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Special Issue Article

The Future of Developmental Psychopathology: Honoring the Contributions of Dante Cicchetti

Cicchetti's organizational-developmental perspective of Down syndrome: Contributions to the emergence of developmental psychopathology and the study of persons with neurodevelopmental conditions

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Abstract

Dante Cicchetti's earliest work, his studies of social-emotional development in infants and children with Down syndrome, set the stage for the emergence of the larger field of developmental psychopathology. By applying basic developmental principles, methodologies, and questions to the study of persons with Down syndrome, Dante took on the challenge of searching for patterns in atypical development. In doing so, he extended traditional developmental theory and introduced a more "liberal" approach that both continues to guide developmentally based research with persons with neurodevelopmental conditions (NDCs), including Down syndrome. We highlight five themes from Dante's work: (1) appreciating the importance of developmental level; (2) prioritizing the organization of development; (3) examining whether developmental factors work similarly in those with known genetic conditions; (4) rethinking narratives about ways of being; and (5) examining the influence of multiple levels of the environment on the individual's functioning. We highlight ways that these essential lessons anticipated present-day research with persons with a variety of NDCs, including Down syndrome, other genetic syndromes associated with intellectual disability, and autism. We conclude with visions to the future for research with these populations as well as for the field of developmental psychopathology more generally.

Keywords: Down syndrome; developmental approach; intellectual disability; neurodevelopmental conditions

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In honoring and extending Dante Cicchetti's legacy in the field of developmental psychopathology, we focus here on his earliest line of research – the study of socio-emotional development in children with Down syndrome. This work began while Dante was an undergraduate with Felisicima Serafica at the University of Pittsburgh, and continued while he was a graduate student with Alan Sroufe at the University of Minnesota, and then as a faculty member at Harvard University. In their focus on children with Down syndrome, these studies differ considerably from much of Dante's subsequent work with children and youth at risk for deleterious outcomes due to environmental experience (e.g., maltreatment) or to the complex interplay of environmental and genetic factors (e.g., depression). However, Dante's work on Down syndrome was foundational in his establishing developmental psychopathology as a formal scholarly discipline.

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The historical context of cicchetti's research on Down syndrome

Dante's studies of persons with Down syndrome came at a time of considerable fractionation in the relevant scholarly fields. The study of persons with intellectual disability, including those with Down syndrome, was largely constrained to the disciplines of special education and psychiatry as developmental psychologists still primarily focused on typically developing persons. Against this backdrop, a developmental approach to intellectual disability was only beginning to emerge (e.g., Zigler, 1967, 1969), as was the notion of a field of developmental psychopathology (e.g., Achenbach, 1974), both of which Dante would go on to transform.

Based on world views from special education and psychiatry, studies of intellectual disability at the time were dominated by perspectives of deficit, pathology, and segregation. In the United States, the study of intellectual disability was largely dominated by

so-called defect or difference theorists, researchers who sought to identify one or more key deficits that could be cited as the source of the lowered IQ scores that define intellectual disability. These researchers typically studied persons with intellectual disability as a single entity, despite its many different causes or etiologies (for discussions and critiques, see Burack et al., 1988; Burack, 1990; Burack & Zigler, 1990; Dykens et al., 2000). Examining performance on various cognitive or behavioral tasks, they compared groups of persons with and without intellectual disability of the same chronological age; and the inevitable finding of worse performance by the participants with intellectual disability was then interpreted as reflecting a primary deficit (for presentations of several of these deficit theories, see Zigler & Balla, 1982).

The situation was somewhat different in the United Kingdom, where a few large-scale longitudinal studies on the development of children with Down syndrome were led by scholars from a range of backgrounds, including Janet Carr, a clinical psychologist, Cliff Cunningham, a former teacher, and Ann Gath, a child psychiatrist. In the first and longest-standing example, Carr studied 54 infants born between December 1963 and November 1964 within three boroughs of London and the county of Surrey. Capitalizing on medical advances that lengthened the life span of persons with Down syndrome well into their adult years, Carr extended her initial plan to study the developmental trajectories of these infants only during their first 4 years of life. Ultimately, she re-examined these individuals at 11 years, 21 years, and then more regularly until they were 50 years old. Publishing primarily in journals on intellectual disability, Carr (1970a, 1970b; Carr & Collins, 2014, 2018; Murphy & Carr, 2020) provided extraordinary amounts of nuanced information about the biological, cognitive, and social development of her participants; their relationships with family members and others; their involvement in various aspects of society; and the effects of aging and dementia. In a research program about the early development of approximately 160 children with Down syndrome from a university research center in Manchester, Cunningham and colleagues (e.g., Berger & Cunningham, 1981, 1983; Glenn et al., 1981) published findings on early vocal behaviors and interactions, social smiling, and eye contact between mothers and infants. These studies were among the first on Down syndrome to appear in mainstream developmental journals such as *Developmental Psychology* and *Child Development*. Based on a cohort of children with Down syndrome in Oxford, Gath and Gumley (1984, 1986a, 1986b, 1987) published a series of papers that appeared primarily in psychiatry journals on the impact of having a child with Down syndrome on families, with a focus on the development of siblings.

