

“FAMILY MATTERS:” FETAL ALCOHOL SPECTRUM DISORDERS AND THE FAMILY

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Information about “family matters” is vital to developing targeted interventions, reducing placement disruption, and enhancing outcome in fetal alcohol spectrum disorders (FASD). The quality of the caregiving environment and family function are associated with long-term outcome in natural history study of individuals with FASD. This article integrates multiple information sources to better understand the role of family factors in the outcome of individuals with FASD, and how the family is affected by raising a child with this lifelong condition. A brief description of the useful informal literature is brought together with a review of the surprisingly limited body of systematic research findings on FASD and caregiver/family function, and new data describing children with FASD and characteristics of their caregivers. Directions for future data-gathering and intervention development emerge from combining what is already known with an exploration of what can be learned from a highly targeted review of family-related data in the wide-ranging, general literature on developmental disabilities, and use of a proposed conceptual framework that joins a developmental systems perspective with a family systems approach.

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A father recently wrote: “As an adoptive parent of (a child with FASD), I am finding that the world is divided in two: those who can see and recognize these children . . . and those to whom this situation remains an invisible epidemic.”

Fetal alcohol spectrum disorders (FASD) are a set of life-long neurodevelopmental disabilities both strikingly overlooked and understudied. This is true even though estimated incidence rates of this identifiable clinical population, including fetal alcohol syndrome (FAS) and conditions across the wider fetal alcohol spectrum, run as high as 9–10 cases per 1,000 live births in the U.S. [Sampson et al., 1997; May and Gossage, 2001]. This estimated incidence rate of FASD is even greater than recent, escalating rates in the U.S. of the well-known condition of autism spectrum disorders [Centers for Disease Control and Prevention (CDC), 2007]. FASD is clearly a global health problem, with many families affected around the world. A slowly growing number of countries have issued guidelines related to the risks of alcohol use during

pregnancy [e.g., Geneva Foundation for Medical Education and Research, 2009]. Yet because of diagnostic challenges and the lack of reliable and consistent data collection techniques, FASD incidence rates internationally are not yet accurately known [British Medical Association, 2007]. Rates are thought to be generally similar to those in the U.S., and data-gathering efforts are underway in many countries with some especially high-risk communities identified [e.g., May et al., 2006, 2007].

Lack of recognition of FASD is remarkable given that economic analysis finds that the full FAS, one condition on the larger fetal alcohol spectrum, is the most costly birth defect in the U.S. [Lupton et al., 2004]. Beyond FAS, additional affected individuals along the spectrum can only further raise family and societal costs. Indeed, initial estimated costs for FASD are very high [e.g., Costs for Canada: Stade et al., 2007]. Lack of awareness of FASD is also striking given that natural history data from several countries document the life-span implications of this condition. “Secondary disabilities” in lifestyle and daily function, such as mental health/behavior problems, trouble with the law, and dependent living have been found to be both frequent and debilitating for affected individuals (especially those on the spectrum but without the full FAS), and for their families and society at large [Streissguth et al., 2004; Spohr et al., 2007].

There are many gaps in our knowledge of this clinical population. In particular, we know little about “family matters,” despite the central, documented importance of the quality and stability of the home environment, and indicators of positive family function, to the ultimate outcome of those with FASD [Streissguth et al., 2004]. Although there is an informal literature, only a small amount of systematic data

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exist to describe the impact on the family of raising a child with FASD. In addition, research is only beginning to provide specifics on how parent and family variables influence outcome of those with FASD. Yet this information is vital to developing targeted interventions, reducing placement disruption, and enhancing the outcome of affected individuals.

This article integrates multiple information sources to better understand “family matters.” A brief description of the useful informal literature is brought together with a review of the surprisingly limited body of systematic research findings on FASD and caregiver/family function, and with new data describing children with FASD and characteristics of their caregivers. Directions for future data-gathering and intervention development emerge from combining what is already known with an exploration of what can be learned from a highly targeted review of family-related data in the wide-ranging, general literature on developmental disabilities, and use of a proposed conceptual framework that joins a developmental systems perspective with a family systems approach.

INFORMAL LITERATURE ON FAMILY ADAPTATION AND FASD

One vital and rich source of information on “family matters” are the thoughts of the families themselves, which point out directions for needed research. Important childrearing and family issues are articulated in first-person accounts, as shared in conferences, online forums, websites, and parent-oriented booklets and newsletters [e.g., SNAP, 1999; VON Canada, 2005; FAS-STAR, 2009; Iceberg, 2009; SAMHSA FASD Center for Excellence, 2009]. Information is also available through a great deal of media attention, and in published collections of parental experiences and clinical wisdom [e.g., Kleinfeld and Wescott, 1993; Kleinfeld et al., 2000; Salmon, 2007]. This extensive but more informal literature ranges world-wide, given the global reach of FASD as a public health problem and flexibility of web-based communication.

