FACIAL ANALYSIS OF CHILDREN WITH MICROCEPHALY IN THE PRIMARY DENTITION

ANÁLISE FACIAL DE CRIANÇAS COM MICROCEFALIA NO ESTÁGIO DA DENTADURA DECÍDUA

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ABSTRACT
Oral health care involved dental development, the stomatognathic system and craniofacial growth in early childhood in order to significantly reduce curative measures. But, what about individuals with microcephaly? The present study explores the morphological diagnosis of these children's faces. The study population corresponded to 7 Brazilian children with microcephaly, without associated syndromes in the complete deciduous dentition. The data collection consisted of the analysis of the facial characteristics obtained from clinical records and photos of the front and profile of the face at rest. In the frontal norm analysis, the predominance of dolichofacial type (100%) was observed. In the lateral norm analysis, there was a predominance of Pattern II children (85.71%) in relation to Pattern I (14.29%). It was also assessed that the proportion of severe Microcephaly (-3 SD) was higher in males (p = 0.04). There was no association between the severity of microcephaly and the facial type (p> 0.05), although the Type II pattern was more prevalent in severe microcephaly (100% of cases). The alert for the longitudinal monitoring of the mandibular growth vector of these individuals is essential, which is significant for the diagnosis and prognosis of orthopedic treatment while there is craniofacial growth.
Keywords: Deciduous Tooth. Face. Microcephaly.

RESUMO
A atenção à saúde oral engloba o desenvolvimento dentário, o sistema estomatognático e o crescimento craniofacial na primeira infância a fim de reduzir significativamente medidas curativas. Mas, e quanto aos indivíduos com microcefalia? O presente estudo explora o diagnóstico morfológico da face destas crianças. A população do estudo correspondeu a 7 crianças brasileiras com microcefalia, sem síndromes associadas e dentadura decídua completa. A coleta de dados consistiu na análise das características faciais obtidas de fichas clínicas e fotos de frente e perfil da face em repouso. Na análise em norma frontal, foi observada a predominância do tipo dolicofacial (100%). Na análise em norma lateral verificou-se o predomínio de crianças Padrão II (85,71%) em relação ao Padrão I (14,29%). Foi avaliado também que a proporção de Microcefalia grave (-3 DP) foi maior no sexo masculino (p= 0,04). Não se observou associação entre a gravidade da microcefalia e o tipo facial (p> 0,05), embora o padrão Tipo II tenha sido mais predominante na microcefalia grave (100% dos casos). É fundamental o alerta para o acompanhamento longitudinal do vetor de crescimento mandibular destes indivíduos, significativo para diagnóstico e prognóstico do tratamento ortopédico enquanto há crescimento craniofacial.
Palavras-chave: Dente Decíduo. Face. Microcefalia.

INTRODUCTION

Microcephaly is a clinical condition in which the child has a significant reduction in the occipitofrontal circumference of the head when compared to others of the same sex and age. In addition to the craniofacial disproportion, it can cause joint malformations of the limbs, changes in neuropsychomotor growth and development, as well as hearing and visual difficulties (LEITE; VARELLIS, 2016; PEREIRA *et al.*, 2017; ARROYO, 2018; DEVAKUMAR *et al.*, 2018). In Brazil, in 2016, the Ministry of Health adopted operational definitions based on the measurement of head circumference (HC). In accordance with the WHO, a normative reference was established, for the first 24-48 hours of life, by the InterGrowth parameters for both sexes. In this new HC reference table, the measurement for newborns (NB) is 31.5 cm for girls and 31.9 cm for boys (INTERGROWTH-21st, [sd]; WORLD HEALTH ORGANIZATION, 2016; BRAZIL, 2017). The WHO standardizes as Microcephaly: NB with an HC of less than -2 standard deviations (SD), that is, more than 2 SD below the average for gestational age and sex. Severe microcephaly: NB with an HC less than -3 SD, that is, more than 3 SD below the average for gestational age and sex (WORLD HEALTH ORGANIZATION, 2016).

