using accessible and open access educational tools such as MY ABILITIES FIRST.[43]

Our research has demonstrated what areas of the ICF parents of children with congenital infections such as CZS consider important. This is extremely important when attempting to establish a COS for CZS. We know that it is essential to also consider the child's own perspective as to what he/she considers to be relevant in outcome measures, however, children with CZS in this case are not yet old enough to express their opinions.

We also acknowledge that parents' perceptions of the needs of their children with CZS will change throughout their lives, and adaptations and further studies will be needed. Professionals will need to remain vigilant as scientific knowledge about the impact of CZS will evolve in the coming years.[44] However, due to the similarity with CP, it is known that in the early years it is important to stimulate the child's development and support caregivers, and over time to support inclusion, independent living, maintaining health and function.[36]

The results obtained in this study must be interpreted taking into account some limitations. The sample was extracted from three Brazilian cities, from public or not-for profit rehabilitation centers, so our findings may not represent the perspectives of all parents of children with CZS and should not be generalized. Our study generated the views of mothers predominantly with very few fathers taking part. They may have a very different perspective.

CONCLUSION

This is a unique study which aims to understand the views and perception of parents with regard to the needs of their children with CZS guided by the ICF. Although the parents did concentrate on issues related to movement abilities of their children, overall their emphasis of concerns was centred around environmental factors. These factors included issues such as; the context of services, systems and policies for prevention and treatment of health problems through rehabilitation as well as factors supporting a healthy lifestyle to promote the physical and psychological well-being and social status of their children.

Our results reinforce the importance of including the parents' perspective in the development of a COS. Parents are the ones who live and experience the main limitations and potential of their children, they highlight the urgent need for environmental changes to improve the lives of children with CZS and their families in Brazil. In the future, when children with CZS are able to express their own opinions, we should also hear their views as to what they consider relevant for outcome measures.

Figure 1 – Study flowchart

Figure 2 – Sociodemographic data of children

Figure 3 – Sociodemographic data for parent/caregivers

Figure 4: ICF categories representing the parents' perspective

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Contributors: VS, MG and EL designed the study. TNCC recruited the participants and conducted the interviews. VS, MG, EL, TNCC reviewed and refined the data. TNCC wrote the article. VS, MG, EL, AM, JST and AGM revised the article. TNCC received qualitative research training through participation in a qualitative health research methods course at the Federal University of Rio Grande do Norte/Brazil.

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Patient consent for publication: Not required

Data availability statement: Data are available upon request to the corresponding author. As data contain information which may lead to the identification of study participants, funding would be required to support anonymisation of the data

Ethics approval: This research was approved by the Ethics and Research Committee of the Faculty of Health Sciences of Trairi/Federal University of Rio Grande do Norte (75053417.1.0000.5568), it was also re-approved at Liverpool University Ethics Committee (#2083)

Patient and public involvement: Families were not involved in the design, recruitment or conduct of the study. However, the results will be presented to families, professionals and managers at each participating center, with a view to discussing strategies to meet the needs of children with CZS and their families.

