



## The obsessive-compulsive trait of Incompleteness in parents of children with autism spectrum disorders

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### ABSTRACT

The obsessive-compulsive behaviors central to Obsessive-Compulsive Disorder (OCD) are not uncommon in Autism Spectrum Disorders (ASD), however the association between these disorders is not yet clear. One construct which may be useful in delineating their overlapping characteristics is “Incompleteness” or a sense of things feeling “not just right”. Incompleteness has been related to a constellation of symptoms in OCD, but its association with ASD has not yet been examined. In this study parents with two or more children with ASD (P-MC) ( $n=115$ ) were compared to an independent sample of parents having a single child with an ASD (P-SC), matched by age and gender, on level of Incompleteness. Results indicate that P-MC parents scored significantly higher in Incompleteness than P-SC parents. Incompleteness scores were also associated with a profile of behaviors in their children with ASD ( $n=357$ ) characterized by high scores on the empirically derived repetitive sensory motor actions and resistance to change domains of the Autism Diagnostic Interview-Revised (Cuccaro et al., 2003). We discuss the implications of Incompleteness found in parents of children with an ASD, as well as its utility as a possible endophenotype for both ASD and OCD.

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### 1. Introduction

Obsessive-compulsive like characteristics have long been noted in individuals with Autism Spectrum Disorders (ASD) beginning with Kanner's (1943) observation that the behavior of children with classical autism is “... governed by an anxiously obsessive desire for the maintenance of sameness that nobody but the child himself may disrupt on rare occasions. Change of routine, of furniture arrangement, of a pattern, of the order in which everyday acts are carried out, can drive him to despair.” (p. 245). In the more than half-century since this seminal work, surprisingly little research has examined the relationship between the repetitive and ritualistic behaviors seen in ASD and the obsessive-compulsive behaviors central to Obsessive-Compulsive Disorder (OCD).

Restrictive and repetitive behaviors can take many forms, including stereotyped movements, tics, compulsions, insistence on sameness, circumscribed interests, and rigid adherence to routines and rituals (American Psychiatric Association (APA), 2000). Although once considered a single domain, there is growing evidence that the restrictive and repetitive behaviors seen in ASD are heterogeneous

in nature. Factor analytic studies using the restricted, repetitive behaviors and interest domain (RRBI) of the Autism Diagnostic Interview-Revised (ADI-R; Lord, Rutter, & Le Couteur, 1994) have demonstrated that such characteristics can be classified as either: (1) repetitive sensory and motor behaviors or (2) resistance to change (Cuccaro et al., 2003; Richler, Bishop, Kleinke, & Lord, 2007; Shao et al., 2003). Repetitive sensory motor actions (RS)—such as rocking and repetitive use of objects—constitute a behavioral domain, regarded as a lower-order category performed solely for the purpose of self-stimulation. Resistance to change (RC) represents a higher-order category with both cognitive and behavioral features associated with the need for sameness in one's routine and environment (Turner, 1999). Studies of ASD have found evidence of familiarity for RS and RC, suggesting that both behaviors may be genetically driven (see Lam, Bodfish, & Piven, 2008; Liu, et al., 2011a). The trajectory of repetitive behaviors in ASD is not entirely clear and there are a handful of longitudinal and cross-sectional studies which have addressed this issue, focusing on both lower-order and higher-order repetitive behaviors. From this body of research, one consistent finding is that RS behaviors are less frequent during adolescence and adulthood than in childhood, while higher-order RC behaviors tend to be less common during childhood and for some individuals with ASD become more prominent with age, most notably for those with higher intelligence levels (Militeri, Bravaccio, Falco, Fico, & Palermo, 2002; Esbensen, Seltzer, Lam, & Bodfish, 2009; McGovern & Sigman, 2005; Richler, Huerta, Bishop,

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& Lord, 2010). However, what is not understood is whether RS behaviors become transformed into higher order RC or whether RS and RC behaviors developed independent of each other.

Restrictive and repetitive behaviors are not unique to ASD and have been associated with a number of developmental, neurological, and genetic disorders. In OCD they appear in the form of compulsions – repetitive ritualized behaviors that the person feels driven to perform either according to rigid self-imposed rules, or in response to obsessions. Obsessions are the cognitive component of OCD – unwanted, recurrent, uncontrollable and anxiety-evoking thoughts that include the fear of contamination, fear of causing harm to another, fear of making a mistake, and the need for symmetry or exactness. Common compulsions include cleaning, checking, arranging, hoarding, counting and repeating (APA, 2000).

Although their etiology remains unknown, ASD and OCD are both considered neurological disorders and family studies suggest that the two may have a common genetic basis. For example, in a study examining obsessive-compulsive behaviors in parents having one or more child with an ASD, Hollander, King, Delaney, Smith, and Silverman (2003) found that autistic probands scoring high on the RRBI domain of the ADI-R ( $\geq 9$ ) were more likely to have one or both parents with obsessive-compulsive traits or OCD, as ascertained by the Yale-Brown Obsessive Compulsive Scale (Y-BOCS; Goodman et al., 1989). Abramson et al. (2005) also examined obsessive-compulsive behaviors in parents of ASD families. Consistent with Hollander et al.'s (2003) findings, these researchers reported that 34.4% of the 155 autism families had first or second-degree relatives with problematic obsessive-compulsive traits or OCD. Using data from a subset of their sample ( $n=69$ ), Abramson et al. compared parents with clinically significant Y-BOCS scores to those with nonclinical Y-BOCS scores based on proband scores from the empirically derived RRBI domains of repetitive sensory motor actions (RS) and resistance to change (RC) on the ADI-R (Cuccaro et al., 2003). It was found that parent Y-BOCS scores were moderately correlated with proband scores on the RC domain, but not with proband scores on the RS domain. The identification of autistic and/or obsessive-compulsive traits in family members may assist with the formation of more homogeneous groups, thus increasing the likelihood of identifying genes common to both ASD and OCD.