From the moment it forms, in the embryonic period, until it reaches its definitive dimension, in skeletal maturity, the face emerges from the skull base, propelled in the three directions of space. In individuals with no congenital malformations involving the face, there is a genetic determinism for the facial configuration, which ends up being a morphogenetics product. Extragenetic factors become more important when congenital malformations mutilate the anatomy (LEWIS; ROCHE; WAGNER, 1985; SILVA FILHO, 1989; CAPELOZZA FILHO, 2005). In early childhood, the face has not yet reached its definitive dimensions. The maxilla and mandible have not reached the final dimension and will show considerable growth until skeletal maturity. However, the tendency is for the facial configuration to remain constant during growth, which is why, at this stage, it should be identified, defining the facial pattern of the individual (ENLOW; HANS, 1998; SILVA FILHO *et al.*, 2008).

However, although craniofacial growth and development are directly associated with genetic factors, they are strongly influenced by the functional pattern of the orofacial muscles. In this context, can this congenital malformation interfere with growth and development? In what magnitude and direction will growth be expressed?

Faced with this reality, with the increase in the number of cases of microcephaly in Brazil and based on scientific concepts that the facial pattern is maintained during growth, knowing the face morphologically by the disposition of the soft tissue of children with microcephaly even in early childhood, will contribute in a meaningful way for health professionals to adequately carry out preventive and interceptive measures in these individuals, fundamental points for defining a realistic prognosis and elaborating a health care protocol. This study aimed to analyze the facial morphological characteristics at rest in the stage of the complete deciduous dentition of children with microcephaly living in Jequié and neighboring municipalities in Bahia.

MATERIAL AND METHODS

This is a cross-sectional clinical study carried out after approval by the Ethics Committee of the State University of Southwest Bahia (UESB) under protocol No. 3,932,364/2019 according to the recommendations contained in the Declaration of Helsinki.

The norms of the guidelines for observational studies were followed as described by Von Elm *et al.* (2009). The study population involved children diagnosed with newborn microcephaly, both sexes, Brazilians, from 3 to 6 years of age, with complete primary dentition. As inclusion criteria, all children should have clinical records, frontal and profile facial photographs at rest and intraoral photographs obtained from the UESB Dentistry Module documentation file from October to December 2019. Children diagnosed with syndromes, as well as those that had a permanent or partially erupted permanent dentition.

The diagnosis of microcephaly (NB with a head circumference less than -2 standard deviations for gestational age and sex) or severe microcephaly (NB with a head circumference less than -3 standard deviations for gestational age and sex) was obtained from the records of these clinical files, as well as data referring to sex, age and color/race (IBGE, 2000; INTERGROWTH-21st, [sd]; WORLD HEALTH ORGANIZATION, 2016). The classification of the complete deciduous dentition was obtained from the clinical records and analysis of the intraoral photographs of these children.

The facial examination of each child consisted of clinical morphological analysis, seen in frontal and profile views, present in the upper, middle and lower facial thirds. The sagittal and vertical facial characteristics were based on the concept of pattern organized by Capelozza Filho (2005). In the sagittal analysis, the face can be grouped into three distinct patterns: Pattern I, Pattern II, Pattern III. The Pattern I reflects a facial balance, in which the maxilla and mandible are well related to each other and a harmonious face. In addition, they have as characteristics: facial symmetry, proportion and balance between the facial thirds, good zygomatic projection, pleasant nasolabial angle, passive lip sealing or discreet interlabial space, harmonious lip-menton curvature, well-defined chin-neck line and angle. Pattern II is defined by a positive sagittal step between the maxilla and the mandible, resulting from mandibular deficiency and/or maxillary dental protrusion, denouncing an excessive facial convexity. Conversely, Pattern III presents a negative sagittal step between the maxilla and the mandible due to mandibular prognathism and/or maxillary deficiency, with reduced facial convexity.

In the frontal view, the face was classified into three morphological types: dolichofacial, mesofacial and brachyfacial. In the dolichofacial configuration, the vertical dimensions of the face prevail over the horizontal dimensions, giving a longer silhouette. In the brachyfacial configuration, the transverse dimensions surpass the vertical dimensions, making up a wider face. The equivalence of the transversal and vertical dimensions characterizes the face as mesofacial. The three configurations described must exhibit characteristics such as symmetry, proportionality between the facial thirds and passive lip sealing with compatibility between the length of the soft lip and the length of the lower third of the face.

The Long Face and Short Face Patterns are discrepancies shown in the vertical direction, and comprise the extrapolation of the variation of normality of the face in the frontal view. The Long Face Pattern portrays a specific characteristic: the lips do not touch, the patient has exposure of the maxillary anterior teeth and gingiva at rest and smiling caused by the excess of the lower third of the face, including the maxilla (CAPELOZZA FILHO, 2005). In the present study, patients analyzed as dolichofacial include the "Long Face" and within brachyfacial patients, the "Short Face" is found.

