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# Crying Without a Cause and Being Easily Upset in Two-Year-Olds: Heritability and Predictive Power of Behavioral Problems

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In order to estimate the influence of genetic and environmental factors on 'crying without a cause' and 'being easily upset' in 2-year-old children, a large twin study was carried out. Prospective data were available for ~18,000 2-year-old twin pairs from the Netherlands Twin Register. A bivariate genetic analysis was performed using structural equation modeling in the Mx software package. The influence of maternal personality characteristics and demographic and lifestyle factors was tested to identify specific risk factors that may underlie the shared environment of twins. Furthermore, it was tested whether crying without a cause and being easily upset were predictive of later internalizing, externalizing and attention problems. Crying without a cause yielded a heritability estimate of 60% in boys and girls. For easily upset, the heritability was estimated at 43% in boys and 31% in girls. The variance explained by shared environment varied between 35% and 63%. The correlation between crying without a cause and easily upset ($r$ = .36) was explained both by genetic and shared environmental factors. Birth cohort, gestational age, socioeconomic status, parental age, parental smoking behavior and alcohol use during pregnancy did not explain the shared environmental component. Neuroticism of the mother explained a small proportion of the additive genetic, but not of the shared environmental effects for easily upset. Crying without a cause and being easily upset at age 2 were predictive of internalizing, externalizing and attention problems at age 7, with effect sizes of .28–.42. A large influence of shared environmental factors on crying without a cause and easily upset was detected. Although these effects could be specific to these items, we could not explain them by personality characteristics of the mother or by demographic and lifestyle factors, and we recognize that these effects may reflect other maternal characteristics. A substantial influence of genetic factors was found for the two items, which are predictive of later behavioral problems.

■ **Keywords:** heritability, temperament, negative emotionality, shared environment

A significant number of children cry persistently in the first 3 to 6 months of life. Prevalence rates vary between 0.3% and 7.7% depending on the definition, but are usually reported to be around 5% (Reijneveld et al., 2001). Children displaying high levels of persistent crying and distress are a source of concern to their parents and a frequent reason to consult a family doctor or pediatrician (Forsyth et al., 1985). After the first months of life, levels of crying decrease and crying behavior becomes a more stable characteristic of the child. Children who frequently fuss or cry are thought of as having a difficult temperament. Difficult temperament shows significant continuity over time and is associated with internalizing and externalizing problem behavior and attention problems later in life (Bates et al., 1998; Caspi et al., 1995; Gjone & Stevenson, 1997; Guerin et al., 1997; Mun et al., 2001).

A recent review of twin and adoption studies reported that the heritability of temperament varied between 23% and 60% (Saudino, 2009). Studies investigating specifically

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the Emotionality and Irritability/Anger subscales, with items describing crying behavior and proneness to distress, yielded heritability estimates of 42–72% (Goldsmith et al., 1997; 1999; Saudino et al., 1995). The remaining part of the variance is mostly explained by unique environmental factors. Shared environment accounts for a small proportion of the variance in most temperament studies and its influence is often considered negligible (Saudino, 2009). The studies included in the review varied regarding sample size, from 50 to 800 individuals.

Most of these studies investigated the heritability of temperament dimensions by means of a sum score of a set of items that are thought to measure an underlying construct like negative emotionality. However, studies on personality have shown that genetic and environmental effects can be facet or item specific; these effects are not picked up when broad dimensions are analyzed (Heath et al., 1989; Jang et al., 1996). A methodological study showed that variance component estimates based on sum scores can differ substantially from the variance components of the underlying latent trait and proposed to use multivariate genetic analysis at the item or symptom level instead of sum scores (Neale et al., 2005). The literature on focal aspects of temperament is rather limited, but one study that estimated the influence of genetic and environmental factors on focal aspects and broader dimensions of temperament, found some genetic or shared environmental influences to be facet specific (Goldsmith et al., 1999).

