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Sensitivity and specificity of an acoustic- and perceptual-based tool for assessing motor speech disorders in French: the MonPaGe-screening protocol

Marina Laganaro¹, Cécile Fougeron², Michaela Pernon^{2,3,8}, Nathalie Levêque^{2,4},
Stéphanie Borel⁵, Maryll Fournet^{1,3}, Sabina Catalano Chiuvé³, Ursula Lopez³,
Roland Trouville², Lucie Ménard⁶, Pierre R. Burkhard³, Frédéric Assal³,
Véronique Delvaux⁷

1. *Faculty of Psychology and Educational Sciences, University of Geneva, Switzerland*
2. *Laboratoire de Phonétique et Phonologie, UMR 7018, CNRS/Univ. Sorbonne Nouvelle, Paris, France*
3. *Geneva University Hospitals and Faculty of Medicine, University of Geneva, Switzerland*
4. *APHP, Département de Neurologie, Hôpital Pitié-Salpêtrière, Centre Référent SLA, Paris, France*
5. *APHP Sorbonne Université, GHU Pitié-Salpêtrière, Service d'ORL & Institut du Cerveau et de la Moelle (ICM), Inserm/CNRS, Paris, France*
6. *Laboratoire de Phonétique, Université de Québec à Montréal, Canada*
7. *FNRS &IRSTL (Language Sciences and Technologies Research Institute), University of Mons, Belgium*
8. *Centre de Référence Maladie Rare pour la maladie de Wilson, Service de Neurologie, Fondation Rothschild, Paris.*

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ABSTRACT

To respond to the need of objective screening tools for motor speech disorders (MSD), we present the screening version of a speech assessment protocol (*MonPaGe-2.0.0s*), which is based on semi-automated acoustic and perceptual measures on several speech dimensions in French. We validate the screening tool by testing its sensitivity and specificity and comparing its outcome with external standard assessment tools. The data from 80 patients diagnosed with different types of mild to moderate MSD and 62 healthy test controls were assessed against the normative data obtained on 404 neurotypical speakers, with *Deviance Scores* computed on seven speech dimensions (voice, speech rate, articulation, prosody, pneumophonatory control, diadochokinetic rate, intelligibility) based on acoustic and perceptual measures. A cut-off of the *MonPaGe total deviance score (TotDevS)* > 2 allowed MSD to be diagnosed with *specificity* of 95% and an overall *sensitivity* of 83.8% on all patients pulled, reaching 91% when very mildly impaired patients were excluded. A strong correlation was found between the MonPaGe TotDevS and an external composite perceptual score of MSD provided by six experts. The MonPaGe screening protocol has proven its sensitivity and specificity for diagnosing presence and severity of MSD. Further implementations are needed to complement the characterization of impaired dimensions in order to distinguish subtypes of MSD.

Introduction

Although both impaired language and impaired speech can be associated with a variety of neurodegenerative pathologies and of acquired brain lesions, clinicians have at one's disposal a large choice of language assessment and screening tools in many languages, while very limited tools are available for the assessment of speech¹. In addition, the few assessment tools available for motor speech disorders (MSD) in clinical practice are mostly based on perceptual judgments. To complement the field of perceptual assessment tools for MSD, a consortium made up of researchers and clinicians created the MonPaGe speech screening battery, which is aimed at going beyond the perceptual-only judgments of speech and enabling more objective, mainly acoustic-based, multiparametric quantification of speech disorders. Here, we test whether the screening version of the protocol (MonPaGe-2.0.0s) can be reliably used as a screening tool for the detection of MSD by assessing its specificity and sensitivity on a group of 80 patients with various patterns of MSD and a group of neurotypical test controls against a normative database gathered from 404 French-speaking healthy adults. Crucially, the potential of the protocol to diagnose the presence of MSD in the patient group is tested by including speakers with mild MSD in the patient cohort. The severity score obtained with the screening protocol is also tested against an external assessment of the speech disorder severity provided by a perceptually-based severity assessment made by six expert judges using a tool widely used in French-speaking clinical settings.

Motor speech disorders and their assessment

Following acquired or degenerative brain disorders, speech can be impaired at different degrees of severity and at different levels of the speech production process. Sub-types of MSD have

¹ Unless otherwise specified, voice is considered to be part of speech.

69 been determined at distinct levels. At a first branching, a distinction is made between dysarthria
70 and apraxia of speech (AoS). Dysarthria further groups several syndromes of impaired speech
71 which are the consequences of impaired control of the neuromuscular commands involved in
72 speech production, giving rise to altered voice and/or articulation and/or prosody and/or speech
73 rate (Duffy, 2005; McNeil, Robin, & Schmidt, 2008). Several subtypes of dysarthria are
74 traditionally distinguished (spastic, flaccid, hypokinetic, hyperkinetic, ataxic and mixed,
75 Darley, Aronson, & Brown, 1975; to which unilateral upper motor neuron dysarthria and
76 undetermined dysarthria have been added in more recent years, Duffy, 2005; 2013). AoS on the
77 other hand, refers to MSD that is not attributed to motor execution and control, but to impaired
78 phonetic planning (Blumstein, 1990; Code, 1998; Darley, Aronson & Brown, 1975; Varley &
79 Whiteside, 2001; Ziegler, 2008, 2009; Wertz, LaPointe & Rosenbek, 1984; McNeil et al.,
80 2008). AoS and dysarthria both result in altered articulation, speech rate and prosody, with an
81 overlap in the distortions observed in the dysarthric and AoS speech profiles.

82 Despite the fact that the two types of MSD share speech distortions and need to be
83 differentiated, most assessment tools have been developed separately for dysarthria and for AoS
84 (Dabul, 2000; Feiken, Hofstede, & Jonkers, 2008, Feiken & Jonkers, 2012; Strand, Duffy Clark,
85 & Josephs, 2014). For instance, the Apraxia Battery for Adults (ABA-2, Dabul, 2000) has been
86 conceived to assess AoS and is composed of tasks focusing on articulation, diadochokinetic
87 rate and non-verbal oral movements, but does not assess other parameters such as voice,
88 prosody or intelligibility; similarly, the AoS Rating Scale (ASRS, Strand et al., 2014) has been
89 conceived to specifically identify and describe the perceptual speech characteristics of AoS. On
90 the other hand, assessment tools have been developed specifically for dysarthria (among others,
91 for English: the Frenchay dysarthria assessment, Enderby & Palmer, 2008, which has been
92 adapted in several languages; for German: BoDys, Ziegler, Schölderle, Staiger, & Vogel, 2015;
93 for Dutch: The Radboud Dysarthria Assessment (RDA), Knuijt, Kalf, van Engelen, de Swart,

& Geurts, 2017; for French: The Batterie d'Evaluation Clinique de la Dysarthrie-BECD, Auzou et al, 2006; 2019, the DIADOLAB, Menin-Sicard & Sicard 2020, see also the partial adaptation of the Frenchay by Ghio, Giusti, Blanc, & Pinto, 2019). These tools may be used to assess AoS also, but they do not always include diadochokinetic rate or a comprehensive assessment of articulation.