Although also providing considerable knowledge about infants and children with Down syndrome, Dante's studies were more experimental and were based in big-picture questions about universal developmental theory. For this reason, Dante's work most converged with that of Ed Zigler (e.g., 1967, 1969), who had a few years earlier proposed a developmental approach to persons with intellectual disability that he contrasted with the various defect theories (for recent reviews, see Burack et al., 2021a, 2021b; Hodapp, 2021). In an early attempt to base the study of intellectual disability in traditional developmental theory, Zigler emphasized that persons with intellectual disability, regardless of intelligence level, traverse developmental milestones in the same order (i.e., the "similar sequence hypothesis"), albeit at slower rates and with lower asymptotes (for discussions, see Hodapp, 1990; Hodapp & Zigler, 1990; Weisz & Zigler, 1979; Zigler & Hodapp, 1986; Weisz et al., 1982). He also hypothesized that, when matched on developmental level (mental age rather than chronological age), groups with and without intellectual disability should function at the same cognitive or linguistic levels (the "similar structure" hypothesis; Weiss et al., 1986; Weisz & Yeates, 1981). Also concerned with the "whole child" and the consequences of the frequent failures that this group experiences, Zigler (1970a) argued that personality and motivational factors needed to be considered in understanding both task and real-world performance of persons with intellectual disability.

Zigler's developmental approach represented an early attempt to base the study of intellectual disability in traditional developmental theory but was limited in its sole focus on the subgroup of children with so-called familial, cultural-familial, or sociocultural-familial intellectual disability (Burack & Zigler, 1990). This group is comprised of children for whom no clear organic cause could be identified for their delays in development. Following Gottesman (1963), who noted that IQ is partially determined by the operation of many inherited genes working together (i.e., "polygenic inheritance"), Zigler (1970b) emphasized that these children were simply those who fell along the very lowest part of the normal distribution of IQ; their intellectual disability was not due to any specific organic etiology per se. The remaining children with intellectual disability were described by Zigler (1967) as having "organic" intellectual disability. This group was comprised of children with Down syndrome or with any of the other approximately 1,100 genetic disorders (Batshaw et al., 2019), associated with intellectual disability. Taking a conservative approach to development, Zigler (1967) asserted, "The general developmental approach is applicable only to the familial [child with intellectual disabilities], and this approach does not speak to the issue of differences discovered between normal children and organic [children with intellectual disability]" (p. 269).

In contrast, in a series of articles and book chapters, Dante emphasized that developmental principles should extend even to the organic group, with his focus on children with Down syndrome. In this regard, Dante's application of developmental theory to the study of children with Down syndrome also contrasted with the Zeitgeist of developmental theorists at the time. Traditional developmental theory, such as articulated by Piaget, had historically been framed almost solely within neurotypical Western frameworks, with only minimal reference to populations with potentially divergent developmental pathways, including those with intellectual disability or from non-Western backgrounds. By the 1970s, however, developmental theory was increasingly being deconstructed by those who emphasized its limitations in accounting for deviations from typical patterns, such as those associated with individual and cultural differences (for a discussion, see Bronfenbrenner et al., 1986). These researchers questioned whether mainstream developmental theory should be maintained in light of the vast differences across persons that are seen in the real world across persons (Kessen, 1984).

Given Zigler's (1967) more conservative developmental approach to intellectual disability and the de-construction of the very idea of development, Dante's commitment to asking big developmental questions about persons with Down syndrome represented a bold theoretical stance. By applying basic developmental principles, methodologies, and questions to the study of persons with Down syndrome, Dante took on the challenge of searching for developmental patterns in atypical development, thereby extending, rather than rejecting, traditional developmental theory.

Dante's scholarship on children with Down syndrome

As early as the mid-1970's, almost a decade before his special issue inauguration of the field of developmental psychopathology

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(Cicchetti, 1984), Dante infused developmental tenets and principles in his research with children with Down syndrome. Fostering the premise that human development follows a universally organized and integrated process, Dante and colleagues highlighted the coherence of development (Motti et al., 1983), that is, the process rather than the pace of development (Cicchetti & Sroufe, 1978). In Dante's early work on the level and quality of play in children with Down Syndrome, individual differences in play were strongly predicted by indexes of affective expressiveness and Bayley Developmental Quotient scores. Moreover, when matched by mental age, children with Down syndrome had the same level of play as typically developing children (Motti et al., 1983).

The issue of developmental level and appropriate comparison strategies was also central to Cicchetti and Sroufe's (1976) consideration of tensions between developmental level (as determined by mental age) and chronological age in the study of the development of laughter among infants with Down Syndrome. They noted that, although potentially delayed, the development of these children proceeds in a similar sequence to non-intellectually disabled infants. In essence, cognitive ability, not chronological age, drove the development of laughter in 4–8-month-old infants with Down syndrome, with development occurring in the same order across 30 developmentally ordered items.

The complexity of applying the developmental approach was further evident in Butterworth and Cicchetti's (1978) findings that attachment behaviors were largely similar to those of typically developing children in terms of sequences and behaviors, although with a few exceptions. The ways in which these exceptions were interpreted by Butterworth and Cicchetti also suggests a deep respect for differences in the experiences of those with Down syndrome (e.g., different interpretation of what it means to be alone); a deep-seated desire to integrate across levels of measurement that included genes, neurotransmitters, cognition, and behavior; and a continued focus on the general universality of developmental sequences and structures. Butterworth and Cicchetti highlighted the reciprocal relationship between typical and atypical development by focusing on how the study of the visual calibration of posture over early development among those with Down syndrome can inform our understanding of the development of these same behaviors in typically developing children.

