Stress trajectories in mothers of young children with Down syndrome

D. E. Most¹, D. J. Fidler², C. Laforce-Booth¹ & J. Kelly³

¹ School of Education, Colorado State University, Fort Collins, CO, USA
² Human Development and Family Studies, Colorado State University, Fort Collins, CO, USA
³ Family & Child Nursing, University of Washington, Seattle, WA, USA

Abstract

Background In this study, we investigated the early development of stress in mothers of children with Down syndrome, compared with mothers of children with developmental disabilities of mixed aetiologies. Growth modelling analyses were used to explore: (1) whether mothers of children with Down syndrome demonstrated distinct patterns of stress during their children’s early development, compared with mothers of children with other developmental disabilities; and (2) whether there was a relation between child behavioural characteristics and the level and rate of change in stress observed in each population.

Method The stress trajectories of mothers of young children with Down syndrome (n = 25) and a mixed-aetiology comparison group (n = 49) were estimated, using growth modelling on data collected at ages of 15, 30 and 45 months.

Results On average, stress in the mixed comparison group was higher at Time 1 and remained unchanged over time, while stress in the Down syndrome group was lower at Time 1 but increased steadily. After taking diagnostic group membership into account, more advanced cognitive-linguistic functioning and lower levels of maladaptive behaviours at all time points were associated with lower levels of maternal stress.

Conclusions These findings suggest that the cognitive-linguistic and behavioural trajectory observed in early development in Down syndrome may contribute to the changes in maternal stress levels observed throughout these early years. Implications for developing targeted and timesensitive family interventions for families of children with Down syndrome are discussed.

Keywords Down syndrome, family stress, early development, growth modelling, longitudinal data analysis

Introduction

Great strides have been made in studying the effects of genetic disorders on development over the past two decades (Dykens & Hodapp 2001). While researchers have continued to uncover the links among gene, brain and behavioural outcomes in genetic disorders such as Down syndrome, there has been a growing interest in the ‘developmental’ component of these developmental disorders. New emphasis has been placed on exploring developmental changes in behavioural phenotypes and the
specific developmental trajectories associated with different genetic disorders. While only a few studies have taken this developmental approach to the 'emergence of behavioural phenotypes', researchers from the neuroconstructivist perspective argue that this approach may contribute substantially to both the scientific understanding of behavioural phenotypes and the development of empirically driven intervention and early education techniques (Karmiloff-Smith 1997, 1998).

Studies to date that have taken this approach to understanding the emergence of behavioural phenotypes over time have found that, some areas of relative strength in middle childhood do not start out initially as areas of relative strength (Paterson et al. 1999). For example, toddlers with Down syndrome do not show relative strengths in visual processing (Fidler et al. 2005), although this strength is pronounced by middle childhood and adolescence (Jarrold & Baddeley 1997; Jarrold et al. 1999; Fidler et al. 2005). Beyond early development, this approach to understanding changes in behavioural phenotypes across development also may be useful for studying changes associated with other stages of development, in particular, adolescence. For example, cross-sectional research suggests that relative strengths in social functioning in children with Down syndrome may decrease with age (Fidler et al. 2005), while vulnerability to psychopathology outcomes increases with age in this population (Dykens et al. 2002; Fidler et al. 2005).

Although this ‘emergence of the phenotype’ approach has begun to be incorporated in behavioural research into the development of different disorders, very little exploration of the concomitant changes in family functioning over time has occurred. If family stress outcomes are related to child characteristics, and there is evidence that this is the case (Hodapp et al. 1997, 1998), then it may be the case that family responses show similar changes in trajectory as the child develops and changes. Most previous reports of stress in families of children with intellectual disability (ID) have taken a cross-sectional approach, and have not, for the most part, included child age as a major focus of exploration. The few exceptions have not used growth modelling (or other more sophisticated methodological techniques for examining change over time), and they have also focused only on outcomes over broad periods of time later in life (Seltzer et al. 1995). Yet, perhaps one of the most vulnerable times for families may be the other end of the lifespan – when the child with the developmental disability is born and during his/her earliest years. This is the time when families are coping with the reality of parenting and caring for a child with special needs, observing and discovering the various areas that are particularly challenging or lagging behind other areas, and struggling to take advantage of the window of opportunity for early intervention.

