

Original Article Artigo Original

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Validation of Content and Reliability of the Protocol for the Evaluation of Acquired Speech Disorders in Individuals with Parkinson's Disease (PADAF)

Validação de conteúdo e confiabilidade do Protocolo de Avaliação dos Distúrbios Adquiridos de Fala em Indivíduos com Doença de Parkinson (PADAF)

Keywords

Dysarthria Speech Parkinson's disease Evaluation Validation Reliability

ABSTRACT

Purpose: To develop a protocol for the evaluation of acquired speech disorders in individuals with Parkinson's disease (PADAF) and to validate its content and determine its inter-judge reliability. Methods: The study was carried out in three stages: in the first one, the protocol was prepared and its content validated through the analysis of seven specialists; in the second, the instrument was applied to 25 individuals with idiopathic Parkinson's disease (PD); in the third and last stage, the inter-judge reliability was determined. Results: The final version of PADAF consisted of 32 items that evaluated breathing, phonation, resonance, articulation, and prosody. It was shown to be valid, with a content validity index (CVI) much higher than that established in the literature, and with perfect agreement in the determination of inter-judge reliability. Conclusion: PADAF for PD individuals was developed and its content was validated, showing perfect instrument reliability.

Descritores

Disartria Fala Doença de Parkinson Avaliação Validação Confiabilidade

RESUMO

Objetivo: Elaborar e realizar a validação de conteúdo, assim como verificar a confiabilidade entre examinador do Protocolo de Avaliação dos Distúrbios Adquiridos de Fala em Indivíduos com Doença de Parkinson (PADAF). Métodos: O estudo foi realizado em três etapas. Na primeira, foi elaborado o protocolo e validado seu conteúdo mediante análise de sete especialistas. Na segunda, aplicou-se o instrumento em 25 indivíduos com doença de Parkinson (DP) idiopática. Na terceira e última etapa, verificou-se a confiabilidade entre-examinador Resultados: A versão final do PADAF foi composta de 32 itens que avaliam a respiração, a fonação, a ressonância, a articulação e a prosódia. Mostrou-se válido, com índice de validade de conteúdo (IVC) bem acima daquele estabelecido na literatura e com perfeita concordância na verificação da confiabilidade entre examinador. Conclusão: O PADAF para indivíduos com DP foi desenvolvido e teve seu conteúdo validado com perfeita confiabilidade do instrumento.

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Received: September 20, 2018

Accepted: January 31, 2019

Study conducted at Departamento de Distúrbios do Movimento, Hospital de Clínicas de Porto Alegre (HCPA), Porto Alegre (RS), Brasil.

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Financial support: nothing to declare.

Conflict of interests: nothing to declare.



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INTRODUCTION

Parkinson's disease (PD) is a chronic neurological disease and has an idiopathic etiology⁽¹⁾. It involves the loss of dopaminergic neurons in the substantia nigra and the deficiency of dopamine in the striatum. It is characterized by the cardinal signs of stiffness, bradykinesia, tremor at rest and postural instability^(2,3).

Studies indicate that up to 90% of people with PD may have speech alterations, showing abnormalities that encompass the system of resonance, respiration, voice, prosody and articulation, believed to originate from damage in motor planning and difficulties in the execution of simultaneous or sequential programs, proper to basal nucleus dysfunctions^(4,5).

The motor control of speech is compromised due to hypokinetic dysarthria, and is characterized by peculiar impairment of verbal expression, reduction of vocal intensity, imprecise articulation, dysprosody, disfluency, less fundamental frequency variation, altered vocal quality such as hoarseness and breathlessness, decreased or increased speed, monotony and vocal⁽⁶⁻¹⁰⁾.

It is important to highlight the value of assessment instruments in patients with neurodegenerative diseases. Few questionnaires are validated to assess speech alterations in PD individuals, and the scarce validated instruments are available only in their original language, mostly English. In several areas of health, the validation of evaluation instruments has been carried out for the purpose of obtaining more accurate and reliable results. Speech therapy can be used to construct better quality instruments⁽¹¹⁻¹³⁾.

In the literature reviewed, we found only one study that was adapted to Portuguese, namely a protocol for evaluating central dysarthria in patients with PD⁽¹⁴⁾, which suggests future studies with modifications in the protocol and its validation in a wider spectrum of subjects and neurological disorders. Other assessments, such as those found in the Mayo Clinic Protocol for Evaluation of Speech Deviations⁽¹⁵⁾ and the Dysarthria Protocol⁽¹⁶⁾, analyze dysarthria, however, without being specifically directed at individuals with PD.