The analysis of the facial morphology at rest was recorded using extraoral photographs of children with Microcephaly selected in forms specially developed for this purpose, as well as the registration data, the stage of the deciduous dentition and the facial characteristics of these patients. Reproducibility was assessed using intra-examiner agreement, performed using the Kappa test. Two researchers formed in orthodontics were trained, calibrated and a pilot study with 5 medical records was performed. Data collection started after an "almost perfect" interexaminer agreement (Kappa > 0.80).

The data were analyzed using the Statistical Package for the Social Sciences SPSS software version 21.0 (SPSS Inc., Chicago, USA). Descriptive statistics were calculated using the measure of central tendency as well as the frequency of sex and microcephaly classification. The inferential analysis between the type of Microcephaly and the independent variables (clinical factors) were determined by the Fischer Exact test, with a significance level of 5%.

RESULTS AND DISCUSSION

Thirteen medical records of children diagnosed with microcephaly were identified in the UESB Dentistry Module archive during the study period. Among these records, it was found that one child had a diagnosis of velo-cardio-facial syndrome (n=1), two had partial eruptions of the first permanent molars (n=2), one had incomplete deciduous dentition (n=1) and two was not within the age proposed in this study (n=2). Thus, 7 children were analyzed, between 3 and 5 years old (mean

age = 3.6 ± 0.22), 3 (42.8%) male and 4 (57.2%) female, brown color, in the stage of complete deciduous dentition, since 2 (28.5%) children diagnosed with microcephaly and 5 (71.5%) with severe microcephaly according to the WHO classification (Table 1).

Table 1 - Data of children selected according to age, sex, color/race (IBGE), denture stage and Microcephaly classification by WHO. Jequié, Bahia, Brazil, 2020

Case	Age	Sex	Cor/ Raça	Denture stage	Microcephaly Classif. (HC/SD)
1	3y 8m	Female	Brown	CD	Severe microcephaly (-3SD)
2	4y 1m	Male	Brown	CD	Severe microcephaly (-3SD)
3	3y 7m	Male	Brown	CD	Severe microcephaly (-3SD)
4	3y 5m	Female	Brown	CD	Microcephaly (-2SD)
5	3y 4m	Female	Brown	CD	Microcephaly (-2SD)
6	3y 6m	Female	Brown	CD	Severe microcephaly (-3SD)
7	3y 6m	Male	Brown	CD	Severe microcephaly (-3SD)

Notes: HC – head circumference; SD – standard deviation; CD – complete deciduous.
Source: the authors.

After the facial morphological analysis at rest, frontal view, it was observed that all children had a disproportion of the upper facial third in relation to the other thirds due to the significant reduction of the occipitofrontal head circumference. In addition to this narrowing of the head circumference, 6 children had a flat forehead (Figure 1A and 1B) and one child with frontal prominence (Figure 2A and 2B).

The seven children were classified, according to the frontal facial type, into brachyfacial, mesofacial or dolichofacial. In this study, all children were classified as dolichofacial (Table 2) (Figure 3). Regarding sagittal patterns, one was considered Pattern I (14.29%) (Figure 4) and six Pattern II (85.71%) (Figure 3) (Table 2).
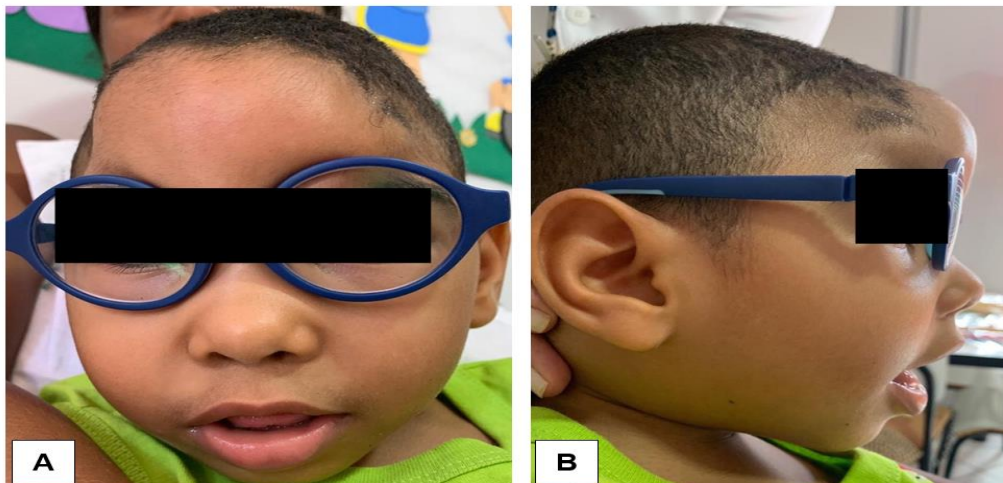
Figure 1 - Facial photographs of a patient at 3 years and 6 months of age with severe microcephaly with a disproportion between the facial thirds