The present study focuses on two items indicating two aspects of negative emotionality, namely crying without a cause (CWC) and being easily upset (EU). Data were available for more than 18,000 two-year-old Dutch twin pairs. The aim of the study was to estimate the heritability of these two specific behaviors and to establish the contribution of the shared and unique environment to these traits. The twins and their parents are participating in a longitudinal survey study of the Netherlands Twin Register. Data on environmental factors and maternal characteristics, were tested for their relation to CWC and EU in an attempt to assess the shared environment of the twins. In addition, it was investigated in a subsample of 8,994 twins whether CWC and EU at age two were predictive of internalizing, externalizing and attention problems at age seven.

## Materials and Methods

### PARTICIPANTS

The Netherlands Twin Register (NTR) is a population-based register that was established at the VU University Amsterdam in 1986. Newborn twins are enrolled in longitudinal survey projects (Bartels et al., 2007a). Parents receive questionnaires by mail until the twins are 12 years old.

Since 1988, two items indicating crying without a cause and being easily upset have been included in the survey that was sent out to all mothers of 2-year-old twins. Over 18,000 parents filled in and returned this questionnaire, with a response rate of 83.4%. In later versions of the questionnaire, respondents were asked to indicate their relationship to the twins; the respondent was the mother of the children in 93.8% of the cases, the father of the children in 5.1% of the cases, and had a relationship specified as 'other' in 1.1% of the cases. The number of twin pairs was almost equally distributed over cohorts 1986–2004.

### PROCEDURES AND INSTRUMENTS

#### Zygosity

If available, DNA or blood-group testing was taken as the conclusive result for zygosity determination. Zygosity for the remaining same-sex twin pairs was determined by a set of questionnaire items filled in by the parents of the twins through ages 3 to 12. This instrument correctly determines zygosity in 93% of same-sex twin pairs (Rietveld et al., 2000). In 11.3% of the cases, zygosity was determined by a single item in the questionnaire that was sent out at age 2 that indicates how much the children look alike. This question gives a correct determination of zygosity in 92% of the cases. For 16 twin pairs, zygosity determination was not available and they were excluded from the study.

#### Measures

The survey sent out at age 2 included two items that describe facets of negative emotionality. The first question, 'Did the children cry without a clear cause?', could be answered with *Rarely*, *Sometimes* or *Often*. The second question, 'Are the children upset for a long time if the usual course of events is disrupted?', could be answered with *Yes, A bit* or *Never/hardly ever*. Both items were rated for each child separately.

Data on parental age, maternal tobacco and alcohol consumption during pregnancy, paternal smoking behavior during pregnancy and gestational age were based on the first survey sent out after registration with the NTR. For the present study, data on maternal tobacco use during pregnancy were coded in two categories: *Yes* and *No*; data on paternal tobacco use during pregnancy were coded in four categories: *No*; *Yes, cigars/pipe*; *Yes, < 10 cigarettes per day*; *Yes, > 10 cigarettes per day*; and data on maternal alcohol use during pregnancy were coded in three answer categories: *No*; *Yes, < 1 glass per week*; and *Yes, > 1 glass per week*. Data on demographic and lifestyle factors and parental personality traits were available from several surveys. Socioeconomic status (SES) was based on a full description of parental occupation at age 3 in two-thirds of the families and coded according to the standard classification of occupations (CBS, 2001). For the remaining families, parental occupational status was based on Goldthorpe's class categories (EPG) combined with information on parental educational level at age 3 (Erikson et al., 1979). Complete data on demographic and lifestyle factors were available for



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13,065 twin pairs. Incomplete data were most often due to a lack of data on SES as a result of drop out from the study at age 3. Data on Neuroticism, Extraversion, Openness, Altruism and Conscientiousness were collected using 60 items of the NEO Five Factor Inventory (NEO-FFI; Costa & McCrae, 1992), which were included in a survey in 2009–2010 sent out to parents and twins aged 18 years and older registered within the NTR. Personality data were available for 1,040 mothers of twins.

