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The place of perceptual analysis of dysarthria in the differential diagnosis of corticobasal degeneration and Parkinson's disease

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Abstract *Objective* To characterize the dysarthria in patients with corticobasal degeneration (CBD) and determine if analysis of speech in isolation helps to distinguish CBD patients from patients with Parkinson's disease (PD). *Methods* 60 subjects were assessed by means of perceptual analysis of speech: 15 patients with CBD, 15 patients with PD and 30 control subjects. A detailed profile was furnished with the help of 33 perceptual items. A global perceptual approach was used to classify patients by judges blind to the medical diagnosis. Rating scales were adapted to quantify the degree of spasticity and hypokinesia in the speech of each patient. *Results* Dysarthria was frequent in CBD even though it remained mild for a long period of time. Group analysis revealed the importance of temporal errors of speech control in CBD patients while voice disturbances were most frequent in PD patients. However, attempts to classify patients according to global perceptual analysis remained below a reasonable level of clinical acceptability. Finally, even though the widespread neuropathological changes suggest that deviant speech dimensions of several types of dysarthria might be found in CBD, evidence for a mixed dysarthria with presence of spastic elements could not be established. *Conclusion* The findings support the view that even though perceptual analysis is mandatory in the management of dysarthric patients, it does not help in the clinical differential diagnosis of CBD.

Key words dysarthria · speech · perceptual analysis · corticobasal degeneration

Introduction

Corticobasal Degeneration (CBD) is a sporadic, progressive disorder characterised by the association of an asymmetrical, dopa-resistant, akinetic-rigid syndrome and signs of cortical dysfunction such as apraxia, alien limb or sensory loss, often accompanied by other movement disorders such as myoclonus or dystonia [1, 2]. The diagnosis is often difficult as CBD has heterogeneous clinical presentations.

Dysarthria is a well recognized complication of parkinsonian disorders, especially multiple system atrophy (MSA) and progressive supranuclear palsy (PSP), but also CBD. However, perceptual characteristics of dysarthria in CBD have not been reported. It has been suggested that careful assessment of dysarthria can provide diagnostic information that assists in localizing

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disease processes [3,4]. This is based on the work of Darley et al. [3] who have classified dysarthria into several types based upon perceptual characteristics and linked these types to anatomic loci in the nervous system. Thus, dysarthria in parkinsonian syndromes such as PSP or MSA is considered mixed with combinations of spastic and hypokinetic components [4–6]. As patients with CBD also have diffuse lesions, we hypothesised that such patients would have mixed dysarthria, differing from hypokinetic dysarthria observed in PD.

Speech can be assessed by different clinical and instrumental tools. In spite of advances in instrumental analysis of dysarthria, perceptual evaluation remains the "gold standard" against which other measures must match up [7]. We therefore performed a perceptual speech analysis in patients with CBD, compared the findings with those in PD and control subjects. Three questions were asked: (1) What are the major perceptual abnormalities in the dysarthria of CBD and do they differ from PD? This step would allow one to determine the precise profile of abnormalities encountered in these patients. (2) Can expert judges distinguish dysarthria in CBD from dysarthria in PD by using a global perceptual approach? (3) Are there deviant spastic speech dimensions in the dysarthria of CBD patients? If those abnormalities are present in CBD, dysarthria could be considered as mixed, with spastic and hypokinetic components as has been found for other parkinsonian syndromes such as MSA and PSP [4–6]. The ultimate aim is to see whether dysarthric deficits can contribute to the differential diagnosis of PD and CBD.

Methods

Subjects

Sixty subjects were enrolled in the study: 15 patients with CBD, 15 patients with PD and 30 control subjects (CS). Patients had to fulfill respectively the modified diagnostic criteria defined by Lang et al. [8] for CBD and the clinical criteria of PD [9]. CBD patients were included prospectively in the present study, before communication disorders precluded exhaustive assessment. They were then matched with PD patients for sex, age and most of all severity of dysarthria as measured by an Intelligibility Score [10].