Exploration of this literature indicates that raising an individual with FASD has many rewards, and there exist a wealth of poignant and even light-hearted stories that take a positive view of the situation. But also highlighted are the dilemmas, costs, and impact of FASD on caregivers and families. Im-

portant issues involved in parenting someone with FASD vary across different “types” of families. Summarized testimony from birth parents, for example, indicates that childrearing is associated with feelings of guilt and shame, financial strain, frustration with the lack of knowledgeable professionals, stress related to the child’s involvement in the judicial system, and multiple time demands [Hope for Women in Recovery Summit, 2005]. There are also common needs across all family types. Town hall meetings held across the U.S. in 2002 and 2003 identified two primary needs: respite care and greater understanding of FASD by various systems of care. All participants noted the need for appropriate services for those with FASD and their families across all systems of care [SAMHSA FASD Center for Excellence, 2004].

One vital and rich source of information on “family matters” are the thoughts of the families themselves, which point out directions for needed research.

Anecdotal data reveal that childrearing presents many challenges to parents of those with FASD. Conduct problems raise questions for caregivers about what really causes misbehavior, and parents struggle with discrepancies between what can be expected from an affected individual based on his or her chronological versus functional age. The challenges in raising someone with FASD appear especially clear at certain developmental timepoints. Clinical wisdom highlights especially difficult times in early elementary school (around 2nd to 4th grade), during the move to middle school, and in the extended transition to young adulthood. Each of these may be thought of as “turning points” in the life trajectories of those with FASD, and families negotiating these turning points may require interventions and parent support specially tailored to these pivotal developmental steps.

Revealed in the informal literature is the very positive impact of both parent support and self-help. There are many informative web-based and hard copy efforts at parent-led community education, peer assistance, and self-help

(e.g., www.nofas.org; www.nofas-uk.org; <http://depts.washington.edu/fadu/Support.Groups.OI.html>). This informal literature, and publications from parent support organizations [e.g., Wilton and Plane, 2006], document that starting in the 1980s, with growing momentum by the early 1990s, specialized FASD parent support networks were developing in the U.S. and abroad. These networks were providing services such as information and referral, phone support, personal advocacy, list serves, lending libraries and publication outlets, and acting as platforms for diagnostic services. Multiple websites sprang up and parent support grew and matured, especially in countries with national leadership efforts. In the U.S. and Canada, for example, FASD-specific parent support organizations now also provide a platform for summer camps, teen groups, social skills groups, respite care, and birth mother networks, and offer access to materials for professional education, school health education, family planning information, and legislative action. In these and other countries, governmental agencies have been helpful in building state-wide, provincial and territorial networks for FASD-related services (see example websites: For U.S.: SAMHSA FASD Center for Excellence: <http://www.fascenter.samhsa.gov>; For Canada: Public Health Agency of Canada: <http://www.phac-aspc.gc.ca/publicat/fasd-fw-etcaf-ca/framework-eng.php>).

In this informal literature, word is being spread about the pressing need for formal services for individuals and their families, and the slowly growing service network. Anecdotal data underscore the usefulness to families of assistance from professionals well-informed and interested in FASD, and discuss what formal intervention services would be most useful. In this literature is lively discussion about family needs, medication, innovative or alternative treatments, and suggestions about how systematic and effective treatment and social services might be set up in various communities in countries across the globe.

IMPORTANCE OF QUALITY OF THE HOME ENVIRONMENT AND FAMILY-RELATED FACTORS ON DEVELOPMENTAL OUTCOME IN FASD

Good quality caregiving and stability of the home environment appear vital to successful outcomes of many individuals with FASD. This was pointed out early on by pioneering intervention

researchers [Giunta and Streissguth, 1988; Weiner and Morse, 1994] who went on to describe characteristics of a good quality caregiving environment and needed supports. Streissguth [1997], in her seminal and still influential book, *Fetal alcohol syndrome: A guide to families and communities*, discussed the even broader role of caregivers (and family) across the lifespan as the primary advocate for the individual with FASD. Kahlberg and Buckley [2007] further acknowledged the role of the caregiver in defining and guiding the school program for the child and adolescent with FASD.