Whether conceived to assess dysarthria or AoS, speech assessment batteries available in clinical practice mostly rely on perceptual classification of speech and voice disorders, often using a large set of feature descriptors, which are well mastered only by SLP with an expertise in motor speech disorders. There are some well-known downsides to the perceptual description of speech features. First, the reliability and reproducibility of perceptual judgment of speech parameters in MSD have often been questioned (Bunton, Kent, Duffy, Rosenbek, & Kent, 2007; Haley et al., 2012). This is in particular the case when speech is assessed perceptually based on specific dimensions, whereas better interrater agreement is observed on overall severity scores, at least for trained raters (Ziegler, Staiger, Schröderle & Vogel, 2017). Although assessment is based on a balance of subjective and objective evaluation in many domains, subjective evaluation is preponderant in the perceptual assessment of specific speech parameters, as they rely on individual internal representations that are based on the natural variability of these speech parameters and that become somewhat shared with training, but not ascertained.

In the light of the acknowledged limitations of perceptual evaluation, acoustic analyses can be of great help to quantify disordered dimensions in a clinical setting. Acoustic analyses have been implemented in *voice* assessment clinical tools (e.g. AVQI: Maryn, De Bodt, & Roy, 2010; Maryn, De Bodt, Barsties, & Roy, 2014; DSI: Wuyts et al., 2000), but have not systematically been applied to other speech parameters such as speech rate, articulation or prosody, nor generalised to assessment tools used in clinical practice. Hence, although global, holistic

perceptual impression represents the very first information available to the clinician in front of the patients, tools are needed to further guide the assessment, combining functional, perceptual judgments with objective acoustic descriptors of impaired speech parameters. One challenge of this approach is to select a limited number of clinically-relevant descriptors, with no claim to be exhaustive with regards to all potential measures. Indeed, there is a constantly growing amount of measures, acoustic analyses and statistical techniques for characterizing a diversity of aspects of the acoustics of dysarthric speech reported in the scientific literature. However, they are typically very specific, quite sophisticated, not implemented as stand-up procedures in the most popular softwares for voice and speech analysis, such as Praat (Boersma & Weenink, 2018) or the Multi-Dimensional Voice Program (MDVP, from Kay Elemetrics Corporation); in short, they are not readily available for clinical purpose.

The MonPaGe speech assessment protocol and the MonPaGe screening tool

The MonPaGe speech assessment protocol (Fougeron et al. 2016, 2018; Pernon et al., 2020) has been developed (1) to provide a comprehensive speech assessment tool for French-speaking adults and (2) to integrate objective descriptors of potentially deviant speech parameters obtained with acoustic analyses. The MonPaGe protocol integrates the main speech parameters used to assess both AoS and dysarthria in clinical assessment tools. These parameters are quantified either perceptually or acoustically, in a way that can be carried out by SLT or other professionals qualified to assess speech but who do not necessarily have an expert competence in acoustic phonetics or speech engineering. The MonPaGe-2.0.0s presented here is a screening version based on a subset of speech dimensions and parameters from the MonPaGe protocol and aimed at detecting speech patterns that deviate from normal conditions. A speech screening tool should allow to determine whether the patient's speech deviates from typical speech, how much it deviates, and on which speech parameters. This usually corresponds to the first step in

clinical assessment. The second step, namely an in-depth speech assessment contributing to the differential diagnosis of subtypes of MSD, goes beyond the aims of the screening tool.

In order to consider MonPaGe-2.0.0s as a valid screening tool for MSD, the tool should be sensitive enough to detect most patients with a clinical diagnosis of MSD, even with mildly disordered speech (high sensitivity), and at the same time it should be specific enough to avoid over diagnosis, i.e. to avert diagnosing healthy speech as disordered (high specificity). In addition, the assessment given with MonPaGe-2.0.0s should present similar trends to the ones observed with other assessment methods which are already widely used in clinical practice, such as perceptual-only evaluations.

Method

Population

Reference group: healthy speakers

For the reference values on the speech dimensions tested in MonPaGe, a group including 404 neurotypical speakers aged 20-93 years was selected from the MonPaGe_HA (for Healthy Adults) database of spoken French (Fougeron et al., 2018). Participants were recruited in 4 French-speaking locations: Mons, Belgium ($n = 104$); Montréal, Canada ($n = 103$); Paris, France ($n = 101$); Geneva, Switzerland ($n = 97$). All participants spoke French as their primary language (mother tongue) and currently used language. The population recruited for the MonPaGe_HA database is balanced within and between countries on sex (209 women and 195 men) and age groups ([20-39], [40-49], [50-59], [60-74], [75+]) (see Fougeron et al., 2018 for more details).

In order to provide normative data based on the largest possible relevant groups of speakers, the entire database was split into three age groups: younger speakers aged 20 to 59 years old

[20-59] (N = 231, 119 women, mean age = 41.81, SD = 11.44), and older speakers further split into two groups [60-74] (N = 94, 48 women, mean age = 66.01, SD = 4.31) and [75-93] (N = 79, 42 women, mean age = 81.71, SD = 4.30). This choice was driven by a first screening of the data which confirmed findings in the literature showing little evidence of age effects on speech between 20 and 60. The two oldest speakers groups include less speakers but were kept as subgroups in response to clinical needs, where appropriate reference values based on such partition are needed for the elderly.

Participants were recruited in the local communities through public advertisements and among relatives. Distribution of educational level (age of finishing school) was wide and similar across countries, with a median age of finishing school at 22 years old (range: 13 to 48). Speakers in the oldest group (>75) were also screened for language and cognitive deficits (with either the e-GeBAS, Chicherio et al., 2019, or MMSE, Folstein, Folstein, & McHugh, 1975).