Figure 1: Study flowchart

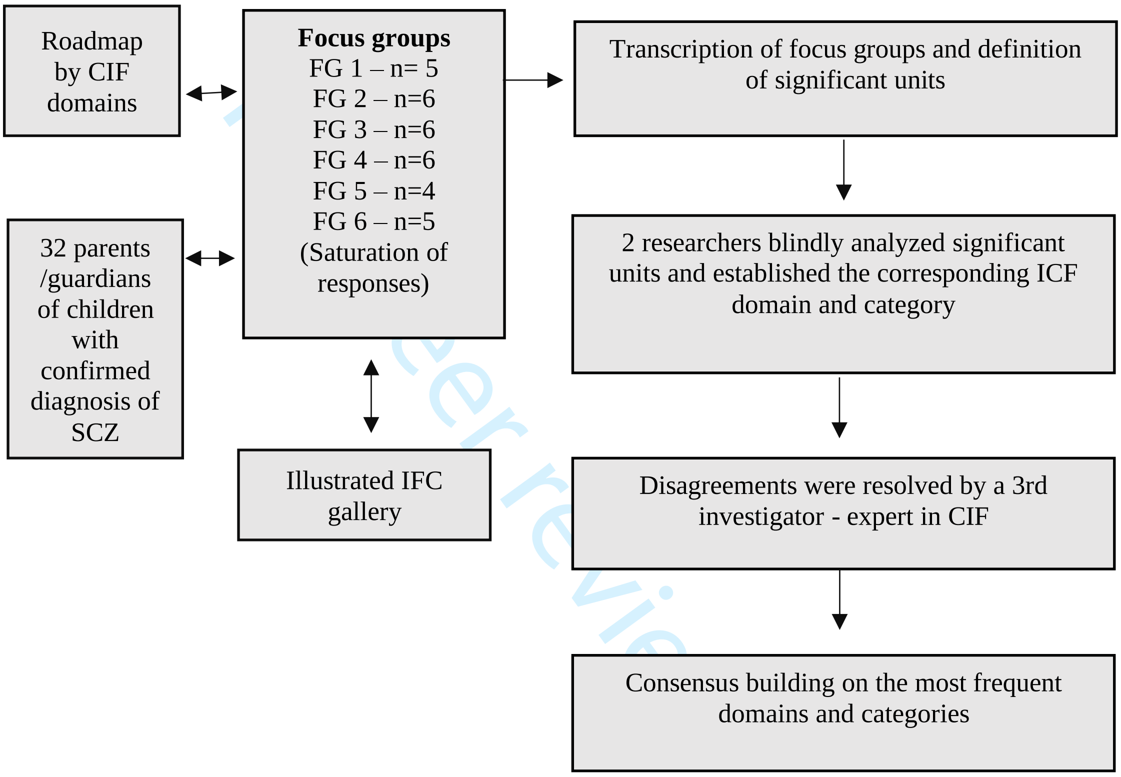


Figure 2 – Sociodemographic data of children

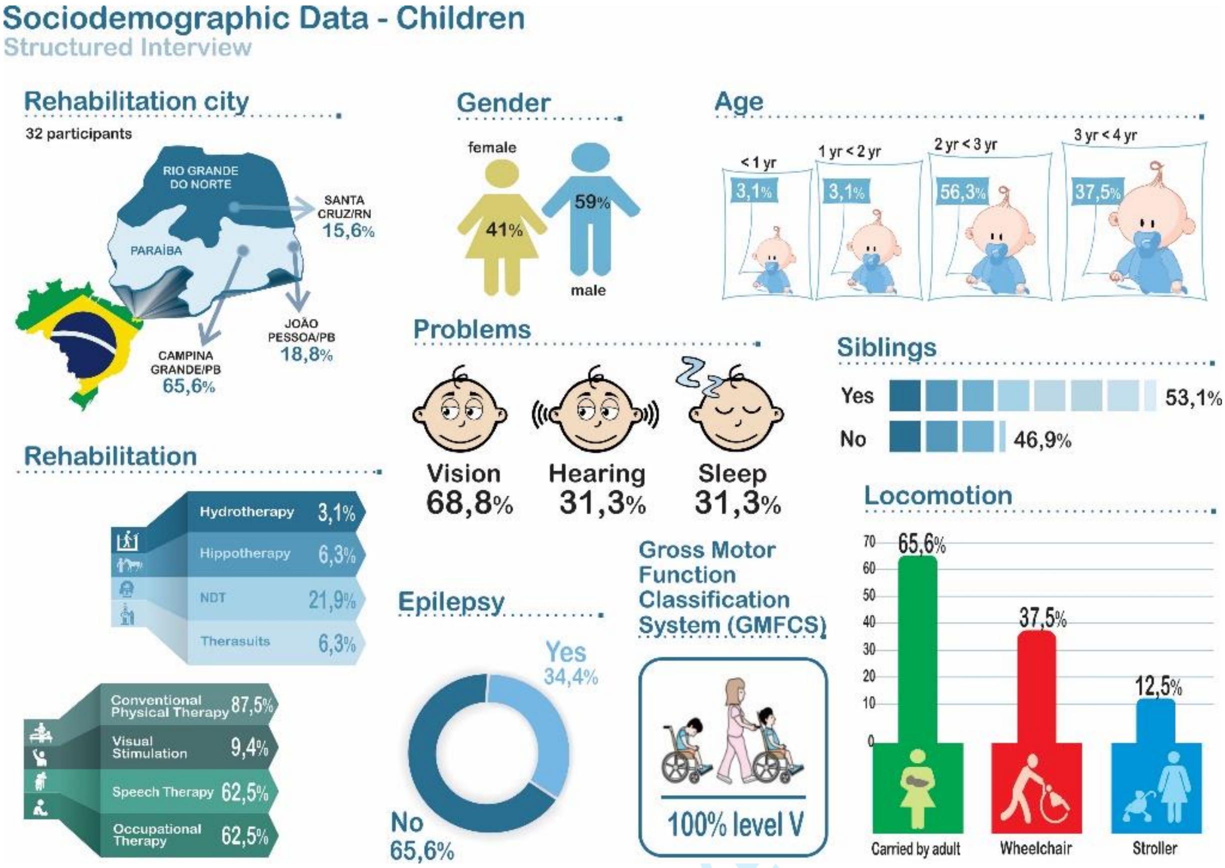


Figure 2 – Sociodemographic data of children

Figure 3 – Sociodemographic data for parent/caregivers

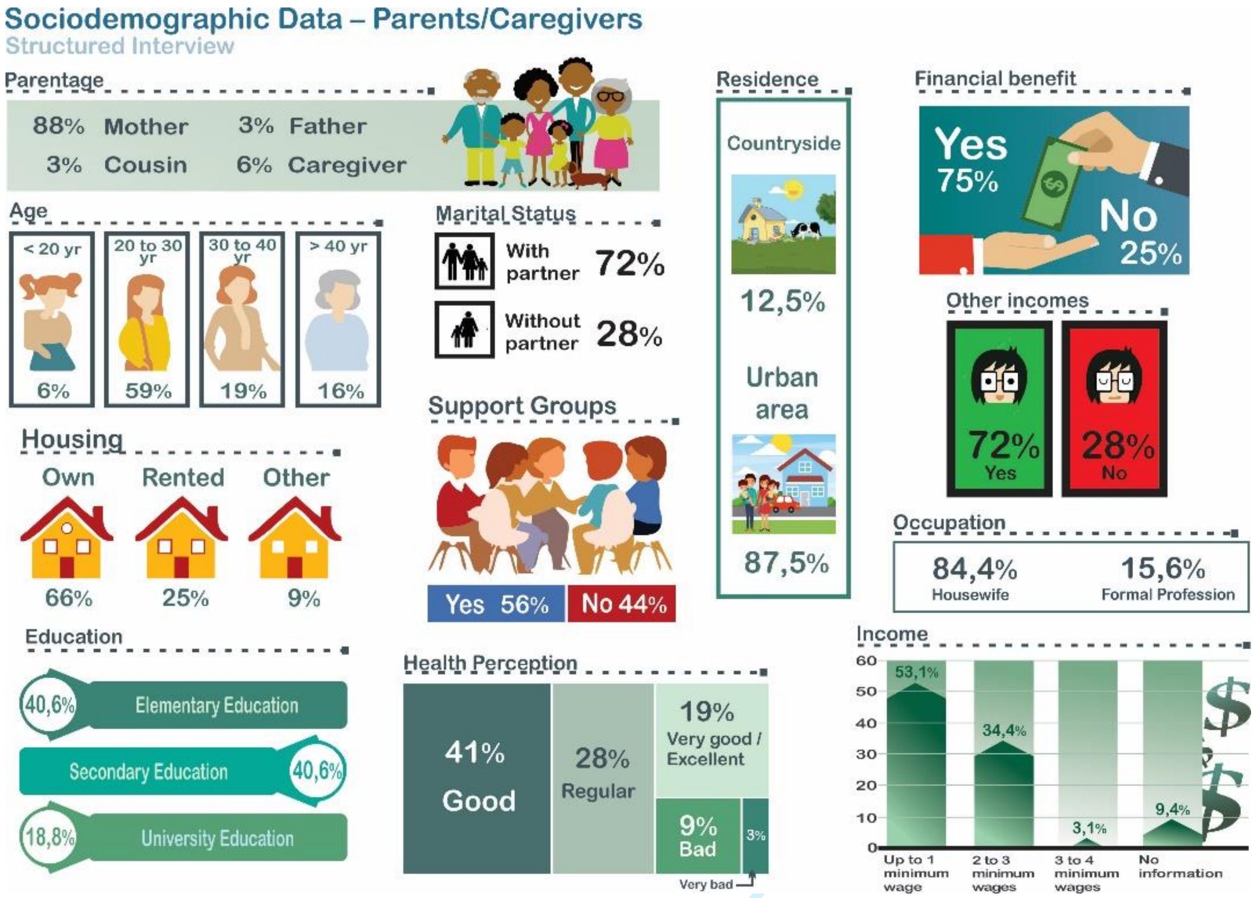


Figure 3 – Sociodemographic data for parent/caregivers

Figure 4: ICF categories representating the parents’ perspective

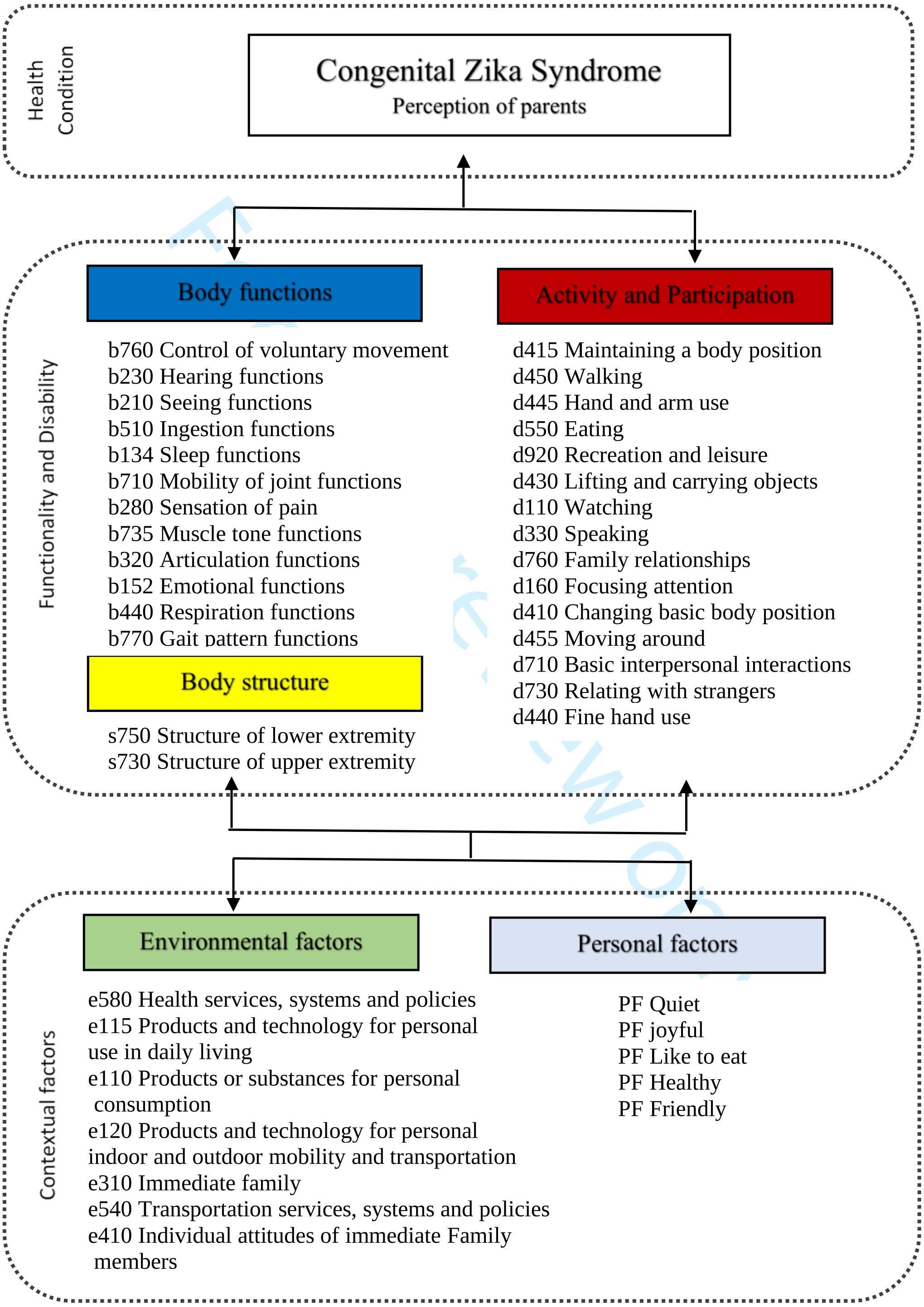


Figure 4: ICF categories representating the parents’ perspective

Supplementary material 1

ZIKA FOCUS GROUPS

Aims of focus groups:

To elicit **what outcomes** are considered important to parents and caregivers of children with Zika virus and therefore what should be included in an outcome measure.

Population:

Parents and/or Caregivers of children who have had infants born with congenital infections or similar conditions who have children from 0-10 years and who are willing to take part in a focus group. We will attempt to sample parents of children with a range of severity of disorders and will attempt to sample from both urban and rural settings.

Framework:

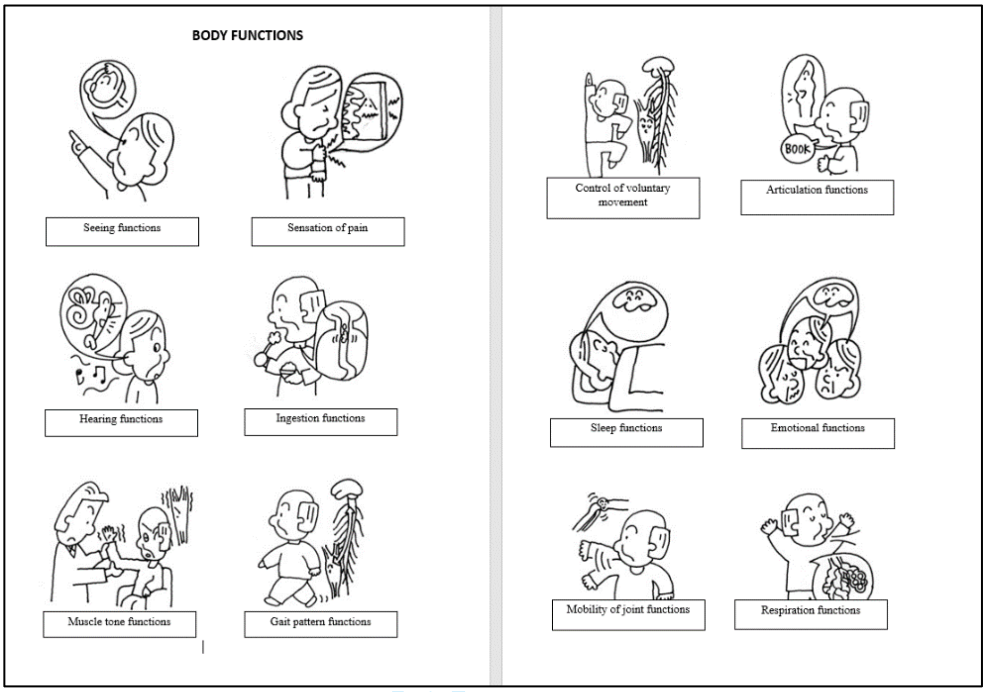
The ICF and ICF Core Sets for children and youth with CP will be used as an overarching framework to ensure a comprehensive representation of outcomes.

