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Parental Stress in Primary Caregivers of Children with Evidence of Congenital Zika Virus Infection in Northeastern Brazil

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Abstract

Background—Despite the well-known role of parents as caregivers, few studies have addressed their health outcomes related to the Zika virus epidemic.

Methods—A cross-sectional study was carried out with 146 primary caregivers of children 15–26 months of age, with laboratory and/or clinical evidence of Zika infection between August and October 2017 in three Brazilian municipalities: João Pessoa and Campina Grande in the state of Paraíba and Fortaleza in the state of Ceará. Caregivers reported on their child's life and health, family circumstances and underwent screening for stress using the Parenting Stress Index-Short Form. Children were evaluated for developmental delays and clinical outcomes. Differences in the prevalence of risk factors between caregivers with high or clinically relevant stress and those with normal stress were evaluated.

Results—Of the 146 participants, 13% (n=19) were classified as having high or clinically relevant stress, all of them mothers. The two risk factors significantly and independently associated with high levels of stress, compared with individuals with normal stress levels, were

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"reporting difficulty in covering basic expenses" (adjusted OR 3.6 (95% CI 1.1–11.8; p = 0.034)) and "having a child with sleep problems" (adjusted OR 10.4 (95% CI 1.3–81.7; p = 0.026)).

Conclusions—Some factors seem to contribute significantly more than others to the level of stress experienced by caregivers of children with evidence of Zika virus congenital infection. Interventions and preventive strategies should also target caregivers, who in turn will be able to respond to the unique characteristics of their child.

Keywords

Zika virus infection; Developmental disabilities; Psychological stress; Coping behavior; Brazil

Introduction

Since the detection of an unexpected increase in cases of viral-associated exanthems in the Northeast Region of Brazil in February 2015 (Brasil 2015a) and given the recognized relationship of microcephaly and Zika virus (ZIKV) infection during pregnancy (Brasil 2015b); Oliveira Melo et al. 2016), there has been a large-scale public health response involving Brazilian government bodies, researchers, and local and international institutions. In April 2015, the circulation of ZIKV was confirmed (de Araújo et al. 2016), followed by the detection of an increase in neurological manifestations, specifically Guillain-Barré Syndrome (Malta et al. 2017), and in October, the detection, in Pernambuco State, of an increased number of children with microcephaly, with brain imaging tests compatible with congenital viral infection and reported exanthems in mothers during their pregnancy (Brasil 2015c).

Following this increase in reports of children with microcephaly, imaging of children with congenital Zika virus infection (CZI) detected cortical atrophy, cerebral calcifications, ventriculomegaly, epilepsy (Moore et al. 2017; Pessoa et al. 2018) and ocular abnormalities (Freitas et al. 2016). Developmental delay, cerebral palsy, seizures, and hearing loss (Malone et al. 2016; Satterfield-Nash et al. 2017) have also been seen in infants and children with CZI.

Previous literature has focused mainly on clinical outcomes following the recent ZIKV outbreak (Kapogiannis et al. 2017; Strafela et al. 2017), but few studies have investigated the mental health and family functioning of the primary caregivers of children affected by Zika. After the birth of an infant with developmental risks, primary caregivers may develop anxiety, depression and parental stress (Alves 2015). Parental stress can be described as the emotional strain experienced by parents in their roles as father or mother and is related to multiple factors including their own personality characteristics, individual stressors, parental health, needs and temperament of the child, family support system and the parent–child bond (Abidin 1992). The pressure of raising a child is an important concept in parenting and is closely related to parents' dysfunction (Östberg and Hagekull 2000), with different impacts on children with physical and mental health challenges (Feizi et al. 2014).

To date, in the context of the ZIKV epidemic, only two publications have reported outcomes related to primary caregiver's mental health (Oliveira et al. 2016, 2017). The mental health of parents and other primary caregivers, an important factor for children's health and development, should be considered at an individual integral health assistance level, which is a right guaranteed by law (Brasil 1990; Pinho et al. 2007). The investigation, Zika Outcomes and Development in Infants and Children (ZODIAC), sought to understand the relationship between children's health and development with primary caregiver stress and functioning among families affected by the 2015 Zika virus outbreak in Brazil (Satterfield-Nash et al. 2017). The present study examines levels of parental stress in primary caregivers of children between 15 and 26 months of age with evidence of CZI and their association with developmental delays, children's clinical outcomes, governmental support and demographic characteristics.

