

# Habilidades do desenvolvimento em crianças com hipotireoidismo congênito: enfoque na comunicação\*\*\*

## Development skills in children with congenital hypothyroidism: focus on communication

Mariana Germano Gejão\*  
Dionísia Aparecida Cusin Lamônica\*\*

\*Fonoaudióloga. Mestre em Fonoaudiologia pela Faculdade de Odontologia de Bauru da Universidade de São Paulo. Endereço para correspondência: Rua Eduardo Vergueiro de Lorena, 5-44, Apto 61 - A - Bauru - SP - CEP - 17012-450 (magejao@yahoo.com.br).

\*\*Fonoaudióloga. Professora Livre-Docente do Departamento de Fonoaudiologia da Faculdade de Odontologia de Bauru da Universidade de São Paulo.

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### Abstract

**Background:** congenital hypothyroidism may cause alterations in the child's global development. **Aim:** to outline the development profile in children with congenital hypothyroidism, focusing on communication, and to verify the influence of clinical history on the outlined profile. **Method:** 35 children, with ages between 2 to 36 months, with congenital hypothyroidism detected by neonatal screening, and who were in treatment for at least one month using hormonal replacement were assessed using the Early Language Milestone Scale (ELM) and the Portage Operation Inventory (POI). The clinical history was obtained in an interview with the family and from the analysis of medical records. **Results:** in the ELM, eleven children presented a poor performance in the expressive auditory function, two in the visual function and one in the receptive auditory function. In the POI, seven children presented a poor performance in the language section, five in cognitive section, four in the motor and social sections and three in the self-care section. There was no correlation between the results obtained in the assessments and the clinical history. **Conclusion:** most of the children presented adequate performances in the evaluated skills. For the children with altered performance, larger deficits were observed in the language section, for the expressive aspects, and in the cognitive section. The influence of clinical history on the development profile was not confirmed. However, a tendency for an adequate performance was observed in those children who underwent neonatal screening, received an early diagnosis and treatment for the congenital hypothyroidism and who received higher doses of thyroxine at the beginning of treatment. The importance of a speech-language follow-up for communication development in this population is highlighted.

**Key Words:** Congenital hypothyroidism; Child development; Language development; Evaluation.

### Resumo

**Tema:** o hipotireoidismo congênito pode acarretar alterações no desenvolvimento global infantil. **Objetivo:** traçar o perfil do desenvolvimento em crianças com hipotireoidismo congênito, enfocando a comunicação, e verificar influências da história clínica no perfil traçado. **Método:** foram avaliadas, por meio da *Early Language Milestone Scale* (ELM) e do Inventário Portage Operacionalizado (IPO), 35 crianças de 2 a 36 meses com hipotireoidismo congênito detectado na triagem neonatal, que realizavam tratamento com reposição hormonal há pelo menos um mês. A história clínica foi obtida por meio de entrevista com familiares e análise de prontuário. **Resultados:** na ELM, onze crianças apresentaram desempenho alterado na função auditiva expressiva, duas na visual e uma na auditiva receptiva. No IPO, sete crianças apresentaram desempenho alterado na área da linguagem, cinco na cognitiva, quatro nas áreas motora e social e três na de autocuidados. Não houve correlação entre os resultados e a história clínica. **Conclusão:** a maioria das crianças apresentou desempenho adequado para as habilidades avaliadas. Para as crianças com desempenho alterado, observou-se maior déficit na área de linguagem, nos aspectos expressivos, e na área cognitiva. Não ficou comprovada a influência da história clínica no perfil do desenvolvimento. Observou-se, entretanto, tendência para desempenho adequado nas habilidades avaliadas entre as crianças que realizaram a triagem neonatal, receberam o diagnóstico e o tratamento para o hipotireoidismo congênito mais precocemente e que receberam dosagem mais elevada de levotiroxina no início do tratamento. Ressalta-se a importância do acompanhamento fonoaudiológico longitudinal do desenvolvimento da comunicação nessa população.

**Palavras-Chave:** Hipotireoidismo congênito; Desenvolvimento infantil; Desenvolvimento da linguagem; Avaliação.

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## Introduction

In congenital hypothyroidism (CH) there is insufficient thyroid hormones production, important for the nervous system development(1). Its detection and early treatment beginning make possible sequels prevention in development skills(2), because it becomes one of the few causes of intellectual deficiency susceptible to prevention(1,3). Its development can be influenced for: gemelar gestation; feminine sex; families who have shown alterations in thyroid gland, among others(4).

