

ORIGINAL ARTICLE

# Description of the linguistic and neurological findings of twins born preterm at two years of age

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**Abstract**

**Introduction:** Prematurity, low birth weight and multiple births are risk factors for structural changes in the brain, as well as for motor, cognitive, social and language developmental disorders.

**Objective:** Describe the linguistic findings and investigate the presence of neurological alterations in preterm twins at two years of age.

**Methods:** This is a cross-sectional study based on data analysis of the medical records of six pairs of low birth weight preterm twins of both sexes, at two years of age, attended at the State Center for the Prevention and Rehabilitation of People with Disabilities - CEPRED, in Salvador-Bahia-Brazil. To this end, information regarding birth; the clinical history; the imaging exams; and the speech, neurological and interdisciplinary assessments were collected.

**Results:** Preterm twins had atypical language development and neurological changes that may compromise oral language development. From the neurological point of view, abnormalities common to the premature newborn, such as peri-intraventricular hemorrhage, were observed.

**Conclusion:** Multiple births associated with prematurity and low birth weight may pose risks to children's language acquisition.

**Keywords:** language development, language development disorders, neurodevelopmental disorders, premature birth, twins.

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## Authors summary

### Why was this study done?

The study was designed to investigate language and neurological changes in preterm twins.

### What did the researchers do and find?

The researchers analyzed clinical data on preterm twins at two years of age and found atypical language development in all children in the sample, even in the absence of central nervous system lesions, extreme low birth weight and extreme preterm birth.

### What do these findings mean?

The presence of language alterations in preterm twin children with or without central nervous system integrity warns of the need for early interventions in order to prevent and/or minimize deleterious effects on child development.

## INTRODUCTION

Prematurity, low birth weight and multiple births are factors that make the brain susceptible to structural changes and increase the chance of deviations in motor, cognitive, social and language development. It is known that the prevalence of prematurity worldwide is around 7.2% and more than 15 million babies are born prematurely each year<sup>1</sup>. Brazil is in tenth position among the countries where most are born premature<sup>2</sup>. There is an increase in multiple pregnancies in Brazil and their occurrence is also associated with high prematurity rates and high incidence of low birth weight (LBW) newborns (NB)<sup>3</sup>.

Preterm newborns (PTNB) have been delayed in development compared to term newborns. In this sense, twinning represents a greater neonatal risk to maternal and child health, due to the association with LBW and other complications arising from prematurity. There is an increased risk of death and neurological disorders in twins, regardless of the influence of prematurity and birth weight, as they are also susceptible to intrauterine growth restriction and neonatal complications<sup>4</sup>.

Research on twin language development has shown how much language acquisition can be altered due to biological and interactional aspects. It is known that most twin children have pre, peri and postnatal complications. Thus, there is a high rate of perinatal mortality in twins, significantly influenced by low birth weight (LBW) and prematurity, often associated with respiratory, metabolic, immunological and neurological alterations<sup>5,6</sup>, in addition to lower Apgar scores in this population<sup>7</sup>.

Premature birth puts the infant brain at increased risk of gray matter and white matter damage, and even in the absence of injury, brain development can be altered with significant structural and microstructural changes that are associated with neurodevelopmental disabilities. The temporal lobe and adjacent regions, which are centers of language development, are particularly vulnerable<sup>8</sup>.

In general, twin pregnancy is associated with preterm birth and risks such as fetal mortality, early perinatal and neonatal mortality, LBW and stay in a Neonatal Intensive Care Unit (NICU)<sup>9</sup>. Twin studies have investigated the importance of factors such as genetic load and environmental influence. Clinical, pathological, and neuroimaging data suggest that preterm infants are exceptionally vulnerable to language center injury during the critical period of brain development and organization<sup>10</sup>.

Thus, the preterm twin's brain remains vulnerable to injuries and morbidities that may occur during prolonged hospitalization<sup>11,12</sup>. Twins may have a typical development,

but many of these children show a delay in oral language acquisition as an effective form of communication. In twin children, complications occurring during childbirth and early childhood are considered as risk factors for child development<sup>13</sup>. Thus, they may later develop cognitive, linguistic or behavioral changes and cannot completely exclude later adverse neurological outcomes<sup>14,15</sup>.

Language acquisition depends on neurobiological and social factors, which include the integrity of brain structures, proper cognitive function, social interaction, and the stimuli received in the child's environment. Deviations in any of these aspects may affect language development<sup>16-18</sup>, hence the importance of studies that evaluate language acquisition of twins in early childhood. Given the need to investigate language acquisition in cases of multiple pregnancies associated with prematurity, the aim of the present study was to describe the linguistic findings and investigate the presence of neurological changes in preterm twins.

