# Reliability and diagnostic efficiency of the Diagnostic Inventory for Disharmony (DID) in youths with Pervasive Developmental Disorder and Multiple Complex Developmental Disorder

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## ABSTRACT

The Pervasive Developmental Disorder-Not Otherwise Specified (PDD-NOS) category is a psychopathological entity few have described and is poorly, and mainly negatively, defined by autism exclusion. In order to limit PDD-NOS heterogeneity, alternative clinical constructs have been developed. This study explored the reliability and the diagnostic efficiency of the Diagnostic Inventory for Disharmony (DID); its concurrent validity with the Vineland Adaptive Behavior Scales (VBAS); and its concordance with Multiplex Complex Developmental Disorder (MCDD) and PDD-NOS. Mean DID interrater reliabilities and internal consistency were good (.58 and .75, respectively). DID diagnostic efficiency yielded by the ROC analysis was very promising using a cut-off score of 12/36 (AUC = .97, sensitivity = .93, specificity = .91). Spearman correlations between the DID total score and the three subscales in the VABS socialization domain were significantly negative, thus confirming an association between Disharmony and impairments in socialization. However, no correlation was found between Disharmony and community daily living skills, likely reflecting a better autonomy in daily activities. Disharmony phenomenology overlapped with Autism Spectrum Disorders (DID and PDD concordance: kappa: .41; $p < .01$). Nevertheless, the Disharmony construct seemed to differ from PDD-NOS and be closer to MCDD. We conclude that Disharmony and MCDD constitute complementary views on the same group of severely impaired children.

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## 1. Introduction

Both clinicians and researchers are facing a huge increase in the prevalence of Pervasive Developmental Disorders (PDD) resulting in part from the expansion of the diagnostic criteria (Matson & Kozlowski, 2011). According to the DSM-IV-TR, the PDD group includes a wide variety of diverse conditions pertaining to the autism spectrum: Autistic Disorder (AD), Asperger

disorder (AS), Rett syndrome, disintegrative disorder of childhood, and PDD Not Otherwise Specified (PDD-NOS)(American Psychiatric Association, 2000; Rapin, 2002). Prevalence estimates are 13 per 10 000 for AD, 2.6 per 10 000 for AS and 20.8 per 10 000 for PDD-NOS (Fombonne, 2005). In France, the concept of what constitutes PDD has been expanded to include Disharmony, a notion introduced by the French Child Psychiatry school (Mises et al., 2002) and until recently conceived of as a distinct entity. Expansion in diagnostic criteria and recognition of PDD as a lifelong disorder has led to even higher figures as shown by recent reviews on prevalence studies (Matson & Kozlowski, 2011). Despite PDD-NOS tending to be more commonly diagnosed than AD, there are fewer studies on the predictive validity and stability of this diagnostic category (Mahoney et al., 1998; Matson & Boisjoli, 2007; Witwer & Lecavalier, 2008), and the developmental trajectories of PDD-NOS are marked with heterogeneity (Chawarska, Klin, Paul, & Volkmar, 2007; Charman et al., 2005; Cox et al., 1999). A recent meta-analysis (Rondeau et al., 2011) showed that PDD-NOS diagnostic stability was lower than AD (pooled Relative Risk was 1.95, $p < .001$). When diagnosed before 36 months PDD-NOS bore a 3-year stability rate of 35%. Examining the developmental trajectories showed that PDD-NOS corresponded to a group of heterogeneous pathological conditions including prodromic forms of later AD, remitted or less severe forms of AD (Leroy et al., 2010; Stone et al., 1999), and developmental delays in interaction and communication (Buitelaar & van der Gaag, 1998).