An age-appropriate version of the Child and Behavior Checklist (CBCL 4-18) was included in the survey that is sent out at age 7. This checklist consists of 120 items that describe several behavioral problems. Parents are asked to rate the behavior of their child during the preceding six months on a three point scale, 0 = *Not true*, 1 = *Somewhat or sometimes true*, 2 = *Very true or often true*. The attention problems subscale describes both hyperactive and inattentive behaviors. The internalizing scale consists of the anxious/depressed, somatic complaints, and withdrawn subscales. The externalizing scale consists of the aggressive and rule-breaking behavior subscales (Achenbach, 1991; Verhulst et al., 1996). Data on CWC and EU at age 2 and internalizing, externalizing and attention problems at age 7 were available for 8,994 twin pairs.

## STATISTICAL ANALYSIS

Monozygotic (MZ) twins are genetically nearly identical while dizygotic (DZ) twins share on average 50% of their segregating genes. A higher resemblance of MZ twins compared to DZ twins for a particular trait is therefore likely to be due to the influence of genetic factors. Environmental factors that are shared between the twins will make MZ and DZ twins resemble each other, whereas unique environmental experiences will cause both MZ and DZ twins to differ from each other. When phenotypic data are available on MZ and DZ twin pairs, the total variance of the trait can be decomposed into variance due to genetic factors (A), common environment (C) shared by children from the same family and unique environment (E). When bivariate data are analyzed, it is possible to detect the causes of covariance between two traits; that is, the proportion of covariance that is due to additive genetic factors, shared environment or unique environment. For instance, using the same logic as before, when the correlation across-traits, across-twins is higher in MZ than in DZ twins (e.g., CWC in one twin and EU in the co-twin), it can be concluded that genetic factors common to both traits partly explain the covariance between the traits. For a more comprehensive summary of the methods in twin research, including tests for sex $\leftrightarrow$ genotype interaction we refer to Martin et al. (1997) and Boomsma et al. (2002).

### Liability Threshold Model

The two categorical outcome variables were analyzed using a liability threshold model. This model assumes that an unobserved liability underlies the measured categories of CWC and EU. The liability can be influenced by an individual's genetic makeup and exposure to environmental influences and is normally distributed with a mean value of 0 and a variance of 1. If a threshold on the liability scale is passed, children enter the next category of the ordinal scale. The thresholds are calculated as the number of standard deviations away from the mean. By definition, the area under the curve corresponds to the probability to be in a certain category (Figure 1). The resemblance between the first and second born of a twin pair is estimated by the correlation for the liability scale, called a polychoric correlation. Polychoric twin correlations were estimated using the software package Mx in a so called saturated model, in which the correlations and thresholds are estimated unconstrained over the different zygosity / sex groups. Then, thresholds were constrained to be equal over the different groups to test for effects of sex and zygosity. The fit of these models was compared with the saturated model. Analyses were performed on raw data using the full information maximum likelihood (FIML) method. The fit of the different models was compared by the log-likelihood ratio test (LRT). The difference in minus two times the log-likelihood (-2LL) between two models has a $\chi^2$ distribution with the degrees of freedom (*df*) equaling the difference in *df* between the two models. As we used a very large dataset, a $p$ value $< .01$ was taken as significant.

Next, a bivariate genetic model (Figure 1) was evaluated. Here, the variance in liability was decomposed using a model with three latent variables: additive genetic factors (A), shared environment (C) and unique environment (E). The variance in liability explained by A, C and E is calculated by squaring the factor loadings, which sum to 1. MZ twins correlate 1 for A and DZ twins correlate on average 0.5. By definition, C correlates 1 in all twin pairs, whereas E correlates 0. In a bivariate model, the genetic factors that influence CWC also load on the liability to EU, and the same is the case for the environmental factors. The Mx software package evaluates the fit of different values of the parameter estimates (i.e., factor loadings and thresholds) and provides estimates that offer the best fit of the model given the data.