Test group 1: Speakers with MSD

80 patients aged 24 to 81 (mean age = 57.1) diagnosed with mild to moderate MSD recruited in France (Pitié-Salpêtrière University Hospital and Lariboisière Hospital, Paris), in French-speaking Switzerland (Geneva University Hospitals) and French-speaking Belgium (Hôpital André Vésale, Charleroi University Hospital) were included for the validation. In order to vary the speech profiles, patients with MSD associated with six different underlying pathologies were included, namely Parkinson disease (PD), Amyotrophic Lateral Sclerosis (ALS), Friedreich ataxia (FA), Kennedy disease (spinal and bulbar muscular atrophy, SBMA), Wilson disease (WD), and post-stroke AoS. The neurological diagnosis was established by neurologists in the hospitals where patients were recruited. To be included in the present study, the patients had to be French native speakers, present mild or moderate acquired or progressive speech

189 difficulty noticed by the patient and a SLP, no or only very mild language impairment. Patients
190 with diagnosed dementia or psychiatric disorders and patients with history of developmental
191 speech and language disorders or hearing impairment were excluded. Table 1 presents the
192 distribution and description of the patients over the six clinical groups with associated basic
193 descriptors.

194 The speech of each of the 80 patients was assessed by six experts (either SLP or clinical
195 phoneticians) on a perceptual basis. A Composite Perceptual (hereby “CP”) score, equivalent
196 to the BECD’s perceptual score (Auzou & Rolland-Monnoury, 2006) was computed based on
197 a perceptual rating of the participant’s speech on five dimensions: voice quality, segmental
198 realization, prosody, intelligibility and naturalness of speech. Each dimension was assessed on
199 a 5-point scale (0: normal to 4: severely impaired) on the recording of a sample of about 2 min.
200 of continuous read speech. The composite perceptual score ranging from 0 to 20 was obtained
201 by summing the five scores.

202
203 *(Table 1 about here)*

204 *Test group 2: healthy control speakers*

205 62 neurotypical French-speaking control participants who were not included in the reference
206 group were recruited in Geneva. They were aged 22 to 88 (mean age = 53.4, 21 men) and had
207 no history of neurological disease or speech and language disorders.

208
209 The study was approved by the local ethic committees at the institutions where the participants
210 were recruited. All participants signed informed consent before participating in the study.

Speech material and speech descriptors

The MonPaGe protocol assesses different speech dimensions based on a variety of tasks (a detailed description of the MonPaGe protocol is provided in Fougeron et al., 2018). Here, a subset of the original speech descriptors was retained for the MonPaGe-2.0.0s screening tool for MSD, providing information on seven dimensions, namely: intelligibility, articulation, pneumophonatory control, voice, prosody, speech rate and diadochokinetic rate. These dimensions have been selected in agreement with the expert SLT associated with the project, based on (i) dimensions that are most used in clinical practice and in other assessment tests and (ii) a balance between the time necessary to assess each dimension and its informativity. Note that the speech descriptors resulting from this procedure, although each relating to a specific dimension, have not the ambition to reflect all the potential disorders on that dimension of speech. Ultimately, they have been selected for their typicality, because they represent the participant's level of competence on that dimension, and/or for their informativity (with respect to the aim of developing a screening tool for MSD), not for the sake of comprehensiveness. For example, prosody is assessed in the extensive MonPaGe protocol via several acoustic and perceptual measures of the distinctive, demarcating and expressive functions of intonation in French, as exhibited by the reading of a tale and the production of selected sentences. In the end, only the ability to express an interrogation via melodic modulations is incorporated in the screening tool. Similarly, pneumophonatory control is indexed by, but by no means reducible to, the indicator of maximum phonation time, which is very commonly used in clinical practice, although its relationship with laryngeal and respiratory physiology is admittedly highly complex (e.g. Solomon et al., 2000).

The seven speech dimensions, the associated speech tasks and the resulting selective descriptors are detailed below. The measurements were carried out off-line using a computerized tool integrating perceptual tasks for scoring intelligibility, coding pseudo-words errors, etc. and a

237 customized Praat script dedicated to human supervision of acoustic measurements.

238 *Intelligibility.* A short intelligibility test was administered in the form of an interactive task
239 between the experimenter and the participant in a face-to-face setting. The participant was asked
240 to instruct the experimenter to place some test-words on a 5x5 grid combining icons of various
241 shapes and colors, using a pre-learned carrier sentence (“Place the word [target_word] on the
242 [color] [shape]” (e.g. ‘Place the word “dog” on the red circle’). The color-shapes combinations
243 were of limited number, while target words were drawn from a database of 437 picturable
244 French words: each target word had one to six competitors (phonological minimal pairs, along
245 five types of phonological contrast, namely place of articulation, voice, manner of articulation,
246 nasality/cluster and vowel quality) within the database and possibly more in the French lexicon.
247 For each session/speaker, a randomization procedure randomly extracted 15 times a target and
248 a specific color-shape combination (corresponding to a particular location on the grid). The
249 participant, but not the experimenter, saw each target picture and corresponding written word
250 on the appropriate grid location on the computer screen and gave instructions to the
251 experimenter, who had to write each target word on a paper grid.

252 The final *intelligibility score* of the participant was computed based on examination of the paper
253 grid, as the number of test words not understood correctly by the experimenter during the
254 interaction (independently of the location on the grid). A rating of 1 was given to incorrect
255 responses, and 0.5 when two responses (including the correct response, e.g. “desert”/”dessert”
256 for “dessert”) were provided.

257 *Articulation.* Articulatory precision was assessed on the production of a set of 50 pseudo-words,
258 covering the articulation of most of the French consonants and vowels as well as consonant
259 clusters.

Pseudoword production was elicited via the bimodal presentation of each target (in an orthographic form on a screen and in an audio form via headphones), to minimise errors due to pseudo-word reading difficulties or to misperception. Regarding the assessment of articulation accuracy, errors on targeted consonants, vowel and syllables were listed based on perceptual assessment following a guided coding procedure for each pseudo-word, where questions were targeted on specific phoneme and on potential types of errors (distortion, substitution, omission, insertion). The computerized tool allowed the raters to play each pseudoword as needed and score targeted phonemes or syllables as correct or incorrect. Overall, 151 targeted phonemes or syllables included in the 50 pseudowords were scored per speaker and a descriptor related to articulation accuracy is expressed in terms of a *number of errors* (from 0 to a maximum of 151). The pseudo-word production is used as a screening for articulatory precision in MonPaGe-2.0 as only the total number of errors is considered and can be easily extracted via the guided computerized scoring tool, but it has been conceived to be used also for further more detailed assessments.

Maximum phonation time (MPT). As a standard measure of pneumo-phonatory control, a descriptor representing maximum phonation time over a sustained vowel was computed. Participants were instructed to produce a sustained vowel /a/ as long as possible after taking a maximal inhalation, at a comfortable pitch and at their habitual loudness. The task was repeated as many times as needed and two productions were recorded. The duration of the two trials of the sustained /a/ production was measured with Praat and the best performance selected as the *maximum phonation time (MPT)*.

