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Characterization of children with congenital zika virus syndrome using the international classification of functioning, disability, and health

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Abstract

Introduction: *The International Classification of Functioning, Disability, and Health (ICF) is useful for evaluating the variability of clinical outcomes in children with congenital Zika virus syndrome (CZVS) and understanding their functional profile.* ***Objective:*** *To characterize body functions of children with CZVS according to the ICF.* ***Methods:*** *This cross-sectional descriptive study included children with CZVS followed up by a physical therapist at a specialized health center. A data collection instrument evaluated sociodemographic and rehabilitation aspects, and the ICF was used.* ***Results:*** *Among ten evaluated children, 60% presented severe disability in tone of muscles of all limbs, and 80% presented moderate disability in mobility of joints generalized.* ***Final considerations:*** *The ICF allowed identifying variability of alterations in children with CZVS; the neuromuscular system was the most affected.*

Keywords: Zika virus. Microcephaly. International Classification of Functioning, Disability, and Health.

1 INTRODUCTION

Zika virus is an arbovirus of flavivirus genus (*Flaviviridae* family) transmitted through the *Aedes Aegypti*[1]. This virus is disseminated across Western regions due to the favorable environment for mosquito propagation, which lays eggs in stagnant water and feeds from human blood2. Vector control is challenging since breeding sites are related to deficits in basic sanitation, garbage coverage, and infrastructure of cities[3-5].

In November 2015, the Brazilian government announced the association between Zika virus and microcephaly and declared a nationwide emergency (Ordinance no. 1.813)[6]. This association was found in the study by Melo et al. 2016, who identified a positive histopathological exam for Zika virus in the amniotic fluid of

two pregnant women from Paraíba (Brazil) and fetal head circumference smaller than expected for gestational age using ultrasonography[7].

Microcephaly is one clinical outcome of the congenital Zika virus syndrome (CZVS)8. Furthermore, ocular anomalies (e.g., cataracts and intraocular calcifications), congenital contractures (e.g., arthrogryposis), congenital dislocation of hip and knee, convulsions, swallowing difficulties, cognitive alterations, and hearing impairments are clinical conditions found even not accompanied by microcephaly[9].

The variety of characteristics presented in CZVS led a multidisciplinary team to search for an instrument adequate for good therapeutic plan since this attribute was not found in instruments assessing only motor development. Therefore, the team considered the International Classification of Functioning, Disability, and Health (ICF) since it follows the biopsychosocial model[10].

The World Health Organization created the ICF, and the World Health Assembly approved this instrument to identify health determinants, such as environmental and personal factors11. The ICF offers a standardized language to compare evolution using codification (i.e., qualifiers indicating the magnitude of health level) and an alphanumeric system composed of four domains ("b" for body function; "s" for body structures; "d" for activities and participation; and "e" for environmental factors)[12].

Araújo (2013) used a multilinear perspective to indicate that the ICF demystified the disease-centered model. The author also reported the analysis of environmental factors as health determinants and suggested the ICF might be statistically and qualitatively applied as a potential indicator of health policies[13].

Specialized Rehabilitation Centers (SRC III) proposed by the Brazilian Ministry of Health are specialized health care centers that can articulate with other health care networks and aim to achieve functionality and increase the autonomy of users[14]. The SRC III combines three types of rehabilitation services (auditory, visual, physical, and intellectual) and performs diagnosis, treatment, concession, adaption, and maintenance of assistive technology[15].

Alternatively, the ICF should be implemented on health services because it presents a dynamic interaction between health problems and contextual factors (e.g., personal and environmental), resulting in an important instrument for

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therapeutic intervention, such as early introduction of assistive technology[16].

Considering the great variability of clinical conditions of children with CZVS and the need for knowledge about their functionality and disability profile, this study aimed to characterize body function of children with CZVS according to the ICF.

2 MATERIALS AND METHODS

This is a quantitative, cross-sectional, and descriptive study conducted with children diagnosed with CZVS and followed by a physical therapist at the SRC III related to a public university of Alagoas (Brazil). The research is part of the study "Characterization of disability profile and functionality of children with congenital Zika virus syndrome" and was approved by the research ethics committee (protocol no 2.678.950). Caregivers of children were informed about the study and signed the informed consent form.

This research included children with CZVS of both sex, aged between zero and three years, and attending the health service. Children with associated genetic syndromes and performing physical therapy treatment for less than three months at SRC III were excluded.

Data were collected during physical therapy follow-up at SRC III. A data collection instrument obtained information regarding sociodemographic and rehabilitation characteristics. Evaluators developed a checklist to apply the ICF in children with the help of the caregiver.

Children with CZVS were characterized using complementary exams and an objective data collection instrument containing sex, caregiver of the child, caregiver educational level, family income, origin of the child, and security benefit.

Due to age of children, caregivers judged situations listed in the ICF. Four qualifiers were used to classify body functions: .0 = no disability (0 to 4%); .1 = mild disability (5 to 24%); .2 = moderate disability (25 to 49%); .3 = severe disability (50 to 95%); and .4 = complete disability (96 to 100%).