Dante identified similar patterns of developmental coherence across domains of functioning. For example, Beeghly and Cicchetti (1997) found both that infants with Down Syndrome and typically developing infants followed the same general sequence of internal state lexicon language skills and a strong association among age, general cognitive functioning, and expressive language ability. In a study of self-recognition, Mans et al. (1978) found that children with Down syndrome and typically developing children showed self-recognition at the same time when matched on cognitive age rather than chronological age. They argued that "In Down Syndrome, the emergence of self-recognition appears to be an orderly and meaningful process. Lower intelligence, per se, does not imply a different developmental sequence." (p. 1249). Although Thompson et al. (1985) found differences in the quality of separation distress among children with Down syndrome versus children without Down syndrome in the strange situation attachment task, they also identified a "consistent organization underlying this aspect of socioemotional responsiveness" for both groups (p.828).

These early observations are all grounded in traditional developmental frameworks (e.g., Piaget, 1970), including those of the study of intellectual disability (Hodapp et al., 1990), and an example of how evidence from an atypical population tells us about the necessity or inevitability of developmental processes. Despite the considerable evidence of universality, Serafica and Cicchetti (1976) warned against studying individuals with intellectual disability as a single group, highlighting that clear patterns of development can only be found by studying etiologically homogeneous groupings such as persons with Down syndrome. Within the context of this type of homogeneous population, Dante and colleagues highlighted the importance of contextualizing development to incorporate the diverse processes and environments that the child is experiencing.

The originality of the contribution of Dante's broadening of the developmental approach is perhaps best illustrated in Cicchetti and Pogge-Hesse's (1982) chapter in Zigler and Balla's (1982) volume, *Mental retardation: The developmental-difference controversy*. In that chapter, the only one on development in persons with an organic form of intellectual disability (i.e., not familial intellectual disability), Cicchetti and Pogge-Hesse invoked a more "liberal," expansive view of development (for a later iteration, see Cicchetti & Ganiban, 1990). Premised on Werner's (1948, 1957) orthogenetic principle, they envisioned a developmental approach that guided research about Down syndrome as well as other conditions associated with intellectual disability. Cicchetti and Pogge-Hesse explicitly extended beyond discussions of the "typical" or "usual" sequences and structures of development that had been applied to persons with familial intellectual disability (Weisz et al., 1982; Zigler & Balla, 1982). Instead, in their critiques of defect/difference theorists, Cicchetti and Pogge-Hesse's (1982) took on more complex developmental questions, such as those about the organization of development and mutual influences between typical and atypical development.

Using persons with Down syndrome as a proxy for all etiological-specific groups of persons with organic forms of intellectual disability (e.g., persons with Williams syndrome or fragile syndrome), Cicchetti and Pogge-Hesse (1982) were setting the stage for a field of developmental psychopathology that Dante would soon articulate. As Dante would later note (Cicchetti, 1984), the basic principles of developmental psychopathology specify that: (1) scholarship about persons with pathology and differences needed to be fused with and informed by developmental theory; and relatedly (2) that typical and atypical development were mutually informative.

Based on these developmental principles and Dante's early studies of the development of infants with Down syndrome, Cicchetti and Pogge-Hesse (1982) argued that the defect approach, and de facto any similar pathologizing approach, "overlooks the possibility that the behavior and development of retarded children is organized, adaptive, and integrated just as is the case for nonretarded children and infants. We know that they are retarded; the important and challenging research questions concern the developmental process" (Cicchetti & Pogge-Hesse, 1982, p. 279). They further noted that "Organically retarded persons are not only 'different' from nonretarded persons . . . They are organized in their own right . . . Thus, a contribution will be made to a general and integrated structural-organismic theory of development that tries to account for development in *all* human beings across *all* behavioral domains" (Cicchetti & Pogge-Hesse, 1982, p. 313). In doing so, Dante highlighted the extent to which the understanding of persons with Down syndrome, like those of any population, is inextricably linked to and informative about the study of the general population. Thus "children with Down syndrome are, indeed, a particularly interesting population to study from a

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developmental view" (Cicchetti & Pogge-Hesse, 1982, p. 293) as they (1) provide rare insight into the sequelae of an extra autosomal chromosome; and (2) allow for a better understanding of true convergences and discontinuities due to the slower cognitive development. These quotes are all precursors to the essential point, or dictum, of Dante's introductory article (Cicchetti, 1984) to the special issue of *Child Development* (Cicchetti, 1984) introducing the field of developmental psychopathology, that, "you can learn more about the normal functioning of an organism by studying its pathology, and, likewise, more about its pathology by studying its normal condition" (Cicchetti, 1984, p. 1).

The relevance of this dictum was evident in the first two publications on intellectual disability that appeared in this journal, the flagship publication of the field of developmental psychopathology. In the first paper, Hodapp and Burack (1990) addressed half of the dictum, "What mental retardation teaches us about typical development: The examples of sequences, rates, and cross-domain relations." They portrayed intellectual disability research as an "experiment of nature" that can be used to both replicate findings from the study of typical development and provide evidence that extends developmental theory. In the second paper, comparing developmental patterns of language, affect, and visuo-spatial skills between children with Down syndrome and those with William Syndrome, Reilly et al. (1990) extended Dante's dictum by comparing two "atypical" populations. That is, just as typical and atypical development can be mutually informative, so too can the development of two different atypical populations.

