Across the continuum of attention skills: a twin study of the SWAN ADHD rating scale

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Introduction: Most behavior checklists for attention problems or attention deficit/hyperactivity disorder (ADHD) such as the Child Behavior Checklist (CBCL) have a narrow range of scores, focusing on the extent to which problems are present. It has been proposed that measuring attention on a continuum, from positive attention skills to attention problems, will add value to our understanding of ADHD and related problems. The Strengths and Weaknesses of ADHD symptoms and Normal behavior scale (SWAN) is such a scale. Items of the SWAN are scored on a seven-point scale, with in the middle ‘average behavior’ and on the extremes ‘far below average’ and ‘far above average’. Method: The SWAN and the CBCL were completed by mothers of respectively 560 and 469 12-year-old twin pairs. The SWAN consists of nine DSM-IV items for Attention Deficit (AD) and nine DSM-IV items for Hyperactivity/Impulsivity (HI). The CBCL Attention Problem (AP) scale consists of 11 items, which are rated on a three-point scale. Results: Children who had a score of zero on the CBCL AP scale can be further differentiated using the SWAN, with variation seen between the average behavior and far above average range. In addition, SWAN scores were normally distributed, rather than kurtotic or skewed as is often seen with other behavioral checklists. The CBCL AP scale and the SWAN HI and AD scale were strongly influenced by genetic factors (73%, 90% and 82%, respectively). However, there were striking differences in genetic architecture: variation in CBCL AP scores is in large part explained by non-additive genetic influences. Variation in SWAN scores is explained by additive genetic influences only. Conclusion: Ratings on the SWAN cover the continuum from positive attention skills to attention and hyperactivity problems that define ADHD. Instruments such as the SWAN offer clinicians and researchers the opportunity to examine variation in both strengths and weaknesses in attention skills. Keywords: Attention deficit, hyperactivity, heritability, twin study.

Attention deficit hyperactivity disorder (ADHD) is characterized by the presence of symptoms of inattention, hyperactivity and impulsivity. It is the most common neuro-developmental disorder of childhood, with prevalences ranging from 4 to 12% in the general population (Brown et al., 2001; Faraone, Sergeant, Gillberg, & Biederman, 2003). The diagnosis is typically made by a trained clinician using information that is collected in several ways, varying from behavior checklists, filled in by for example parents or teachers, to interviews and observations by trained psychiatrists. The overlap in diagnoses among the different measures of attention problems, such as the Child Behavior Checklist’s (CBCL; Achenbach, 1991) Attention Problem Syndrome (AP) and DSM-IV interviewed-based ADHD, is moderate to high (Hudziak, Copeland, Stanger, & Wadsworth, 2004; Kasius, Ferdinand, van den Berg, & Verhulst, 1997; Derks, Hudziak, Dolan, Ferdinand, & Boomsma, 2006b).

A feature of most behavior checklists is the strict and narrow range of ratings on the problem items. Possible scores on the AP scale of the widely used CBCL, for example, are 0, 1 or 2, indicating that a child shows certain behavior (0) not at all, (1) sometimes, or (2) often. Similarly, the Rutter scale has a scoring range of 0 to 3 (Rutter, Tizard, & Whitmore, 1970), and the DuPaul ADHD rating scale a range of 0 to 2 (DuPaul, 1981). When data are collected with these instruments in the general population the distribution of scores is often skewed. This is due to the fact that only a small percentage of subjects have serious attention problems while the majority of the children score in the very low range or have zero problem symptoms. As a result, there is no possibility of studying variance at the other end of the distribution, e.g., those children who have above average or excellent skills in the attentional, hyperactive/impulsive domains.

The skewness seen in regular measures of ADHD may be avoided through the use of a relatively new ADHD scale named the Strengths and Weaknesses of ADHD symptoms and Normal behavior scale (SWAN; Swanson et al., 2006). The SWAN is based on the 18 ADHD items listed in the DSM-IV. What sets it apart from other checklists is that each item is scored on a seven-point scale with ‘average behavior’ scored in the middle and ‘far below average’ and ‘far...
above average’ at the extremes. Because the SWAN measures both the strength and weakness characteristics of ADHD it is expected that it yields a normal distribution of scores in the general population (Swanson et al., 2006). This broader range of scores might provide additional information about the nature of attention problems.