Data were analyzed and deleted to ensure confidentiality of participants. Descriptive statistics was performed using the Statistical Package for the Social Sciences (SPSS) version 22.0 and represented as absolute (N) and relative (%) frequencies, mean, standard deviation, and minimum and maximum values.

3 RESULTS AND DISCUSSION

Ten children with CZVS (mean age of 18.20 ± 1.81 months; range, 10 to 20 months) participated in the study. Sample consisted of an equal number of male and female children, and half of them resided in the same municipality of attendance. Most caregivers (60%) had more than nine years of schooling, and all families had income below one minimum wage. Table 1 describes the characteristics of children with CZVS and their caregivers.

Sex was homogeneous in this study, different from Ventura et al. (2016) and Avelino and Ferraz (2018), which consisted of 60% (six) and 62.5% (five) female children, respectively[17,18]. In contrast, Petribu et al. (2017) studied a sample with 59% (22) male children[19].

The primary caregiver in this study was the mother and another family member with more than nine years of schooling. In contrast, Marinho et al. (2016) observed mothers with incomplete high school[20]. Family income of all children with CZVS in this study was less than one minimum wage, similar to Félix and Farias (2018) and Ferreira et al. (2018), in which parents lived with one monthly minimum wage from the child security benefit. In the present study, 20% (two) of children did not receive the security benefit[16,21], corroborating Gonçalves et al. (2018), in which 36.59% (15) of the sample did not receive the benefit[22]. These data follow the Brazilian Ordinance no. 58 (June 3rd, 2016) and the Provisional Measure no. 894 (September 4th, 2019), considering socioeconomic aspects described in our study[23,24].

The Brazilian Ministry of Social Development and Fight against Hunger recommended using the ICF to evaluate people with disabilities and access the Benefit of Continuous Provision of Social Assistance (BCP) since it structures complex evaluation systems and globally classifies with standardized language. This recommendation provides "a fair model for granting the benefit, following a global trend and considering medical and social models to access BCP"[25].

The Integrated Guidelines for Surveillance, Health Care, and Prevention of Congenital Syndrome associated with Zika virus in Alagoas (Brazil) was developed to guide health services. Among orientations, Health Information Systems request notification regarding serological screening, rapid tests, physical exams (including neurological exam), head circumference in the first 24 hours of life, imaging exams (e.g., transfontanellar ultrasound and cranial computed

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tomography for diagnostic confirmation), and neonatal screening (e.g., biological, hearing, and ocular)[26]. Children presenting alterations according to diagnosis should be referred to specialized health services[27]. The SRC III is an authorized and specialized health care service that covers 12 municipalities of the first health region[26].

The Brazilian Ministry of Health recommends early imaging exams for children at the first suspicion or confirmed exposition to Zika virus. Although computed tomography reconstructs skull bones three-dimensionally, this exam is poorly chosen. Cranial magnetic resonance is the reference exam in complex cases[14].

The protocol established by the State Department of Health of Alagoas chose computed tomography as imaging exam[26]. Findings of computed tomography in this study were categorized into calcification, ventriculomegaly, and lissencephaly. Results showed that 60% (6) of children did not have imaging data registered on medical records or with caregivers. Of caregivers with registered imaging data of children, 75% (3) presented the three mentioned alterations, 25% (1) presented only ventriculomegaly, and 25% (1) did not present calcifications.

Melo et al. (2016) observed two clinical cases with cerebral calcification, cerebral atrophy, corpus callosum dysgenesis, asymmetric cerebral hemispheres, severe unilateral ventriculomegaly, and disorganization of the brainstem[7]. Teixeira et al. (2020) suggested intracranial calcification, ventriculomegaly, and brain atrophy were responsible for signs and symptoms presented by children with CZVS[28]. These studies corroborate our findings regarding main neuroimaging alterations (cerebral calcification and ventriculomegaly).

One of two children that had electroencephalogram data presented alterations consistent with epilepsy. Only one child presented calcifications in the magnetic resonance imaging.

Only three children performed neonatal screenings for CZVS (e.g., hearing and ocular tests). Of these, one presented an altered ocular test (strabismus). After the outbreak of neurological manifestations, many cases reported optic neuritis[29]. França (2018) observed ophthalmological alterations, especially in the macula and optic nerve, in eight children with mean age similar to the present study (20.5 months)[30].

In our study, half of children with CZVS did not present cardiac exams. From

the other half, 40% (4) of children presented cardiac alterations. Interatrial communication and patent foramen ovale were observed in 60% (3) and 20% (1) of the studied children, respectively. Only 30% (3) of children had abdominal ultrasound data and normal results. Not all children had clinical data available for analysis, despite the Brazilian Ministry of Health and the State Department of Health recommend tests and screenings. The ICF was used to register the presence and magnitude of alterations found in body functions of children with CZVS (Table 2).

Half of children with CZVS were classified with no disability (.0) regarding seeing functions (b210), and the other 50% were classified with mild (3), moderate (1), or severe (1) disabilities. Seeing disabilities impair activities and participation of children since vision is important to explore the environment.

Most (90%) children presented no disability in hearing function (b230). In contrast, 60% of children presented severe disability (.3) in tone of muscles of all limbs (b7354), and 80% (8) presented moderate disability (.2) in mobility of joints generalized (b7102). Both were the most observed alterations.