In the past, other proposals have been formulated to catch subgroups of children with less severe pervasive developmental trajectories than autism. In the US, the Yale Child Study Center team described the Multiplex Developmental Disorder (MDD) focusing on the clinical phenomenology of patients who presented a later age of onset and a better outcome (Cohen, Paul, & Volkmar, 1987). Symptoms were distributed into three separate domains: emotional, social and cognitive. In the Netherlands, Buitelaar and van der Gaag (1998) individualised a subtype of PDD-NOS, Multiple Complex Developmental Disorder (MCDD) that paralleled the Yale team construct and for which they validated a criteria scoring algorithm. In Sweden, Christopher Gillberg described a syndrome named Deficit in Attention, Motor control and Perception (DAMP) (Gillberg, 2003). The syndrome encompassed Attention-Deficit-Disorder-Not-Otherwise-Specified and motor coordination disorder with normal or borderline intelligence. The condition was compatible with comorbidities such as autism spectrum disorder, major depression or oppositional defiant disorder. Finally, in France, the concept of Disharmony was used to describe a developmental disorder characterized by the association of focal cognitive delay, emotional disturbances and learning difficulties. The phenomenology has been regrouped under two diagnostic labels, psychotic Disharmony (Mises et al., 2002) and cognitive Disharmony (Gibello, 2010) according to the hypotheses underlying the core psychopathological processes: impairments in self construction and discrepancies in cognitive development respectively.

There remains a need to better define PDD, especially PDD-NOS and improve both positive and differential diagnoses (Matson & Boisjoli, 2007; Rondeau et al., 2011). In order to achieve better diagnostic procedures research has to be done in developing proper assessment instruments (Matson, Nebel-Schwalm, & Matson, 2007). Among the previously mentioned proposals, only MCDD received proper empirical validation. In keeping with the proposal from the Yale Child Study Centre, the Netherlands Group evinced a set of symptom patterns in children with PDD-NOS which they labelled as a specific entity, "Multiple Complex Developmental Disorder" (MCDD). This entity was defined by (a) important social and communication difficulties, (b) emotional dysfunction, (c) cognitive disturbances, (d) a risk for schizophrenia in adolescence, and (e) a higher biological reactivity to stress. The group also validated the MCDD criteria in a sample of outpatients. Concurrent validity was assessed towards the Child Behavior Check List (Buitelaar and van der Gaag, 1998; De Bruin, De Nijs, Verheij, Hartman, & Ferdinand, 2007; Lahuis et al., 2008; Van der Gaag et al., 1995).

Tordjman et al. (1997) set up an Expert Work Group to study the French and US concepts. They identified a set of operationalised diagnostic criteria for each of the two constructs, MDD and Disharmony that the present study aimed to compare. They highlighted the similarities in the potential cases despite differences in the proposed criteria. The French Disharmony puts clinical and psychodynamic elements on the same level, while the US MDD concept proposed a more descriptive model and appears potentially more relevant for research (Tordjman et al., 1997).

The first aim of the present study was to explore the reliability and the diagnostic efficiency of the Diagnostic Inventory for Disharmony (DID) and its concurrent validity with the Vineland Adaptive Behavior Scales. In addition we aimed at studying the concordance between Disharmony, MCDD and PDD-NOS.

## 2. Methods

### 2.1. Sample

The study was conducted in accordance with the Hospital's Research Ethics Board regulations on two-day care units in a University teaching Hospital Child and Adolescent Psychiatry Department. The study concerned all consecutive admission for children at risk for Disharmony over a period of 18 months in 2009–2010. Inclusion required at least the presence of two diagnostic criteria for Disharmony. The resulting sample was composed of 41 six- to thirteen-year-old children (mean age: 10.02; SD: 2.10) with 39 boys and 2 girls. Participants' performance IQ ranged from 47 to 116 (mean 86.23; SD 16.62). Two children presented with Mental Retardation with performance quotient <70. All children were severely impaired. Total score on the Children's Global Assessment Scale (CGAS) ranged from 25 to 50 (mean score < 40). Following diagnostic assessment thirty children with Disharmony and eleven without Disharmony were included. Main characteristics of the sample are summarized in Table 1.