These data suggest the need for studies with diagnostic objectives, considering parameters that can help in the differential diagnosis and therapeutic procedures aimed at the evaluation of these disorders. Thus, a protocol for evaluating acquired speech disorders in individuals with PD (PADAF) was developed, through the compilation and adaptation of the protocols, and with the authorization of the authors of the study on Portuguese adaptation of a protocol for evaluating central dysarthria in PD patients, proposed by Fracassi et al.⁽¹⁴⁾. Therefore, the aim of this study was to develop PADAF and to validate its content and determine its interjudge reliability.

METHODS

The study was approved by the Research Ethics Committee of a referral university hospital in the area under no 150.339, and the subjects who agreed to participate in the study signed an informed consent form.

The protocol was performed according to the following steps: 1. Content validation: The first version of the Protocol was prepared by consulting the literature (14-16) and on the basis of the clinical experience of two specialists in orofacial motricity and voice, both are postdoctoral researchers in the area. The items and subitems were proposed regarding aspects related to the motor bases of speech (respiration, phonation, resonance, articulation and prosody), and the possibilities of responses for each item. Subsequently, this version was submitted to seven invited speech therapists, all specialists with at least three years of experience in speech therapy, and 57.1% of them had more than ten years of experience in the area of dysarthria, as well as more than 30 hours available per week for work in the area. These specialists analyzed the protocol in relation to items, subitems and possible responses. After the analysis, each item was classified as to clarity, according to a four-point Likert scale: 1 = not clear; 2 = little clear; 3 = clear; and 4 = very clear.

If the examiners gave a score of 1 or 2, the items needed to be reformulated. For these items, they could also make suggestions within 30 days, and in this way, the contents could be redone, perfected and submitted for a new evaluation. Also, the examiners were given explanations related to the guidelines established in the protocol. The second version of the protocol was then obtained and sent back to the examiners for analysis, which resulted in its final version.

The examiners still performed a final evaluation (checklist) of PADAF. The variables analyzed were: usefulness, feasibility, objectivity, clinical-scientific aspects, accuracy, clarity, instructional sequence of items, vocabulary, comprehensiveness and organization; they were classified according to a Likert-type scale: 1 = bad, 2 = regular, 3 = good, 4 = very good.

Content validation was performed through the application of the individual content validity index (CVI-I) equation and the total content validity index (CVI-T). CVI-I was calculated by means of the sum of concordance of the items that were marked by "3" or "4" divided by the total number of responses. The calculation of CVI-T was calculated as the sum of all CVI-I values, divided by the number of items of the instruments. Consensually, a minimum agreement of CVI-I and CVI-T of 0.75 was considered acceptable⁽¹⁷⁾.

2. Application of PADAF: The final version of the protocol was applied in 25 patients with a clinical diagnosis of idiopathic PD who underwent clinical follow-up at the Movement Disorders Outpatient Clinic of Hospital Clinicas de Porto Alegre (HCPA), RS, Brazil, from August 2015 to June 2016. The evaluation was filmed and lasted approximately 40 minutes. Patients included in the study had been diagnosed according to the criteria of the London Brain Bank(18) in the H&Y 2 and H&Y 3 stages of the Hoehn & Yahr Scale (1967)⁽¹⁹⁾ and on medication for the disease. Patients excluded from the study were those who showed altered oral comprehension, auditory or visual impairment that prevented the accomplishment of the tasks, those who were off their medication at the time of evaluation, and those who refused to participate in the study. A sample of 21 patients was initially estimated to detect a 0.5 difference in effect size, with a power of 80% and a significance level of 5%⁽²⁰⁾.

3. Interjudge reliability: This compares the results obtained by the examiners for the same individual. The protocol was applied by two examiners on the same day. The examiners did not talk during the tests, because the reliability coefficient could be influenced. The reliability of PADAF was analyzed using kappa agreement analysis, which is an indicator of adjusted agreement that varies from "minus 1" to "plus 1" - where the closer to 1, the better is the level of agreement between observers. Its distribution and the respective levels of interpretation are: <0.00 = no agreement; 0.00 to 0.20 = minimum agreement; 0.21 to 0.40 = reasonable agreement; 0.41 to 0.60 = moderate agreement; 0.61 to 0.80 = substantial agreement; 0.81 to 1.00 = perfect agreement⁽²¹⁾. As a criterion of acceptance, the agreement was established with values higher than 0.61 between the examiners.

