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Influence of congenital heart disease on the neuropsychomotor development of infants

Influência da cardiopatia congênita no desenvolvimento neuropsicomotor de lactentes

Influencia de la cardiopatía congénita en el desarrollo neuropsicomotor de los lactantes

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ABSTRACT | Congenital heart defects (CHD) are among the main causes of morbidity and mortality in infants who has this impairment may present delays in neuropsychomotor development (NPMD). This study assesses the influence of CHD on NPMD of infants. This is an observational study assessing neuropsychomotor development performed by Bayley Scales of Infant and Toddler Development – BSID-III. The Brazilian Economic Classification Criteria was used to verify the socioeconomic status of the families and also the maternal and infants' clinical conditions were verified in the medical discharge report and in the child's health handbook. For the association between the quantitative and qualitative variables with the NPMD, the Spearman's correlation coefficient and the likelihood ratio test were used. A total of 18 infants were assessed, with a predominance of females (72.2%). Most mothers (47.1%) had complete high school or incomplete higher education, with a mean age of 27.2±5.5 years. There was a correlation between the BSID-III scales and the quantitative variables analyzed: motor scale with weight (p=0.02 and r=0.54) and oxygen therapy (p=0.009 and r=−0.591); besides that, the qualitative variables correlation were: motor scale and socioeconomic condition (p=0.015), motor scale and Interatrial Communication – IAC (p=0.023) and language with IAC scales (p=0.038). CHD influences the delay of NPMD, mainly for motor aspect. Furthermore, weight, diagnosis of IAC, use of oxygen therapy and socioeconomic status were considered the main risk factors for the delay in NPMD.

Keywords | Heart Defects, Congenital; Child Development; Infant.

RESUMO | As cardiopatias congênitas (CC) estão entre as principais causas de morbimortalidade na primeira infância e os lactentes com essa condição podem apresentar atrasos no desenvolvimento neuropsicomotor (DNPM). O objetivo deste estudo foi avaliar a influência da CC no DNPM de lactentes. Trata-se de um estudo observacional com avaliação do desenvolvimento neuropsicomotor realizada pela Bayley Scales of Infant and Toddler Development (BSID-III). As condições maternas e clínicas dos lactentes foram verificadas no relatório de alta médica e na caderneta de saúde da criança, e a condição socioeconômica das famílias pelo Critério da Classificação Econômica Brasil. Para associar as variáveis clínicas e o DNPM foram utilizados o coeficiente de correlação de Spearman e o teste de razão de verossimilhança. Foram avaliados 18 lactentes, com predomínio do sexo feminino (72,2%). A maioria das mães (47,1%) possuía ensino médio completo ou

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superior incompleto, com média da idade de 27,2±5,5 anos. Houve correlação das escalas do BSID-III com as variáveis quantitativas analisadas: escala motora com o peso (p=0,02 e r=0,54) e com uso de oxigenoterapia (p=0,009 e r=−0,591); já para as variáveis qualitativas as associações foram entre: escala motora e condição socioeconômica (p=0,015), escala motora e comunicação interatrial – (CIA) (p=0,023) e escala da linguagem e CIA (p=0,038). A CC influenciou o DNPM, principalmente no aspecto motor. Além disso peso, diagnóstico de CIA, uso de oxigenoterapia e condição socioeconômica foram considerados como principais fatores de risco para o atraso no DNPM.
Descritores | Cardiopatias Congênitas; Desenvolvimento Infantil; Lactente.