Genetic studies showed that variation in ADHD is strongly influenced by genetic factors, with heritability estimates ranging from 70 to 90% (Rietveld, Hudziak, Bartels, van Beijsterveldt, & Boomsma, 2004; Hudziak, Rudiger, Neale, Heath, & Todd, 2000; Faraone & Doyle, 2002; Nadder, Silberg, Eaves, Maes, & Meyer, 1998) for both attention deficit and hyperactivity/impulsivity. The prevalence of ADHD tends to be higher in boys than in girls, but there is no evidence for substantial sex differences in the relative importance of genetic or environmental influences (Derks, Hudziak, & Boomsma, 2006a).

Heritability of ADHD appears to be the same for extreme cases of ADHD as for individual differences in the normal population, suggesting that attention problems are normally distributed with ADHD being on the tail of the distribution (Levy, Hay, McStephen, Wood, & Waldman, 1997). Most genetic studies on ADHD found no significant influences of common environment (i.e., the environment that is shared by members of a family) but suggested, based on a pattern of DZ twin correlations being lower than half the MZ twin correlations, the influence of contrast effects or genetic non-additivity (i.e., dominance or epistasis effects). Contrast effects may arise because of competitive social interaction among siblings, or because parents compare the behavior of their twins and stress differences between them (Eaves et al., 1997; Simonoff et al., 1998; Nadder et al., 1998; Van den Oord, Verhulst, & Boomsma, 1996; van Beijsterveldt, Verhulst, Molenaar, & Boomsma, 2004; Eaves, 1976). Low DZ correlations can also indicate the influence of genetic dominance (i.e., non-additive genetic effects) as is reported by, for example, Rietveld, Hudziak, Bartels, van Beijsterveldt, and Boomsma (2003a), Martin, Scourfield, and McGuffin (2002), Derks, Hudziak, van Beijsterveldt, Dolan, and Boomsma (2004) and Thapar, Harrington, Ross, and McGuffin (2000). Interestingly, teacher ratings do not indicate the presence of dominance or contrast effects, suggesting that only in parental data do these phenomena play a role. In parental ratings, however, the results are inconclusive and seem to vary across instruments, age (of the twins) and methods (Derks et al., 2006a). For example, Rietveld et al. (2004) reported in a longitudinal study contrast effects at age 3 and effects of dominance at ages 7, 10 and 12. Thapar et al. (2000) found significant contrast effects on the Rutter scale (Rutter et al., 1970) and, in addition, significant dominance effects on the DuPaul ADHD rating scale (DuPaul, 1981).

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Methods

Subjects and procedure

The subjects are Dutch twins whose parents voluntarily registered with the Netherlands Twin Registry (NTR) when the twins were born (Boomsma, 1998; Boomsma et al., 2002b). All twin pairs are participating in a longitudinal study in which surveys are sent to their parents and teachers (Bartels et al., 2007). Parents are asked to fill in the CBCL for their twins at ages 3, 7, 10 and 12.

Of the total NTR population, data on the SWAN were collected for two samples of approximately 12-year-old...
children. Twin mothers were asked to complete the SWAN (N = 681 pairs). The first sample consisted of 177 Dutch twin pairs who were born between 1990 and 1992 and who participated in a longitudinal study on Cognition, Attention and Attention Problems (Polderman et al., 2006). Data on the SWAN were collected when the twins were 12 years old (mean age = 12.42, SD = .16). The sample is unselected with respect to attention problems. Invitation to participate in this study was based on age and a sample equally distributed across sex and zygosity. Zygosity was determined on the basis of DNA polymorphisms. None of the children suffered from severe physical or mental handicaps. Parents signed an informed consent form.