A series of models was tested. First, an ACE model was fitted to the data that allowed estimates to differ in boys and girls. Then, the loadings on the A, C and E factors were constrained to be equal between sexes. Next, the AE and CE model were tested against the ACE model. Finally, we aimed to identify specific risk factors that could underlie the shared environment. Associations were tested between CWC and EU and birth cohort, socioeconomic status, parental age, maternal tobacco and alcohol consumption during pregnancy, paternal smoking behavior during pregnancy and gestational age. The thresholds of CWC and EU were regressed on these variables by including them in the bivariate model as fixed effects on the



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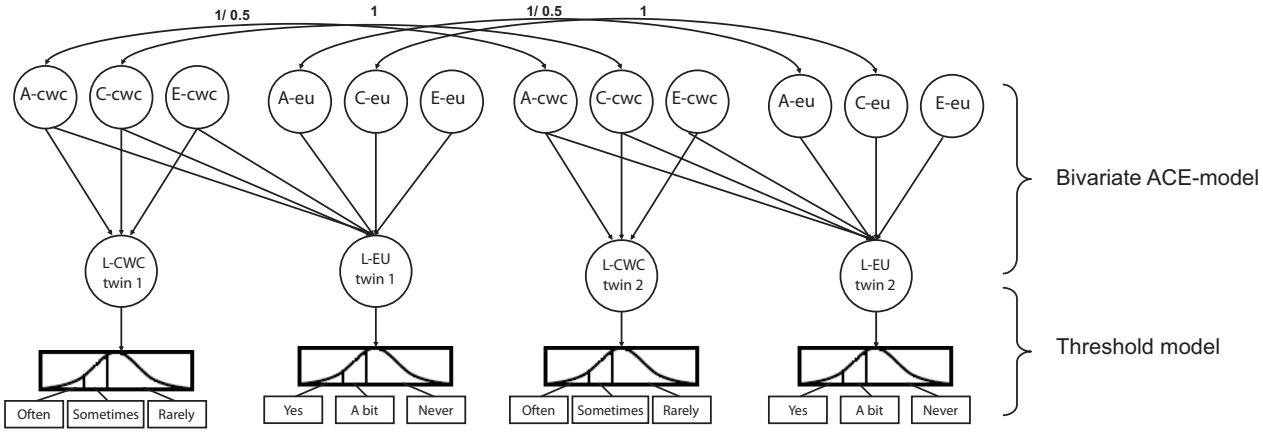


**FIGURE 1**

Path diagram of the bivariate model with the latent factors A, C and E and their influence on the liability to CWC and EU in twin 1 and 2 as modeled in the liability threshold model.

Note: A = additive genetic factors, C = shared environment, E = unique environment, L=liability to the trait, CWC = crying without a cause, EU = easily upset.

thresholds. These predictors were then dropped one by one and tested for a difference in model fit. Following the same procedure, it was tested if personality characteristics measured in a subsample of the mothers influenced the ratings of the children's behavior.

To test whether CWC and EU were predictive of internalizing, externalizing and attention problems at age 7, the children who were crying without a cause *Often* were compared to the children who were crying without a cause *Rarely* and *Sometimes*, and the children who were easily upset *Yes*, were compared to the children who were easily upset *A bit* or *Never/hardly ever*. Data from two individuals within a twin pair are not independent, therefore effect sizes were calculated for first born twins only. Effect sizes were calculated in terms of Cohen's *d*: the difference between two means divided by the pooled *SD*s for those means (Cohen, 1988).

## Results

Crying Without a Cause was reported to occur *Rarely* in 64.2%, *Sometimes* in 27.2% and *Often* in 8.6% of the cases. Children were Easily Upset *Yes* in 5.9%, *A bit* in 34.8% and *Never/hardly ever* in 59.3% of the cases. These frequencies are in line with general reports on the frequency of crying behavior (Reijneveld et al., 2001).

In the saturated model, thresholds could be constrained to be equal over all groups without a significant worsening of the fit ($p = .063$), except for the threshold of girls who were part of an opposite-sex (DOS) twin pair ($p < .001$). Thus, there were no overall sex differences in item responses, but girls with a male co-twin were consistently rated as less easily upset and less often crying without a cause than girls with a female co-twin. The mirror effect was not observed, that is, boys with a female co-twin do not differ from boys with a male co-twin. The phenotypic correlation for CWC and EU was 0.36 in boys and girls.