Cognitive and motor deficits have been observed, mainly when the treatment is late and/or the hormonal alteration is more serious(5-12). The neuropsychomotor development altered may cause gaps in perceptive, cognitive, linguistics, social and self-care areas(13-14). It has been observed language delay, articulatory, phonological, morphosintatics and understanding alterations, vocabulary reduced and nomination difficulty(11-12,15).

The longitudinal attendance of these individuals' development has been jutting out(11,16-18) and, although it has been observed language alterations with late and early treatment beginning(19), the speech and hearing pathology performance is unforeseen in the attendance routine(2). With the early detection there is the possibility of the necessary stimulation anticipation for the global development(20) and low cost prevention programs establishment(21-22).

Ahead to the exposed, it was aimed to draw the children's development skills profile with CH diagnosed and treated early, focusing on communicative skills, and to verify clinical history influences in development skills.

## Method

It was obtained the approval of the Faculdade de Odontologia de Bauru of the Universidade de São Paulo Committee of Ethics in Research (protocol 53/2005) and Signature of Free and Illustrious Consent according to 196/96 Resolution. The study was developed in partnership with one of the six centers of São Paulo State Neonatal Screening (NS), accredited by Health Ministry, that assists the São Paulo Center West population with approximately 371 collection put in 206 cities.

Thirty five children among 2-36 months formed the casuistry according to the inclusion criteria: to possess CH diagnosis; to accomplish treatment and attendance for CH in Neonatal Screening Program (NSP), with appropriate adhesion, according to Health Ministry criteria; to be in medical treatment for CH for a superior period to 1 month; not to present other alterations genetic or neurological proven, which are not part of the CH picture.

Of the 57 registered children, in the described age group, it was excluded 22 because of the accomplish treatment and attendance for CH in their origin cities or inadequate adhesion to the program.

The clinical history was collected through medical records analysis and interview with the children's legal responsible (Table 1).

To draw the development skills profile it was used the procedures:

ELM scale application - early language milestone scale(23).

Language screening scale of children with 0-36 months, thoroughly used in national studies. It presents development marks contained in the auditory expressive functions (AE), auditory receptive (AR) and visual (V). AE is subdivided in content (babble, isolated words, sentences) and speech intelligibility. AR includes pré-linguistic auditory behaviors and verbal commands understanding. V includes visual following behaviors, answer to facial expressions and symbolic aspects.

The results application and analysis followed that proposed by the instrument. Vertical line was drawn along the answer protocol, in chronological age in months. All of the items that crossed this line in the three functions were appraised. The success or failure determination in each item was accomplished by report, direct behavior test or spontaneous behavior observation consideration. The results were analyzed in limit level (success obtaining in three higher consecutive items). To analyse its application, it was obtained failure in three consecutive items. The linguistic development was considered typical when the limit value in the three appraised functions corresponded to the child's chronological age. The same was considered for the three functions separately, making possible to evaluate the AE, AR and V language performance in an independent way.

TABLE 1. Casuistry characterization referring the historical clinic.

Casuistry characterization	n (%)	
data collection age (months)	2-12	16 (45.72)
	13-24	10 (28.57)
	25-36	9 (25.71)
Sex	Feminine	23 (65.71)
	Masculine	12 (34.29)
birth weigh (grams)	860-2.500*	6 (17.14)
	2.501-3.900	29 (82.86)
NS age (days)	2-7	16 (45.71)
	8-30	16 (45.71)
	31-60	1 (2.86)
	61-240	2 (5.71)
diagnosis age (days)	8-30	18 (51.43)
	31-60	12 (34.28)
	61-270	5 (14.29)
treatment begining age (days)	8-30	12 (34.28)
	31-60	17 (48.57)
	61-240	6 (17.14)
NS thyrotropin hormone level ( $\mu$ ui/ml)	6-20	16 (45.72)
	20-368	19 (54.28)
thyroxine dose in the treatment begining (mg/day)	6,50-25,00	23 (65.71)
	37,50-75,00	12 (34.29)
socioeconomic level	Inferior –low	9 (25.71)
	Superior-low	25 (71.43)
	Inferior- Medium	1 (2.86)
other risk factors for development alterations besides CH**	Yes	23 (65.71)
	No	12 (34.29)

\*Low weight<sup>(25)</sup>

\*\* Gestacional intercurrance and/or premature birth and/or birth low weight and/or health problems after birth.

Inventory portage operationalized application - IPO(24).

It was accomplished by legal responsible report and target behaviors observation. It was used answer protocol and ludic and pedagogic materials organized in agreement with the IPO demand. All of the children were appraised in motor, linguistics, cognitive, social and self-care areas, considering their age group. The infantile stimulation area was applied in children with development operation inferior to 4 months.

