# The Virtual Man Project's CD-ROM "Voice Assessment: Speech-Language Pathology and Audiology & Medicine", Vol.1

**Millena Maria Ramalho Matta VIEIRA[1], Giédre BERRETIN-FELIX[2], Alcione Ghedini BRASOLOTTO[2]**

1- BS, Speech-Language pathologist.
2- BS, MSc, PhD, Assistant Professor of the Department of Speech-Language Pathology and Audiology, Bauru School of Dentistry, University of São Paulo, Bauru, SP, Brazil.

**Corresponding address:** Alcione Ghedini Brasolotto - Departamento de Fonoaudiologia da Faculdade de Odontologia de Bauru, Universidade de São Paulo - Alameda Octávio Pinheiro Brisolla, 9-75 - 17012-901 - Bauru, SP - Brasil - Phone: +55 14 32358332 - Fax: +55 14 32358460 - e-mail: alcione@fob.usp.br

**Received: November 12, 2009 - Accepted: March 14, 2010**

## ABSTRACT

The CD-ROM "Voice Assessment: Speech-Language Pathology and Audiology & Medicine" was developed as a teaching tool for people interested in the production of the spoken or sung human voice. Its content comprises several subjects concerning the anatomy and physiology of spoken and sung voice. A careful assessment becomes necessary in order to ensure the effectiveness of teaching and learning educational materials, whether related to education or health, within the proposal of education mediated by technology. Objective: This study aimed to evaluate the efficacy of the Virtual Man Project's CD-ROM "Voice Assessment: Speech-Language Pathology and Audiology & Medicine", as a self-learning material, in two different populations: Speech-Language Pathology and Audiology students and Lyrical Singing students. The participants were instructed to study the CD-ROM during 1 month and answer two questionnaires: one before and another one after studying the CD-ROM. The quantitative results were compared statistically by the Student's t-test at a significance level of 5%. Results: Seventeen out of the 28 students who completed the study, were Speech-Language Pathology and Audiology students, while 11 were Lyrical Singing students (dropout rate of 44%). Comparison of the answers to the questionnaires before and after studying the CD-ROM showed a statistically significant increase of the scores for the questionnaire applied after studying the CD-ROM for both Speech-Language Pathology and Audiology and Lyrical Singing students, with $p<0.001$ and $p<0.004$, respectively. There was also a statistically significant difference in all topics of this questionnaire for both groups of students. Conclusion: The results concerning the evaluation of the Speech-Language Pathology and Audiology and Lyrical Singing students' knowledge before and after learning from the CD-ROM allowed concluding that the participants made significant improvement in their knowledge of the proposed contents after studying the CD-ROM. Based on this, it is assumed that this didactic material is an effective instrument for self-learning of this population.

**Key words**: CD-ROM. Distance education. Assessment. Voice.

## INTRODUCTION

The use of information technology in the areas of education and health with teaching and assistance purposes has continuously increased. Technology-assisted education is a teaching modality that employs didactic resources, presented as different information bases. It can involve several means of communication facilitating the self-learning process[8] and the students' independence because they will be able to study according to their own capacity, anywhere and at any time[12]. The use of these pedagogical learning strategies, also denominated as collaborative strategies, is very

effective to increase the motivation level of the participants and for the accomplishment of the proposed activities[6].

Under the coordination of the Professor Chao Lung Wen, the Virtual Man Project is part of the category of learning goals of the Medical School of the University of São Paulo (FM/USP), described as a powerful iconographic resource that helps learning, considering that it facilitates understanding in relation to a specific subject[3]. As a result of a partnership among the Medical School of the University of São Paulo (FM/USP), Bauru School of Dentistry of the University of São Paulo (FOB/USP) and Federal University of São Paulo (UNIFESP), a CD-ROM denominated "Voice Assessment: Speech-Language Pathology and Audiology & Medicine", vol. 1, was developed under the coordination of Professor György Mikɛlós Böhm (FM/USP). This volume comprises the following subjects: upper commands and innervations of the phonation tract; functional anatomy of the larynx; physiological functions of the larynx; vocal tract (forming and vibrating); sound articulation (vowels and consonants) and singing. It is a kind of technology that allows observing the speech tract in a three-dimensional and dynamic way, enabling the comprehension of the speech complexity in a simpler way. In addition to the iconographic communication of the Virtual Man Project, the CD-ROM contains sounds, films, several illustrations and texts[16]. This material has been developed to be applied as a didactic instrument for both undergraduate and graduate students, and anyone interested in the production of the human voice, spoken or sung.

The elaboration of a didactic material of any nature always demands previous analysis in order to promote actual learning facilities to the students[15]. The evaluation of an educational software, for example, is a step of fundamental importance for reaching the proposed goals and comprise the teaching-learning problem that has motivated its creation[17].

The aim of the present study was to analyze the efficacy of the Virtual Man Project's "Voice Assessment: Speech-Language Pathology and Audiology & Medicine", vol. 1, as a self-learning material for two distinct populations.

## MATERIAL AND METHODS

The present study was reviewed and approved by the Department of Speech-Language Pathology and Audiology and by the FOB/USP's Research Ethics Committee (Process #120/27). The participants received verbal and written explanations about the study purposes and signed an informed consent form.

Thirty students attending the 1st semester of the Speech-Language Pathology and Audiology course at FOB/USP and 30 students attending the 1st-6th year of the Lyrical Singing course at the Drama and Music Conservatory "Dr. Carlos de Campos", in the city of Tatuí, SP, were invited to participate. At first, 50 students accepted taking part in the study, being 23 Speech-Language Pathology and Audiology students and 27 Lyrical Singing students. The following exclusion criteria were adopted: previous contact with the content of the CD-ROM under study; and not having access to a computer with CD reader and without specific softwares for images and sounds.

Initially, a questionnaire denominated "before CD-ROM questionnaire" containing one open and multiple-choice questions was handed to the students. All questionnaires had 4 options of answers (a, b, c and d), the last one being "I do not know the answer". The questionnaires were different according to each group. For the Speech-Language Pathology and Audiology students, 44 questions were elaborated about the following topics of the CD: upper commands and innervation of the phonation tract (17 questions), functional anatomy of the larynx (7 questions), physiology of the larynx (14 questions) and human voice sound articulation (6 questions). For the Lyrical Singing students, 36 questions were elaborated about the following topics of the CD: upper commands and innervation of the phonation tract (6 questions), functional anatomy of the larynx (7 questions), physiology of the larynx (14 questions) and sung voice (8 questions).

All students received a Virtual Man Project's "Voice Assessment: Speech-Language Pathology and Audiology & Medicine", vol. 1, and an auxiliary study guide, which was elaborated with some

questions related to the topics of the CD for both courses. The Speech-Language Pathology and Audiology students were oriented to study the following subjects: upper commands and innervations; propositional vocalization, emotional vocalization, important skull nerves for phonation, larynx (functional anatomy: cartilages, ligaments and articulations; extrinsic muscles; intrinsic muscles, vocal fold; glottis configuration), physiology of the larynx (functions of the larynx, phonation, types of voice, vocal tract (forming and vibrating), human voice (sound articulation: vowels and consonants). The Lyrical Singing students were oriented to study the following subjects: larynx (vocal fold, glottis configuration) physiology of the larynx (functions of the larynx, phonation, types of voice, vocal tract (forming and vibrating) singing (phonation mechanisms and some aspects of sung voice).

After answering the "before CD-ROM questionnaire" and receiving the didactic material (CD-ROM and the auxiliary study guide), a period of 1 month was granted to the participants to study. After that period, the participants were contacted in person or by e-mail or telephone to schedule a date for answering a second questionnaire, denominated "after CD-ROM questionnaire". This second questionnaire was elaborated with the exact same questions of the "before CD-ROM questionnaire" and some other open questions, which aimed at knowing the opinions and suggestions of the students about the CD-ROM as well as any problems they found during the study.

As a strategy for the students to study the material within the established period, they were informed that the FOB/USP's Department of Speech-Language Pathology and Audiology would provide them an attendance certificate at the end of the research and another certificate of good performance for the ones who reached the score of 75% or higher on the "after CD-ROM questionnaire".

The t-test for dependent samples was applied for analysis of the quantitative results (comparison of the answers of the questionnaires applied before and after the CD-ROM), as it was considered significant $p<0.05$. An analysis of the qualitative results was performed by means of questions referring to the participants' suggestions and the problems they faced when studying the CD-ROM.

## RESULTS AND DISCUSSION

From the 50 students that initially accepted taking part in the study, only 28 (56%) remained until the end of the investigation. From these 28 participants, 17 (60.71%) were Speech-Language Pathology and Audiology students and 11 (39.28%) were Lyrical Singing students. There were only four male students, one from the Speech-Language Pathology and Audiology course and three from the Lyrical Singing course. The mean ages were 18.94 and 27.72 years for the Speech-Language Pathology and Audiology and Lyrical Singing students. The dropout rate was 44%, being 16 students who from the Lyrical Singing course and 6 students from the Speech-Language Pathology and Audiology course. The students who retired from the study alleged that they did not have enough time to study the CD-ROM. Although the study of the CD-ROM has not been proposed as a formal course, the results are in agreement with the literature, which points a dropout rate around 50%[5,13,14] for technology-assisted educational courses, lack of time being the most frequently reported reason for giving up.

Considering that the maximum score to be reached by the Speech-Language Pathology and Audiology students on the "before CD-ROM questionnaire" was 44 points, the mean of correct answers of all students was 5.94 (13.35%), with of minimum of zero and maximum of 12 points (27.27%). On the "after CD-ROM questionnaire", the students had 20.59 (46.79%) mean of correct answers, with minimum of 11 (25%) and maximum of 39 points (88.63%). Comparison of the results of both questionnaires showed a statistically significant increase ($p<0.001$) on the scores of the "after CD-ROM questionnaire". Statistically significant differences were also present in all topics of this questionnaire (Table 1).

The results of the "before CD-ROM questionnaire" for the Lyrical Singing students, which had as maximum value 36 points, the mean of correct answers was 6,45 (17,91%), with

minimum of 1 (2.77%) and maximum of 16 points (44.44%). On the "after CD-ROM questionnaire", the mean of correct answers was 15.73 (43.69%), with minimum of 1 (2.77%) and maximum of 29 points (80.55%). Comparison of the results of both questionnaires showed a statistically significant increase ($p<0.004$) on the scores of the "after CD-ROM questionnaire". Statistically significant differences were also present in all topics of this questionnaire (Table 2).

It is possible to notice that the maximum score for the "before CD-ROM questionnaire" reached by the Speech-Language Pathology and Audiology students was similar to the minimum reached for the "after CD-ROM questionnaire". Similarly, better results were observed in the "after CD-ROM questionnaire" by the Lyrical Singing students when compared to the "before CD-ROM questionnaire", demonstrating the effectiveness of the study also for this group. Through a written evaluation of a study proposal employing the CD-ROM, statistically significant differences were also observed before and after the test, demonstrating that the elaborated material helped the student learning2. A study that examined the impact of an online course on healthcare professionals' competency in infection prevention using questionnaires applied before and after the course for knowledge evaluation, concluded that the participants made statistically significant increases in their perceptions of competency in infection control following the course. Additionally, most of the participants were very pleased and reported that the acquired knowledge was useful and relevant for their professional practice[1].

For the Speech-Language Pathology and Audiology students, the topic in which there were more correct answers in the "before and after CD-ROM questionnaires" was articulation of the human voice sounds (before = 16.66% and after = 52.94%). The topics that presented the largest number of incorrect answers in the "before CD-ROM questionnaire" were functional anatomy of the larynx and physiology of the larynx (84.87%). In the "after CD-ROM questionnaire", the topic that presented more incorrect answers was physiology of the larynx (55.46%). The option "I do not know the answer" was employed more frequently on the topic upper commands and innervation of the phonation tract (73.35%) in the "before CD-ROM questionnaire" and on the topic physiology of the larynx (18.48%) in the "after CD-ROM questionnaire".

For the Lyrical Singing students, the topic that presented more correct answers in the "before and after CD-ROM questionnaires" was sung voice (before = 24.24% and after = 49.49%). The topic that presented the largest number of incorrect answers in the "before CD-ROM questionnaire" was functional anatomy of the larynx (89.61%). In the "after CD-ROM questionnaire", the topic that presented more incorrect answers was physiology of the larynx (61.03%). The option "I do not know the answer" was employed more frequently on the topic functional anatomy of the larynx in both the before (80.51%) and after (24.02%) CD-ROM

**Table 1**- Results before and after CD-ROM study, by topics – Speech-Language Pathology and Audiology students

| Topics | Mean | SD | Min. | Max. | T | p |
|---|---|---|---|---|---|---|
| Upper Commands - Before | 1.71 | 1.4 | 0 | 5 | - 7.217 | <0.001* |
| Upper commands - After | 7.71 | 3.46 | 3 | 17 | | |
| Functional Anatomy - Before | 1.06 | 1.03 | 0 | 3 | - 5.66452 | <0.001* |
| Functional Anatomy - After | 3.59 | 1.58 | 1 | 7 | | |
| Larynx Physiology - Before | 2.12 | 1.69 | 0 | 5 | - 5.87169 | <0.001* |
| Larynx Physiology - After | 6.24 | 2.82 | 2 | 11 | | |
| Sound Articulation - Before | 0.94 | 1.14 | 0 | 3 | - 6.96262 | <0.001* |
| Sound Articulation - After | 3.24 | 1.25 | 1 | 6 | | |
| Total Before | 5.94 | 3.49 | 0 | 12 | - 7.71699 | <0.001* |
| Total After | 20.59 | 7,3 | 11 | 39 | | |

*Statistically significant difference ($p<0.05$)

questionnaires.

The mean of correct answers for the Speech-Language Pathology and Audiology students in the "after CD-ROM questionnaire" was 20.59 and for the Lyrical Singing students was 15.75. It was observed a relevant improvement of 14.65 points for the Speech-Language Pathology and Audiology students and 9.30 points for the Lyrical Singing students. It appears that the items that presented the largest number of correct answers for both groups were those that were directly related to their area of study.

It was observed that one Lyrical Singing student presented more correct answers in the "before CD-ROM questionnaire" (2 correct) than "after CD-ROM questionnaire" (1 correct). In addition, another student presented the same results for before and after the CD-ROM (16 correct answers). This was not observed for the Speech-Language Pathology and Audiology students. Educational courses mediated by technology require mature, self-confident and self-motivated students that are normally better applied to professionals who are already working and intend to be successful in their careers or are constantly being required for updating knowledge[7]. It is known that motivation is a determining factor for the success of the learning process. In addition, it is important that the students organize their time and space for studying, as well as developing autonomous learning strategies, so that they may become the active subject of their cultural background, developing the learning process in any environment[4].

Referring to the frequency of the study of the CD-ROM, most students of both groups reported that they studied from 3 to 4 times the material. Although a quantitative analysis was not performed for such purpose, the score reached by the students in relation to the frequency of the study indicates that it did not interfere on the performance of the students, because the ones who had the higher frequency of study were not the ones who presented the highest scores.

In the beginning of the "after CD-ROM questionnaire", there were questions referring to the CD-ROM use itself as well as space for the participants presenting suggestions and/or problems faced during the study. For the questions regarding to static and dynamic images, most of the answers were Very good and Excellent. About the texts of the CD-ROM, most answers were Very good. There were no answers Poor or Regular for those items. About the theoretical content, most of the evaluations were Good and Very good, and there was no Poor evaluation.