The polychoric twin correlations and the cross-trait cross-twin correlations (the correlation between CWC in one twin and EU in the co-twin) were estimated in the saturated model (Table 1). The within-trait and the cross-trait twin correlations were higher in MZ than in DZ twins, providing evidence for genetic influences on CWC and EU and on the covariation between the traits. However, MZ correlations were never twice as high as DZ correlations, indicating a role for shared environmental influences. Next, the ACE model was evaluated. The factor loadings were significantly different for boys and girls for EU, but not for CWC. Therefore, it was tested separately for males and females whether the influence of A, C and E on EU and the cross loadings from CWC on EU could be dropped from the model. The influence of A and C could not be dropped from the model without a significant worse fit, both for CWC and EU. None of the cross loadings could be dropped from the model (Table 2).

Parameter estimates are provided in Table 3. The correlation between the additive genetic factors influencing CWC and the genetic factors influencing EU was 0.36 in boys and 0.41 in girls. The correlation between shared

**TABLE 1**

Polychoric Twin Correlations Based on Maximum Likelihood Estimates of a Liability model with Three Answer Categories

| | Number of twin pairs | CWC twin correlation | EU twin correlation | Cross-trait cross-twin correlation |
|---|---|---|---|---|
| MZM | 2884 | 0.95 | 0.94 | 0.33 |
| DZM | 3165 | 0.64 | 0.73 | 0.24 |
| MZF | 3228 | 0.96 | 0.94 | 0.34 |
| DZF | 2861 | 0.64 | 0.78 | 0.25 |
| DOS | 6153 | 0.66 | 0.75 | 0.24 |

Note: CWC = crying without a cause, EU = Easily upset, MZM = monozygotic males, DZM = dizygotic males, MZF=monozygotic females, DZF = dizygotic females, DOS = dizygotic opposite-sex twin pairs



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**TABLE 2**

Model Fitting Results for the Bivariate Model of Crying Without a Cause (CWC) and Easily Upset (EU)

| Model | -2 times likelihood | Df | Comparison | $X^2$ difference | Difference in df | *p* value |
|---|---|---|---|---|---|---|
| 1. ACE model with sex differences on the path loadings | 101009.297 | 72762 | | | | |
| 2. ACE model no sex differences | 101034.315 | 72769 | Model 1 | 25.018 | 7 | < .001 |
| **3. ACE model no sex differences CWC path loadings** | **101018.424** | **72764** | **Model 1** | **9.127** | **2** | **.01** |
| 4. AE-ACE model | 101367.745 | 72765 | Model 3 | 349.321 | 1 | < .001 |
| 5. ACE-AE model (M) | 101472.907 | 72766 | Model 3 | 454.483 | 2 | < .001 |
| ACE-AE model (F) | 101545.533 | 72766 | Model 3 | 527.109 | 2 | < .001 |
| 6. CE-ACE-model | 102629.857 | 72765 | Model 3 | 1611.433 | 1 | < .001 |
| 7. ACE-CE-model (M) | 102130.111 | 72766 | Model 3 | 1111.687 | 2 | < .001 |
| ACE-CE-model (F) | 102128.917 | 72766 | Model 3 | 1110.493 | 2 | < .001 |
| 8. Drop A cross loading (M) | 101152.551 | 72765 | Model 3 | 134.127 | 1 | < .001 |
| Drop A cross loading (F) | 101135.759 | 72765 | Model 3 | 117.335 | 1 | < .001 |
| 9. Drop C cross loading (M) | 101098.258 | 72765 | Model 3 | 79.834 | 1 | < .001 |
| Drop C cross loading (F) | 101098.317 | 72765 | Model 3 | 79.893 | 1 | < .001 |
| 10. Drop E cross loading (M) | 101106.749 | 72765 | Model 3 | 88.325 | 1 | < .001 |
| Drop E cross loading (F) | 101073.005 | 72765 | Model 3 | 54.581 | 1 | < .001 |

Note: The best model is specified in bold font. As the factor loadings of EU showed significant sex differences, it was tested separately for males (M) and females (F) whether the influence of A, C, E on EU and the cross loadings of CWC on EU could be dropped from the model. CWC = crying without a cause, EU = easily upset, A = additive genetic factors, C = shared environment, E = unique environment, M = males, F = females.

environment influencing CWC and the shared environment influencing EU was 0.36 in boys and 0.34 in girls.