## METHODS

This is a cross-sectional study with a sample composed of six pairs of preterm twins (12 children), of both sexes, evaluated at two years of age, who met the following inclusion criteria: records of speech therapy and interdisciplinary care in medical records; presence of imaging results and neurological reports; information regarding birth, gestational age, birth weight and any complications. Excluded from this study were children whose information above was not in the medical records and the pairs in which children were diagnosed with Down Syndrome (DS) and autistic spectrum disorder (ASD).

This study was approved by the Research Ethics Committee of the Secretariat of Health of the State of Bahia (SESAB), under opinion 310.813/13 and amendment 1.952.793/17. Followed the ethical guidelines of research with human beings, according to CNS Resolution 466/12.

The study was conducted through analysis of medical records data at the State Center for the Prevention and Rehabilitation of People with Disabilities - CEPRED, a reference unit of the Unified Health System (SUS) in the state of Bahia. All institutional records selected had the consent of users and/or legal guardians (under 18 years old) to use their data in scientific research.

Data regarding speech, neurological and interdisciplinary assessment were analyzed. For this, we collected information regarding birth, such as gestational age (GA), birth weight, sex and Apgar; the clinical history;

the results of imaging exams; and the basic assessments of the interdisciplinary team, checking the pre, peri and postnatal complications of the children. Among the total number of users assisted in the early intervention sector, from 2014 to 2018, the medical records of all twin assisted during this period were analyzed. From this, the above inclusion criteria were applied. Data analysis was performed descriptively and presented through tables.

## RESULTS

The sample consisted of six pairs of preterm newborns with LBW of both sexes. For the analysis of linguistic and neurological development we considered the evaluation performed at two years of age. Two pairs of twins were excluded from this study due to the occurrence of other associated conditions, such as DS and ASD. Birth weight ranged from 1158g to 2298g, with a median of 1,623.5g; and GA ranged from 29 to 36 weeks, with a median of 32.5 weeks (Table 1).

**Table 1:** Descriptive analysis of the variables birth weight and gestational age.

Variable	Median	1st quartile	3rd quartile	Maximum	Minimum
Birth weight (g)	1.623.5	1.269.8	2.005.0	2.298.0	1.158.0
GA (weeks)	32.5	31.8	34.0	36.0	29.0

Source: research data. Legend: g – grams; GA – gestational age.

For the analysis of socioeconomic aspects, maternal education and the monthly family income described in the medical record were considered. It was observed that 41.7% of the sample reported having 10 to 12 years of study and 25% of six to nine years of study. It is noteworthy that this information was absent in 33.3% of the sample. Regarding monthly income, 16.7% reported receiving a minimum wage, 5.5% from one to two minimum wages and 16.7% from two to three minimum wages (Table 2).

Regarding language development, 100% of children presented atypical performance, *ie*, incompatible with the expected for chronological age; 58.3% of the sample consisted of female children and 41.7% male; 41.7% had very low weight (VLW) and 58.3% LBW, no child was classified as extremely low weight (ELW) or with adequate birth weight. Regarding gestational age, 41.7% was classified as very preterm (VPT) and 58.3% as moderate/late preterm (MTM/LPT). There was no child classified as extreme preterm (EPT) in the sample (Table 3).

**Table 2:** Family socioeconomic data (n = 12).

Variable	n	%
Maternal education (in years)		
Not included *	4	33.3
1 to 5	0	0
6 to 9	3	25
10 to 12	5	41.7
13 to 18	0	0
More than 19	0	0
Monthly income		
Not include*	0	0
Less than 1 minimum wage **	0	0
1 minimum wage	2	16.7
1 to 2 minimum wages	8	5.5
2 to 3 minimum wages	2	16.7
3 to 4 minimum wages	0	0
More than 5 minimum wages	0	0

Source: research data.

\*Absence of information in the medical record

\*\* The minimum wage in force in 2018 was considered: R\$954.00.

According to the neurological report and the image exams contained in the medical records, 33.3% of the children did not present alterations, 16.6% grade I periventricular hemorrhage (PIVH), 8.33% grade II and grade III PIVH, and 33.3% PIVH grade IV associated with

**Table 3:** Frequency distribution of language development, gender, birth weight, gestational age, neurological diagnosis and zygosity variables.