Questionnaire:

1. If you think about your child as an individual, what **personal characteristics** are important about him/her? (Moderator provide an example) **PERSONAL FACTORS**
2. If you think about the **daily activities**, (Moderator provide an example, show pictures of activities)
    a. What activities your child is able to do?
    b. What activities does your child find hard/difficult to do?
3. If you think about the **physical and social environment** (**family, school, and neighborhood)** of your child, (Moderator provide an example, show pictures, ask HOME, SCHOOL, COMMUNITY)
    a. What do you find **helpful or supportive** in facilitating participation in different activities?
    b. What **challenges or barriers** does your child experience?
4. If you think about the **body** of your child, which body parts cause your child some difficulties, if any? (Moderator provide an example, may be a picture of the body and they can mark areas with an X)
5. If you think about the **body** of your child, what parts of his/her body do not work the way it supposes to, if any? (Moderator provide an example, may be a list from the ICF Core Sets then they can mark functions from the list) **BODY FUNCTIONS**
6. Tell me about the **biggest problems** for you at the moment.
7. If people were looking into treatments for your child, what are the things you might **hope to improve:**
    a. In terms of their **daily activities?**
    b. In terms of making it **easier for your child to participate** in daily activities (in the neighbourhood, at home with the family and at school or nursery, if they attend)?

Supplementary material 2

Visual cards with images from the ICF illustrated gallery



Consolidated criteria for reporting qualitative studies (COREQ): 32-item checklist

Domain 1: Research team and reflexivity			
Personal Characteristics			
1	Interviewer/facilitator	Which author/s conducted the interview or focus group?	The first and the last authors conducted the focus groups. **Pag 6**
2	Credentials	What were the researcher's credentials? E.g. PhD, MD	Msc Candidate - nurse; PhD- Physical Therapist. **Pag 1**
3	Occupation	What was their occupation at the time of the study?	Msc candidate - nurse; Professor.
4	Gender	Was the researcher male or female?	Female.
5	Experience and training	What experience or training did the researcher have?	She was training by the last author, who has a lot of experience conducting focus group in Brazil and Spain. She was following previous qualitative studies as a trainee before start her research.
Relationship with participant			
6	Relationship established	Was a relationship established prior to study commencement?	No. **Pag 22**
7	Participant knowledge of the interviewer	What did the participants know about the researcher? e.g. personal goals, reasons for doing the research	Participants were informed that they would be interviewed by a nurse who was conducting research in partnership with the university of Liverpool, with the aim of developing a set of outcome measures for children with CZS. **Pag 5**
8	Interviewer characteristics	What characteristics were reported about the interviewer/facilitator? e.g. Bias, assumptions, reasons and interests in the research topic	Participants were informed that they would be interviewed by a nurse who was conducting research in partnership with the university of Liverpool, with the aim of developing a set of outcome measures for children with CZS. **Pag 5**

Domain 2: study design			
Theoretical framework			
9	Methodological orientation and Theory	What methodological orientation was stated to underpin the study? e.g. grounded theory, discourse analysis, ethnography, phenomenology, content analysis	The methodological orientation used for the study was the current model of the World Health Organization (WHO), the International Classification of Functioning (ICF). The data were analyzed considering the linking rules methodology proposed by Cieza et al. (2019). **Pag. 07**
Participant selection			
10	Sampling	How were participants selected? e.g. purposive,	Participants were purposively selected. **Pag. 05**

		convenience, consecutive, snowball	
11	Method of approach	How were participants approached? e.g. face-to-face, telephone, mail, email	Participants were nvited via phone call or in person at the rehabilitation clinics. Participants were interviewed face to face, through focus groups. **Pag. 05**
12	Sample size	How many participants were in the study?	Six focus groups were conducted with 32 caregivers of children with severe neurodevelopmental disorders associated with CZS. **Pag. 06**
13	Non-participation	How many people refused to participate or dropped out? Reasons?	Of the 36 parents identified and invited to the study, 32 agreed to participate. **Pag. 06**
Setting			
14	Setting of data collection	Where was the data collected? e.g. home, clinic, workplace	Data were collected at the rehabilitation clinic linked to each participant. **Pag. 06**
15	Presence of non-participants	Was anyone else present besides the participants and researchers?	In the focus groups helded in Campina Grande the psychologist accompanied the interviews.
16	Description of sample	What are the important characteristics of the sample? e.g. demographic data, date	Information on children are in the Figure 2 and the characteristics of the families are in the Figure 3. **Pag. 09**
Data collection			
17	Interview guide	Were questions, prompts, guides provided by the authors? Was it pilot tested?	A questionnaire adapted from a previous ICF-based qualitative study was used to conduct the focus group, covering the different ICF domains. **Pag. 06**
18	Repeat interviews	Were repeat interviews carried out? If yes, how many?	In the sixth focus group, the content started to repeat itself, when the saturation criterion was established. **Pag. 06**
19	Audio/visual recording	Did the research use audio or visual recording to collect the data?	All groups were recorded and later transcribed in full. **Pag. 07**
20	Field notes	Were field notes made during and/or after the interview or focus group?	Yes, the researcher used a field diary.
21	Duration	What was the duration of the interviews or focus group?	The duration of the focus groups ranged from 60 to 90 minutes **Pag. 07**
22	Data saturation	Was data saturation discussed?	We continued to do focus groups until saturation was reached where the testimonies became repetitive, predictable, and were not providing any additional information. Saturation was achieved by the sixth group. **Pag. 06**
23	Transcripts returned	Were transcripts returned to participants for comment and/or correction?	No

Domain 3: analysis and findingsz			
Data analysis			
24	Number of data coders	How many data coders coded the data?	The transcribed content was analyzed by three researchers experienced in qualitative research (TC, EL, VS). **Pag. 07**
25	Description of the coding tree	Did authors provide a description of the coding tree?	They split the content into significant units with using NVIVO software, version 11.0 (QSR International, 2019), and associated them with each ICF domain and category using the WHO's linking rules (Cieza et al, 2019) as follows: TC and EL, trained in ICF, performed a blind reading of the content and established the ICF domain and category for each significant unit of the interview to which it was judged to be relevant. VS, a specialist in ICF and with extensive experience in childhood disability, analyzed the disagreements of the two researchers, establishing consensus in relation to the domain/category of the ICF regarding the significant units. **Pag. 07**
26	Derivation of themes	Were themes identified in advance or derived from the data?	The themes were identified from the study data using the ICF model. **Pag. 07**
27	Software	What software, if applicable, was used to manage the data?	NVIVO software, version 11.0 (QSR International, 2019). **Pag. 07**
28	Participant checking	Did participants provide feedback on the findings?	No.
Reporting			
29	Quotations presented	Were participant quotations presented to illustrate the themes / findings? Was each quotation identified? e.g. participant number	The participants' quotes were presented to illustrate the themes without identification. **Pag 10-14**
30	Data and findings consistent	Was there consistency between the data presented and the findings?	Yes. **Pag 9-14**
31	Clarity of major themes	Were major themes clearly presented in the findings?	Yes. **Pag 9-14**
32	Clarity of minor themes	Is there a description of diverse cases or discussion of minor themes?	Yes. **Pag 9-14**

How Congenital Zika Virus impacted my child's functioning and disability: a Brazilian qualitative study guided by the ICF

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How Congenital Zika Virus impacted my child's functioning and disability: a Brazilian qualitative study guided by the ICF

Taynah Neri Correia Campos, Verónica Schiariti, Melissa Gladstone, Adriana Melo, Jousilene Sales Tavares, Adriana Gomes Magalhães & Egmar Longo

Correspondence to:

Egmar Longo, PhD
Graduate Program in Collective Health – Federal University of Rio Grande do Norte – UFRN-FACISA
Rua Vila Trairi S/N, Centro, Santa Cruz, RN
59200-000, Brazil, +5584 32912411
egmarlongo@yahoo.es

Taynah Neri Correia Campos, MsC
Graduate Program in Collective Health
Federal University of Rio Grande do Norte – UFRN-FACISA
Rua Vila Trairi S/N, Centro, Santa Cruz, RN
59200-000, Brazil

Verónica Schiariti, PhD
Adjunct Associate Professor – Division of Medical Sciences
University of Victoria
Victoria, BC, Canada

Melissa Gladstone, PhD
Professor of International Child Health and Neurodevelopmental Paediatrics
Department of Women and Children's Health
Institute of Translational Medicine
University of Liverpool
Eaton Road, Liverpool, L12 2AP, UK

Adriana Melo, PhD
Instituto de Pesquisa Professor Joaquim Amorim Neto
Campina Grande – Paraiba Brazil

Jousilene Sales Tavares, PhD
Instituto de Pesquisa Professor Joaquim Amorim Neto
Campina Grande – Paraiba Brazil

Adriana Gomes Magalhães, PhD
Graduate Program in Rehabilitation Sciences – Federal University of Rio Grande do Norte – UFRN-FACISA
Rua Vila Trairi S/N, Centro, Santa Cruz, RN
59200-000, Brazil

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Abstract

Introduction: The Zika Virus outbreak in Brazil has had devasting social, medical and financial consequences for families. Both researchers and clinicians are measuring longer term outcomes to understand the impact of the Zika on child development, functioning and disability. Outcomes and tools used to measure them are very varied and we are unclear how meaningful they are to families and children. This study aimed to identify the parents' perspectives on relevant areas of functioning and disability that should be included as outcome measures for children with congenital Zika Syndrome (CZS), as guided by the International Classification of Functioning, Disability and Health (ICF).

Methods: This qualitative study included parents or caregivers of children aged 0 to 5 years with confirmed CZS from two states in northeastern Brazil. Interviews were conducted using focus groups. Content mapping followed the WHO's ICF linking rules. Three raters analyzed the content using NVIVO 11.

Results: Thirty-two caregivers participated in six focus groups, 88% were mothers with an average age of 30 years. Most children were male (59%) and all were level V (severe) to on the Gross Motor Function Classification System (GMFCS). Overall, 825 themes were mapped to 36 ICF categories. Although parents mentioned areas across all ICF domains, they reported that areas of mobility, eating and recreation were most relevant for them. In addition, environmental factors were highly identified as barriers, specifically services, policies and access to assistive devices. The most predominant facilitators within the environment were; immediate family support, kind relationships with therapists and support from the extended family.