The control group included 30 native French speakers matched for age and sex with no history of a central nervous system or ENT disorder, speech disturbance, respiratory disease or a significant hearing or visual impairment. General demographic characteristics of the three groups are summarized in Table 1. Detailed clinical description of patients with CBD is reported in Table 2. Ten of these patients have previously been reported [11].

Methods

All patients were assessed using an intelligibility score (IS) in order to match patients according to the severity of speech impairment. We

Table 1 Demographic variables of the three groups

	CS	PD	CBD
Number	30	15	15
Sex, male/female	16/14	8/7	8/7
Age, years*	65 (7)	70 (6)	70 (7)
Disease Duration, years*	–	7.9 (3.2)	3.8 (1.3)
Intelligibility score (/24)*	24	20 (3)	20 (3)

* Values presented are means (standard deviations)
CS control subjects; *PD* Parkinson's disease; *CBD* Corticobasal degeneration

Table 2 Description of the CBD patients

Case	1	2	3	4	5	6	7	8	9	10	11	12	13	14	15
Demographic variables															
Age, years	73	77	78	73	70	78	67	71	67	69	65	53	78	56	69
Sex	M	M	F	M	M	F	M	F	M	F	M	F	F	F	M
Disease duration, years	2	5	1.5	0.5	4	3.5	1.5	5	2	5	5	2	3	5	4
Side of initial symptom	L	R	R	L	R	L	R	R	L	L	R	R	L	L	L
Intelligibility score	24	23	23	22	20	20	19	19	17	15	14	24	20	22	23
Clinical features															
Asymmetric parkinsonism	+	+	+	+	+	+	+	+	+	+	+	+	+	+	+
Myoclonus	–	–	+	–	–	+	–	–	–	+	–	–	+	–	–
Tremor (postural or action)	+	–	+	+	–	+	–	–	+	+	+	–	+	+	+
Dystonia	–	+	–	+	+	+	–	+	–	+	+	+	–	+	+
Limb apraxia	+	+	+	+	+	+	+	+	+	+	+	+	+	+	+
Sensory Loss	–	–	–	–	–	+	–	+	–	+	+	–	+	+	–
Alien limb	–	–	–	–	+	–	–	–	+	+	–	–	–	–	–
Supranuclear gaze palsy	–	–	–	+	–	+	–	–	+	–	+	+	+	+	–
Pyramidal signs	–	–	–	–	+	+	+	+	+	+	+	–	–	+	–
Dysarthria	–	+	+	+	+	+	+	+	+	+	+	–	+	+	+
Dysphagia	–	+	–	–	–	–	–	–	–	+	–	+	+	–	–

found it more appropriate to match patients according to the severity of dysarthria, as with similar disease durations patients with CBD often have more severe motor signs than patients with PD. This intelligibility score is a French adaptation of the Frenchay Dysarthria Assessment [12]. It is a score on 24 points where 24 corresponds to normal speech. The IS is a global measure which reflects the severity of dysarthria: a score between 18 and 23 corresponds to mild dysarthria (everything is correctly identified but speech shows perceptual impairments such as hypophonia, dysprosody or rate abnormalities); between 12 and 17, dysarthria is moderate and when the IS is below 12, dysarthria is considered as severe [13].

Speech samples were recorded from all 60 subjects. Fifty eight subjects read a standard paragraph of simple expository prose. In one case, a sample of conversational speech was used as the patient had difficulties in reading and in another case, repetition of sentences was used as spontaneous speech was too poor. Recording was done in a quiet room using a low noise unidirectional microphone maintained at a mouth-to-microphone distance of 30 cm. Thirty second samples were extracted to have standardized length passages, corresponding to the beginning of the paragraph. Each subject was identified by a number and the order of presentation was randomized for the listening task.

The speech samples of twenty speakers were scored twice in order to calculate intralistener reliability. These twenty samples were randomly embedded in the series of 60 samples. They corresponded to 5 CBD patients, 5 PD patients and 10 control subjects. A total of 80 samples was thus obtained.

