Genetic and Environmental Influences on Temperament in the First Year of Life: The Puerto Rico Infant Twin Study (PRINTS)

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Three dimensions of temperament — difficult temperament, unadaptablility and unsociability — were assessed in the first year of life by maternal interview in twins born in Puerto Rico during 2001 and 2002. Eight hundred and sixty-five eligible mothers (80%) were traced and interviewed. Model-fitting results showed that additive genetic factors and the individual specific environment contributed to variation in all three dimensions. In addition, the pattern of variances and correlations suggested that sibling contrast effects influence ratings of difficult temperament. Moderate effects of the shared environment contributed to ratings of adaptability and sociability. There was a significant genetic correlation between difficult temperament and unadaptability. Genetic and environmental effects do not differ significantly between boys and girls. The study is the first population-based study of Puerto Rican twins and one of few to attempt the assessment of behavior in the first year. Preliminary results for difficult temperament and sociability were consistent with those in other populations and ages. In contrast, a significant effect of the shared environment on the temperamental trait of unadaptability has not been reported previously.

It is becoming increasingly apparent that serious behavioral and emotional problems are present in very young children, as early as preschool age (Angold & Egger, 2004; Campbell, 1995; Derks et al., 2004; Keenan & Wakschlag, 2002; Lavigne et al., 1996; McDonnell & Glod, 2003; Zeanah, 2000). While many young children manifest developmentally appropriate symptoms of aggression, noncompliance and anxiety, some children experience persistent and severe behavioral and emotional symptoms that impair the child's current and future functioning and development. While there has been a debate in the field of infant/young child mental health about the applicability of the *Diagnostic and Statistical Manual of Mental Disorders* (4th ed.; DSM-IV; American Psychiatric Association, 1994) criteria for identifying psychopathology in very young children (Campbell et al., 1995; Emde et al., 1993), there is a growing recognition that impairing mental health symptoms and syndromes appear early in life. Because of the obvious need for early intervention, researchers are seeking to identify potentially important early markers for these serious behavioral and emotional disorders in young children. Understanding the emergence of specific symptoms and patterns of symptoms across early childhood is a critical first step in defining targets for clinical interventions and preventative efforts.

This research goal is not new. Nearly 40 years ago, Thomas and Chess began their influential studies of early reaction patterns (Thomas & Chess, 1977; Thomas et al., 1968) for understanding the development of disordered behavior in young children (Thomas & Chess, 1977; Thomas et al., 1968). It was thought that these 'early emerging' behavioral traits, later termed *temperament* would be prognostic of later maladjustment (Rothbart & Bates, 1998). Since then, longitudinal studies have linked infant temperament (generally after 4 months of age) to later psychopathology (Guerin et al., 1997; Rothbart & Bates, 1998) even

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as far as young adulthood (Caspi et al., 1996; Kagan & Snidman, 1999). Despite the apparent link between early temperament and later behavioral problems, the etiological mechanisms are not yet well understood. The study of genetic factors in these early behaviors represents a promising approach for a deeper understanding of the mechanisms underlying these behaviors during the course of early development.

Despite varying definitions and labeling of temperamental constructs (Rothbart et al., 2000), twin studies have consistently established the influence of genetic factors on nearly all displays of infant temperament (Cherny et al., 1994; Cyphers et al., 1990; Emde & Hewitt, 2001; Emde et al., 1992; Matheny, 1989): activity level, negative affect and effortful control (Cherny et al., 1994), distress to novelty and stranger distress (Cherny et al., 1994), behavioral inhibition (Emde et al., 1992; Matheny, 1989; Robinson et al., 2004), shyness (Cherny et al., 1994; Cherny et al., 2001) and anxiety (Rothbart, 1981). However, whereas negative emotionality or infant 'difficultness' is best accounted for by genetic factors, positive emotionality is influenced by the shared environment (Goldsmith et al., 1997). Other substantive findings from the study of infant twins demonstrate: (1) the absence of genetic effect on temperament during the neonatal period (Riese, 1990); (2) the genetic relationship between early infant temperament and later behavioral problems (Goldsmith & Lemery, 2000; Schmitz et al., 1999); (3) the contribution of genetic influences to longitudinal stability in temperament (Plomin et al., 1993; Saudino & Cherny, 2001); and (4) the influence of environmental effects (primarily individual-specific) on developmental change (Saudino & Cherny, 2001).

Beyond estimating the overall role of genetic and environmental factors in infant behavior, twin studies also offer the unique opportunity to examine more complex processes that underlie the development of disordered behavior in early childhood. To date, however, there is not yet a study of infants that is large and sufficiently powerful enough to address issues related to heterogeneity across ages and gender, and to examine underlying mechanisms of gene–environmental interplay that are so critical to understanding the development of abnormal behavior (Eaves et al., 1977; Eaves et al., 1986; Eaves et al., 2003; Kendler & Eaves, 1986; Rutter & Silberg, 2001; Silberg et al., 2001). Because the complex genetic and social processes that lead to serious behavioral and emotional problems in children are likely to start as early as conception, an understanding of these processes requires that studies begin very early in life.