The second sample consisted of 504 Dutch twin pairs, aged between 10 and 13 years old (mean age = 11.71; SD = .77) who were born between 1989 and 1994 and participated in a study of Attention Problems (Derks et al., 2006b). For this sample subjects were selected from an initial sample of 6191 twin pairs on the basis of their maternal CBCL ratings (T-scores; Mean = 50, SD = 10) at ages 7, 10, and 12 years. Subjects were excluded if maternal ratings were available only at one time-point, or if they suffered from a severe handicap, which disrupts daily functioning. Twin pairs were selected if at least one of the twins scored high on AP (affected pairs) or if both twins scored low on AP (control pairs). A high T-score was defined as a T-score above 60 at all available time-points (age 7, 10, and 12 years) and a T-score above 65 at least once. A low score was defined as a T-score below 55 at all available time-points. The control pairs were matched with the affected pairs on the basis of sex, cohort, maternal age, and socio-economic status. T-scores were computed in boys and girls separately. In other words, girls were selected if they scored low or high compared to other girls, and boys were selected if they scored low or high compared to other boys. This procedure resulted in the selection of an equal number of boys and girls. Zygosity for 403 twin pairs was determined on the basis of DNA polymorphisms. The remaining twin pairs, zygosity was based on a 10-item questionnaire. Zygosity determination using this questionnaire is almost 95% accurate (Rietveld et al., 2000). Parents signed an informed consent form.

Mothers of children of the first sample completed the SWAN when their children performed a neuropsychological test battery at the Vrije Universiteit. Mothers of children of the second sample received and returned the SWAN by mail. Of the first sample the data for 9 twin pairs were missing, and of the second sample the data for 99 twin pairs were missing. Twelve twin pairs participated in both studies. Of these 12 twin pairs, the questionnaires of one or both studies were selected at random. The combination of both samples then resulted in a sample of 224 MZ twin pairs and 337 DZ twin pairs (N = 561 pairs).

Maternal CBCL data (age 12) were collected as part of the parental surveys by the NTR every two years (total N = 6191 twin pairs for cohorts 1989–94). For the current sample CBCL data were available for 469 twin pairs.

**Instruments**

The CBCL (Achenbach, 1991) is a behavioral checklist for parents to report the frequency and intensity of behavioral and emotional problems of their children. Parents are instructed to rate the child’s behavior over the last six months with 0 if the behavior is not true, 1 if the behavior is sometimes or somewhat true, and 2 if the behavior is very or often true. The Attention Problem scale of the CBCL consists of 11 items, so the maximum score on this scale is 22. The more attention problems a child has, the higher his or her score on the Attention Problem scale.

The SWAN (Swanson et al., 2006) employs 18 items on a seven-point scale ranging from ‘far below average’ (1) to ‘far above average’ (7) to allow for ratings of relative strengths (above average) as well as weaknesses (below average). The first nine items correspond to the Attention Deficit scale and the last nine items to the Hyperactivity/Impulsivity scale. The maximum score on a SWAN scale is 63. The more attention problems a child has, the lower his or her score on the SWAN rating scales.

**Analyses**

Of the total sample (N = 561 pairs), one part was unselected with respect to attention problems and one part was selected based on longitudinal scores on the CBCL-Attention Problem scale. The selection procedure described above resulted in an under-representation of twins with moderate CBCL scores. Data-weighting was used to take account of the fact that the sample was not a random sample (Heath, Madden, & Martin, 1998). With this method, the CBCL scores at age 12 and the SWAN scores of the sample were re-weighted so that the distribution of the problem behavior scores was the same as the distribution in the original sample. Using logistic regression analyses, the probability of being included in the selected sample was predicted for each twin pair based on their longitudinal CBCL-AP scores. As a result of our selection procedure, this probability was higher for twin pairs with high or low CBCL-AP scores than for twin pairs with moderate CBCL scores. Therefore, in the selected sample, twin pairs with a low probability of participation were underrepresented. To correct for this under-representation, these pairs received a higher weight than twin pairs with a high probability of participation. The logistic regression analyses and the calculation of weights were performed in SPSS (11.5; SPSS Inc., 2002). The weights were then used for the ensuing analyses in the statistical software package Mx (Neale, Boker, Xie, & Maes, 2003). The weights were entered as the fixed variable in the model and twin-pair scores were re-weighted by this variable.