Methods

This cross-sectional descriptive study was carried out from August to October 2017, in two Northeastern States in Brazil, Paraíba and Ceará, which had the highest incidence of microcephalic infants reported in Brazil in 2016 and 2017 (Brasil 2017). For each child between 15 and 26 months of age with evidence of CZI, the primary caregiver was invited to participate as the person most responsible for the decision-making and daily care of this child.

In Paraíba, children were selected from a subset of participants in a previous case–control study (Krow-Lucal et al. 2018). They lived in Eastern Paraíba (regions I and II) and had either laboratory evidence of ZIKV infection, clinical evidence of ZIKV infection, or both. Laboratory evidence included enzyme-linked immunosorbent assay (IgM ELISA), plaque reduction neutralization test (PRNT), or ZIKV antibody titers/dengue antibody titers ratio < 1 for CZI. Clinical indication was defined as microcephaly at birth (< 32 cm if ≥ 37 weeks of gestation or, if preterm, < 3rd percentile for gestational age and sex, according to the International Consortium for Fetal and Neonatal Growth for the 21st Century—NTERGROWTH-21st) (International Fetal and Newborn Growth Consortium for the 21st Century, 2017), being small (head circumference to length ratio ≥ 0.65 and head circumference < 3rd percentile), or disproportionate (head circumference to length ratio ≤ 0.65) (Krow-Lucal et al. 2018). In Ceará, children were selected from cases with suspected CZI that had been reported in the Registry of Public Health Events-Microcephaly (RESP), were living in Fortaleza metropolitan area, and had either laboratory evidence of CZI or an available sample for laboratory testing collected at birth.

This investigation was approved by the National Committee for Ethics in Research (CONEP) and was carried out in accordance with current national ethics recommendations and have therefore been performed in accordance with the ethical standards laid down in the 1964 Declaration of Helsinki and its later amendments. Adult participants signed a written consent form for themselves and another for the child in their care. No identifiable individual data were shared or published.

The current study is part of the ZODIAC investigation, which also involved a number of assessments of enrolled children, such as growth, head circumference, neurological, vision, blood tests and developmental tests (Satterfield-Nash et al. 2017); if indicated, children were referred for computed tomography brain scan and audiological examination. The validated Brazilian Portuguese version of the 3rd edition of the Ages and Stages questionnaire (ASQ) was used to evaluate child developmental function (Filgueiras et al. 2013; Squires et al. 2009). The ASQ is a screening instrument composed of 21 questionnaires used to evaluate children from 1 month to 5 years of age. Each questionnaire is designed for a certain age range and consists of 5 domains: communication, gross motor coordination, fine motor coordination, problem solving, and personal-social. For this study, the ASQ protocol has been modified (Kotzky et al. 2019): instead of using the age-specific questionnaire for each child as is usual for the ASQ, all evaluations started with the 6 months of age questionnaire, for each domain, and then proceeded to the next questionnaire when all the milestones for that specific domain were reached. This adapted protocol was used because of the challenges of starting with the administration of the questionnaire appropriate for a child's biological age and working backward, given the severe developmental delays among the children studied.

Developmental Quotients (DQ) scores were calculated for each domain, transformed into z-scores, and adjusted to the distribution of ASQ-3 scores for Brazilian children (Filgueiras et al. 2013). These DQ z-scores indicated how far above or below a child's development score was, compared to children of the same biological age, in standard deviations (SD). After evaluation, children were classified, based on ASQ cutoff points by SDS (Squires et al. 2009), into three groups according to DQ z-scores: group 1 (global delay), which had DQ z-scores greater than or equal to 2 SD below the mean in all 5 domains; group 2 (some delay), which had DQ z-scores greater than or equal to 1 SD below the mean in at least one domain but not greater than 2 SD below the mean in all domains; Group 3 (no delay), which had DQ z-scores that were less than 1 SD below the mean on all domains.

Children's neurological function was assessed using the Hammersmith Infant Neurological Examination (HINE). The HINE is a standardized exam that can be used to evaluate children from 2 to 24 months of age and contains 26 items in 5 domains. The sum of the scores in each domain provides a global score, ranging from 0 to 78. It can be used to diagnose cerebral palsy (CP), type and severity of CP, and severity of motor impairment. For this study, children with a score below the 40 point cut-off (Romeo et al. 2015) were classified as having severe motor impairment.