How might early developmental factors in Down syndrome impact family outcomes over the first few years of life? While there have been few explorations of stress – let alone changes in stress – in the earliest years of development, it may be that some factors associated with early development in Down syndrome impact family outcomes in specific ways that are not common to parents of all young children with developmental disabilities. If this is the case, then understanding changes in family stress over the first few years of a child’s life may be highly clinically relevant, especially in the planning of Individualized Family Service Plans. It may be that families of children with Down syndrome have a distinct pattern of stress over time that puts them at risk for clinically high levels of stress at specific points during their children’s development. With a better understanding of this pattern of trajectories, it may be possible to incorporate targeted and time-sensitive family interventions and support planning for times when families are experiencing their greatest degree of vulnerability.

In this study, we investigated the early development of stress in mothers of children with Down syndrome, compared with mothers of children with developmental disabilities of mixed aetiologies. Assessments of stress and other developmental measures were taken at three specific time points, when the child was 12–15, 30 and 45 months of age.

Growth modelling analyses were then used to explore: (1) whether mothers of children with Down syndrome demonstrated distinct patterns of stress during their children’s early development, compared with mothers of children with other developmental disabilities; and (2) whether there is a relation between child behavioural characteristics and the level and rate of change in stress observed in each population.
Method

Participants

Participants were 74 families with children with developmental delays. For the Down syndrome group (n = 25), nearly all child participants (21) had a genetic diagnosis of trisomy. For the mixed-aetiology comparison group (n = 49), 73.5% (n = 36) had idiopathic (non-specific) developmental delays. The remaining children in the mixed comparison group had other diagnoses, but not Down syndrome (Spina Bifida, n = 6; Tetralogy of Fallot, n = 1; Prader–Willi syndrome, n = 1; Williams syndrome, n = 1; VACTERL syndrome, n = 1; Translocation, chromosomes 11 & 15, n = 1; Duchenne Muscular Dystrophy, n = 1; Tubrous Sclerosis, n = 1). Demographic information by diagnostic group (Down syndrome; mixed aetiology) is shown in Table 1.

There is an ongoing debate in the study of research on ID syndromes regarding appropriate comparison groups. The approach used here was to try to represent the population of individuals with ID as a whole in the comparison group. Dykens et al. (2000) note that, ‘comparisons with groups with mixed aetiologies directly test whether a behavioural feature is characteristic of people with mental retardation in general or instead to the specific etiological group under study’ (p. 247). Thus, a mixed group of children — such as the one used in this study — would draw from children with non-specific (familial/environmental) ID, children with other genetic syndromes, children with pre-, peri-, and post-natal defects, and children with no identifiable aetiology for their ID, without overrepresentation of any group.

The diagnostic groups were matched on Bayley raw scores at Time 1, t (70) = 0.39, P = 0.69 (Down syndrome M = 68.58, SD = 15.35; Mixed M = 70.17, SD = 16.40; see Table 2). As per Mervis & Klein-Tasman (2004), raw scores were used for these analyses because a large percentage of children in both groups performed at the floor (49) of the available standard scores for the Bayley. Our matching procedures, which involved collecting data at the same time point for each child, made it possible to compare across groups on raw scores. In addition, raw scores have been used by a large number of studies of cognition in Down syndrome (Byrne et al. 1995; Cupples & Iacono 2000; Kay-Raining Bird et al. 2000; Boudreau 2002; Laws & Gunn 2002; Fidler et al. 2005).

No meaningful between-group differences were observed for child gender or ethnicity (see Table 1). Families of children in this study in both groups were predominantly Caucasian. The majority of mothers in each group were married at Time 1. However, the two groups did differ in two important ways. Families of children with Down syndrome had higher mean incomes, and mothers of children with Down syndrome were older compared with mothers of children in the comparison group. Because of these

