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Metacognitive knowledge of olfactory dysfunction in Parkinson's disease

Theresa L. White [a,b], Abbas F. Sadikot [c], Jelena Djordjevic [c,*]

[a] Le Moyne College, United States
[b] SUNY Upstate Medical University, United States
[c] Department(s) of Neurology and Neurosurgery, Montreal Neurological Institute and Hospital, McGill University, Canada

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ABSTRACT

It is well known that patients with Parkinson's Disease (PD) suffer from olfactory impairments, but it is not clear whether patients are aware of their level of deficit in olfactory functioning. Since PD is a neurodegenerative disorder and its progression may be correlated with olfactory loss (Ansari & Johnson, 1975; but see also Doty, Deems, & Stellar, 1988), it is possible that these patients would be subject to metacognitive errors of over-estimation of olfactory ability (White & Kurtz, 2003). Nineteen non-demented PD patients and 19 age-matched controls were each given an objective measure of olfactory identification (the UPSIT, Doty, Shaman, Kimmelman, & Dann, 1984) and a subjective measure involving a questionnaire that asked them to self-rate both their olfactory function generally and their ability to smell each of 20 odors, 12 of which were assessed on the UPSIT. All of the PD patients showed impaired olfactory ability, as did 7 of the controls, according to the UPSIT norms. Self-rated and performance-based olfactory ability scores were significantly correlated in controls ($r = .49$, $p = .03$) but not in patients with PD ($r = .20$, $p = .39$). When the 12 odors common to both the self-rated questionnaire and UPSIT were compared, PD patients were less accurate than controls ($t(36) = -4.96$, $p < .01$) at estimating their own ability and the number of over-estimation errors was significantly higher ($t_{\text{one-tailed}}(29) = 1.80$, $p = .04$) in PD patients than in the control group, showing less metacognitive awareness of their ability than controls. These results support the idea that olfactory metacognition is often impaired in PD, as well as in controls recruited for normosmic ability (Wehling, Nordin, Espeseth, Reinvang, & Lundervold, 2011), and indicate that people with PD generally exhibit over-estimation of their olfactory ability at a rate that is higher than controls. These findings imply that PD patients, unaware of their olfactory deficit, are at greater risk of harm normally detected through olfaction, such as smoke or spoiled foods.

1. Introduction

Parkinson's Disease (PD) is a neurodegenerative disorder that affects between 7 and 10 million people worldwide (Parkinson's Disease Foundation, 2015). Patients with PD suffer from a wide range of motor and non-motor symptoms (e.g., Chaudhuri, Healy, & Schapira, 2006; Müller, Reichmann, Livermore, & Hummel, 2002). The motor deficits include bradykinesia (slowness of movement), rigidity, balance, gait impairment, and tremor (Parkinson, 2002 [1817]; Postuma et al., 2015). The non-motor symptoms include disorders of mood, cognition, gastrointestinal function, and sensory systems, including olfaction (Halliday, Barker, & Rowe, 2011). While traditionally seen as primarily a dopaminergic disorder affecting the substantia nigra and striatum, current understanding of the disease extends to multiple neurotransmitter systems and the underlying neuropathology is far more complex than initially thought (Sulzer & Surmeier, 2013).

Olfactory dysfunction is one of the earliest non-motor symptoms of PD and thus is a predictor of disease onset (Postuma, Gagnon, & Montplaisir, 2010), albeit a somewhat imprecise one, since a number of other neurological conditions are also associated with olfactory loss (Haehner et al., 2007; Ross et al., 2008). Olfactory deficits in the PD population are extremely prevalent, with some estimates indicating that over 90% of patients suffer from some impairment of their sense of smell (Doty, 2012a; Haehner et al., 2009). The olfactory system is broadly impaired in people with PD, with bilateral difficulties including higher olfactory thresholds (Ansari & Johnson, 1975; Quinn, Rossor, & Marsden, 1987), diminished olfactory identification and discrimination (Müller, Müngersdorf, Reichmann, Strehle, & Hummel, 2002; Ward, Hess, & Calne, 1983), and impaired olfactory memory (Lehrner, Brucke, Dal-Bianco, Gatterer, & Kryspin-Exner, 1997). The olfactory deficits in this patient population are severe (Doty, 2012b, p. 534), which can be a useful part of a differential diagnosis

* Corresponding author at: Montreal Neurological Institute, 3801 Rue University, Montreal, QC H3A 2B4, Canada.
E-mail address: jelena.djordjevic@mcgill.ca (J. Djordjevic).

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as other motor disorders generally do not have accompanying olfactory loss to this extent (Doty et al., 1993).

Normal olfactory functioning is essential to a variety of daily activities, many of which contribute to safety and improve quality of life (Croy, Nordin, & Hummel, 2014; Kurtz, Emko, White, Belknap, & Kurtz, 1995). These activities include detection of dangerous substances such as natural gas, identification and regulation of suitable nutritional substances (Stevenson, 2010), as well as the enjoyment in a number of contexts. Given that olfactory ability is impaired in patients with PD, it is possible, even likely, that the important day-to-day functions of olfaction are also impaired in this population, similarly to people suffering olfactory loss due to other aetiologies (Hummel & Nordin, 2005; Miwa et al., 2001).

Although one perhaps cannot compensate for a diminished quality of life, knowing that one has a deficit in olfactory ability allows an individual to take compensatory measures to improve safety in daily life. People with diminished olfactory abilities can take steps to protect themselves from dangers normally detected via olfaction, such as labeling stored food with dates to avoid spoiled food or installing and regularly verifying detectors for natural gas leaks. However, these steps can only be taken if someone is metacognitively aware that they have an olfactory deficit.