Structural equation modeling, as implemented in Mx (Neale et al., 2003), was used for the genetic analyses. Mx provides parameter estimates by maximizing the raw data likelihood. The goodness of fit of nested models is evaluated by hierarchic likelihood ratio (\( \chi^2 \)) tests. Specifically, the \( \chi^2 \) statistic is computed by taking twice the difference between the log-likelihood of the full model and the log-likelihood of a reduced model (\( \chi^2 = -2(\text{LL}_0 - \text{LL}_1) \)). The associated degrees of freedom are computed as the difference in degrees of freedom between the two hierarchic models (Neale & Cardon, 1992). In a saturated model, means and standard deviations and phenotypic twin correlations were estimated.
The total variation of each variable can be decomposed into sources of additive genetic variance (A), non-additive genetic variance (dominance, D), common environmental variance (C) and unique environmental variance (E). A is due to additive effects of different alleles, D is due to non-additive genetic effects reflecting interaction effects between alleles of the same gene locus, C is due to environmental influences shared by members of a family, and E is due to environmental influences not shared by members of a family. E also includes measurement error and is therefore always included in the models. The effects of C and D in the classical twin design are confounded; C will decrease differences between MZ and DZ covariances while D will increase the differences. Therefore C and D cannot be estimated simultaneously.

A first impression of the relative importance of each component is obtained by inspecting the within-twin-pair correlations. MZ correlations as high as DZ correlations indicate only common and unique environmental influences and no genetic sources of variance. MZ correlations twice as high as DZ correlations indicate additive genetic influences. DZ correlations higher than half the MZ correlations designate common environmental influences while DZ correlations lower than half the MZ correlations point to dominance or contrast effects (Boomsma, Busjahn, & Peltonen, 2002a). Contrast and dominance effects can theoretically be distinguished by making use of the fact that contrast effects lead to differences in variances in MZ and DZ twins while non-additive genetic effects do not (Carey, 1986).

Results

Because there is no evidence for sex differences in heritability for ADHD (Derks et al., 2006a), data from male and female twins for both zygosities were combined in the analyses. There were no significant differences in means and variances between MZ and DZ twins for the CBCL ($\chi^2 (4) = 7.73, p = .102$) or for the SWAN/HI ($\chi^2 (4) = 3.24, p = .518$) and SWAN/AD scale ($\chi^2 (4) = 6.71, p = .152$). Means and standard deviations of the CBCL-AP scale and the HI and AD scale of the SWAN are shown in the upper part of Table 1. SWAN scores were normally distributed for both scales, covering the continuum across the strengths and weaknesses of ADHD characteristics, while CBCL-AP scores were skewed. In Figure 1 the histograms for the CBCL AP scale, the SWAN/HI scale and SWAN/AD scale are shown.

Table 1 Means (including the effects of sex on the means), SD and twin correlations of scores on the Attention Problem scale of the CBCL, and Hyperactivity/Impulsivity and Attention Deficit scale of the SWAN

<table>
<thead>
<tr>
<th></th>
<th>CBCL AP</th>
<th>SWAN Hyperactivity/Impulsivity</th>
<th>SWAN Attention Deficit</th>
</tr>
</thead>
<tbody>
<tr>
<td>Means boys/girls (SD)</td>
<td>3.09/2.33 (2.97)</td>
<td>43.9/45.6 (8.63)</td>
<td>44.0/45.7 (8.08)</td>
</tr>
<tr>
<td>Twin correlations (N pairs)</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>MZ</td>
<td>.67 (190)</td>
<td>.91 (221)</td>
<td>.85 (218)</td>
</tr>
<tr>
<td>DZ</td>
<td>.25 (269)</td>
<td>.43 (335)</td>
<td>.38 (331)</td>
</tr>
</tbody>
</table>

Figure 1 Distribution of scores of the CBCL AP scale, the SWAN Hyperactivity/Impulsivity scale and SWAN Attention Deficit scale

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Skewness and kurtosis of the CBCL were 1.76 and 4.50, of the SWAN/HI scale these were .10 and .06 respectively and of SWAN/AD these were -.13 and .06.