<table>
<thead>
<tr>
<th></th>
<th>Down syndrome (n = 25)</th>
<th>Mixed aetiologies (n = 49)</th>
</tr>
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<tbody>
<tr>
<td>Child gender (% male)</td>
<td>68</td>
<td>65</td>
</tr>
<tr>
<td>Ethnicity (%)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>European American</td>
<td>88</td>
<td>86</td>
</tr>
<tr>
<td>Asian/Pacific Islander</td>
<td>4</td>
<td>2</td>
</tr>
<tr>
<td>African American</td>
<td>4</td>
<td>2</td>
</tr>
<tr>
<td>Other</td>
<td>4</td>
<td>10</td>
</tr>
<tr>
<td>Family income (Time 1)</td>
<td>45 000</td>
<td>30 000</td>
</tr>
<tr>
<td>Median (IQR/2)</td>
<td>16 875</td>
<td>16 875</td>
</tr>
<tr>
<td>Mother’s age (years)</td>
<td>34.60</td>
<td>31.59</td>
</tr>
<tr>
<td>Mother’s education (years)</td>
<td>14.72</td>
<td>13.65</td>
</tr>
<tr>
<td>Married (%)</td>
<td>88</td>
<td>82</td>
</tr>
</tbody>
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IOR, interquartile range.

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demographic differences, family income, maternal education and maternal age were included in all subsequent analyses.

Procedure

Families were recruited from 17 participating clinics or agencies providing early intervention services for infants with diagnosed disabilities, or follow-up services for infants at biomedical risk. A staff member at each agency identified infants who were the appropriate age, and then gave these infants’ mothers a descriptive pamphlet about the study, and asked if a member of our research team could telephone them to describe the study in more detail. Project staff obtained the names and telephone numbers of mothers who agreed to be called, and they contacted and recruited the families by phone (a few families contacted the project after hearing about the study from a friend, or obtaining information from a poster about the study). The staff did not recruit mothers of infants with significant sensory impairments, due to assessment difficulties.

At 12 months of age, an evaluator conducted a home visit to enrol the family into the study, to collect initial data about the family and child, and to assess the child’s development. At 15 months of age, mothers completed questionnaires during a home visit.

<table>
<thead>
<tr>
<th>Time 1 (12–15 months)</th>
<th>Down syndrome</th>
<th>Mixed</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>$n$ = 25</td>
<td>$n$ = 49</td>
</tr>
<tr>
<td>PSI</td>
<td>M (SD)</td>
<td>M (SD)</td>
</tr>
<tr>
<td>Total PSI</td>
<td>212.42 (37.09)</td>
<td>241.93 (35.59)</td>
</tr>
<tr>
<td>Bayley mental scale raw score</td>
<td>68.58 (15.35)</td>
<td>70.17 (16.40)</td>
</tr>
<tr>
<td>Child temperament</td>
<td>121.51 (17.00)</td>
<td>124.04 (21.62)</td>
</tr>
<tr>
<td>Time 2 (30 months)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>PSI total</td>
<td>233.37 (44.24)</td>
<td>244.43 (37.91)</td>
</tr>
<tr>
<td>Bayley mental scale raw score</td>
<td>103.24 (18.40)</td>
<td>115.11 (32.30)</td>
</tr>
<tr>
<td>Total CBCL $t$-scores</td>
<td>51.08 (7.54)</td>
<td>54.83 (10.83)</td>
</tr>
<tr>
<td>MacArthur CDI dev age</td>
<td>16.09 (3.52)</td>
<td>19.50 (7.55)</td>
</tr>
<tr>
<td>Time 3 (45 months)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>PSI total</td>
<td>244.05 (51.51)</td>
<td>242.23 (47.49)</td>
</tr>
<tr>
<td>DAS/Bayley standard scores</td>
<td>50.92 (6.04)</td>
<td>75.74 (20.91)</td>
</tr>
<tr>
<td>Total CBCL $t$-score</td>
<td>54.36 (8.92)</td>
<td>53.20 (12.04)</td>
</tr>
<tr>
<td>PLS total standard scores</td>
<td>56.44 (7.02)</td>
<td>79.30 (22.17)</td>
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<tr>
<th>Measure</th>
<th>Down syndrome M (SD)</th>
<th>Mixed M (SD)</th>
</tr>
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<tbody>
<tr>
<td>CBCL</td>
<td>51.08 (7.54)</td>
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</tr>
<tr>
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<tr>
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CBCL, Child Behavior Checklist; CDI, Communicative Development Inventory; DAS, Differential Abilities Scale; PLS, Pre-school Language Scale.

These two visits constitute Time 1 for the purposes of this study. At 30 and 45 months of age, children visited the laboratory playroom to assess child developmental outcomes and to obtain questionnaire data from mothers.