Over the past few decades, both comparative frameworks have been central to cutting-edge research in persons with intellectual disability. For example, Landau and colleagues have utilized findings from persons with Williams syndrome to provide compelling cases for a better general understanding of various aspects of development including the organization of spatial representation (Landau, 2012), spatial language (Landau & Zukowski, 2003) and spatial cognition (Landau & Hoffman, 2005); and the disassociations between intuitive physics and psychology (Kamps et al., 2017) and of vision for perception and vision for action (Dilks et al., 2008). Similarly, Mervis (2012) highlighted the extent to which evidence from children with Williams syndrome provides a strong case for the interdependence of language and cognition throughout development. Cross-etiology comparisons have grown to be essential to the study of the development across many areas of functioning, including attention (Cornish & Wilding, 2010), language (Hofmann & Müller, 2021), visual processing (Ly & Hodapp, 2005), and maladaptive behavior (Dykens & Kasari, 1997).

Dante's questions as precursors of scholarship about persons with neurodevelopmental conditions

Just as Dante's early work on Down syndrome provided a guiding framework for the emergence and growth of developmental psychopathology, it largely anticipated contemporary research among persons with NDCs including Down syndrome, other genetic syndromes, and autism. In discussing advances from our own and others' research, we again invoke Dante's seminal conceptualizations, and essential quotes, from his work with children with Down syndrome.

Conceptualization 1: appreciate the importance of developmental level

"... the important and challenging research questions concern the developmental process.."

For the study of group comparisons, Dante extended the idea of mental age matching, or considering developmental level, in research to include individuals whose intellectual disability is due to an organic etiology. This lesson remains critical to understanding unique facets and universals in development and has clinical implications as well. One example involves children with fetal alcohol effects (FAEs) who are often described as inattentive – more than 90% of children with FAE are diagnosed with ADHD and many of those children are placed on medications.

To assess attentional difficulties and contrast clinical measures of attention, Lane et al. (2014) compared children with FAE to a group of children whose development was typical and did not include exposure to alcohol. Although the group of children with FAE had cognitive abilities in the average range, they were in the low average range and as a result, Lane et al. (2014) compared the groups on the basis of developmental level rather than chronological age to account for these IQ differences. On the Conners (1997), a parent report of inattention, impulsivity and hyperactivity, the children with FAE were rated as being in the clinically significant range. In a clinical setting, this would be a strong indicator that a diagnosis of ADHD combined presentation might be appropriate (if this occurred across contexts, was reported by one or more people, and impaired the child's functioning). In contrast, on the Test of Everyday Attention for Children (TEA-Ch) (Manly et al., 1998), the children with FAE performed in a manner that was largely similar to the typically developing children when the groups were compared on the basis of developmental level or mental age. Together, these findings highlight the critical importance of the metric by which comparisons are made for those with lower cognitive abilities and the relatively high potential for empirical findings to have real clinical impacts. In one scenario, when attention (in this case parent reported behaviors indicative of attention problems) was constructed relative to expectations based on chronological age, children with FAE would likely be diagnosed with a chronic psychiatric condition, for which medications are the front-line treatment. Conversely, in another, when attention abilities (in this case measured on the basis of a standardized test) were understood relative to children's developmental level, they would be expected to score within the average range. The metric of comparison, chronological or developmental age, is critical and requires care and consideration in both the choice of matching measure and the interpretation of research findings (Burack et al., 2004; Russo et al., 2021).

Despite the self-evident significance of appropriate developmental matching, neuroscience researchers of intellectual disability have not embraced this approach as extensively as could be hoped (Burack et al., 2016a). This field is often compromised by studies in which comparisons between individuals with and without intellectual disability that are based on chronological, rather than mental age, lead to spurious interpretations of group differences. As lower IQ is pathognomonic to genetic syndromes associated with intellectual disability, chronological age-based comparisons tell us nothing about the specific syndromes being examined (Burack et al., 2023a). Rather, they tell us only that those with an intellectual disability are performing less well than would be expected for their age, the very definition of intellectual disability.

Conceptualization 2: prioritize the organization of development, even among children with different genetic conditions

"Organically retarded persons are not only 'different' from nonretarded persons . . . They are organized in their own right . . . "

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Dante's studies on the development of infants and infants with Down syndrome within an organizational perspective were paradigms for the extensive study of "behavioral phenotypes" of different conditions associated with intellectual disability (e.g., Cornish & Wilding, 2010; Dykens et al., 2000). In this framework, we document ways in which specific etiological groups show their own particular, etiology-related cognitive-linguistic-adaptive organization or patterns of strengths or weaknesses across domain. Throughout this work, caveats have been provided that (a) not every child with a particular syndrome would show that syndrome's "characteristic" strengths and weaknesses; and (b) not every etiological group would differ from every other group on all behaviors (Hodapp, 2021).

As etiology-specific research progressed, researchers began to pay greater attention to development "into" the fully formed behavioral phenotypes. Across multiple studies by different research groups, etiology-specific performance of persons with Down syndrome was evident, including in executive functions (Hodapp & Fidler, 2021), which relate to the prefrontal cortex, hippocampus, and cerebellum (Edgin, 2013; Nadel, 2003), and for which lower-than-MA performance could be seen on neuropsychological tasks that involve working memory, planning, shifting, and inhibitory control. In addition to closer ties to their neuropsychological functioning, researchers are now exploring how infants and toddlers with Down syndrome progress in executive functions over the infancy period. For example, Schworer et al. (2022) compared infants with Down syndrome to typically developing, MA-matched infants (mean age = 8.62 months), on a set of executive function "precursor tasks" (i.e., attending to an object, shifting attention, and planning to perform an object like reaching). As expected by the behavioral profile, the infants with Down syndrome performed worse than the MA-matches on these precursor tasks. When a subset of the infants with Down syndrome were then examined 6 months later on an actual executive function task (the "A not B" object permanence test), performance levels on precursor tests at Time 1 were related to actual executive function tasks six months later at Time 2.