As a large influence of C was found, several demographic and lifestyle factors were tested for their influence on CWC and EU in a subsample of twin families ($n$ = 13,065), the results are shown in Table 4. Gestational age was the only predictor that could not be dropped from the model without resulting in a significant worse fit. However, the differences in A, C and E loadings in the model including all versus no predictors were very small. Moreover, the predictors influenced the factor loadings of A on CWC and EU rather than the factor loadings of C. Finally, maternal NEO scores were examined as predictors of CWC and EU ($n$ = 1,040), the results are shown in Table 5. Only Neuroticism of the mother significantly predicted EU, but not CWC. Including all personality scales as predictors in the model reduced the proportion of variance due to genetic factors with 2%. Again, the influence of shared environment was unchanged.

**TABLE 3**

Percentage of Variance and Covariance Explained by Additive Genetic Influences (A), Common Environment Shared by Twins (C) and Unique Environment (E) as Estimated in the Best Model Specified in Table 2

| Variance component | A (%) | C (%) | E (%) |
|---|---|---|---|
| CWC | 60.4 | 34.7 | 4.9 |
| EU (M) | 42.5 | 51.7 | 5.8 |
| EU(F) | 30.5 | 63.1 | 6.4 |
| Covariance CWC-EU explained (M) | 49.8 | 41.6 | 8.6 |
| Covariance CWC-EU explained (F) | 48.4 | 43.8 | 7.8 |

Note: CWC = crying without a cause, EU = easily upset, M = males, F = female.

The CWC and EU items were predictive of internalizing, externalizing and attention problems at age 7: first-born twins who were often crying without a cause and/or easily upset scored significantly higher on all three scales, as is depicted in Figure 2. Effect sizes were .30, .29 and .28 for CWC and .42, .38 and .41 for EU respectively. The polychoric correlations were .15, .15, and .13 for CWC and .19, .15 and .15 for EU.

## Discussion

In this large sample of 2-year-old twins, the heritability of CWC and EU was estimated between 30% and 60%. Shared environment explained 35–63% of the variance in CWC and EU. The unique environment, which by definition includes measurement error, explained only a small proportion of variance, around 5%. The large estimate of shared environmental influence was not explained by the effect of birth cohort, socioeconomic status, parental age, gestational age, parental smoking behavior and alcohol use during pregnancy, neither by personality characteristics of the mother.

Previous studies on the heritability of crying behavior after the first 3-6 months of life are mostly performed within the temperament framework. The Emotionality and Irritability/Anger subscales showed heritability estimates between 42–72% and negligible influence of shared family environment, both in studies using parental ratings and studies utilizing laboratory observations (Goldsmith et al., 1997; 1999; Saudino et al., 1995). The most obvious difference between the current and previous studies is that we assessed two specific behaviors with two items on a 3-point scale, whereas other studies utilized extensive temperament questionnaires and analyzed the according dimensions. It is therefore possible that we have detected



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**TABLE 4**

Results of the Regression of the Thresholds of CWC and EU Ratings on Several Demographic and Lifestyle Factors

| | CWC | | EU | |
|---|---|---|---|---|
| Model | Chi square | *p* value | Chi square | *p* value |
| 1. Drop birth cohort | 0.049 | .826 | 0.231 | .631 |
| 2. Drop SES | 4.211 | .040 | 2.832 | .092 |
| 3. Drop maternal age | 0.372 | .542 | 7.138 | .008 |
| 4. Drop paternal age | 1.420 | .233 | 0.020 | .887 |
| 5. Drop maternal smoking behavior | 0.321 | .571 | 1.531 | .472 |
| 6. Drop paternal smoking behavior | 0.226 | .634 | 0.004 | .951 |
| 7. Drop maternal alcohol use | 0.030 | .863 | 1.399 | .237 |
| 8. Drop gestational age | 27.306 | < .001 | 15.343 | < .001 |
| 9. Drop all | 41.739 | < .001 | 44.813 | < .001 |

Note: CWC = crying without a cause, EU = easily upset.