RESUMEN | Las cardiopatías congénitas (CC) se encuentran entre las principales causas de morbimortalidad en la primera infancia, y los lactantes con esta afección pueden tener retrasos en el desarrollo neuropsicomotor (DNPM). El presente estudio tuvo el objetivo de evaluar la influencia de las CC en el DNPM de los lactantes. Este es un estudio observacional en el cual se evaluó el desarrollo neuropsicomotor utilizando la Bayley scales of infant and toddler development (BSID-III). Las condiciones maternas y clínicas de los lactantes se obtuvieron en el informe de alta médica y en la libreta de salud del niño, y el estado socioeconómico de las familias en el Criterio de Clasificación Económica de Brasil. Para asociar las variables clínicas y el DNPM, se utilizaron el coeficiente de correlación de Spearman y la prueba de razón de probabilidad. Se evaluaron a 18 lactantes, con un predominio del sexo femenino (72,2%). La mayoría de las madres (47,1%) tenían la secundaria completa o la educación superior incompleta, con una edad promedio de 27,2±5,5 años. Hubo una correlación entre las escalas BSID-III y las variables cuantitativas analizadas: escala motora con el peso (p=0,02 y r=0,54) y con el uso de oxigenoterapia (p=0,009 y r=−0,591); para las variables cualitativas, las asociaciones fueron entre: escala motora y estado socioeconómico (p=0,015), escala motora y comunicación interauricular (CIA) (p=0,023) y escala de lenguaje y CIA (p=0,038). Las CC influyeron en el DNPM, principalmente en el aspecto motor. Además, el peso, el diagnóstico de CIA, el uso de oxigenoterapia y el estado socioeconómico fueron considerados los principales factores de riesgo para el retraso en el DNPM.
Palabras clave | Cardiopatías Congénitas; Desarrollo Infantil; Lactante.

INTRODUCTION

Congenital malformations are among the main causes of mortality in early childhood, and congenital heart defect (CHD) represents 40% of them[1]. The incidence of CHD ranges around eight in every thousand live births, according to the World Health Organization (WHO)[1], and these defects are often associated with pediatric emergency demands due to needs for hospital admissions and surgical procedures[2,3].

Previous studies have shown how much CHD can affect motor[3,4,5], cognitive and language development of infants[5]. It is known that neuropsychomotor development (NPMD) can be influenced from biological, psychological, social and environmental factors[6]. However, surgical interventions, especially when performed in the first year of life, and prolonged hospital admissions significantly affect cognitive and motor development, with repercussions in early childhood and even in adult life.

Considering that children affected by a heath defect need differentiated treatment, which often includes prolonged hospital admissions, it is necessary to assess possible changes in NPMD, as well as to recognize possible risk variables in order to intervene as early as possible.

Thus, this study investigates the influence of CHD on NMD of infants.

METHODOLOGY

This is a cross-sectional observational study carried out between November 2017 and February 2018. The infants assessed were assisted in the outpatient clinic of child cardiology of the Hospital de Clínicas de Uberlândia of the Universidade Federal de Uberlândia/MG (HCU-UFU). Legal guardians, after reading and understanding the study, signed the free and informed consent form. At the end of the infant's assessment, all families were instructed about the possible sensory-motor stimulation that could be made in the period to reduce neuropsychomotor delay. Infants with significant delay in DNPM were referred for the institution specific care.

Participants

Those infants diagnosed with cyanotic and non-cyanotic CHD, aged between one and 18 months, who were under follow-up at the HCU-UFU outpatient clinic were included in this study.

The exclusion criteria adopted were: infants who did not meet all five scales of the neuropsychomotor development assessment instrument Bayley Scales of Infant and Toddler Development, Third Edition (BSID-III), infants who had some acute pathological process on the day of test and those who possessed some associated genetic disease.

Sampling calculation

Normative scores based on population studies with a mean score of 100 points and standard deviation of 15 points for each domain of the BSID-III scale[7] were considered. It was also considered that the target population would be 123 infants (total number of patients with CHD under follow-up in the HCU-UFU outpatient clinic), with a seven points as margin of error in the questionnaire score and 95% confidence level. Thus, the minimum sample size, according to the methodology suggested by Fonseca and Martins[8], was 16 participants.