Notes: a narrow and flat upper third (A and B) is observed.
Source: the authors.

Figure 2 - Facial photographs of a patient at 3 years and 7 months of age with severe microcephaly without equivalent facial thirds

Notes: there is a narrow upper third with frontal prominence (A and B).
Source: the authors.

Figure 3 - Facial photographs of a patient with severe microcephaly at 3 years and 6 months of age. Pattern II, dolichofacial (A and B)

Source: the authors.

Figure 4 - Facial photographs of a patient with microcephaly at 3 years and 5 months of age. Pattern I, dolichofacial (A and B)

Source: the authors.

Table 2 - Data of children selected according to age, sex, morphological characteristics of facial pattern and type and microcephaly classification by WHO. Jequié, Bahia, Brazil, 2020

| Case	Age	Sex	Facial Type	Facial Pattern	Microcephaly Classif. (HC/SD)
1	3y 8m	Female	Pattern II	D	Severe microcephaly (-3SD)
2	4y 1m	Male	Pattern II	D	Severe microcephaly (-3SD)
3	3y 7m	Male	Pattern II	D	Severe microcephaly (-3SD)
4	3y 5m	Female	Pattern I	D	Microcephaly (-2SD)
5	3y 4m	Female	Pattern II	D	Microcephaly (-2SD)
6	3y 6m	Female	Pattern II	D	Severe microcephaly (-3SD)
7	3y 6m	Male	Pattern II	D	Severe microcephaly (-3SD)

Notes: HC- head circumference; SD- standard deviation; D - Dolichofacial
Source: the authors.

The analysis of the chin-neck line and angle was not possible because they were not defined in most facial photographs of the selected individuals.

Fisher's Exact statistical test was applied to assess possible associations between the clinical parameters (gender, facial type, facial pattern) according to the microcephaly classification by WHO (Table 2). By applying Fisher's Exact test, it was shown that the proportion of severe Microcephaly (-3 SD) was higher in males (p= 0.04) (Figure 5). There was no association between the severity of microcephaly and the facial type (p> 0.05), although Pattern II was more prevalent in severe microcephaly (100% of the cases).

Figure 5 - Frequency of microcephaly severity by sex
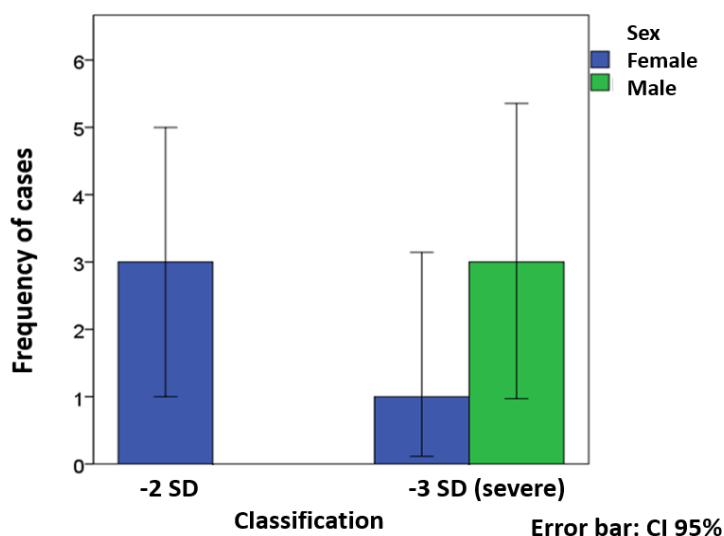
Source: the authors.