Interviews were conducted with primary caregivers of these children using questionnaires to collect sociodemographic data, receipt of government assistance, level of difficulty in covering basic expenses, and perceptions of child's breastfeeding, sleep, vision, hearing and swallowing. Primary caregivers were also screened for depression and stress. The short form of the 4th edition of the Parenting Stress Index (PSI-SF), validated in Brazilian Portuguese (Pereira et al. 2016), was used to assess parental stress. The PSI-SF is used to screen parental stress in the parent/child relationship, identifying need for follow-up services. It consists of 36 questions divided into three domains (parental anxiety, dysfunctional parent-child interaction, and difficult child) and allows the calculation of a total parental stress index. Each domain has 12 items rated on a 5-point Likert scale: "I totally agree" = 1, "I agree" =2, "I am not sure"=3, "I disagree" =4, and "I totally disagree" =5. Scores can be calculated separately for each domain, ranging from 12 to 60 points. The total score was used in this study, calculated by summing the 3 domain totals, ranging from 36 to 180 points, and then converting this number to a percentile according to the developer's guidance (Abidin 2012). In general, the normal range of scoring is within the 16th and 84th percentiles. Scores in the 85th to 89th percentiles are considered high and scores in the 90th percentile or higher are considered clinically relevant (Abidin 2012).

Our main dependent variable was the level of parental stress of primary caregivers of children with developmental delays possibly related to CZI. Independent variables included: ASQ delay groups, receipt of government assistance, difficulty in covering basic expenses, child's current microcephaly status, child's age and overall HINE score. Primary caregivers' socio-demographic characteristics (gender, relationship to the child, age, race, education, family income and employment challenges) and primary caregiver's perception of child's health (eating, sleeping, vision and hearing problems) were also included as independent variables.

Descriptive exploratory analysis was done by examining and comparing the frequencies of the variables. We used Chi-square (χ^2) and Fisher's exact test to test the association between stress level and selected risk factors. We also calculated crude and adjusted odds ratios (OR) and their 95% confidence intervals, using logistic regression, and an alpha level of 0.05, to assess the difference in risk between the two stress levels considered in this study (normal and high or clinically relevant). The Mantel–Haenszel stratified analyses test was used to analyze modification effects among the variables based on the background social and epidemiological knowledge. We crossed 'difficulty in covering basic expenses' with 'parenting stress', controlling by 'government assistance'. And we crossed 'developmental delays' with 'parenting stress', controlling by 'difficulty in covering basic expenses'. We performed Shapiro–Wilk normality tests to identify normally distributed data, however, if the variables tested was not normally distributed, we used non-parametric tests (Mann–Whitney) and for continuous normally distributed variables, we used parametric tests (Student's t-test). The variables selected for multivariable analysis were those with a p-value lower than 0.20 and that didn't presented collinearity with other variables, assessed by a correlation matrix. The 'stepwise backward LR' method was used to select variables, adopting the alpha level of 0.20 for input and 0.05 for the output of variables in the model. Binary logistic regression was used to examine if covariates were predictors of primary caregiver stress.

Data were collected using Research Electronic Data Capture software (REDCap), and analyzed using IBM SPSS Statistics 25.0 software. Each family (child and primary caregiver) was anonymized using a unique identifier ID.

Results

In Ceará, 52 eligible primary caregivers were identified. Of these, 18 (34.6%) could not be contacted, five (9.6%) refused to participate or did not attend the evaluations, and four (7.7%) were excluded from the sample due to missing data on critical fields for classification at the time of validation and database cleaning. In Paraíba, 273 eligible primary caregivers were identified. Of these, 75 (27.5%) could not be contacted, 76 (27.8%) refused to participate or did not attend the evaluations and one participant was excluded from the sample because of inconsistent information. In all, 146 primary caregivers were evaluated, 25 (17.1%) from Ceará and 121 (82.9%) from Paraíba.

According to the distribution of the total score of the PSI-SF test, primary caregivers were separated into normal stress (n= 127; 87.0%) and high or clinically relevant stress (n= 19; 13.0%). The majority (99.3%) of the primary caregivers were female and the median age was 28 (ranging from 16 to 62) years of age. Among children, 72 (49.3%) were female and the median age was 22 (ranging from 15 to 26) months of age. Additional demographic data are shown in Table 1.