Voice. Voice-related measures were based on a sustained production for 2-3 seconds of the vowel /a/ at a comfortable height and loudness and on the reading of a 7 syllables sentence composed of only voiced sounds (“Mélanie vend du lilas” - [melanivãdylila], ‘*Melanie sells*

284 *lilac*'). All measurements were computed with Praat, using a semi-automatic procedure: a
285 customized Praat script was developed to guide the non expert user so that s/he can adjust the
286 relevant settings for optimal results at each stage of the procedure.

287 A first set of standard descriptors of voice quality was taken on the first 2 seconds of the
288 sustained /a/ vowel. These included the two short-term (cycle-to-cycle) measures of vocal
289 instability in terms of frequency and amplitude: jitter and shimmer, respectively. These were
290 computed with Praat as the five-point Period Perturbation Quotient (*a_Jitter-PPQ5*) and the
291 11-point Amplitude Perturbation Quotient (*a_Shimmer-APQ11*). Instability in vocal fold
292 vibration over the whole 2-second window was assessed in terms of f0 standard deviation
293 (*a_SDf0*), potentially allowing for the detection of vocal tremor. Presence of a noise component
294 during the vowel was measured in terms of a harmonic-to-noise ratio (*a_HNR*) and possible
295 dysphonia was also assessed with the smoothed cepstral peak prominence measure (*a_CPPs*)
296 (Hillenbrand et al., 1994).

297 A second set of descriptors related to the speaking voice was measured on the longer continuous
298 read speech sample "Mélanie vend du lilas". On the f0 time series computed over the whole
299 sentence, mean (*speaking_meanf0*) and standard deviation (*speaking_SDf0*) were computed.
300 The smoothed cepstral peak prominence was also taken from the whole voiced sentence
301 (*speaking_CPPs*).

302 *Prosodic contrast.* The production of an assertive vs. interrogative prosodic contrast was tested
303 on a four syllables fully voiced sentence 'Laurie l'a lu' ([loʁilaly], 'Laurie read it'). The
304 sentence was first presented on the screen as a declarative and read by the speaker. Then the
305 speaker was asked to say the same sentence again but asking a question, while the sentence was
306 presented with a question mark.

307 The prosodic contrast between the two modalities was computed in terms of a difference in f0
308 modulation over the sentence. In the interrogative condition marked by a large final rise of f0
309 in French, expected a large delta in f0 range between the first half and the second half of the
310 sentence is expected. This ambitus was expected to be smaller in the assertive condition,
311 although it was not inexistent if the speaker produces a large f0 fall at the end of the assertion,
312 or an initial rise at the beginning of it. Therefore, the prosodic contrast was expressed as the
313 difference between the two modalities in these f0-range deltas between the beginning and the
314 end of the sentence. The recorded sentences were automatically split into two parts, and f0
315 range was computed in semitones for the first and second parts. The descriptor for prosodic
316 contrast achievement was thus computed as:

317 $(f0_{range@end} - f0_{range@begin})_{question} - (f0_{range@end} - f0_{range@begin})_{assertion}$

318 *Speech rate.* Speech rate was assessed on a short sentence reading task. The duration of the
319 sentence “Mélanie vend du lilas” ([melanivãdylila], ‘Melanie sells lilac’) was measured with
320 Praat and a simple measure of speech rate was obtained by dividing the expected number of
321 phonemes (14 phonemes) by this duration.

322 *Diadochokinetic (DDK) rate.* Maximum repetition rates with oral *diadochokinetic* tasks are
323 often used in clinical practice to test the ability to make alternating articulatory movements in
324 quick and accurate succession. Seven items, which vary in terms of phonological complexity,
325 were used here. They included standard sequences used to compute alternative motion rate
326 (AMR) with the repetition of a CV syllable (AMR_{CV}) or a CCV syllable (AMR_{CCV}). Different
327 CV and CCV syllables were used to target alternating movements with different articulators:
328 jaw/lips with /ba/, front part of the tongue with /de/, tongue body with /go/, constrictions with
329 /kla/, and front to back with /tɾa/. Finally, a repetitive sequence /badego/ was used to compute
330 a sequential motion rate (SMR_{CV}). Participants were instructed to produce these sequences in a

continuous manner for at least five second as fast and as accurately as possible.

The number of phonemes produced over an interval of about 4 seconds of continuous repetition was used as an index of DDK rate for AMR_{CV} , AMR_{CCV} , SMR_{CV} . This interval was selected from the onset of the speech waveform and was manually adjusted to the right in order not to cut the last syllable if needed. Also, in order to capture difficulties in the repetition of the same syllables (AMR) vs. the repetition of a sequence of three syllable (SMR) which could be found for speakers with AoS for instance (e.g. Ziegler, 2002), we also computed the difference between the sequential motion rate and the alternative motion rate averaged over all CV sequences: $(SMR_{CV} - AMR_{CV})$.

Procedure and analyses

All the participants underwent the MonPaGe protocol following the standard assessment procedure with speech and speech-language pathologists in clinical settings for the patients and in a standard room at the University for the controls. The MonPaGe protocol was run on a laptop and speech samples were recorded using either a head-mounted or a table microphone depending on the place of recruitment. This variability in recording reflects the variations across clinical settings and has also been introduced in the recordings of the reference group (see Fougeron et al., 2018). The assessment takes about 20 minutes with typical speakers and 30 minutes with patients with moderate MSD (Pernon et al., 2020), and the same duration is necessary for scoring.

Constitution of the norms from the reference healthy speakers group

The productions of the 404 healthy speakers reference group were analyzed in order to obtain normative values. For each descriptor, missing data and extreme values (below and above the 1st/99th percentiles) were first removed. In order to account for sex or age-group effects on the

norms, linear models were first computed in R with Sex (male/female) and Age-group ([20-59], [60-74], [75+]) as between-subject factors and the various descriptors as dependent variables. Whenever an effect of Sex or an interaction with Age-group was found, further analyses were carried out for the male and female speakers separately. Inter-group differences for the Age-group predictor were tested with Tukey post-hoc tests. Results from these statistical analyzes are provided in the supplementary material. Normative values for each descriptor was then computed by subgroups accordingly. Since groups split or merging were done according to the sex or age effects, the number of participants retained for the computation of normative values varied according to the descriptor considered.

Computation of deviance scores

For each descriptor, the speech of the participants in the two test groups (patients and healthy controls) were compared to the normative values of the reference population, according to its sex and age group.