This present analysis introduces a genetically informed study of infant temperament in a large population-based sample of very young Puerto Rican twins. Apart from the obvious opportunity to study behavioral development in a understudied population, this sample of twins offers a unique opportunity for beginning the study of the complex interplay of genes and environment that underlies the development of psychopathology in young children.

Methods

Sample Ascertainment and Structure

In addition to the scientific value afforded by a distinct cultural context, Puerto Rico offers a series of practical advantages for population-based twin studies that are not shared with the North American mainland. The island is relatively small and well populated, and has excellent systems of communication. It has proved relatively easy to ascertain twins from birth records in the earliest months of life and participation rates are far higher than is common at the current time in the mainland US.

The Puerto Rico Neo-Natal Twin Registry (PRNTR) was initially established with the assistance of the Puerto Rico Department of Health (PRDH). On the mainland United States (US), the convenience of ascertainment from electronic birth records is offset by the delay in consolidating records. This often means that delays of a year or more intervene between birth and initial recruitment. In our study we were assisted greatly by the fact that the PRDH agreed to deliver a preliminary database containing basic twin birth information comprising names, addresses, and telephone numbers. We were thus able to contact and interview the twin families within the first 3 months of birth.

Population Demographics

From a total of 1079 multiple births in Puerto Rico from 2001 to 2002 we were able to interview 898 families for a response rate of 83%. A total of 538 (60%) of these interviews were done face-to-face. Twenty-nine of these families included triplets and were not included in the present analysis. Twin ages ranged from 0 to 32 months, with the median age being 6 months. The majority (99%) of parental respondents reported themselves to be the mother of the twins. Twelve per cent reported a single-parent family where no father was present. Twenty-one per cent of maternal respondents reported having obtained an education level of less than 9th grade; 19% reported having a high school degree or equivalent, such as a General Education Diploma (GED); and 59% reported having some level of college education. In households where both parents were present, 26% of fathers achieved an education level of less than 9th grade; 25% received a high school degree or GED; and 49% received college-level education or beyond. At the time of interview, 39% of maternal respondents reported themselves as either actively working (in full- or part-time jobs) or on temporary leave from a permanent job. Sixty-one per cent of mothers defined as *unemployed* identified themselves, with the majority reporting themselves as homemakers. Of households reporting the presence of both parents, 82% of fathers reported having employment, and 18% were unemployed.

Table 1

Results of Latent Class Analysis of Zygosity Items in Like-Sex Twins

Item	Response	Endorsement frequency (%)	
		DZ	MZ
Latent class frequency		56.4	43.6
Mistaken for one another (wave 1)	Rarely	63.9	22.1
	Sometimes	25.0	32.8
	Often	11.1	45.1
Parents opinion of zygosity (wave 1)	DZ	96.0	12.1
	MZ	4.0	87.9
Mistaken for one another (wave 2)	Rarely	62.6	11.1
	Sometimes	23.7	35.9
	Often	13.7	52.8
Parents opinion of zygosity (wave 2)	DZ	100.0	0.6
	MZ	0.0	99.4
Twins confused by mother (wave 2)	Rarely	87.1	16.2
	Sometimes	12.9	28.1
	Often	0.0	55.7
Can differentiate picture? (wave 2)	No	6.5	64.3
	Yes, hard	5.5	24.9
	Yes, easy	88.0	10.8
Believe twins identical (wave 2)	No	98.6	4.4
	Yes	1.4	95.6
As alike as two drops of water (wave 2)	No	89.8	36.2
	Yes	10.2	63.8

Assessment

Rothbart and Bates (1998) consider reactivity and self-regulation as key temperamental constructs of early development: 'Early reactive patterns of emotionality and approach are overlain by fearfulness in the first year and contribute to subsequent behavioral inhibition of action and emotion. Thereafter, individual differences in self-regulatory processes beginning around 18 months, which include effortful control, continue to develop across the preschool period'. Three specific dimensions of temperament are defined: (1) difficult temperament; (2) shyness/extraversion; and (3) from 18 months forward, self-regulatory processes that reflect the ability to sustain attention and inhibit behavior.

To capture early reactivity, and later behavioral inhibition and self-regulation, we planned two waves of assessment, separated by an interval of 1 year. The second wave is nearly completed. The Infant Characteristics Questionnaire (ICQ), was used in both phases of the study. The ICQ has adequate test–retest reliability and validity, and correlates moderately with observer ratings of difficult temperament, and other rating scales assessing distress to limitations (difficultness) and fear (unadaptability; Bates et al., 1979; J. E. Bates, personal communication, 2004). The translation and adaptation of the ICQ into Spanish and to the Puerto Rican culture involved a comprehensive process guided by a conceptual model (Flaherty, 1987; Matías-Carrelo et al., 2003) that focused on cross-cultural equivalence in three dimensions: semantic, content and technical equivalence. This model was used successfully in the translation of several diagnostic instruments in Puerto Rico (Bravo et al., 1991; Bravo et al., 1993; Matías-Carrelo et al., 2003). A similar approach was taken in assessing zygosity in the Puerto Rican sample. For example, the question for assessing zygosity in Caucasian samples, 'Are your twins as alike as two peas in a pod?', was translated into Spanish as 'as alike as two drops of water'.