There are systematic research data to support the importance of caregiving and the home environment to the outcome of those with FASD. A large-sample, retrospective, cross-sectional natural history study by Streissguth et al. [2004] provided data to support the central role of caregiving and home stability in the outcome of those with FASD. Streissguth et al. identified percentage of time spent in a stable/nurturing home environment as the most influential “protective factor,” reducing by three- or four-fold the odds for occurrence of four of five adverse life outcomes or “secondary disabilities” for individuals with FASD (e.g., disrupted school experiences, trouble with the law, etc.). Other important, family-related factors that reduced the odds for occurrence of all or almost all adverse outcomes were years spent per household by age 18, and whether an individual was ever a victim of physical or sexual abuse, or domestic violence.

Yet despite the demonstrated importance of family factors to outcome among those with FASD, natural history research, and subsequent archival data, have shown that a positive and stable family environment during childhood is not typical for this population. In fact, very high rates of environmental risk factors characterize clinical samples of children who have been exposed prenatally to alcohol or diagnosed with FASD, and these children often have other prenatal exposures. This means that most children with FASD experience “double jeopardy” because they also have lived in poor caregiving environments. In data archived in a large clinical database, alcohol-exposed young children (birth to 8 years; $n = 781$) [Olson et al., 2007] and school-aged children with FASD (6–12 years; $n = 573$) [Coggins et al., 2007] were found to be disproportionately subject to negative or unpredictable caregiving environments. For example, Olson et al. found that 84.5% of the younger sample

had other prenatal exposures, and 82.6% had clinical rankings indicating either “high” or “some” postnatal environmental adversity. Coggins et al. found similar levels of psychosocial risk among the older children.

Children experiencing the “double jeopardy” of a very difficult caregiving environment who also have FASD do more poorly than children who only experience maltreatment. The impact of cooccurring FASD and an adverse family environment on developmental outcome was directly examined in a cross-sectional study of U.S. children in the child welfare system, aged 6–16 years ($n = 274$), 40% receiving a diagnosis on the fetal alcohol spectrum. Of these children, 97% were rated as moderately to severely traumatized by postnatal experiences, and the sample was divided into “FASD + trauma” (40%) and “no FASD + trauma groups” (60%). Compared to children who had only been

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traumatized, Henry et al. [2007] found that children with FASD who had also been traumatized did more poorly on classroom-oriented neurodevelopmental measures (attention, memory, language) and an intellectual estimate. They also scored lower on both parent behavior ratings (signs of oppositional behavior, problems related to Attention Deficit/Hyperactivity Disorder (ADHD), social problems, and total problems) and teacher behavior ratings (problems related to ADHD and total problems).

Large-sample, longitudinal, population-based prospective studies of maternal alcohol use are usually focused on moderate to heavier social drinking, rather than the problem drinking often seen in clinical samples. Such studies can

statistically adjust for confounding factors, and are more representative of the community at large than are clinical samples. Similarly to clinical studies, these longitudinal studies reveal that prenatal alcohol exposure is related to poorer outcome, and prenatal exposure is also often associated with postnatal family risks which themselves can adversely affect development [e.g., Sood et al., 2001]. But carefully-designed longitudinal studies also document that caregiving can have a positive impact on the outcome of those born alcohol-exposed. Jacobson et al. in a large longitudinal study of 7½-year-olds prenatally exposed to alcohol, found that a more positive home environment in elementary school was associated with less severe alcohol effects on certain aspects of cognition, suggesting that raising a child in a more intellectually stimulating or enriched home environment may buffer some of the observed effects of prenatal alcohol exposure [Jacobson et al., 2004].

PRENATAL ALCOHOL EXPOSURE, FASD, AND PARENTING

To develop effective parenting and family-level interventions for individuals with FASD and young children born alcohol-exposed, factors pivotal to family adaptation and developmental outcome of affected individuals must be identified through systematic research.

A Developmental Systems Study

Developmental systems research is uniquely able to identify these pivotal variables, which can help in intervention planning. Pioneering developmental systems research carried out by O'Connor et al. examined heavily alcohol-exposed young children and their caregivers, who may continue to drink and/or who may experience psychological distress such as depression.