Each speech sample was rated on a series of 33 dimensions. The original version of this scale had 32 dimensions with eight categories: pitch (four dimensions), loudness (four dimensions), vocal quality (seven dimensions), resonatory dysfunction (two dimensions), respiration (three dimensions), prosody (six dimensions), articulation (five dimensions) and intelligibility (one dimension) [14]. We have added a last dimension on the general impression of "bizarreness" of speech, corresponding to the loss of the naturalness of speech.

Each dimension was rated on a five-point equal-appearing interval scale of severity with 0 representing normal speech for this dimension and 4 representing severe deviation from normal. However, for the rating of rate, plus and minus signs were used to note the accelerated or slowed aspect of speech, for example –4 corresponded to severely slowed speech. Similarly, for pitch, positive values corresponded to pitch higher than normal and negative values for pitch lower than normal. Finally, intensity was also scored with a plus or minus sign.

Speech samples were rated by two judges (CO, PA) who were blind to the clinical status of the subjects (CBD, PD or CS). Judges were considered "experts", in terms of their experience with dysarthria. They were given unlimited time to listen to and rate the speech samples. For each sample, once all dimensions had been scored by each judge, a consensus score was obtained for each dimension. If the two judges had given the same score, this score was retained for the consensus. If there was a one point difference, the mean of the two scores was retained. If there was more than one point difference, the sample was listened again in order to obtain an agreement between the two judges and that score was retained. The same procedure was repeated for the 80 samples.

Interlistener agreement was defined as the percentage of ratings where the 2 judges gave exactly the same score and was calculated on the sixty original samples. Intralistener agreement was defined as the percentage of ratings where the same judge gave exactly the same score and was calculated using the twenty subjects whose samples were repeated twice.

The consensus ratings for each subject were used for the perceptual profiles of the three groups. The mean scale value (MSV) for each dimension was determined by calculating the mean ratings in each group: CBD, PD, CS. For the pitch, rate and intensity, absolute values were used. Student's t test was used to search for differences between the CBD and PD groups ($p < 0.05$).

In order to search for the discriminatory capacity of a global perceptual approach (Question 2), the same judges had to classify subjects in one of the three categories (CS, PD or CBD) according to their overall impression. Scores of correct classification were obtained and Kappa coefficient measured.

The third step consisted of searching for a spastic component in the speech of CBD patients which would help in classifying them as mixed dysarthria. The detailed perceptual assessments were thus used to obtain hypokinetic and spastic scores by adapting our items from the University of Michigan classification [4–6]. Briefly, Kluin and colleagues [4–6] analyzed speech by rating the severity of the deviant speech dimensions according to empirical criteria that were inspired by the classification of the Mayo Clinic [3]. Three lists of dimensions were constructed, corresponding to spastic, hypokinetic and ataxic dysarthria with each having 10 dimensions scored from 0 (unaffected) to 3 (severely affected). Weighting factors (WF) were secondarily applied to emphasize the deviant speech dimensions most characteristic of each type of dysarthria. Each patient had thus three scores ranging from 0 to 48, reflecting the degree of hypokinesia, spasticity and ataxia in speech, with higher scores corresponding to more severe impairment (for example, for the hypokinetic dysarthria: the maximum score of 48 resulted from the addition of the items low volume (3 points X 3WF) + monopitch (3 points X 2 WF) + loudness decay (3 points X 2 WF) + short rushes of speech (3 points X 2 WF) + rate (3 points X 2 WF) + imprecise consonants (3 points X 1 WF) + breathy voice (3 points X 1 WF) + palilalia (3 points X 1 WF) + inappropriate silences (3 points X 1 WF) + reduced stress (3 points X 1 WF). Table 3 shows the deviant speech dimensions used to obtain spastic and hypokinetic scores (the ataxic score was not used as CBD patients have no major involvement of the cerebellum). We chose not to use the item reduced stress as this is not relevant to French. We therefore have only 9 items for spastic dysarthria (with a maximum score of 42) and 9 for hypokinetic dysarthria (with a maximum score of 45).

