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Low dimensional temporal organization of spontaneous eye blinks in adults with developmental disabilities and stereotyped movement disorder

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ABSTRACT

This study investigated the mean rate and time-dependent sequential organization of spontaneous eye blinks in adults with intellectual and developmental disability (IDD) and individuals from this group who were additionally categorized with stereotypic movement disorder (IDD + SMD). The mean blink rate was lower in the IDD + SMD group than the IDD group and both of these groups had a lower blink rate than a contrast group of healthy adults. In the IDD group the n to $n + 1$ sequential organization over time of the eye-blink durations showed a stronger compensatory organization than the contrast group suggesting decreased complexity/dimensionality of eye-blink behavior. Very low blink rate (and thus insufficient time series data) precluded analysis of time-dependent sequential properties in the IDD + SMD group. These findings support the hypothesis that both IDD and SMD are associated with a reduction in the dimension and adaptability of movement behavior and that this may serve as a risk factor for the expression of abnormal movements.

Stereotypic movement disorders (SMD) are particularly prevalent in individuals with intellectual and developmental disability (IDD) (Baumeister & Forehand, 1973; Cooper & Dourish, 1990; Sprague & Newell, 1996), though stereotypy is also evident in a number of other population groups and species, including human infants (Thelen, 1996) and caged animals (Berkson, 1967; Mason & Rushen, 2007). The pathophysiological basis of stereotypies in neurodevelopmental disorders is not well understood. There is growing evidence, however, that these behaviors are associated with alterations in cortical-basal ganglia circuitry (Lewis, Tanimura, Lee, & Bodfish, 2007). This is consistent with long-standing observations that dopamine agonists (e.g., amphetamine) acting on the basal ganglia reliably induce stereotyped behavior in a wide range of animal species (see Cooper & Dourish, 1990). In addition, stereotyped behavior that has been shown to be a predictable consequence of early social deprivation in non-human primates was shown to be associated with dopamine receptor supersensitivity (Lewis, Gluck, Beauchamp, Keresztury, & Mailman, 1990), loss of dopamine innervation in striatum and dopamine cells in substantia nigra, and decreases in striatal neuropeptide staining (Martin, Spicer, Lewis, Gluck, & Cork, 1991).

It has been proposed that spontaneous blink rate is a valid and robust behavioral index of the status of dopaminergic function in the system (Bodfish, Powell, Golden, & Lewis, 1995; Lewis, Gluck, Bodfish, Beauchamp, & Mailman, 1996). The

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mean rate of spontaneous eye blinks has been shown to vary over changes of age through the life span and as a function of particular disease states (Caplan, Guthrie, & Komo, 1996; Schellini, Sampaio, Hoyama, Cruz, & Padovani, 2005; Zametkin, Stevens, & Pittman, 1979). Importantly, in the context of this paper, the mean rate of spontaneous eye blinks has been shown to be lower in adults diagnosed with stereotyped movement disorder (SMD) (Bodfish et al., 1995; MacLean et al., 1985; Roebel & MacLean, 2007).

The hypothesis of a relation of the mean duration between eye blinks and the dopaminergic status of the system provides a natural and relatively convenient experimental behavioral index of pathophysiology of SMD. However, the mean of cyclical processes does not capture or exploit the richness of the dynamics of a time series and may even be misleading as to the dynamics and clinical status of a system (Bassingthwaighte, Liebovitch, & West, 1994; West, 2006). For example, there may be no difference between the mean heart rate of individuals over a segment of time but the heart rate could differ significantly in the time- and frequency-dependent structure (Lipsitz & Goldberger, 1992). Thus, an examination of the dynamics of a time series of spontaneous eye blinks beyond the mean rate could in principle provide a more informative and robust evaluation of SMD and related abnormal movements in IDD.

A striking example of this time series approach in repetitive movement disorders is that of Peterson and Leckman (1998) who studied the time-dependent relations of the time intervals between tics through a variety of analysis methods. The results showed that the tic intervals were not statistically independent and that the frequency distribution of the tic interval duration followed an inverse power law.[1] These findings together with those from other analyses gave preliminary evidence for the presence of a fractal, deterministic and possibly chaotic processes in the tic time series. Furthermore, this study showed that time series methods are useful in the analysis of repetitive movement disorders and the characterization of symptom severity.

In a series of previous studies investigating the dynamics of stereotyped movements in posture and action tasks we have shown that the sequential properties of the movement time series reveal important properties of the system dynamics and differences between population groups with IDD (see Newell, 1996, 1997). A general finding is that the dimension of the movement output is progressively lower in instrumental tasks in individuals who display SMD + IDD when contrasted with individuals with IDD who do not exhibit SMD (Bodfish, Parker, Lewis, Sprague, & Newell, 2001). Expressed another way, SMD progressively reduces the number of functional degrees of freedom (dimension) of the motor behavior beyond that of individuals with IDD but not recognized as exhibiting SMD, even in goal-oriented movement tasks.

In this study we examined the organization of the spontaneous eye blinks of individuals with IDD and of age-matched healthy adults, a contrast that has not been made directly. In the IDD group we also contrasted the organization of eye blinks of individuals categorized with SMD and those that were not. Based on the previous findings of the progressive reduced dimension of motor behavior in IDD and IDD + SMD (Newell, 1996, 1997) it was hypothesized that the organization of eye blinks would also show reduced dimensionality with IDD that would be further reduced with SMD. Thus, we anticipated that there would be a reduction in both the mean blink rate and the dimension of the dynamics of natural eye blinks in intellectual and developmental disability (IDD) that would be magnified in individuals also diagnosed with SMD.

1. Methods

1.1. Participants

The participant pool was composed of two groups: one with intellectual and developmental disability (IDD) and a contrast group of age-matched healthy adults. The IDD group was further sub-divided into two subgroups: participants with comorbid stereotyped movement disorder (IDD + SMD) and the remaining participants who did not display stereotyped behaviors (IDD). The participants in the IDD group resided at a public residential facility and were recruited from a larger group of persons with IDD who resided at the facility ($N = 290$).

The recruitment was based on the following criteria: (a) adult between the ages of 18 and 60 years, (b) diagnosed with severe IDD, (c) ambulatory, (d) free of chronic medical conditions including seizure disorders, (e) not receiving psychotropic medication, and (f) parent or guardian provided informed consent for participation in this study. The IDD only subgroup had 25 participants, 16 males and nine females (mean (SD) age = 48.68 (9.58) years). The IDD + SMD subgroup included eight participants, six males and two females (mean (SD) age = 46.1 (9.71) years), and it was defined as those individuals from the IDD group, who: (a) received a minimum score of 2 on the Stereotype (STY) Checklist (used to identify presence of stereotyped movements during a standard motor exam, Bodfish et al., 1995); and (b) received a minimum score of 8 on the behavior problems inventory (BPI) Stereotype Subscale (used to determine minimum cut-off for frequency/severity of stereotyped movements, Rojahn, Matson, Lott, Esbensen, & Smalls, 2001). Diagnosis of IDD in both the IDD and the IDD + SMD groups was determined based on chart review (recent cognitive and adaptive behavior assessments) and using DSM-IV criteria for mental retardation.

[1] In an inverse power law the distribution of the data set is inversely proportional to the frequency of the form $1/f^i$ where f is a frequency and i is a positive exponent called the spectral coefficient. When the data are presented in a log–log format the data appear as a straight line over a certain range. See West (2006).