To minimize subject burden, researchers employed a shortened version of the ICQ in the first wave. The 15 temperament items were rated on a 7-point scale, with higher scores reflecting increasing difficultness/fussiness, unadaptability and unsociability. The full 24-month version is used in wave 2. With the exception of a small set of items from the second wave to assist in the validation of zygosity assessment, the current report focuses only on the first wave for which the first 2 years of data collection are complete.

Data Analysis

Preliminary psychometric properties of the instrument, for example, underlying factor structure, for this population are comparable to those for a sample of US

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Caucasians. However, the scores, particularly of unadaptability and unsociability are somewhat lower than those found in other samples.

Zygosity Assignment

In most large-sample behavior–genetic studies such as ours, like-sex twin pairs are categorized either as monozygotic (MZ) or dizygotic (DZ) on the basis of a series of items relating to zygosity such as parental beliefs about zygosity and perceptions of similarity. When validated against diagnosis based on blood groups or DNA markers this approach typically classifies about 95% of pairs correctly (Kasriel & Eaves, 1976).

In our study, we used the items summarized in Table 1 to assign zygosity to the twins at wave 1. Where available, zygosity-related questions from wave 2 supplemented the information used to assign zygosity to twins in the wave 1 analyses. wave 1 items are repeated at wave 2 and more items are added at wave 2. Our algorithm for zygosity assignment was developed in two stages. First, we fitted two-class latent class model to all the zygosity items for all like-sex twin pairs on both waves. The model assumes that there are two groups of twins in the sample of like-sex pairs: MZ and DZ. The latent class analysis estimates the frequencies of the two zygosity classes and the probability that a parent will endorse each item for both types of twin. We used a program written in SAS (Version 6.12; SAS, 1996) for the latent class analysis (L. J. Eaves, unpublished) that maximizes the likelihood over all pairs allowing for missing data. Only like-sex pairs were included in the analysis of zygosity since unlike-sex pairs are unquestionably dizygotic.

At wave 1, an estimated 62% of mothers of putative DZ twins say they ‘rarely’ mistake their twins for one another, while only 11% of mothers of likely MZ twins claim rarely to mistake their twins.

In practice, the zygosity of any particular pair is not known with complete certainty. Instead, we estimate the probability that a pair will be MZ or DZ based on the available questionnaire data. This approach is comparable to that used for zygosity diagnosis using genetic markers that is described in most introductory texts on human genetics (e.g., McKusick, 1969). These are the ‘posterior probabilities’ of zygosity as they are computed after the responses to the zygosity items are known for a given pair.

Ideally, we would like these probabilities to be either 0 or 1, corresponding to the absolutely perfect separation of MZ and DZ twins into two groups. In practice, the posterior probabilities vary between the ideal limits, some being close to 1 or 0 but many falling short of either extreme and a significant handful of ‘doubtful’ cases where there is little evidence to support one zygosity assignment rather than another. The distribution of probabilities shows that 16.2% of like-sex pairs have less than 1% chance of being DZ (i.e., p [MZ] > 99%) and 18.3% like-sex pairs have greater than 99% chance of being DZ, based on available parental ratings.

Using the Posterior Probabilities of Zygosity in Genetic Data Analysis

In the past, zygosity was typically assigned to each pair based on the most likely category, which is based in turn on questionnaire responses. Genetic modelfitting then specifies separate expected covariance structures for MZ and DZ pairs and applies one or the other to every pair based on this zygosity assignment. However, this approach often discards a number of pairs for whom zygosity cannot be assigned with adequate certainty and does not deal adequately with the fact that even the best of algorithms for zygosity assignment by questionnaire has a significant risk of error even for some of the most likely assignments.

We follow a recent suggestion (Neale, 2003) that uncertainty about zygosity can be reflected in the computation of the likelihood of each twin pair’s responses by adding together the likelihoods based on the assumption of each zygosity type weighted by the posterior probabilities that the pair is MZ or DZ, $p$mz or $p$dz, based on the information available. The likelihood of each pair is the weighted sum of the likelihoods of obtaining the outcome measures for both MZ and DZ twins. Obviously, if zygosity can be assigned with complete certainty, either $p$mz = 1 or $p$dz = 1 for every pair, the weighted formula reduces to the traditional approach that separates putative MZ and DZ twins, assigning different expectations to each zygosity (e.g., Neale & Cardon, 1992). The new approach has the advantage of using each pair in a way that reflects the different degrees of certainty about the zygosity of each pair.