Measures

Demographics Questionnaire

Mothers provided information at the 12-month visit (and updated it at 30 and 45 months, when appropriate), regarding the study child’s birth order, gender and ethnicity; the mother’s age, education and marital status; and family income.

Bayley Scales of Infant Development (BSID-II; Bayley 1993)

A trained evaluator administered the revised BSID-II to the study children at 12 months and 30 months, yielding scores on the mental scale, the motor scale and the behaviour rating scale (BRS) for each time point. The mental and motor scales assess the child’s cognitive, language, personal-social, fine and gross motor development. The BRS assesses the quality of the child’s behaviour during testing in the areas of attention/arousal; orientation/engagement toward the
tasks, examiner and caregiver; emotional regulation; and quality of movement.

**Differential Abilities Scale (DAS, Elliott 1990)**

Cognitive performance at 45 months was indexed by the standard scores on either the DAS or the Bayley mental scales (if the child was functioning below the 2.5 years developmental level). The DAS is a battery of cognitive tests for children and adolescents aged from 2.5 to 17 years.

**MacArthur Communicative Development Inventory (CDI) (Fenson et al. 1993)**

The CDI is a standardized parent report measure that assesses a child’s lexical development. The toddler scale (words and sentences) was administered at 30 months (Time 2).

**Infant Temperament Questionnaire**

Mothers completed the Revised Infant Temperament Questionnaire (ITQ; Carey & McDevitt 1978) to provide an evaluation of their children’s temperament at 12 months. The 95-item ITQ was reduced to 39 items for the National Institute of Child Health and Human Development (NICHD) study, and this version was used in the present report.

**Child Behavior Checklist (Achenbach 1991a,b)**

At Time 2 (30 months), mothers completed the 99-item Child Behavior Checklist (CBCL; Achenbach et al. 1987) to assess behaviour problems in 2- to 3-year-old children.

**Pre-school Language Scale (PLS-3; Zimmerman et al. 1992)**

The PLS-3 was administered at 45 months. The PLS-3 examines a balanced and developmentally appropriate range of language behaviours, including the items that separately load on factors of vocabulary, morphology, syntax and integrative thinking.

**Parenting Stress Index (PSI; Abidin 1995)**

The PSI, which was administered at all three time points, is designed to measure both the degree to which the parent finds the child’s behaviours aversive or stress-inducing (child domain), and the degree to which the parent’s life is stressful more broadly (parent domain). Higher scores on the PSI indicate more negative characteristics.

**Analyses**

The first aim of the study was to describe trajectories of maternal stress over time, with particular attention to possible differences between mothers of young children with Down syndrome and mothers of children with developmental disabilities of mixed aetiologies. The second specific aim was to explore how patterns of changes in maternal stress are related to various child and family characteristics. The techniques of individual growth modelling (Singer & Willett 2003) are well suited to address these objectives, and it is this analytic framework that guided the analyses and the multilevel modelling language presented in the Results section.

The first set of analyses presented in the Results section describes individual empirical growth plots and fitted plots of maternal stress for each child. These analyses shed light on the general patterns (e.g. trends, functional form) of changes in stress over time. Fitted trajectories were also used to describe differences among diagnostic groups and to characterize the heterogeneity within groups.

The second set of analyses used the multilevel model for changes to explore the relation between various child and family characteristics and stress trajectories. Specifically, we examined predictors of the level of stress (especially at 45 months, the final time point) and the rate of change in stress over the 30-month study period. Child predictors included measures of IQ, problem behaviour and language development. The family characteristics under consideration were Time 1 measures of family income, mother’s education and age. As the child characteristics were of primary interest, the family characteristics were considered control variables.

Four sets of estimated growth models are presented. The primary purpose of estimating models in this exploratory context was to generate parsimonious prediction equations for stress. In all cases, stress was modelled as a linear function of age, and the age variable was centred at 45 months because of the
interest in estimating the relations between child characteristics and the final status of maternal stress (i.e. stress at Time 3). In the first set of models, only diagnostic group was considered as a predictor. In the next three sets of models, 15-, 30- and 45-month child characteristics, respectively, were considered. Within each set of models, each child characteristic was initially considered individually as a predictor (of the level of stress and rate of change in stress). A best-fitting prediction equation was established for each set of models by considering all child characteristics simultaneously as predictors of the level and rate of change in stress over time. The best-fitting equation, in each case, had the smallest deviance and only retained predictors with substantively meaningful partial relations (i.e. after controlling for the other predictors) with stress. Proportional reductions in the variance components (an approximation to explained variance) were computed for each model, with particular attention to reductions in the between-person variability in the level and rate of change in stress accounted for by child characteristics. Computing a proportional reduction requires some sort of baseline model, and as there were a few missing predictor values at each time point, each set of models included its own baseline unconditional growth model (i.e. a model in which the only predictor of stress was age over time). The parameter estimates were essentially the same across all baseline models.