Results

Overall mean interjudge agreement was 91 % (96.3 % for control subjects and 86.1 % for patients in CBD and PD groups). Mean intrajudge agreement was 88 %.

Table 3 Deviant Speech Dimensions for hypokinetic and spastic components of dysarthrias (adapted from the University of Michigan Classification)

Hypokinetic	Spastic
Low volume (3)	Strained voice (3)
Monopitch (2)	Low pitch (2)
Loudness decay (2)	Harsh voice (2)
Short rushes of speech (2)	Rate (2)
Rate (2)	Modification of phoneme duration (1)
Imprecise consonants (1)	Imprecise consonants (1)
Breathy voice (1)	Monoloudness (1)
Palilalia (1)	Hypernasality (1)
Inappropriate silences (1)	Monopitch (1)

The numbers in parentheses indicate weighting factors used in computing numerical scores for the deviant speech dimensions

Detailed perceptual profiles

Mean Scale Values (MSV) for CS were below 0.5 points for all speech dimensions. Table 4 shows the MSV for the speech dimensions above 0.5 points for the CBD and PD groups, as well as the number of occurrence. The mean speaking rate for CBD patients was 1.2/4. Ten patients had slow rate, while two patients had fast rate. In the PD group, the mean speaking rate was 1.0/4: 8 patients had slow rate and 5 patients had fast rate. For the pitch dimension, only PD patients had MSV > 0.5: nine patients had low pitch and two patients had high pitch. There was no significant difference between the CBD and PD groups for any dimension.

Correct classification rates with a global approach

Data concerning classification by expert judges according to their global impression is reported in Table 5. The overall rate of correct classification is of 67.5 %. Ninety three percent of control subjects were correctly classified while correct classifications rates were 47 % for PD patients and 37 % for CBD patients. Kappa coefficient was 0.46.

Table 4 Mean scale values (MSV) and occurrence (N) of the speech deviations found in the CBD and PD groups (only items with MSV > 0.5 are reported)

Speech deviations	CBD		PD	
	MSV	N	MSV	N
In the two groups				
Global dysprosody	1.6	12	1.3	11
"Bizarreness"	1.5	12	0.9	11
Rate	1.2	11	1.0	13
Monoloudness	1.1	13	1.1	13
Monopitch	1.0	10	1.0	10
Decrease of loudness level	0.9	11	1.0	11
Imprecise consonants	0.9	12	0.6	9
Only in the CBD group				
Prolonged intervals	0.8	7		
Only in the PD group				
Pitch level			0.9	11
Breathy voice			0.7	8
Harsh voice			0.7	8

Table 5 Correct diagnosis with a global approach on perceptual abnormalities of speech

	Number of correct predicted diagnosis		
	CS	PD	CBD
Medical diagnosis of CS (/60)*	56	2	2
Medical diagnosis of PD (/30)*	9	14	7
Medical diagnosis of CBD (/30)*	5	14	11

* Subjects are classified twice as each judge predicts the medical diagnosis

Arguments for mixed dysarthria in CBD

Fig. 1 summarizes the distribution of spastic and hypokinetic scores in the 3 groups of subjects. In control subjects (CS), the mean value for the spastic score was 2.15 (SD 2.95) and the mean value for the hypokinetic score was 0.9 (SD 1.33). These scores were respectively 9.6 (SD 6.40) and 9.2 (SD 6.9) for the PD group and 9.9 (SD 6.02) and 8.3 (SD 4.5) for the CBD group.

Discussion

Dysarthria was frequent in our series of CBD patients, present in 13 out of 15 subjects. While a frequency of 55 % has been reported in a review of the literature [15], dysarthria is much more frequent when it is prospectively and systematically assessed [11, 16]. Speech impairment remains mild for a long period even when general motor impairment is relatively severe [16].

There is no clear cut argument in the CBD literature for the effect of disease duration on the severity of dysarthria [11, 16, 17].

