

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

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- 5 protocol
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28 ABSTRACT

29 To respond to the need of objective screening tools for motor speech disorders (MSD), 30 we present the screening version of a speech assessment protocol (MonPaGe-2.0.0s), 31 which is based on semi-automated acoustic and perceptual measures on several speech 32 dimensions in French. We validate the screening tool by testing its sensitivity and 33 specificity and comparing its outcome with external standard assessment tools. The data 34 from 80 patients diagnosed with different types of mild to moderate MSD and 62 healthy 35 test controls were assessed against the normative data obtained on 404 neurotypical 36 speakers, with Deviance Scores computed on seven speech dimensions (voice, speech 37 rate, articulation, prosody, pneumophonatory control, diadochokinetic rate, intelligibility) based on acoustic and perceptual measures. A cut-off of the MonPaGe total deviance 38 39 score (TotDevS) > 2 allowed MSD to be diagnosed with specificity of 95% and an overall 40 sensitivity of 83.8% on all patients pulled, reaching 91% when very mildly impaired 41 patients were excluded. A strong correlation was found between the MonPaGe TotDevS 42 and an external composite perceptual score of MSD provided by six experts. The 43 MonPaGe screening protocol has proven its sensitivity and specificity for diagnosing 44 presence and severity of MSD. Further implementations are needed to complement the 45 characterization of impaired dimensions in order to distinguish subtypes of MSD.

47 Introduction

Although both impaired language and impaired speech can be associated with a variety of 48 49 neurodegenerative pathologies and of acquired brain lesions, clinicians have at one's disposal 50 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 51 52 available for motor speech disorders (MSD) in clinical practice are mostly based on perceptual 53 judgments. To complement the field of perceptual assessment tools for MSD, a consortium 54 made up of researchers and clinicians created the MonPaGe speech screening battery, which is 55 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 56 57 whether the screening version of the protocol (MonPaGe-2.0.0s) can be reliably used as a 58 screening tool for the detection of MSD by assessing its specificity and sensitivity on a group 59 of 80 patients with various patterns of MSD and a group of neurotypical test controls against a 60 normative database gathered from 404 French-speaking healthy adults. Crucially, the potential 61 of the protocol to diagnose the presence of MSD in the patient group is tested by including 62 speakers with mild MSD in the patient cohort. The severity score obtained with the screening 63 protocol is also tested against an external assessment of the speech disorder severity provided 64 by a perceptually-based severity assessment made by six expert judges using a tool widely used 65 in French-speaking clinical settings.

66 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 rate (Duffy, 2005; McNeil, Robin, & Schmidt, 2008). Several subtypes of dysarthria are 73 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.

99 Whether conceived to assess dysarthria or AoS, speech assessment batteries available in clinical 100 practice mostly rely on perceptual classification of speech and voice disorders, often using a 101 large set of feature descriptors, which are well mastered only by SLP with an expertise in motor 102 speech disorders. There are some well-known downsides to the perceptual description of speech 103 features. First, the reliability and reproducibility of perceptual judgment of speech parameters 104 in MSD have often been questioned (Bunton, Kent, Duffy, Rosenbek, & Kent, 2007; Haley et 105 al., 2012). This is in particular the case when speech is assessed perceptually based on specific 106 dimensions, whereas better interrater agreement is observed on overall severity scores, at least 107 for trained raters (Ziegler, Staiger, Schrölderle & Vogel, 2017). Although assessment is based 108 on a balance of subjective and objective evaluation in many domains, subjective evaluation is 109 preponderant in the perceptual assessment of specific speech parameters, as they rely on 110 individual internal representations that are based on the natural variability of these speech 111 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

118 perceptual impression represents the very first information available to the clinician in front of 119 the patients, tools are needed to further guide the assessment, combining functional, perceptual 120 judgments with objective acoustic descriptors of impaired speech parameters. One challenge of 121 this approach is to select a limited number of clinically-relevant descriptors, with no claim to 122 be exhaustive with regards to all potential measures. Indeed, there is a constantly growing 123 amount of measures, acoustic analyses and statistical techniques for characterizing a diversity 124 of aspects of the acoustics of dysarthric speech reported in the scientific literature. However, 125 they are typically very specific, quite sophisticated, not implemented as stand-up procedures in 126 the most popular softwares for voice and speech analysis, such as Praat (Boersma & Weenink, 127 2018) or the Multi-Dimensional Voice Program (MDVP, from Kay Elemetrics Corporation); 128 in short, they are not readily available for clinical purpose.