When the students were asked if they faced any difficulty in exploring the CD-ROM, only 2 (11.76%) Speech-Language Pathology and Audiology students and 4 (36.36%) Lyrical Singing students answered affirmatively. The problems were related to the computer, for example: incompatible programs for the CD-ROM, "slow computer", bad functioning of the videos, and computer technical failure.

Regarding the question on the problems faced

**Table 2-** Results before and after CD-ROM study, by topics – Lyrical Singing students

| Topics | Mean | SD | Min. | Max. | T | p |
|---|---|---|---|---|---|---|
| Upper Commands - Before | 0.82 | 0.75 | 0.00 | 2.00 | -4.3788 | <0.001* |
| Upper commands - After | 2.73 | 1.10 | 1.00 | 5.00 | | |
| Functional Anatomy - Before | 0.64 | 0.81 | 0.00 | 2.00 | -4.98098 | <0.001* |
| Functional Anatomy - After | 3.18 | 1.54 | 0.00 | 5.00 | | |
| Larynx Physiology - Before | 2.64 | 1.69 | 1.00 | 6.00 | -2.29661 | <0.045* |
| Larynx Physiology - After | 5.45 | 3.59 | 0.00 | 12.00 | | |
| Sung Voice Before | 2.18 | 2.40 | 0.00 | 6.00 | -2.49723 | <0.032* |
| Sung Voice After | 4.55 | 2.38 | 0.00 | 9.00 | | |
| Total Before | 6.45 | 4.48 | 1.00 | 16.00 | -3.75671 | <0.004* |
| Total After | 15.73 | 7.42 | 1.00 | 29.00 | | |

*Statistically significant difference (p<0.05)

about the theoretical content of the CD-ROM, 1 (5.88%) Speech-Language Pathology and Audiology student did not answer the question; 5 (29.41%) Speech-Language Pathology and Audiology students and 7 (63.63%) Lyrical Singing students reported that they did not face any problems. The problems reported by the participants referred to the type of vocabulary contained in the CD-ROM, which was considered very technical and specific; the immaturity of the students to understand complex subjects for being students of the first year of college, and some topics that were too long.

From a total of 17 Speech-Language Pathology and Audiology students, only 7 (41.17%) gave some suggestions for the CD-ROM and from the 11 Lyrical Singing students, only 6 (54.54%) gave some suggestions. The suggestions given by the Speech-Language Pathology and Audiology students were: "It would be better if the videos were a little slower"; "Make them accessible on Windows Vista"; "Although the texts were narrated before the videos, it would be interesting to have a narration during the videos"; "More pictures are necessary, so that the texts can be more explored"; "Pronunciation of just the sound of the phoneme, without the sound of the consonants"; "Make the CD run in other programs". The suggestions given by the Lyrical Singing students: "Employ a simple language and explain the technical terms"; "Less texts and more explanations presenting the tables, more graphics and clips, all simultaneously"; "Allow another type of access as DVD"; "Elaborate an easier language"; "Add more explanations about respiration"; "Add more pictures".

Only 2 students, one from each group, obtained a score higher than 75% in the "after CD-ROM questionnaire". The Speech-Language Pathology and Audiology student obtained 88.63% score and the Lyrical Singing student 80.55%.

Overall, the aspects related to static and dynamic images, texts and theoretical content were positively evaluated by the students. Similarly, the subjective evaluation of the participants of a multimedia program about pleural drainage[10] presented some results like excellent, very good and good, concerning the aspects related to informatics and content. Another study about the development and evaluation of a multimedia system about irrigation[9] required 56 people to evaluate a CD-ROM in three different ways: as a whole, on the didactic aspect and only one chapter of the CD-ROM. The educational role of the software was evaluated in relation to the facility to access to the screen; the sequence of the units of the CD-ROM; the amount of information; the quality of the videos, audio, digital pictures, graphic animation and written information; if the examples presented were important for the student to learn; if the material corresponded to the expectations of the user in both theoretical and practical terms. The evaluation indicated that the developed multimedia system may be applied as a didactic resource for teaching and learning purposes[9].

There is no doubt that it is necessary to evaluate all the possible aspects of the materials that are employed for teaching and learning, whether they are concerned to health or education. Thus, studies must regard not only the elaboration and creation, but also the evaluation of the material. The follow up during all steps of the study of the CD-ROM by the learner may reduce the motivational problems found and allows for expanding the knowledge about its use.

An aspect that may contribute for understanding the ability of learning from the use of the CD-ROM refers to its usability, and this should be addressed in further studies. Usability is a set of factors that ensure that the products are easy to manage, efficient and pleasant, according to the perspective of the user[11]. Employing some strategies to evaluate usability enables the comprehension for some ways of learning for each user and also the difficulties faced during the process. Although it has not been possible to employ usability strategies on the present study, an auxiliary guide with questions referring to the CD-ROM topics was supplied to help students organizing themselves. In addition, the students were free to contact the researcher for clarification about the use of the CD-ROM.

Another very important aspect of this investigation was having two distinct groups evaluating the CD-ROM, employing traditional

methodologies and modern methodologies that use the technological resources in order to compare the ways students study and learn, as well as the teaching methods.

The teaching tool evaluated in this investigation contributes to the knowledge of some subjects referring to the assessment of voice, its upper commands and innervation of the larynx, anatomy and physiology, articulation of the voice sound and sung voice. The findings of the present investigation contributed to validate the efficacy of this didactic material for the population under study and this CD-ROM was proven useful for both in loco and at distance teaching modalities. Diverse populations can benefit from this material, including doctors, healthcare and education professionals, ordinary people, students and even patients.

## CONCLUSION

The results concerning the evaluation of the Speech-Language Pathology and Audiology and Lyrical Singing students' knowledge before and after learning from the CD-ROM allowed concluding that the participants made significant increase in their knowledge of the proposed contents after studying the CD-ROM. Based on this, it is assumed that this didactic material was an effective instrument for self-learning of this population.

## ACKNOWLEDGEMENTS

First of all we are very grateful to the Tutorial Educational Program (PET) in Speech-Language Pathology and Audiology from the Ministry of Education that enabled the accomplishment of this study as well as the creators of the Virtual Man Project, the coordinator Professor György Mikelós Böhm and the responsible for the Virtual Man Project Professor Chao Lung Wen, both from FMUSP.

We would also like to thank the coordinators of the Speech-Language Pathology and Audiology course of Bauru School of Dentistry of the University of São Paulo and the Lyrical Singing course of the Drama and Music Conservatory "Dr. Carlos de Campos" in the city of Tatuí, SP, for the authorization and collaboration for the optimal research development. Special thanks to the students because without their collaboration our study could not be done.

## REFERENCES

1- Atack L, Luke R. Impact of an online course on infection control and prevention competencies. J Adv Nurs. 2008;63(2):175-80.
2- Blasca WQ, Bevilacqua MC. A multimídia como uma nova proposta de ensino da audiologia. Salusvita. 2006;25(3):113-26.
3- Böhm GM, Chao LW. Estação digital médica e estágio rural multiprofissional: proposta de estratégia de integração regional através de ação de cidadania, resgate social e inclusão digital [online]. Apr 2005. [cited 2010 March 20]. Available from: http://www.estacaodigitalmedica.com.br/estacaodigitalmedica/pdf/EstacaoDigitalMedica_EstagioRural.pdf.
4- Maia MC, Meirelles FS, Pela SK. Análise dos índices de evasão nos cursos superiores a distância do Brasil [online]. Apr 2004. [cited 2010 March 20]. Available from: http://www.abed.org.br/congresso2004/ por/pdf/073-TC-C2.pdf.
5- Marques IR, Marin HF. Enfermagem na web: o processo de criação e validação de um web site sobre doença arterial coronariana. Rev Latinoam Enferm. 2002;10(3):298-307.
6- Medélez Ortega E, Burgun A, Le Duff F, Le Beux P. Collaborative environment for clinical reasoning and distance learning sessions. Int J Med Inform. 2003;70(2-3):345-51.
7- Moran JM. Avaliação do ensino superior a distância no Brasil [online]. 2007. [cited March 20 2010]. Available from: http://www.eca.usp.br/prof/moran/avaliacao.htm.
8- Oliveira MA. Educação a distância como estratégia para a educação permanente em saúde: possibilidades e desafios. Rev Bras Enferm. 2007;60(5):585-9.
9- Oliveira RA, Mota RD, Farias CV, Bastos LN, Ramos MM. Desenvolvimento e avaliação de sistema multimídia para ensino e aprendizado de irrigação. Rev Bras Eng Agríc Ambient. 2002;6(3):553-6.
10- Perfeito JA, Forte V, Giudici R, Succi JE, Lee JM, Sigulem D. Desenvolvimento e avaliação de um programa multimídia de computador para ensino de drenagem pleural. J Bras Pneumol. 2008;34(7):437-44.
11- Pimenta DN, Diniz HM, Andrade MA, Oliveira PR, Silva JF, Dias JC, et al. A importância do ergodesign na avaliação de CD-ROM sobre dengue e doença de chagas na educação em saúde. Trab Educ Saúde. 2008;6(1):147-67.
12- Reynolds PA, Mason R, Eaton KA. Remember the days in the old school yard: from lectures to online learning. Brit Dent J. 2008;204(8):447-51.
13- Ribeiro MA, Lopes MH. Desenvolvimento, aplicação e avaliação de um curso a distância sobre tratamento de feridas. Rev Latinoam Enferm. 2006;14(1):77-84.
14- Sanches LM, Lopes MH. Educação a distância sobre cardioversão e desfibrilação para enfermeiros. Rev Bras Enferm. 2008;61(5):583-8.
15- Souza TR. A avaliação como prática pedagógica [online]. 2002. [cited 2009 Jun 18]. Available from: http://www.abed.org.br/congresso2000/texto01.doc.
16- Voz: fonoaudiologia e medicina. Projeto Homem Virtual. Vol 1 [CD-ROM]. FOB-USP/UNIFESP/FM-USP São Paulo: 2006.
17- Zem-Mascarenhas SH, Cassiani SH. Desenvolvimento e avaliação de um software educacional para o ensino de enfermagem pediátrica. Rev Latinoam Enferm. 2001;9(6):13-8.

# Temporomandibular disorders, voice and oral quality of life in women

**Tatiane Cristina PEREIRA[1], Alcione Ghedini BRASOLOTTO[2], Paulo César CONTI[3], Giédre BERRETIN-FELIX[2]**

1- MSc Student, Department of Speech-Language Pathology and Audiology, Bauru School of Dentistry, University of São Paulo, Bauru, SP, Brazil.
2- MSc, PhD, Assistant Professor, Department of Speech-Language Pathology and Audiology, Bauru School of Dentistry, University of São Paulo, Bauru, SP, Brazil.
3- DDS, PhD, Associate Professor, Department of Prosthodontics, Bauru School of Dentistry, University of São Paulo, Bauru, SP, Brazil.

**Correspondig address:** Giédre Berretin-Felix - Faculdade de Odontologia de Bauru - Universidade de São Paulo - Departamento de Fonoaudiologia - Alameda Dr. Octávio Pinheiro Brisolla, 9-75 - C.P. 73 - 17012-901 - Bauru, SP, Brasil - Phone: +55-14-3235-8332 - e-mail: gfelix@usp.br

**Received: December 16, 2009 - Modification: March 24, 2010 - Accepted: April 01, 2010**

## ABSTRACT

Some studies have shown a relationship between temporomandibular disorders (TMD) and dysphonia, as well as quality of life in oral health. Objective: The purpose of this study was to investigate the correlation between severity of vocal self-perception and TMD severity and the correlation between oral health-related quality of life impairment and TMD severity. Material and methods: Thirty-three women aged 20 to 40 years, with or without complaint of dysphonia, were recruited at the Bauru campus of the University of São Paulo, Brazil, and the local community. All participants were subjected to an investigation of quality of life related to dental and speech aspects by the application of Oral Health Impact Profile-short form (OHIP-14) and the Voice-Related Quality of Life (V-RQOL) protocol. Also, a questionnaire was applied to detect the presence and severity of TMD. Results: There was significant correlation between TMD and quality of life for all aspects analyzed in the oral health protocol, except for function and physical limitation ($p>0.05$). There was negative correlation between TMD and voice-related quality of life in the total score ($p=0.007$) as weel as physical ($p=0.008$) and socio-emotional aspects ($p=0.017$). In addition, there was statistically significant correlation between TMD and vocal self-perception ($p=0.037$). Conclusion: There is an association between TMD severity, voice-related and oral health-related quality of life. It is important to investigate in future studies the vocal self perception as well as the oral and voice conditions in patients with TMD.

**Key words**: Temporomandibular joint disorders. Voice. Quality of life.

## INTRODUCTION

Temporomandibular disorders (TMD) result from abnormal functioning of the masticatory muscles, temporomandibular joints (TMJs), associated structures or both[8]. TMD are considered as a multifactorial manifestation and can be related to parafunctional habits such as tooth clenching and/or bruxism, head or neck traumas, unstable bite, postural problems, and emotional stress, among others[24].

Individuals with TMD may present headaches or neck pain, TMJ noises, tinnitus or ear fullness, crepitation[10] on opening or closing the mouth, opening limitation, and difficulties in chewing[9,11] and on the speech[5,6]. TMD can even influence individual's psychosomatic characteristics reducing their quality of life[2].

The Oral Health Impact Profile (OHIP) is a scaled index developed in Australia to measure oral health related to quality of life. This questionnaire has been used in its German version to characterize quality of life related to oral health in individuals with TMD, and scores of the questionnaire indicated damages in different categories of diagnosis, with greater emphasis on the psychosocial axis[16]. The OHIP has also been applied in Slovak patients,

resulting that individuals with TMD have more physical symptoms related to injury, and greater commitment of the oral health-related quality of life with the increase of age[27].

In addition to the TMD impact on orofacial functions and the individuals' quality of life, vocal changes are signs and symptoms commonly associated with TMD cases. This occurs due to the fact that TMD etiologic factors are common to dysphonia, such as excessive tension in the cervix and orofacial region and mouth opening restriction, since mandibular movement limitation during speech can affect voice acoustics[27].

In cases of dysphonia, quality of life and emotional condition are also compromised. In the last years, a number of scales have been developed[19], making possible the individual self-assessment about the severity of one's voice problem. The Voice-Related Quality of Life (V-RQOL) protocol assesses individual's perception of the voice problem impact on one's life[14]. The Brazilian version of the V-RQOL, named "*Qualidade de Vida em Voz – QVV*", has been proven a valid, reliable and sensitive instrument that specifically assesses patients with voice problems[12,13].

Several studies have been carried out to elucidate the relationship between emotional conditions and TMD or dysphonia, but very limited research has been done with individuals presenting both conditions. In addition, the application of quality of life protocols has brought important contributions to the understanding of the impact of the problems presented by patients as well the therapeutic approach in their lives.

The goal of this study was to investigate the correlation between the severity of vocal self-perception and TMD severity, between voice-related quality of life and TMD severity, and between oral health-related quality of life impairment and TMD severity.

## MATERIAL AND METHODS

### Ethical Aspects

The study was approved by the Research Ethics Committee of Bauru School of Dentistry, University of São Paulo (Protocol #173/2007). All women recruited for the study were clearly informed about the research purposes and their acceptance to participate was expressed by their signature on an informed consent form.