A series of studies traced how developmental and relationship problems emerged in infancy within the parent-child interaction. In a lower-risk sample, women who drank more heavily during pregnancy had infants displaying higher levels of negative affect than those with less prenatal exposure. In addition, these heavier drinkers interacted in ways that were less responsive and developmentally stimulating to their babies, and their children displayed higher levels of insecure attachments in later infancy. These problems continued as children moved through toddlerhood and preschool, and were associated with increased internalizing behavior problems by the time the

children reached age 5–6 years [O'Connor et al., 1993; O'Connor and Kasari, 2000; O'Connor, 2001]. O'Connor and Kasari [2000] found further intricacies in the developmental process. Not only did mothers who drank more during pregnancy have children reporting more depressive features, but they also found that if the mother was depressed, the child was more likely to acknowledge higher levels of depressive symptoms. Prenatal alcohol exposure and maternal depression had additive predictive effects. Findings could not be explained by the mother's current drinking practices.

In a separate higher-risk sample, O'Connor et al. found that when children have higher levels of cumulative risk in the larger social context, such as poverty or living with a single parent, this negative developmental trajectory worsens [O'Connor et al., 2002]. Of interest, 80% of children exposed prenatally to alcohol showed insecure attachment (a pivotal developmental feature) while, even in this high-risk sample, only 36% of the nonexposed children displayed attachments rated as insecure. When mothers provided a high level of emotional support (another key developmental factor), their children had better coping skills and more secure attachment relationships [O'Connor and Paley, 2006].

Caregiver Stress

Recent FASD-related research has focused on caregiver stress, a key family-level construct. As Plant and Sanders [2007] point out from more general study of developmental disabilities, heightened caregiver stress is related to other negative family factors, such as coercive parent-child interactions, increased risk of family maladjustment, and parental depression. As Olson et al. [2007] suggest, the caregiver stress of raising a child with FASD may be different from that in other developmental disability groups. FASD carries an emotional overlay, because it is a birth defect that could have been prevented, and the disruptive influence of parental substance abuse will always, in some way, be part of the family history. The question for intervention planning is how caregiver stress is related to other variables within the developmental process, so stress can be reduced.

Paley et al. [2005] used a developmental systems perspective and path analysis to examine relationships between prenatal alcohol exposure, child externalizing behavior, caregiver-

child interaction, current maternal alcohol consumption, and parenting stress. They studied a high-risk sample of 42 biological mothers of 4- to 6-year-old children with varying degrees of prenatal alcohol exposure, oversampled for heavier use, none with the full FAS. Current maternal alcohol use was not associated with prenatal alcohol exposure or level of externalizing behavior. Mothers of children with clinically significant levels of externalizing behaviors reported significantly higher levels of child-related and total stress (using the Parenting Stress Index (PSI)) than did those whose children's behavior did not reach clinical cutoffs. Other child and demographic variables, such as child or maternal intelligence quotient (IQ), child gender and ethnicity, marital status, SES, caregiver years of education, and indicators of family size, did not contribute to the association between child externalizing behaviors and maternal stress. Of importance, a model in which maternal stress was presumed to relate to less supportive mother-child interactions and higher levels of current alcohol consumption, which were in turn expected to relate to greater child externalizing behavior, did not fit the data. Instead, the best fit to the data was a model in which children with higher levels of prenatal alcohol exposure showed more externalizing behavior problems which, in turn, were associated with greater maternal stress. In addition, there was a direct path from parent report of fewer family resources to higher levels of maternal stress.

A second study by Paley et al. [2006] examined 100 children with prenatal alcohol exposure and their parents; 71% living with adoptive parents, and the remainder with one or both birth parents. Using parent questionnaires or structured parent interview, these investigators found that child-related parenting stress, again assessed with the PSI, increased in association with the degree of child externalizing behavior problems. But greater child-related stress was also related to increases in internalizing behavior problems, and to decreases in both executive and adaptive functioning. In fact, the child's reported level of executive functioning was the strongest predictor of child-related parental stress. Neither child-related nor parenting role-related caregiver stress was associated with child IQ or presence of the full FAS diagnosis. Assessing family-level variables of interest, they found that child-related stress was associated with adoptive parent status, but not with ad-

equacy of family resources, while parenting-role related stress was independently associated with both birth parent status and fewer family resources. Paley et al. [2006] have suggested that the connection between parental stress and raising a child with FASD is related not to a child's diagnostic condition on the fetal alcohol spectrum (or even to IQ), but rather to the actual functional impairments of the child with FASD. They comment that it may be the "child's cognitive limitations in planning and organizing their behavior to engage in effective problem-solving and their ability to perform developmentally appropriate tasks in everyday life that were especially taxing to parents" (p 401).

Level of Support for Adaptive Function as a Measure of Caregiver Burden

Another pivotal factor to family adaptation is the degree of support that parents and the family must mobilize to help a child succeed day-to-day, with a greater degree of support creating caregiver burden. The question for intervention planning is how to help caregivers bear this burden, including setting appropriate funding levels for social services such as respite.