Variables	Percentage (%) n=12
Language development	
Typical performance	0
Atypical performance	100
Sex	
Female	58.3
Male	41.7
Birth weight	
Extreme Low Weight	0.0
Very Low Weight	41.7
Low Weight	58.3
Gestational age classification	
Extreme preterm	0.0
Very preterm	41.7
Moderate/Late Preterm	58.3
Neurological diagnosis	
No changes	33.3
Grade I PIVH	16.6
Grade II PIVH	8.33
Grade III PIVH	8.33
Grade IV PIVH associated to PVL	33.3
Zygosity	
Monozygous	50
Dizygotic	50

Source: research data. Legend: PIVH – peri-intraventricular hemorrhage; LPV – periventricular leukomalacia.

periventricular leukomalacia (PVL). Regarding zygosity, 50% of the sample presented monozygotic and 50% dizygotic twinning (Table 3). There are clinical reports of neuropsychomotor developmental delay in all children. No difference in language performance was observed between the first and second twin.

## DISCUSSION

The preterm twins investigated at two years of age presented language development incompatible with that expected for their age group, considering the stages of language skills acquisition. Changes in oral language in this population may manifest as language delays or disorders, according to the presence or absence of associated pathologies<sup>19</sup>.

Another twin study at two years of age correlated performance in imitation, vocabulary, play and social behavior with aspects of the environment shared by twins, prompting reflection on the role of the family in promoting developmental stimuli. Significant genetic correlation between imitation and vocabulary was also evidenced, suggesting that different aspects of cognition are linked by sets of overlapping genes<sup>20</sup>.

When investigating the communicative skills of twins diagnosed with language delay, it was found that they had a lag in global and language development, highlighting the interference of genetic and environmental factors in child development<sup>21</sup>. For some children, these difficulties may persist throughout childhood and adolescence and thus have a significant impact on their achievement and quality of life<sup>22,23</sup>.

Preterm birth has been associated with a higher risk of language delays<sup>24</sup>. When assessing motor, adaptive, social and language behavior in triplets, there was a delay in all these domains, with language being the most affected domain<sup>25</sup>. The present study agrees with these findings, since deviations in the twin language acquisition were evidenced. Research examining language development in 310 twin pairs at the age of four found that individual differences in language development substantially overlap with genetic influences, underscoring the importance of investigating environmental factors that affect both language and language cognition<sup>26</sup>. In contrast, another study states that the effects of twinning at two years of age diminish from four to six years, suggesting that twins have delays in initial language acquisition, which can be overcome at later ages<sup>27</sup>.

There was no difference in language performance in relation to twin birth order and zygosity, since all children had atypical language development. Thus, it is necessary to investigate the possible effects of time of birth and zygosity on twin language acquisition in larger samples, as well as whether early effects persist in children over time. This finding agrees with another study<sup>28</sup> that also did not observe a significant difference between the first and second twin when evaluating them at school age.

The same occurred in a study that performed cognitive, language and motor development assessment<sup>29</sup>, thus agreeing with the present findings. In an extensive cohort it was observed that preterm twins underperformed language assessment at five years of age when compared to

full-term twins. However, no significant differences were noted regarding birth order<sup>30</sup>.

Another relevant aspect refers to the fact that all children in the sample had a delay in neuropsychomotor development, even without alterations diagnosed in neurological exams. Children with neuropsychomotor developmental delay have more difficulties to act independently during interactive and dialogical activities and, therefore, there are influences on language development<sup>25</sup>.

On the other hand, in another study<sup>28</sup>, the group of preterm twins had a performance similar to those of single-gestation preterm infants, suggesting that prematurity was a more important influencing factor on visuomotor and overall motor performance than twinning. Thus, concrete opportunities to make their repertoire viable are lost, generating gaps in the perceptive, cognitive, linguistic and social areas, with repercussions on the other dimensions of child development<sup>31</sup>. Early motor performance in childhood may be an indicator of brain connectivity, meaning maturity of developmental pathways between various brain regions<sup>32</sup>.

Regarding the interaction, peculiar characteristics are observed regarding the quantity and quality in the language of these children. In general, the twin brothers' experiences are differently experienced in interpersonal circumstances that do not favor linguistic interaction with other interlocutors, thus leading to an atypical process of language development<sup>25,33</sup>. Other studies show consistent evidence of the influence of genetic factors on language acquisition, with variations according to children's age<sup>34</sup>. Moreover, there is evidence that the close relationship between twin brothers is a factor that impedes the proper development of oral language<sup>35</sup>.