Conclusions: Although parents emphasized issues related to mobility, their greatest concerns involved environmental factors, such as access and quality of health and social services, systems and policies. These results reinforce the importance of including parents' perspectives when selecting or developing outcome measures for CZS.

Strengths and limitations of this study

- This is the first ICF-based qualitative study describing caregivers' perceptions on relevant areas of function in children with CZS in Brazil.
- Few studies have previously undertaken research to understand parent's views on the issue of what outcome measures should be considered in neurodevelopmental disorders.
- We had limited representation of men within our sample as the main caregiver was predominantly women.
- Our study was limited in terms of numbers of participants and region in Brazil, limiting the generalizability of our study.

INTRODUCTION

The Zika virus (ZIKV) was identified in Brazil at the beginning of 2015. During this time, rates of microcephaly and other congenital brain abnormalities increased, suggesting a causal relationship between the two.[1] Since then, clinical reports have documented the teratogenic effect of ZIKV in pregnant women, particularly when infected in the first trimester.[2,3,4] The most prominent anomalies for children with congenital ZIKV is microcephaly, but other manifestations such as; spasticity, seizures, eating difficulties, irritability, ocular abnormalities, hearing loss, calcifications, cortical disorders and ventriculomegaly have been described.[5, 6, 7] There is still no clear definition of exactly what is included in Congenital Zika syndrome. As a result, surveillance studies are underway to identify the wide~~r~~ spectrum of congenital malformations which may be associated with ZIKV infection.[8,9]

A precise estimate of ZIKV infection incidence is difficult to determine due to variations in case ascertainment between countries and only a small number of children identified with manifestations of the more severe congenital ZIKV syndrome.[10] There are currently 3,406 confirmed cases of children with microcephaly and/or other neurological signs due to CZS in Brazil, with a further 2,596 cases under investigation.[11]

Given the severity of the syndrome, the scientific community has mobilized efforts to understand the mechanisms of this health condition and to measure long term health and neurodevelopmental outcomes of children with this condition. Presently, a large variety of heterogenous tools and outcomes are being utilized, often which causes confusion and lack of ability to conduct meta-analyses and syntheses of data. Furthermore, most tools chosen are those chosen by epidemiologists and researchers who may have little understanding of the what is most relevant to parents and families with children with this condition. Primarily, tools to measure child development are used; not always that relevant when functioning for these children may be very limited. As such, creating a standardized core outcome set (COS) for CZS will reduce the heterogeneity of the studies and further enable clearer synthesis and data sharing between studies.[12]

The Core Outcomes Measures in Effectiveness Trials (COMET) is an initiative aimed at identifying and creating a core set of outcomes for any clinical health situation. This is often conducted through a process of systematic reviews of outcomes measured, consensus work as well as the involvement of families who support the development of these outcomes in order to ensure that researchers consider outcomes that are most relevant and appropriate to the patient's needs.[13, 14] As part of COS development, a framework is usually utilized to enable outcomes to be classified and considered for finalization in a core outcome set. In the case of neurodisability and neurodevelopmental disorders, a helpful framework can be the International Classification of Functioning, Disability and Health (ICF) which provides a standard language and framework for health states.[14] The ICF includes both functions and structure of the body and activities and participation of the child and can be used as a guiding framework providing a common language for describing the dynamic interaction between the person functional abilities and the role of the environment.[15]

Currently, there is no COS for children with CZS. To fill this gap, we aim to develop a COS for children with Zika and other congenital infections. This project is coordinated by the University of Liverpool in partnership with the University of Victoria (Canada) and Federal University of Rio Grande do Norte - UFRN-FACISA (Brazil). This paper describes a qualitative study that is part of the 1st phase of the COS for CZS project.[16] The specific objective of this qualitative study is to identify the parents' perspectives on relevant areas of functioning and disability which should be included as outcome measures for children with CZS, guided by the ICF.

METHODS

This study used a qualitative methodology with an exploratory approach through focus groups. We chose to use a qualitative approach as it enables investigation and an in-depth understanding of the perspectives and experiences of parents/caregivers of children with CZS. Focus groups, in particular, provide the potential to explore and clarify points of view and reveal dimensions of understanding which would be less easily accessible in other data collection formats.[17, 18]

This research was approved by the Ethics and Research Committee of the Faculty of Health Sciences of Trairi/Federal University of Rio Grande do Norte (2.357.552), it was also re-approved at Liverpool University Ethics Committee (#2083) and developed in three Brazilian rehabilitation centers for CZS in the states of Rio Grande do Norte (Physical Therapy School Clinic at UFRN-FACISA, in Santa Cruz) and Paraíba (Center for the Care of Children with Microcephaly at the Professor Joaquim Amorim Neto Research Institute - IPESQ, in Campina Grande, and Physical Therapy School Clinic at UNIPE, in Joao Pessoa). Parents/guardians signed a Consent Form after being provided with an information sheet which was read out to them. Parents also signed consent for voice recording and use of images.

Participants with important common characteristics were purposively selected by members of the research team, through active search by phone call or direct personal approach. The inclusion criteria included: 1) being parents/caregivers of children with confirmed diagnosis of CZS by polymerase chain reaction (PCR) or presumed diagnosis based on obstetric ultrasound, transfontanellar ultrasound, computed tomography (CT) and magnetic resonance imaging (MRI), 2) child with congenital CZS aged zero to five years , and 3) living in the area covered by the study (states of Rio Grande do Norte and Paraíba) and attending the respective rehabilitation services.

The rehabilitation centers are public services linked to research institutions and higher education in the region. This study did not include parents whose children had congenital syndromes due to other etiologies.

All parents responded to a sociodemographic questionnaire, applied by the researchers prior to the focus groups with information about; their relationship with the child, marital status, age, income, government benefit, education, housing and self-reported health, in addition to information from children about gender, age, mobility, rehabilitation, frequency day care centers or schools, and difficulties with vision, hearing, sleep and epilepsy. This questionnaire was to ensure good understanding prior to rolling it out to all respondents. Two research assistants were involved in the data collection. In each center, the physiotherapist who accompanied the children classified their motor abilities using the GMFCS. This is an age-specific scheme designed for children with cerebral palsy (CP) based on five levels of gross motor function, ranging from level I (most able) to level V (least able)[19] and provided some clear information on the severity of the children whose parents took part in the study. The study flowchart can be viewed in Figure 1. We used the consolidated criteria for notification of qualitative studies (COREQ) guidelines to guide us with our study methodology and write up.

Insert Figure 1 here

The focus groups were carried out between September/2018 and January/2019, by research assistants (TC, EL), trained both in qualitative studies and in conducting focus groups. We continued to do focus groups until saturation was reached – where testimonies became repetitive, predictable, and were not providing any additional information. Saturation was achieved by the sixth group. We did not exceed six parents/caregivers per group in order to enable effective participation of the participants and appropriate discussion of themes. Of the 36 parents identified and invited to the study, 32 agreed to participate.

A questionnaire adapted from a previous ICF-based qualitative study was used to conduct the focus group, covering the different ICF domains (Supplementary material 1).[20] Visual tags were produced with images from the ICF illustrated gallery (Supplementary material 2)[21] to assist in understanding the concepts, as studies indicate that visualization has the potential to support data production during the qualitative interview process.[22] The duration of the focus groups ranged from 60 to 90 minutes. The participants were given a therapeutic toy developed by research team at the end of the groups, called "fun fishing", in order to thank the parents for participating in the study. They were encouraged by the professional team to use it as a therapeutic toy for stimulation of their children at home and to promote fun. All groups were recorded and later transcribed in full. Names of participants and any identifying characteristics were excluded from the transcripts. The transcribed content was analyzed by three researchers experienced in qualitative research (TC, EL, VS). They split the content into significant units with using NVIVO software, version 11.0 (QSR International, 2019), and associated them with each ICF domain and category using the WHO's linking rules[23] as follows: TC and EL, trained in ICF, performed a blind reading of the content and established the ICF domain and category for each significant unit of the interview to which it was judged to be relevant. VS, a specialist in ICF and with extensive experience in childhood disability, analyzed the disagreements of the two researchers, establishing consensus in relation to the domain/category of the ICF regarding the significant units.

Next, the content was analyzed by the researchers who interpreted the data through an inductive and deductive analysis process based on a permanent dialogue throughout the process to ensure consistency and reliability of the interpretations. Any verbatim which did not exceed "five mentions" was discarded as it was felt not to be representative enough of the data. The frequency of each ICF category in the statements of parents/caregivers was used as a parameter to determine the order of importance of ICF domains/categories. After this process, the content was translated from Brazilian Portuguese into English by a bilingual member of the research team and the content checked by two others, to ensure that there was no loss of meaning.

Aspects of credibility, transferability, reliability and confirmability were considered during all stages in order to guarantee the quality of the qualitative research.[24] Credibility was ensured by thoroughly conducting six focus groups until saturation was reached, with subsequent blind analysis, review and triangulation, and then consensus being reached after meetings and debates regarding the interpretation of results. The researchers have robust training in the area of childhood disabilities and in the use of the ICF for neurodisabilities. Transferability occurred through observation of the rigor to which the research group stopped to describe how the data was obtained through the focus groups, the selection and description of the sample, which in this case involved the participants. Reliability was guaranteed by detailing the data collection processes, presenting the multiple steps followed by the researchers, and showing how the pair analysis and data interpretation took place. A measure of reliability through using the kappa statistic was also used to verify the agreement between the judges, with results indicating almost perfect agreement (0.916) considering 95% confidence intervals. Finally, verification was achieved through a peer discussion of the data at each stage of the analysis by the research team.

Patient and public involvement: Families were not involved in the design, recruitment or conduct of the study. However, the results will be presented to families, professionals and managers at each participating center, with a view to discussing strategies to meet the needs of children with CZS and their families.