A **deviance score** (*DevS*) spanning from 0 to 4 was defined for each descriptor depending on its distance from the reference value. The standard limit from the normative data was fixed at centile 5 (Brooks, Sherman, Iverson, Slick, & Strauss, 2011) and five degrees of *DevS* are defined, with *DevS* = 0 standing for no-deviant (within normal range) and *DevS* = 4 for excessively deviant, according to the position of the speaker's descriptor value relative to the reference distribution. Cut-off values for defining these deviance degrees are determined according to either the tails of the distribution (c5/c95 and c1/c99, for the *DevS* = 1 and *DevS* = 2 respectively) and further severity, beyond c1/c99, is computed based on the inter-centile distance between c50 and c5/c95, as detailed in Table 2.

(Table 2 about here)

For the dimensions of voice quality and DDK performance, which rely on several descriptors, a composite deviance score was further computed as detailed in Table 3. Finally, each participant's speech is globally characterized by the *MonPaGe total score* (*TotalDevS*, ranging from 0 to 32); which corresponds to the sum of the deviant scores on the seven speech dimensions, where larger scores indicate more deviance in speech.

(Table 3 about here)

The detailed procedures for the assessment of sensitivity, specificity and external validity are described along with the results.

Results

Reference values

The normative values for each descriptor obtained from the 404 healthy speakers reference group are presented in Appendix A (Tables A.I and A.II). Normative values for each descriptor and for each age/sex subgroup are presented in terms of mean, median and standard deviation, except for intelligibility and segmental errors for which median and maximum error rates are provided. Distribution tails of the population's scores are also given with the 1st/99th, 5th/95th and 10th/90th percentiles, in order to provide possible cut-off scores of the normal performance limits. According to the descriptor, the tails of interest for the determination of the cut-off scores are either on the upper or lower sides (e.g. for error values, alteration is to be found in the upper tail while 0 error is normal).

Specificity and sensitivity of MonPaGe

Appendix B and C present the deviant scores per speech dimension and *TotalDevS* for each of the 80 patients and 62 healthy test controls, respectively. The *MonPaGe TotalDevS* ranges from 0 to 12 in the patient group and from 0 to 4 in the healthy test control group.

The cut-off score for a diagnosis of MSD with MonPaGe was defined by plotting sensitivity versus specificity for the few possible cut-off scores (from 1 to the maximum MonPaGe *TotalDevS* in the test control group). The best cut-off score was a **MonPaGe *TotalDevS* > 2** with a *specificity* of 95.2% and *sensitivity* of 83.8%. Figure 1 illustrates how sensitivity varies according to patients' severity as assessed externally with the Composite Perceptual Score. It can be observed that sensitivity raises to 92% when excluding patients with very mild impairments (CP score $\leq 4 / 20$).

(Figure 1 about here)

For the *external validity*, the composite perceptual score of severity was considered the clinical gold standard. Pearson correlation computed between the MonPaGe *TotalDevS* and the external CP score on the 80 patients is $r(80) = .737$, $p < .0001$.

Discussion

The sensitivity and specificity in the diagnosis of MSD of the MonPaGe screening protocol based on a set of semi-automated acoustic measures and on targeted perceptual measures was assessed on the speech from 80 French-speaking patients diagnosed with a diversity of mild to moderate MSD and 62 healthy test controls against the normative data of 404 neurotypical adult speakers. On each of the seven speech dimensions (voice, speech rate, articulation, prosody, pneumophonatory control, diadochokinetic rate, intelligibility) a deviance score (*DevS*) was calculated for each participant with reference to the normative data and an overall deviance

score, the MonPaGe *TotalDevS*, was computed as the sum of all *DevS*. With a cut-off score at MonPaGe *TotalDevS* > 2, the *specificity* of the tool was excellent (95%), and its sensitivity was very high, in particular for the mild to moderate patients (sensitivity > 92% when the patients with very mild MSD are excluded). Along with the strong correlation between the MonPaGe *TotalDevS* and an external composite perceptual assessment of severity obtained from 6 expert judges, the present results indicate that the MonPaGe protocol is a reliable screening tool for assessing the presence of MSD and its severity.

The current study has two crucial features related to the specificities of the analyses in the MonPaGe protocol and to the population involved in the study, which will be discussed in further detail below.

Guided objective analyses of impaired speech

As exposed in the introduction, the available clinical assessment tools for MSD are mostly based on perceptual judgment, which are bound to rely heavily on subjective evaluation. For these reasons the MonPaGe assessment protocol relies mainly on descriptors extracted from the acoustic signal for five out of the seven dimensions (voice, MPT, speech rate, prosodic contrast and DDK rate). For the other two dimensions (intelligibility and articulation accuracy), the descriptors are not objective in the sense of ‘acoustic’, but are not purely subjective either. In the functional intelligibility testing, the clinician writes down his/her understanding of the word pronounced by the patient and the matching with the intended word produced by the patient is done *off-line* during the scoring procedure, a procedure which has been used previously to assess intelligibility (Kent, 1992; Miller, 2013; Yorkston & Beukelman, 1978; Weismer, 2008). Here the intervention of a human listener is needed since the very notion of intelligibility refers to how adequately the intended targets are actually perceived by an interlocutor. In the assessment of articulation accuracy, the scoring of segmental errors on pseudo-words is

essentially relying on perceptual judgments. It should be noticed however that this assessment is also done off-line: the clinician follows a guided coding procedure for each pseudo-word, where questions are targeted on specific phonemes and on potential types of distortions. To answer these questions, the clinician/scorer can listen to each production as many times as necessary. Inter-rater agreement was evaluated for pseudo-word scoring, as this measure involves some amount of subjective assessment. Two different raters (an expert and a recently qualified SLP) independently scored the 151 target phoneme/syllables in the pseudo-word module for 20 patients (5 with PD, 5 ALS, 5 WD and 5 AoS). The mean inter-rater agreement on the 3020 observations was very high (98.94%, Cohens' Kappa = 0.89). With this off-line scoring procedure we thus obtained a very high inter-rater agreement between two clinicians on a significant subpart of the validation data.