In the bivariate analysis, primary caregivers with high or clinically relevant parental stress presented 4.8 (95% CI 1.5–15.4) times the odds of difficulty covering basic expenses and 4.8 (95% CI 1.8–13.5) times the odds of employment challenges (quit job or cutting down on work hours because of the child's health), when compared to those with normal stress level. On the other hand, no statistically significant associations were found between stress level and primary caregiver's age (OR= 1.3; 95% CI 0.5–3.5), race (OR=0.8; 95% CI 0.3–2.7), education level (OR= 1.6; 95% CI 0.6–4.2) or government support—cash, food or child care (OR= 1.1; 95% CI 0.3–4.2).

Regarding caregiver's level of stress by child's characteristics, the odds of presenting high or clinically relevant stress was significantly greater among those who cared for children with sleeping problems (OR 12.9; 95% CI 1.7–99.6). No statistically significant association was found between stress level and child's vision problems (OR 1.2, 95% CI 0.4–4.1), hearing problems (OR 2.3; 95% CI 0.2–23.5), eating or swallowing problems (OR 2.4; 95% CI: 0.9 to 6.5) or with 'global developmental delay' on ASQ (OR 3.5; 95% CI: 0.9–13.4).

After adjusting for key variables (ASQ group and government support), in the logistic regression model, the variables 'sleeping problems' (OR 10.4; 95% CI= 1.3–81.7) and 'difficulty covering basics' (OR 3.6; 95% CI 1.1–11.8) were significant predictors of high or clinically relevant stress among primary caregivers.

Discussion

In our univariate analysis, primary caregivers of children in the ASQ 'global delay' group had higher odds of high or clinically relevant stress when compared to primary caregivers of children without such delay, which is consistent with Hayes and Watson's findings in parents of children with autism (Hayes and Watson 2013). In the multivariable analysis, however, "sleeping problem" was the only child-related variable significantly associated with stress in the primary caregiver which showed no statistical association with developmental delay.

Contrary to our expectations and dos Santos Oliveira's observations regarding anxiety and depression (Oliveira et al. 2017), no statistical relationship was found between CZI-associated microcephaly and parental stress. It is important to highlight the difference in the time elapsed between the births of the children and the data collections of the two studies. In the present study, children were evaluated were between 15 and 26 months of age, a period in which parental psychological adaptation and resilience may have developed, as Halstead points out in mothers of children (Halstead et al. 2018).

Another possibility is to analyze, like Hastings, the family as a complex system in which all members are likely to be influenced by each other and by external factors, not only by having a child with developmental delays (Hastings 2016). Several studies point to a bidirectional relationship between parental stress and child behavior, such as Woodman and colleagues who studied these relationships longitudinally over a 15-year period, reporting differences between life stages and the nature of the behavioral problem (internalized or externalized) (Woodman et al. 2015).

During the study period and only in Ceará, the government offered psychological assistance to mothers of children with microcephaly, while the child received routine health care. We did not analyze the potential benefits of these services; however, our findings suggest a need for psychological assistance and other types of government support among these children and their families.

This study has some limitations. First, regarding comparison and discussion of the findings, few national studies have evaluated the physical and mental health of the primary caregivers of children born in the context of the ZIKV epidemic. Therefore, the findings of this study were compared to others with themes of parental stress, but with different exposure factors or diagnoses such as Down Syndrome, autism and presence of externalizing behavior problems. Second, information about participants' state and municipalities of residence were not included in the analyses, and structural differences between locations and availability of social support that could possibly buffer parental stress were not measured. Third, parental self-report could introduce several response biases that could not be ruled out or controlled for in the present study. Fourth, the relationships that were observed are limited by the reliance on cross-sectional designs and might be better explicated using longitudinal designs. And finally, we acknowledge the low response rate, for which the reason may have been stress itself, since caregivers were going through delicate moments with the infants. A higher response rate would have increased the power to detect associations of the variables of interest with parental stress, such as "feeding/swallowing problems" and "global development delay on ASQ".