Table 3 summarizes the distribution of probabilities that like-sex pairs are dizygotic (p[DZ]) for the Puerto Rican sample based on the available questionnaire data. The corresponding MZ probabilities are simply p[MZ] = 1–p[DZ]. 16.2% of like-sex pairs have < 1% chance of being DZ (i.e., p[MZ] > 99%). Of the like-sex pairs, 18.3% have greater than 99% chance of being DZ, based on available parental ratings. We note that 20.7% fall into the intermediate range, where the probability of being DZ, based on

Table 2

Maximum Likelihood Estimates of Twin Correlations for Assessment of Difficult Temperament, Unadaptability, and Unsociability in Puerto Rican Twins During First Year of Life

	Twin correlation		
Twin type	Difficult	Unadaptable	Unsociable
MZm	.610	.562	.678
DZm	.175	.373	.337
MZf	.596	.638	.646
DZf	-.014	.385	.381
DZmf	.060	.463	.393

Note: Unlike-sex pairs are ordered so that the male twin is ‘Twin 1’.

the profile of questionnaire responses, lies between 6% and 84%.

Results

Preliminary Modeling of Genetic and Environmental Effects

The present analysis focused on scale scores derived for the three ICQ subscales, which were subjected to a square root transformation to minimize the effects of heterosecedasticity. The (6 × 6) covariance matrices for each of the five twin groups were estimated by maximum likelihood (ML), allowing for the unreliability of zygosity assignment. ML estimates of the twin correlations were derived from the covariance matrices. Table 2 gives the twin correlations for the three scale scores.

The raw twin correlations suggest the causes of individual differences in difficult temperament are different from those affecting unadaptability and sociability. Difficult temperament shows strong evidence of twin contrast effects that magnify twin differences or the effect of genetic nonadditivity (Carey, 1986; Eaves, 1976). Such effects reduce the correlations of DZ twins relative to those of MZ twins. Unadaptability and unsociability show larger DZ correlations relative to MZs, possibly indicating either the effects of the home or caregiver environment. It is also conceivable that such an effect arises from the maternal ratings. Whereas the findings for difficult temperament are consistent with many previous studies, the detection of a shared environmental influence on unadaptability has not been shown previously.

Univariate Analyses of Individual Temperament Scales

Initial univariate analysis of the three temperament scales focused on estimating the overall effects of additive and nonadditive genetic differences (A and D), the effects of the environment common to family members (C) and specific to individuals within each family (E). The effects of sibling interaction are reflected in the magnitude and sign of parameter (B). If sibling interaction exaggerates intrapair differences (contrast), B is negative. If sibling interaction increases intrapair similarity (cooperation), B is positive. At this age it is probably unlikely that there is direct reinforcement of behavior between twins, but that such an effect could arise through the process of maternal perceptual processes.

A preliminary grasp of the relative importance of these causes of variation is obtained by fitting a series of four models to the raw data (ACE, ADE, AE, ABE). These models can be fitted to the data either ignoring the effects of sex on the trait means and variances or allowing for heterogeneity in the effects of genes and environment across sexes ('sex limitation' models; Eaves et al., 1977). The statistical program Mx (Neale et al., 1999) that yields maximum likelihood estimates of scale means and the parameters of each gene–environment model for twin covariances is used. Nested models can be compared by likelihood-ratio chi-squares that assess the improvements in fit associated with selected increases in model complexity.

Table 3

Summary of Model-Fitting Results (Chi-Square Tests of Significance) and Best Fitting Univariate Models (Parameter Estimates) for Three Scales of Temperament in Puerto Rican Twins During the First Year of Life

Trait	Change in chi-square (*df*)			
	Sex-differences Model	None	Scalar	Nonscalar
Difficult	[C]	—	7.70(2)	6.31(1)
temperament	D	20.39(1)	7.75(3)	1.32(1)
	B	23.66(1)	8.81(3)	0.04(1)
Unadaptability	C	9.18(1)	0.67(3)	2.03(1)
	[D]	—	0.33(2)	9.27(1)
	B	9.26(1)	0.67(3)	1.93(1)
Unsociability	C	2.94(1)	0.62(3)	0.27(1)
	[D]	—	0.47(2)	1.75(1)
	B	2.95(1)	0.29(3)	0.13(1)

Trait	Parameter estimate				
	A	C	D	E	B
Difficult	44.3	0.0	—	55.7	—
	0.0	—	58.6	41.4	—
	74.9	—	—	25.1	–0.155
Adaptable	33.9	25.2	—	40.9	—
	63.6	—	0.0	36.4	—
	44.2	—	—	55.8	0.109
Sociable	48.9	14.4	—	36.7	—
	65.9	—	0.0	34.1	—
	56.7	—	—	43.3	0.057

Notes:1. All models include additive genetic effects (A) and individual-specific environmental effects (E). Models are identified by the addition of the third parameter to reflect either dominant genetic effects (D), environmental effects common to family members (C) or the mutual interaction (positive or negative) between twins (B).

2. When the estimate of the third parameter is zero, it is included in square brackets thus: [C] or [D] and the model reduces to the two parameter baseline A, E model.

3. Models of 'scalar' sex differences allow for all parameters in the model to have distinct values in boys and girls but do not allow for expression of different genes or common environmental effects in the two sexes.

4. Models of 'nonscalar' sex differences include an additional parameter to reflect sex-specific genetic and/or common environmental effects.