129 The MonPaGe speech assessment protocol and the MonPaGe screening tool

130 The MonPaGe speech assessment protocol (Fougeron et al. 2016, 2018; Pernon et al., 2020) 131 has been developed (1) to provide a comprehensive speech assessment tool for French-speaking 132 adults and (2) to integrate objective descriptors of potentially deviant speech parameters 133 obtained with acoustic analyses. The MonPaGe protocol integrates the main speech parameters 134 used to assess both AoS and dysarthria in clinical assessment tools. These parameters are 135 quantified either perceptually or acoustically, in a way that can be carried out by SLT or other 136 professionals qualified to assess speech but who do not necessarily have an expert competence 137 in acoustic phonetics or speech engineering. The MonPaGe-2.0.0s presented here is a screening 138 version based on a subset of speech dimensions and parameters from the MonPaGe protocol 139 and aimed at detecting speech patterns that deviate from normal conditions. A speech screening 140 tool should allow to determine whether the patient's speech deviates from typical speech, how 141 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 thedifferential 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.

- 151
- 152 Method

153 **Population**

154 Reference group: healthy speakers

155 For the reference values on the speech dimensions tested in MonPaGe, a group including 404 156 neurotypical speakers aged 20-93 years was selected from the MonPaGe_HA (for Healthy 157 Adults) database of spoken French (Fougeron et al., 2018). Participants were recruited in 4 158 French-speaking locations: Mons, Belgium (n = 104); Montréal, Canada (n = 103); Paris, France 159 (n = 101); Geneva, Switzerland (n = 97). All participants spoke French as their primary language 160 (mother tongue) and currently used language. The population recruited for the MonPaGe_HA 161 database is balanced within and between countries on sex (209 women and 195 men) and age 162 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

165 [20-59] (N = 231, 119 women, mean age = 41.81, SD = 11.44), and older speakers further split 166 into two groups [60-74] (N = 94, 48 women, mean age = 66.01, SD =4.31) and [75-93] (N = 167 79, 42 women, mean age = 81.71, SD = 4.30). This choice was driven by a first screening of 168 the data which confirmed findings in the literature showing little evidence of age effects on 169 speech between 20 and 60. The two oldest speakers groups include less speakers but were kept 170 as subgroups in response to clinical needs, where appropriate reference values based on such 171 partition are needed for the elderlies.

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).

177

178 Test group 1: Speakers with MSD

179 80 patients aged 24 to 81 (mean age = 57.1) diagnosed with mild to moderate MSD recruited 180 in France (Pitié-Salpêtrière University Hospital and Lariboisière Hospital, Paris), in French-181 speaking Switzerland (Geneva University Hospitals) and French-speaking Belgium (Hôpital 182 André Vésale, Charleroi University Hospital) were included for the validation. In order to vary 183 the speech profiles, patients with MSD associated with six different underlying pathologies 184 were included, namely Parkinson disease (PD), Amyotrophic Lateral Sclerosis (ALS), 185 Friedreich ataxia (FA), Kennedy disease (spinal and bulbar muscular atrophy, SBMA), Wilson 186 disease (WD), and post-stroke AoS. The neurological diagnosis was established by neurologists 187 in the hospitals where patients were recruited. To be included in the present study, the patients 188 had to be French native speakers, present mild or moderate acquired or progressive speech difficulty noticed by the patient and a SLP, no or only very mild language impairment. Patients with diagnosed dementia or psychiatric disorders and patients with history of developmental speech and language disorders or hearing impairment were excluded. Table 1 presents the distribution and description of the patients over the six clinical groups with associated basic 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

62 neurotypical French-speaking control participants who were not included in the reference group were recruited in Geneva. They were aged 22 to 88 (mean age = 53.4, 21 men) and had 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.