As Zika is an emerging disease of interest with recently defined clinical implications, there are still many questions to be answered about the consequences of CZI, especially in the long term health and development for children, their parents, and other primary caregivers. This study described indicators of parental stress in this context, contributing to knowledge about the extent of health and social concerns among families affected by the ZIKV epidemic. Providing more targeted support on child's behavioral issues, specifically sleeping problems as shown in this study, may help reduce parental stress in families of children with developmental delays, including CZI.

Other longitudinal studies with larger populations may add external validity to the findings. Also other modeling techniques are encouraged, such as Directed Acyclic Graph (DAG), working with causal pathways. In addition to actions that support the health and development of affected children, it may be necessary to structure a network that supports mothers and other caregivers concomitantly with their emotional needs and demands. Addressing parental stress should be an integral part of comprehensive care for the families of all children, especially those presenting with developmental delays.

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Abbreviations

ASQ	Ages and stages questionnaire
CONEP	National Committee for Ethics in Research
CP	Cerebral palsy
CZI	Congenital Zika infection
DAG	Directed acyclic graph
DQ	Developmental quotients
ELISA	Enzyme-linked immunosorbent assay
INTERGROWTH	International Consortium for Fetal and Neonatal Growth for the 21st century
HINE	Hammersmith infant neurological examination
OR	Odds ratio
PSI	Parenting stress index
PRNT	Plaque reduction neutralization test
REDCap	Research Electronic Data Capture software
RESP	Registry of Public Health Events-Microcephaly

SD	Standard deviation
ZIKV	Zika virus
ZODIAC	Zika outcomes and development in infants and children

References

Abidin RR (1992). The determinants of parenting behavior. Journal of Clinical Child Psychology, 21(4), 407–412. 10.1207/s15374424jccp2104.

Abidin RR (2012). Parenting stress index short form—Fourth Edition (PSI-4-SF) (4th ed.). Lutz, FL: Psychological Assessment Resources.

Alves GMAN (2015). Master's deegree: Indicadores de estresse, ansiedade e depressão de mães de bebês com risco ao desenvolvimento. Faculdade de Ciéncias: Universidade Estadual Paulista Julio de Mesquita Filho.

Brasil. (1990). Lei n° 8.080, de 19 de setembro de 1990. Lei Orgânica da Saúde. Dispõe sobre as condições para a promoção, proteção e recuperação da saúde, a organização e o funcionamento dos serviços correspondentes e dá outras providências. Brasília.

Brasil. Ministério da Saúde. Portaria GM n° 1.813, de 11 de novembro de 2015. Declara Emergência em Saúde Pública de importância Nacional (ESPIN) por alteração do padrão de ocorrência de microcefalias no Brasil, Pub. L. No. Portaria GM n° 1.813, de 11 de novembro de 2015 (2015a). Diário Oficial da República Federativa do Brasil, Brasilia (DF), 2015 nov 12;Seção 1:51.

Brasil. Ministério da Saúde. Secretaria de Vigilância em Saúde. (2015b). Nota informativa n0 01/2015 – COES microcefalias. Brasília.

Brasil. Ministério da Saúde. Secretaria de Vigilância em Saúde. (2015c). Protocolo de Vigilância e Resposta A Microcefalia Relacionada À Infecção Pelo Vírus Zika. Versão, 1(2), 70. 10.7554/eLife.15272.

Brasil. Ministério da Saúde. Secretaria de Vigilância em Saúde. (2017). Monitoramento integrado de alterações no crescimento e desenvolvimento relacionadas à infecção pelo vírus Zika e outras etiologias infecciosas, da Semana Epidemiológica 45/2015 até a Semana Epidemiológica 02/2017. Boletim Epidemiológico, 48(6), 1–18. 10.1590/S1415-790X2004000400010.

de Araújo TVB, Rodrigues LC, Ximenes R. A. de A., Miranda-Filho D. de B., Montarroyos UR, de Melo APL, … Martelli CMT (2016). Association between Zika virus infection and microcephaly in Brazil, January to May, 2016: preliminary report of a case–control study, 16, 1356–1363. 10.1016/S1473-3099(16)30318-8

de Pinho LB, Kantorsk LP, Saeki T, de Duarte M, & C. L, & Sousa J. de. (2007). A integralidade no cuidado em saúde: um resgate de parte da produção científica da área. Revista Eletrônica de Enfermagem, 09(03), 835–846.