### Participants

The study sample was composed by 33 women aged 20 to 40 years (mean age: 25.61 years), with or without dysphonia symptoms, who were recruited at the Bauru campus of the University of São Paulo and the local community.

The following exclusion criteria were used[18]: 1. Two or more teeth missing (excluding third molars); 2. Use of removable denture; 3. Presence of occlusal risk, such as such anterior open bite, unilateral crossbite, overjet greater than 6 mm, and centric relation (CR) slide to an intercuspal position (IP) greater than 5 mm[25]; 4. Presence of severe psychiatric, neurological or motion disorders; 5. Presence of dental pain and unsatisfactory periodontal health; 6. History of head and neck cancer or hormonal problems; 7. Smoking history; 8. Participants who had received or were receiving radiation therapy, those using antidepressants and anticonvulsants, and those who had undergone laryngeal surgery.

### Methods

All selected women were subjected to lifestyle investigation by the application of the Brazilian version of the OHIP-short form (OHIP-14)[21] questionnaire and the Brazilian version of the V-RQOL protocol (QVV). In addition, their self-perception about the importance of the voice was registered[13]. A questionnaire for determination of the presence and severity of TMD was also used[7].

### Presence and severity of TMD

The entire sample was requested to fill out a questionnaire for clinical interview containing personal information (name, age, sex, address and telephone number) and questions about symptoms related to the main symptoms of TMD. This questionnaire[7] was developed based on preexisting questionnaires, and was applied to the women without any examiner interference. The participants answered to 10 questions to assess the severity of their TMD signs and

symptoms :

1. Do you feel any difficulty on opening the mouth?
2. Do you feel any difficulty on moving your jaw sideways?
3. Do you feel any discomfort or muscle pain when you chew?
4. Do you frequently have headaches?
5. Do you feel any pain in your neck and/or shoulders?
6. Do you feel any pain inside your ears or next to them?
7. Do you notice any noise in your TMJ?
8. Do you consider your bite "abnormal"?
9. Do you use only one side of your mouth to chew?
10. Do you feel any pain on your face when you wake up?

Three possibilities of answers were offered: "Yes", "No" or "Sometimes". Each "Yes" received the value 2, each "Sometimes" received the value 1 and the value 0 was given for each "No" answer. Questions 4, 6 and 7 received value 3 when the answer "Yes" was given to bilateral or intense symptoms, 2 if symptoms were unilateral or soft, 1 for "Sometimes" and zero for the answer "No"[7]. The sum of values allowed classifying the individuals regarding TMD severity as follows: sum of values from 0 to 3: absence of TMD; sum of values from 4 to 8: mild TMD, sum of values from 9 to 14: moderate TMD, sum of values from 15 to 23: severe TMD.

### Quality of life evaluation

Quality of life-related issues were investigated by asking the women to fill out questionnaires at their homes in a condition of privacy. The questionnaires were delivered to the research subjects, who were mostly recruited at the Bauru community. The subjects filled out the questionnaires without interference of the examiner, who was though always available to help them with any doubt.

Both the OHIP-14 and the QVV questionnaires were used to measure the quality of life. The OHIP-14[21] contains 14 questions that measure the individual's perception about the impact of their oral conditions on their well-being in the recent months. The results obtained for each question were distributed on a 5-point scoring system (never = 0, almost never = 1, sometimes = 2, almost always = 3 and always = 4). For each of the 7 categories of the questionnaire (functional limitation, physical pain, physical discomfort, physical limitation, psychological limitation, social limitation and incapacity) the mean value assigned for two questions from each categorie was calculated. The final score was calculated by summing the mean values assigned to the questions, totalizing a maximum score of 56 points.

The QVV[13] is a voice-related quality of life protocol with the purpose of understanding how a voice problem can affect one's daily activities. It displays a list of possible voice-related issues, and the individuals can respond how their voice was during the last two weeks (1 = excellent, 2 = very good, 3 = good, 4 = reasonable and 5 = bad), representing the vocal self-perception. The questionnaire contains 10 questions that were answered by the participants.

Among the 10 issues, 6 of them (items 1, 2, 3, 6, 7 and 9) cover the physical and functional domain and 4 of them (items 4, 5, 8 and 10) cover the socio-emotional domain. The scale contains 5 response options that correspond to how much each item is considered a problem by the patient, as follows: 1 = it is not a problem, 2 = it is a small problem, 3 = it is a middle/moderate problem, 4 = it is a major problem, and 5 = it is huge problem. Patients filling out this questionnaire are instructed to answer each question according to the severity of their problem.

According to the authors' proposed calculations, QVV domains can be calculated separately, using the following equations:

Physical functionality: $$100 - \frac{(\text{full score} - 6)}{24} \times 100$$

Socioemotional: $$100 - \frac{(\text{full score} - 4)}{16} \times 100$$

The full score ranges from 0 (zero) to 100. The higher the value, the better the quality of life. For this calculation, the following equation is used:

$$100 - \frac{(\text{fullscore} - 10)}{40} \times 100$$

### Statistical Analysis

The characterization of the sample was done based on the TMD severity, obtained by the clinical interview questionnaire, through percentage.

For characterization of the quality of life aspects, the OHIP-14 and QVV questionnaires were applied and the mean, median, standard deviation, minimum and maximum values as well as the values of the first and third quartile were used.

Spearman's Correlation was used to determine the association between TMD presence and severity with the QVV and OHIP-14 results, adopting 5% of significance level.

## RESULTS

The results of the TMD clinical interview questionnaire showed that 8 (24%) participants were considered TMD-free, 15 (46%) had mild TMD, 6 (18%) had moderate TMD and only 4 (12%) were considered as having severe TMD.

The descriptive QVV, OHIP-14 and vocal self-perception measures for the women participating in the study (Table 1) showed a good quality of life. Table 2 presents the results related to the correlation of TMD and the women's quality of life.

As shown in Table 2, when the OHIP-14 aspects were associated with TMD, all aspects, except functional limitation (p=0.326) and physical limitation (p=0.187), had statistical significance, and there was a trend to positive correlation between TMD and social limitation (p=0.053). For the rest of aspects analyzed, values of mean positive correlations were detected (0.38 to 0.45 range). In addition, there was negative correlation between the TMD and voice-related quality of life on the total score (p=0.007), physical functioning domain (p=0.008), and socio-emotional domain (p=0.017) as this statistically significant correlation. In this way for this study, the higher the TMD reduced voice-related quality of life and oral health care, as well as the opposite. There were statistically significant positive correlation between TMD and vocal self perception (p=0.037), demonstrating

**Table1-** Descriptive results of the QVV, OHIP-14 and vocal self-perception questionnaires for the women participating in the study

| | | Mean ± standard deviation | Median | Minimum | Maximum | 1st quartile | 3rd quartile |
|---|---|---|---|---|---|---|---|
| OHIP - 14 | Functional Limitation | 0.17 (± 0.39) | 0.00 | 0.00 | 1.50 | 0.00 | 0.00 |
| | Physical Pain | 0.64 (± 0.82) | 0.50 | 0.00 | 3.00 | 0.00 | 1,00 |
| | Psychological Discomfort | 0.26 (± 0.56) | 0.00 | 0.00 | 2.00 | 0.00 | 0.00 |
| | Physical Limitation | 0.11 (± 0.32) | 0.00 | 0.00 | 1.50 | 0.00 | 0.00 |
| | Psychological Limitation | 0.26 (± 0.44) | 0.00 | 0.00 | 1.50 | 0.00 | 0.50 |
| | Social Limitation | 0.09 (± 0.32) | 0.00 | 0.00 | 1.50 | 0.00 | 0.00 |
| | Disability | 0.09 (± 0.38) | 0.00 | 0.00 | 2.00 | 0.00 | 0.00 |
| | Total | 3.21 (± 4.54) | 1 | 0.00 | 18.00 | 0.00 | 5 |
| QVV | Physical | 94.68 (± 8.43) | 100.00 | 66.60 | 100.00 | 91.60 | 100.00 |
| | Socio-emotional | 98.67 (± 3.04) | 100.00 | 87.50 | 100.00 | 100.00 | 100.00 |
| | Total | 96.29 (± 5.97) | 100.00 | 77.50 | 100.00 | 92.50 | 100.00 |
| | Vocal self-perception | 2.91 (± 0.77) | 3.00 | 1.00 | 4.00 | 3.00 | 3.00 |

**Table 2 -** Temporomandibular disorders (TMD) presence and severity correlation with the results of QVV, OHIP-14 and vocal self-perception questionnaries

| | | R | p |
|---|---|---|---|
| TMD X OHIP-14 | Functional Limitation | 0.18 | 0.326 |
| | Physical Pain | 0.42 | 0.015 * |
| | Psychological Disconfort | 0.45 | 0.009 * |
| | Physical Limitation | 0.24 | 0.187 |
| | Psychological Limitation | 0.38 | 0.030 * |
| | Social Limitation | 0.34 | 0.053 |
| | Disability | 0.41 | 0.018 * |
| | Total | 0.43 | 0.012 * |
| TMD X QVV | | | |
| | Physical | -0.45 | 0.008 * |
| | Socio-emotional | -0.41 | 0.017 * |
| | Total | -0.46 | 0.007 * |
| TMD X Vocal self-perception | | 0.36 | 0.037 * |

R: Correlation values. *statistically significant at p<0.05.

that the higher the TMD severity, the greater self perception of vocal problems by the women analyzed in this study.

## DISCUSSION

TMD is more common in women aged 20 to 40 years, which may be accompanied by muscle-skeletal tension, decrease of voice quality, as well as oral health problems, with an impact on quality of life. However, the findings in literature about the relationship between these aspects are scarce. Thus, in this study two questionnaires of quality of life relating to speech (QVV) and oral health (OHIP-14) were applied to verify if vocal self perception, those related to life and oral health voice present relationship with the severity of TMD.

The study was carried out in a female population because the literature shows a predominance of TMD symptoms in women from 21 to 40 years of age[17]. It has also been shown that the prevalence of TMD is significantly higher among younger women[1-15], with a female-to-male ratio of 5:1[1-8,10-15,21]. In addition, women are more sensitive to not stimuli pain, aged between 24 to 33 years experience changes in their social role, which provokes physical and mental tensions, which could induce the vices of inappropriate mandible use[20]. Another hypothesis is related to an increased occurrence of ligamentary inertia, due to hormonal changes and the level of stress in women[30]. Some authors believe that women are more concerned with health and thus seek treatment more frequently[23]. However, the participants of this study did not sought for treatment related to the TMD.

The application of the TMD questionnaire found the presence of TMD in 25 out of 33 women (76%), and this occurrence was higher than that of a previous study in which TMD was found in 60.63% of the women studied[28]. With regard to TMD severity, the majority of women presented mild TMD (45.45%), supporting the findings of a previous study[29]. Further research might help elucidating the determinants of this occurrence.

Studies that used of OHIP to investigate the quality of life conditions in individuals with TMD[16-20,22-26] differed from the present study because we used the short version of the questionnaire (OHIP-14). This quality of life protocol has seven dimensions of impact and the results showed statistically significant relationship between TMD and the following aspects: physical pain, psychological discomfort, psychological limitation and disability (p<0.05). These data agree with those of a previous work[25], which showed a relationship between the presence of pain, as

well as the occurrence of two or more TMD problems associated with the quality of life index presented by individuals, and other data where the results of the quality of life are related also to psychosocial axis and the presence of chronic pain[16].

TMD severity was found to be associated with the presence of changes in the vocal quality[27]. Application of the voice-related quality of life questionnaire and the question about self-perception of vocal problems showed statistically significant results regarding the voice-related quality of life in subjects with TMD, most of them presenting mild TMD.

In addition, vocal self-perception showed positive correlation with the presence of TMD, agreeing with previous data that revealed high incidence of oral disturbances in patients with TMD, including problems concerning speech system[12], as well as monotone voice quality, hypernasality, hoarse, rough and breathy voice in TMD individuals[4].

Limitations of the present investigation were not differentiating the various sub-types of TMD and the cross-sectional aspect of the observation. Further studies on this subject are necessary to better understand the exact association between the studied variables.

## CONCLUSION

In the surveyed group of women, there was correlation between TMD and oral health-related quality of life for the following aspects: functional limitation, physical, psychological pain, physical limitation, physical discomfort and disability, as well as on voice-related quality of life questionnaire physical aspects, and socio-emotional, and following with respect to the vocal self perception.

## ACKNOWLEDGEMENTS

This work was financed by the State of São Paulo Research Foundation (FAPESP), process nº. 2008/05373-1.

## REFERENCES

1- Ash MM. Current concepts in aetiology, diagnosis and treatment of TMJ and muscle dysfunction. J Oral Rehabil. 1986;13(1):1-20.
2- Barros V M, Seraidarian PI, Côrtes MI, Paula LV. The impact of orofacial pain on the quality of life of patients with temporomandibular disorder. J Orofacial Pain. 2009;23(1):28-37.
3- Behlau M, Oliveira G, Santos LM, Ricarte A. Validation in Brazil of self- assessment protocols for dysphonia impact. Pró-Fono. 2009;21(4):326-32.
4- Bianchini EM. Relações das disfunções da articulação temporomandibular com a articulação da fala. Rev Dent Press Ortodon Ortop Maxilar. 2000;5(1):51-9.
5- Bianchini EM, Paiva G, Andrade CR. Mandibular movement patterns during speech in subjects with temporomandibular disorders and in asymptomatic individuals. Cranio. 2008;26(1):50-8.
6- Bianchini EM, Paiva G, Andrade CR. Mandibular movements in speech: interference of temporomandibular disorders according to pain indexes. Pró-Fono. 2007;19(1):7-18.
7- Conti PC, Ferreira PM, Pegoraro LF, Conti JV, Salvador MC. A across-sectional study of prevalence and etiology of signs and symptoms of temporomandibular disorders in high school and university students. J Orofac Pain. 1996;10(3):254-62.
8- Conti PC, Pertes RA, Heir GM, Nasri C, Cohen HV, Araújo CR. Orofacial pain: basic mechanisms and implication for successful management. J Appl Oral Sci. 2003;11(1):1-7.
9- Felício CM, Mazzetto MO, Perri Angote Dos Santos C. Masticatory behavior in individuals with temporomandibular disorders. Minerva Stomatol. 2002;51(4):111-20.
10- Felício CM, Melchior Mde O, Ferreira CL, Silva MA. Otologic symptoms of temporomandibular disorder and effect of orofacial myofunctional therapy. Cranio. 2008;26(2):118-25.
11- Felício CM, Melchior Mde O, Silva MA, Celeghini RM. Masticatory performance in adults related to temporomandibular disorder and dental occlusion. Pró-Fono. 2007;19(2):151-8.
12- Felicio CM, Rodrigues da Silva MA, Mazzetto MO, Centola AL. Myofunctional therapy combined with occlusal splint in treatment of temporomandibular joint dysfunction-pain syndrome. Braz Dent J. 1991;2(1):27-33.
13- Gasparini G, Behlau M. Quality of life: validation of the Brazilian version of the Voice-Related Quality of Life Measure (V-RQOL). J Voice. 2009;23(1):76-81.
14- Hogikyan ND, Sethuraman G. Validation of an instrument to measure voice-related quality of life (V-RQOL). J Voice. 1999;13:557-69.
15- Janal MN, Raphael KG, Nayak S, Klausner J. Prevalence of myofascial temporomandibular disorder in US community women. J Oral Rehabil. 2008;35(11):801-9.
16- John MT, Reissmann DR, Schierz O, Wassell RW. Oral health-related quality of life in patients with temporomandibular disorders. J Orofac Pain. 2007;21(1):46-54.
17- Junqueira PS. A importância do trabalho fonoaudiológico integrado a dentistas e psicólogos nas disfunções da articulação temporomandibulares. [Mestrado]. São Paulo: Pontifícia Universidade Católica; 1990.
18- Lavigne GJ, Rompré PH, Montplaisir JY. Sleep bruxism: validity of clinical research diagnostic criteria in a controlled psysomnographic study. J Dent Res. 1996;75(1):546-52
19- Murry T, Medrado R, Hogikyan ND, Aviv JE. The relationship between ratings of voice quality and quality of life measures. J Voice. 2004;18(2):183-92.
20- Nogueira MF. Disfunção da articulação temporomandibular (DTM) e mastigação. Uma relação de causa e efeito. Recife: CEFAC; 2001.
21- Oliveira BH, Nadanovsky P. Psychometric properties of the Brazilian version of the Oral Health Impact Profile-short form. Community Dent Oral Epidemiol. 2005;33(4):307-14.