5. Models that include 'B' assume that the strength of twin interaction is not affected by the sex composition of the pair.

6. Estimates of A, C, D and E effects are expressed as percentages of the total phenotypic variance based on the model that assumed there are no sex differences in genetic and environmental effects. The effect of sibling interaction is expressed as the (reciprocal) effect of each twin on the others' phenotype. Parameters denoted by dashes (—) are not included in the particular model. Parameters with zero values are constrained at their lower bound by the data.

The summary statistics for model comparison are given for each scale in Table 3. In each case, the baseline model includes only two parameters, A and E that are assumed to be homogeneous in boys and girls. In Table 3, this model is denoted by the dash (—) in the corresponding cell. Effects of sex on genetic and environmental effects are indicated by the changes in chi-square associated with allowing the effects of A,

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C, D and E to differ across sexes ('scalar' sex differences) and by the addition of a further parameter ('nonscalar' sex differences) to reflect sex-specific genetic and environmental influences that inflate the correlations of like-sex DZ twins relative to unlike-sex DZ twins (Eaves, 1976). The addition of parameters under the scalar and nonscalar sex limitation models does not appear to result in a significant improvement in fit for any of the three temperamental traits. The parameter estimates under the best-fitting univariate models without sex differences are also summarized in Table 3. The AE model with contrast represents the best-fitting model to difficult temperament. An AE model with a cooperative effect that reflects a positive effect of one twin's rating on the other twin's fits only slightly better to the unadaptability and sociability scales than one that includes a shared environmental parameter.

Multivariate Analysis of the Three Temperamental Scales

The basic genetic analyses point towards a model for early temperament that includes both genetic and environmental effects. There is evidence that both genetic and individual specific environmental influences are responsible for some modest correlation between the different temperamental domains. Either nonadditive genetic effects or, more likely, twin contrast effects, also affect difficult temperament and some shared environmental effects or twin imitation or cooperative effects affect adaptability and sociability. A 'final provisional multivariate' model was tested that included additive genetic (A) and unique environmental (E) variances and covariances for all three temperament scales, contrast effects on difficult temperament only (B) and shared environmental effects (C) affecting only unadaptability and sociability and their covariance. Separate means were fitted to boys and girls, but the covariance structure was assumed to be the same in both sexes. This model has six means (three variables in each sex) and 10 parameters accounting for the covariance structure, 16 parameters in all. For this model, –2ln (l) was 14,700.045. A saturated model for the means and covariances of the five twin groups has 30 (5 × 6) means and 105 (5 × 21) components of variance and covariance, or 135 parameters that yield –2ln (l) = 14,601.348. The difference is a chi-square of 98.70 for 119 *df* ($p > 50\%$), suggesting an excellent fit of the parsimonious final model to the observed means and covariance structure. We note that a model replacing the shared environmental effects by twin imitation received less support [–2ln (l) = 14,714.311]. The estimated contributions of A, C and E to the temperament variables are given in Table 4.

The genetic and environmental correlations among the three temperamental traits are shown in Table 5. The unique environmental correlations are small. This finding is fairly typical and is commonly assumed to be due to the relatively large errors of measurement that contribute to E and are likely to be independent across traits. The genetic correlations are larger. There is clearly a substantial genetic component common to difficult temperament (D) and unadaptability (U) that far outweighs any environmental communality.

Table 4.

Estimated Contributions (% Phenotypic Variance) of Genetic and Environmental Effects to Variation in Infant Temperament During the First Year of Life

	Source of variation (% total)		
Temperament trait	Additive genetic (A)	Shared x genetic (A)	Unique environment (E)
Difficult	75.2	—	24.8
Unadaptable	31.5	21.0	40.5
Unsociable	54.8	9.9	35.3

Note: The contributions of A and E to difficult temperament are calculated without including the additional variance created by twin contrast effects (b = –0.162).

Discussion

In contrast to the US mainland, it has been possible in Puerto Rico to ascertain and interview a substantial population of twin families within the first year of life. The current report summarizes the results from two years' ascertainment of 865 same-sex and opposite-sex male and female infant twin pairs. The results of our study are based on maternal ratings rather than observational assessment and could, therefore, introduce spurious shared environmental or contrast effects. However, the model-fitting analysis suggests strongly that genetic differences contribute significantly to differences in temperament even in the first year of life. We conclude that different genes are responsible for much of the variation in the different dimensions of temperament, inferring that genetic factors contribute very early on to the differentiation of the principal dimensions of temperament that are recognized in older children. However, there is a large (greater than .5) genetic correlation between difficult temperament and unadaptability. At this stage, and with this moderately large sample size, we were unable to detect any marked sex differences in the contributions of genetic and environmental factors.

Overall our results are consistent with previous studies in other populations in demonstrating the importance of genetic and unique environmental influences on infant temperament. The finding of contrast effects in ratings of difficult temperament pervades many studies of negative emotionality of difficultness (Saudino, 2003). Although the effect of contrasting twins (i.e., rating twins more differently than they really are) can spuriously inflate estimates of heritability, correcting for contrast effects indicate genuine effects of the genes on this trait. We have also recently shown that evocative genotype–environment correlation, in which the behavior of parents is a response to the behavior of a 'difficult' child can also result in spurious contrast effects, providing a theoretical framework for

Table 5.