Feizi A, Najmi B, Salesi A, Chorami M, & Hoveidafar R (2014). Parenting stress among mothers of children with different physical, mental, and psychological problems. Journal of Research in Medical Sciences, 19(2), 145–152. [PubMed: 24778669]

Filgueiras A, Pires P, Maissonette S, & Landeira-Fernandez J (2013). Psychometric properties of the Brazilian-adapted version of the Ages and Stages Questionnaire in public child daycare centre. Early Human Development, 89(8), 561–576. 10.1016/j.earlhumdev.2013.02.005. [PubMed: 23507472]

Freitas BDP, Dias JRDO, Prazeres J, Sacramento GA, Ko AI, Maia M, & Belfort R (2016). Ocular findings in infants with microcephaly associated with presumed zika virus congenital infection in Salvador, Brazil. JAMA Ophthalmology, 134(5), 529–535. 10.1001/jamaophthalmol.2016.0267. [PubMed: 26865554]

Halstead E, Ekas N, Hastings RP, & Griffith GM (2018). Associations between resilience and the well-being of mothers of children with autism spectrum disorder and other developmental disabilities. Journal of Autism and Developmental Disorders, 48(4), 1108–1121. 10.1007/s10803-017-3447-z. [PubMed: 29322385]

Hastings RP (2016). Do children with intellectual and developmental disabilities have a negative impact on other family members? The case for rejecting a negative narrative a. International Review of Research in Developmental Disabilities (Vol. 50). Elsevier Ltd. 10.1016/bs.irrdd.2016.05.002.

Hayes SA, & Watson SL (2013). The impact of parenting stress: A meta-analysis of studies comparing the experience of parenting stress in parents of children with and without autism spectrum disorder. Journal of Autism and Developmental Disorders, 43(3), 629–642. 10.1007/s10803-012-1604-y. [PubMed: 22790429]

International Fetal and Newborn Growth Consortium for the 21st Century. (2017). Standards for newborns and references for very preterm infants. Recuperado de https://intergrowth21.tghn.org/.

Kapogiannis BG, Chakhtoura N, Hazra R, & Spong CY (2017). Bridging knowledge gaps to understand how Zika virus exposure and infection affect child development. JAMA Pediatrics, 171 (5), 478–485. 10.1001/jamapediatrics.2017.0002. [PubMed: 28241263]

Kotzky K, Allen JE, Robinson LR, Satterfield-Nash A, Bertolli J, Smith C, … Peacock G (2019). Depressive symptoms and care demands among primary caregivers of young children with evidence of congenital Zika virus infection in Brazil. The Journal of Developmental and Behavioral Pediatrics. 10.1097/DBP.0000000000000666.

Krow-Lucal ER, de Andrade MR, Cananéa JNA, Moore CA, Leite PL, Biggerstaff BJ, … Arena JF (2018). Association and birth prevalence of microcephaly attributable to Zika virus infection among infants in Paraíba, Brazil, in 2015–16: a case-control study. The Lancet Child & Adolescent Health, 2(3), 205–213. 10.1016/S2352-4642(18)30020-8. [PubMed: 30169255]

Malone RW, Homan J, Callahan MV, Glasspool-Malone J, Damodaran L, Schneider ADB, … Weaver S (2016). Zika virus: Medical countermeasure development challenges. PLoS Neglected Tropical Diseases, 10(3), 1–26. 10.1371/journal.pntd.0004530.

Malta JMAS, Vargas A, Leite P. L. e, Percio J, Coelho GE, Ferraro AHA, … Saad E (2017). Síndrome de Guillain-Barré e outras manifestações neurológicas possivelmente relacionadas à infecção pelo vírus Zika em municípios da Bahia, 2015. Epidemiologia e Serviços de Saúde, 26(01), 09–18. 10.5123/S1679-49742017000100002.

Moore CA, Erin Staples J, Dobyns WB, Pessoa A, Ventura CV, Borges da Fonseca E, … Rasmussen SA (2017). Characterizing the pattern of anomalies in congenital Zika syndrome for pediatric clinicians. JAMA Pediatrics, 171(3), 288–295. 10.1001/jamapediatrics.2016.3982. [PubMed: 27812690]

Oliveira Melo AS, Malinger G, Ximenes R, Szejnfeld PO, Alves Sampaio S, & Bispo De Filippis AM (2016). Zika virus intrauterine infection causes fetal brain abnormality and microcephaly: Tip of the iceberg? Ultrasound in Obstetrics and Gynecology. 10.1002/uog.15831.