The analyses were performed in SPSS (Statistical Package for Social Sciences), version 20.0.

RESULTS

Twenty-five patients with PD (52% females) were evaluated and there were no difficulties regarding the application of the protocol. Data on the demographic and clinical variables of the participants are shown in Table 1.

The final composition of PADAF (Annex 1) had 32 questions that assessed respiration, phonation, resonance, articulation and prosody.

The first content analysis was performed by seven expert examiners, described in Table 2. It was necessary to make a change in item 11 (phonation-loudness) by inserting the sign (+/-), where (+) signified that loudness increased and where (-) indicated that loudness decreased, and in item 19 by adding the description "opening" to jaw (jaw opening).

After these alterations, the protocol was again sent to the examiners for a second analysis, described in Table 3.

The final analysis (checklist) of PADAF is described in Table 4.

Reliability, determined using the kappa value in each motor base of speech, showed perfect agreement, as indicated in Table 5.

DISCUSSION

The purpose of this study was to develop of PADAF and to validate its content and determine its reliability. This evaluation involved the aspects related to the motor bases of speech (respiration, phonation, resonance, articulation and prosody).

The selection of the items that made up the proposal of this protocol was based on the literature related to the area⁽¹⁴⁻¹⁶⁾ and the training and experience of seven speech therapists who analyzed the protocol in relation to the items, subitems and possible answers, classifying them as to their clarity. In addition, they performed a final evaluation (checklist) of PADAF. After the analysis, they classified each according to a four-point Likert-type scale, and finally, CVI was determined.

The validation of the content of an instrument is performed through evaluations by different examiners. The items are analyzed in relation to the content and relevance of the objectives of interest, and the examiners also make suggestions on how much to suppress, add or change in the item⁽²²⁾. Some studies perform content validation only by qualitative analysis based on the evaluation of a committee of specialists^(23,24), while others

Table 1. Demographic and clinical characteristics

Variable	Mean(SD)
Age (years)	63.5 (±13.6)
Schooling (years)	8.3 (±5.3)
Disease duration (years)	8.4 (±5.4)

SD: standard deviation

Table 2. Content Validity Index (CVI) - First analysis

Tuble 2: Content validity mack (CVI)	10	
Items on the evaluation instrument	Examiners in agreement n=7	CVI-I
1. Respiration – rate	7	1
Respiration – rate Respiration – inspiration and expiration	7	1
Respiration – phonotactics and articulatory	7	1
coordination	,	'
4. Phonation – pasty voice	7	1
5. Phonation – hoarse voice	7	1
6. Phonation – breathy voice	7	1
7. Phonation – harsh voice	7	1
8. Phonation – trembling voice	7	1
9. Phonation – wet voice	7	1
10. Phonation – asthenic voice	7	1
11. Phonation – loudness	5	*0.71
12. Phonation – pitch (+/-)	6	0.86
13. Phonation – vocal quality variation	6	0.86
14. Resonance - velar movement	7	1
15. Resonance – nasal emission	6	0.86
16. Resonance - Gutzman test	6	0.86
17. Articulation - lip movements (/i/, /u/, and /pa/)	6	0.86
18. Articulation - tongue (ka/ta)	6	0.86
19. Articulation – jaw opening	5	*0.71
20. Articulation - diadochokinesia	7	0.86
21. Articulation – isolated vowels and words	6	0.86
22. Articulation – plosives	6	0.86
23. Articulation – nasal	6	0.86
24. Articulation – fricatives	6	0.86
25. Articulation – liquid	6	0.86
26. Articulation – vibrants	6	0.86
27. Articulation - consonant meeting	6	0.86
28. Articulation – spontaneous speech	7	1
29. Articulation - speed (+/-)	7	1
30. Prosody – affirmative phrase	7	1
31. Prosody – interrogative phrase	7	1
32. Prosody – exclamative phrase	6	0.86
	CVI -T	0.93

CVI-I: individual content validity index. CVI-T: total content validity index. *: Items with CVI-I values below the expected. n: sample number.

consider quantitative analysis to be of great importance^(25,26). In the present study, both analyses were performed.