212 Speech material and speech descriptors

213 The MonPaGe protocol assesses different speech dimensions based on a variety of tasks (a 214 detailed description of the MonPaGe protocol is provided in Fougeron et al.. 2018). Here, a 215 subset of the original speech descriptors was retained for the MonPaGe-2.0.0s screening tool 216 for MSD, providing information on seven dimensions, namely: intelligibility, articulation, 217 pneumophonatory control, voice, prosody, speech rate and diadochokinetic rate. These 218 dimensions have been selected in agreement with the expert SLT associated with the project, 219 based on (i) dimensions that are most used in clinical practice and in other assessment tests and 220 (ii) a balance between the time necessary to assess each dimension and its informativity. Note 221 that the speech descriptors resulting from this procedure, although each relating to a specific 222 dimension, have not the ambition to reflect all the potential disorders on that dimension of 223 speech. Ultimately, they have been selected for their typicality, because they represent the 224 participant's level of competence on that dimension, and/or for their informativity (with respect 225 to the aim of developing a screening tool for MSD), not for the sake of comprehensiveness. For 226 example, prosody is assessed in the extensive MonPaGe protocol via several acoustic and 227 perceptual measures of the distinctive, demarcating and expressive functions of intonation in 228 French, as exhibited by the reading of a tale and the production of selected sentences. In the 229 end, only the ability to express an interrogation via melodic modulations is incorporated in the 230 screening tool. Similarly, pneumophonatory control is indexed by, but by no means reducible 231 to, the indicator of maximum phonation time, which is very commonly used in clinical practice, 232 although its relationship with laryngeal and respiratory physiology is admittedly highly 233 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.

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

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

260 Pseudoword production was elicited via the bimodal presentation of each target (in an 261 orthographic form on a screen and in an audio form via headphones), to minimise errors due to 262 pseudo-word reading difficulties or to misperception. Regarding the assessment of articulation 263 accuracy, errors on targeted consonants, vowel and syllables were listed based on perceptual 264 assessment following a guided coding procedure for each pseudo-word, where questions were 265 targeted on specific phoneme and on potential types of errors (distortion, substitution, omission, 266 insertion). The computerized tool allowed the raters to play each pseudoword as needed and 267 score targeted phonemes or syllables as correct or incorrect. Overall, 151 targeted phonemes or 268 syllables included in the 50 pseudowords were scored per speaker and a descriptor related to 269 articulation accuracy is expressed in terms of a number of errors (from 0 to a maximum of 151). 270 The pseudo-word production is used as a screening for articulatory precision in MonPaGe-2.0 271 as only the total number of errors is considered and can be easily extracted via the guided 272 computerized scoring tool, but it has been conceived to be used also for further more detailed 273 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).*

281 Voice. Voice-related measures were based on a sustained production for 2-3 seconds of the 282 vowel /a/ at a comfortable height and loudness and on the reading of a 7 syllables sentence 283 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).

A second set of descriptors related to the speaking voice was measured on the longer continuous read speech sample "Mélanie vend du lilas". On the f0 time series computed over the whole sentence, mean (*speaking_meanf0*) and standard deviation (*speaking_SDf0*) were computed. The smoothed cepstral peak prominence was also taken from the whole voiced sentence (*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 1'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 (f0range@end-f0range@begin)question - (f0range@end-f0range@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 /tua/. 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

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

332 The number of phonemes produced over an interval of about 4 seconds of continuous repetition 333 was used as an index of DDK rate for AMR_{CV}, AMR_{CCV}, SMR_{CV}. This interval was selected 334 from the onset of the speech waveform and was manually adjusted to the right in order not to 335 cut the last syllable if needed. Also, in order to capture difficulties in the repetition of the same 336 syllables (AMR) vs. the repetition of a sequence of three syllable (SMR) which could be found 337 for speakers with AoS for instance (e.g. Ziegler, 2002), we also computed the difference 338 between the sequential motion rate and the alternative motion rate averaged over all CV 339 sequences: (SMR_{CV}- AMR_{CV}).

340 Procedure and analyses

341 All the participants underwent the MonPaGe protocol following the standard assessment 342 procedure with speech and speech-language pathologists in clinical settings for the patients and 343 in a standard room at the University for the controls. The MonPaGe protocol was run on a 344 laptop and speech samples were recorded using either a head-mounted or a table microphone 345 depending on the place of recruitment. This variability in recording reflects the variations across 346 clinical settings and has also been introduced in the recordings of the reference group (see 347 Fougeron et al., 2018). The assessment takes about 20 minutes with typical speakers and 30 348 minutes with patients with moderate MSD (Pernon et al., 2020), and the same duration is 349 necessary for scoring.