Data collection instruments

Bayley Scales of Infant and Toddler Development, Third Edition

The BSID-III is an instrument considered as gold standard that is widely used to assess the development of children between one month and 42 months of age[7]. It has been translated and adapted to Portuguese[9] and it is in process validation in Brazil. It includes all aspects of NPMD with accurate data and excellent reliability standard. The test is subdivided into five domains: (1) cognitive, with 91 items; (2) language, which is divided into two subtests (receptive communication, with 49 items and expressive communication, with 48 items; (3) motor (subdivided into gross motor ability, with 72 items and fine motor ability, with 66 items); (4) social-emotional; and (5) adaptive behavior, the latter two being obtained from filling the scales by caregivers or parents of the child[7,9].

Compound scores follow the patterns of population-normative studies that found an average of 100 points and standard deviation of 15 points for cognitive, language, global motor, social-emotional and adaptive behavior scales; that is, scoring 15 points below average represents that the child has a slight delay, and two standard deviations (30 points) below average, relevant delay. The receptive communication, expressive communication, fine motor and gross motor scales were assessed with balanced score, in which a standard deviation (three points) below average represents slight delay, and two standard deviations (six points), represents relevant delay.

Brazilian Economic Classification Criteria Scale

The Brazilian Economic Classification Criteria (CCEB—*Critério de Classificação Econômica Brasil*) is used to categorize households and their residents into social classes, according to the Brazilian Association of Research Companies (ABEP—*Associação Brasileira das Empresas de Pesquisa*), based on the socioeconomic survey conducted by the Brazilian Institute of Public Opinion and Statistics (IBOPE—*Instituto Brasileiro de Opinião Pública e Estatística*) in 2015. The method establishes the number of points that a household receives by presenting a particular good or service and their weight according to the number of possessed goods and services. The classification is made according to the six classes (A, B1, B2, C1, C2 and ED), added to the points of a given domicile[10].

Procedures

The patients' legal guardians were approached while they waited for routine appointment with the cardiologist to invite and to clarify about the application of BSID-III and questionnaires. The evaluation occurred so it did not hinder the pre-established order of care.

Initially, information about maternal and infant clinical data was collected by the hospital discharge report and the child's health handbook.

The assessment of BSID-III occurred while the guardians were answering the adaptive behavior scale, the socio-emotional scale and the socioeconomic questionnaire (CCEB).

Data were collected individually in a room on the same sector for about 90 minutes. The infant's fatigue and stress were respected with pauses during the evaluation, and rescheduling was not necessary.

Data were collected by two trained and prepared researchers, with the technical rigor required by the

scale. Data interpretation was performed and conferred by two other researchers who did not participate in the application of the instrument.

Data analysis

The Shapiro-Wilk test was performed, and non-normality of the data was observed, so non-parametric tests were used. Spearman's correlation coefficient[11] were used to evaluate the association between quantitative variables with NPMD. The associations of qualitative variables with the NPMD were analyzed using the likelihood ratio test.

All tests were used with a 5% significance level. The procedures were performed using the SPSS software, version 20.

RESULTS

A total of 20 infants were assessed and two were excluded, one for not fulfilling all BSID-III scales and the other for being very sleepy at the time of data collection.

The mother was characterized as primary caregiver for all infants. As for these, 10 had been hospitalized at birth, most of them (22.2%) due to respiratory distress; six were readmitted, four for cardiac surgery, one for pneumonia and one for respiratory distress and bronchitis; five required some assisted ventilation; 10 infants required oxygen therapy; three required physical therapeutic care; and two of them needed speech therapy. Every infant had Apgar score higher than six in the fifth minute of life. Other maternal and infant clinical characteristics are found in Table 1

Table 1. Maternal and infant characteristics of the selected sample (n=18)

Maternal characteristics	n (%)	Mean (SD)
Age (years)		27.2±5.5
Schooling (n=17)*		
Illiterate/incomplete elementary school	0 (0%)	
Elementary school	3 (17.6%)	
Incomplete high school	2 (11.8%)	
High School/incomplete higher education	8 (47.1%)	
Higher education	4 (23.5%)	
Neonatal characteristics		
Sex		
Male	5 (27.8%)	
Female	13 (72.2%)	
Type of birth		
Vaginal birth	3 (16.7%)	
Caesarean section	15 (83.3%)	
Prematurity	3 (16.7%)	
Chronological age (months)		8.5±5.1
Corrected age (months)		8.3±5
Weight (grams)		2,770.5±667.2
Hospitalization at birth (days)		12.3±20.4
Oxygen therapy (hours)		182.8±475.9
Assisted ventilation (days)		9.4±35.2
Readmission (days)		12.6±26.5
Type of heart defect		
Cyanotic	4 (22.2%)	
Non-cyanotic	14 (77.8%)	

*Total sample for schooling.