Jirikowic et al. [2008] studied adaptive function in a group of 25 younger children (aged 5–8 years) with FASD compared to a typically-developing group of 23 peers using parent ratings from the Scales of Independent Behavior-Revised (SIB-R). A SIB-R rating of the degree of adaptive support needed by the child is a better predictor of service intensity than either the child's level of adaptive function or maladaptive behavior alone. For instance, a higher-functioning child with severe behavior problems may be more difficult to support than a lower-functioning child without problematic behavior. Nearly half (48%) of the children with FASD needed the second and third most intensive levels of "extensive" or "frequent" support. No children with typical development required these burdensome levels of support. An additional 39% of children with FASD required support at the next level, termed "limited support." Providing extensive, frequent, or even limited support for a child's everyday functioning is a significant caregiver burden. In contrast, while three-quarters (74%) of the children with typical development needed only the lowest levels of "intermittent" or "infrequent" support, only a small number (13%) of the children with FASD could manage with "inter-

mittent” support for adaptive behavior and none could manage with “infrequent” support.

Data on Needs and Parenting Experiences in Different Family Types

A few researchers have begun systematic analysis of qualitative data to derive participant-generated themes of family impact, parenting experiences and perceived family needs among different types of families raising individuals with FASD. Birth, foster, and adoptive parents have been queried. Their experiences identify pivotal factors to family adaptation in these different family types. The question for intervention planning is how to adjust treatments according to family type.

Salmon [2008] conducted detailed analysis of small-sample, open-ended interview data using a constant comparative method to reveal the “lived experiences” of eight multiparous, middle to high socioeconomic status biological mothers in New Zealand. This information has universal relevance. Using a feminist standpoint theory, several themes emerged, including that all women feared for their child’s future. First, according to birth mothers’ report, the larger social world seemed to view FASD as the birth mother’s fault, while the “lived experience” of the birth mothers was actually that they lacked knowledge about FASD and drank before they knew they were pregnant. Second, birth mothers reported feeling abandoned by medical and health professionals. While they desired an accurate diagnosis, they felt they were left to deal with problems on their own, and had to independently develop an ability to understand and cope with their child’s neurodevelopmental problems. Third, birth mothers reported they lacked support from the educational system for themselves and their children, in that they were not listened to, and their child’s problems were not understood or responded to within the classroom. Finally, birth mothers reported being seen by the larger social world as the cause of their children’s criminal behavior. Their perceptions were that police unduly blamed them and actually displayed little or no knowledge of FASD.

Narrative analysis of small-sample interview data on the experiences of foster and adoptive parents raising a child with FAS in the U.S. reveals seven common themes that have universal relevance. As with birth parents, these document wide-ranging family impact,

but show different concerns: the need for constant vigilance; effects on marriages; child management concerns, parental issues (such as feelings of inadequacy and slow parenting adjustment with time and experience); complexities of interactions with the professional community; medical implications; and emancipation concerns [Morrisette, 2001].

Qualitative study with foster parents in Canada used interesting “concept mapping” methodology, and yielded participant-generated results that were grouped and further subjected to multidimensional scaling and cluster analysis. Results have universal relevance, and show many similarities but also some differences from the larger

Recent systematic FASD intervention studies have all involved caregivers as change agents in efforts to improve the behavioral outcome and targeted skills of preschool and school-aged children with FASD [Interventions for Children with FASDs Research Consortium, 2009], with promising results.

fostering literature. Three questions were asked of a group of 63 licensed foster parents raising children with FASD about their motivations for fostering, needs for successful placement, and what might be causes for a placement breakdown. Motives for fostering included witnessing positive changes while they cared for children, helping children focus on their strengths, using their own parenting experience to good effect, earning an income, wanting to help children with disabilities, and helping children stay connected with their families and communities [Brown et al., 2007b]. Needed for successful placement was the foster parents’ ability to provide structure and a high level of organization, the right kind of personality and skills and a good understanding of FASD in the foster parent, and the availability of family and neighbor social

support and funding. These foster parents also noted the need for professionals informed about FASD and the child’s care team, and for peer advice and networking [Brown et al., 2005]. Foster parents reported they would end a placement if the child’s behavior became unmanageable despite attempts to make the placement work, or if the child was at risk to harm others at home. They also noted they would end a placement because of burnout, if demands were too great or there were inadequate resources, if they had not been given sufficient information, or if they felt they were being taken for granted [Brown et al. 2007a].