During the development of an instrument, one of the points discussed in this evaluation is the number and qualification of the examiners. It is recommended that a minimum of five and a maximum of ten people participate in this process⁽²³⁾. In this decision, the characteristics of the instrument and the training,

Table 3. Content Validity Index (CVI) - Second analysis

Table 3. Content validity index (CVI) – Second a	Halysis	
Items on the evaluation instrument	Examiners in agreement n=7	CVI-I
1. Respiration – rate	7	1
Respiration – inspiration and expiration	7	1
3. Respiration – phonotactics and articulatory coordination	7	1
4. Phonation – pasty voice	7	1
5. Phonation – hoarse voice	7	1
6. Phonation – breathy voice	7	1
7. Phonation – harsh voice	7	1
8. Phonation – trembling voice	7	1
9. Phonation – wet voice	7	1
10. Phonation – asthenic voice	7	1
11. Phonation – loudness	7	1
12. Phonation – pitch (+/-)	6	0.86
13. Phonation – vocal quality variation	6	0.86
14. Resonance – velar movement	7	1
15. Resonance – nasal emission	6	0.86
16. Resonance - Gutzman test	6	0.86
17. Articulation - lip movements (/i/, /u/, and /pa/)	6	0.86
18. Articulation – tongue (ka/ta)	6	0.86
19. Articulation – jaw opening	5	0.86
20. Articulation – diadochokinesia	7	0.86
21. Articulation – isolated vowels and words	6	0.86
22. Articulation – plosives	6	0.86
23. Articulation – nasal	6	0.86
24. Articulation – fricatives	6	0.86
25. Articulation – liquid	6	0.86
26. Articulation – vibrants	6	0.86
27. Articulation – consonant meeting	6	0.86
28. Articulation – spontaneous speech	7	1
29. Articulation – speed (+/-)	7	1
30. Prosody – affirmative phrase	7	1
31. Prosody – interrogative phrase	7	1
32. Prosody – exclamative phrase	6	0.86
	CVI -T	0.91

CVI-I: individual content validity index. CVI-T: total content validity index. n: sample number.

qualification and availability of the necessary professionals need to be taken into account(27,28).

The first proposed version was analyzed by the examiners who suggested some minor adjustments related to the subitems, which helped make the proposal more effective and clearer. After this modification, the second and final version of the instrument was obtained. In addition, the specialists still performed a final evaluation (checklist). According to some authors, the assessment of the instrument by competent examiners experienced in the specific area that is intended to be tested is essential and should be considered in the process of validating the content (24,26).

In this study, therefore, for the validation of the content, besides performing the qualitative validation described above, we validated quantitative content, as done by other authors^(29,30) who report that content validation is essential in the process of developing and adapting assessment instruments. However, it

Table 4. Content Validation Index – final analysis (checklist)

Final opinion - Check	dist	
Requirements of the evaluation instrument	Examiners in agreement n=7	CVI-I
1. Utility	7	1
2. Practicality	7	1
3. Objectivity	6	0.86
4. Clinical-scientific aspect	6	0.86
5. Precision	6	0.86
6. Clarity	6	0.86
7. Instructional sequence of items	7	1
8. Vocabulary	7	1
9. Scope	6	0.86
10. Organization	7	1
	CVI -T	0.93

CVI-I: individual content validity index. CVI-T: total content validity index. n: sample number

Table 5. Kappa value

	Kappa value
	(k)
Respiration	
	0.82
Phonation	
	1.00
Resonance	
	1.00
Articulation	
	0.93
Prosody	
	0.88
Total	
	1.00

has limitations because it is a subjective process. Thus, its use does not eliminate the need to apply additional psychometric measurements.

In this study, CVI was used to measure the percentage of concordance between the examiners who analyzed the first, second and last version of the protocol. CVI is a widely used method in the health area, where it determines the proportion or percentage of evaluators who are in agreement on certain aspects of the assessment instrument and its items. It allows one to initially analyze each item individually and then the instrument as a whole. This method employs a Likert scale with a score of one to four to assess relevance/representativeness. We considered a minimum agreement of CVI-I and CVI-T of 0.75. In the first analysis of the examiners, two subitems showed an agreement below 0.75, and therefore, the protocol was perfected according to the suggestions, and forwarded to the specialists. In its second and last version, indices above the acceptable minimum were obtained. Thus, the protocol proved to be valid in its content, with a percentage of concordance above that established in the literature(17,24,26). Therefore, the content of the instrument proposed

in this study was considered a valid and accurate measure for the 32 items evaluated.