Genetic and Environmental Correlations Between Temperament Measures

	Correlation		
Trait pair	Additive genetic (A)	Shared x genetic (A)	Unique environment (E)
D-U	.586	—	.085
D-S	.145	—	–.026
U-S	.167	–.195	.137

Note: D = Difficult, U = Unadaptable, S = Unsociable

the widespread finding of very low DZ correlations in infant twin studies (Eaves & Silberg, 2005).

Our present results also reveal moderate effects of the shared environment on the maternal ratings of sociability and adaptability. Shared environmental influences have been associated with differences in sociability or positive emotionality previously (Cherny et al., 1994; Goldsmith et al., 1997), possibly reflecting indices of the care-giving environment. This is also consistent with the recent findings of shared environmental influences on attachment security in very young children (Bokhorst et al., 2003).

We have also detected some potentially important differences in the Puerto Rican community, specifically the detection of a shared environmental influence on adaptability. This may be explained by increased statistical power for detecting an effect of the shared environment in this large sample of twins. Alternatively, we may be assessing a quite different temperamental process given the very young age of the sample — the expression of genuine unadaptability or behavioral inhibition, characterized by shyness, fear, and withdrawal in novel situations, is thought to occur later in development. Nevertheless, preliminary analyses of the ratings of unadaptabiltiy in the second wave of the study indicate that this effect endures into the second year (J. L. Silberg, unpublished).

The finding of a shared environmental influence on adaptability coincides with studies that have shown significant differences in the social interaction between Puerto Rican and Caucasian mothers and their young children (Harwood et al., 1995). Whereas Caucasian mothers value mastery and self-sufficiency in young children, Puerto Rican mothers emphasize positive social interaction and respect of others. This early emphasis on community and social interaction in the Puerto Rican culture has been linked to lower rates of conduct problems in older Hispanic children (Bird et al., 2001) — we speculate that these same family/community factors that have an ameliorative effect on behavioral problems in older children may begin their influence quite early in development in effecting infant temperament. It is these kinds of cultural differences that make Puerto Rico a potentially important 'laboratory' for studying how family and/or community environmental factors may protect children by minimizing their risk for later behavioral maladjustment.

This data also shows significant genetic overlap between difficult temperament and unadaptability. This finding is consistent with the view that infant 'difficultness' reflects two patterns of emotionality: (1) frustration to stimulation deficit related to later externalizing behaviours; and (2) a heightened sensitivity to aversive stimuli related to later internalizing disorders. As we are planning on following this sample up into the preschool period, we will have the opportunity to test whether this common genetic liability to infant difficultness and unadaptability represents an important precursor to both kinds of behaviors in later development.

Genetic effects on behavior seem to be as important in infants as they are in later childhood and adult life. Beyond that, there are numerous gaps and uncertainties that will only be addressed by a much larger study targeted more towards the state-of-the art assessment of clinically significant outcomes within a broader context that takes into account contextual, family–environmental, cognitive and temperamental factors.

Ideally, future extension of this study will: (1) resolve the ambiguity of evidence for the role of the shared environment and its change with age; (2) detect heterogeneity in the effects of genes over sexes (sex-limited gene expression), across ages (G × age interaction), and environmental contexts (G × E interaction); (3) resolve differences in the roles of genes and environment between behavioral differences in the normal range and clinically significant outcomes; (4) identify specific environmental factors that contribute to clinically significant outcomes and detect their interaction and correlation with genetic differences; (5) resolve genetic and nongenetic patterns of symptoms and comorbidity among infant behavioral problems and their changes with age; and (6) provide the first clues about the generalizability of findings based on Caucasians to a Latino population.

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References

American Psychiatric Association. (1994). *Diagnostic and statistical manual of mental disorders* (4th ed.). Washington, DC: Author.

Angold, A., & Egger, H. L. (2004). Psychiatric diagnosis in preschool children. In R. DelCarmen-Wiggins & A. Carter (Eds.), *Handbook of infant, toddler, and*

https://doi.org/10.1375/twin.8.4.328 Published online by Cambridge University Press

preschool mental health assessment (pp. 123–139). New York: Oxford University Press.

Bates, J. E., Freeland, C. B., & Lounsbury, M. L. (1979). Measurement of infant difficultness. *Child Development, 50,* 794–803.

Bird, H. R., Canino, G. J., Davies, M., Zhang, H., Ramirez, R., & Lahey, B. B. (2001). Prevalence and correlates of antisocial behavior among three ethnic groups. *Journal of Abnormal Child Psychology, 29,* 465–478.

Bokhorst, C. L., Bakermans-Kranenburg, R. M., Fearon, P., van IJzendoornm, M. H., Fonagy, P., & Schuengel, C. (2003). The importance of shared environment in mother–infant attachment security: A behavioral genetic study. *Child Development, 74,* 1769–1782.