The result obtained in each of the BSID-III scales was organized into means with their respective standard deviations. Out of all neuropsychomotor development skills assessed, the only one that was interpreted as discrete delay was the general motor ability (M=82.06; SD=21.77) by composite score, and fine motor ability (M=7.72; SD=3.51) and gross motor (M=6.28; SD=4.52) by balanced score.

Table 2. Scores obtained by applying the Bayley III scale

	Mean	Standard Deviation
Cognitive*	91.11	18.20
Language*	91.28	23.29
Receptive language**	9.33	4.45
Expressive language**	7.78	4.21
General motor*	**82.06**	21.77
Fine motor**	**7.72**	3.51
Gross motor**	**6.28**	4.52
Social-emotional*	95.56	19.39
Adaptive behavior*	90.11	20.72

*Composite score. **Balanced score.

Each NPMD ability evaluated by BSID-III was correlated with maternal sociodemographic and infant variables to identify possible risk factors for delay in NPMD of the cardiopathic infant.

The motor scale presented a statistically significant correlation with birth weight and oxygen therapy time.

A statistically significant correlation was also observed between the motor scale and socioeconomic status and diagnosis of Interatrial communication (IAC) heart defect; and the scale of language with the diagnosis of IAC heart defect.

Table 3. Association between items of the Bayley-III scale (composite score) with the quantitative characteristics of the selected sample (n=18)

	Cognition	Language*	General motor	Social-emotional	Adaptive behavior
Chronological age (months)					
Correlation	−0.124	−0.102	0.155	−0.453	−0.199
p value	0.6238	0.6858	0.5398	0.0591	0.4292
Corrected age (months)					
Correlation	−0.100	−0.151	0.122	−0.428	−0.253
p value	0.6932	0.5500	0.63	0.0767	0.3103
Mother age					
Correlation	0.308	0.105	0.32	0.155	0.154
p value	0.2293	0.6886	0.20	0.5521	0.5544
Weight (grams)					
Correlation	0.289	0.096	0.54	−0.268	−0.116
p value	0.2453	0.7049	***0.02**	0.2828	0.6455
Hospitalization at birth (days)					
Correlation	−0.449	0.023	−0.09	−0.066	0.025
p value	0.0615	0.9273	0.7021	0.7949	0.9212
Assisted ventilation (days)					
Correlation	0.120	0.141	−0.129	0.260	0.208
p value	0.6347	0.5781	0.6108	0.2977	0.4075
Oxygen therapy (hours)					
Correlation	−0.343	−0.309	−0.591	−0.210	−0.277
p value	0.1634	0.2115	***0.0097**	0.4027	0.2663
Readmission (days)					
Correlation	0.130	0.094	−0.192	0.217	0.270
p value	0.6202	0.7200	0.4610	0.4019	0.2942

*Correlation with p<0.05; likelihood ratio test (Test G).

Table 4. Association between the items of the Bayley-III scale (composite score) with the qualitative characteristics of the selected sample (n=18)

		Cognition	Language*	General motor	Social-emotional	Adaptive behavior
Sex	p value	0.537	0.152	0.504	0.081	0.619
Mother's education level	p value	0.464	0.483	0.136	0.719	0.734
Type of birth	p value	0.436	0.799	0.417	0.137	0.103
Type of heart defect	p value	0.113	0.224	0.281	0.335	0.319
Other comorbidity*	p value	0.060	0.274	0.759	0.117	0.206
Physical therapy	p value	0.694	0.510	0.578	0.988	0.229
Speech therapy	p value	0.051	0.478	0.443	0.913	0.632
ABEP	p value	0.257	0.662	**0.015**	0.594	0.451
Heart defect classification (CIA)	p value	0.607	**0.038**	**0.023**	0.093	0.981

*Presented among the comorbidities: renal agenesis, tracheostomy, autoimmune hemolytic anemia, neonatal hypothyroidism, intrauterine growth restriction, tachycardia, umbilical hernia; likelihood ratio test (Test G). IAC: Interatrial communication.