**TABLE 5**

Results of the Regression of the Thresholds of CWC and EU Ratings on the NEO Scores of the Mother

| | CWC | | EU | |
|---|---|---|---|---|
| Model | Chi square | *p* value | Chi square | *p* value |
| 1. Drop neuroticism | 3.799 | 0.051 | 30.054 | < .001 |
| 2. Drop rxtraversion | 0.020 | 0.888 | 0.030 | .864 |
| 3. Drop openness | 0.390 | 0.532 | 0.020 | .888 |
| 4. Drop agreeableness | 0.020 | 0.887 | 0.573 | .449 |
| 5. Drop conscientiousness | 0.656 | 0.418 | 1.200 | .273 |
| 6. Drop all | 5.486 | 0.360 | 37.405 | < .001 |

Note: CWC = crying without a cause, EU = easily upset.

effects that are unique to these specific behaviors. Multivariate item genetic analyses of temperament questionnaires that are more commonly used should clarify which genetic and shared environmental effects are shared among the items and which are item-specific. In general, large studies are needed to gain sufficient power to detect C (Posthuma & Boomsma, 2000). Our study has by far the largest sample size; therefore it is possible that previous studies did not detect an influence of C due to the smaller sample sizes. However, these studies report very low, sometimes even negative DZ correlations and these cannot be explained by small sample sizes (Saudino et al., 2000). Other explanations for the difference between our findings and the findings of previous studies on the importance of the shared environment may include different approaches: we used a liability threshold model, whereas other studies used a continuous variable.

No specific risk factors were identified that explained the shared environmental contribution in this cohort. No association between birth cohort, socioeconomic status, parental age, maternal tobacco and alcohol consumption during pregnancy, paternal smoking behavior during pregnancy and gestational age and CWC and EU item scores was observed. The only predictor that reached significance in our study was gestational age. The association between preterm birth and difficult temperament has been described before (Washington et al., 1986); however, in our study, its effect on the A, C and E loadings was very small. Most studies investigating specific environmental factors that are shared between family members, like parental style and family functioning, have not established an association with temperament measures (Daniels et al., 1984).

One possible explanation for large effects of C is 'rater bias'. This is the systematic error that occurs when raters consistently over- or underestimate behavioral scores. If a rater then has to report on more than one child, scores can become correlated due to characteristics of the rater and in an ACE model this will appear like 'C' (Bartels et al., 2007b). To explore this form of rater bias, the influence of maternal personality characteristics on CWC and EU ratings was estimated. A significant association was found only for Neuroticism and EU. However, this effect did not explain our large estimate of the influence of C. The model including Neuroticism of the mother yielded slightly lower A loadings and not lower C loadings. It is therefore probable that the influence of Neuroticism of the mother on EU ratings is due to genes that are shared between the mother and her children. In addition, the bivariate analysis showed that the correlation of CWC and EU ($r = .36$), was for 50% and 48% due to shared genetic factors and for 42% and 44% due to shared environment influencing both traits, in boys and girls respectively. If a large proportion of our estimate of shared environmental influence would be due to rater bias one would expect a higher correlation between the items, and the covariance between the items to be mostly explained by the shared environment that is common to both traits. Still, the large correlated influences of C on both items may reflect rater characteristics that we did not account for in this study.

CWC and EU at age two were found to be predictive of internalizing, externalizing and attention problems at age 7. Several previous studies have investigated the association between difficult temperament or emotionality and later behavioral problems. Significant associations have been reported in many studies, but the correlations reported differ widely (Bates et al., 1998; Caspi et al., 1995; Gjone & Stevenson, 1997; Guerin et al., 1997; Mun et al., 2001). This might be due to the differences across studies with regard to age of measurement and instruments used. A more comprehensive comparison might be found in the literature on crying behavior in the first year of life conceptualized as 'regulatory problems'. A recent meta-analysis on the association of regulatory problems in the first year of life and later behavioral problems reported effect sizes of .50 (95% CI 0.27 to 0.73), .56 (95% CI 0.31 to 0.82), and .42 (95% CI 0.06 to 0.77) when comparing children with and without crying problems on ratings of internalizing, externalizing and attention problems later in life (Hemmi et al., 2011). Altogether,