Bravo, M., Canino, G., Rubio-Stipec, M., & Woodbury, M. (1991). A cross-cultural adaptation of a diagnostic instrument: The DIS adaptation in Puerto Rico. *Culture, Medicine, and Psychiatry, 15,* 1–18.

Bravo, M., Woodbury-Fariña, M., Canino, G., & Rubio-Stipec, M. (1993). The Spanish translation and cultural adaptation of the Diagnostic Interview Schedule for Children (DISC) in Puerto Rico. *Culture, Medicine, and Psychiatry, 17,* 329–344.

Campbell, S. B. (1995). Behavior problems in preschool children: A review of recent research. *Journal of Child Psychology and Psychiatry, 36,* 113–149.

Carey, G. (1986). Sibling imitation and contrast effects. *Behavior Genetics, 16,* 319–341.

Caspi, A., Moffitt, T. E., Newman, D. L., & Silva, P. A. (1996). Behavioral observations at age 3 years predict adult psychiatric disorders. *Archives of General Psychiatry, 53,* 1033–1039.

Cherny, S. S., Fulker, D. W., Corley, R., Plomin, R., & DeFries, J. C. (1994). Continuity and change in infant shyness from 14 to 20 months. *Behavior Genetics, 24,* 365–379.

Cherny, S. S., Saudino, K. J., Fulker, D. W., Plomin, R., Corley, R., & DeFries, J. C. (2001). The development of observed shyness from 14 to 20 months: Shyness in context. In R. Emde & J. K. Hewitt (Eds.), *Infancy to early childhood: Genetic and environmental influences on developmental change* (pp. 269–282). Oxford, UK: Oxford University Press.

Cyphers, L. H., Phillips, K., Fulker, D. W., & Mrazek, D. A. (1990). Twin temperament during the transition from infancy to early childhood. *Journal of the American Academy Child and Adolescent Psychiatry, 29,* 392–397.

Derks, E. M., Hudziak, J. J., Beijsterveldt, C. E. M., Dolan, C. V., & Boomsma, D. I. (2004). A study of genetic and environmental influences on maternal and paternal CBCL syndrome scores in a large sample of 3-year-old Dutch twins. *Behavior Genetics, 34,* 571–583.

Eaves, L. J. (1976). A model for sibling effects in man. *Heredity, 36,* 205–214.

Eaves, L. J., Last, K. A., Martin, N. G., & Jinks, J. L. (1977). A progressive approach to non-additivity and genotype-environmental covariance in the analysis of human differences. *The British Journal of Mathematical and Statistical Psychology, 30,* 1–42.

Eaves, L. J., Long, J., & Heath, A. C. (1986). A theory of developmental change in quantitative phenotypes applied to cognitive development. *Behavior Genetics, 16,* 143–162.

Eaves, L. J., & Silberg, J. L. (2005). Parent–child feedback predicts sibling contrast: Using twin studies to test theories of parent-offspring interaction in infant behavior. *Twin Research and Human Genetics, 8,* 1–4.

Eaves, L. J., Silberg, J. L., & Erkanli, A. (2003). Resolving multiple epigenetic pathways to adolescent depression. *Journal of Child Psychology and Psychiatry, 44,* 1006–1014.

Emde, R. N., Bingham, R. D., & Harmon, R. J. (1993). Classification and the diagnostic process in infancy. In C. H. Zeanah, Jr. (Ed.), *Handbook of infant mental health* (pp. 225–235). New York: Guilford Press.

Emde, R. N., & Hewitt, J. K. (2001). *Infancy to early childhood: Genetic and environmental influences on developmental change.* Oxford, UK: Oxford University Press.

Emde, R. N., Plomin, R., Robinson, J., Corley, R., DeFries, J. C., Fulker, D. W., Reznick, J. S., Campos, J., Kagan, J., & Zahn-Waxler, C. (1992). Temperament, emotion, and cognition at fourteen months: The MacArthur Longitudinal Twin Study. *Child Development, 63,* 1437–1455.

Flaherty, J. A. (1987). Appropriate and inappropriate research methodologies for Hispanic mental health. In M. Gaviria (Ed.), *Health and behavior: Research agenda for Hispanics* (pp. 177–186). Chicago: University of Illinois Press.

Goldsmith, H., Buss, K. A., & Lemery, K. (1997). Toddler and childhood temperament: Expanded content, stronger genetic evidence, new evidence for the importance of the environment. *Developmental Psychology, 33,* 891–905.

Goldsmith, H., & Lemery, K. (2000). Linking temperamental fearfulness and anxiety symptoms: A behavior-genetic perspective. *Biological Psychiatry, 48,* 1199–1209.

Guerin, D. W., Gottfied, A. W., & Thomas, C. W. (1997). Difficult temperament and behavior problems: A longitudinal study from 1.5 to 12 years. *International Journal of Behavioral Development, 21,* 71–90.

Harwood, R. L., Miller, J. G., & Irizarry, N. L. (1995). *Culture and attachment: Perceptions of the child in context.* New York: The Guilford Press.