It is important to note that for Time 1 and Time 2 growth models, Bayley raw scores were used to connote IQ. For time 3 growth models, standard scores were used to translate across the Bayley and the DAS for children of different functioning levels.

Results

Trajectories of stress

We began with an examination of an empirical growth plot of stress for each participant family. A simultaneous inspection of all of the plots revealed variability across participants in the level (degree) of stress at any given time and in changes in stress over time. Increases in stress over time were more common than decreases, although in many instances, there was little change over time. The biggest increases tended to be larger in magnitude than the biggest decreases. Excluding a single outlier on each end of the spectrum, the largest decreases in stress were only 60% as large as the largest increases. It also appeared that, for most of the participants, a linear functional form would adequately characterize the trajectory of changes in stress over time.

Each participant’s growth trajectory was summarized by fitting a separate Ordinary Least Square (OLS) regression to each person’s data, assuming linear changes in stress over time. The within-person regression models were then used to generate fitted trajectories. A plot of each participant’s fitted trajectory, grouped by diagnosis, is presented in Fig. 1. It shows that both fitted levels of stress and rates of change in stress varied across participants. Although it is not presented here, an individual $R^2$ statistic was computed for each participant as a method of quantifying the quality of fit. In general, the $R^2$ values were very high, suggesting that a linear trajectory reasonably captured the changes in stress over time.

![Figure 1 OLS fitted trajectories of stress by diagnostic group. DS, Down syndrome; OLS, Ordinary Least Square; PSI, Parenting Stress Index.](image-url)
Stress trajectories in Down syndrome vs. mixed comparison group

Figure 1 shows the substantial within-diagnostic group heterogeneity in trajectories of stress. In both diagnostic groups, there were cases in which stress increased, decreased and changed little over time. However, the trajectories also differed by diagnostic group. There was a much higher proportion of positive slopes (i.e. increases in stress over time) in the Down syndrome group (~76%) than in the mixed comparison group (~51%).

This difference only tells part of the story, however, as the range in slopes differed by diagnostic group. While the range in magnitude of the positive slopes within each diagnostic group was similar, the larger (in magnitude) negative slopes in the mixed comparison groups were more than three times as large as those in the Down syndrome group. Even in the few cases where mothers in the Down syndrome group experienced a decrease in stress over time, the magnitude of the decrease was notably smaller than the decreases experienced in the mixed comparison group. In sum, the individual fitted OLS trajectories suggest that, in the mixed comparison group, there was more variability in the rates of change in stress over time (from large increases to large decreases) than in the Down syndrome group, in which rates of change in stress ranged from large increases to essentially flat or very slight decreases over time.

Growth models

Diagnostic group

We next explored the relation between various child and family characteristics and stress trajectories. The first model estimated was an unconditional growth model (Singer & Willett 2003), assuming a linear change trajectory. A linear functional form is assumed for all growth models presented, both because the descriptive analyses suggested that it would be quite reasonable and because of statistical constraints. The results of the unconditional growth model are presented in Table 3. In order to have the intercept correspond to the level of stress at Time 3, time was centred at 45 months. The estimated level-2 variance components suggest that there is substantial variability between participants in final status and rate of change.

The next model estimated (see Table 3) included diagnostic group as a predictor of both final status and rate of change in stress over time. The results indicated that level of stress at 45 months was not dissimilar across diagnostic groups. With a point estimate of −0.0065, the average monthly rate of change in stress over time for the mixed comparison group was not significantly different from zero. However, the average monthly changes in stress in the Down syndrome group was 1.05 units higher. So, while stress in the mixed comparison group remained flat on average over time, it increased approximately one
point per month on average in the Down syndrome group. This is equivalent to an average 30 point increase in stress in the Down syndrome group over the study period. Fitted trajectories reflecting this difference are presented in Fig. 2. Note that it displays average trajectories of stress over time in each diagnostic group and does not depict the within-diagnostic group heterogeneity in trajectories discussed earlier. The decrease in the level-2 variance component for the rate of change, from 0.76 in the unconditional growth model to 0.53 in the current model, indicates that approximately 30% of the variability across participants in rates of change in stress can be accounted for by diagnostic group membership.