These findings have both conceptual and clinical implications. Theoretically, they illustrate the construct of "developmental cascades," another developmental phenomenon discussed by Dante. As described by Masten and Cicchetti (2010), "Developmental cascades refer to the cumulative consequences for development of the many interactions and transactions occurring in developing systems that result in spreading effects across levels, among domains at the same level, and across different systems or generations" (Masten & Cicchetti, 2010, p. 491). In this preliminary work, cascades are evident among infants with Down syndrome, with the "spreading effects" evident across time (from T1 to T2) and from precursor to actual executive function tasks (Malachowski & Needham, 2023).

These findings also lead to intervention implications. In what they refer to as a "syndrome-informed micro-intervention," Fidler et al. (2021) launched a brief caregiver-facilitated intervention concerning goal-directed infant reaching, among the earliest examples of executive functions. Infants with Down syndrome were randomly assigned to intervention and control groups, with mothers in each group interacting with their infants around infant toys for 5–10 minutes each day. Each toy was presented to the infant at eye level and shaken, tapped, or moved to make the toy optimally salient. Infants would then reach out toward the desired toy. The only difference was that the intervention infants wore "Sticky Mittens," or Velcro mittens that latched on to the infant toys making the toy easier for the infant to grasp. At posttreatment in which no infants had the use of the Sticky Mittens, the infants in the treatment group showed more goal-directed reaches to the desired object and contacted the object more quickly (shorter mean latency), with large effect sizes. Fidler et al., also showed that the optimal window for the intervention involved infants with Down syndrome who were from 5 to 10 months of age; younger and older infants showed less improvement. Although preliminary, etiology-based interventions seem both feasible and effective.

Conceptualization 3: examine whether developmental factors work similarly in those with known genetic conditions

"...the behavior and development of retarded children is organized, adaptive, and integrated—just as is the case for nonretarded children and infants..." (Cicchetti & Pogge-Hesse, 1982)

In the developmental literature on neurotypical persons, the development of IQ is thought to be linked to polygenic inheritance. In this framework, parents who are tall typically have tall children but can have one or more short children, just as two parents with above-average intelligence can have a child of average or even below average intelligence. The relative deficit in height or in intelligence can occur not because of any specific genetic mutations, but because both height and cognitive ability are complex, largely heritable, traits stemming from polygenic effects. In most cases, however, the height or intelligence of parents is correlated with height-intelligence of their biological offspring.

Although the IQs of persons with the familial form of intellectual disability are also thought to conform to the same processes of inheritance (e.g., Zigler, 1967, 1969), known genetic causes of intellectual disability, such as Down syndrome, are often viewed as immutable conditions that are impervious to social and familial influences. They are commonly thought to be defined solely by the genetics of the condition and devoid of genetic influences that are not associated with the syndrome. However, we now know that this is not the case. This was exemplified in research with persons with Turner syndrome (X,0), a whole chromosomal aneuploidy on the sex chromosomes, resulting in a deletion of a second X (or Y) chromosome. Although Turner Syndrome shows wide phenotypic variability, one of the common physical phenotypes is short stature. However, 50 years ago, Brook et al. (1974) observed that the adult height of females with Turner syndrome, while some 3 standard deviations below the mean height of both parents, highly correlated to bi-parental mean height ($r = .84$). Several other researchers have reported similar findings. In each case, despite a shift from bi-parental mean height of between 2 and 4 standard deviations, the parent–offspring correlations are preserved, and remain similar to what one would expect to find in the general population (r's ranging from .42 to .84; see Lemli & Smith, 1963; Brook et al., 1974; 1977; Holl et al., 1994; Massa & Vanderschueren-Lodeweyckx, 1991; Rochiccioli et al., 1994). This is also the case for Prader–Willi syndrome (15q 11–13) – although offspring are between 1 and 2 standard deviations shorter than their parents, the parent–offspring correlation remains what one would expect under typical circumstances ($r = \sim .50$). Persons with Klinefelter syndrome (47, XXY) are often above average in height owing to this aneuploidy, but here again, while they are some 2 standard deviations taller than the mean height of their parents, the parent–offspring correlation is preserved ($r = .64$; Brook et al., 1977).

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These types of connections between parental and offspring characteristics occur as well in other traits, such as cognitive or social ability. One example involves 16p11.2 deletion, a microdeletion syndrome comprising some 26 genes, resulting in wide phenotypic variability (Stefansson et al., 2014; Moreno De Luca et al., 2014). In this case, some 25% of offspring with the deletion are diagnosed with autism spectrum disorders (ASDs) and approximately 25% with intellectual disability (Hanson et al., 2014; Moreno De Luca et al., 2014). However, although most persons with this deletion do not reach clinical thresholds for ASD or intellectual disability, within-family correlations are preserved. Thus, when compared to first degree relatives (parents and non-carrier siblings), the persons with the16p11.2 deletion demonstrated a shift of −1.7 standard deviations in full-scale IQ, even as the intraclass correlation was significant ($r = .42$). In terms of social development as well, scores on the Social Responsiveness Scale (a dimensional scale of autistic traits) showed connections among the persons with the deletion, parents, and the non-carrier sibling. Relative to bi-parental mean and sibling scores, the persons with the deletion were shifted more than 2 standard deviations (they were more symptomatic), even as the intraclass correlation was again significant ($r = .52$). This shift-but-spared pattern of correlations was also observed for fine motor ability (Moreno-De-Luca et al., 2015).