Several studies point out that although there are increases in craniofacial growth after early childhood, especially in the vertical direction and in the mandible, deviations in the growth pattern can already be identified and refer us to interceptive protocols in an attempt to reestablish a favorable morphological environment for adequacy of craniofacial growth (ENLOW; HANS, 1998; SILVA FILHO *et al.*, 2008; NIEMI *et al.*, 2019). In accordance with these principles, in this study, the facial morphology of patients with an average age of 3 years and 6 months (SD $=\pm0.22$) was analyzed using photographs, as it proved to be the most viable and ethical method for defining facial characteristics of children with microcephaly with neuropsychomotor impairment.

The most important limitations of this study can be represented by the small number of children studied, the absence of previous studies and the quality of the facial photographs. Due to the neurological impairment of the children with microcephaly studied, it was not possible to obtain dental occlusion at the time the photographs were taken. The resting position was associated with relaxation of the orofacial muscles, mandibular clockwise rotation and atypical tongue interposition.

In relation to sex dimorphism, in the present study, it was observed notably in the severity of microcephaly, where there was a greater manifestation of severe Microcephaly (-3 SD) in males. Although 100% of severe microcephaly cases had a predominance of Pattern II, there was no association between the severity of microcephaly and the facial type (p> 0.05). This can be explained by the small sample size, which compromised the power to detect differences between these variables. It is worth mentioning that, although in this study the characterization of the small number of the studied population was a significant limiting variable, the alert for the longitudinal monitoring of the mandibular growth vector of these patients, which is significant for the diagnosis and prognosis of craniofacial growth and development, is essential (PEREIRA *et al.*, 2017; MARQUES *et al.*, 2018). Marques *et al.* (2018), when analyzing the stomatognathic system functions of babies with microcephaly, suggest future studies that seek to describe the orofacial functional state of microcephalic patients due to their importance in understanding the craniofacial growth of children with microcephaly.

Previous epidemiological surveys on the facial morphology of Brazilian children without craniofacial anomalies and/or syndromes, in the deciduous dentition, found that in almost two thirds of the children evaluated, there was a predominance of 69.9% (TRALDI *et al.*, 2015) and 63.22% (SILVA FILHO *et al.*, 2008) of Pattern I in relation to Patterns II and III. In the present study, with a methodology similar to that of previous research, the results obtained in the sagittal analysis of the face of children with microcephaly revealed a predominance of Pattern II. The morphological definition of Pattern II in 85.71% of children with microcephaly in this study configured disharmony in the sagittal relationship between the apical bases, with a positive sagittal step between the maxilla and the mandible, showing an excessive face convexity. The maxilla and/or the maxillary incisors were projected forward in the sagittal direction, with clockwise mandibular rotation. These facial characteristics were also assessed in a comparative study among children in the deciduous dentition with/without a history of night snoring and the absence/presence of passive lip sealing, where they observed a statically significant increase in facial convexity in mouth breathing children (NIEMI *et al.*, 2019). This correlation of facial pattern with orofunctional disorder highlights the need for further investigations in children with microcephaly as Marques *et al.* (2018) analyzed the oral functional characteristics of 26 children aged 12, 15 and 18 months of age, diagnosed with microcephaly, with no association of any disorder or congenital malformation. Patients showed changes in facial muscle tone, breathing and swallowing.

In the present study, the study population had no diagnosis of the syndrome. In the frontal norm analysis of this study, the vertical dimensions of the face prevailed over the horizontal dimensions, classifying all children as dolichofacial. There was an absence of passive lip sealing with exposure of the maxillary anterior teeth and tongue interposition between the anterior teeth, dazzling a possible predisposition for the long face, evidenced by the increase in the lower third of the face in relation to the others. However, Silva Filho *et al.* (2008), in an epidemiological survey with 2009 Brazilian children in early childhood, verified that, regarding the frontal characteristic of the face, the mesofacial type was predominant (64.56%) in relation to dolichofacial (21.90%) and brachyfacial (13.54%). The authors, when also evaluating the distribution of frontal facial types within the sagittal facial patterns, showed that the dolichofacial type tends to manifest more frequently in Pattern II. In the present study, there was an agreement on this clinical finding in 85.71% of children with microcephaly.

The greater the change in orofacial structures, the more compromised the therapeutic evolution of the child with microcephaly can become, given the facial morphological limitations. It is worth emphasizing the importance of early investigation of facial growth and development in the sagittal and vertical direction of these individuals.