22- Oliveira W. Disfunções temporomandibulares. São Paulo: Artes Médicas; 2002.
23- Poveda Roda R, Bagan JV, Díaz Fernández JM, Hernández Bazán S, Jiménez Soriano Y. Review of temporomandibular joint pathology. Part I: classification, epidemiology and risk factors. Med Oral Patol Oral Cir Bucal. 2007;12(4):E292-8.
24- Pullinger AG, Seligman DA, Gorbein JA. A multiple logistic regression analysis of the risk and relative odds of temporomandibular disorders as a function of common occlusal features. J Dent Res. 1993;72(6):968-79.
25- Reissmann DR, John MT, Schierz O, Wassell RW. Functional and psychosocial impact related to specific temporomandibular disorder diagnoses. J Dent. 2007;35(8):643-50.
26- Rener-Sitar K, Celebic A, Stipetic J, Marion L, Petricevic N, Zaletel-Kragelj L. Oral health related quality of life in Slovenian patients with craniomandibular disorders. Coll Antropol. 2008;32(2):513-7.
27- Silva AM, Morisso MF, Cielo CA. Relationship between the severity of temporomandibular disorder and voice. Pro Fono. 2007;19(3):279-88.
28- Tomacheski DF, Barboza VL, Fernandes MR, Fernandes F. Disfunção temporomandibular: estudo introdutório visando estruturação de prontuário odontológico. Publ UEPG Ci Biol Saúde. 2004;10(2):17-25.
29- Verri FR, Garcia AR, Zuim PR, Almeida EO, Falcón-Antenucci RM, Shibayama R. Avaliação da qualidade do sono em grupos com diferentes níveis de desordem temporomandibular. Pesqui Bras Odontopediatria Clín Integr. 2008;8(2):165-9.
30- Zanini CF. Os hábitos parafuncionais na disfunção da articulação temporo-mandibular. Porto Alegre: CEFAC; 1999.

# The maturational process of the auditory system in the first year of life characterized by brainstem auditory evoked potentials

**Raquel Beltrão AMORIM[1], Raquel Sampaio AGOSTINHO-PESSE[1], Kátia de Freitas ALVARENGA[2]**

1- BS, Audiologist of the Speech-Language Pathology and Audiology Clinic and Student of the Master's Program in Speech-Language Pathology and Audiology, Bauru School of Dentistry, University of São Paulo, Bauru, SP, Brazil.
2- BS, MSc, PhD, Associate Professor, Department of Speech-Language Pathology and Audiology, Bauru School of Dentistry, University of São Paulo, Bauru, SP, Brazil.

**Corresponding address:** Kátia de Freitas Alvarenga - Faculdade de odontologia de Bauru - Departamento de Fonoaudiologia - Alameda Dr. Octávio Pinheiro Brisolla, 9-75 - Vila Universitária - 17012-901 - Bauru - São Paulo - Brasil - Phone: +55 14 3234-8332 - Fax: +55 14 3235-8464 - e-mail: katialv@fob.usp.br

**Received: October 17, 2009 - Accepted: February 19, 2010**

## ABSTRACT

The study of brainstem auditory evoked potentials (BAEP) allows obtaining the electrophysiological activity generated in the cochlear nerve to the inferior colliculus. In the first months of life, a period of greater neuronal plasticity, important changes are observed in the absolute latency and inter-peak intervals of BAEP, which occur up to the completion of the maturational process, around 18 months of life in full-term newborns, when the response is similar to that of adults. Objective: The goal of this study was to establish normal values of absolute latencies for waves I, III and V and inter-peak intervals I-III, III-V and I-V of the BAEP performed in full-term infants attending the Infant Hearing Health Program of the Speech-Language Pathology and Audiology Course at Bauru School of Dentistry, Brazil, with no risk history for hearing impairment. Material and Methods: The stimulation parameters were: rarefaction click stimulus presented by the 3$^{A}$ insertion phone, intensity of 80 dBnHL and a rate of 21.1 c/s, band-pass filter of 30 and 3,000 Hz and average of 2,000 stimuli. A sample of 86 infants was first divided according to their gestational age in preterm (n=12) and full-term (n=74), and then according to their chronological age in three periods: P1: 0 to 29 days (n=46), P2: 30 days to 5 months 29 days (n=28) and P3: above 6 months (n= 12). Results: The absolute latency of wave I was similar to that of adults, generally in the 1st month of life, demonstrating a complete process maturity of the auditory nerve. For waves III and V, there was a gradual decrease of absolute latencies with age, characterizing the maturation of axons and synaptic mechanisms in the brainstem level. Conclusion: Age proved to be a determining factor in the absolute latency of the BAEP components, especially those generated in the brainstem, in the first year of life.

**Key words:** Auditory brainstem evoked responses. Infant. Neuronal plasticity.

## INTRODUCTION

The research of brainstem auditory evoked potentials (BAEPs) allows obtaining the electrical activity generated in the cochlear nerve up to the brainstem through stimulation, with the recording of five waves. Waves I and II are generated in the cochlear nerve[6], wave III, in the neurons which emerge from the complex of cochlear nuclei[9,10,19], waves IV and V, in the upper lateral lemniscus, the latter followed by a negative contingent termed slow negative 10 (SN10) deriving from the depolarization of the inferior colliculus[7,18].

The auditory system presents maturational and developmental patterns that are reflected in the

possibility of recording the amplitude, measured in micro-volts (Ωv), and the latency, measured in milliseconds (ms), of the auditory evoked potentials (AEP). Electrophysiological studies for the auditory system have demonstrated that the maturation of the structures occurs from the periphery to the core, without following a hierarchical pattern[3,14]. In the first months of life, a period of greater neuronal plasticity, important changes are observed in the absolute latency and inter-peak intervals of BAEP, which occur up to the completion of the maturational process, around 18 months of life, in full-term newborns, when the response is similar to that of adults.

In the clinical practice, BAEP analysis is performed by the latencies of waves I, III and V, and values of inter-peak I-III intervals, which reflects the functional state of the hearing nerve and low region of the brainstem. While III-V reflects the higher and central region, I-V encompasses the structure of both intervals[8].

It is thus possible, through the BAEP research, to evaluate the maturation of the auditory nerve and brainstem, and verify the occurrence of an abnormal development process in preterm newborns or with risk indicators[5,8,16]. Hence, the absolute latency and the inter-peak intervals must be precisely determined for each period of development and according to the evaluation protocol utilized, since the BAEP are exogenous potentials, totally dependent on the characteristics of the stimulus utilized to evoke the response.

This study aimed at characterizing the changes in absolute latencies and inter-peaks of the BAEP generated by click stimulus, in the first year of life of normal infants.

## MATERIAL AND METHODS

After approval by the Ethics Committee of Bauru Dental School, University of São Paulo (Protocol #114/2005), this transversal cohort study analyzed absolute latencies for waves I, III and V and inter-peak intervals I-III, III-V and I-V of the BAEP performed in infants with no risk history for hearing impairment attending the Infant Hearing Health Program of the Speech-Language Pathology and Audiology Course at Bauru School of Dentistry. The normal peripheral hearing was determined by means of a battery of tests, carried out according to the period, including otoacoustic emissions, immittance measures, visual reinforcement audiometry and evaluation of the hearing behavior. A sample of 86 infants was first divided according to their gestational age in preterm (n=12) and full-term (n=74), and then according to their chronological age in three periods: P1: 0 to 29 days (n=46), P2: 30 days to 5 months 29 days (n=28) and P3: above 6 months (n= 12). For BAEP analysis, the rarefaction click stimulus was presented by the 3 Ω insertion phone, with intensity of 80 dBnHL and a presentation rate of 21.1 c/s, with a band-pass filter of 30 and 3,000Hz and average of 2,000 stimuli, Navigator Pro Bio-logic System Corp, version 4.2.0. The BAEP were captured through ECG disposable electrodes (MEDITRACE$_{TM}$ 200),

**Table 1-** Descriptive analysis of absolute latencies for waves I, III and V for groups of full-term and preterm infants, according to the gestational age and period

| GA/P | ABSOLUTE LATENCIES | | | | | |
|---|---|---|---|---|---|---|
| | I | | III | | V | |
| | Mean | SD | Mean | SD | Mean | SD |
| Preterm/P1 | 1.80 | 0.35 | 4.47 | 0.75 | 6.66 | 0.55 |
| Preterm/P2 | 1.60 | 0.20 | 4.26 | 0.18 | 6.32 | 0.24 |
| Preterm/P3 | 1.62 | 0.20 | 4.09 | 0.27 | 6.23 | 0.29 |
| Full term/P1 | 1.67 | 0.28 | 4.49 | 0.47 | 6.77 | 0.54 |
| Full term/P2 | 1.71 | 0.30 | 4.32 | 0.33 | 6.50 | 0.33 |
| Full term/P3 | 1.71 | 0.20 | 3.97 | 0.28 | 6.23 | 0.30 |

GA: gestational age; P: Period; SD: standard deviation

with EEG conductive paste (Tem $20_{TM}$), placed after cleaning the skin with ECG/EEG abrasive gel (NUPREP). The impedance level was kept between 1 and 3 KΩ for the electrodes: the active electrode was positioned in $F_z$, the reference electrodes in $M_1$ and $M_2$, and the ground electrode in Fpz, which allowed the ipsilateral and contralateral recording of the response.

For statistical purposes, a descriptive analysis of the variables was done, and the Student's t-test and two-way analysis were used. A significance level of 5% was set for all analyses.

## RESULTS

The result of the Student's t-test for comparison between the right and left ears of all infants showed no statistically significant difference for either the absolute latencies (wave I: p=0.717; wave III: p=0.883; wave V: p=0.384) or the inter-peak interval values (I-III: p=0.105; III-V: p=0.375; and I-V: p=0.573). Thus, statistical analysis was carried out taking into account the individual and not the ears separately.

Tables 1 and 2 present the descriptive

**Table 2-** Descriptive analysis for inter-peak interval values I-III, III-V and I-V, for the groups of full-term and preterm infants, according to the gestational age and period

| GA/P | INTER-PEAK INTERVAL VALUES | | | | | |
|---|---|---|---|---|---|---|
| | I-III | | III-V | | I- V | |
| | Mean | SD | Mean | SD | Mean | SD |
| Preterm/P1 | 2.66 | 0.43 | 2.19 | 0.22 | 4.85 | 0.28 |
| Preterm/P2 | 2.66 | 0.21 | 2.06 | 0.19 | 4.72 | 0.26 |
| Preterm/P3 | 2.48 | 0.20 | 2.14 | 0.23 | 4.61 | 0.25 |
| Full term/P1 | 2.80 | 0.49 | 2.25 | 0.50 | 5.05 | 0.75 |
| Full term/P2 | 2.61 | 0.18 | 2.18 | 0.20 | 4.79 | 0.26 |
| Full term/P3 | 2.12 | 0.64 | 2.49 | 1.14 | 4.61 | 0.54 |

GA: gestational age; P: Period; SD: standard deviation

**Table 3-** Results of the two-way analysis of variance for comparison of the variables gestational age and period, and their interaction for the absolute latencies and inter-peak intervals

| Wave/Interval | p Value | | |
|---|---|---|---|
| | Gestational age | Period | Interaction |
| I | 0.666 | 0.574 | 0.299 |
| III | 0.925 | 0.008* | 0.720 |
| V | 0.400 | 0.010* | 0.762 |
| I-III | 0.413 | 0.007* | 0.241 |
| III-V | 0.180 | 0.394 | 0.664 |
| I-V | 0.558 | 0.254 | 0.879 |

*p<0.05: statistically significant.

**Table 4-** Results of Tukey's test for absolute latencies of waves III and V and inter-peak I-III interval value

| Wave | P1 (0 to 29 d) | P2 (30 d to 5 m29 d) | P3 (>6 m) |
|---|---|---|---|
| III | 4.48[a] | 4.29[b] | 4.03[c] |
| V | 6.71[a] | 6.41[b] | 6.23[b] |
| I-III | 2.73[a] | 2.63[a] | 2.30[b] |

P: Period; Same letters in the columns indicate no statistically significant difference at 5%; d= days; m= months

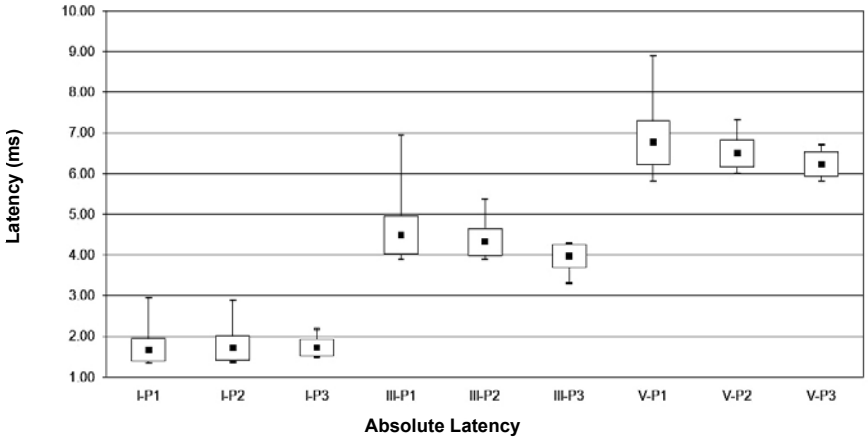


**Figure 1-** Mean, minimum, maximum and standad deviation values of absolute latencies for waves I, III and V, for the full-term infants according to the period

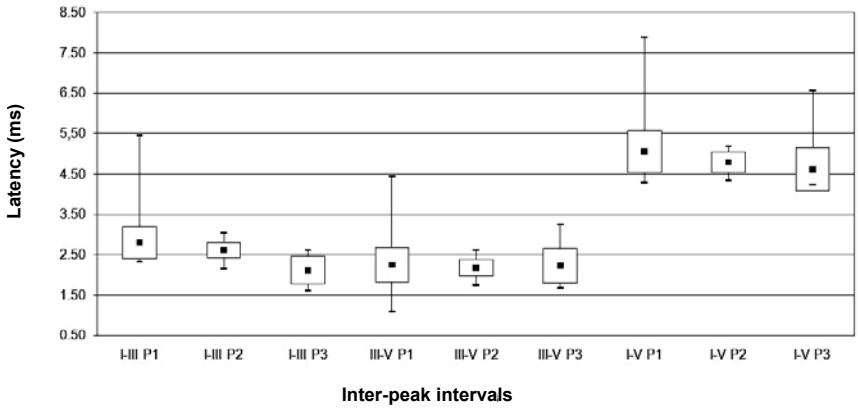


**Figure 2-** Mean, minimum, maximum and standad deviation values of inter-peak I-III, III-V and I-V interval values, for the full-term infants according to the period