350 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 354 norms, linear models were first computed in R with Sex (male/female) and Age-group ([20-355 59], [60-74], [75+]) as between-subject factors and the various descriptors as dependent 356 variables. Whenever an effect of Sex or an interaction with Age-goup was found, further 357 analyses were carried out for the male and female speakers separately. Inter-group differences 358 for the Age-group predictor were tested with Tukey post-hoc tests. Results from these statistical 359 analyzes are provided in the supplementary material. Normative values for each descriptor was 360 then computed by subgroups accordingly. Since groups split or merging were done according 361 to the sex or age effects, the number of participants retained for the computation of normative 362 values varied according to the descriptor considered.

363 *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.

367 A deviance score (DevS) spanning from 0 to 4 was defined for each descriptor depending on 368 its distance from the reference value. The standard limit from the normative data was fixed at 369 centile 5 (Brooks, Sherman, Iverson, Slick, & Strauss, 2011) and five degrees of DevS are defined, 370 with DevS = 0 standing for no-deviant (within normal range) and DevS = 4 for excessively 371 deviant, according to the position of the speaker's descriptor value relative to the reference 372 distribution. Cut-off values for defining these deviance degrees are determined according to 373 either the tails of the distribution (c5/c95 and c1/c99, for the DevS = 1 and DevS = 2374 respectively) and further severity, beyond c1/c99, is computed based on the inter-centile 375 distance between c50 and c5/c95, as detailed in Table 2.

377 (Table 2 about here)

378

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.

384 (Table 3 about here)

385 The detailed procedures for the assessment of sensitivity, specificity and external validity are386 described along with the results.

387 Results

388 *Reference values*

389 The normative values for each descriptor obtained from the 404 healthy speakers reference 390 group are presented in Appendix A (Tables A.I and A.II). Normative values for each descriptor 391 and for each age/sex subgroup are presented in terms of mean, median and standard deviation, 392 except for intelligibility and segmental errors for which median and maximum error rates are 393 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 394 395 limits. According to the descriptor, the tails of interest for the determination of the cut-off scores 396 are either on the upper or lower sides (e.g. for error values, alteration is to be found in the upper 397 tail while 0 error is normal).

399 Specificity and sensitivity of MonPaGe

400 Appendix B and C present the deviant scores per speech dimension and *TotalDevS* for each of
401 the 80 patients and 62 healthy test controls, respectively. The *MonPaGe TotalDevS* ranges from
402 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 $\leq 4/20$).

410 (*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.

414 **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 422 score, the MonPaGe *TotalDevS*, was computed as the sum of all *DevS*. With a cut-off score at 423 MonPaGe *TotalDevS* > 2, the *specificity* of the tool was excellent (95%), and its sensitivity 424 was very high, in particular for the mild to moderate patients (sensitivity > 92% when the 425 patients with very mild MSD are excluded). Along with the strong correlation between the 426 MonPaGe *TotalDevS* and an external composite perceptual assessment of severity obtained 427 from 6 expert judges, the present results indicate that the MonPaGe protocol is a reliable 428 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.

432 Guided objective analyses of impaired speech

433 As exposed in the introduction, the available clinical assessment tools for MSD are mostly 434 based on perceptual judgment, which are bound to rely heavily on subjective evaluation. For 435 these reasons the MonPaGe assessment protocol relies mainly on descriptors extracted from the 436 acoustic signal for five out of the seven dimensions (voice, MPT, speech rate, prosodic contrast 437 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 438 439 the functional intelligibility testing, the clinician writes down his/her understanding of the word 440 pronounced by the patient and the matching with the intended word produced by the patient is 441 done off-line during the scoring procedure, a procedure which has been used previously to 442 assess intelligibility (Kent, 1992; Miller, 2013; Yorkston & Beukelman, 1978; Weismer, 2008). 443 Here the intervention of a human listener is needed since the very notion of intelligibility refers 444 to how adequately the intended targets are actually perceived by an interlocutor. In the 445 assessment of articulation accuracy, the scoring of segmental errors on pseudo-words is

446 essentially relying on perceptual judgments. It should be noticed however that this assessment 447 is also done off-line: the clinician follows a guided coding procedure for each pseudo-word, 448 where questions are targeted on specific phonemes and on potential types of distortions. To 449 answer these questions, the clinician/scorer can listen to each production as many times as 450 necessary. Inter-rater agreement was evaluated for pseudo-word scoring, as this measure 451 involves some amount of subjective assessment. Two different raters (an expert and a recently 452 qualified SLP) independently scored the 151 target phoneme/syllables in the pseudo-word 453 module for 20 patients (5 with PD, 5 ALS, 5 WD and 5 AoS). The mean inter-rater agreement 454 on the 3020 observations was very high (98.94%, Cohens' Kappa = 0.89). With this off-line 455 scoring procedure we thus obtained a very high inter-rater agreement between two clinicians 456 on a significant subpart of the validation data.