The connections to familial genetics can be further extended to real-world adaptation, Evans and Uljarević (2018) examined whether the IQs of persons with DS were correlated with parental education level. Parental education is a particularly informative variable as it reflects both some margin of cognitive ability and the broader sociodemographic and cultural-familial milieu. Evans and Uljarević used a combined cross-sectional (two age groups: 4–12 years; 12–21 years) and longitudinal (over a 2-year period) design. As predicted given previous findings, bi-parental education levels were correlated with the IQs of the offspring with Down syndrome. The findings indicated that these associations were particularly strong for verbal IQ in the older age group and at Time 2; the weakest in the younger cohort at Time 1. However, overall, the patterns were fairly consistent for full-scale, verbal and non-verbal IQ, with median r values of approximately .50.

From these studies, and despite the clear impact of various genetic anomalies on various domains of functioning, persons with Down syndrome are still impacted by a variety of familial factors. We cannot argue definitively whether these factors involve polygenic effects, or whether some impact of environment mediates the developmental outcomes of children with such conditions. What is clear, however, is that similar factors are likely at play in determining outcomes of children with NDCs as with neurotypical children. The story is complicated, as the mutability of functional domains may vary depending on the degree of heritability of a given trait; some heritable traits may be more genetically complex, and some may be more influenced by environmental factors. Still, despite the presence of a known genetic condition, the development of children with and without NDCs are similarly impacted by multiple, and likely interacting, genetic and environmental factors.

Conceptualization 4: rethink narratives about ways of being

"we should study not "does × have y" but rather "in what way does × use y "and does a particular mental structure interact with other mental structures in the same way in one group as in another?" (Cicchetti and Pogge-Hesse, 1982).

Dante's invoking of Werner's (1957) orthogenetic principle that development is adaptive, organized, hierarchical, and universal implies that an organism, and the behaviors it generates at any given time point, reflect the cumulative endpoint of an organized, systematic developmental process. This type of perspective is the essence of the opposition of developmental theorists to deficit models of intellectual disability and is central to the rethinking of narratives about autistic persons who historically have been studied with the emphasis on what they cannot do or accomplish (for a discussion, please see Bottema-Beutel et al., 2021). Over the past four decades, prominent deficit models include those related to theory of mind (Baron-Cohen et al., 1985), weak central coherence (Frith & Happé, 1994), and various aspects of attention (Burack, 1994; Klin et al., 2002). However, these narratives have been revised considerably in relation to: (1) evidence that autistic persons process, and therefore, act on the world in different ways than do neurotypical persons; and (2) the advent of the neurodiversity movement emanating from the voices of both the autistic and research communities (for a review, see Pellicano & den Houting, 2022). Although the term neurodiversity was coined by Judy Singer in 1990 to underscore the value of different minds, many precursors of the notion can be seen in Dante's writings. The titles of his (more than 700) published papers do not directly mention the term, however, the fundamental tenets of developmental psychopathology are consistent with a deep respect for, and valuing of, divergence.

Within the scholarly literature, the current narratives about autistic persons are increasingly focused on their unique abilities, cognitive styles, perceptual biases, and motivation in interacting with the people and world around them (Burack et al., 2016; Happé and Frith, 2020). This type of narrative has been presented several times over the past few decades despite the more common lay, clinical, medical, and scholarly emphases on pathology. For example, Schultz and colleagues (Grelotti et al., 2002; Schultz et al., 2000) argued that the fusiform gyrus and related brain areas are used effectively by autistic people, but in different ways and to process different objects than might be seen in the neurotypical population. In developing their enhanced perceptual functioning model, Mottron and Burack (2001; Mottron et al., 2006) highlighted that rather than being a deficit, the unique way that autistic people process information in the world may lead to certain advantages on specific types of tasks. This type of evidence even led to the reformulating of primary deficit theories, most notably the weak central coherence theory of autism (Happé & Frith, 2006).

In borrowing from Cicchetti and Pogge-Hesse (1982), Burack et al. (2016) sought to recast the literature on attention in autism within the framework of "how, rather than on how well" autistic people attend within their environments. They reviewed several examples in which the performance of autistic persons on tasks of attention and perception could be, and in some cases were, interpreted as evidence of deficit may be reframed as an alternative, and often more adaptive or efficient, style of processing and way of being. In this reshaped narrative, the consideration of context and development are essential in understanding how biases, styles, and ways of being of autistic people affect their attention in the real world. Burack et al. highlighted that cases of competition among attentional demands, preferences, biases, and perspectives might lead autistic persons to filter, orient, or selectively attend to some features over others. Although what is being attended to in any particular context may appear atypical to the outside observer, one should

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not infer that any particular attentional skill is impaired or better or worse than for any other person. In one extension of that approach, Brodeur et al. (2018) and Burack and Brodeur (2020) argued that the attentional style of autistic persons is particularly utilitarian, a way of being that can lead to greater efficiency and in turn any number of benefits, including those related to education or employment.