Figure 2 shows a scatter plot of SWAN scores on the y-axis and CBCL AP scores (at age 12) on the x-axis. Children who score high on the CBCL AP scale also show many HI or AD problems on the SWAN. Notable is the fact that children who show no variation (i.e., score zero) on the CBCL AP scale show variation on the SWAN. This pattern was similar for the HI and AD scale of the SWAN. The correlation between the CBCL and the SWAN/HI scale and the SWAN/AD scale were .38 and .42 respectively.

Twin correlations of MZ and DZ twin pairs of the CBCL AP scale and of the SWAN scales are shown in the lower part of Table 1. The twin correlation pattern of the CBCL AP scale showed DZ correlations lower than half the MZ correlations, which pointed to dominance or contrast effects. Because contrast effects cause different variances in MZ and DZ twins and therefore lead to different prevalences of Attention Problems among these groups, contrast effects were only included if the variances of MZ and DZ twins were different. This was not the case in the present data, so a model with dominance effects was tested for the CBCL data. The results showed that a model with additive (A, 21%) and non-additive (D, 52%) genetic effects and unique environmental influences described the data best. The broad heritability (i.e., A + D) estimate was 73%. To test whether the weighting procedure influenced these estimates we performed the same analyses in all available mother ratings of the CBCL at age 12 from which the SWAN samples originally were selected (birth cohort 1989–94; N = 2869 twin pairs). Estimates for A, D and E of the CBCL AP scale were not significantly different in the larger NTR sample and the current sample ($\chi^2 = 2.94$, df = 3, $p = .40$), indicating that the weighting method resulted in the correct parameter estimates in a selected sample. For both SWAN scales the DZ correlations were about half the MZ correlations, indicating additive genetic influences and unique environmental influences, and no influences of common environment, genetic dominance or contrast effects. Model fitting confirmed that a model with additive genetic and unique environmental sources of variance described the data well for the SWAN/HI scale and the SWAN/AD scale (see Table 2).

Heath et al. (1998) pointed out that as a result of data weighting, $\chi^2$ tests are biased. To investigate the direction of this bias we performed simulation analyses. These showed that the statistical test, in which the AE model is compared to the saturated model, is too conservative. When the AE model is the correct model ($H_0 = true$), we would normally expect to reject this model with a probability of 5% (Type-I error rate). In the simulations, it appeared that the probability of rejecting the AE model, given that the AE model is the correct model, is too high (65%). The fact that the AE models for both SWAN scales were not rejected therefore provides strong evidence that these models fitted well to the data. The heritability estimates were 90% for the Hyperactivity/Impulsivity scale and 82% for the Attention Problems scale.

**Discussion**

In this study the distribution and genetic architecture of the Strengths and Weaknesses of ADHD symptoms and Normal behavior Scale (SWAN; Swanson et al. 2006) was investigated. The SWAN is a questionnaire measuring Hyperactivity/Impulsivity (HI) and Attention Deficit (AD) with item scores on

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*Figure 2* Scatter plot with CBCL Attention Problem scores on the x-axis and SWAN-Hyperactivity/Impulsivity scores on the y-axis, and scatter plot with CBCL Attention Problem scores on the x-axis and SWAN-Attention Deficit scores on the y-axis. 

*Note:* Maximum score on the CBCL Attention Problem scale is 22. The more Attention Problems a child has, the higher his or her score on the Attention Problem scale. Maximum score on the SWAN is 63. The more Hyperactive/Impulsive or Attention Deficit problems a child has, the lower his or her score on the SWAN.
a seven-point scale, ranging from ‘average behavior’ to the extremes ‘far below average’ and ‘far above average’. So in contrast to most other checklists the SWAN scores cover the strengths as well as the weaknesses of a child, ranging from severe hyperactivity to normal activity and from serious attention deficits to a high level of attention. As a result, scores on the SWAN rating scales show a normal distribution in general population samples. The SWAN was compared to the attention problem (AP) scale of a widely used regular checklist, namely the CBCL. Such checklists often have skewed distributions because the range of responses to questions about problems is constrained to only a few possibilities (e.g., ‘never’, ‘sometimes’, ‘often’) and the majority of children in general population samples show no attention problems. The present study demonstrated that especially children who score zero (i.e., ‘never’), and hence show no variation on the AP scale of the CBCL, do show substantial variation on the ratings of the SWAN. The normal distribution of problem scores of the SWAN is particularly an improvement when assessing problems of hyperactivity and attention deficit in general population samples. It offers, for example, significant potential advantages in gene-finding expeditions, and in studies of quantitative endophenotypes. The correlation between the CBCL AP scale and the HI and AD SWAN scale was −.36 and −.43 respectively. However, this is probably an underestimation, as for the CBCL AP scale the variance of children who score zero is missing.