Overall, the MonPaGe protocol takes advantage of objective descriptors of impaired speech parameters obtained via semi-automatic acoustic analyses or via targeted/guided coding of the intended production. Acoustic analysis has the advantage of releasing the clinicians from subjective descriptions, but it has the drawback of being time-consuming and relying on specific expertise. An assessment tool combining perceptual and easy-to-obtain acoustic information seems to represent a good balance taking advantage of the two approaches while minimizing their respective drawbacks. For MonPaGe-2.0.0s, a semi-automatic acoustic analyses routine was developed to be easily performed with minimal intervention and minimal acoustic-phonetic knowledge. These interventions typically required to check the automatic segmentation of the onsets-offsets of analysis windows in the audio files, and to adjust some key parameters in the case of noisy recordings. Then, the automatic extraction of acoustic measures on the various speech parameters, followed by their automated comparison with normative data defined on a large reference population (404 speakers) free the clinicians from the many pitfalls of a heuristic, subjective approach to clinical evaluation. To date, the MonPaGe-2.0.0s screening is

based on a relatively limited set of well-defined acoustic descriptors and is quite performant.

Objective measures should also facilitate a fast and effective communication among clinicians and should ensure a good/quality screening of MSD even when done by clinicians without expert competence in acoustics and phonetics.

Validation on a variety of MSD

The few available MSD assessment tools often lack a validation procedure, or they have been validated on very specific subtypes of MSD (e.g. progressive AoS vs. progressive aphasia in Strand et al., 2014; dysarthria associated with Parkinson disease in Cardoso et al., 2017) or on cohorts of patients with a large variety of dysarthria subtypes but without a grouping approach (Knuijd et al., 2017; DeBiagi et al., 2018). In the current study, the MonPaGe screening tool has been validated on a group of 80 patients encompassing a large diversity of types of MSD, including AoS following stroke, dysarthria associated to PD, to ALS, to FA, to WD and to Kennedy disease. Thus, as a screening tool, the MonPaGe protocol presents overall good performance in the diagnosis of the presence and severity of impaired speech for a variety of patterns of MSD. There are however some speech parameters which need to be improved in future implementations of MonPaGe. First, 25 patients with MSD displayed impaired intelligibility scores, but some patients with severe impairment of articulation displayed intelligibility scores within the normal range. This observation may be related to the difficulty to measure intelligibility and call for further improvement, although the relationship between intelligibility and articulatory errors is not necessarily linear as articulatory errors may be predicted by the listener and therefore comprehensible (Coppens-Hofman et al., 2016). Second, prosodic contrasts and maximum phonation time scores were deviant in very few patients. This may be due to low sensitivity of these specific tasks or to the fact that these parameters were within normal range in our group of patients with mild to moderate MSD and should be

495 specifically clarified in future. Finally, a further step will also be to achieve differential
496 diagnosis for different subtypes of MSD by enriching the screening tool with specific speech
497 descriptors and possibly additional speech tasks/materials.

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Declaration of interest statement

The authors report no conflict of interest

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645 Tables

646 **Table 1.** Patient distribution over the 6 clinical groups and associated descriptors: sex, age,
647 origin and severity measured with the Composite Perceptual (CP) score.

648 **Table 2.** Calculation of deviance scores (*DevS*) used in the MonPaGe screening protocol
649 (c=centiles, SD=standard deviations from the reference distribution)

650 **Table 3.** Descriptors and deviant scores for each of the 7 speech dimensions

651

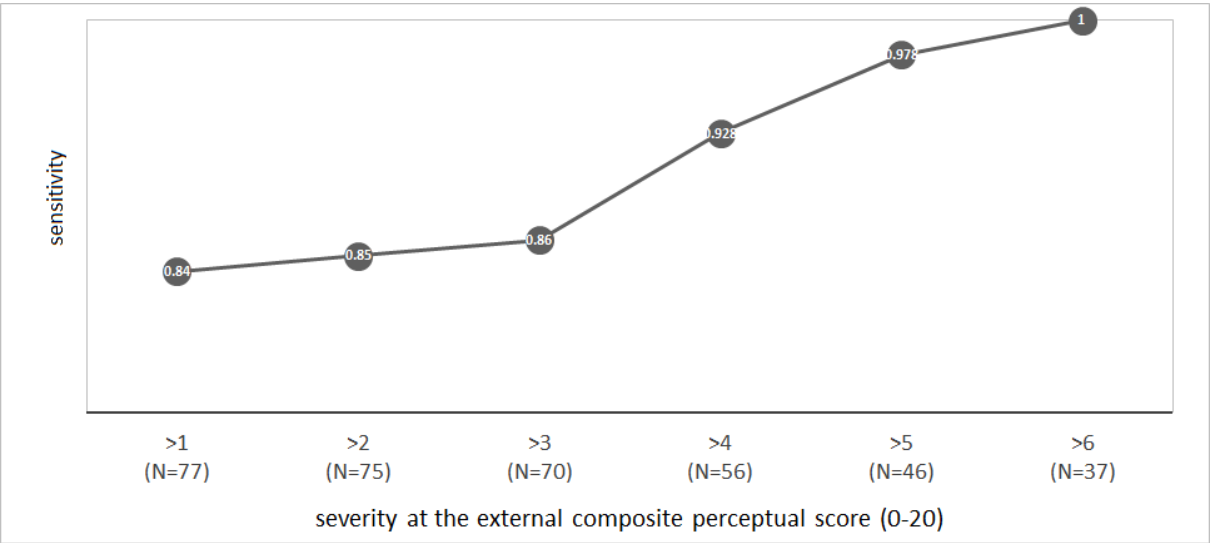
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654 **Figure caption**

655 **Figure 1.** sensitivity of the MonPaGe screening protocol, with a MonPaGe *TotalDevS* cut-off
656 score at >2, according to severity level of the population assessed with the external composite
657 perceptual score, ranging 0 to 20 (where 1 to 6 is considered mildly impaired, 7 to 13 moderate,
658 14 to 16 severe and > 16 very severe).

659



660

661 **Appendices**

662 **Appendix A.**

663 **Table A.I.** Normative values for the descriptors for the 7 dimensions in terms of mean, standard
664 deviation (sd) and median, and distribution tails (1st, 5th, 10th, 90th, 95th and 99th percentiles).
665 Tails that are informative for the determination of the DevS are presented in bold.

666

667 **Table A.II.** Reference values for Intelligibility (in terms of number of word not recognized over
668 15 words) and articulation accuracy (in terms of segmental errors over 151 targets). Centiles 1,
669 5, 50, 95, 99 and maximum error.

670

671 **Appendix B.**

672 Deviant scores (DevS) per dimensions and MonPaGe total deviance score (TotalDevS) for each
673 of the 80 patients.

674

675 **Appendix C.**

676 Deviant scores (DevS) per dimension and MonPaGe total deviance score (TotalDevS) for each
677 of the 35 test controls.

678

680 **Table A.I.** Normative values for the descriptors for the 7 dimensions in terms of mean,
681 standard deviation (sd) and median, and distribution tails (1st, 5th, 10th, 90th, 95th and 99th
682 percentiles). Tails that are informative for the determination of the *DevS* are presented in
683 bold.