To determine interjudge reliability, the results obtained by the evaluators for the same individual were compared using the kappa coefficient (k), which has been recommended to evaluate concordance measures between evaluators in the area of health. The agreement of the protocol was perfect for all items of the protocol, both for the total kappa value (k = 1.00), and for all of its subitems. The lowest concordance coefficient occurred for the subitem respiration (k = 0.82) and prosody (k = 0.86). We believe that these subitems showed the lowest value because of their complexity. The assessment of respiration may involve many factors, such as morphological (facial type, and nasal, oral and intraoral region) and functional (respiratory mode and type) issues, and it is necessary to understand that what matters is the result of this whole set, requiring a more succinct evaluation. Prosody probably involves several factors, such as syllable accent, intensity variation, duration of word emphasis, variation in syllable duration, and speech rate, and also evaluations of emotional prosody and posture, where a less extensive assessment is needed as well.

Regarding the limitations of this study, only part of the protocol was validated, that is, the content. However, our results indicate that further studies are warranted, which are already under way for construct validation of PADAF in individuals with PD and other neurological diseases. The Protocol is currently being applied in research involving myasthenia gravis and hereditary spastic paraplegia. Based on preliminary results, it has been shown to be a useful, efficient, and important protocol in these diseases and can therefore be used in other populations by validating and adapting the score according to the different clinical manifestations of each disease. For this reason, it is important to highlight that the protocol was elaborated with a greater number of items in the subsystems that are commonly altered in PD, phonation and articulation, which may influence the total protocol score for the severity of the disorder, when applied.

This study is important because validated and reliable protocols that evaluate acquired speech disorders in PD individuals are rare. Comparing it with other protocols, PADAF presents more complete, directive and functional items in each subsystem, as well as presenting, in addition to the total sum of all subsystems, total score in each motor base, being able to identify which of these bases changed more, assisting in differential diagnosis and guiding rehabilitation. In Brazil, there are no validated and reliable instruments in this area. These assessments are essential both for a more accurate diagnosis and for a more effective therapeutic intervention, contributing to the improvement of communication in a general way and positively impacting the quality of life of these patients.

CONCLUSION

PADAF for PD individuals was developed and its content validated, and its reliability was found to be in perfect agreement. Understanding these procedures is essential for researchers and professionals in the area who are concerned with increasingly

using valid and reliable measures and instruments appropriate for a given population.

ACKNOWLEDGEMENTS

The authors thank all patients with PD seen at the Movement Disorders Clinic of HCPA for their collaboration, the invited speech pathologists for their analysis of the Protocol, and FIPE/HCPA (Research Incentive Fund) for its support of this research.

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Author contributions

MP, CRMR, MRO contributed to the research and realization of the article, through data collection, statistical analysis and bibliographic review.

Annex 1. Protocol for the evaluation of acquired speech disorders in individuals with Parkinson's Disease - PADAF

PROTOCOLO DE AVALIAÇÃO DOS DISTÚRBIOS ADQUIRIDOS DE FALA EM PACIENTES COM DOENÇA DE PARKINSON (PADAF)

Nome:		Data da a	valiação:
Identidade:	Tel.:		
Data de nascimento:			
Escolaridade:	Tabagista	: () Sim () Não	
Tempo da doença	H&Y* ECP**	* Sim () Não () Tempo	
UPDRS*** Med	dicação:	() or	n () off
Queixa (tempo):	Comorbi	idades:	
	mulação cerebral profunda. ***Es		nção da Doença de Parkinson.
Sugere-se que o protocolo sej	a filmado para analisar com mais p	recisão os dados.	
1: normal ou ausente	assifique, assinalando um valor de 1 a e; 2: leve; 3: moderado; 4: grave. O sina sinal de "-"deve ser usado para indica	al "+" deve ser utilizado para	indicar aumentado ou agudo,
() Inspiração e expiração força () Coordenação pneumofond		ação audível, estridor inalat Assalto" e contar de 20 a	normalmente) tório e grunhidos ao final da respiração) 30 (verificar excesso ou insuficiência
	Soma dos escore	es do subsistema	
	Respiração: análise indio gra Normal Leve (Moderad Grave (*	au: I (até 3) (4 a 6) do (7 a 9)	
II – FONAÇÃO (emissão da Qual caminho você fez para co Qualidade vocal () Voz pastosa () Voz rouca () Voz soprosa () Voz áspera () Voz trêmula () Voz molhada () Voz astênica () Loudness (+/-) () Pitch (+/-) variação da qualidade vocal () Vogal – curva melódica)		ala espontânea de acordo	o com a resposta à seguinte questão:
	Soma dos escore	es do subsistema	٦
	Fonação: análise indica co		1
	Normal		

III – RESSONÂNCIA

() Movimento velar - /a/ e /â/ (alternadamente) e sustentação da movimentação velar com manutenção do /a/ por 5 segundos.

