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Speech dysprosody but no music 'dysprosody' in Parkinson's disease

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ABSTRACT

Parkinson's disease is characterized not only by bradykinesia, rigidity, and tremor, but also by impairments of expressive and receptive linguistic prosody. The facilitating effect of music with a salient beat on patients' gait suggests that it might have a similar effect on vocal behavior, however it is currently unknown whether singing is affected by the disease. In the present study, fifteen Parkinson patients were compared with fifteen healthy controls during the singing of familiar melodies and improvised melodic continuations. While patients' speech could reliably be distinguished from that of healthy controls matched for age and gender, purely on the basis of aural perception, no significant differences in singing were observed, either in pitch, pitch range, pitch variability, and tempo, or in scale tone distribution, interval size or interval variability. The apparent dissociation of speech and singing in Parkinson's disease suggests that music could be used to facilitate expressive linguistic prosody.

1. Introduction

Impairment of singing would perhaps not be considered the most salient symptom of a movement disorder such as Parkinson's disease (PD). The expressive qualities of music, however, depend largely upon the same features which characterize expressive linguistic prosody: pitch, rhythm, and sound intensity, aspects of speech which can be severely impaired in PD (Sapir, 2014). Nevertheless, clinical assessment does not generally probe the singing abilities of these patients, and even more significant, scientific investigation of the issue is almost non-existent. One study, based solely on the singing of a scale, has suggested that Parkinson patients are no longer able to sing accurately (Rigaldie, Nespoulous, & Vigouroux, 2006).

It has previously been remarked that music is a useful tool for the study of the functional organization of the brain (Zatorre, 2005). That is particularly so in the case of Parkinson patients for whom the facilitating effects of music on gait have been well documented (Bernatzky, Bernatzky, Hesse, Staffen, & Ladurner, 2004; de Bruin et al., 2010; Hayashi, Nagaoka, & Mizuno, 2006; Ito et al., 2000; McIntosh, Brown, Rice, & Thaut, 1997; Rubinstein, Giladi, & Hausdorff, 2002; Thaut et al., 1996). Understanding how this takes place should be a main concern of current neuroscientific research. The immediate aim of the present study was to determine whether the previously demonstrated impairments of expressive linguistic prosody were paralleled by similar melodic impairments in patients' singing. Our hypothesis was that the well-documented effect of music on the gait of Parkinson patients is not specific to locomotion, but that it extends to vocal behavior as well. We therefore expected the singing of patients to be quite similar to that of healthy individuals, while their speech is not.

PD is a progressive movement disorder characterized by the loss of the dopaminergic neurons in the substantia nigra. Main motor symptoms of the disease are bradykinesia, rigidity, and tremor (Bartels & Leenders, 2009; Jankovic, 2008), symptoms that can be partly alleviated by dopamine repletion (Connolly & Lang, 2014). Remarkably, however, patients may exhibit improvement in walking speed and stride length while listening to music, particularly music with a salient beat (Dalla Bella, Benoit, Farrugia, Schwartze, & Kotz, 2015; Hove & Keller, 2015; Lim et al., 2005). While they may be severely impaired in their ability to walk, under the influence of 'groovy' music (Madison, 2006) some patients are even able to dance (Volpe, Signorini, Marchetto, Lynch, & Morris, 2013). The innate, largely human, capacity for musical beat induction (Fitch, 2012; Grahn & Brett, 2007; Honing, 2012; Large & Snyder, 2009; Schachner, Brady, Pepperberg, & Hauser, 2009) seems to play a role in the elicitation and synchronization of movement in patients with Parkinson's disease, apparently circumventing neural circuits devastated by the disease (Grahn & Brett, 2009).

Besides bradykinesia, rigidity, and tremor, PD is characterized by hypokinetic dysarthria, a term referring to a variety of speech

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abnormalities such as reduced volume, poor enunciation, and 'flat' prosody (Cheang & Pell, 2007; Fox & Ramig, 1997; Walsh & Smith, 2012). Prosody may be defined as the patterned distribution of stress, intonation and other phonatory features in speech (Scott, Caird, & Williams, 1984). Wennerstrom (2001) calls it 'the music of everyday speech'. Her reference to the term 'music' is an allusion to the word $\pi\rho o\sigma\omega\delta\acute{\iota}\alpha$ (prosodia) whose original meaning connoted the ancient Greek practice of singing poetry instead of reciting it. Prosodic cues are used to convey emotion (Adolphs, 2002) as well as to resolve syntactic ambiguities (Steinhauer, Alter, & Friederici, 1999). The monotone character of patients' speech (Holmes, Oates, Phyland, & Hughes, 2000) suggests falsely that they are uninterested and emotionally detached (Benke, Bösch, & Andree, 1998; Mikos et al., 2009; Pitcairn, Clemie, Gray, & Pentland, 1990).

Impairment of expressive linguistic prosody, one of the most conspicuous features of Parkinsonian dysarthria (Skodda, Rinsche, & Schlegel, 2009), makes it difficult for patients to be understood (Blonder, Gur, & Gur, 1989; Pell, Cheang, & Leonard, 2006). They are frequently unable to make an audible distinction between compound nouns (a greenhouse) and noun phrases (a green HOUSE) or to emphasize salient words in a sentence (Where do you think YOU are going?). Patients do not succeed in producing the rising pitch that distinguishes a question from a statement (Darkins, Fromkin, & Benson, 1988; Pell et al., 2006). Sentences exhibit incongruent contour patterns, going up and down at the wrong places (MacPherson, Huber, & Snow, 2011), and breathing pauses do not always take place at syntactic boundaries (Huber, Darling, Francis, & Zhang, 2012).