		Sex	Age Group s	n	mean	sd	media n	1st	5th	10th	90th	95th	99th
VOICE	a_Jitter- PPQ5 (%)	f	20-93	173	0.25	0.13	0.22	0.09	0.11	0.13	0.41	0.47	0.65
		m	20-59	107	0.27	0.14	0.25	0.1	0.12	0.16	0.41	0.47	0.89
			60-74	44	0.3	0.15	0.26	0.1	0.12	0.13	0.55	0.63	0.69
			75-93	30	0.37	0.2	0.32	0.15	0.16	0.17	0.59	0.79	0.89
	a_Shimm er- APQ11 (%)	f	20-59	102	3	1.98	2.51	0.9	1.02	1.17	5.89	7.1	8.59
			60-93	67	4.56	3	4.09	1.06	1.22	1.37	8.51	9.48	12.57
		m	20-59	108	4.63	2.56	3.84	0.93	1.58	1.89	8.5	9.03	11.12
			60-93	75	5.85	2.84	5.52	1.29	1.91	2.35	9.71	11.28	11.81
	a_SDF0 (Hz)	f	20-59	103	3.77	2.05	3.31	1.14	1.41	1.8	6.5	7.4	10.58
			60-74	38	4.63	2.14	4.19	1.37	2	2.43	7.67	9.33	9.83
			75-93	29	4.96	2.91	3.75	2.02	2.36	2.57	10.69	11.52	11.71
		m	20-74	152	2.92	1.8	2.41	0.95	1.18	1.26	4.99	6.61	8.95
			75-93	29	5.09	2.64	4.53	1.58	1.98	2.45	8.32	9.07	10.98
	a_CPPs (dB)	f	20-93	171	15.55	2.32	15.32	11.13	11.74	12.69	18.89	19.42	20.94
		m	20-59	105	18.17	2.35	18.08	12.55	14.6	14.96	21.47	21.97	22.32
			60-93	75	16.49	2.85	16.38	11.08	12.19	12.91	20.44	21.76	22.5
	Speaking _SDF0	f	20-93	177	28.58	9.73	27.23	11.88	15.1	16.61	41.74	45.83	55.02

	(Hz)	m	20-93	170	18.87	7.09	17.4	8.86	9.69	10.82	29.12	31.46	39.37
	Speaking _CPPs (dB)	f	20-59	101	10.52	1.57	10.47	7.47	8.01	8.57	12.5	13.17	13.84
			60-74	36	10.9	1.49	10.59	8.42	8.83	9.16	12.88	13.37	13.9
			75-93	31	9.87	1.96	9.43	7.41	7.69	7.81	12.33	12.81	14.74
		m	20-93	171	11.21	2.03	11.02	7.71	8.09	8.52	14.1	14.97	15.47
MPT	MPT (s)	f	20-93	202	14.65	5.82	14.08	5.05	6.69	7.93	21.83	25	32.33
		m	20-93	187	16.62	6.77	15.35	6.57	7.7	9.22	25.71	29.35	36.55
PROSODY	Melodic contrast Q-A (st)	f+m	20-59	228	6.59	4.46	6.98	-1.57	-1.05	0.08	11.7	13.89	17.2
			60-93	161	4.69	5.4	4.7	-5.97	-4.22	-2.46	11.74	13.8	15.98
SPEECH RATE	Rate_Sen tence (ph/s.)	f	20-59	119	11.15	1.67	11.16	7.7	8.41	8.86	13.22	13.78	15.12
			60-93	89	9.75	1.95	9.58	6.23	6.59	6.91	12.31	13.08	13.51
		m	20-59	106	11.53	1.9	11.69	7.08	8.34	8.98	14.04	14.67	14.81
			60-93	79	10.18	1.99	10.15	6.34	6.86	7.14	12.72	13.05	14.72
DDK RATE	AMR _{cv}	f	20-59	117	10.38	1.73	10.64	5.97	7.12	8.06	12.39	12.72	13.36
			60-93	89	9.29	2.08	9.13	5.19	5.61	6.2	12.16	12.54	13.31
		m	20-59	108	10.89	1.82	11.1	6.13	7.44	8.64	13.01	13.65	13.91
			60-93	78	9.44	2.16	9.7	4.9	5.61	6.5	12.15	12.67	14.04
	AMR _{ccv}	f	20-59	119	13.23	2.63	13.51	7.48	8.17	9.57	16.31	17.59	18.54
			60-93	90	11.24	2.71	11.18	5.65	6.85	7.79	14.32	15	17.19
		m	20-59	106	13.88	2.74	14.24	7.15	9.13	10.31	17.01	17.97	18.94
			60-93	78	11.66	2.95	11.52	6.31	6.61	7.67	15.83	16.5	17.27
	SMR _{cv} (/badego /)	f	20-59	117	11.67	2.3	11.88	5.32	7.64	8.61	14.22	14.76	16.39
			60-93	88	9.84	2.58	9.95	5.23	5.83	6.84	12.96	13.75	15.41
		m	20-59	109	12.18	2.43	12.59	4.89	7.8	8.65	15.17	15.68	16.49
			60-93	78	10.33	2.68	9.98	5.26	6.12	7.11	13.77	15.17	15.66

SMR _{CV} - AMR _{CV}	f+m	20-59	222	1.3	1.55	1.41	-2.48	-1.09	-0.74	3.2	3.74	4.71
		60-74	91	0.94	1.52	1.12	-3.3	-1.86	-0.71	2.77	3.365	3.827
		75-93	78	0.6	1.08	0.55	-1.52	-1	-0.66	1.82	2.47	3.34

684

685

Table A.II. Reference values for Intelligibility (in terms of number of word not recognized over 15 words) and articulation accuracy (in terms of segmental errors over 151 targets). Centiles 1, 5, 50, 95, 99 and maximum error.