457 Overall, the MonPaGe protocol takes advantage of objective descriptors of impaired speech 458 parameters obtained via semi-automatic acoustic analyses or via targeted/guided coding of the 459 intended production. Acoustic analysis has the advantage of releasing the clinicians from 460 subjective descriptions, but it has the drawback of being time-consuming and relying on specific 461 expertise. An assessment tool combining perceptual and easy-to-obtain acoustic information 462 seems to represent a good balance taking advantage of the two approaches while minimizing 463 their respective drawbacks. For MonPaGe-2.0.0s, a semi-automatic acoustic analyses routine 464 was developed to be easily performed with minimal intervention and minimal acoustic-phonetic 465 knowledge. These interventions typically required to check the automatic segmentation of the 466 onsets-offsets of analysis windows in the audio files, and to adjust some key parameters in the 467 case of noisy recordings. Then, the automatic extraction of acoustic measures on the various 468 speech parameters, followed by their automated comparison with normative data defined on a 469 large reference population (404 speakers) free the clinicians from the many pitfalls of a 470 heuristic, subjective approach to clinical evaluation. To date, the MonPaGe-2.0.0s screening is

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

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

475 Validation on a variety of MSD

476 The few available MSD assessment tools often lack a validation procedure, or they have been 477 validated on very specific subtypes of MSD (e.g. progressive AoS vs. progressive aphasia in 478 Strand et al., 2014; dysarthria associated with Parkinson disease in Cardoso et al., 2017) or on 479 cohorts of patients with a large variety of dysarthria subtypes but without a grouping approach 480 (Knuijd et al., 2017; DeBiagi et al., 2018). In the current study, the MonPaGe screening tool 481 has been validated on a group of 80 patients encompassing a large diversity of types of MSD, 482 including AoS following stroke, dysarthria associated to PD, to ALS, to FA, to WD and to 483 Kennedy disease. Thus, as a screening tool, the MonPaGe protocol presents overall good 484 performance in the diagnosis of the presence and severity of impaired speech for a variety of 485 patterns of MSD. There are however some speech parameters which need to be improved in 486 future implementations of MonPaGe. First, 25 patients with MSD displayed impaired 487 intelligibility scores, but some patients with severe impairment of articulation displayed 488 intelligibility scores within the normal range. This observation may related to the difficulty to 489 measure intelligibility and call for further improvement, although the relationship between 490 intelligibility and articulatory errors is not necessarily linear as articulatory errors may be 491 predicted by the listener and therefore comprehensible (Coppens-Hofman et al., 2016). Second, 492 prosodic contrasts and maximum phonation time scores were deviant in very few patients. This 493 may be due to low sensitivity of these specific tasks or to the fact that these parameters were 494 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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512 **Declaration of interest statement**

513 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,
- 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)
- **Table 3.** Descriptors and deviant scores for each of the 7 speech dimensions
- 651