Oliveira SJGS, de Melo ES, Reinheimer DM, Gurgel RQ, Santos VS, & Martins-Filho PRS (2016). Anxiety, depression, and quality of life in mothers of newborns with microcephaly and presumed congenital Zika virus infection. Arch Women' Ment Health. 10.1007/s00737-016-0654-0.

Oliveira SJGS, dos Reis CL, Cipolotti R, Gurgel RQ, Santos VS, & Martins-Filho PRS (2017). Anxiety, depression, and quality of life in mothers of newborns with microcephaly and presumed congenital Zika virus infection: a follow-up study during the first year after birth. Archives of Women's Mental Health, 20(3), 473–475. 10.1007/00737-017-0724-y.

Östberg M, & Hagekull B (2000). A structural modeling approach to the understanding of parenting stress. Journal of Clinical Child Psychology, 29(4), 615–625. 10.1207/S15374424JCCP2904_13. [PubMed: 11126638]

Pereira LM, Viera CS, Toso BRGO, Carvalho ARS, & Bugs BM (2016). Validação da escala Índice de Estresse Parental para o portugues do Brasil. Acta Paulista de Enfermagem, 29(6), 671–677. 10.1590/1982-0194201600094.

Pessoa A, van der Linden V, Yeargin-Allsopp M, Carvalho MDCG, Ribeiro EM, Van Naarden Braun K, … Moore CA (2018). motor abnormalities and epilepsy in infants and children with evidence of congenital Zika virus infection. Pediatrics, 141(Supplement 2), S167–S179. 10.1542/peds.2017-2038F. [PubMed: 29437050]

Romeo DM, Ricci D, Brogna C, & Mercuri E (2015). Use of the Hammersmith Infant Neurological Examination in infants with cerebral palsy: A critical review of the literature. Developmental Medicine and Child Neurology, 58(3), 240–245. 10.1111/dmcn.12876 [PubMed: 26306473]

Satterfield-Nash A, Kotzky K, Allen J, Bertolli J, Moore CA, Pereira IO, … Peacock G (2017). Health and Development at Age 19–24 Months of 19 children who were born with microcephaly and laboratory evidence of congenital Zika virus infection during the 2015 Zika virus outbreak—Brazil, 2017. MMWR Morbidity and Mortality Weekly Report, 66(49), 1347–1351. 10.15585/mmwr.mm6649a2. [PubMed: 29240727]

Squires J, Twombly E, Bricker D, & Potter L (2009). ASQ-3 user's guide (3rd ed.). Washington: Paul H. Brookes Pub.

Strafela P, Vizjak A, Mraz J, Mlakar J, Pizem J, Tul N, … Popovic M (2017). Zika virus-associated micrencephaly: A thorough description of neuropathologic findings in the fetal central nervous system. Archives of Pathology and Laboratory Medicine, 141(1), 73–81. 10.5858/arpa.2016-0341-SA. [PubMed: 27726416]

Woodman AC, Mawdsley HP, & Hauser-cram P (2015). Research in Developmental Disabilities Parenting stress and child behavior problems within families of children with developmental disabilities : Transactional relations across 15 years. Research in Developmental Disabilities, 36, 264–276. 10.1016/j.ridd.2014.10.011.

Significance statement

What is already known on this subject?

Until the end of this article, only two publications have demonstrated outcomes related to the mental health of caregivers in the context of the recent Zika virus epidemic. The pressure of raising a child is closely related to parents' dysfunction, with different impacts on children with physical and mental health challenges.

What this study adds?

We report on parental stress in primary caregivers of children with evidence of congenital Zika virus infection and its relation to children's developmental delays and clinical outcomes. Contributing to build knowledge about the extent of health and social concerns of the Zika epidemic among affected families.