That impairments of expressive linguistic prosody in PD are not only due to a general loss of motor abilities can be deduced from the accompanying impairments of receptive prosody (Monetta, Cheang, & Pell, 2008; Pell & Leonard, 2003; Pell & Monetta, 2008; Schröder et al., 2006). Patients fail to recognize prosodically communicated emotion (Ariatti, Benuzzi, & Nichelli, 2008; Dara, Monetta, & Pell, 2008; Gray & Tickle-Degnen, 2010; Schröder, Nikolova, & Dengler, 2010; Ventura et al., 2012; Yip, Lee, Ho, Tsang, & Li, 2003), and when meaning is being signaled by prosodic inflection, they may even fail to understand what is being said (Blonder et al., 1989; Lloyd, 1999; Pell, 1996; Scott et al., 1984).

A few studies have investigated patients' recognition of emotions in music (Lima, Garrett, & Castro, 2013; van Tricht, Smeding, Speelman, & Schmand, 2010). To our knowledge, however, no studies have investigated the role of auditory cues in Parkinson patients' comprehension of music. On the other hand, a recent study showed that patients' speech processing (as indicated by EEG) could be enhanced by first listening to music that exhibited a metric similarity to the spoken text. Semantic and syntactic processing of a trochaic text was significantly better when the spoken text was preceded by marching music than when it was preceded by a waltz (Kotz & Gunter, 2015). Moreover, in two different case studies, it has been observed that intelligibility could be enhanced by singing. Intelligibility was poorer when the text was spoken than when it was sung (Ferriero, Bettoni, Picco, Massazza, & Franchignoni, 2013; Kempler & van Lancker, 2002).

These results suggest that both expressive and receptive 'prosodic' aspects of music are spared in PD and that music might have a similar facilitating effect on vocal behavior as it has on gait, a hypothesis that was explored in the present study by comparing the singing of Parkinson patients with that of healthy controls matched for age and gender. The focus of the study was not on expert music performance, but rather on the general vocal ability of non-musicians to sing a familiar tune or to improvise a melodic continuation to an antecedent phrase.

Our main hypothesis was that during the vocal rendition of familiar melodies and the singing of improvised melodic continuations to antecedent phrases, no differences between patients and healthy individuals would be observed. In order to confirm the presence of expressive linguistic prosodic impairments in the patient group, recordings were made of spontaneous oral autobiographical narratives and the rhythmic recitation of song lyrics. Dysprosody was assessed in two randomized aural discrimination tests in which ten neurologically skilled assessors (five senior neurologists and five residents in neurology) listened to soundbites of the autobiographical narratives of all participants and differentiated patients from healthy controls on the basis of aural perception. Dysprosody was further quantified by digital speech analysis of the recordings.

2. Material and methods

The present study was approved by the Medical Ethics Committee of the University Medical Center Groningen, Groningen, The Netherlands. All participants gave written informed consent in accordance with the Declaration of Helsinki (2008), prior to participation. In addition, patients gave written informed consent granting access to classified information concerning their medication.

2.1. Participants

Fifteen Parkinson patients, many of whom played a music instrument or sang in a choir, but none professionally, were recruited for this study: six males and nine females, mean age (±SD): 65 (±8) years. Fifteen healthy participants, mean age 65 (±8) years, with similar musical interests, matched for age and gender, were recruited as controls. Patients were recruited via the local patient society as well as by advertisement on the website of the Dutch Parkinson Society. Of the nineteen patients who responded, four patients were excluded on the basis of additional pathology (CerebroVascular Accident), treatment (Deep Brain Stimulation), career (semiprofessional musician), and in one case, general inability to sing. Eight patients had left-asymmetric symptom involvement, of whom one was affected bilaterally at the time of testing. Seven patients had right-asymmetric involvement, of whom one was affected bilaterally at testing. For ethical reasons, patients were not requested to refrain from taking their normal doses of (dopamine repletion) medicine.

As patients were recruited from all over The Netherlands, data were acquired in the homes of the participants by one of the researchers (RH) who holds a master's degree in Human Movement Science as well as two degrees in music performance. Acquisition in the homes of the patients made on-the-spot disease quantification on the Unified Parkinson's Disease Rating Scale (Movement Disorder Society Task Force on Rating Scales for Parkinson's Disease, 2003) impossible. The visiting researcher (RH) estimated the Hoehn & Yahr score during acquisition (Hoehn & Yahr, 1967) and obtained written consent from each patient permitting the participating neurologist (BMdJ) to acquire medical information from the patient's consulting neurologist which, however, did not consistently include UPDRS scores. Based on the available information, BMdJ established the Hoehn & Yahr scores and computed the LEDD (Levodopa Equivalent Daily Dose). Mean disease duration was 7.3 years (±3.5); mean Hoehn & Yahr score: 2 (±0.19); mean LEDD: 835 (±537). Individual Hoehn & Yahr scores, disease duration (years since diagnosis), and LEDD are reported in Table 1.

2.2. Experimental protocol

2.2.1. Speech tasks

While the assumption was that Parkinson patients would suffer from dysprosody, it was important to confirm its presence in the