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Original Article

Quantitative assessment of motor speech abnormalities in idiopathic rapid eye movement sleep behaviour disorder

Jan Rusz [a,b,*], Jan Hlavnička [a], Tereza Tykalová [a], Jitka Bušková [b,c], Olga Ulmanová [b], Evžen Růžička [b], Karel Šonka [b]

[a] Faculty of Electrical Engineering, Department of Circuit Theory, Czech Technical University in Prague, Prague, Czech Republic
[b] Department of Neurology and Centre of Clinical Neuroscience, Charles University in Prague, First Faculty of Medicine, Prague, Czech Republic
[c] National Institute of Mental Health, Czech Republic

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ABSTRACT

Objective: Patients with idiopathic rapid eye movement sleep behaviour disorder (RBD) are at substantial risk for developing Parkinson's disease (PD) or related neurodegenerative disorders. Speech is an important indicator of motor function and movement coordination, and therefore may be an extremely sensitive early marker of changes due to prodromal neurodegeneration.
Methods: Speech data were acquired from 16 RBD subjects and 16 age- and sex-matched healthy control subjects. Objective acoustic assessment of 15 speech dimensions representing various phonatory, articulatory, and prosodic deviations was performed. Statistical models were applied to characterise speech disorders in RBD and to estimate sensitivity and specificity in differentiating between RBD and control subjects.
Results: Some form of speech impairment was revealed in 88% of RBD subjects. Articulatory deficits were the most prominent findings in RBD. In comparison to controls, the RBD group showed significant alterations in irregular alternating motion rates ($p = 0.009$) and articulatory decay ($p = 0.01$). The combination of four distinctive speech dimensions, including aperiodicity, irregular alternating motion rates, articulatory decay, and dysfluency, led to 96% sensitivity and 79% specificity in discriminating between RBD and control subjects. Speech impairment was significantly more pronounced in RBD subjects with the motor score of the Unified Parkinson's Disease Rating Scale greater than 4 points when compared to other RBD individuals.
Conclusion: Simple quantitative speech motor measures may be suitable for the reliable detection of prodromal neurodegeneration in subjects with RBD, and therefore may provide important outcomes for future therapy trials.

1. Introduction

Idiopathic rapid eye movement sleep behaviour disorder (RBD) is a parasomnia characterised by dream-enactment behaviours associated with REM sleep without muscle atonia [1]. Recent studies have shown that patients diagnosed with RBD are at increased risk for developing α-synucleinopathy, particularly Parkinson's disease (PD) or dementia with Lewy bodies (DLB), and less frequently multiple system atrophy (MSA) [2–4], with risk estimates of 33.1% at five years, 75.7% at 10 years, and 90.9% at 14 years after onset [5]. This high conversion rate to neurodegenerative disease provides a unique opportunity to observe the clinical development of parkinsonism or cognitive impairment [6]. Identifying predictive markers of neurodegeneration is essential [4,6] as they could provide invaluable information for future trials and disease-modifying therapies before the onset of motor and cognitive symptoms [7].

Motor speech disorder is a common clinical manifestation occurring in 70%–100% of patients with PD, DLB, and MSA, and typically appears early in the course of disease [8–11]. Hypokinetic dysarthria tends to be the dominant subtype in PD and DLB, whereas ataxic–hypokinetic dysarthria prevails in MSA [8–11]. Hypokinetic dysarthria affects primarily phonatory, articulatory, and prosodic speech subsystems, and may be related to numerous deviant dimensions such as reduced vocal loudness, poor voice quality, harshness, articulatory undershoot of vowels and consonants, dysrhythmia, articulatory decay, monopitch, monoloudness, variability of speech rate, and dysfluency [12]. Ataxic dysarthria is characterised by distorted articulation, reduced speech rate, and deviant prosodic modulations, particularly rhythmical irregularities during fast repetitive productions of syllables [12].

* Corresponding author. Department of Neurology and Centre of Clinical Neuroscience, Charles University in Prague, First Faculty of Medicine, Kateřinská 30, 12000 Prague, Czech Republic. Tel.: +420 224 352 287; fax: +420 224 311 081.
E-mail address: rusz.mz@gmail.com (J. Rusz).

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Speech disorders may be a prodromal sign of PD, as speech dysfunction is present in up to 90% of de novo PD patients [13–15]. Family members of PD patients have perceptually noted changes in speech before the diagnosis was established [16], and previous studies have reported cases in which reduced intonation variability was observed several years before the onset of the first motor symptoms [17]. Furthermore, ultrasonic vocalisation deficits were among the first prodromal markers of motor dysfunction in a murine model of PD [18]. Based on the Unified Parkinson's Disease Rating Scale, Postuma et al. [6] estimated that vocal and facial akinesia is the earliest indicator of parkinsonism in RBD patients, followed by rigidity, gait abnormalities, limb bradykinesia, and tremor.