In trying to disentangle the neural mechanisms that may underlie some of these unique ways in how autistic people attend, Russo et al. (2012) and Kaplan-Kahn et al. (2021) leveraged the exquisite temporal resolution of Event-Related Potentials or ERPs. ERPs reflect the activity of synchronized postsynaptic potentials that can be measured at the scalp via electrodes in response to a particular event. One of the most commonly measured ERP components is the N400 effect, which is a negative deflection that occurs around 400ms after a stimulus that is incongruent with its semantic context. The most classic example is provided by Kutas and Hillyard (1980), the first to document this effect, who presented participants with sentences that either did (e.g., he spread the bread with butter) or did not (e.g., he spread the bread with socks) end as would be expected. Compared to congruent sentences, incongruent sentences yielded this negativity around 400ms.

The N400 effect, which was not found among autistic persons in several studies (for a review, see Russo et al., 2012), was often cited in relation to the rather delayed language development commonly seen among autistic persons. However, Russo et al. (2012) noted that the behavioral performance of autistic youth on N400 tasks (when it was measured) did not differ from that of IQ and/or age matched youth, leaving open the question of how autistic people actually solved this semantic problem. To answer this question, they developed a simple task where they asked participants to press one button if the sound and image of an animal (e.g., a barking dog) matched and a different button if the sound and picture did not match (e.g., a barking cat). Rather than looking for the presence or absence of an N400, Russo et al. (2012) examined modulations comparing congruent and incongruent trial types in early, perceptual timeframes, based on the premise (e.g., Mottron & Burack, 2001) that autism is associated with an increased reliance on perceptual processes. They replicated the findings of others as (a) the behavioral performance of the autistic and the nonautistic participants was the same and (b) the nonautistic, but not the IQ and age matched autistic, group demonstrated an N400. However, they also found that autistic youth differentiated congruent and incongruent trials by 100ms post stimulus, a full 300ms before their nonautistic peers. These findings are consistent with the emphasis that it's not "how well" but rather *how one uses* what they have to process the world that matters and is scientifically informative.

In addition to highlighting the unique ways underlying how autistic individuals process their environments, Kaplan-Kahn et al. (2021) assessed whether perception interacted with semantic processes in nonautistic individuals. They asked college students without autism to complete the same animal congruence task described above while they measured ERPs. At the end of the task, the participants completed the Autism Quotient, a self-report questionnaire of autistic traits. The brain's differentiation between barking dogs and barking cats at 100ms mediated the relationship between attention to detail as measured by the AQ and the N400 effect suggesting a similar interaction between cognitive/neural processes in autistic and nonautistic persons.

Conceptualization 5: examine the influence of multiple levels of the environment on the individual's functioning

"*a contribution will be made to a general and integrated structural-organismic theory of development that tries to account for development in all human beings across all behavioral domains*" (Cicchetti & Pogge-Hesse, 1982, p. 313).

In his early work, Dante highlighted the central role of relationships, particularly parent–child relationships, in shaping social and emotional development. In particular, caregiver attachment was considered central to the development of social competence in children. However, Dante emphasized the need to consider social competence, the ability to effectively navigate social situations, build positive relationships, and engage in age normative social behaviors (Dodge, 1985; Rose-Krasnor, 1997), within diverse social environments. This includes an understanding of how cultural contexts influence socialization practices and shape social interactions in the moment as well as over the span of a child's development. In this framework, the assessment of social competence involves examining multiple dimensions and identifying key skills such as empathy, cooperation, conflict resolution, and the ability to establish and maintain connections with others.

Social competence is context-dependent, as individuals demonstrate different levels of competence across different situations. They must skillfully coordinate multiple processes (e.g., cognition, emotion) and integrate contextual factors (e.g., social norms, setting) to adequately meet the social demands of particular situations (e.g., peer-to-peer or parent–child interaction) (Crnic, 1990; Iarocci et al., 2007, 2008). However, how well an individual navigates different social contexts and the specific relational challenges they may encounter is also dependent on their social partner. An understanding of the relational aspects of social interactions, particularly within the context of culturally diverse social partners, can help us extend Dante's valuable work on identifying both risk and protective factors and the potential for resilience in the social development of children with NDCs.

Autism may offer a critical window into the dynamic and relational nature of social interactions and adaptation. In one example of how cultural or, in the case of autism, subcultural differences may influence social interactions is the 'double empathy problem' introduced by the autistic researcher, Damian Milton (2012). Milton highlights a mutual struggle to understand and connect between autistic and nonautistic individuals. That is, difficulties in communication and understanding are reciprocal, as both autistic and nonautistic individuals may experience challenges in comprehending each other's communication styles and social cues. The concept draws parallels with the idea of cultural differences in communication. Just as individuals from different cultures may experience challenges in understanding each other's communication styles, autistic individuals, who may be considered a subculture in mainstream society, may face similar challenges due to differences in their cognitive and perceptual styles.

Recognizing the relational nature of social relationships (and the potential challenges) between autistic persons and their social partners fosters a better understanding and inclusivity. It suggests that efforts to improve communication should be bidirectional, with an emphasis on mutual accommodation and appreciation for diverse communication styles. Thus, social competence is closely tied to social awareness and sensitivity, as an individual's awareness of social cues and their ability to adapt to the emotions and needs of others are critical to effective and inclusive social

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behavior. The social acceptance and inclusion of people with NDCs rely on a thorough understanding of how both neurodivergent and neurotypical individuals behave in social relationships. For example, autistic individuals' social abilities may not be the only factors influencing their social success or lack thereof. Rather, the social perceptions and behaviors of nonautistic persons contribute to autistic individuals' social difficulties.