Anosognosia, which can be defined as a deficit in metacognitive knowledge or a decrement in awareness of one's own thought processes (Rosen, 2011), can manifest as either an over- or under-estimation of ability (White & Kurtz, 2003). Over-estimation of ability is of greater concern, because it can lead people to erroneously believe that they are able to detect environmental threats via an impaired sensory or cognitive system. Over-estimation of ability is reasonably common in neurodegenerative diseases; for example, estimates of anosognosia are as high as 80% for memory ability in Alzheimer's Disease (AD; Agnew & Morris, 1998). Awareness of different deficits within a particular neurodegenerative disorder can vary; metacognitive knowledge for some aspects of a neurological condition do not necessarily indicate awareness of other symptoms, as patients with PD show varying degrees of awareness in cognitive, social, and functional deficits (Leritz, Loftis, Crucian, Friedman, & Bowers, 2004). Anosognosia seems to be partially the result of deficits in memory (Duke, Seltzer, Seltzer, & Vasterling, 2002) in which new information about abilities are not updated, but emotion and motivation are also involved in the creation of this syndrome (e.g., Michon, Deweer, Pillon, Agid, & Dubois, 1994). Learning about one's abilities begins with accurate perception of either successes or failures during task performance and the initiation of a self-evaluative process to update stored semantic representations of self. People with anosognosia seem to have an inability to update the representation of their aptitudes that may be mediated by memory, emotion, or other cognitive factors (Rosen, 2011).

In contrast to the awareness of other sensory modalities, olfactory awareness appears to be impaired in a significant proportion of the general population. Though the majority of individuals are well aware of their olfactory abilities, a sizable number of middle and older-aged individuals who believe themselves to be normosmic actually show some level of olfactory deficit (Nordin, Monsch, & Murphy, 1995). Awareness of olfactory loss seems to be inversely related to age, so that older individuals are less aware of their olfactory loss (Wehling, Nordin, Espeseth, Reinvang, & Lundervold, 2011). In addition, individuals who are unaware of their olfactory loss have also been reported to score more poorly on tests of cognitive and neurological functioning than other similarly aged individuals (Devanand et al., 2000; Wehling et al., 2011).

In addition to the healthy general population, metacognitive errors regarding olfactory ability are also seen in a variety of neurological disorders, such as AD (Devanand et al., 2000) and the Parkinson Dementia Complex of Guam (Doty et al., 1991). Even patients presenting to a Smell and Taste Disorders Clinic may be unaware of the extent of their olfactory deficits (White & Kurtz, 2003). In this population, however, metacognitive awareness of olfactory ability seems to be less a function of age, and more related to the rate at which olfactory loss occurred. Patients who experience a rapid loss of their sense of smell, such as through a head injury, tend to under-estimate their olfactory ability, while those who lost their sense of smell more gradually, such as through age-related loss, tend to over-estimate their olfactory performance (White & Kurtz, 2003).

While olfactory impairment in PD is well documented, specific awareness of patient decline in olfactory functioning is considerably less well explored. Doty, Deems, and Stellar provided evidence on this topic in 1988, suggesting that than 80% of people with PD failed to appreciate their own dysfunction level. However, metacognition was not the focus of that study, and as such, only a single, general question about chemosensory functioning was asked to those participants ("Do you suffer from any smell or taste problems?"), rather than a thorough investigation. Furthermore, neither the medical community nor the patients themselves were particularly aware of the association between olfactory impairment and PD in the 1980s. Today, olfactory deficits are recognized regularly as one of the non-motor symptoms of the disease.

The present study seeks to expand the exploration of anosognosia in PD by examining awareness of olfactory loss in more detail. We addressed three questions. First, since olfactory loss is now a well-known symptom of PD (Doty, 2012a), we questioned whether the level of anosognosia was as high as previously reported (Doty, Deems, & Stellar, 1988). Next, we wondered about the types of metacognitive errors that might be made by people with PD, i.e. whether under-estimations or over-estimations of ability would be more frequent. Since PD is a slow and progressive disorder that has been reported to be correlated with olfactory loss (Ansari & Johnson, 1975), we predicted that these patients would therefore be subject to errors of over-estimation of olfactory ability (White & Kurtz, 2003). Lastly, because previous assessment of anosognosia of olfactory ability in PD was gathered as the result of a general question (Doty et al., 1988), patients might have responded relative to gustation, olfactory detection, or olfactory identification. We wanted to examine whether asking specific questions regarding olfactory identification ability would yield a different pattern of metacognitive status from such a general question, since some evidence has been provided that framing of the question may be important to estimates (Wehling, Lundervold, & Nordin, 2014).

In the present study, we assessed metacognitive ability by directly comparing olfactory performance with self-report of olfactory ability (both general and specific estimates) as a means of answering these questions. Self-report of ability is one of the typical ways to measure anosognosia (Rosen, 2011). Self-report has been proposed as a potential alternative to direct testing of olfactory ability by physicians, who are often pressed for time in examining patients. Initial reports indicate good levels of prediction of olfactory performance (Takebayashi et al., 2011), although other studies have reported that self-estimates of olfactory ability are relatively undependable (Bahar-Fuchs, Moss, Rowe, & Savage, 2011; Shu et al., 2009). The design of the present study will enable examination of the validity of self-report as a quick method of evaluating olfactory disorders, as well as consideration of the awareness of olfactory loss.

The aim of the present study was to assess olfactory metacognition in patients with PD and a group of healthy volunteers. Based on the previous literature and the nature of the disorder, we hypothesized that anosognosia would be highly prevalent in these patients, and would be particularly characterized by errors of