

A Twin Study of Attention-Deficit/Hyperactivity Disorder Dimensions Rated by the Strengths and Weaknesses of ADHD-Symptoms and Normal-Behavior (SWAN) Scale

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Background: When symptom rating scales are used in the general population, there is severe skewness, with many individuals having no symptoms. While this has major implications for genetic designs that require extremely discordant and concordant (EDAC) siblings, little is known of the genetics of scales which seek to differentiate within the “no ADHD symptom” group.

Methods: Parents of Australian twins completed two attention-deficit/hyperactivity disorder (ADHD) questionnaires, the Australian Twin Behaviour Rating Scale (ATBRS), based on conventional DSM-IV symptom scores, and the Strengths and Weaknesses of ADHD-Symptoms and Normal-Behavior (SWAN) scale, which includes above-average performance on attention and activity. The two scales were compared in two age groups of same-sex twins, 528 pairs aged 6 to 9 and 488 pairs aged 12 to 20.

Results: Parents reported higher levels of activity and attention in their twins when reporting using the SWAN scale than when using the ATBRS, and while the monozygotic (MZ) correlations were similar on both scales, the dizygotic (DZ) correlations were consistently higher on the SWAN. On DSM-IV based scales, parents exaggerated differences within those sibling pairs in the “with few ADHD symptoms” category.

Conclusions: The SWAN may provide a more realistic description of the ADHD phenotype for the selection of twin and sibling pairs for genetic analysis.

Key Words: ADHD, EDAC, genetic analysis, SWAN, symptom rating scales, twins

Attention-deficit/hyperactivity disorder (ADHD) is one of the most common psychiatric disorders in childhood. Graetz et al (2001) surveyed 3597 Australian children and adolescents (6 to 17 years of age) and found that approximately 7.5% of the group sampled had ADHD. Attention-deficit/hyperactivity disorder is defined by elevated levels of inattention and/or hyperactive and impulsive behavior, and the DSM-IV (American Psychiatric Association 1994) recognizes three distinct subtypes within ADHD, the inattentive, hyperactive-impulsive, and combined forms. Graetz et al (2001) found that the inattentive subtype of ADHD was more commonly identified than the hyperactive-impulsive or combined subtypes.

Many twin studies (reviewed in Bennett et al, *in press*) indicate the very substantial genetic component to ADHD. However, there has been some questioning of whether the DSM-IV subtypes are the most realistic approach to ADHD for the purposes of genetic analyses, and one alternative proposed is that of empirically based latent classes (Todd 2000). Such latent classes have proved robust at both the phenotypic (Rasmussen et al 2002) and genetic (Rasmussen et al 2004) levels across Missouri and Australian twin studies where the families were

identified and assessed in very different ways by telephone interview and rating scale, respectively.

But there is a much broader issue for genetic studies than just the DSM-IV classification of symptoms. Whether assessed by rating scales or interview, most scales measuring ADHD are based on the 18-item diagnostic criteria of the DSM-IV (Murphy and Adler 2004). Such scales assess ADHD using a three-, four-, or five-point scale to rate symptom severity from, for example, 0 = never or not at all to 3 = symptom occurs very often. In a general population, most people will have a low score on many of the items, indicating that they have low or no attention problems. If, as described by Levy et al (1997), ADHD comprises a continuous dimension of behavior, then it is likely that some individuals will actually be performing better than average on attention behaviors. Therefore, the use of such rating systems can result in limited (and skewed) data about individuals who are considered to be unaffected by ADHD, as they are all assigned the same score of 0.

This has major implications for molecular genetic studies of ADHD and other behaviors. Increasingly, genetic studies of behavior are selecting sibling pairs using the Extreme Discordant and Concordant (EDAC) design to maximize power to detect linkage (Kirk et al 2000). While concordant affected pairs are easily identified using conventional DSM-IV scales, the ability to define concordant unaffected pairs and extremely discordant pairs will be limited. The same problem goes for association studies. For example, in their study of selection strategies for quantitative trait locus (QTL) mapping in pooled DNA samples, Jawaid et al (2002) demonstrated the most effective comparison was of the top 27% with the bottom 27% of the sample. There are many examples in ADHD (including the present study; Table 1) where much more than 27% of a population sample will have a score of 0.

Swanson et al (2005) developed the Strengths and Weaknesses of ADHD-Symptoms and Normal-Behavior (SWAN) scale

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Table 1. Monozygotic and Dizygotic Twins Mean Scores in Two Age Groups on the Australian Twin Behaviour Rating Scale and Strengths and Weaknesses of ADHD-Symptoms and Normal-Behavior Rating Scale

	ATBRS (Range 0 to 3)		SWAN (Range -3 to +3)		SWAN Recoded (Range 0 to 3)	
	Inattention	Hyperactivity-Impulsivity	Inattention	Hyperactivity-Impulsivity	Inattention	Hyperactivity-Impulsivity
Younger Age Group						
MZ	.63 (.54)	.75 (.46)	-.50 (.89)	-.53 (.86)	.12 (.40)	.08 (.35)
DZ	.66 (.58)	.76 (.49)	-.50 (.93)	-.52 (.84)	.14 (.44)	.09 (.34)
Older Age Group						
MZ	.65 (.62)	.46 (.54)	-.71 (1.17)	-.86 (1.17)	.16 (.47)	.11 (.38)
DZ	.65 (.61)	.44 (.51)	-.64 (1.15)	-.75 (1.12)	.18 (.53)	.10 (.40)