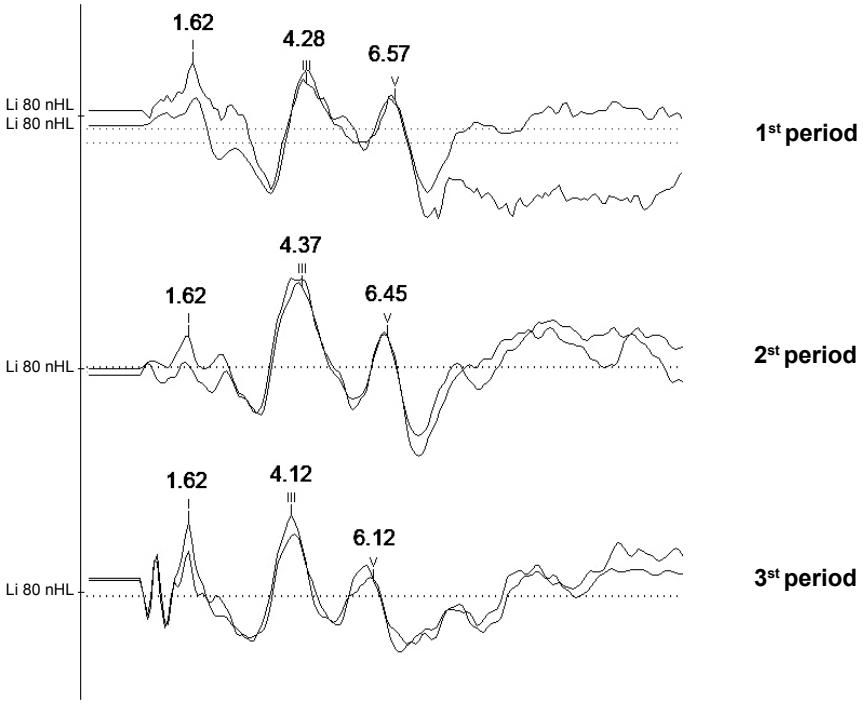


**Figure 3-** Recording of brainstem auditory evoked potentials (BAEP) with the respective latencies for waves I, III and V, of inter-peak interval values I-III, III-V and I-V, for the full-term infants according to the period

analysis (mean and standard deviation) of the absolute latencies for waves I, III and V and values of inter-peak intervals I-III, III-V and I-V, respectively, according to the gestational age and analysed periods.

The results of the two-way analysis of variance for comparison of the variables gestational age, period and their interaction for the absolute latencies of waves I, III and V and inter-peak interval values I-III, III-V and I-V, are presented in Table 3. Table 4 shows the results of Tukey's test for the absolute latencies of waves III and V and inter-peak I-III interval value.

Due to the reduced casuistic of the preterm group, the normality characterization was performed taking into account the full-term group. Figures 1 and 2 present the minimum, maximum, mean and SD values of absolute latencies for waves I, III and V, and inter-peak interval values I-III, III-V and I-V, respectively, obtained in the in full-term infants according to the period. Figure 3 presents the recorded BAEP with the respective absolute latencies for waves I, III and V, and values of inter-peak intervals I-III, III-V and I-V, in the three periods analysed.

## DISCUSSION

In the present study, there was no statistically significant difference between the right and left ears for the absolute latencies and inter-peak values, which indicates that the maturational process occurs in a similar manner in both, with no inter-aural difference, corroborating the data in the literature[1,17].

No difference was seen for the absolute latencies of waves I, III and V, and the inter-peak interval values, when comparing full-term and preterm infants (Table 3). This finding must be analyzed with caution due to the small sample size in the preterm group. However, this similar behavior of absolute latencies and inter-peak values in preterm and term children has been described[4], though it is not consistent with other studies[2,17].

There was no significant difference (p=0.666) for the absolute latency of wave I among periods analyzed in this study (Table 3). The absolute latency of wave I was similar to that of adults (1.67±0.28 ms), already in the first period studied, remaining similar in the subsequent periods, demonstrating that the maturational process of the distal portion of the auditory nerve is practically complete in the first month of live[5,12] (Table 1). Clinically, this is an important information since the delay in the absolute latency of wave I might aid the clinician in determining the presence of alteration in the peripheral function, involving the middle and/or inner ear.

On the other hand, the absolute latencies of waves III and V and inter-peak values I-III, III-V and I-V tended to diminish as the period increased (Tables 1 and 2), with a significant correlation for wave III (p=0.008) in P2 (p=0.015) and P3 (p=0.000), and between P2 and P3 (p=0.032); for wave V (p=0.009) in P2 (p=0.000) and P3 (0.000), and for interval I-III (p=0.006) in P3 (p=0.000), and between P2 and P3 (p=0.004), characterizing the myelinization of axons and maturation of the synaptic mechanisms at the brainstem level[8,11,15]. The absolute latency of wave III showed to be similar to that of adults, in the third period, 4.09±0.27 ms for the preterm group and 3.97±0.28 ms for the full-term group. This result demonstrates that the maturational process in the region of the cochlear nucleus in the lower portion of the brainstem is complete, in the first year of life. However, the lateral lemniscus area, the upper portion of the brainstem, represented by wave V, 6.23±0.29 ms for the preterm group and 6.23±0.30 ms for the full-term group, will keep its development in the second year of life[13,15]. These findings confirm that the maturation is peripheral-central/caudal-rostral, occurring in different speeds in the structures of the brainstem and in different phases of development[5,8,17].

This way, the following values of absolute latencies for each period, expressed in ms, respectively, can be adopted for the analysis of BAEP recording of full-term infants: wave I - 1.67±0.28/1.71±0.30/1.72±0.20; wave III - 4,49±0.47/4,32±0.33/3.97±0.28; wave V - 6.77±0.54/6.50±0.33/6,23±0.30, with a proportional decrease of inter-peak values I-III 2.80±0.49/2.61±0.18/2,12±0.64, III-V

2.25±0.50/2,18±0.20/2.49±1.14 and I-V 5.05±0.75/4.79±0.26/4.61±0.54.

The knowledge of this process is determinant for the speech pathologist to undertake an accurate analysis of the brainstem auditory evoked potential accomplished in full-term infants.

## CONCLUSION

Age was proven to be determinant in the absolute latency and inter-peak interval values of the brainstorm auditory evoked potentials (BAEP) components, especially those generated in the brainstem, within the first year of life.

## REFERENCES

1- Beagley HA, Sheldrake JB. Differences in brainstem response latency with age and sex. Br J Audiol. 1979;12(3):69-77.

2- Chiang MC, Chou YH, Wang PJ. Auditory brainstem evoked potentials in healthy full-term and pre-term infants. Chang Gung Med J. 2001;24(9):557-62.

3- Eggermont J, Ponton C. Auditory-evoked potential studies of cortical maturation in normal hearing and implanted children: correlations with changes in structure and speech perception. Acta Otolaryngol. 2003;123(2):249-52.

4- Eggermont JJ, Salamy A. Maturational time course for the ABR in preterm and full term infants. Hear Res. 1988;33(1):35-47.

5- Guilhoto L, Quintal V, Costa M. Brainstem auditory evoked response in normal term neonates. Arq Neuropsiquiatr. 2003;61(4):906-8.

6- Hall JW. New handbook for auditory evoked response. Boston: Pearson Allyn & Bacon; 2007.

7- Hashimoto I, Ishiyama Y, Yoshimoto T, Nemoto S. Brain-stem auditory-evoked potentials recorded directly from human brain-stem and thalamus. Brain. 1981;104(Pt 4):841-59.

8- Jiang Z, Brosi D, Wu Y, Wilkinson A. Relative maturation of peripheral and central regions of the human brainstem from preterm to term and the influence of preterm birth. Pediatr Res. 2009;65(6):657-62.

9- Levine RA, Gardner JC, Fullerton BC, Stufflebeam SM, Carlisle EW, Furst M, et al. Effects of multiple sclerosis brainstem lesion on soud lateralization and brainstem auditory evoked potentials. Hear Res. 1993;68(1):73-88.

10- Moller AR, Janetta PJ. Auditory evoked potentials recorded intracranially from the brain in man. Exp Neurol. 1982;78(1):144-57.

11- Moore J, Guan Y, Shi S. Axogenesis in the human fetal auditory system, demonstrated by neurofilament immunohistochemistry. Anat Embryol (Berl). 1997;195(1):15-30.

12- Moore J, Linthicum FJ. The human auditory system: a timeline of development. Int J Audiol. 2007;46(9):460-78.

13- Moore J, Ponton C, Eggermont J, Wu B, Huang J. Perinatal maturation of the auditory brain stem response: changes in path length and conduction velocity. Ear Hear. 1996;17(5):411-8.

14- Ponton C, Eggermont J, Kwong B, Don M. Maturation of human central auditory system activity: evidence from multi-channel evoked potentials. Clin Neurophysiol. 2000;111(2):220-36.

15- Ponton C, Moore J, Eggermont J. Auditory brain stem response generation by parallel pathways: differential maturation of axonal conduction time and synaptic transmission. Ear Hear. 1996;17(5):402-10.

16- Shih L, Cone-Wesson B, Reddix B. Effects of maternal cocaine abuse on the neonatal auditory system. Int J Pediatr Otorhinolaryngol. 1988;15(3):245-51.

17- Sleifer P, Costa S, Cóser P, Goldani M, Dornelles C, Weiss K. Auditory brainstem response in premature and full-term children. Int J Pediatr Otorhinolaryngol. 2007;71(9):1449-56.

18- Sousa LCA, Piza MRT, Alavarenga KF, Cóser PL. Eletrofisiologia da audição e emissões otoacústicas. São Paulo: Tecmed; 2008.

19- Starr A, Hamilton AE. Correlation between confirmed sites of neurological lesions and abnormatilies of far-fiel auditory brainstem response. Electroencephalogr Cli Neurophysiol. 1976;41(6):595-608.

# Reference values of nonword repetition test for Brazilian Portuguese-speaking children

**Simone Rocha de Vasconcellos HAGE[1], Márcia Aparecida GRIVOL[2]**

1- PhD, Speech Language Pathologist, Assistant Professor, Department of Speech-Language Pathology and Audiology, Bauru School of Dentistry, University of São Paulo, Bauru, SP, Brazil.
2- Speech Language Pathologist, Graduate student, Department of Speech-Language Pathology and Audiology, Bauru School of Dentistry, University of São Paulo, Bauru, SP, Brazil.

**Corresponding address:** Simone R.V. Hage - Faculdade de Odontologia de Bauru - USP - Departamento de Fonoaudiologia - Alameda Dr. Octávio Pinheiro Brisolla 9-75 - 17102-901 - Bauru, SP - Brasil - Phone: +55 (14) 3235 8332 - Fax: +55-14-3235-8323 - e-mail: simonehage@usp.br

**Received: October 15, 2009 - Modification: February 19, 2010 - Accepted: March 14, 2010**

## ABSTRACT

Evaluation of the phonological working memory (PWM) through repetition of nonwords can provide important information on the linguistic abilities of children, thus differentiating those with and without communication disorders. Objective: The aim of this study was to obtain reference values in the Nonword Repetition Test (NWRT) in order to investigate the performance of children without language disorders concerning this type of memory. Material and Methods: The study was conducted on 480 normal children of both genders aged 4 years to 8 years and 11 months, attending preschool and elementary school. The NWRT consisted of repeating 20 (children up to 4 years) or 40 (for children aged 5 years or more) invented words with 2 to 5 syllables. The results were subjected to descriptive statistical analysis. Comparison between ages and between the number of syllables in nonwords was performed by the Tukey's multiple-comparison test and one-way analysis of variance, at a significance value of $p<0.05$. Results: There was statistically significant difference ($p<0.05$) in performance between children of different age groups, except between 7- and 8-year-olds. The analysis also showed statistically significant difference ($p<0.05$) in the number of syllables between the different age groups. Conclusions: The reference values obtained indicated an improvement in performance with the increase of age of children, indicating an improvement in the storage of verbal material in the PWM. The performance was worsened with the increase in the number of syllables in words, demonstrating that the greater the number of syllables, the greater is the difficulty in storing verbal material.

**Key words**: Memory. Language development. Speech-language pathology.

## INTRODUCTION

The Psycholinguistic Model (PLM) has decisively influenced the way to assess and treat language disorders in the last decade[8]. This model has been proven efficient because it explains how human beings process information coming to their senses, access the words stored in their lexicon and use the mental representations that encode information, thus understanding the nature of language disorders[6,9,19,23]. This model considers all processes involved in the act of communicating, from the primary level, involving the input and output of verbal information, up to the third, which corresponds to the level of cognitive operations of more complex language[17].

These processes include the working memory, which plays a significant role in the maintenance of thinking and learning, verbal comprehension and lexicon access[11,14,16]. It is a system for processing and storing information on a short-term basis, organized into four components, namely the central executive, two work subsystems - the phonological and visuospatial loop -, and the episodic buffer[2,5,8]. The phonological loop stores and manipulates material based on speech

and has two components: the phonological storage, which receives information through direct (auditory presentation) and indirect ways (visual presentation); and the reverberation process or subvocal test, which occurs serially in real time and acts to restrain the natural decay of phonological storage. One of the primary functions of the phonological loop or phonological working memory (PWM) is to store unfamiliar sound patterns, until a record of more permanent memory becomes consistent[1,4].

The PWM has a fundamental role in acquiring language skills in children[8,13] and its deficit has been suggested as the origin of linguistic difficulties in children with specific language impairment[1,15,16,18,24,25].

In the clinical context, the PWM is evaluated by two procedures: digit span (repeating sequences of numbers in direct and inverse order) and repetition of words or nonwords (NW). The repetition of NW is indicated as a more reliable test for the PWM, because the verbal material input is unknown and hence not subject to lexical influences[3,4,10,21].

Thus, considering the lack of instruments based on the Portuguese language for assessment of the PWM, the objective of this study was to obtain reference values for the Nonword Repetition Test (NWRT), investigating if there are differences in the performance of children without language disorders in different age groups, as well as if the increase of syllables of nonwords impairs their repetition.

## MATERIAL AND METHODS

The study was conducted on 537 children aged 4 years to 8 years and 11 months, of both genders, being 274 girls and 263 boys. Fifty-seven children were excluded due to the detection of problems in oral or written communication during sample selection. Thus, the study involved a final sample of 480 children, 231 boys and 249 girls, attending preschools and elementary schools in the São Paulo state countryside, according to the following inclusion criteria: no history of deficits in oral and written language, as reported on interviews with parents and teachers, who answered a questionnaire containing questions to check if the child had communication, hearing or school disturbances; phonological system compatible with chronological age, as assessed by the Task of Phonology of the Test of Children Language (ABFW)[26]; and, for children in the literacy process, punctuation appropriate to the age and schooling on the subtest of reading of the TDE - School Performance Test [22]. Informed written consent approved by the local Institutional Review Board was obtained from patients regarding the specific procedure and the use of their data for research purposes.