**Time 1 predictors**

Child IQ at 12 months, child temperament at 12 months and the parent control variables were the Time 1 predictors considered. The best-fitting model (see Table 4) did not include any predictors of rate of change in stress other than diagnostic group. There were, however, significant predictors of the level of stress. Among the parent control variables, only family income significantly improved the fit of the model. The child predictors, IQ and temperament, were both associated with the level of stress, controlling for all other predictors in the model. Approximately 18% of the variability across participants in stress levels can be accounted for by diagnostic group membership, family income, and child IQ and temperament at 12 months.

**Time 2 predictors**

Child IQ, problem behaviour (total CBCL) and language age (MacArthur CDI developmental age) at 30 months were the Time 2 predictors, considered along with the parent control variables. Due to the high degree of association between IQ and language age, two different best-fitting models were estimated; one with IQ and one with language age (see Table 5). All of the child predictors, IQ, language age, problem behaviour at 30 months, and diagnostic group were associated with stress at 45 months, controlling for all other predictors in the model. Other than diagnostic group, only IQ was associated with the rate of change in stress.

As was the case with the Time 1 predictors, after controlling for child characteristics, most of the estimated effects of diagnostic group on the level and rate of change remained similar to the uncontrolled effects. The only modest change was in the model in which IQ was controlled. In the model with IQ, the decreases in the level-2 variance components indicate that approximately 59% of the variability across participants in stress at 45 months and 55% of the variability in the rate of change in stress over time can be accounted for by child characteristics. The comparable values in the model with developmental age are 52% and 30%.

**Time 3 predictors**

Child IQ, problem behaviour and language (PLS-3 adjusted total language standard scores) at 45 months were the Time 3 predictors, considered along with the parent control variables. As was the case with the predictors at 30 months, due to the high degree of association between IQ and language at 45 months, two different best-fitting models were estimated; one with IQ and one with language (see Table 6). IQ, language and problem behaviour at 45 months were associated with stress at 45 months and the rate of change in stress, controlling for all other predictors in the model. The magnitudes of the effects of IQ and problem behaviour on stress at 45 months are quite similar to those of the same predictors at 30 months. The effects on the rate of change in stress, however, are somewhat different.

Unlike the results of the Time 1 and Time 2 model, after controlling for Time 3 child characteristics, the
relations between diagnostic group and stress were not the same as in the uncontrolled model. Specifically, once IQ or language at 45 months was taken into account, diagnostic group was not a predictor of the rates of change in stress over time. Diagnostic group, however, was a predictor of the level of stress. Holding constant IQ or language at 45 months and problem behaviour, stress was, on average, approximately 25 points lower in the mixed comparison group.
While either the IQ or language model may be useful for prediction purposes, given the limited overlap between diagnostic groups in the distribution of IQ or language scores at 45 months, these results should not be interpreted as suggesting that diagnostic group is not associated with rates of change in stress. Rather, the association between IQ or language at 45 months and rates of change in stress can be understood as an effect of diagnostic group membership. In the model with IQ, approximately 59% of the variability across participants in stress at 45 months and 78% of the variability in the rate of change in stress over time can be accounted for by diagnostic group membership, IQ and problem behaviour at 45 months. The comparable values in the model with developmental age are 61% and 77%.

Summary

The individual fitted OLS trajectories (see Fig. 1) suggested that there was more variability in the rates of change in stress of time in the mixed comparison group than in the Down syndrome group, and no families in the Down syndrome group experienced substantial decreases in stress during the study period. On average, stress in the mixed comparison group remained unchanged over time, while it increased in the Down syndrome group (see Fig. 2).