The motors skills refer to postures reactions; development neuropsychomotor and use of hands. The cognitive ones refer to adaptations front of problems; practical problems solution; sensorial-motors adjustments; whole decomposition in parts, its reintegration and relationships perception. The linguistic area includes facial expressions; gestures, vocalizations, words and sentences use; imitation and understanding. The socialization skills refer to social expressions and cooperation in activities and self-care, refer to the independence in routine activities. The infantile stimulation area refers to the tactile, visual and auditory sensibility, visual and auditory location and to the neuropsychomotor development.

Firstly, the age group items corresponding to the child's chronological age were appraised and when the performance was insufficient the previous age group was applied. It was determined, in each development area, the number of items that should be accomplished by the child for every month of each age group.

To verify the clinical history influence in the evaluations results, the casuistry characterization data were correlated with the performance in ELM Scale and IPO.

To present the development skills profile it was used descriptive statistics (absolute and relative

frequency values). To analyze the clinical history influence in development skills it was used: Mann Whitney, Fisher and Contingency qui-square tests, with significance level fastened of  $p < 0,05$ . The agreement among the casuistry performance in ELM Escala and IPO language area was measured through the kappa Statistics.

## Results

Twenty-four children (68.57%) obtained appropriate global performance for the chronological age in ELM Escala and 27 (77.14%) in IPO and, 11 children (31.43%) obtained altered performance in ELM Scale and 8 (22.86%) in IPO.

In ELM Scale, 68.57% presented appropriate performance for AE function, 97,14% for AR and 94,29% for V. In IPO, 91.43% obtained appropriate performance for self-care area, 88.57% for socialization and motor, 85.71% for cognition, 80.00% for language and 75.00% for infantile stimulation. This last one was just applied in 12 children (Table 2).

Of the eleven children with altered performance in ELM Scale, nine presented deficits just in AE function; one in AR and V and one in the three functions. Of the eight children with altered performance in IPO, three presented deficits in the five skills; two in language; 1 in motor, linguistics, cognitive and social skills; one in the self-care, linguistics and cognitive skills and 1 in motor, linguistics and cognitive skills. Alteration in cognitive and linguistic area was verified in six children and specifically in linguistic area in eight.

The agreement among the casuistry performance in ELM Scale and in IPO language area, was 0.559 (kappa Statistics). Clinical history influences were not observed in children's performance (Table 3).

TABLE 2. Casuistry distribution according to ELM Scale functions and IPO areas performance.

Performance	ELM Scale functions			IPO area					
	Auditory Expressive n (%)	Auditory Receptive n (%)	Visual n (%)	Self-care n (%)	Socialization n (%)	Motor n (%)	Cognition n (%)	Language n (%)	Infantile Stimulation* n (%)
Adequate	24 (68.57)	34 (97.14)	33 (94.29)	32 (91.43)	31 (88.57)	31 (88.57)	30 (85.71)	28 (80.00)	9 (75.00)
Altered	11 (31.43)	1 (2.86)	2 (5.71)	3 (8.57)	4 (11.43)	4 (11.43)	5 (14.29)	7 (20.00)	3 (25.00)
Total	35 (100.00)	35 (100.00)	35 (100.00)	35 (100.00)	35 (100.00)	35 (100.00)	35 (100.00)	35 (100.00)	12 (100.00)

\* Applied in children with inferior development to four months.

TABLE 3. Clinical history influence statistical analysis in children's performance in ELM Scale and IPO.

Clinical history	Performance	
	ELM Scale (p)	IPO (p)
data collection age <sup>a</sup>	0.487	0.608
sex <sup>b</sup>	0.709	1.000
birth weight <sup>a</sup>	0.606	0.542
NS age <sup>a</sup>	0.291	0.133
diagnosis age <sup>a</sup>	0.109	0.246
treatment beginning age <sup>a</sup>	0.285	0.665
NS thyrotropin hormone level <sup>a</sup>	0.570	0.479
thyroxine dose in the treatment beginning <sup>a</sup>	0.091	0.198
socioeconomic level <sup>c</sup>	0.788	0.615
other risk factors for development alterations, besides CH <sup>b*</sup>	1.000	1.000

a: Mann Whitney Test; b: Fisher Test; c: Contingency qui-square Test.

\* Gestacional intercurrente and/or premature birth and/or birth low weight and/or health problems after birth.

## Discussion

It was observed CH larger frequency in feminine sex. Studies pointed 2:1(16) and 4:1(3) proportion for the feminine sex and considered it a risk factor for CH(4).