One of our interests was the contribution of the additional variance of normally ‘low scoring’ children to the underlying sources of variance of ADHD. We speculated that adding the variance of the ‘strength part’ of the SWAN might manifest the influences of common environment (C) as it is possible that these abilities are due to parental style or, for example, school systems. The only prior genetic study on parental ratings of the SWAN (Hay et al., 2007) showed substantial influences of C. In this study, SWAN data of a younger and an older sample, both consisting of around 500 twin pairs, were analyzed. DZ correlations in this study were unusually high (ranging between .50 and .78), and consequently the heritabilities were unusually low; 31% for hyperactivity in the older sample, for example. Their samples, however, were heterogeneous regarding age, especially in the older sample (i.e., age in the young sample ranged between 6 and 9, and in the older sample between 12 and 20).

The current study did not replicate the findings of Hay et al. (2007) as no evidence for common environmental influences was found. The DZ correlations were about half the MZ correlations and model fitting showed that additive genetic and unique environmental factors explained the variance of both SWAN scales. The heritability estimates (90% for HI and 82% for AD) were somewhat higher but comparable to the CBCL AP scale (73%) and to many previous studies on attention problems (Bartels et al., 2004; Rietveld et al., 2004, 2003a; Nadder et al., 1998; Nadder, Silberg, Rutter, Maes, & Eaves 2001; Derks et al., 2006b; Hudziak et al., 2000; Thapar, Hervas, & McGuffin, 1995; Thapar et al., 2000; Levy et al., 1997; Levy, McStephen, & Hay, 2001). The results for the CBCL AP scale in this sample, however, showed significant effects of dominance, while these effects were not found in the SWAN scales. The debate has been whether the low DZ correlations that are often found in twin studies on ADHD and attention problems are caused by non-additive genetic (i.e., dominance) effects or contrast effects. Studies in the past have shown evidence for both possibilities (Derks et al., 2006a). However, assuming that parents prefer to emphasize differences between their children rather than similarities, parental rater bias (and hence contrast effects) might be amplified by the narrow range of possible scores on regular checklists. The fact that no such effects were found in the SWAN ratings supports this idea. Instead of forcing a choice between the ‘often’ or ‘never’ possibility of most checklists, the wider range of scores of the SWAN makes it possible to rate one twin just a little bit more or less hyperactive than his or her co-twin. Consequently, contrast effects in this case might be avoided. However, to detect reliably (small) contrast effects or effects of dominance, larger
sample sizes (i.e., >1000 pairs) than the current one are needed (Rietveld, Posthuma, Dolan, & Boomsma, 2003b).

To summarize, the current results firstly demonstrated that the SWAN rating scale, in contrast to the CBCL, yields a normal distribution of scores covering the strength part as well as the weakness part of attention. This makes it a very useful instrument for examining variation of (hyper) activity and attention (problems) in the general population. In the search for more highly refined phenotypes, it appears that the SWAN offers added benefits by also obtaining ratings on positive attentional skills. These include the added statistical power that is gained in genetic and endophenotypic studies using a full quantitative trait. In addition, it might be an attractive option for clinicians to offer parents, because they can not only score the weaknesses of their child but also report on their strengths. Hay et al. (2007) also concluded that the SWAN might provide a more ‘realistic description of the ADHD phenotype’ than the ratings of problems do. Secondly, both SWAN scales showed a very high heritability estimate, but did not find any evidence of genetic dominance, or contrast effects. The extended range of scores on the SWAN (compared to regular checklists) that makes it possible to differentiate between children on a wider scale might be due to this.

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