For the NWRT[12] (Appendix 1), all 480 boys and girls enrolled in the study were asked to repeat either 20 (children up to 4 years) or 40 (for children aged 5 years or more) invented words with 2 to 5 syllables. The NWRT was created based on the phonological structure of Portuguese language spoken in Brazil. It is divided in two parts, the first for children aged 3 and 4 years, consisting of 20 invented words with Portuguese phonemes, and the second for individuals above 5 years of age, consisting of 40 invented words with Portuguese phonemes, both containing sequences of 2 to 5 syllables. All invented words were paroxytone, because most words in Portuguese are also paroxytone, and were prepared containing different orders of the following phonemes: 6 occlusive (/ p /, / t /, / k, / b /, / d /, / g /), 3 nasal (/ m /, / n /, / ŋ /), 6 fricative (/ f /, / v /, / ʃ /, / ç /, s /, / z /) and 2 liquids (/ l /, / R /), as well as 5 closed vow els (/ a /, / e /, / i /, / o /, / u /). The syllabic pattern used for children aged 3 and 4 years was C + V (C = consonant, V = vowel) and V + C; and for those above 5 years the pattern was C + V, V + C, C + V + C, C + C + V. The nonwords were prepared with the aid of combinatorial analysis, and the phonemes were combined in different positions in the nonwords, namely in the beginning, middle and end.

The list of nonwords was applied without visual clues, in the same vocal intensity, by a single examiner. The instructions were clearly provided to enhance the understanding: "I speak and you repeat" or "You speak after me", "Now we are going to play 'follow the leader', the

**Appendix 1**

Test of phonological working memory – Nonwords
Designed by Prof. PhD Simone Hage
Personal information:
Name: _____
Birth date:_____ Age:_____ Educational level /school: _____
Complain: _____
Examiner: _____ Date: _____

NONWORD TEST
Scoring:
**2 points (P) when repeated correctly in the first time**
1 point (P) when repeated correctly in the second time
0 point (P) when unable to repeat in the first two attempts
Observations:
Intonation – all words are paroxytones.
An adequate repetition is considered when emitted in an identical manner as the examiner. However, it may be considered correct in case of replacement of the vowel "e" by "i" in the end of words, or also closed vowels "e, o" by open vowels "é, ó".
If a phonological disorder is observed, the processes should be recorded. In these cases, the replacement or omission of a phoneme during the repetition will not be considered a mistake.
Instructions: "I will say some words that do not exist. You should pay attention because you will repeat as I said. I will say it once and you will repeat it. It may be slightly strange, but it won't take long. Attention, let's go!"
For children aged 3 and 4 years:

| Nonwords | Answer | Points | Nonwords | Answer | Points |
|---|---|---|---|---|---|
| 01. faque | | | 05. patofe | | |
| 02. vano | | | 06. daverra | | |
| 03. tabi | | | 07. fideco | | |
| 04. dalo | | | 08. balico | | |
| 05. sito | | | 10. zupanho | | |
| Partial score (2 syllables) | | | Partial score (3 syllables) | | |
| Nonwords | Answer | Points | Nonwords | Answer | Points |
| 11. patifevo | | | 16. polanhosaba | | |
| 12. bacuvipe | | | 17. guimalebiza | | |
| 13. farrebitu | | | 18. verripimeno | | |
| 14. valonigo | | | 19. patofelica | | |
| 15. laboquefu | | | 20. bozicalode | | |
| Partial score (4 syllables) | | | Partial score (5 syllables) | | |

TOTAL
Above 5 years of age:

| Nonwords | Answer | Points | Nonwords | Answer | Points |
|---|---|---|---|---|---|
| 01. toli | | | 11. rossola | | |
| 02. erba | | | 12. porquijo | | |
| 03. guchi | | | 13. deitiva | | |
| 04. deico | | | 14. querrefo | | |
| 05. binha | | | 15. senuno | | |
| 06. ruris | | | 16. cholapes | | |
| 07. chefu | | | 17. gromelha | | |
| 08. prido | | | 18. vunhébe | | |
| 09. zuga | | | 19. churéga | | |
| 10. ratros | | | 20. jutrisbe | | |
| Partial score (2 syllables) | | | Partial score (3 syllables) | | |
| Nonwords | Answer | Points | Nonwords | Answer | Points |
| 21. munhocossi | | | 31. pedalhofame | | |
| 22. ritossila | | | 32. islogaguta | | |
| 23. merbufita | | | 33. ribomaniga | | |
| 24. feituninha | | | 34. duvoupilhepo | | |
| 25. zojilibo | | | 35. chotinecapu | | |
| 26. lusvanicha | | | 36. zanovelopus | | |
| 27. diruzeto | | | 37. dilepazina | | |
| 28. plesmizigo | | | 38. bitrujalico | | |
| 29. guilheravi | | | 39. sujemitóssa | | |
| 30. brapitelo | | | 40. flesbaroguido | | |
| Partial score (4 syllables) | | | Partial score (5 syllables) | | |

TOTAL

**Table 1-** Descriptive measures of the performance of children in the Nonword Repetition Test, considering the total scores obtained

| Age | Subjects | Mean | Median | Minimum | Maximum | Lowest quartile | Superior quartile | Standard deviation |
|---|---|---|---|---|---|---|---|---|
| 4 years old | 106 | 34 [A] | 34 | 22 | 40 | 31 | 37 | 3.89 |
| 5 years old | 94 | 58 [B] | 58 | 37 | 80 | 52 | 67 | 9.28 |
| 6 years old | 80 | 68 [C] | 70 | 47 | 79 | 65 | 73 | 7.93 |
| 7 years old | 117 | 74 [D] | 74 | 60 | 80 | 72 | 76 | 4.05 |
| 8 years old | 83 | 74 [D] | 76 | 61 | 80 | 72 | 78 | 4.62 |

* Ages with the same letter in the mean are not statistically different.

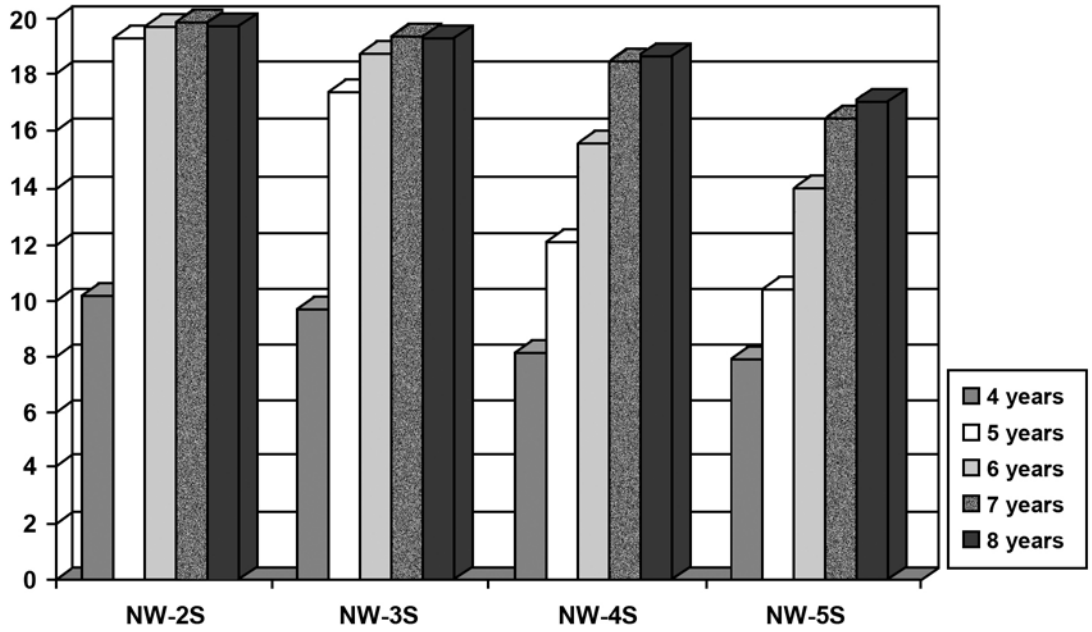


**Figure 1-** Descriptive measures of the performance of children aged 4 to 8 years according to the variable number of syllables. Legend: NW- nonword; S- syllable

leader will speak words that do not exist and you will repeat them". The child was scored 2 (two) points when the nonwords were repeated correctly in the first time, 1 (one) point when they were repeated correctly in the second time, and 0 (zero) point when the child was unable to repeat the nonwords correctly in two attempts.

The results were subjected to descriptive statistical analysis. Comparison between ages and between the number of syllables in nonwords was performed by the Tukey's multiple-comparisons test and one-way analysis of variance, at a significance value of $p<0.05$.

## RESULTS

The results showed statistically significant difference in performance between children of different age groups, except between seven and eight years (4 years <5 years <6 years < 7 years = 8 years).

The results showed that the performance was statistically different depending on the number of syllables of nonwords ($F=206.1$, $p<0.001$). The greater the number of syllables in nonwords, the worse was the children's performance in their repetition.

## DISCUSSION

The achievement of reference values for national evaluation tools is fundamental for the advancement of research in Brazil, particularly in the area of language, since the culture and language structure are important variables when testing cognitive and linguistic abilities[20].

The instrument of this study was designed in accordance with the structure of the Brazilian language spoken in Brazil in order to obtain indices that can be used as reference for the evaluation of children with language problems, since lexical, syntactic and phonological difficulties have been related to deficits in PWM[1,16,25]. The PWM formed the theoretical basis for construction of this instrument because it allows the establishment of hypotheses on the mechanisms underlying the development of language - both in normal and pathological operations - and proposes strategies for the assessment and intervention that consider the various cognitive processes underlying the processing of linguistic information, such as PWM[6,9,19,23].

The choice of tests involving the repetition of nonwords was based on studies[3,4,10,21] that reported that the skills of PWM are more reliably assessed by repetition of this index, because the verbal material presented is not subject to lexical influences. The repetition of nonwords by children requires a connection between their system of perceptual analysis and phonological planning, and the perceptual analysis provides the sequence of phonemes that cannot be generated in the lexicon[10].

The descriptive measures obtained in this study showed that, with the increase in age, children were more efficient in the accomplishment of NWRT, with progressive scores in the median and minimum value (Table 1). There was statistically significant difference between the performances of children of different age groups, except between 7- and 8-year-olds (Table 1), although the performance of eight-year-old children was on top of most descriptive measures. The expansion of memory with age is attributed to the increased speed of "subvocal recall" and is well related with the increase in language skills, typical of child development[8,13]. It is necessary to verify the age from which this performance is in decline, because seniors have memory decline, including in the verbal aspect[27].

Regarding the comparison between the number of syllables in nonwords, the results showed statistically significant difference between all of them (two syllables> three syllables> four syllables> five syllables) for the different age groups (Figure 1). The findings are consistent with the study of Santos and Bueno[21] (2003), who found that the extent of nonwords is reflected in the subvocal test component of the MPWM, since the children's performance decreased as the number of syllables of nonwords increased. Thus, the greater the number of syllables, the greater the difficulty in storing verbal material in

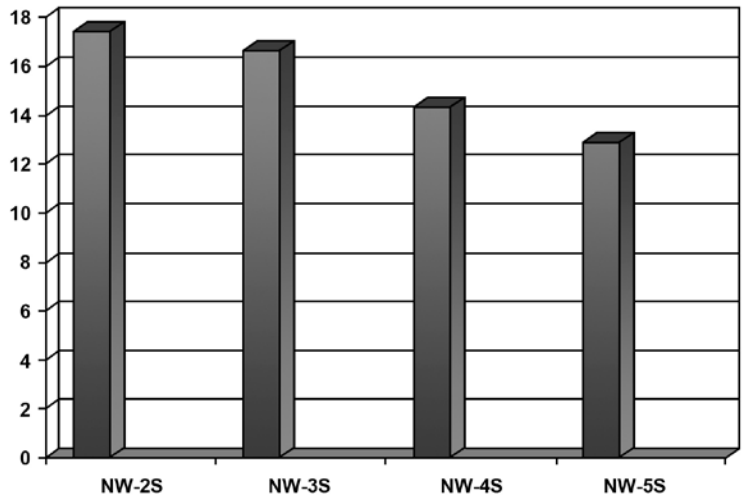


**Figure 2-** Comparative measurements between the numbers of syllables of nonwords.
Legend: NW- nonword; S- syllable

the memory (Figure 2).

## CONCLUSIONS

The reference values obtained indicated that the performance improves with the increase in age of children, indicating an increase in the storage of verbal material in the phonological working memory. There was worsening of the performance with the increase in the number of syllables in nonwords, demonstrating that the difficulty in storing the verbal material increased with the increase of the number of syllables. The results of this study may serve as parameters in the evaluation of children with language disorders and aid in diagnosing the nature of the possible linguistic deficit.

## ACKNOWLEDGEMENTS

The authors acknowledge the financial support from the São Paulo State Research Foundation (Processes #2003/03247-5, 2006/03097-1).

## REFERENCES

1- Archibald LM, Gathercole SE. Short term and working memory in specific language impairment. Int J Lang Commun Disord. 2006;41(6):675-93.
2- Baddeley A, Hitch GJ. Working memory. In: Bower G, editor. The psychology of learning and motivation. New York: Academic Press; 1974. p. 47-89.
3- Baddeley A. Working memory. Oxford: Oxford University Press, Clarendon Press; 1986.
4- Baddeley A, Gathercole SE, Papagno C. The phonological loops as a language learning device. Psychol Rev. 1998;105(1):158-73.
5- Baddeley A. The episodic buffer: a new component of working memory? Trends Cogn Sci. 2000;4(11):417-23.
6- Baker E, Croot K, McLeod S, Paul R. Psycholinguistic models of speech development and their application to clinical practice. J Speech Lang Hear Res. 2001;44(3):685-702.
7- Cervera-Mérida JF, Ygual-Férnandez A. Intervención logopédica em los transtornos fonológicos desde el paradigma psicolinguístico del processamento del habla. Rev Neurol. 2003;36(1):39-53.
8- Gathercole SE, Baddeley AD. Working memory and language. Hillsdale: Lawrence Erlbaum Associates; 1993.
9- Gillon GT. Follow-up study investigating the benefits of phonological awareness intervention for children with spoken language impairment. Int J Lang Commun Disord. 2002;37(4):381-400.
10- Gonçalvez CS. A interferência da memória de trabalho fonológica no desenvolvimento da linguagem. Rev Fonoaudiologia Brasil. 2002;2(1):10-8.
11- Grieve J. Memória. In: Neuropsicologia em terapia ocupacional: exame da percepção e cognição. 2 ed. São Paulo: Santos; 2005. p. 55-64.
12- Hage SR, Grivol MA. Desempenho de crianças normais falantes do português em prova de memória de trabalho fonológica. Cad Comun Ling. 2009;1(1):11-22.
13- Hulme C, Thomson CM, Lawrence A. Speech rate and development of short–term memory span. J Exp Child Psychol. 1984;38(2):241-53.
14- Lent R. Pessoas com história: as bases neurais da memória e aprendizagem. In: Cem bilhões de neurônios: conceitos fundamentais de neurociência. São Paulo: Atheneu; 2001. p. 587-618.
15- Mainela-Arnold E, Evans JL. Beyond capacity limitations: determinants of word recall performance on verbal working memory span tasks in children with SLI. J Speech Lang Hear Res. 2005;48(4):897-909.
16- Montgomery JW. Working memory and compreension in children with specific language impairment: what we know so far. J Commun Disord. 2003;36(3):221-31.
17- Narbona J, Chevrie-Miller C. Avaliação neuropsicológica. In: Narbona J, Chevrie-Miller (orgs). A linguagem da criança: aspectos normais e patológicos. São Paulo: Artmed; 2005. p. 146-70.
18- Norrelgen F, Lacerda F, Forssberg H. Temporal resolution of auditory perception and verbal working memory in 15 children with language impairment. J Learn Disabil. 2002;35(6):539-45.
19- Pascoe M, Stackhouse J; Wells B. Phonological therapy within a psycholinguistic framework: promoting change in a child with persisting speech difficulties. Int J Lang Commun Disord. 2005;40(2):189-220.
20- Puyuelo M, Rondal JA, Wiig EH. Evaluación del lenguaje. Barcelona: Masson; 2002.
21- Santos FH, Bueno, O. Validation of the Brazilian Children's Test of Pseudoword Repetition in Portuguese speakers aged 4 to 10 years. Braz J Med Biol Res. 2003;36(11):1533-47.
22- Stein LM. Teste de desempenho escolar: manual para aplicação e interpretação. 1.ed. São Paulo: Casa do Psicólogo; 1994.
23- Waters D, Hawkes C, Burnett E. Targeting speech processing strengths to facilitate pronunciation change. Int J Lang Commun Disord. 1998;33:469-74.
24- Weismer SE, Evans J, Hesketh LJ. An examination of verbal working memory capacity in children with specific language impairment. J Speech Lang Hear Res. 1999;42(5):1249-60.
25- Weismer SE, Tomblin JB, Zhang X, Buckwalter P, Chynoweth JG, Jones M. Nonword repetition performance in school-age children with and without language impairment. J Speech Lang Hear Res. 2000;43(4):865-78.
26- Wertzner HF. Prova de fonologia. In: Andrade CR, Befi-Lopes DM, Fernandes FDM; Wertzner, HF. ABFW: teste de linguagem infantil nas áreas de fonologia, vocabulário, fluência e pragmática. São Paulo: Pró-Fono; 2004.
27- Yassuda MS, Batistone SST, Fortes AG, Neri AL. Treino de memória no idoso saudável: benefícios e mecanismos. Psicologia: reflexão e crítica. 2006;19(3):470-81.