DISCUSSION

The main result of this study is in agreement with the previously published evidence, showing that heart disease negatively influences the NPMD of infants[12-16]. Different abilities were affected, including motor and language.

Our results showed that from all averages scored by infants on the BSID-III scale, the only one interpreted as a discrete delay was that obtained for general motor ability. Regarding the subtests, the averages scored in the gross and fine motor abilities also presented a slight delay by the balanced score. Polat et al.[17] found similar results when assessing children between one and 72 months old regarding gross and fine motor ability. Delay was observed in these abilities for children with heart defect when compared to the control group using the Denver II developmental screening test.

Miller et al.[16] associated NPMD delay with inadequate metabolism and brain maturation that children with heart defect may present, even before undergoing a surgery for heart disease correction . In contrast, Snookes et al[4], by a systematic review that covered articles assessing cognitive and motor aspects of child development after heart surgery during early childhood, stated that brain damage is one of the complications of long-term CHD.

Our study presented no significant association between the types of heart defect (cyanotic and non-cyanotic) and NPMD. Few studies carried out this comparison. Williams et al.[13] showed more delay in the cognitive and language aspects of infants with cyanotic CHD when they assessed, with the BSID-III, 18-month-old infants with heart defect, besides observing lower intrauterine growth rate in them.

A significant association between the diagnosis of IAC heart disease and delays in language acquisition was observed. From the 18 participants, eight presented IAC, and half of them were delayed in language, so that the greatest delay was observed in expressive communication. Other authors also found delays in the language mastery of infants with CHD[5,17].

The diagnosis of IAC, researched alone, was also related to general motor ability (p=0.023). Half of the infants with this diagnosis presented significant delay (score less than or equal to 69). Few studies have sought to assess the diagnosis of IAC with child development, perhaps because the diagnosis is not often exclusive to IAC, or because this studies consider that each cardiology center presents patients' profiles according to resources and treatments offered at the place.

It was also observed that weight showed a positive correlation with general motor ability. Edwards et al.[18] also reported a high prevalence of changes in motor coordination in infants born with very low weight/premature birth, and these changes were observed until school age and adolescence. The same occurs in fine motor subtest, as observed by Cahill-Rowley and Rose[19], in which infants with very low birth weight (<1,500 g and <32 weeks) showed worse results in fine motor ability compared to typical infants.

A negative association between the oxygen therapy and its time of use was observed with the general motor development of the evaluated children with CHD, i.e., the longer the time of oxygen therapy, the lower the score in general motor ability. Newborns with bronchopulmonary dysplasia usually present compromised weight-height development, since they have low nutritional input and higher energy needs[20], which can compromise NPMD.

Although factors related to hospital care influence NPMD[21], our study presented no correlation between delay in NPMD with number of days of hospitalization at birth, use of assisted ventilation and readmission.

Regarding maternal and socio-environmental factors, only the socioeconomic aspect (p=0.015) interfered in the NPMD. The results show that the lower the socioeconomic status, the lower the score obtained by infants in motor ability. Defilipo et al.[22] found similar results regarding socioeconomic factor when they assessed 239 typical infants between three and 18 months old. Possibly this association is due to the lower opportunities of motor stimuli of lower-income families (limited physical space and less resources, such as toys).

CONCLUSIONS

Congenital heart defect compromises the NPMD of infants up to 18 months old, especially in general, fine and gross motor skills. Furthermore, birth weight, IAC diagnosis, use of oxygen therapy and socioeconomic status may also influence the development of these infants.

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