An objective, quantitative assessment of speech in RBD is currently lacking. Speech evaluation is inexpensive, noninvasive, and simple to administer, and acoustic analyses provide objective, sensitive, and quantifiable information for the precise assessment of various deviant speech dimensions [13]. In addition, current advances in information and communication technologies have provided speech assessment the unique opportunity to be considered a simple screening test for the development of parkinsonism [13,14]. However, speech abnormalities in RBD should first be well explored.

Therefore, the aims of the current study were as follows: (1) to propose an acoustic methodology that would be sensitive to potential motor speech deficits in RBD; (2) to quantitatively characterise speech disorders in RBD; (3) to determine the most salient features of speech dysfunction in RBD, and to estimate their specificity and sensitivity in differentiating between RBD and healthy control subjects; and (4) to explore the relationship between speech and clinical findings to provide deeper insight into the pathophysiology of speech dysfunction in RBD.

2. Methods

2.1. Patients

A total of 16 consecutive Czech patients (10 men, 6 women), mean age 65.6 years [standard deviation (SD) 7.0 years], diagnosed with idiopathic RBD according to the International Classification of Sleep disorders diagnostic criteria, second edition [19], were included in the study. The examination consisted of detailed medical and pharmacological history, neurological assessment, and night polysomnography from 10 PM to 6 AM during a 1-day hospitalisation. Polysomnographic features of RBD were analysed from the chin and tibialis superficialis muscles according to the American Association of Sleep Medicine scoring rules [20]. Five subjects were treated with antidepressants before the diagnosis of RBD was established (Table 1), but only two subjects were receiving antidepressants at the time of the diagnostic polysomnography (RBD07, RBD15). Diagnostic investigation revealed that seven subjects fulfilled the criteria of obstructive sleep apnoea [19]. The mean apnoea/hypopnoea index (AHI) was 9.1 (SD 6.9); the AHI did not exceed the value of 20 in any RBD patient. The average number of periodic limb movements in sleep (PLMS) per one hour was 16.1 (SD 34.5); a value greater than 15 was found in four RBD patients (RBD05, RBD12, RBD14, RBD16).

At the time of speech investigation, nine of 16 patients were treated by clonazepam at bedtime to alleviate symptoms of RBD. None of the patients complained of motor or cognitive difficulties or had a history of treatment with antiparkinsonian medication or any other therapy influencing sleep, cognition, or motor features. All patients were examined by a movement disorders specialist (O.U.) and scored according to the Unified Parkinson's Disease Rating Scale motor subscore (UPDRS III). The clinical characteristics of the RBD subjects are summarised in Table 1.

The healthy control group consisted of 16 sex- and age-matched subjects (10 men, 6 women), mean age 65.6 years (SD 7.0 years), with no history of neurological or communication disorders or abnormalities of sleep. Each participant provided written informed consent. The study was approved by the Ethics Committee of the General University Hospital in Prague, Czech Republic, and has therefore been performed in accordance with the ethical standards laid down in the 1964 Declaration of Helsinki.

2.2. Speech examination

Speech recordings were performed in a quiet room with a low ambient noise level, in the afternoon, using a head-mounted condenser microphone (Beyerdynamic Opus 55, Heilbronn, Germany) placed approximately 5 cm from the subject's mouth. Speech signals were sampled at 48 kHz with 16-bit resolution. Recording was performed in each subject during a single session with a speech specialist (J.R.). All participants were instructed to perform three

Table 1
Clinical characteristics of RBD patients.

Patient no.	Sex	Age (y)	RBD symptoms duration (y)	Antidepressant therapy before RBD diagnosis	UPDRS III motor score	UPDRS III speech item 18	Clonazepam (mg/day)
RBD01	F	73	1	None	5	1	0.125
RBD02	M	54	8	None	1	0	0
RBD03	M	66	11	None	7	0	0.5
RBD04	M	59	3	None	2	0	0
RBD05	M	75	16	None	10	0	0.5
RBD06	M	70	10	None	7	1	0.5
RBD07	F	64	3	SSRI, NaSSA	9	1	0.5
RBD08	F	71	5	SNRI	12	0	0.5
RBD09	M	69	5	None	2	0	0
RBD10	F	57	1	None	3	0	0
RBD11	F	68	1	SSRI	7	0	1
RBD12	F	67	5	SSRI	2	0	0.125
RBD13	M	68	11	None	4	0	2
RBD14	M	65	12	None	1	0	0
RBD15	M	51	10	SSRI, SARI	0	0	0
RBD16	M	73	5	None	3	0	0
Mean (SD)		65.6 (7.0)	6.7 (4.6)		4.7 (3.6)	0.19 (0.40)	0.36 (0.53)

Abbreviations: F, female; M, male; NaSSA, noradrenergic and specific serotonergic antidepressant; RBD, rapid eye movement sleep behaviour disorder; SARI, serotonin antagonist reuptake inhibitor; SD, standard deviation; SNRI, serotonin–noradrenaline reuptake inhibitor; SSRI, selective serotonin reuptake inhibitor; UPDRS III, Unified Parkinson's Disease Rating Scale motor subscore.

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