The study of Leite et al. (2018) conducted a hearing screening with 45 children (mean age of 10 months), and most did not present cochlear or middle ear disorders[31]. In contrast, Leal et al. (2016) observed sensorineural hearing loss possibly related to Zika virus infection in a study with 70 children and described 5.8% of cases of hearing loss without other potential cause[32]. Leite et al. (2018) stated that newborns with a suspected infection should perform a hearing test, corroborating our study since most children did not present hearing screening test[31].

A mild alteration in heart function (b410) was observed in 30% of evaluated children. Costa (2018) observed 54.2% (32) of children had cardiac alterations; however, severity was not reported. Digestive functions (b515) were altered in 20% (2) of children (moderate disability), and 70% (7) did not present disabilities[33]. However, the health team must be aware of the possibility that children born with Zika virus present altered digestive function because most congenital syndromes cause digestive dysfunctions[34]. Moreover, Prata-Barbosa et al. (2019) suggest a direct relationship between CZVS and low weight[35].

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The Brazilian Ministry of Health highlights the increased muscle tone (flexors or extensors)[14], corroborating our study (60% of children with severe disability) and the study of Cardoso et al. (2019)(hypertonia found in 84.2% [16] of 19 studied children). This author also observed ataxia in half of the studied children, dyskinesia in eight children, some degree of asymmetry in six children, and irritability in eight children[36]. Altered muscle tone may trigger eating difficulties and lead to respiratory changes[23]. Although 80% (8) of children did not present altered respiratory functions (b440), attention and recommendations to this condition are needed. In this sense, the Department of Health of Alagoas emphasizes the importance of the health team in conducting recommendations about posture of children during and after feeding[26].

A cross-sectional study conducted recruited 34 children diagnosed with microcephaly due to CZVS to characterize the profile of disability and functionality. Although no children presented altered sleep functions (b134), 64.7% and 76.5% of them presented severe disability (.3) for items b710 (mobility of joints generalized) and b735 (tone of muscle of all limbs), respectively, corroborating the present study16. A case series with 18 children reported the association between Zika virus infection and arthrogryposis of hip, knee, ankle, and wrist joints[37]. These alterations may be related to peripheral motor nerve damage and fetal akinesia, resulting in joint stiffness and arthrogryposis.

4 CONCLUSIONS

The study demonstrated moderate to severe alterations in several body systems, especially neuromuscular (tone, mobility, and strength). Changes in seeing, hearing, sleep, and heart functions emphasize the systemic impairment and the need for attention of professionals to the variability of impairments in children with CZVS.

Barriers found in this study were related to availability of services, systems, policies for transport and health professionals, and difficulties accessing screening and complementary exams, despite recommendations of the Brazilian Ministry of Health. These factors directly affect activities and participation and prognosis of the child.

The ICF has the potential to indicate factors that need intervention, therapeutic strategies, and public health policies related to comprehensive childcare, encouraging

the use of the ICF among different professionals. The ICF also facilitates transdisciplinary communication between health, management, and administrative professionals to optimize the care of children with CZVS and their access to public policies.

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Table 1 – Characterization of children with congenital Zika virus syndrome and their family.

Variable	Number	Percentage
Child sex		
Female	5	50%
Male	5	50%
Child caregiver		
Mother	2	20%
Father	-	-
Grandmother	2	20%
Mother and another family member	6	60%
Caregiver educational level		
Illiterate	2	20%
≤ 4 years of study	-	-
5 to 8 years of study	2	20%
≥ 9 years of study	6	60%
Family income		
< 1 MW	10	100%
> 1 and < 2 MW	-	-
> 2 and < 3 MW	-	-
Child residence		
Same municipality of attendance	5	50%
Outside municipality of attendance	5	50%
Receiving security benefit		
Yes	8	80%
No	2	20%

MW: minimum wage.

Source: research data.

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Table 2 - Qualifiers designated to body function codes of children with congenital Zika virus syndrome.

Codes	.0	.1	.2	.3	.4
b210 Seeing functions	5 (50%)	3 (30%)	1 (10%)	1 (10%)	-
b230 Hearing functions	9 (90%)	1 (10%)	-	-	-
b440 Respiration functions	8 (80%)	2 (20%)	-	-	-
b134 Sleep functions	7 (70%)	1 (10%)	2 (20%)	-	-
b410 Heart functions	7 (70%)	3 (30%)	-	-	-
b515 Digestive functions	7 (70%)	1 (10%)	2 (20%)	-	-
b7354 Tone of muscles of all limbs	1 (10%)	1 (10%)	2 (20%)	6 (60%)	-
b7102 Mobility of joints generalized	1 (10%)	1 (10%)	8 (80%)	-	-

0: NO disability (no, absent, scarce) - 0 to 4%; xxx.1: MINOR disability (mild, small) 5 to 24%; xxx.2: MODERATE disability (average) 25 to 49%; xxx.3: SEVERE disability (great, extreme) 50 to 95%; xxx.4: COMPLETE disability (total) 96 to 100%.

Source: research data.