RESULTS

Six focus groups were conducted with 32 caregivers of children with severe neurodevelopmental disorders associated with CZS. Information related to children can be found in Figure 2 and the characteristics of families are in Figure 3. The average age of the children was 32.5 (SD = 6.2) months, with a predominance of males, all with severe motor impairment on, the GMFCS. Although none of the children walked, only 37.5% of them had a wheelchair.

Insert Figure 2 here

Insert Figure 3 here

Thirty-six categories of the ICF were identified, as demonstrated in Figure 4 (personal factors were expressed only minimally). This figure represents the parents' perspectives on relevant areas of functioning and disability of their children.

Insert Figure 4 here

The parents mentioned relevant areas that covered all ICF domains, but a greater diversity of categories was observed in the domains of activities and participation and body functions. However, the environmental factors domain led the ranking of 12 categories, as can be seen in Table 1.

Insert Table 1 here

	CATEGORY	QTY	DOMAIN
1º	e580 Health services, systems and policies	59	Environmental Factors
2º	b760 Control of voluntary movement functions	37	Body Functions
3º	e115 Products and technology for personal use in daily living	33	Environmental Factors
4º	d415 Maintaining a body position	33	Activities and Participation
5º	e110 Products or substances for personal consumption	31	Environmental Factors
6º	b230 Hearing functions	31	Body Functions
7º	b134 Sleep functions	31	Body Functions
8º	d450 Walking	28	Activities and Participation
9º	e120 Products and technology for personal indoor and outdoor mobility and transportation	25	Environmental Factors
10º	b210 Seeing functions	24	Body Functions
11º	d445 Hand and arm use	23	Activities and Participation
12º	s750 Structure of lower extremity	22	Body Structures

Table 1: Table demonstrating the ranking of the 12 most frequent categories of the ICF mentioned by parents

Table 2 shows the distribution of categories related to the environmental factors domain in detail.

Insert Table 2 here

Environmental Factors		
e580 Health services, systems and policies	59	33,0%
e115 Products and technology for personal use in daily living	33	18,4%
e110 Products or substances for personal consumption	31	17,3%
e120 Products and technology for personal indoor and outdoor mobility and transportation	25	14,0%

e310 Immediate Family	16	8,9%
e540 Transportation services, systems and policies	9	5,0%
e410 Individual attitudes of immediate family members	6	3,4%
TOTAL	179	100,0%

Table 2: Table of areas of the ICF considered important for parents within the area of Environmental factors

Among the categories, **Health services, systems and policies** (e580) was the barrier most expressed by parents, receiving 59 mentions. This category includes the prevention and treatment of health problems, the provision of rehabilitation services and the promotion of a healthy lifestyle. The following verbatim demonstrates a mother's desire to have access to rehabilitation services in her own city.

> *"Also that in Alagoas state has been the same possibilities that here in Campina, to don´t need to move from one place to other, it´s so complicated spend 3 months here, searching treatment that there it should have. I don´t know they can´t leave this treatment there, to every states have, for us don´t need to move a lot to do a treatment. For them could at least sit, maintain their trunk, catch and walk."* Mother 20.

Other barriers reported in this area included; lack of information or professionals specialized in the management of CZS and the difficulty in transport to travel to rehabilitation services in other larger cities. Examples of this are evidenced in the statements below.

> *"I guess that it would help a lot if the access is facilitated, in our city, if I had physiotherapy in my city, sure I would go more often."* Mother 03.

> *"I would like that science produces medicines for this disease, because it is a new disease. Another thing, a great difficulty for me, is the absence of answers…. you go to see a doctor with your child, and the doctor is stalling, he never tells you the reality…."* Mother 24.

> *"If our city would have treatment…. moving is tiring. And if when we come, we were taken care of…. We suffer from these experiences, some days I have spent many hours travelling…."* Mother 04.

Some parents pointed to assistive products and technologies as facilitators. They described how adapted or specially designed equipment improved the functioning of their children and contributed to greater participation;

> *"For E*, to facilitate activities, first I put her glasses on… once I take her glasses off, her vision becomes worse…. if I put the glasses on, she becomes much more animated."* Mother 06.

The support of the nuclear family was expressed by participants of all focus groups as a major facilitator to their environment, as described below.

> *"What helps? I have a lot of support from my husband and my sister. I live close to my sister… my parents don´t live in the same city as me, but they always are present…. Always… from the beginning they have been present. My family helps me a lot, and this helps me, when I want to go out and can´t take her with me… I leave her with my sister. The family support, it´s the main factor that helps me, because alone, it´s hard."* Mother 14.

Table 3 presents the categories referring to the activities and participation domain.

Insert Table 3 here

Activities and Participation		
d415 Maintaining a body position	33	14,5%
d450 Walking	28	12,3%
d445 Hand and arm use	23	10,1%
d550 Eating	21	9,3%
d920 Recreation and leisure	19	8,4%
d430 Lifting and carrying objects	15	6,6%
d110 Watching	15	6,6%
d330 Speaking	12	5,3%
d760 Family relationships	11	4,8%
d160 Focusing attention	11	4,8%

d410 Changing basic body position		9	4,0%
d455 Moving around		9	4,0%
d710 Basic interpersonal interactions		8	3,5%
d730 Relating with strangers		7	3,1%
d440 Fine hand use		6	2,6%
	TOTAL	227	100,0%

Table 3: Table demonstrating areas of the ICF within activity and participation found as important to parents

As shown in Table 3, the most often expressed categories were those related to movement of children, such as **Maintaining a body position - d415**, **Walking - d450**, and **Hand and arm use - d445**. Such aspects were pointed out by the parents as being responsible for limiting activity and restricting participation in daily activities, as evidenced in the statements below.

> *"M* has difficulty maintaining body position, when I put him in standing position supported by the wall, he can stay a long time. Sitting.. he also doesn´t sit alone, but if you put him in sitting.. he stays there…He doesn´t go from lying down position to sitting, but from sitting to lying, he can."* Mother 15.
>
> *"Eating… she holds it but doesn´t know how put it in her mouth."* Mother 01.
>
> *"His major difficulties are to grasp things. He doesn´t grasp and we need to put in his hands and then he holds it. If he needs to pick up something, he can not do it, but he holds and object if I open his hand for him"* Mother 08.
>
> *"I guess it´s so difficult for these children; they don´t walk and don´t sit yet. If, when I arrive in a place and it has a wheelchair I try and sit him in it, he still can not sit in it."* Mother 17.

Finally, the areas of Body Function and Structure identified most by parents is shown in Table 4.

Insert Table 4 here

Body Functions			
b760 Control of voluntary movement functions		37	16,70%
b230 Hearing functions		31	14,00%
b210 Seeing functions		24	10,80%
b510 Ingestion functions		19	8,60%
b134 Sleep functions		31	14,00%
b710 Mobility of joint functions		18	8,10%
b280 Sensation of pain		16	7,20%
b735 Muscle tone functions		15	6,80%
b320 Articulation functions		10	4,50%
b152 Emotional functions		8	3,60%
b440 Respiration functions		7	3,20%
b770 Gait pattern functions		6	2,70%
	TOTAL	222	100%
Body Structures			
s750 Structure of lower extremity		22	78,60%
s730 Structure of upper extremity		6	21,40%
	TOTAL	28	100%

Table 4: Table demonstrating the categories of d Body Functions and Structures (within the ICF) identified as important by parents

Control of voluntary movement - b760 was the most common category mentioned in the domain of functioning. Many parents expressed concerns about their child's voluntary movements (both simple and more complex) as well as their child's difficulties with coordination as well as use of their upper and lower limbs.

> *"Control of movements… L* has a lot of difficulties, she can´t lift her arm or her hand. With regards to her ability to move, she also has difficulties, she can´t lift her arm and hand. She doesn´t have a lot problems with her legs, but she can´t move her body."* Mother 24.

Parents also pointed out problems related to; **Hearing functions - b230**, **Seeing functions - b210** and **Sleep functions - b134**.

> *"Her hearing is only good on one side, on the other side, it doesn´t work. We are doing a raffle to earn money to buy a device, because when she was examined it showed that she can hear with her left ear, but on the right, she doesn´t hear anything."* Mother 22.
>
> *"About his vision, he doesn´t see everything, his vision is low, he has difficulties."* Father 01.
>
> *"He has sleep difficulties, but when he falls on sleep, he sleeps the whole night."* Mother 28.

With regards to "body structure", the category relating to **Structure of lower extremities- s750** most commonly emerged within our parent focus groups. Many parents described issues with foot positioning and how this prevented walking and standing.

> *"The knees, it is something that I guess she won´t do. And her feet, to walk, that will be a great difficulty, because she has a dislocation. We don´t know if she will walk or if she will need a wheelchair for the rest of her life."* Mother 27.
>
> *"About S*'s little foot… when I put him in standing position, his foot turns to the side."* Mother 10.

DISCUSSION

This pioneering research is the first to describe the perceptions of parents and caregivers regarding their understanding of the issues for their children with congenital CZS as placed within the ICF framework. Parents/caregivers highlighted the importance of environmental factors and motor function for their children with CZS. These factors must therefore, be taken into account when constructing a recommended core set of outcomes to be measured in CZS. The use of the ICF as our guide for this study has enabled us to provide a holistic framework for considering perspectives that extend beyond the issues with body functions and structures for children with CZS.