Data concerning dysarthria in CBD are rare and generally focus on its frequency. The present work is the first detailed study on the perceptual characteristics of speech in CBD. Our first question aimed to furnish a perceptual speech profile of a group of French patients

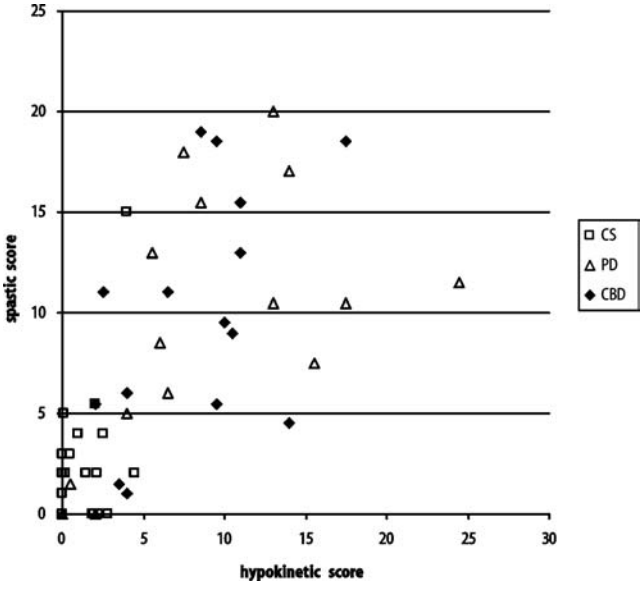

Fig. 1 Distributions of hypokinetic and spastic scores in control subjects (CS), parkinsonian patients (PD) and patients with corticobasal degeneration (CBD)

with CBD, compared with patients with PD and control subjects. Interlistener and intralistener reliability was high, supporting the validity of perceptual analysis. Dysprosody was frequent in both CBD and PD patients and encompassed several parameters such as monoloudness, monopitch, slow rate and global prosody. These are very frequent but unspecific abnormalities in dysarthria. The profile for patients with PD was largely similar to profiles for hypokinetic speakers of other languages such as English [3, 18] or Cantonese [19]. Particularly affected dimensions were those related to vocal quality such as harsh or breathy voice or pitch abnormalities. In contrast, patients with CBD presented with much less vocal abnormalities. It is interesting to note that their speech was considered "bizarre" in 80% of cases. The most frequently observed explanation was abnormalities in the temporal organization of speech with fluency disorders, slow rate and sometimes prolongations in the duration of phonemes. This suggests that even at an early stage some CBD patients might have distinctive speech characteristics that could help in the differential diagnosis. Unfortunately, while perceptual characteristics of dysarthria in PD have been well established, much less is known about dysarthria in other Parkinsonian syndromes. To our knowledge, there is no similar description of the perceptual abnormalities in CBD. Kluin and colleagues [4–6] have presented similar perceptual works concerning PSP and MSA patients. However, results cannot be compared as these authors obtained global composite scores rather than precise perceptual profiles.

The purpose of the second part of this work was to examine the degree with which experienced judges can use perceptual analysis alone to identify specific types of dysarthria. Many clinicians who assess patients with dysarthria routinely attempt to analyze perceptual features of speech to determine dysarthria types. The usefulness of perceptual evaluation as an aid to identify lesion locus has been described [3]. If perceptual evaluation is to be considered an effective and reliable aid in diagnosis and classification, it becomes clinically relevant to determine the accuracy of this method when used in isolation. However, few studies have so far tried to diagnose specific dysarthria types with perceptual analysis alone [7, 20, 21]. Judges blinded to the medical diagnoses listened to audiotapes containing several types of dysarthria and were asked to derive the neurological disease or the dysarthria type for each patient: whatever the response procedure overall accurate identification rates were low, between 19% and 56% [20]. The authors suggested that an overall impression after listening to the sample rather than an analysis of isolated dimensions might lead to more successful classification. Chenery [7] tried to distinguish between ataxic, hypokinetic and spastic dysarthrias using a discriminant function analysis: five speech dimensions correctly classified 89% of cases. However, it was the statistical combination of these items which led to correct classification as no one item contributed massively to differential diagnosis when entered in isolation. These studies therefore call into question the validity of using results of perceptual assessment alone to diagnose types of dysarthria. In the present work, PD patients should classically have hypokinetic dysarthria and those with CBD will be in the mixed category due to diffusion of lesions to sites other than the basal ganglia (motor cortex, pyramidal tract).