Caregivers as Change Agents in Outcome of Children with FASD

Data indicate that caregiving can impact developmental outcome among individuals with FASD. The question for intervention planning is how to make that happen. Recent systematic FASD intervention studies have all involved caregivers as change agents in efforts to improve the behavioral outcome and targeted skills of preschool and school-aged children with FASD [Interventions for Children with FASDs Research Consortium (ICFRC), 2009], with promising results. The five studies included in the ICFRC each used a different intervention that always included some type of parent support and education to improve the caregiving environment, with four studies involving the parent directly with the child in skill-building practice or behavior management techniques. These interventions all aimed to increase parent knowledge of FASD and related topics, and variously attempted to change parenting attitudes and self-reported behavior, observed parenting skills, or parent involvement in assisting child learning. Several interventions explicitly worked to help parents understand their child’s functional impairments and connect families with needed community resources. Some of these interventions were highly individualized and could be adjusted for the different needs of family types.

In all studies, parent knowledge increased and some projects found altered caregiver attitudes and behavior. In all studies, some degree of positive change was seen in child outcome, suggesting alterations in the caregiving environment may ameliorate the deficits seen in FASD. Improvement was seen in: direct child testing of knowledge of appropriate social behavior, and parent report of social skills and problem

behavior, (children aged 6–12; Study no. 1); parent report of problem behaviors and direct child testing of mathematics outcomes (children aged 3–10 years; Study no. 2); parent overall rating of behaviors related to executive function (children aged 6–11; Study no. 3); and parent rating of child disruptive behavior (younger children aged 3–7; Study no. 4; children aged 5–11 with serious challenging behavior; Study no. 5).

NEW DATA ON CHARACTERISTICS OF FAMILIES RAISING CHILDREN WITH FASD AND BEHAVIOR PROBLEMS

Presented here are new descriptive data to learn more about caregiver characteristics and family needs. The question for intervention planning is how to tailor and refine treatments based on specifics about the population to be served.

Baseline data have been drawn from our systematic research on a behavioral consultation intervention, Study no. 5 in the ICFRC discussed above, called the “Families Moving Forward Program.” This intervention was designed for families raising preschool and school-aged children with FASD and very clinically concerning externalizing behavior problems. Children were recruited from a large clinical database of individuals with FASD in the U.S. ($N \approx 1,500$ at time of recruitment).

While not necessarily representative of all families raising individuals with FASD, this high-need group is important to understand. Based on prospective longitudinal data, and secondary analysis of large datasets (including community samples), there is a strengthening case for an association (perhaps causal) between prenatal alcohol exposure and an increased risk for symptoms of conduct problems. This association is likely mediated by environmental effects, but with prenatal alcohol exposure still making unique contributions [e.g., Baer et al., 1998; Sood et al., 2001; Disney et al., 2008]. Designing treatments for families raising children with FASD who are already showing significant externalizing symptoms in the preschool and elementary school-age years is a high priority. These families are most likely to be seen by mental health, school, social services, and medical providers, and need appropriately tailored and effective treatments.

Diagnosis and Demographics

Highlights of data from our first randomized control trial of 52 families

Table 1. Sample Characteristics for Children with FASD and Behavior Problems and Their Primary Caregivers (N = 52)

Variables	Mean (SD)	Percentage
Child age at enrollment	8.53 (2.03)	
% Child gender (male)		51.9
Ethnicity ^a		
% White/Non-Hispanic		50.0
% Black		7.7
% White/Hispanic or White/Mexican		3.8
% Native Ancestry		3.8
% Mixed Ethnicity		34.6
% Primary caregiver married/living with partner		75.0
Primary caregiver years of education		
11 years or less		3.8
12 years		30.8
13–16		46.2
17 years or more		19.2
Yearly household income in \$ ^b		
\$15,000 or less		11.5
\$16,000–\$39,000		15.4
\$40,000–\$59,000		28.9
\$60,000–\$79,000		15.4
\$80,000–\$99,000		13.4
\$100,000 or more		15.4
Family type (defined as type of primary caregiver) ^c		
% Biological parent (mother or father) ^d		11.5
% Biological grandparent (grandmother or grandfather)		11.5
% Adoptive parent (mother or father)		48.0
% Foster parent		15.4
% Legal guardian		3.8
% Other primary caregiver type (relative placement (e.g., sister, cousin), stepmother, or nonbiological grandparent)		9.6
Number of children currently in home (including child) (range 1–7)	2.67 (1.28)	
% Primary caregiver scoring within normal limits for current drinking (AUDIT ^e <8)		100.0
Number of significant earlier stresses experienced by child (range 0–9)	5.19 (2.36)	