	<i>Age Groups</i>	<i>median</i>	<i>1st</i>	<i>5th</i>	<i>10th</i>	<i>90th</i>	<i>95th</i>	<i>99th</i>	<i>maximum</i>
# words not recognized	20-74	0	0	0	0	1	1	2	3
	75-93	0	0	0	0	1	1.5	3.5	5
# segmental errors	20-59	0	0	0	0	1	2	4	5
	60-74	0	0	0	0	3	4	5	5
	75-93	1	0	0	0	4	5	6	6

691 **Appendix B.** Deviant scores (*DevS*) per dimensions and MonPaGe total deviance score
692 (*TotalDevS*) for each of the 80 patients.

patient	age	gender	group	Intelligibility	Articulation	MaxPhonation Time	Voice						Prosody	Speech Rate	DDK				MonPaGe TotalDevS
							d1	d2	c	b	a2	a1			A	B	C	D	
Fri01	50	F	FA																9.0
Fri02	38	F	FA																8.5
Fri03	36	F	FA																1.0
Fri04	31	F	FA																7.0
Fri05	37	F	FA																3.0
Fri06	24	M	FA																4.0
Fri07	45	M	FA																14.0
Fri08	46	M	FA																6.5
Fri09	29	M	FA																11.5
Fri10	40	M	FA																12.5
Wil01	29	M	WD																6.0
Wil02	44	M	WD																7.0
Wil03	27	M	WD																6.5
Wil04	33	M	WD																5.5
Wil05	31	M	WD																4.0
Wil06	35	M	WD																4.0
Wil07	49	M	WD																3.5
Wil08	25	M	WD																8.5
Wil09	44	F	WD																8.0
Wil10	35	M	WD																3.0
ALS01	61	F	ALS																6.5
ALS02	45	F	ALS																7.0
ALS03	66	F	ALS																1.5
ALS04	58	M	ALS																6.0
ALS05	60	M	ALS																4.0
ALS06	46	M	ALS																0.5
ALS07	63	M	ALS																6.0
ALS08	52	M	ALS																6.5
ALS09	46	M	ALS																5.0
ALS10	53	M	ALS																6.0
Ken01	56	M	SBMA																2.0
Ken02	70	M	SBMA																4.0
Ken03	66	M	SBMA																3.0
Ken04	66	M	SBMA																4.0
Ken05	72	M	SBMA																0.0
Ken06	58	M	SBMA																4.5
Ken07	60	M	SBMA																4.0
Ken08	76	M	SBMA																4.0
Ken09	64	M	SBMA																3.0
Ken10	73	M	SBMA																3.5
Par01	72	F	PD																3.0
Par02	81	F	PD																2.5
Par03	75	F	PD																7.0
Par04	79	F	PD																3.5
Par05	69	F	PD																5.0
Par06	74	F	PD																9.0
Par07	65	M	PD																3.0
Par08	76	M	PD																2.5
Par09	58	M	PD																0.0
Par10	65	M	PD																4.5
Par11	73	F	PD																2.5
Par12	73	M	PD																0.0
Par13	59	M	PD																2.5
Par14	56	M	PD																2.5
Par15	70	F	PD																4.0
Par16	55	M	PD																1.5
Par17	55	M	PD																0.5
Par18	64	M	PD																6.0
Par19	69	M	PD																3.5
Par20	70	M	PD																0.0
ALS11	73	M	ALS																1.5
ALS12	72	M	ALS																3.0
ALS13	72	M	ALS																3.5
ALS14	81	F	ALS																3.0
ALS15	74	M	ALS																7.5
ALS16	73	F	ALS																5.5
ALS17	71	F	ALS																1.0
ALS18	77	F	ALS																0.5
ALS19	75	F	ALS																4.5
ALS20	75	F	ALS																7.0
AoS01	54	M	AoS																10.0
AoS02	24	M	AoS																2.5
AoS03	60	M	AoS																8.5
AoS04	61	F	AoS																4.0
AoS05	32	F	AoS																7.0
AoS06	72	F	AoS																3.0
AoS07	43	F	AoS																5.0
AoS08	54	F	AoS																7.5
AoS09	72	F	AoS																11.5
AoS10	53	M	AoS																6.5

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Appendix C.

Deviant scores (*DevS*) per dimension and MonPaGe total deviance score (*TotalDevS*) for each of the 62 test controls.

participant	gender	age	Intelligibility	Articulation (Pseudow)	MaxPhonation Time	Voice						Prosody	Speech Rate	DDK				MonPaGe Total DevScore
						a1	a2	b	c	d1	d2			A	B	C	D	
CTRL1	M	38	1											#	#	#	#	3.0
CTRL2	M	45		1														2.0
CTRL3	F	31		1														2.0
CTRL4	F	26																0.0
CTRL5	F	25																0.0
CTRL6	F	33																2.0
CTRL7	F	29			1													1.0
CTRL8	M	30																0.0
CTRL9	F	26											1		#			1.5
CTRL10	F	82																1.5
CTRL11	M	65																0.0
CTRL12	F	60																0.0
CTRL13	F	62																0.0
CTRL14	M	50																1.0
CTRL15	M	63																0.0
CTRL16	F	64																0.0
CTRL17	M	68																0.0
CTRL18	F	60																0.0
CTRL19	F	48																0.0
CTRL20	F	47		2														2.0
CTRL21	F	56																0.0
CTRL22	F	65																0.0
CTRL23	F	61																0.0
CTRL24	M	73																0.0
CTRL25	F	79																0.0
CTRL26	M	50																0.0
CTRL27	M	25																0.0
CTRL28	F	54																0.0
CTRL29	F	65			2													2.0
CTRL30	F	59																0.0
CTRL31	M	80																0.0
CTRL32	F	62																0.5
CTRL33	F	76																2.5
CTRL34	F	76																0.0
CTRL35	M	77																0.0
CTRL36	F	25																0.0
CTRL37	F	67																0.0
CTRL38	F	28																0.0
CTRL39	F	50																0.0
CTRL40	F	83																0.0
CTRL41	M	35																0.0
CTRL42	F	60																0.0
CTRL43	M	58																0.0
CTRL44	F	82																0.0
CTRL45	F	27																2.0
CTRL46	F	55											2					2.0
CTRL47	M	35																0.0
CTRL48	M	47																0.0
CTRL49	F	82																0.0
CTRL50	M	62		1														1.0
CTRL51	F	53																1.0
CTRL52	F	81																0.5
CTRL53	M	88			1													1.5
CTRL54	F	27																0.0
CTRL55	M	24		2														2.0
CTRL56	F	25																0.0
CTRL57	F	22																0.0
CTRL58	F	70		2														2.0
CTRL59	F	72																0.0
CTRL60	M	70																0.0
CTRL61	M	72																0.5
CTRL62	M	27																0.0

within normal range (0)
 mildly deviant (1)
 moderate deviant (2)
 severe deviant (3)
 excessive deviant (4)
 missing data

Voice:
 a1 /a/ F0 SD
 a2 Speaking F0 SD
 b /a/ Jitter
 c /a/ Shimmer
 d1 /a/ CPPS
 d2 Speaking CPPS

DDK:
 A CCV AMR
 B CV AMR
 C CV SMR
 D (CV SMR) - (CV AMR)