CONCLUSION

According to the results obtained in the present study, the characterization of facial Pattern II with a predominance of vertical growth shows a disproportion of the apical bases, maxilla and mandible. Additional studies with a larger number of samples and functional evaluation of breathing, swallowing and chewing are necessary to investigate the craniofacial morphology in order to provide early diagnosis and treatment for these children.

REFERENCES

ARROYO, H. A. Microcefalia. **Medicina - Suplemento II**, v. 78, n. Supl. 2, p. 94-100, 2018.

BRASIL. MINISTÉRIO DA SAÚDE. **Orientações integradas de vigilância e atenção à saúde no âmbito da Emergência de Saúde Pública de Importância Nacional : procedimentos para o monitoramento das alterações no crescimento e desenvolvimento a partir da gestação até a primeira infância, relacionadas à infecção pelo vírus Zika e outras etiologias infeciosas dentro da capacidade operacional do SUS** [recurso eletrônico], 2017. Disponível em: http://bvsms.saude.gov.br/publicacoes/orientacoes_emergencia_gestacao_infancia_zika.pdf. Acesso em: 28 maio 2020.

CAPELOZZA FILHO, L. **Diagnóstico en ortodoncia**. Maringá: Dental Press Editora, 2005.

DEVAKUMAR, D. *et al*. Infectious causes of microcephaly: epidemiology, pathogenesis, diagnosis, and management. **The Lancet Infectious Diseases**, v. 18, n. 1, p. 1-13, 2018.

ENLOW, D.; HANS, M. **Noções básicas sobre crescimento facial**. São Paulo: Santos, 1998.

INSTITUTO BRASILEIRO DE GEOGRAFIA E ESTATÍSTICA. **Censo 2000**. p. 1-28, 2000. Disponível em: https://www.ibge.gov.br/censo/censomundo.shtm. Acesso em: 28 maio 2020.

INTERGROWTH-21st. **Sobre INTERGROWTH-21st** (Portuguese). Disponível em: https://intergrowth21.tghn.org/about/sobre-intergrowth-21st-portuguese/. Acesso em: 28 maio 2020.

LEITE, C. N.; VARELLIS, M. L. Z. Microcefalia E a Odontologia Brasileira. **Journal Health Núcleo de Pesquisa e Extensão em Política, Planejamento, Organização e Práticas em Saúde**, v. 1, n. 2, p. 297-304, 2016.

LEWIS, A.; ROCHE, A.; WAGNER, B. Puberal sourts in cranial base and mandible. Comparisons with individuals. **Angle Orthodontist**, v. 55, n. 1, p. 17-30, 1985.

MARQUES, R. S. *et al*. Achados clínicos faciais em bebês com microcefalia. **Odonto**, v. 25, n. 49, p. 17-27, 2018.

NIEMI, P. *et al*. Association between snoring and deciduous dental development and soft tissue profile in 3-year-old children. **American Journal of Orthodontics and Dentofacial Orthopedics**, v. 156, n. 6, p. 840-845, 2019.

PEREIRA, S. M. S. *et al*. Zika Vírus e o Futuro da Odontologia no Atendimento a Pacientes com Microcefalia. **Revista de Investigação Biomédica**, v. 9, n. 1, p. 58, 2017.

SILVA FILHO, O. G. Padrão facial do paciente portador de fissura pré-forame incisivo unilateral completa. **Revista Brasileira de Cirurgia**, v. 79, n. 4, p. 197-200, 1989.

SILVA FILHO, O. G. *et al*. Padrão facial na dentadura decídua: Estudo epidemiológico. **Revista Dental Press de Ortodontia e Ortopedia Facial**, v. 13, n. 4, p. 45-59, 2008.

TRALDI, A. *et al*. Evaluation of facial morphology and sagittal relationship between dental arches in primary and mixed dentition. **Dental Press Journal of Orthodontics**, v. 20, n. 4, p. 63-70, 2015.

VON ELM, E. *et al*. The Strengthening the Reporting of Observational Studies in Epidemiology (STROBE) Statement: Guidelines for reporting observational studies. **Urology Today International Journal**, v. 2, n. 2, p. 806, 2009.

WORLD HEALTH ORGANIZATION. **Screening, assessment and management of neonates and infants with complications associated with Zika virus exposure in utero Interim guidance**, 2016. Disponível em: http://www.who.int/csr/resources/publications/zika/assessment-infants/en/ Acesso em: 14 dez. 2019.