# Communicative and psycholinguistic abilities in children with phenylketonuria and congenital hypothyroidism

**Mariana Germano GEJÃO[1], Amanda Tragueta FERREIRA[2], Greyce Kelly SILVA[3], Fernanda da Luz ANASTÁCIO-PESSAN[4], Dionísia Aparecida Cusin LAMÔNICA[5]**

1- MSc, Speech Language Pathologist; Department of Speech-Language Pathology and Audiology, Bauru School of Dentistry, University of São Paulo, Bauru, SP, Brazil.
2- Speech Language Pathologist, Graduate student; Department of Speech-Language Pathology and Audiology, Bauru School of Dentistry, University of São Paulo, Bauru, SP, Brazil.
3- MSc; Speech Language Pathologist; Municipal Public Service, Uru, SP, Brazil.
4- Speech Language Pathologist, Graduate student; Neonatal Screening Laboratory of the Association of Parents and Friends of Special Needs Individuals (APAE), Bauru, SP, Brazil.
5- PhD, Associate Professor; Department of Speech-Language Pathology and Audiology, Bauru School of Dentistry, University of São Paulo, Bauru, SP, Brazil.

**Corresponding address:** Dionísia Aparecida Cusin Lamônica - Via Puccini, 1-16 - Residencial Tívoli I - Bauru - SP - Caixa Postal: 17053-095 - Phone: + 55 14 3235-8232 - e-mail: dionelam@uol.com.br

**Received: February 19, 2010 - Accepted: November 04, 2009**

## ABSTRACT

The Neonatal Screening for Inborn Errors of Metabolism of the Association of Parents and Friends of Special Needs Individuals (APAE) - Bauru, Brazil, was implanted and accredited by the Brazilian Ministry of Health in 1998. It covers about 286 cities of the Bauru region and 420 collection spots. Their activities include screening, diagnosis, treatment and assistance to congenital hypothyroidism (CH) and phenylketonuria (PKU), among others. In 2005, a partnership was established with the Department of Speech-Language Pathology and Audiology, Bauru School of Dentistry, University of São Paulo, Bauru, seeking to characterize and to follow, by means of research studies, the development of the communicative abilities of children with CH and PKU. Objective: The aim of this study was to describe communicative and psycholinguistic abilities in children with CH and PKU. Materials and Methods: Sixty-eight children (25 children aged 1 to 120 months with PKU and 43 children aged 1 to 60 months with CH) participated in the study. The handbooks were analyzed and different instruments were applied (Observation of Communication Behavior, Early Language Milestone Scale, Peabody Picture Vocabulary Test, Gesell & Amatruda's Behavioral Development Scale, Portage Operation Inventory, Language Development Evaluation Scale, Denver Developmental Screening Test, ABFW Child Language Test-phonology and Illinois Test of Psycholinguistic Abilities), according to the children's age group and developmental level. Results: It was observed that the children with PKU and CH at risk for alterations in their developmental abilities (motor, cognitive, linguistic, adaptive and personal-social), mainly in the first years of life. Alterations in the psycholinguistic abilities were also found, mainly after the preschool age. Attention deficits, language and cognitive alterations were more often observed in children with CH, while attention deficits with hyperactivity and alterations in the personal-social, language and motor adaptive abilities were more frequent in children with PKU. Conclusion: CH and PKU can cause communicative and psycholinguistic alterations that compromise the communication and affect the social integration and learning of these individuals, proving the need of having these abilities assisted by a speech and language pathologist.

**Key words:** Phenylketonurias. Congenital hypothyroidism. Communication. Child.

## INTRODUCTION

Neonatal screening program (NSP) is the popular name attributed to the Neonatal Screening for Inborn Errors of Metabolism, which has the objective of detecting early congenital hypothyroidism (CH) and phenylketonuria (PKU), among other alterations that can cause intellectual deficiency[7].

The NSP of the Association of Parents and Friends of Special Needs Individuals/Bauru (APAE-Bauru) was implanted and accredited by Brazilian Ministry of Health in 1998. It covers approximately 286 cities of the Bauru area, totalizing 420 collection spots. Their activities include screening, diagnosis and long-term assistance for CH and PKU. The multidisciplinary team for the assistance to the individuals is composed by a pediatrician, an endocrinologist, a nutritionist, a psychologist, a neurologist, a social assistant, a speech language pathologist and a biochemist. This is a pioneering work in this area because the speech language pathologist is not included in the team of professionals proposed by the Ministry Health. However, studies have shown communicative, psycholinguistic, cognitive, motor and personal-social developmental alterations, even in children with early beginning of treatment[3,5,10,15,18,22,25,27-29].

CH is a systemic metabolic disturbance caused by insufficient production of thyroid hormones due to thyroid gland malformation or alterations in hormonal biosyntheses[20,21]. These hormones have great influence in the central nervous system (CNS) development because the vascularization, myelinization, dendritic trees, synapse formation, neuronal migration and genes expression depend on them[3,22-25].

PKU is an autosomal recessive disorder, resulting from the mutation of the gene located in chromosome 12q22-24.1[6]. PKU is caused by the lack of an enzyme known as phenylalanine hydroxylase. This enzyme is responsible for converting the amino acid phenylalanine to a second amino acid, tyrosine, in the liver[27]. The alterations found in the brain tissue of individuals with PKU are nonspecific and of diffuse nature, and might compromise the CNS maturation, produce flaws in the myelinization, and interfere in the biochemical processes that affect some neurotransmitters.

The objective of this study was to describe communicative and psycholinguistic abilities in children with CH and PKU.

## MATERIAL AND METHODS

After approval by the Research Ethics Committee (Protocol #14/2005) of the Bauru School of Dentistry, University of São Paulo, the parents were asked to sign an informed consent form according to 196/96 Resolution. The study was developed in partnership with one of the six São Paulo State NSP centers, accredited by the Ministry of Health.

The criteria for the participants' eligibility were: having early diagnosis[7] for CH (TSH above 10 μIU/mL and T4 free below 0.75 mg/dL) or for PKU[7,9] (PHE levels above 4 mg/dL); attending periodic follow up according to the national guidelines[1]; not presenting other congenital or acquired alterations apart from those of CH and/or PKU; being aged less than 120 months for PKU and 60 months for CH.

Sixty-eight individuals of both genders aged 1 to 120 months were enrolled, being 25 children in the PKU group (PKUG) and 43 children in the CH group (CHG). The clinical history was collected by review of the medical records. The following evaluation instruments were used according to the age group:

*Early Language Milestone Scale[11] (ELMS): to evaluate the visual, receptive auditory and expressive auditory functions of children under 36 months of age.

*Peabody Picture Vocabulary Test[12] (PPVT): to evaluate the receptive vocabulary of children over 36 months of age.

*Gesell and Amatruda's Behavioral Development Scale[17] (GABDS): to evaluate the adaptive motor, fine motor, gross motor, language and personal-social behavior of children under 72 months of age.

*Portage Operation Inventory[30] (POI): to evaluate the motor, language, cognition, socialization and self-car behavior of children

under 72 months of age.

*Language Development Evaluation Scale[21] (LDES): to evaluate the receptive and expressive language development of children under 83 months of age.

*Denver Developmental Screening Test[13] (DDST-II): to evaluate the fine motor, adaptive motor, gross motor, language and personal-social behavior of children under 72 months of age.

*ABFW Child Language Test-phonology[2] – (ABFW): to evaluate the phonology of children over 36 months of age.

*Illinois Test of Psycholinguistic Abilities[4] (ITPA): to evaluate the 12 psycholinguistic abilities subtests of children over 36 months of age (auditory reception, visual reception, auditory association, visual association, auditory memory, visual memory, auditory closure, visual closure, grammatical closure, verbal expression, manual expression, sounds combination).

*Observation of Communication Behavior (OCB): to evaluate the communicative function, comprehension, dialogue maintenance, symbolic play and time attention in all children of the study.

Descriptive statistical analysis was used in the results obtained for the CHG and PKUG. The Spearman's correlation test was used to determine the correlations among the instruments employed in the study. A significance level of 5% was set for all analyses.

## RESULTS

Figure 1 presents, in percentage, the results of the alterations in the abilities evaluated in the DDST-II of 43 children with CH and 17 children with PKU.

Figure 2 presents, in percentage, the results of the alterations in the abilities evaluated in the POI of 43 children with CH and 17 children with PKU.

Figure 3 presents, in percentage, the results of the alterations in the abilities evaluated in the GABDS of 43 children with CH and 17 children with PKU.

Figure 4 presents, in percentage, the results of the alterations in the abilities evaluated in the ELMS (expressive auditory, receptive auditory and visual) of 35 children with CH and 12 children with PKU; in the LDES of 43 children with CH and 17 with PKU; in the PPVT of 8 children with CH and 13 with PKU; and in the ABFW of 8 children with CH and 13 with PKU.

Figure 5 presents, in percentage, the results of the alterations in the abilities evaluated in the ITPA of 12 children with CH and 15 with PKU.

In the OCB, the CHG and PKUG presented verbal order comprehension, protesting, requesting, offering and informing functions, and symbolic play allowing dialogical activities. In both groups, the children demonstrated difficulty in attention time maintenance (39.5% for CHG and 64% for PKUG). The PKUG also presented