Despite the number of new cases of CZS stabilizing, the complex clinical presentation of CZS and its effect on children's health means that both standardized and individualized care is needed.[25] Researchers need to consider a holistic approach to their measurement of outcomes when following these children. The diversity of needs of these children (and families) with CZS and neurodevelopmental disabilities is wide.[26]

Several ICF categories were identified by parents as very relevant and which must be considered and advocated for within the global context for the creation of a COS for CZS. This is particularly the case for researchers who will be measuring the effect of rehabilitation and support interventions for families.[27] In particular, the importance that parents place on environmental factors for the wellbeing of their children demonstrates the real need to include this environmental context as an aspect to be assessed in the follow-up of children with CZS.[28,29]

Our study has demonstrated that the lack of trained professionals to support families is a major issue for parents that needs to be addressed.[30] Furthermore, parents expressed a great need for information about the condition of their children.[31] The Centers for Disease Control and Prevention (CDC) has produced a pocket guide summarizing pediatric guidelines on initial assessment and outpatient treatment for CZS.[32] Resources such as this could be more often provided for parents, particularly if given in a caring and sensitive manner with space for parents to reflect and ask questions. It is clear that a carefully planned public health approach which targets the needs of parents and families and provides adequate care and support for children and families, is still lacking.[33, 34] Addressing the environmental issues which hinder many parents and families' every day functioning is the most crucial aspect of this, and the one that will make the most difference for families and children quality of life.

Many parents/caregivers in our study expressed a desire to receive treatment and support in a single place or city. Moving from place to place for services and different types of rehabilitation all the time, is a real barrier for families. Parents in our study signalled a definite need for reorganizing health services in a way which would provide more of a "one stop" approach for supporting them. This has been highlighted in other studies.[35] Furthermore, lack of access to services (mainly due to distance, cost and lack of availability) was a massive issue for families of children with CZS.[36] From our study, it is clear that services for support and rehabilitation in a single location would minimize problems of low attendance and/or avoidance of services. Lack of attendance interferes hugely with enabling good outcomes when using focused, context-based and evidence-based rehabilitation.[37]

Parents who took part in our study voiced particular concerns around aspects of their children's movement. This permeated the domains of activities and participation, functions and body structure and was, of course, expressed as a limiting factor in their participation in daily and leisure activities. It was clear that parents were disappointed and desperate for a change in mobility of their children and had had little counselling about adaptations to support children in their daily lives through other means. The focus still seems to be on the perspective of "fixing" the disability through therapies.[38]

Alongside this, parents voiced their desires relating to gait acquisition (walking) in the context of their comparison of their children with typical children. This is well documented in previous research with children with CP.[39] We know that many parents who have children who will never walk often still resist early introduction of mobility aids (such as adapted toy cars or wheelchairs). This is despite the fact that studies involving children with CP with severe motor impairment demonstrate very positive results when using these adaptations in terms of autonomy, self-esteem, social skills and participation.[40] Recent studies have shown that promoting participation in children with physical disabilities can, in itself, result in improved body function and structure.[41]

Our findings also showed that parents concentrate on the child’s impairments and that parents have high expectations for healing or a cure. Unfortunately, in Brazil, there is little acceptance of disabled children for what they are within their context. This negative view of disability is common for parents with children with similar conditions.[42] Parents in Brazil tend to focus on their children’s ability to perform daily activities as well as the environmental barriers at home, school and in the community. This demonstrates a need for action, including education, at a local and national level to change attitudes towards disability. One example which could be advocated is the use of accessible and open access educational tools such as MY ABILITIES FIRST.[43]

Our research has demonstrated what areas of the ICF parents of children with congenital infections such as CZS consider important. This information is vital to have when attempting to establish a COS for CZS. We also know that it is essential to also consider the child’s own perspective as to what he/she considers to be relevant in outcome measures, however, children with CZS have limited communication and in our case, were not yet old enough to express their opinions.

We acknowledge that parents' perceptions of the needs of their children with CZS will change throughout their lives with further studies being necessary to understand this over time. Professionals will need to remain vigilant as scientific knowledge about the impact of CZS will evolve in the coming years.[44] However, due to the similarity with CP, many professionals working with these children in the early years are focusing on ways to provide devices to aid with functioning, stimulate child development, prevent worsening of health conditions and to support caregivers. Over time the focus will shift to supporting inclusion, independent living, and to continuing to maintain health and function.[36]

There were a number of limitations in our study which must be taken into account. The sample was extracted from three Brazilian cities, from public or not-for profit rehabilitation centers. In this way, our findings may not represent the perspectives of all parents of children with CZS and should not be generalized. Our study generated the views of mothers predominantly with very few fathers taking part. Fathers may have a very different perspective and may have provided very different results to our study. Finally, within this study we were unable to take the views of the children themselves into account. This would be important and relevant to consider in the future.

CONCLUSION

This is a unique study which aims to understand the views and perception of parents with regard to the needs of their children with CZS guided by the ICF. Although the parents did concentrate on issues related to movement abilities of their children, overall their emphasis was centred around environmental factors. These factors included services, systems and policies for prevention and treatment of their children as well as factors which would enable a healthy lifestyle to promote the physical and psychological well-being and social status of their children.

Our results reinforce the importance of including the parents' perspective in the development of a COS. Parents are the ones who live and experience the main limitations and potential of their children and in our study, they highlight the urgent need for environmental changes to improve the lives of children with CZS and their families in Brazil. In the future, when children with CZS are able to express their own opinions, we should also hear their views as to what they consider relevant for outcome measures.

Figure 1 – Study flowchart

Figure 2 – Sociodemographic data of children

Figure 3 – Sociodemographic data for parent/caregivers

Figure 4: ICF categories representing the parents' perspective

Contributors: VS, MG and EL designed the study. TNCC recruited the participants and conducted the interviews. VS, MG, EL, TNCC reviewed and refined the data. TNCC wrote the article. VS, MG, EL, AM, JST and AGM revised the article. TNCC received qualitative research training through participation in a qualitative health research methods course at the Federal University of Rio Grande do Norte/Brazil.

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Competing interests: None declared**.**

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Patient consent for publication: Not required

Data availability statement: Data are available upon reasonable request. Data are available upon reasonable request from the corresponding author subject to restrictions to preserve anonymity and personal privacy. These data are not publicly available as they contain information that could compromise research participant privacy/consente.

Ethics approval: This research was approved by the Ethics and Research Committee of the Faculty of Health Sciences of Trairi/Federal University of Rio Grande do Norte (75053417.1.0000.5568), it was also re-approved at Liverpool University Ethics Committee (#2083)

Patient and public involvement: Families were not involved in the design, recruitment or conduct of the study. However, the results will be presented to families, professionals and managers at each participating center, with a view to discussing strategies to meet the needs of children with CZS and their families.