Control subjects were correctly classified in 93.3% of cases, suggesting that judges can easily distinguish abnormal speech even when dysarthria is mild. Correct classification rates of CBD and PD were below a reasonable level of clinical acceptability even though the Kappa analysis reveals that these scores are better than simple random assignment. Frattali and Sonies [16] reported that 57% of their patients had a predominantly hypokinetic profile and that there was a trend for mixed rather than pure dysarthrias from 3 years of disease duration or longer, probably owing to the involvement of multiple motor systems as the disease progresses. In the present study, only one third of the CBD patients were correctly classified. However, among the CBD patients with 3 years of duration or longer, more than half of them (5/9) were correctly identified. It can thus be argued that patients included in the present study had mostly mild dysarthria making it difficult to use such a clinical approach and that more specific abnormalities could be found in CBD patients with a long disease duration. However, two arguments can counter this suggestion. First, clinical relevance of differential diagnosis of speech motor types is most important in the early stages of the disease and not when all clinical signs become prominent. Second, CBD patients with more advanced disease often have communication disorders other than dysarthria such as echolalia or speech apraxia which help the diagnosis more than the characteristics of dysarthria per se [22]. In summary, even though detailed analysis of speech in groups of patients with CBD and PD reveal some differences, a global approach is not sufficient to distinguish between the two groups.

In the third part of our work, we searched for the presence of spastic elements in the speech of patients with CBD by using the method developed by Kluin and colleagues to analyze speech in PSP and MSA. A first study conducted with 44 patients with PSP revealed that all patients had a mixed dysarthria with deviant speech dimensions reflecting a combination of two or more of the following types of dysarthria: spastic, hypokinetic and ataxic [6]. In a second study with 14 patients with post mortem confirmed diagnosis of PSP, all patients had mixed dysarthria with hypokinetic and spastic dysarthria and 9 patients had also ataxic components

[4]. Finally, a similar study was conducted on 46 patients with MSA: again, all patients had dysarthria with combination of hypokinesia, ataxia or spasticity [5].

Adaptations of these clinical scales were used to assess spastic and hypokinetic components of speech in patients with CBD and PD: the two groups of patients could not be distinguished. In fact, patients with PD had spastic scores similar to those observed in CBD patients while theoretically they should have hypokinetic and not spastic abnormalities. This brings into question the validity of the approach proposed by Kluin. In fact, several factors can explain this result. The choice of the criteria for the three scales was empirical, based on clinical experience with some redundancy as abnormalities of rate, monopitch and imprecise consonants are present in both the spastic and hypokinetic scales. Most important, these scales have been directly tested on patients supposed to have mixed dysarthria, such as PSP or MSA patients, without confirming their validity in homogeneous groups such as pure cerebellar subjects that should have specific and isolated perturbations of the ataxic scale. Therefore, further work is necessary to improve the validity of such scales before using them to identify elements of mixed dysarthria in patients with parkinsonian syndromes.

In conclusion, detailed group analysis of speech underlines the predominance of vocal abnormalities in PD while temporal aspects of speech seem more disturbed in CBD. However, correct classification rates using a global perceptual approach are clearly below a reasonable level of clinical acceptability for use in differential diagnosis. The role of perceptual analysis in conjunction with instrumental measures such as acoustic analysis requires further study. Finally, we could not distinguish the profiles of PD and CBD patients using spastic and hypokinetic scales. Further studies are necessary to determine the validity of such an approach in large and homogeneous groups of patients.

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