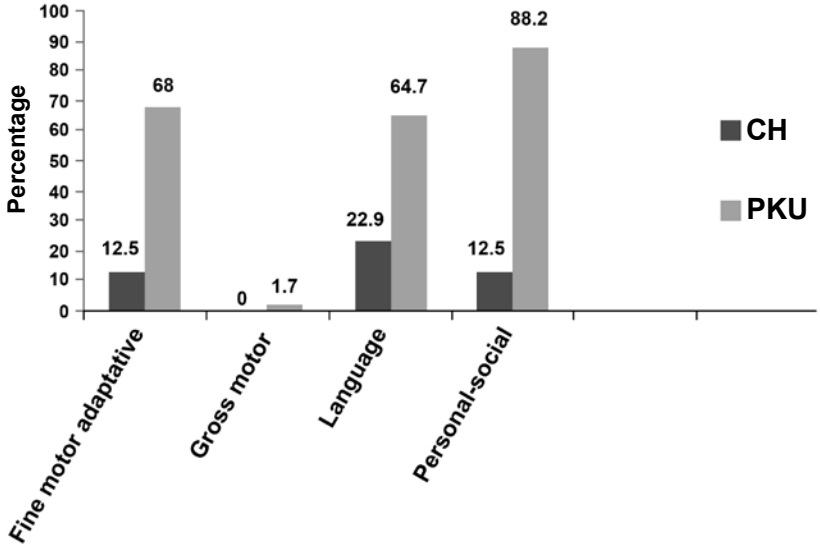


**Figure 1-** Percentage of children with alterations in the abilities of the DDST-II

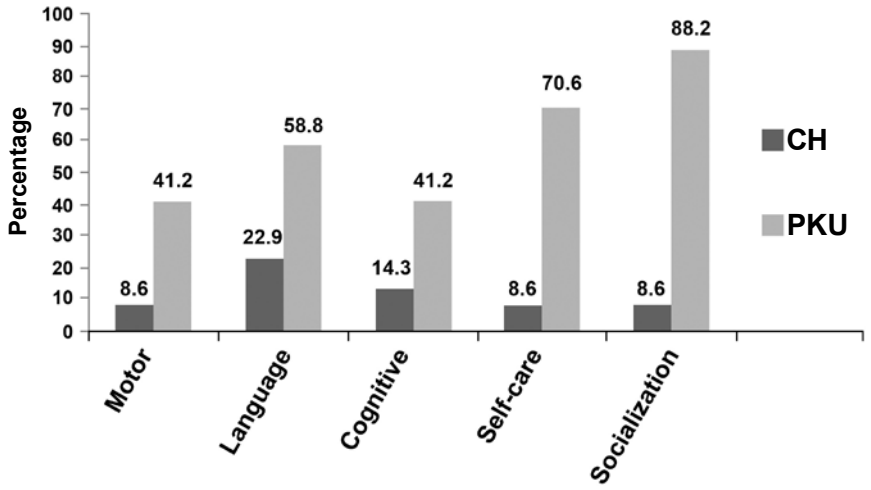


**Figure 2-** Percentage of children with alterations in the abilities of the POI

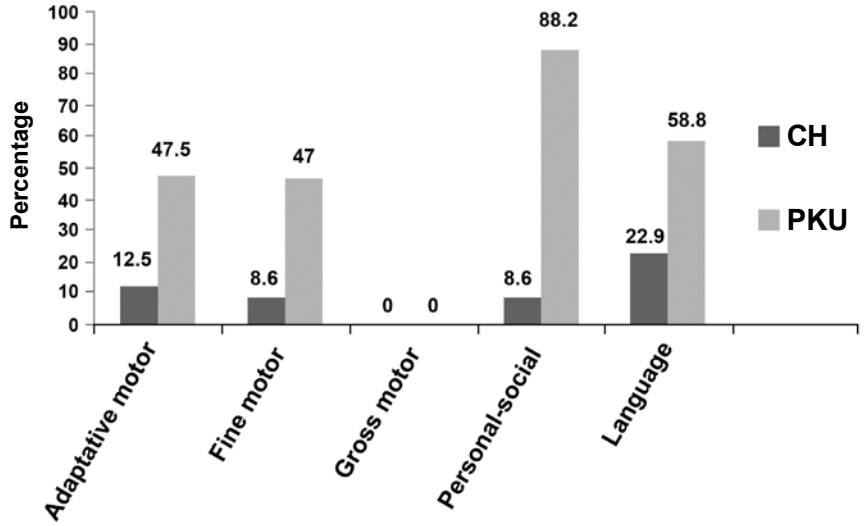


**Figure 3-** Percentage of children with alterations in the abilities of the GABDS

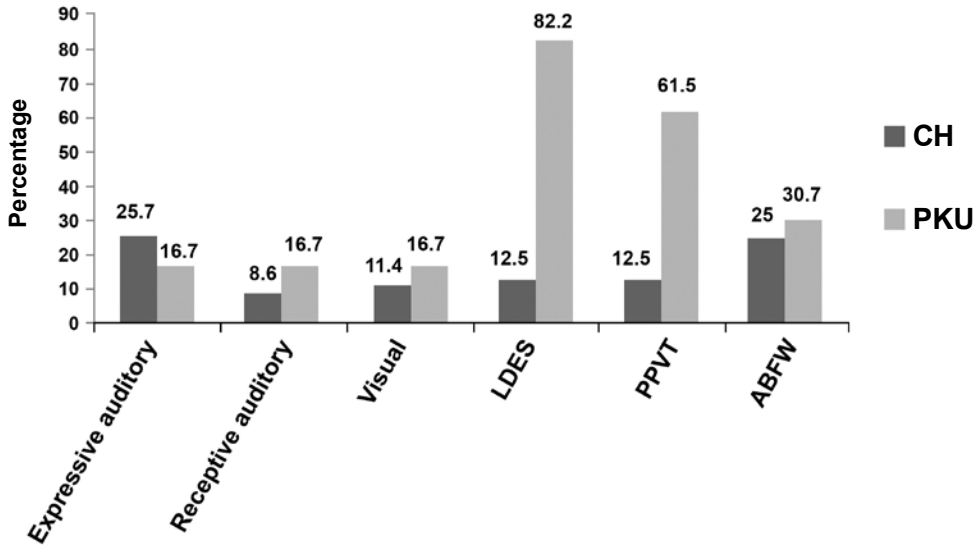


**Figure 4-** Percentage of children with alterations in the abilities of the ELMS, LDES, PPVT and ABFW

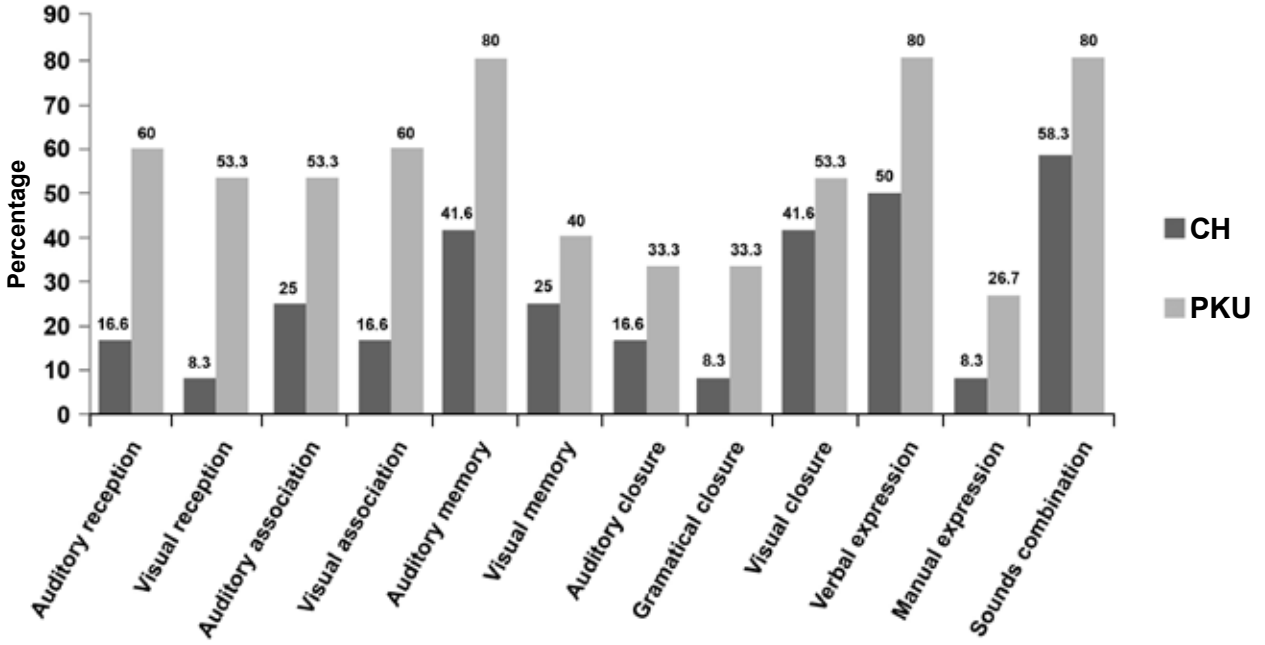


**Figure 5-** Percentage of children with alterations in the abilities of the ITPA

hyperactivity (32%). These data were confirmed by the review of the clinical history.

There was statistically significant correlation among the evaluation instruments, which means that the instruments used in the study had similar capacity to evaluate the same ability.

## DISCUSSION

Analyzing the Figures 1 to 3, the CHG presented worse performance in the language area and PKUG in the personal-social area, followed by the language and motor fine-adaptive areas. The literature refers that delays in the oral language acquisition are frequent in CH[3,15,24] and that individuals with PKU are of risk for alterations in their personal-social[5], psycholinguistic[10,22,29] and fine motor coordination[14] abilities. Few works focused the language specifically[28]. The language is a superior mental function, which depends on the CNS integrity, sensorial, perceptual, cognitive and maturational processes, and the environment influence[15]. The development field influence on the child's general performance has been emphasized. In other words, the language is the mediator for the child planing actions and interacting with the social environment. Therefore, alterations in the receptive or expressive language performance affect other development fields, mainly the adaptive and personal-social skills, interfere in the language development. As far as the expressive aspects are concerned (ELMS, LDES, ABFW, ITPA-verbal expression), CHG and PKUG did not present significant alterations for the phonology, but for the language use.

It has been reported that the longer the period of insufficient thyroid hormone production, the more severe and more extensive the cerebral damages because there will be alterations in the neuronal connections reducing the stimulus transmission capacity[3,18,20,24]. Individuals who cannot maintain the recommended phenylalanine levels can present alterations in the chemical mechanisms of the solid neurotransmitters with pre-frontal and/or left hemisphere dysfunction[29], which affects the general learning.

Comparing the performance of CHG and PKUG in PPVT and LDES (Figure 4), PKUG presented more extensive damages. These data were confirmed by the Spearman's correlation test, proving that children that failed in one of the tests, in a given ability, also failed in the correlated abilities in another test. It means that these instruments were sensitive to detect the appraised ability profile and to confirm the tested hypothesis. It is emphasized that CH treatment is accomplished by hormonal replacement[7,19,23,24],

while PKU treatment involves a phenylalanine-poor diet[3,5-7,10,16,27], making PKU control much more complex, especially in older children.

In ITPA, the most extensive damages were observed in CHG compared to PKUG. In OCB, difficulty was verified in the attention time maintenance for CHG and PKUG, which also presented hyperactivity. It is inferred that the results in ITPA suffered influence of the difficulty in the attention time maintenance. These symptoms have been extensively discussed in the literature[1,3,8,14,18,25-26]. In other words, attention deficit disorders, impulsiveness and hyperactivity are observed in CH[3,20,25] and PKU[1,14,22,28], and brings interference in attention, concentration, memory, analysis, synthesis abilities and executive functions performance, which depends on the appropriate CNS operation.

In this study, measurement of TSH, T4 and phenylalanine levels was done periodically during the treatment and the values were not correlated. Neurological imaging exams were also performed during the course of the treatment. These analyses are objective of future studies.

With the exposed so far, it is inferred that the treatment accomplished by the SNP in children with CHG and PKUG was efficient for the prevention intellectual deficit, but not for prevention of alterations in the communicative and psycholinguistic abilities that compromise the communication and interfere in the social integration and learning, indicating the need of assistance for these abilities by a speech language pathologist.

## CONCLUSION

In the studied population, children with CH and PKU presented alterations in their developmental abilities (linguistics, personal-social and fine-adaptive motor), mainly in the first years of life. There were significant alterations mainly in the visual and auditory psycholinguistic abilities of school children. The importance of having these children assisted is emphasized, seeking the prevention of communicative and psycholinguistic alterations as well as the increase of their social integration in the family and school environment.

## ACKNOWLEDGEMENTS

The authors thank the Neonatal Screening Laboratory of the Association of Parents and Friends of Special Needs Individuals of Bauru, SP, for the partnership in the study of the development of children with CH and PKU. This work was supported by FAPESP (Grant# 2007/00130-0) and CNPq (Grant# 473570/2006-8 and 308121/2006-6).

## REFERENCES

1- Anderson PJ, Wood SJ, Francis DE, Coleman L, Anderson V, Boneh A. Are neuropsychological impairments in children with early-treated phenylketonuria (PKU) related to white matter abnormalities or elevated phenylalanine levels? Dev Neuropsychol. 2007;32(2):645-8.

2- Andrade RF, Béfi-Lopes DM, Fernandes FD, Wertzner HF. ABFW – teste de linguagem infantil nas áreas de fonologia, vocabulário, fluência e pragmática. São Paulo: Pró-Fono; 2000.

3- Bargagna S, Astrea G, Perelli V, Rafaneli V. Neuropsychiatric outcome in patients with congenital hypothyroidism precautiously treated: risk factors analysis in a grup of patients from Tuscany. Minerva Pediatr. 2006;58(3):279-87.

4- Bogossian MA. Teste de Illinois de habilidades psicolingüística: crítica do modelo mediacional e de diversos aspectos da validade do instrumento [Doutorado]. Rio de Janeiro: Fundação Getúlio Vargas; 1984.

5- Bosh AM, Tybout W, van Spronsen FJ, De Valk HW, Wijburg FA, Grootenhuis MA. The course of life and quality of life of early and continuously treated Dutch patients with phenylketonuria. J Inherit Metab Dis. 2007;30(1):29-34.

6- Brandalize SR, Czeresnia D. Avaliação do programa de prevenção e promoção da saúde de fenilcetonúricos. Rev Saúde Pública. 2004;38(2):300-6.

7- Brasil. Ministério da Saúde. Manual de normas técnicas e rotinas operacionais do programa nacional de triagem neonatal. Brasília: Ministério da Saúde; 2002. [Cited at 2006 Jul 25]. Available from: http://dtr2001.saude.gov.br/sas/dsra/MANUAL%202002%200456%20Neo%20Natal-%2006.JUN02.pdf

8- Brumm VL, Azen C, Moats RA, Stern AM, Broomand C, Nelson MD, et al. Neuropsychological outcome of subjects participating in the PKU adult collaborative study: a preliminary review. J Inherit Metab Dis. 2004;27:549-66.

9- Campos MV, Campos B. Disfunção tireoidiana no recém-nascido. Acta Med Port. 2003;16:348-50.

10- Channon S, Goodman G, Zlotowitz S, Mockler C, Lee PJ. Effects of dietary management of phenylketonuria on long-term cognitive outcome. Arch Dis Child. 2007;92(3):213-8.

11- Coplan J. Early language milestone scale. 2nd ed. Austin: PRO-ED; 1993.

12- Dunn LM, Padilla ER, Lugo DE, Dunn LM. Test de vocabulário en imágenes Peabody: adaptación hispanoamericana. Circle Pines: American Guindance Service; 1986.

13- Frankenburg WK, Dodds J, Archer P, Shapiro H, Bresnick B. The Denver II: a major revision and restandardization of the Denver Developmental Screening Test. Pediatrics. 1992;89(1):91-7.

14- Gassió R, Artuch R, Vilaseca MA, Fusté E, Colome R, Campistol J. Cognitive functions and the antioxidant system in phenylketonuria patients. Neuropsychology. 2008;22(4):426-31.

15- Gejão MG, Lamônica DAC. Habilidades do desenvolvimento em crianças com hipotireoidismo congênito: enfoque na comunicação. Pró-Fono. 2008;20(1):25-30.

16- Giovannini M, Verduci E, Salvatici E, Fiori L, Riva E. Phenylketonuria: dietary and therapeutic challenges. J Inherit Metab Dis. 2007;30(2):145-52.
17- Knobloch H, Passamanick B. Gesell e Amatruda: diagnóstico do desenvolvimento. Rio de Janeiro: Atheneu; 1990.
18- Leuzzi V, Pansini M, Sechi E, Chiarrotti F, Carducci C, Levi G, et al. Executive function impairment in early-treated PKU subjects with normal mental development. J Inherit Metab Dis. 2004;27(2):115-25.
19- Manglik AK, Chatterjee N, Ghosh G. Umbilical cord blood TSH levels in term neonatal: a screening tool for congenital hypothyroidism. Indian Pediatr. 2005;42(10):1029-31.
20- Manríquez MO, Nagel LB, Vivanco XW. Evaluación neurológica em pacientes com hipotiroidismo congênito diagnosticado por rastreo neonatal. Rev Chil Pediatr. 1998;69(2):56-9.
21- Menezes, ML. Avaliação do desenvolvimento da linguagem. Rio de Janeiro: Fiocruz; 2004.
22- Moyle JJ, Fox AM, Arthur M, Bynevelt M, Burnett JR. Meta-Analysis of neuropsychological symptoms of adolescents and adults with PKU. Neuropsychol Rev. 2007;17(2):91-101.
23- Nunes MT. Hormônios tireoidianos: mecanismo de ação e importância biológica. Arq Bras Endocrinol Metab. 2003;47(6):639-43.
24- Rovet JF. Congenital hypothyroidism: long-term outcome. Thyroid. 1999;9(7):741-8.
25- Simons WF, Fuggle PW, Grant DB, Smith I. Educational progress, behavior, and motor skills at 10 years in early treated congenital hypothyroidism. Arch Dis Child. 2004;77(3):219-22.
26- Smith ML, Klim P, Hanley WB. Executive function in school-aged children with phenylketonuria. J Dev Physical Disabilities. 2000;12(4):317-32.
27- Van Spronsen FJ, Burgard P. The truth of treating patients with phenylketonuria after childhood: the need for a new guideline. J Inherit Metab Dis. 2008;31(6):673-9.
28- VanZutphen KH, Packman W, Sporri L, Needham MC, Morgan C, Weisiger K, et al. Executive functioning in children and adolescents with phenylketonuria. Clin Genet. 2007;72(1):13-8.
29- Waisbren SE, Noel K, Fahrbach K, Cella C, Frame D, Dorenbaum A, et al. Phenylalanine blood levels and clinical outcomes in phenylketonuria: a systematic literature review and meta-analysis. Mol Genet Metab. 2007;92(1-2):63-70.
30- Williams LC, Aiello AL. O Inventário Portage operacionalizado: intervenção com famílias. In: Tavares J, org. Resiliência e Educação. São Paulo: Cortez; 2001.