653654 Figure caption

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



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	

679 Appendix A.

- 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
		f	20-93	173	0.25	0.13	0.22	0.09	0.11	0.13	0.41	0.47	0.65
	a_Jitter-		20-59	107	0.27	0.14	0.25	0.1	0.12	0.16	0.41	0.47	0.89
	PPQ5 (%)	m	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	f	20-59	102	3	1.98	2.51	0.9	1.02	1.17	5.89	7.1	8.59
	er-		60-93	67	4.56	3	4.09	1.06	1.22	1.37	8.51	9.48	12.57
	APQ11	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
Щ	a_SDf0		20-59	103	3.77	2.05	3.31	1.14	1.41	1.8	6.5	7.4	10.58
VOIG	(Hz)	f	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	f	20-93	171	15.55	2.32	15.32	11.13	11.74	12.69	18.89	19.42	20.94
	(dB)	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
	Sneaking		20-59	101	10.52	1.57	10.47	7.47	8.01	8.57	12.5	13.17	13.84
_CPPs		f	60-74	36	10.9	1.49	10.59	8.42	8.83	9.16	12.88	13.37	13.9
	(dB)		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
ът П	- (s)	f	20-93	202	14.65	5.82	14.08	5.05	6.69	7.93	21.83	25	32.33
Σ	TAM	m	20-93	187	16.62	6.77	15.35	6.57	7.7	9.22	25.71	29.35	36.55
7	Melodic		20-59	228	6.59	4.46	6.98	-1.57	-1.05	0.08	11.7	13.89	17.2
ROSOD	contrast	f+m											
Ы	Q-A (st)		60-93	161	4.69	5.4	4.7	-5.97	-4.22	-2.46	11.74	13.8	15.98
	Rate Sen	f	20-59	119	11.15	1.67	11.16	7.7	8.41	8.86	13.22	13.78	15.12
H RATE	tence		60-93	89	9.75	1.95	9.58	6.23	6.59	6.91	12.31	13.08	13.51
SPEECH	(ph/s.)	m	20-59	106	11.53	1.9	11.69	7.08	8.34	8.98	14.04	14.67	14.81
• 7			60-93	79	10.18	1.99	10.15	6.34	6.86	7.14	12.72	13.05	14.72
	AMRcv	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
		f	20-59	119	13.23	2.63	13.51	7.48	8.17	9.57	16.31	17.59	18.54
RATE	AMRccv		60-93	90	11.24	2.71	11.18	5.65	6.85	7.79	14.32	15	17.19
DDK		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
	SMRcv	f	20-59	117	11.67	2.3	11.88	5.32	7.64	8.61	14.22	14.76	16.39
	(/badego		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

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

- **Table A.II.** Reference values for Intelligibility (in terms of number of word not recognized over
- 687 15 words) and articulation accuracy (in terms of segmental errors over 151 targets). Centiles
- 688 1, 5, 50, 95, 99 and maximum error.