^aThis article reports ethnicity or racial origin with more specificity than offered in other papers from this study, and emphasizes mixed ethnicity. Children are only reported as belonging to a specific category (e.g., White/Non-Hispanic; Black, Native ancestry, etc.) if both biological parents were reported with that ethnicity or racial origin. “Native Ancestry” indicates American Indian, Alaskan Indian, and or Canadian Indian. “Mixed ethnicity” includes multiple combinations of ethnicities or racial origins from the table above, and instances where the ethnicity or racial origin of one of the parents was reported as “other” or “unknown.” There are many different combinations, some describing only one child. When data from this study have been reported in less detail or alongside other studies using common definitions, different categorization methods for ethnicity have been applied.

^bThese data are based on the primary caregiver’s estimate of exact yearly household income, which differs slightly from reported data grouped categorically. The minimum reported exact yearly household income was \$10,000 and the maximum was \$250,000.

^cIt is possible to redefine family type in other ways. For instance, families self-reported their “type” of family into categories such as “kinship care, informal,” “kinship care, adoption,” and other categories. Percentages are fairly similar, though not identical, when different definitions of family type are used.

^dThe percentage of children living with their biological mother was only 5.8% of the total sample of 52 children.

^eThe AUDIT is the Alcohol Use Disorders Identification Test [Babor et al., 1989]. This is a measure developed by the World Health Organization to assess any current high-risk drinking. The AUDIT was only administered to the primary caregiver in this study.

are included here. In these data, 25% of the children had a diagnosis of the FAS (or partial FAS) and 75% had diagnoses falling on the larger fetal alcohol spectrum (which might be termed as alcohol-related neurodevelopmental disorder, or ARND). Data in Table 1 reveal clearly the striking demographic diversity in our sample, exemplified by the wide variety of ethnicities and family types. Such diversity appears characteristic of families raising individuals with FASD. Overall, the current sample was not comprised of high-risk families, unlike those in many clinical studies. As seen in Table 1, 75% of those in the sample were two-caregiver families (married/living with partner), the average educational

level of primary caregivers (most of whom were female) was beyond high school, most families were in the middle to higher-income range, and no primary caregivers reported alcohol use disorders. However, these families were raising children with both prenatal and accumulated postnatal risk. For example, these alcohol-exposed children often had other prenatal exposures or other risks, such as poor prenatal care. Growing up, these children had experienced an average of about five significant stresses (range from 0 to 9) on a measure developed for the study, such as neglect, violence toward the child, living with parental substance abuse, parental separation or divorce, living in poverty, or major traumas.

Neurodevelopmental and Adaptive Deficits Among Children with FASD

True to the definitions of FAS and ARND, these children had clear evidence of significant (but individually variable) neurodevelopmental disabilities in multiple domains. They definitely showed the functional impairment, Paley et al. see as central to caregiver stress. On average, their estimated intellectual status was within the average range (mean Kaufman-Brief Intelligence Test [K-BIT; Kaufman and Kaufman, 1990], IQ estimate = 94.27), though with about one-quarter (23%) of the sample falling more than 1 SD below the mean. Yet, as one example of their learning problems, when tested on “everyday memory” using the Rivermead Behavioral Memory Test, Children’s Version [Wilson and Ivani-Chalian, 1991], over half of this sample of children showed performance that was considered borderline/poor memory (36.5%) or moderately/severely impaired (21.2%). As is characteristic of FASD, this group of children had low adaptive function (mean Vineland Adaptive Behavior Scale (VABS) Composite = 66.00) [Sparrow et al., 1984], as reported by their parents, quite discrepant from their average estimated IQ. As the data [Jirikowic et al., 2008] point out, these were children requiring considerable adaptive support from their parents.

Behavior Problems and Caregiver Burden

At baseline, before treatment, these children with FASD showed very real and complicated behavior problems. The group overall scored on average above the clinical cutoff (T-score ≥ 64) for both internalizing and externalizing problems, using an age-appropriate version of the standardized caregiver questionnaire called the Child Behavior Checklist (CBCL) [Achenbach and Rescorla, 2000, 2001]. In this sample, 80.8% had CBCL Externalizing Behavior Problem scores at or above the clinical cutoff (as compared, for example, to 26% in the Paley et al. [2006] sample). The burden of caregiving, before treatment, was demonstrated in the difficulties these children showed across situations and activities, both at home and, to a lesser extent, at school. For example, their parents reported an average of 12 (of 16) situations in which the children showed problems (e.g., “in public places,” “when visitors are in your home”), with an average severity score of 5.08 (moderate; possible range 1–9), using the Home Situations Questionnaire [Barkley and Murphy, 2005].