### 1.1. Incompleteness

One construct which may be useful in delineating the overlapping characteristics seen in OCD and ASD is “Incompleteness”. Incompleteness was first introduced into the OCD literature over a century ago by Pierre Janet, who described this experience as ‘les sentiments d’incomplétude’—literally, feelings of incompleteness – an inner sense of imperfection connected with the perception that actions or intentions have been incompletely achieved (see Pitman, 1987, for a translated précis). This sense of feeling that things are “not just right” can be experienced through all sensory modalities including visual (e.g., appearance of objects in one’s environment), auditory (e.g., preference for sameness in ambient noise), tactile (touching or tapping), and proprioceptive (e.g., having to “even up” actions). Incompleteness can also be applied to more complex cognitive experiences (e.g. expressing one’s thoughts unambiguously, in the best words) (Summerfeldt, 2007). A number of studies have demonstrated the association between Incompleteness and a particular constellation of OCD symptoms comprising symmetry, counting, repeating, and ordering (Coles, Heimberg, Frost, & Steketee, 2005; Ecker & Gonner, 2008; Pietrefesa & Coles, 2008; Summerfeldt, 2007; Tolin, Brady, & Hannan, 2008). A common theme cutting across these symptoms is the person’s sense of being driven to arrange/order

objects in their environment to quell feelings or sensations of things being “not just right”, suggesting that Incompleteness may be the chief underlying motivator (Summerfeldt, 2007).

Evidence suggests that Incompleteness may be a heritable trait. For example, Moore, Smith, Shevlin, and O’Neill (2010) recently conducted a study with a sample of 517 adolescent twins (48% males; 52% females), ranging in age from 11 to 16 years. Adolescents completed the Short Leyton Obsessional Inventory – Children’s Version (Short LOI-CV; Bamber, Tamplin, Park, Kyte, & Goodyer, 2002) assessing a multidimensional model of obsessive-compulsive behaviors which encompasses three domains: obsessions/incompleteness, numbers/luck, and cleanliness. Twin analyses revealed a heritability rate of 60% in adolescent boys for the domain of obsessions/incompleteness. However, it should be noted that the Short LOI-CV was developed as a screener for OCD in adolescents and contains only 11 items. Of the 11 items, only four refer to the obsessions/incompleteness domain. While this research offers additional insight on the trait of Incompleteness, further research is needed to confirm the heritability of this construct.

A number of OCD symptoms related to Incompleteness have been noted in individuals with ASD. In a study comparing 50 adults with autistic disorder to an age and gender-matched group of adults with OCD using the Y-BOCS symptom checklist, McDougle et al. (1995) found that the adults with autistic disorder endorsed more items related to repetitive ordering, hoarding, touching, tapping, or rubbing, and self-damaging behaviors. Of note is that over half ( $n=35$ ) of the autistic participants in McDougle et al.’s sample also met DSM-IV criteria for mental retardation, including 15 participants who were classified as nonverbal. In a more recent study Russell, Mataix-Cols, Anson, and Murphy (2005) compared 40 adults with high-functioning autism or Asperger syndrome having average intelligence to 45 individuals meeting DSM-IV criteria for OCD. Based on Y-BOCS scores, it was found that both groups were comparable in the overall frequency of obsessions and compulsions, but the OCD group reported significantly more somatic obsessions, as well as the compulsions of repeating and checking. Of the ASD sample, approximately 25% met criteria for comorbid OCD. Taken together these findings suggest that although IQ and/or verbal ability may play a major role in obsessive-compulsive symptom expression in individuals with an ASD, the types of symptoms most often observed include the ones most associated with Incompleteness in OCD samples.

The Incompleteness construct may be of value in efforts to understand the overlapping features of OCD and ASD. Although Incompleteness has been increasingly recognized in the OCD literature, to date no research has examined its association with ASD. In light of this, the present study had two goals. The first was to examine whether level of Incompleteness in parents with more than one child with an ASD would differ from parents with a single child with an ASD.

The idea that it is meaningful to compare multiplex (MF; having two or more children with an ASD) and singleton (SF; having only one child with an ASD) families arises from the literature on familiarity of traits and symptoms. To determine possible genetic linkage in ASD a number of studies have focused on multiplex families based on the assumption that these families are genetically more robust than singleton families. In support of this, Losh, Childress, Lam, and Piven (2008) examined sub clinical autistic traits as part of the broad autism phenotype in parents from 25 MF in comparison to parents from 40 SF. These researchers found that for the majority of parents from MF, both parents displayed autistic traits; however, parents from SF were more likely to display these traits by either one, both, or neither parents. Virkud, Todd, Abbacchi, Zhang, and Constantino (2008)

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