	Age Groups	median	1st	5th	10th	90th	95th	99th	maximum
# words	20-74	0	0	0	0	1	1	2	3
not recognized	75-93	0	0	0	0	1	1.5	3.5	5
#	20-59	0	0	0	0	1	2	4	5
segmental	60-74	0	0	0	0	3	4	5	5
errors	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	dz di Voice b a2 a1	Prosody	Speech Rate	ррк в	MonPaGe TotalDevS	
Fri01	5	0 F	FA	2	3	0	000000	0	2	2120	9.0	within normal range (0)
FriO2	3	8 F	FA	0	\$	1		0	2	5 1 2 0	8.5	/ mildly deviant (1)
Fri03	3	6 F	FA	0	0	0	0000 10	0	0	0010	1.0	moderate deviant (2)
Fri04	3	7 F	FA	0	5	0	<u>NO</u> O O O O O O	0	2	2120	7.0	severe deviant (3)
Fri06	2	4 M	FA	0	5	0	000000	0	0	1100	4.0	encessive demand (1)
Fri07	4	5 M	FA	e.	5	2	400000	1	e e	\$ 100	14.0	missing data
Fri08	4	6 M	FA	0	5	0		0	1	5120	6.5	
Fri109	4	0 M	FA	2	4	2	0 10 0 0 0 0 0 0 0 0 0 0 0 0 0 0 0 0 0	0	3 2	5200	11.5	01 /a/ F0 SD
Wil01	2	9 М	WD	0	4	0	000000	0	1	1100	6.0	a2 Speaking F0 SD
Wil02	4	4 M	WD	æ	3	0	000020	0	1	0000	7.0	b /a/ Jitter
Wil03	2	7 M	WD	0	4	0	001000	0	1	1100	6.5 E E	c /a/ Shimmer
Wilos	3	1 M	WD	1	3	0	0000000	0	0	0000	5.5 4.0	d2 Speaking CPPS
Wil06	з	5 M	WD	0	4	0	000000	0	0	0000	4.0	
Wil07	4	9 M	WD	0	3	0		0	0	1000	3.5	DDK:
Wil08	2	5 M	WD	5	3	0	001020	0	0	1010	8.5	
Wil10	3	5 M	WD	0		0	000000	0	0	0000	3.0	C CV SMR
ALSO1	6	1 F	ALS	1	\$	0	400000	0	0	1000	6.5	D (CV SMR) - (CV AMR)
ALSO2	4	5 F	ALS	0	\$	0	001000	0	2	2110	7.0	
ALSO3	6	6 F	ALS	0	0	0	200001	0	0	0000	1.5	FA Friedreich Ataxia
ALS04 ALS05	6	ом	ALS	0	2	0	0000000	0	2	0020	4.0	ALS Amyotrophic Lateral Sclerosis
ALS06	4	6 M	ALS	0	0	0	100000	0	0	0000	0.5	PD Parkinson disease
ALS07	6	зм	ALS	1	3	0	4 Nº 0 0 0 0	0	0	0000	<u>6.0</u>	AoS Apraxia of Speech (post stroke)
ALSO8	5	2 M	ALS	0	4	0	000000	0	0	3120	6.5	SBMA Spinal and Bulbar Muscular Atrophy
ALS09 ALS10	-4	3 M	ALS	0	3	0	0000000	0	2	2120	5.0 6.0	(Kennedy disease)
Ken01	5	6 M	SBMA	0	0	0	400000	0	0	0000	2.0	
Ken02	7	οм	SBMA	0	9	0	000000	1	0	0000	4.0	
Ken03	6	6 M	SBMA	0	3	0		0	0	0000	3.0	
Ken04	7	2 M	SBMA	0	3	0	100000	0	0	0001	4.0	
Ken06	5	8 M	SBMA	1	5	0	001100	0	0	0000	4.5	
Ken07	6	οм	SBMA	1	S	0	000000		0	0000	4.0	
Ken08	7	6 M	SBMA	1	5	0	000000	0	0	0000	4.0	
Ken10	7	3 M	SBMA	0	5	0	0000000	0	0	0000	3.0	
Par01	7	2 F	PD	1	0	2	000000	0	0	0000	3.0	
Par02	8	1 F	PD	0	0	0	004010	0	0	0000	2.5	
Par03	7	5 F	PD	2	3	0	001000	0	0	1020	7.0	
Par04 Par05	6	9 F	PD	1	2	0	301000	0	0	00000	5.0	
Par06	7	4 F	PD	0	9	0	001010	0	e.	3 2 3 0	9.0	
Par07	6	5 M	PD	æ	0	0	000000	1	0	0000	3.0	
Par08	5	6 M	PD	2	0	0	0 14 0 0 0 0	0	0	0001	2.5	
Par10	6	5 M	PD	0	2	0	003020	0	0	0000	4.5	
Par11	7	ЗF	PD	0	0	0	001122	0	0	2000	2.5	
Par12	7	зм	PD	0	0	0	000000	0	0	0000	0.0	
Par14	5	6 M	PD	0	0	0	101020	0	0	1000	2.5	
Par15	7	O F	PD	0	0	0	RI <mark>- 1 4</mark> 5 5 5	0	0	0000	4.0	
Par16	5	5 M	PD	0	1	0	000000	0	0	1000	1.5	
Par17 Par18	6	4 M	PD	0	0	0	000010	0	0	0000	0.5	
Par19	6	9 M	PD	0	0	0	0000000	0	0	1000	3.5	
Par20	7	ом	PD	0	0	0	000000	0	0	0000	0.0	
ALS11	7	зм	ALS	0	0	0	N. 0 0 1 2 1	0	0	0000	1.5	
ALS12	7	2 M	ALS	1	0	2	000000	0	0	0000	3.0	
ALS14	8	1 F	ALS	0	0	0	N. 0 4 1 2 2	0	0	0000	3.0	
ALS15	7	4 M	ALS	0	1	2	100032	0	0	2000	7.5	
ALS16	7	3 F	ALS		3	0	000003	0	0	3000	5.5	
ALS17	7	1 F 7 F	ALS	0	0	0	0 0 0 0 0 3 3 0 0 0 1 0 0	0	0	0000	1.0	
ALS19	7	5 F	ALS	0	0	0	203342	0	0	0000	4.5	
ALS20	7	5 F	ALS	0	0	0	2000 32	\$	0	5000	7.0	
AoSO1	5	4 M	AoS	1	4	0	000000	0	5	1102	10.0	
AoS02 AoS03	2	<u>4 М</u> ом	AoS	1	0	- 0		0	1	0001	8.5	
AoSO4	6	1 F	AoS	0	5	0	001000	0	0	0001	4.0	
AoSO5	з	2 F	AoS	0	1	0	000002		3	2101	7.0	
AoSO6	7	2 F	AoS	0	3	0	000000	0	0	0000	3.0	
AoSO8	4	3 F 4 F	AoS	1	- 4	0	0000000	0	2	0001	7.5	
AoS09	7	2 F	AoS	0	4	2	000000	0	3	3 2 2 0	11.5	
AoS10	5	зм	AoS	1	4	0	100010	0	0	0001	6.5	

694 Appendix C.

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