Table 1

Clinical and demographic characteristics of caregivers and their children with evidence of congenital Zika virus infection according to parental stress level

Variable	Parental stress level				OR	CI (95%)	p-value	OR[d]	CI (95%)	p-value
	High or Clinically Relevant (n = 19)		Normal (n = 127)							
	n	%	n	%						
Caregiver's characteristics										
Sex[c]										
Female	19	100.0	126	99.2	0.3	0.0–3.6	0.732[b]	-	-	-
Male	0	0.0	1	0.8	Ref					
Relationship[c]										
Mother	19	100.0	120	94.5	1.3	0.2–11.2	1.000[b]	-	-	-
Other relationship	0	0.0	7	5.5	Ref					
Age (years)										
<24	8	42.1	45	35.4	1.3	0.5–3.5	0.573[a]	-	-	-
≥24	11	57.9	82	64.6	Ref					
Race										
Black	15	78.9	104	81.9	0.8	0.3–2.7	0.755[b]	-	-	-
Non-Black	4	21.1	23	18.1	Ref					
Education level										
No formal schooling to 8 years	8	42.1	40	31.5	1.6	0.6–4.2	0.359[a]	-	-	-
9 +	11	57.9	87	68.5	Ref					
Household income per month (Brazilian Real)										
<R$500	15	83.3	95	79.2	1.0	0.3–3.4	1.000[b]	-	-	-
≥R$500	3	16.7	25	20.8	Ref					
Difflculty covering basic expenses										
Yes	15	78.9	55	43.7	4.8	1.5–15.4	0.004[a]	3.6	1.1–11.8	0.034
No	4	21.1	71	56.3	Ref					
Employment challenges										

Variable	Parental stress level				OR	CI (95%)	p-value	OR[d]	CI (95%)	p-value
	High or Clinically Relevant (n = 19)		Normal (n = 127)							
	n	%	n	%						
Yes	12	63.2	33	26.0	4.9	1.8–13.5	0.001[a]	-	-	-
No	7	36.8	94	74.0	Ref					
Government support										
Yes	16	84.2	105	82.7	1.1	0.3–4.2	1.000[b]	1.1	0.3–4.4	0.943
No	3	15.8	22	17.3	Ref					
Child’s characteristícs										
Sex										
Female	9	47.4	63	49.6	0.9	0.4—2.4	0.856[a]	-	-	-
Male	10	52.6	64	50.4	Ref					
Age (months)										
15 to 22 months of age	13	68.4	72	56.7	1.7	0.6–4.6	0.334[a]	-	-	-
23 to 26 months of age	6	31.6	55	43.3	Ref					
Head circumference Classification										
Microcephalic	7	36.8	40	31.5	1.3	0.5–3.5	0.642[a]	-	-	-
Not microcephalic	12	63.2	87	68.5	Ref					
Development delay (ASQ)										
Global delay	11	57.9	47	37.0	3.5	0.9–13.4	0.054[a]	3.0	0.7–12.4	0.136
Some delay	5	26.3	35	27.6	2.1	0.5–9.6	0.460[b]	2.0	0.4—9.2	0.400
No delay	3	15.8	45	35.4	Ref					
Motor impairment (HINE)										
Yes	8	42.1	35	27.6	1.9	0.7–5.2	0.195[a]	0.9	0.2–4.6	0.851
No	11	57.9	92	72.4	Ref					
Ealing or swallowing problems										
Yes	9	47.4	34	27.0	2.4	0.9–6.5	0.070[a]	0.8	02–2.1	0.737
No	10	52.6	92	73.0	Ref					
Sleep problems										

Variable	Parental stress level				OR	CI (95%)	p-value	OR[d]	CI (95%)	p-value
	High or Clinically Relevant (n = 19)		Normal (n = 127)							
	n	%	n	%						
Yes	18	94.7	74	58.3	12.9	1.7–99.6	0.002[a]	10.4	1.3–81.7	0.026
No	1	5.3	53	41.7	Ref					
Vision problems										
Yes	4	22.2	23	18.9	1.2	0.4—4.1	0.751[b]	-	-	-
No	14	77.8	99	81.1	Ref					
Hearing problems										
Yes	1	5.6	3	2.5	2.3	0.2–23.5	0.430[b]	-	-	-
No	17	94.4	118	97.5	Ref					

The odds ratio (OR) and 95% confidence interval are for the risk of presenting high or clinically relevant levels of stress given the presence of the risk factor (Ref is the reference group)

[a] χ^2 test

[b] Fisher's Exact Test

[c] Variable artiflcialized for containing value equal to zero

[d] Odds Ratio adjusted for eating or swallowing problem, Motor impairment (HINE), Developmental Delays (ASQ) and govemment support

[e] R$500.00= 154.32 US$ by 2017