Kagan, J., & Snidman, N. (1999). Early childhood predictors of adult anxiety disorders. *Biological Psychiatry, 46,* 1536–1541.

Kasriel, J., & Eaves, L. J. (1976). The zygosity of twins: Further evidence on the agreement between diagnosis by blood groups and written questionnaires. *Journal of Biosocial Science, 8,* 263–266.

Keenan, K., & Wakschlag, L. (2002). Can a valid diagnosis of disruptive behavior disorder be made in preschool children. *The American Journal of Psychiatry, 159,* 351–358.

Kendler, K. S., & Eaves, L. J. (1986). Models for the joint effect of genotype and environment on liability to psychiatric illness. *American Journal of Psychiatry, 143,* 279–289.

Lavigne, J. V., Gibbons, R. D., Christoffel, K. K., Arend, R., Rosenbaum, D., Binns, H., Dawson, N., Sobel, H., & Isaacs, C. (1996). Prevalence rates and correlates of psychiatric disorders among preschool children. *Journal of the American Academy of Child and Adolescent Psychiatry, 35,* 204–214.

Matheny, A. P. (1989). Children's behavioral inhibition over age and across situations: Genetic similarity for a trait during change. *Journal of Personality, 57,* 215–235.

Matías-Carrelo, L., Chavez, L. M., Negron, G., Canino, G., Aguilar-Gaxiola, S., & Hoppe, S. (2003). The Spanish translation and cultural adaptation of five outcome measures. *Culture, Medicine, and Psychiatry, 27,* 291–313.

McDonnell, M. A., & Glod, C. (2003). Prevalence of psychopathology in preschool-age children. *Journal of Child and Adolescent Psychiatric Nursing, 16,* 141–152.

McKusick, V. A. (1969). *Human genetics*. Englewood Cliffs, NJ: Prentice-Hall.

Neale, M. C. (2003). A finite distribution mixture model from data collected from twins. *Twin Research, 6,* 235–239.

Neale, M. C., Boker, S. M., Xie, G., & Maes, H. H. (1999). *Mx: Statistical modeling* (5th ed.). Richmond, VA: Department of Psychiatry, Medical College of Virginia.

Neale, M. C., & Cardon, L. R. (1992). *Methodology for genetic studies of twins and families*. Dordrecht, the Netherlands: Kluwer Academic.

Plomin, R., Emde, R. N., Braungart, J. M., Campos, J. J., Corley, R., Fulker, D. W., Kagan, J., Reznick, J. S., Robinson, J., Zahn-Waxler, C., & DeFries, J. C. (1993). Genetic change and continuity from fourteen to twenty months: The MacArthur Longitudinal Twin Study. *Child Development, 64,* 1354–1376.

Riese, M. L. (1990). Neonatal temperament in monozygotic and dizygotic twins. *Child Development, 61,* 1230–1237.

Robinson, J., Kagan, J., Reznick, J., & Corley, R. (2004). The heritability of inhibited and uninhibited behavior: A twin study. *Developmental Psychology, 28,* 1030–1037.

Rothbart, M. K. (1981). Measurement of temperament in infancy. *Child Development, 52, 569–578.*

Rothbart, M. K., Ahadi, S. A., & Evans, D. E. (2000). Temperament and personality: Origins and outcomes. *Journal of Personality and Social Psychology, 78,* 122–135.

Rothbart, M. K., & Bates, J. E. (1998). Temperament. In W. Damon & N. Eisenberg (Eds.), *Handbook of child psychology* (5th ed.; pp. 105–176). New York: John Wiley & Sons.

Rutter, M. L., & Silberg, J. L. (2001). Gene–environment interplay in relation to emotional and behavioral disturbance. *Annual review of Psychology, 53,* 490.

SAS. (1996). *SAS user's guide: Statistics* (Version 6.12). Cary, NC: SAS Institute, Inc.

Saudino, K. J. (2003). Parent ratings of infant temperament: Lessons from twin studies. *Infant Behavior and Development, 26,* 100–107.

Saudino, K. J., & Cherny, S. S. (2001). Sources of continuity and change in observed temperament. In R. N. Emde & J. K. Hewitt (Eds.), *Infancy to early childhood: Genetic and environmental influences on developmental change* (pp. 89–110). Oxford, UK: Oxford University Press.

Schmitz, S., Fulker, D. W., Plomin, R., Zahn-Waxler, C., Emde, R., & DeFries, J. C. (1999). Temperament and problem behaviour during early childhood. *International Journal of Behavioral Development, 23,* 333–355.

Silberg, J. L., Neale, M. C., Rutter, M. L., & Eaves, L. J. (2001). Genetic moderation of environmental risk for depression and anxiety in adolescent girls. *British Journal of Psychiatry, 179,* 116–121.

Thomas, A., & Chess, S. (1977). *Temperament and development*. New York: Brunner/Mazel.

Thomas, A., Chess, S., & Birch, H. G. (1968). *Temperament and behavior disorders in children*. New York: New York University Press.

Zeanah, C. H. (2000). *Handbook of infant mental health*. New York: Guilford.