The development skills more dephased belonged to the language area, reinforcing that children with CH have risk for alterations in linguistic development(11). Studies also observed alterations in individuals' communicative performance with HC(5,11-13). Among the linguistic skills, the AE function was shown more committed in relation to AR and V. Study with CH also observed expressive language skills inferior to the receptive ones(11).

The statistical agreement among the performance in ELM Scale and in IPO language area showed that the scale was more sensitive in children's identification with alterations in the communicative development, revealing the importance of the application of more than one evaluation instrument.

The cognitive ability was the second more committed area. There is great concern about the development attendance of this ability, because the main CH sequel is the intellectual deficiency(1). Studies with individuals that accomplished early and appropriate treatment for CH, revealed cognitive alterations absence(6,9,11-12,17-18). The cognitive compromising accompanied the linguistic compromising, showing relationship between the cognitive and linguistic development.

Motor skills alterations were observed, according to literature(11-12). There are reports that motor alterations in children with CH persisted until the adult age, emphasizing the longitudinal attendance importance(9).

The socialization and self-care skills were shown altered, according to the literature (10). Deficits in these skills happen due to the practice lack in daily tasks, proportionate for the family actions in cares for the child. For interfering in the environmental exploration quality, in the interaction and relationships that the child establishes in the atmosphere, the motor, linguistic and cognitive compromisings observed, might have influenced the children's global development(13-14).

It was not statistically proven that the clinical history has influenced in the appraised skills. The age difference in the age groups among the children with appropriate and altered performance for ELM Scale and IPO was not significant statistically.

The sex and birth weight variables were not

significant for differences in children's performance. In spite of not being important risk factor for the CH occurrence(4), the low birth weight can interfere in the development(25).

Tendency for appropriate performance was observed in the appraised skills, among the children that accomplished NS, received diagnosis and began treatment for CH earlier. However, that difference in the performance was not statistically significant. Absence of this correlation was also found in other studies(8-9), once the treatment beginning age was also early. It is stood(jutting) out that by definition, CH is one of the few causes in that the intellectual deficiency may be forewarned, when treated earlier(1).

The difference in the thyrotropin levels average obtained in the NS in children with appropriate and altered performance in ELM Scale and IPO was not statistically significant. However, studies have been observing inverse relationship among CH gravity in the diagnosis and performance in cognitive, linguistics and motor skills(5,9,11-12,17). This result can suggest that in spite presenting different hormonal levels in NS, the hormonal replacement treatment has been adapted to prevent infantile development sequels.

Tendency for appropriate performance was observed in the appraised skills among the children that received higher dosage of thyroxine in the treatment beginning. However, that difference in the performance was not statistically significant, suggesting that the children began treatment in early ages and with appropriate dosages to prevent alterations current of the thyroid hormones lack in the organism. The literature has been describing correlation between thyroxine dosage and performance in cognitive, motor and learning skills(5,7-8,18). These correlations happen due to the largest difference among the hormonal replacement dosages studied by these authors when compared to the observed in this study.

The children's performance with different socioeconomic levels didn't differ significantly for the two used instruments. The fact of there being just a child in the medium-inferior socioeconomic condition might have contributed to that association absence. Correlation between socioeconomic level and children with CH performance in motor, cognitive and linguistics skills were described in the literature(6,12,17). In these studies there was homogeneity in the number of individuals in each socioeconomic level.

The children's performance with other risk factors for development alterations, besides CH, didn't differ significantly from performance of those that didn't present other risk factors.

This NSP has been accomplishing the established norms for Neonatal Screening National Program(2), that seek to identify metabolic alterations and to prevent deficiencies. However, development alterations frequently happen, contemplating in the general learning skills.

The study discoveries suggest the need of the Speech Hearing and Language Pathologist insert in the NSP professionals team that accompany longitudinally the individuals' development with HC, once these children have the risk for linguistic development alterations and, therefore, they need communicative development attendance(11,19). This way, the Speech Hearing and Language Pathologist performance will contribute to the early prevention and detection of possible alterations in the infantile development, as already outstanding in the literature(20-22).

## Conclusion

In this study, most of the children (68.57%) presented appropriate performance for the motor, cognitive, linguistics, social and self-care skills. For children with altered performance in development skills, larger deficit was observed in the language skills, mainly in what concerns to the expressive aspects and cognitive skills. It was not proven the clinical history influence in the profile drawn for the development skills. However, tendency was observed for appropriate performance in the appraised skills among the children that accomplished NS, received diagnosis and began treatment for CH earlier and that received higher thyroxine dosage in the medical treatment beginning of CH. These children's longitudinal attendance will contribute to the complementation of the discoveries in this study.

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