Scheerer et al. (2022) attempted to improve nonautistic teenagers' views of autistic people by teaching them about autism in a way to which they can more easily relate. They then assessed both whether the quality and quantity of contact with autistic people predicted these perceptions and whether their views of autistic people depend on how they view their own social skills. Scheerer et al., found that nonautistic high school students showed negative attitudes toward autistic people, but that these views could be improved by the informative presentation about autism. They also found that the youth who reported higher quality interactions with autistic people had less negatively biased attitudes toward them, but those who self-reported higher social competence rated autistic people the most negatively. One possible reason is that they are the most likely to value 'normative' social skills and be more judgmental about those who socialize in a different way. These findings helps us better understand how the social perceptions of neurotypical youth toward autistic youth may contribute to their social challenges.

Knowledge about the attitudes and perceptions of neurotypical individuals toward neurodivergent individuals can contribute to efforts aimed at reducing stigma and discrimination. This was highlighted by the finding that the social perceptions in the neurotypical youth were malleable with a deeper understanding of autistic people. By addressing misconceptions and fostering empathy, we can create social attitudes and behaviors that are mutually accepting and supportive. To date we have focused almost exclusively on neurodivergent people and their social "disabilities" without much consideration of neurotypical influences on neurodivergent people's social behavior/outcomes. The research on the social (dis)abilities of neurodivergent people would benefit from Dante's idea of understanding typical and atypical development within a wholistic framework. This approach views individuals of all neurotypes as valuable in their own right, contributing to the social diversity essential for human resilience.

Dante's influence on the future of developmental psychopathology and the study of persons with neurodevelopmental conditions

Begun while he was an undergraduate and emphasized during the early part of his career, Dante Cicchetti's contributions to developmental approaches to the study of Down syndrome and intellectual disability more generally is almost immeasurable (for a brief discussion, see Burack et al., 2023b). Dante infused the field with a rigorous scientific methodology encapsulated in theoretical sophistication (for a discussion, see Wagner et al., 1990). Dante allowed us to ask questions that we had never before considered and integrate ideas that we had never before connected as he synthesized the study of persons with Down syndrome across disparate disciplines and ways of thinking. Even as a graduate student, Dante's vision and call for action of a developmental framework for understanding of children with Down syndrome, and de facto any NDC, was evident in the title of his dissertation, *Affective development in Down's syndrome infants: An organizational perspective* (Cicchetti & Sroufe, 1976). This thread would continue through all his work in the field. And, although his scholarship in this area largely culminated in the publication of the seminal Cicchetti and Beeghly (1990b) edited volume, "Down syndrome: A developmental perspective," his primary ideas remained: (1) research needs to be etiology specific in order to best assess development; (2) development needs to be studied across domains simultaneously to reflect its complexity and organization in real-world context; (3) developmental matching needs to be fine-tuned to account for unique patterns of development; (4) developmental research needs to be guided by questions that tap into group and individual ways of being, styles, and biases; and (5) developmental changes need to be considered in relation to the influences of multiple levels of the environment. As Dante has taught, these essential developmental lessons will allow us to appreciate how *all* children develop within their environments, empowering us to unravel the transactional nature of development – the presence of changes or lack thereof, and the influences of diverse processes within and outside of the developing child (Cicchetti & Beeghly, 1990a).

Such advances also foreshadow perspectives that are becoming increasingly prominent as time goes on. Dante's ideas about the reciprocal value of typical and atypical development align well with more humane outlooks that underlie the current neurodiversity perspective. Proponents highlight that all people experience and interact with the world around them in multiple ways; there is no one "correct" way of thinking, learning, or behaving, and differences are not viewed as deficits. Social acceptance and inclusion of people with developmental disabilities rely on a thorough understanding of how both neurodivergent and neurotypical individuals behave in social relationships.

Even as the studies described in this paper highlight Dante's influence, his impact will resonate for decades to come, and his contributions remind us of the rich landscape of questions still to be asked. For example, we still have much to learn about the development of individuals with any one or more of the 1,100 genetic conditions associated with intellectual disability, and even more to learn about how this development impacts and is impacted by the connections and interactions of various "levels" of genes-brain-behavior (Dykens, 2021). We also need to remind ourselves and advocate to others about the fundamental impact of methodological choices such as how and to whom we make comparisons between those with and without NDCs. When framed within a developmental context, rigorous research can ultimately be translated into meaningful interventions and policy initiatives that allow us to make substantive impact in the lives of persons with NDCs and their families (Abbeduto et al., 2023; Spiker, 1990).

As we conclude we note that we have only begun to elaborate on the many ideas that Dante has provided us. In the years to come, the depth of Dante's lessons will continue to guide the field, to impact research, education, psychology, psychiatry, and policy. Dante's legacy will allow us, his contemporaries, and those that follow, to ask the big questions and to center his larger view of development, in both how we frame questions and in how we interpret our findings and share them with the broader community. The ultimate goal, of course, is to provide the most essential information to inform our collaborations with persons with NDCs and their family members as we work together to promote their well-being and successes as well the betterment of a more inclusive and empowering society.

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