After taking diagnostic group membership into account, stronger cognitive-linguistic skills and lower levels of maladaptive behaviour at all time points were associated with lower levels of stress. IQ at 30 months and all child characteristics at 45 months were associated with rates of change in stress. Because of the minimal overlap between diagnostic groups in the distribution of cognitive-linguistic characteristics at 45 months, these associations also can be understood as resulting from group membership status. The associations between child characteristics and stress did not vary by diagnostic group. Controlling for diagnostic group membership and other child characteristics, including the parent control variables (except family income in the Time 1 model) did not result in an improved model. Across all models, a
high proportion of the variability in the level and rate of change in stress over time can be accounted for by diagnostic group membership and other child characteristics.

Discussion

This study explored stress trajectories in families of young children with Down syndrome and in families of young children with developmental disabilities of mixed aetiologies. Families participated in this study at three time points: when children were 12–15, 30 and 45 months old. As such, this study is among a small number of studies to explore changes in stress over time in families of children with developmental disabilities, and it is unique among these studies in its focus on changes in maternal stress across the early developmental years of the child with disabilities.

Although the two diagnostic groups were equated on developmental functioning (cognition and behaviour) at Time 1, findings from this study suggest that families of children with Down syndrome experience a different stress trajectory during the early developmental years than do families of children with other developmental disabilities. Families of children with Down syndrome showed lower levels of maternal stress as measured by the PSI at Time 1, when children were 12–15 months old. However, by the third time point (45 months), levels of maternal stress in the Down syndrome group, on average, were equal to those in the mixed comparison group. It is important to note that these findings represent overall trends in each diagnostic group, and within-diagnostic group variability was observed in these stress trajectories, despite these overall between-diagnostic group patterns.

Findings from this study also suggest that the specific cognitive-linguistic and behavioural patterns observed in early development in Down syndrome may contribute to the changes in maternal stress levels observed throughout these early years. The growth modelling analyses performed in this study indicated that at each time point, cognitive-linguistic and behavioural factors were associated with maternal stress levels. This suggests that the early emergence of the Down syndrome behavioural phenotype may play an important role in shaping maternal experience. As behavioural patterns become more pronounced (cognitive delays, language delays, maladaptive behaviours) during the first 3 years of development in children with Down syndrome, maternal stress levels increase. Clinically speaking, this suggests that stress in families of children with Down syndrome may present differently at various time points during a child’s early development. While parents may show relatively lower stress levels when their children are emerging from infancy into toddlerhood, they may be at increased risk for higher stress levels as the children enter their early pre-school years.

At first glance, these findings may seem to contradict with previous studies that report lower levels of stress in families of children with Down syndrome when compared with families of children with other developmental disabilities. In this study, we found that by Time 3, equal levels of stress were reported in both diagnostic groups, while previous studies have reported that when matched for overall functioning level, families of children with Down syndrome tend to show lower levels of reported family stress (Fidler et al. 2000; Hodapp et al. 2002). There is one critical difference, however, that distinguishes the present study from previous reported findings. Previous studies primarily have selected diagnostic groups for comparison with Down syndrome that are matched on developmental level at a particular time point in middle childhood, adolescence or early adulthood. In this study, we prospectively followed two diagnostic groups of children (Down syndrome and mixed aetiologies) throughout their earliest childhood years. Diagnostic groups were equated on Bayley mental scale raw scores at Time 1; however, as children in each diagnostic group developed, they did not show similar patterns of progress along cognitive or language-related dimensions. As a result, we were able to capture the dynamic process of how development interacts with the family experience, and how starting points in early development in Down syndrome may lead to unexpected endpoints as a result of the behavioural trajectories associated with a particular genetic disorder. As such, this study contributes to the line of research on the 'Down syndrome advantage' in family functioning, where several studies have reported lower levels of stress and higher levels of rewardingness in the parenting experience (Holroyd & McArthur 1976; Mink et al.)
Uncovering this dynamic interplay was only possible with the use of longitudinal data. In fact, it could be hypothesized that if different IQ-matched comparison groups were selected for use at each time point in this study, we might have observed the pattern of comparatively lower levels of maternal stress in the Down syndrome group, similar to what has been reported previously. It is important to consider that equal levels of stress were observed between diagnostic groups at Time 3, when by this time, the group of children with Down syndrome was considerably lower functioning than the children in the mixed comparison group. Thus, the longitudinal nature of this study offers a different window into the experiences of families of children with Down syndrome during the first few years of their children’s life, and how they may be at risk for a unique trajectory of stress that is linked to the emergence of their children’s phenotypic characteristics.