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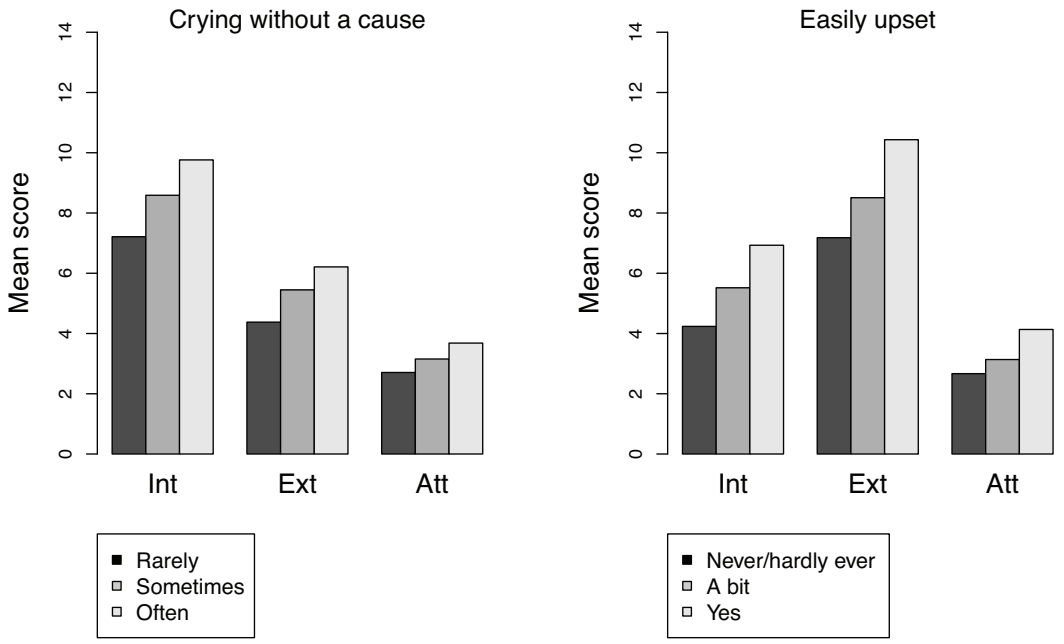


**FIGURE 2**

Mean internalizing (Int), externalizing (Ext) and attention (Att) problems scores at age seven for first born twins per answer category of the items Crying Without a Cause and Easily Upset.

the effect sizes found in the present study seem slightly smaller but comparable to the effect sizes found in previous studies.

A limitation of the present study lies in the measurement instrument used, as only two items were assessed. However, the vast size of the cohort under investigation and its embedding in a longitudinal study design is a unique advantage of the present study, as it allowed us to test for factors possibly underlying the shared environment of twins. Furthermore, the longitudinal aspect of the study made it possible to establish that CWC and EU are predictive of later behavioral problems and thus likely capture an important aspect of infant behavior.

Much debate has been spent on the issue of the validity of parental ratings versus observer or laboratory ratings of child behavior. Although these ratings tend to differ substantially, parental reports have the overwhelming advantage of in depth knowledge of the child's behavior over different time points and situations and are therefore still considered of utter importance. Moreover, parent rated data can be collected in large numbers, whereas laboratory measures are always limited in size due to practical infeasibilities.

A somewhat surprising result emerged from tests of prevalence and thresholds: females from DZ opposite-sex pairs were rated as less easily upset and less often crying without a cause than female same-sex twins. This effect was not seen in their brothers. One could consider the possibility that having a male co-twin is protective for girls, but the observed effect might also represent a rater effect. Rietveld et al. reported a comparable phenomenon with regard to hyperactivity and attention problems; opposite-sex female twins were rated less hyperactive than same-sex female twins (Rietveld et al., 2003). As these behaviors are more common in boys than in girls, this may cause parents to rate the female co-twin as less difficult when there is a brother as a comparison, a so called contrast effect. However, overall sex differences with regard to negative emotionality were not found in the present study nor in a large meta-analysis that assessed sex differences in temperament, including Negative Affectivity (Else-Quest et al., 2006). Despite the absence of sex differences, a contrast effect seems the most likely explanation for our findings.

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