ATBRS, Australian Twin Behaviour Rating Scale; SWAN, Strengths and Weaknesses of ADHD-Symptoms and Normal-Behavior; MZ, monozygotic; DZ, dizygotic.

to help overcome the limitations identified in the earlier rating scales of ADHD. While also based on the 18 ADHD items in the DSM-IV (American Psychiatric Association 1994), the SWAN was designed to measure a wider range of population variation by extending the four-point rating scale to seven points, using $-3 =$ far above average to $+3 =$ far below average in severity. This extended rating system allows reporting of areas where individuals perform well above average, as well as areas where they are struggling. Extending the range of responses results in additional data on individuals who are unaffected by ADHD and does not truncate the data. Therefore, the full range of behavior in the general population is measured.

This study aims to investigate the genetic utility of the SWAN by comparing twin data from the SWAN to that of a traditional four-point scale for the assessment of DSM-IV ADHD, the Australian Twin Behaviour Rating Scale (ATBRS).

Methods and Materials

Two different-aged samples of Australian twins were selected to investigate differences between the two scales of ADHD, given indications (Hay et al 2004) that there may be differences in the genetic determinants of ADHD during development. Both samples came from the Australian Twin ADHD Project (ATAP) described in Levy and Hay (2001). This research was approved both by the Curtin University Human Research Ethics Committee and by the Australian Twin Registry (ATR).

Younger Age Group

In 2001, a cohort of 1042 families were approached through the Australian Twin Registry. Of these, 707 families completed the ATBRS and the SWAN rating scale of ADHD. The data from 528 consenting families with same-sex twins are analyzed in the current article. Opposite-sex pairs were excluded to avoid complications of modeling around the gender difference in symptom number (Levy et al 1996; Rhee et al 2001). Referred to as the younger age group, these families had twins aged between 6 and 9 years (mean age = 7.6 years, SD = .91). There were approximately equal numbers of male and female twin pairs (262 and 266 pairs, respectively) in the sample with 52% being monozygotic (MZ) twin pairs and 48% being dizygotic (DZ) twin pairs.

Older Age Group

In 1999, data were collected from 887 twin pairs aged 12 to 20 years (mean age = 15.2 years, SD = 2.54), the older age group. These were the initial ATAP cohort, studied from 1991 when all 4- to 12-year-old twin pairs in the Australian Twin Registry were screened (Levy et al 1996). The data from 488 families with

same-sex twins and complete data are analyzed in the current study. Forty-nine percent of the sample was male. Approximately 60% of this group were MZ twin pairs and 40% were DZ twin pairs. The slightly lower rate of DZ twins in this older sample is consistent with MacFarlane and Blondel (2005), who reported there has been a marked increase in DZ twin rates since the 1980s and only a small increase in the rates of MZ twins.

Assessment of Zygosity

Zygosity was established using discriminant function analysis based on a questionnaire by Cohen et al (1975). This scale had six questions on similarity of features and six questions on frequency of confusion by the mother. There was individual follow-up where ambiguity remained. A more detailed description of this scale can be found in Levy et al (1997) and Hay et al (2001).

Assessment of ADHD

The data on ADHD were collected by mailed questionnaires sent to parents for completion. Parents were asked to provide information on the behavior of their twins and similar-aged siblings, though the latter are not analyzed here. The two ADHD scales were the ATBRS and the SWAN.

The Australian Twin Behaviour Rating Scale is based on the DSM-IV criteria for the diagnosis of ADHD. The relationship of this measure to diagnosis of ADHD by formal psychiatric interview has been described in Levy et al (1997, 2005). More discussion of the scale and its genetics can be found in Levy et al (2001). It contains 18 items measuring ADHD behaviors, which ask a parent to rate their child's behavior on a four-point scale from "never" to "very much or very often." For example, each parent was asked to answer the following about their children: "Has trouble following through on instructions and doesn't finish schoolwork, chores or duties?" Parents indicated their response as 0 = not at all, 1 = just a little or sometimes, 2 = pretty much or often, or 3 = very much or very often. The individual's total score on the nine inattention and nine hyperactivity-impulsivity items were then averaged to range from 0 to 3, with a higher score indicating a higher level of ADHD symptoms. Previous comparison with interview data indicate this questionnaire provides a more conservative symptom score, with parents identifying more ADHD problems at interview (Levy et al 1997).

The Strengths and Weaknesses of ADHD-Symptoms and Normal-Behavior scale contains 18 reworded items to measure ADHD. Based on the previous Swanson, Nolan, and Pelham (SNAP) rating scale (Swanson 1992), items were reworded from the categorical approach of ATBRS (This child: "Has trouble following through on instructions and doesn't finish schoolwork,

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