There are several implications for this study. First, these findings may be important to consider when shaping early family intervention services in this population. It may be that families of children with Down syndrome necessitate a different pattern of services to address the changes in stress levels that may emerge as the child transitions from toddlerhood to pre-schooler status. In particular, it may be important to increase the knowledge and skill level of professionals in this area, so they can anticipate possible or even probable increased parental stress levels as children with Down syndrome age. With increased knowledge and skills, professionals can address increased stress levels by providing their support and understanding, by helping parents emotionally adapt to child characteristics, and by helping parents access additional support and services that can decrease stress.

A second implication for this study relates to how the research and practice communities conceptualize family experience in different diagnostic groups. Between-diagnostic group differences in stress trajectories also may be explained in ways that relate to the ‘road to the diagnosis’ in Down syndrome vs. other developmental disabilities. Families of children with Down syndrome receive a diagnosis either at birth or shortly thereafter, often even before the child is born. Thus, parents have the opportunity to begin the process of assimilating this information into their expectations, understanding their parenting role, and in many cases, beginning a coping process. However, many parents of children with mixed developmental disabilities do not have diagnoses for their children – and perhaps no expectation of atypicality in their children’s development – and begin to observe delays in their children throughout infancy without explanation.

Beyond these implications for practice, this study is part of the larger movement of research that explores the interplay between behavioural phenotypes in different genetic disorders and family outcomes. As such, this study contributes to our understanding of the indirect effects of genetic disorders on family functioning (Hodapp 1999), by adding a dynamic, longitudinal component to this theoretical approach. To date, most studies of the indirect effects of behavioural phenotypes of families have explored effects cross-sectionally (Fidler et al. 2000; Hodapp et al. 2002). Yet, contextual theorists have argued for decades that there is a transactional nature to the bidirectional effects of parents and children on one another (Sameroff 1975). Including a temporal component to the study of functioning in families of children with genetic disorders deepens our understanding of how different syndromes impact families in unique and important ways.

There are several important limitations to this study that should be considered. First, this study was conducted with relatively small samples of children in each diagnostic group. Small samples reduce the precision with which all parameters are estimated, and caution should be exercised in generalizing the results beyond the members of the observed sample. As such, this study warrants replication with larger sample sizes, especially in the Down syndrome group. Because of the developmental nature of this study – where children were assessed at 12–15, 30 and 45 months – it was not possible to include identical measures at all time points. For example, the PLS was used at Time 3, but would not have been appropriate to use at Time 1. Thus, it is important to interpret the developmental data with a measure of caution, as the use of scores from different measures may have limited utility (e.g. the use of standard scores at Time 3 for IQ from Bayley and DAS). In
addition, many children at Time 1 and Time 2 showed standard scores at the floor for the Bayley, so it was necessary to use raw scores for these analyses (as has been done previously in studies of development in children with Down syndrome). Again, this limits the interpretability of the developmental findings in this study.

It is also important to note that the two diagnostic groups differed demographically from one another in important ways from the outset. Specifically, mothers of children with Down syndrome were approximately 3 years older compared with mothers of children in the mixed comparison group, and median. Family income was approximately $15 000 higher in the Down syndrome group. This pattern of differences is not uncommon in studies in which families of children with Down syndrome are compared with families of children with other developmental disabilities (Cahill & Glidden 1997). Yet, given that these demographic characteristics often are observed in families of children with Down syndrome, it would not be appropriate to compare them with an artificially constructed comparison group. Rather, these dimensions were included as potential predictors in all growth modelling analyses, and only one demographic characteristic – income – was a statistically significant predictor of the level of stress after taking into account Time 1 child characteristics. While higher income was associated, on average, with lower levels of stress, the stress differentials associated with a difference of thousands of dollars of income were quite minimal. Further, prediction equations for stress that included child characteristics at Time 2 or Time 3 were not improved by the inclusion of any parent variables.

Even given these issues, this study constitutes an important step toward better understanding the interplay between development in children with Down syndrome and family outcomes. As one of the first to explore the connection between changes in development and family stress, findings from this study suggest that the early emergence of the Down syndrome behavioural phenotype may play an important role in shaping early family experience. Future studies should continue to explore the role of the Down syndrome behavioural phenotype, including cognition, language, behaviour – and other features including personality and motivation – in shaping family experiences over time.

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