Leve (11 a 20) Moderado (21 a 30) Grave (31 a 40)

() Emissão nasal: (mamão x papai - pau x mau - Papai pediu pipoca - Amanhã mamãe amassará mamão - Vovó viu a uva - A fita de filó é verde)

() Prova de Gutzman – manter o /i/ prolongado com oclusão intermitente do nariz durante a expiração. Conclusão (adequada/hipernasal ou hiponasal leve, moderada ou acentuada):_____

Soma dos escores do subsistema

Ressonância: análise indica comprometimento de grau:

Normal (até 3) Leve (4 a 6) Moderado (7 a 9) Grave (10 a 12)

IV – ARTICULAÇÃO

Solicitar que o paciente repita (observar se há dificuldade na execução de movimentos, lentidão, fraqueza, imprecisão e incoordenação durante a produção dos fonemas e/ou dificuldade na programação, sequencialização e presença de inúmeras tentativas de posicionar adequadamente os fonemas durante a produção de fala).

()	Movimentos de lábios i/u e pa (com som e sem som, cinco vezes seguidas)
()	Língua (ka/ta) – alternadas (velocidade crescente)
()	Mandíbula – abertura
()	Diadococinesia (repetir separadamente várias vezes /PA/ /TA/ /KA/ e, depois, rapidamente, várias vezes, PATAKA
()	Vogais isoladas e nas palavras (A E I O U - Meia - Pia - Boia - Baú)
()	Plosivas (Dedo - Caco - Batata - Pato - Pipoca)
()	Nasais (Mão - Ninho - Menino - Mãe - Anão)
()	Fricativas (Sapo - Vaso – Fogão - Chave - Gema)
()	Líquidas (Lápis - Milho - Lua - Olho - Bolo)
()	Vibrantes (Carta - Amor - Coração - Árvore - Arara)
()	Encontros consonantais (Prato - Blusa - Flores - Fralda – Cobra – Vidro)
()	Fala espontânea de acordo com a seguinte resposta: Como é a sua rotina? (observar distorções, inteligibilidade)

Soma dos escores do subsistema

Articulação: análise indica comprometimento de grau:

Normal (até 13) Leve (14 a 26) Moderado (27 a 39) Grave (40 a 52)

V - PROSÓDIA

Frases (repetição sem indução de entonação – solicitar ao paciente que leia as frases em tom de afirmação, interrogação e exclamação, sem dar o modelo – verificar ritmo, velocidade, intervalos prolongados, jatos de fala, redução ênfase/entonação). Se o paciente tiver dificuldades, o avaliador poderá dar o modelo, mas não pontuar nem fazer observação, assim como se for analfabeto.

() Choveu muito neste fim de semana (afirmação)
() Ela vai viajar nas férias? (interrogação)
() Hoje é meu dia de sorte! (exclamação)

() Velocidade (+/-) (Leitura do texto "O assalto").

Soma dos escores do subsistema

Prosódia: análise indica comprometimento de grau:

Normal (0 a 3) Leve (4 a 6) Moderado (7 a 9) Grave (10 a 12)

SOMA TOTAL DOS ESCORES DOS SUBSISTEMAS

Análise geral de todos os subsistemas indica comprometimento de distúrbio de fala de grau:

Normal (até 32) Leve (33 a 64) Moderado (65 a 96) Grave (97 a 128)

Texto "O assalto":

O Senhor João Rocha, funcionário da empresa de segurança Vigiar, foi morto segunda-feira à noite, em Porto Alegre. Quatro assaltantes usavam máscaras e um deles portava uma metralhadora importada. Os detetives da polícia estão examinando depoimentos de testemunhas oculares. Uma testemunha disse que ele era muito corajoso, pois atacou o assaltante armado e travou uma tremenda luta.