It was observed that a large number of children presented neurological alterations, such as PIVH. Brain abnormalities in preterm twins, including PIVH and PVL, are generally associated with concomitant neuronal/axonal changes, and may be accompanied by posthemorrhagic infarction, cerebellar injury, and posthemorrhagic hydrocephalus<sup>36</sup>. The risk for language disorders is associated with the risk of changes in motor and social development, ie delays that cause negative consequences for the child's adaptation and socialization process<sup>37</sup>. In addition, the presence of perinatal PIVH, especially in the most severe cases (grades III and IV) is associated with central and language hearing disorders<sup>38</sup>. The prognosis of PIVH varies according to the degree of injury. In grade I and II hemorrhages, the child often progresses without noticeable neurological abnormalities; grade III may evolve to ventriculomegaly or hydrocephalus, with incidence of cerebral palsy and intellectual disability; Grade IV children have high mortality, especially when large lesions occur in low gestational age infants<sup>39</sup>.

With regard to neurological development, detection of white matter volume abnormalities may be a significant predictor of cognitive and language development at two years of age<sup>40</sup>. A longitudinal study from 18 to 24 months and then at five years of age showed that the perinatal brain characteristics of preterm infants may influence later functional development. In addition, prematurity and

perinatal risk factors found in the sample had an impact on brain tissue volumes and their growth between birth and equivalent term age<sup>41</sup>. These findings emphasize the need for multiprofessional follow-up in order to observe the child's overall development and not in a fragmented or isolated area.

The children in the study sample presented other non-linguistic risk factors, such as socioeconomic and cultural factors, which may act together or in isolation, favoring such changes. Little is known about the real effects of low socioeconomic status on language development in preterm infants, but studies suggest that there is a negative effect and may increase the risk for speech-language disorders<sup>42</sup>.

This study highlights the importance of monitoring the development of preterm twins and early intervention. Thus, the systematic speech-language follow-up of these cases is of paramount importance to identify the demands of each child and to verify, if any, whether or not oral language changes persist. Studies with larger samples and longitudinal follow-up are recommended to verify the linguistic performance of twins after two years of age. It is also suggested that research be conducted to identify if language development in this population is more influenced by twins, prematurity or low birth weight.

The contribution of this study was to verify if oral language acquisition occurred typically in preterm twins, in an age group in which acquisition, in its primordial functions, is under development. In addition, it identified

which neurological changes exist in the investigated cases. This knowledge emphasizes the need for early intervention in cases where there is a risk for delays in child development. However, it is important to highlight some limitations of the study. The first is the absence of data from a control group under the same experimental conditions, allowing to compare the linguistic performance of children with their full-term peers and single gestation. The second is related to the sample size and study design, since the sample consists of six twin pairs and the data analyzed are secondary, although they were analyzed according to the same parameters.

## ■ CONCLUSION

The present study demonstrated that multiple births associated with prematurity and low birth weight may be related to language development disorders at two years of age. As all children had atypical language development, there was no difference in language performance regarding gestational age, birth weight, gender, birth order and zygosity. All children had neuropsychomotor developmental delay, even without central nervous system lesions. A large number of children with neurological disorders, such as PIVH.

There is a need to increase the number of national studies on this topic in order to investigate the risk factors for changes in oral language development and their implications for child development.

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## Resumo

**Introdução:** A prematuridade, o baixo peso ao nascer e os nascimentos múltiplos são fatores de risco para alterações estruturais no cérebro, assim como para desvios no desenvolvimento motor, cognitivo, social e de linguagem.

**Objetivo:** Descrever os achados linguísticos e investigar a presença de alterações neurológicas em gemelares nascidos pré-termo, aos dois anos de idade.

**Método:** Trata-se de um estudo do tipo corte transversal, realizado por meio da análise de dados dos prontuários de seis pares de gêmeos nascidos pré-termo, com baixo peso ao nascer, de ambos os sexos, aos dois anos de idade, atendidos no Centro Estadual de Prevenção e Reabilitação da Pessoa com Deficiência – CEPRED, em Salvador-Bahia-Brasil. Para tanto, foram coletadas as informações referentes ao nascimento; a história clínica; os exames de imagem; e as avaliações fonoaudiológica, neurológica e interdisciplinar.

**Resultados:** Os gemelares nascidos pré-termo apresentaram desenvolvimento linguístico atípico e alterações neurológicas que podem comprometer o desenvolvimento da linguagem oral. Do ponto de vista neurológico, foram observadas anormalidades comuns ao recém-nascido prematuro, como a hemorragia peri-intraventricular.

**Conclusão:** Os nascimentos múltiplos associados à prematuridade e ao baixo peso ao nascer podem oferecer riscos à aquisição da linguagem da criança.

**Palavras-chave:** desenvolvimento da linguagem, transtornos do desenvolvimento da linguagem, transtornos do neurodesenvolvimento, nascimento prematuro, gêmeos.

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