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Figure 1: Study flowchart

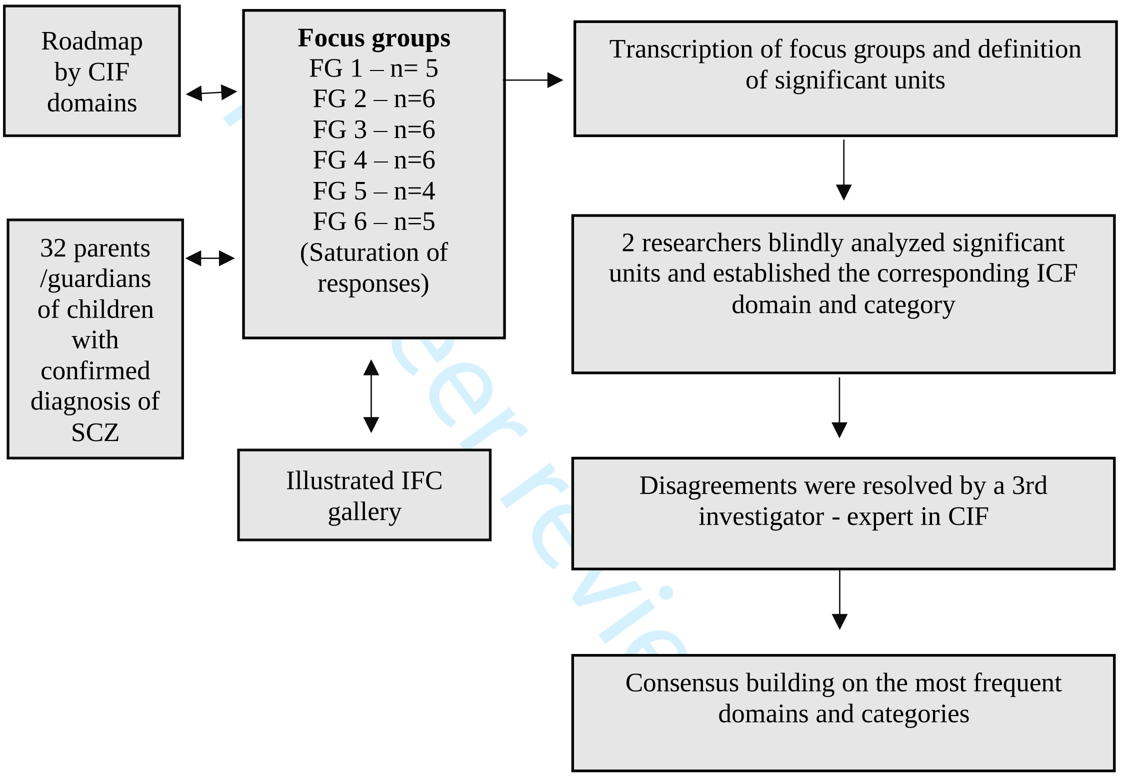


Figure 2 – Sociodemographic data of children

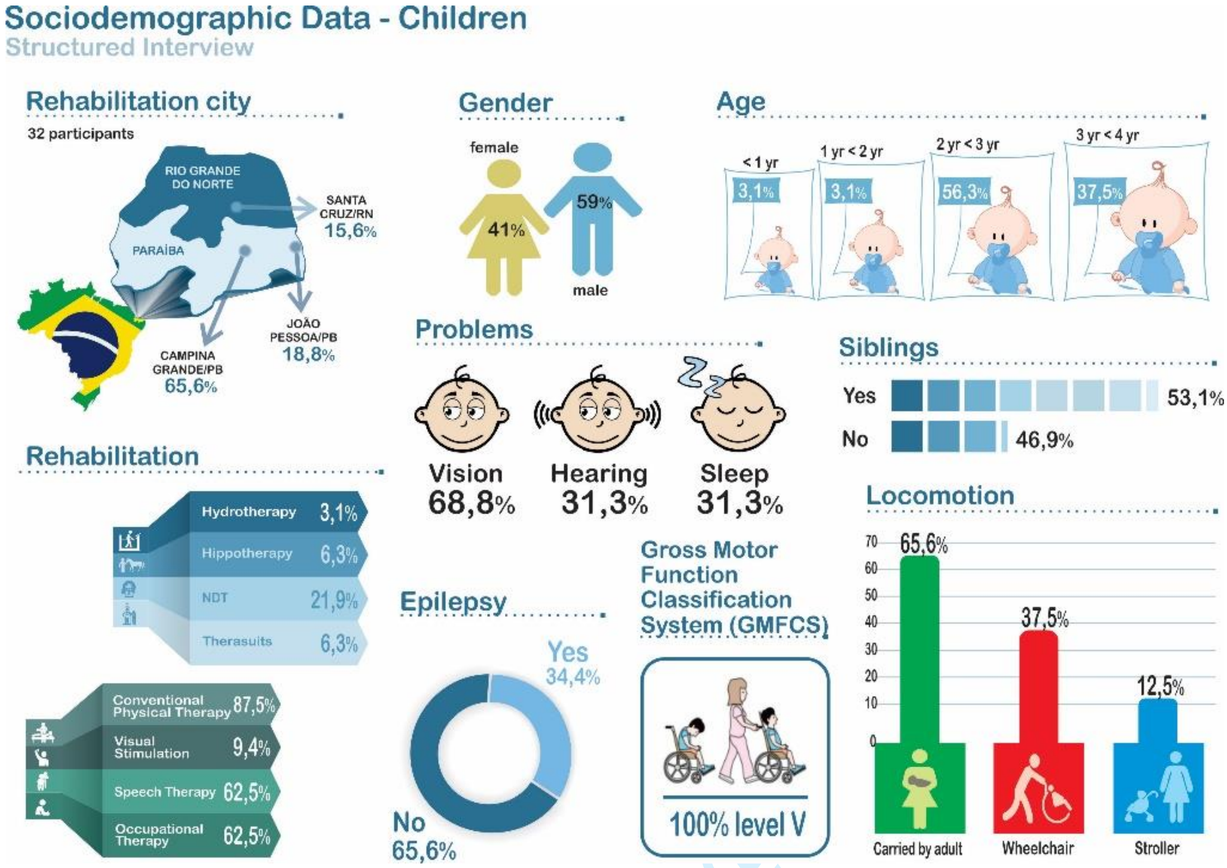


Figure 2 – Sociodemographic data of children

Figure 3 – Sociodemographic data for parent/caregivers

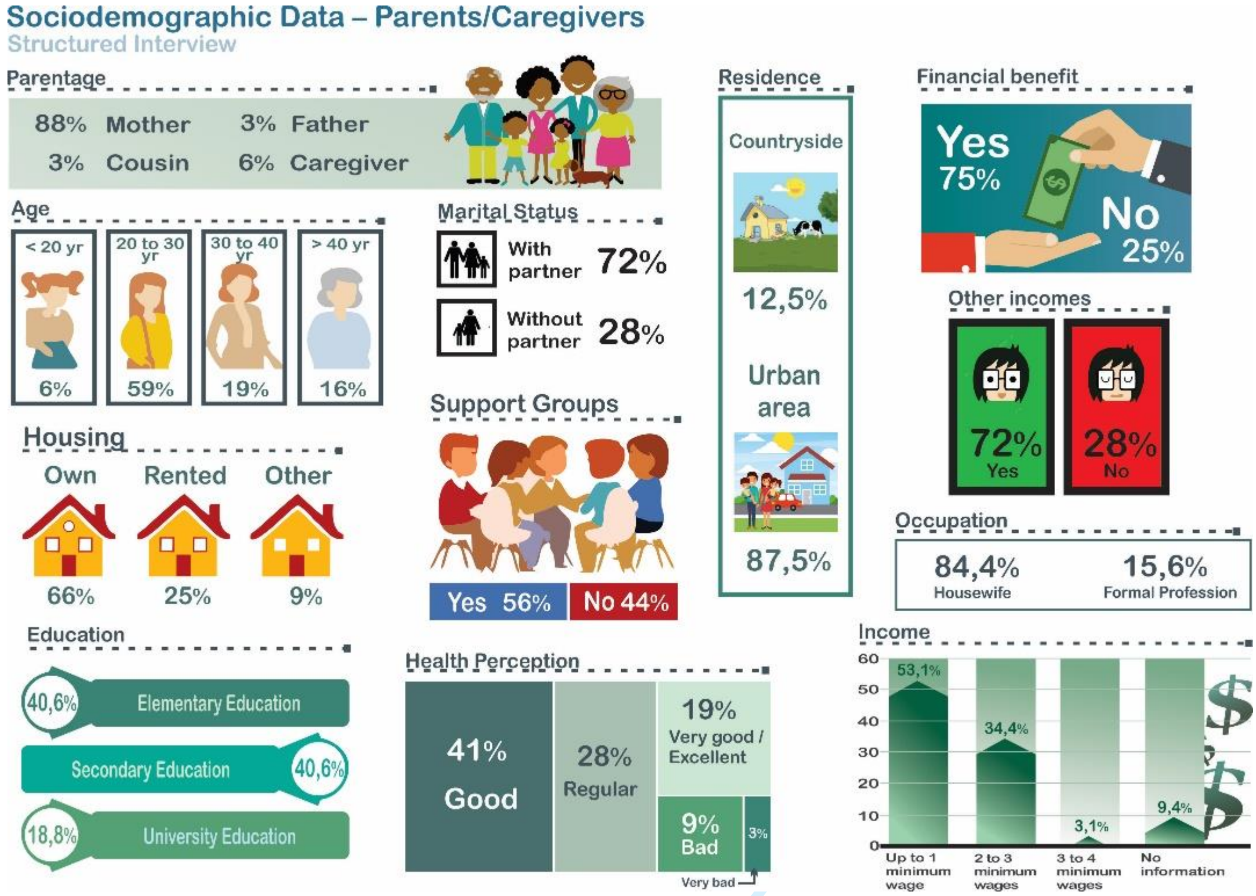


Figure 3 – Sociodemographic data for parent/caregivers

Figure 4: ICF categories representating the parents’ perspective

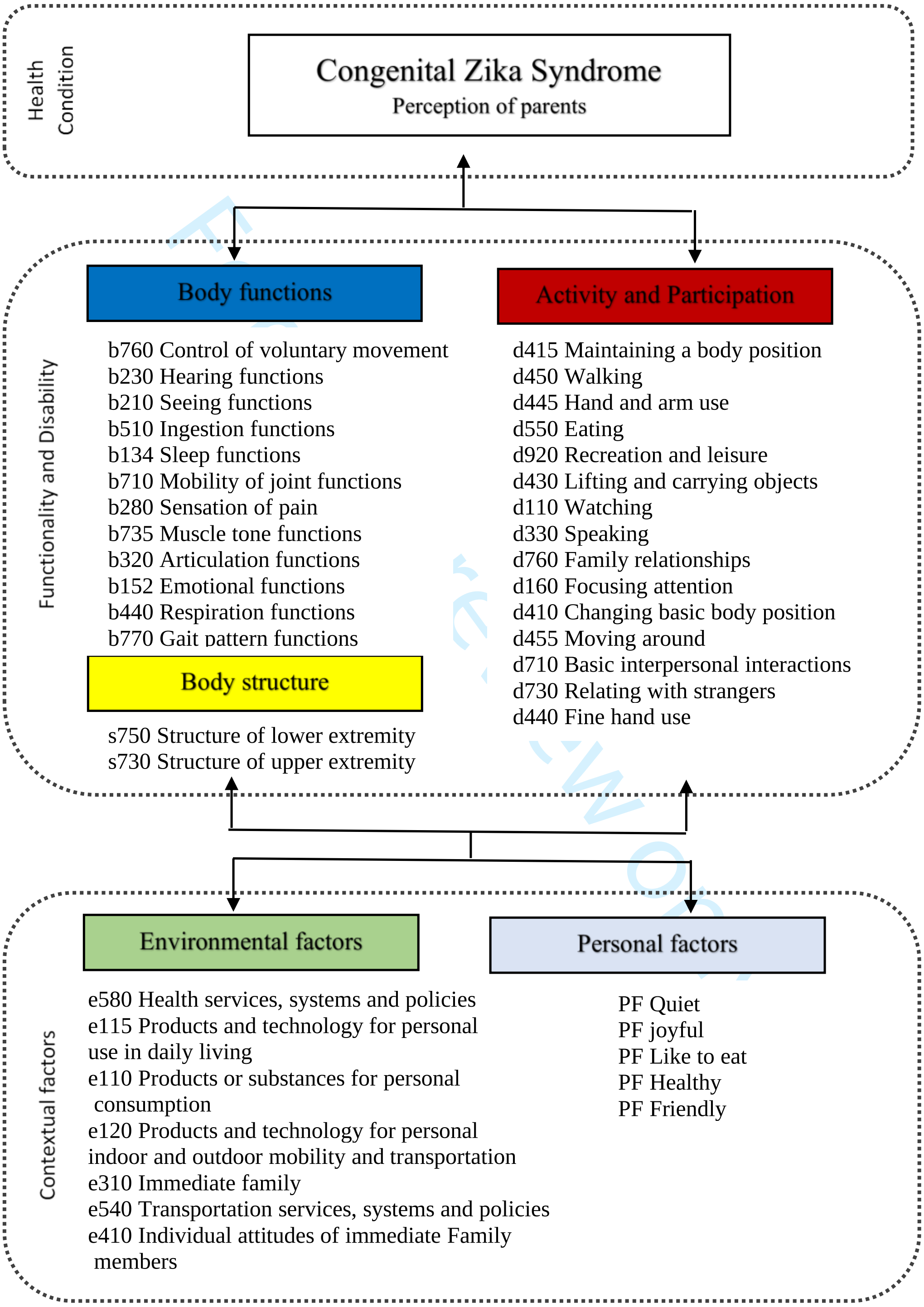


Figure 4: ICF categories representating the parents’ perspective

Supplementary material 1

ZIKA FOCUS GROUPS

Aims of focus groups:

To elicit **what outcomes** are considered important to parents and caregivers of children with Zika virus and therefore what should be included in an outcome measure.

Population:

Parents and/or Caregivers of children who have had infants born with congenital infections or similar conditions who have children from 0-10 years and who are willing to take part in a focus group. We will attempt to sample parents of children with a range of severity of disorders and will attempt to sample from both urban and rural settings.

Framework:

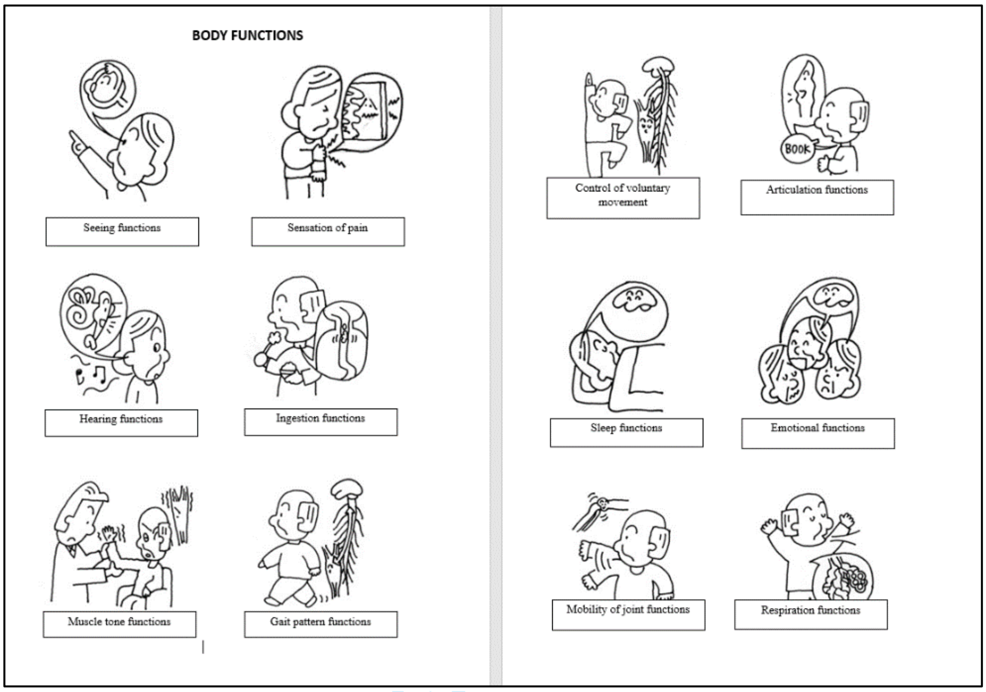
The ICF and ICF Core Sets for children and youth with CP will be used as an overarching framework to ensure a comprehensive representation of outcomes.

Questionnaire:

1. If you think about your child as an individual, what **personal characteristics** are important about him/her? (Moderator provide an example) **PERSONAL FACTORS**
2. If you think about the **daily activities**, (Moderator provide an example, show pictures of activities)
    a. What activities your child is able to do?
    b. What activities does your child find hard/difficult to do?
3. If you think about the **physical and social environment (family, school, and neighborhood)** of your child, (Moderator provide an example, show pictures, ask HOME, SCHOOL, COMMUNITY)
    a. What do you find **helpful or supportive** in facilitating participation in different activities?
    b. What **challenges or barriers** does your child experience?
4. If you think about the **body** of your child, which body parts cause your child some difficulties, if any? (Moderator provide an example, may be a picture of the body and they can mark areas with an X)
5. If you think about the **body** of your child, what parts of his/her body do not work the way it supposes to, if any? (Moderator provide an example, may be a list from the ICF Core Sets then they can mark functions from the list) **BODY FUNCTIONS**
6. Tell me about the **biggest problems** for you at the moment.
7. If people were looking into treatments for your child, what are the things you might **hope to improve:**
    a. In terms of their **daily activities?**
    b. In terms of making it **easier for your child to participate** in daily activities (in the neighbourhood, at home with the family and at school or nursery, if they attend)?

Supplementary material 2

Visual cards with images from the ICF illustrated gallery



Consolidated criteria for reporting qualitative studies (COREQ): 32-item checklist

Domain 1: Research team and reflexivity			
Personal Characteristics			
1	Interviewer/facilitator	Which author/s conducted the interview or focus group?	The first and the last authors conducted the focus groups. **Pag 6**
2	Credentials	What were the researcher's credentials? E.g. PhD, MD	Msc Candidate - nurse; PhD- Physical Therapist. **Pag 1**
3	Occupation	What was their occupation at the time of the study?	Msc candidate - nurse; Professor.
4	Gender	Was the researcher male or female?	Female.
5	Experience and training	What experience or training did the researcher have?	She was training by the last author, who has a lot of experience conducting focus group in Brazil and Spain. She was following previous qualitative studies as a trainee before start her research.
Relationship with participant			
6	Relationship established	Was a relationship established prior to study commencement?	No. **Pag 22**
7	Participant knowledge of the interviewer	What did the participants know about the researcher? e.g. personal goals, reasons for doing the research	Participants were informed that they would be interviewed by a nurse who was conducting research in partnership with the university of Liverpool, with the aim of developing a set of outcome measures for children with CZS. **Pag 5**
8	Interviewer characteristics	What characteristics were reported about the interviewer/facilitator? e.g. Bias, assumptions, reasons and interests in the research topic	Participants were informed that they would be interviewed by a nurse who was conducting research in partnership with the university of Liverpool, with the aim of developing a set of outcome measures for children with CZS. **Pag 5**

Domain 2: study design			
Theoretical framework			
9	Methodological orientation and Theory	What methodological orientation was stated to underpin the study? e.g. grounded theory, discourse analysis, ethnography, phenomenology, content analysis	The methodological orientation used for the study was the current model of the World Health Organization (WHO), the International Classification of Functioning (ICF). The data were analyzed considering the linking rules methodology proposed by Cieza et al. (2019). **Pag. 07**
Participant selection			
10	Sampling	How were participants selected? e.g. purposive,	Participants were purposively selected. **Pag. 05**

		convenience, consecutive, snowball	
11	Method of approach	How were participants approached? e.g. face-to-face, telephone, mail, email	Participants were nvited via phone call or in person at the rehabilitation clinics. Participants were interviewed face to face, through focus groups. **Pag. 05**
12	Sample size	How many participants were in the study?	Six focus groups were conducted with 32 caregivers of children with severe neurodevelopmental disorders associated with CZS. **Pag. 06**
13	Non-participation	How many people refused to participate or dropped out? Reasons?	Of the 36 parents identified and invited to the study, 32 agreed to participate. **Pag. 06**
Setting			
14	Setting of data collection	Where was the data collected? e.g. home, clinic, workplace	Data were collected at the rehabilitation clinic linked to each participant. **Pag. 06**
15	Presence of non-participants	Was anyone else present besides the participants and researchers?	In the focus groups helded in Campina Grande the psychologist accompanied the interviews.
16	Description of sample	What are the important characteristics of the sample? e.g. demographic data, date	Information on children are in the Figure 2 and the characteristics of the families are in the Figure 3. **Pag. 09**
Data collection			
17	Interview guide	Were questions, prompts, guides provided by the authors? Was it pilot tested?	A questionnaire adapted from a previous ICF-based qualitative study was used to conduct the focus group, covering the different ICF domains. **Pag. 06**
18	Repeat interviews	Were repeat interviews carried out? If yes, how many?	In the sixth focus group, the content started to repeat itself, when the saturation criterion was established. **Pag. 06**
19	Audio/visual recording	Did the research use audio or visual recording to collect the data?	All groups were recorded and later transcribed in full. **Pag. 07**
20	Field notes	Were field notes made during and/or after the interview or focus group?	Yes, the researcher used a field diary.
21	Duration	What was the duration of the interviews or focus group?	The duration of the focus groups ranged from 60 to 90 minutes **Pag. 07**
22	Data saturation	Was data saturation discussed?	We continued to do focus groups until saturation was reached where the testimonies became repetitive, predictable, and were not providing any additional information. Saturation was achieved by the sixth group. **Pag. 06**
23	Transcripts returned	Were transcripts returned to participants for comment and/or correction?	No

Domain 3: analysis and findingsz			
Data analysis			
24	Number of data coders	How many data coders coded the data?	The transcribed content was analyzed by three researchers experienced in qualitative research (TC, EL, VS). **Pag. 07**
25	Description of the coding tree	Did authors provide a description of the coding tree?	They split the content into significant units with using NVIVO software, version 11.0 (QSR International, 2019), and associated them with each ICF domain and category using the WHO's linking rules (Cieza et al, 2019) as follows: TC and EL, trained in ICF, performed a blind reading of the content and established the ICF domain and category for each significant unit of the interview to which it was judged to be relevant. VS, a specialist in ICF and with extensive experience in childhood disability, analyzed the disagreements of the two researchers, establishing consensus in relation to the domain/category of the ICF regarding the significant units. **Pag. 07**
26	Derivation of themes	Were themes identified in advance or derived from the data?	The themes were identified from the study data using the ICF model. **Pag. 07**
27	Software	What software, if applicable, was used to manage the data?	NVIVO software, version 11.0 (QSR International, 2019). **Pag. 07**
28	Participant checking	Did participants provide feedback on the findings?	No.
Reporting			
29	Quotations presented	Were participant quotations presented to illustrate the themes / findings? Was each quotation identified? e.g. participant number	The participants' quotes were presented to illustrate the themes without identification. **Pag 10-14**
30	Data and findings consistent	Was there consistency between the data presented and the findings?	Yes. **Pag 9-14**
31	Clarity of major themes	Were major themes clearly presented in the findings?	Yes. **Pag 9-14**
32	Clarity of minor themes	Is there a description of diverse cases or discussion of minor themes?	Yes. **Pag 9-14**