Psychiatric Conditions

As in other clinical samples, these were children with high rates of co-occurring psychiatric conditions (though it is important to be careful in interpreting these data, because the medical condition of FASD must be taken into account in how children’s symptoms are interpreted and diagnosed). Using a structured psychiatric interview [C-DISC-4; DISC Development Group, 2000; Shaffer, 2000], data for diagnoses over the past year revealed the following proportions of children meeting full criteria for a “positive” diagnosis: 74.0% for attention deficit/hyperactivity disorder (ADHD); 72.5% for oppositional defiant disorder (ODD); and 27.4% for anxiety disorders. Rates of other psychiatric conditions were lower. In these data, the same child might have one or more disorders.

Positive Child Characteristics

Descriptive data gave a glimpse of the many positives of these children with FASD. A set of positively-oriented questions were drawn from the Child and Adolescent Functional Assessment Scale [CAFAS; Hodges, 2000] and Preschool and Early Childhood Functional Assessment Scale [PECFAS; Hodges, 1999]. On these questions, half or more of the caregivers reported the following characteristics were “very true” of their children, starting with the most frequent: takes pride in doing activities independently; participates in family activities; willing to accept household routines; willing to take help from the caregiver; and seeks help from the caregiver. That these children were engaged with their families and willing to receive (and even seek) help are potentially powerful protective factors. For these parents, the “special benefits” of raising a child with FASD were poignantly revealed in interviews, such as a parent who described their child with FASD as a “teacher” of what is important in life.

Parenting Attitudes: Caregiver Stress and Distress

As in other samples of families raising children with FASD or general developmental disabilities, parenting stress was clinically elevated, as measured with the Parenting Stress Index (PSI) [Abidin, 1995]. But in contrast to data on a sample at higher socioeconomic risk studied by Paley et al. [2006], clinical elevations in this sample were seen only in the area of child-related stress, with 92% of primary caregivers (of all family types) reporting

child-related stress above the clinical cutoff. Compared to PSI norms, these caregivers, on average, rated their parenting role-related stress and overall life stress as well within normal limits. A number of caregivers, though, reported experiencing significant psychological distress, using the Symptom Checklist 90-Revised [Derogatis, 1994]. Caregiver distress has not been previously studied in this population. Before treatment, a notable 23% of these caregivers scored at a level indicating therapeutic intervention would be suggested using the Symptom Checklist 90-Revised [Derogatis, 1994]. In an additional sample of 37 families currently under study, where more fine-grained data on caregiver psychological status was obtained, 10.8% of caregivers fell above clinical cutoffs before treatment on the Center for Epidemiology Scale for Depression [Radloff, 1977]. It should be noted that only 25% of this sample of 52 families had been able to access respite care over the year before treatment to alleviate caregiver burden.

Differences Between Family Types

Table 1 presents percentages of different family types, using relationship of the primary caregiver as one definition of family type. Table 1 makes clear that there were many different family types. Although data should not be over-interpreted given sample size limitations, it is useful to explore a few subgroup differences between family types, omitting the “other” category in this exploration. Children not with adoptive parents (and especially those currently living with foster parents) were over-represented among those experiencing earlier significant life stress (e.g., stress score $> \sim 5$). Primary caregivers who were birth parents or foster parents were over-represented among those endorsing concerning levels of psychological distress. Primary caregivers with direct biological relationships to the child (e.g., birth mothers, birth fathers, and/or grandparents) significantly more often reported monitoring their children less fully when compared to adoptive and foster parents, though no other group differences on self-reported parenting behavior were found.

Parenting Attitudes: Parenting Sense of Competence and Family Needs

An important area of caregiver attitudes is a sense of satisfaction and efficacy in parenting, assessed in our study with the Parenting Sense of Competence Scale (PSOC) [Johnston and

Table 2. Top Unmet Important Family Needs from Caregivers Raising Children with FASD and Behavior Problems

Percentage Indicating Need is Unmet	Type of Family Need
69.2%	Discuss feelings about my child with someone who has gone through the same experience.
61.7%	Have help in preparing for the worst.
60.8%	Have enough resources for myself or the family.
58.8%	Have help in remaining hopeful about my child's future.
58.0%	Get a break from my problems and responsibilities.
52.9%	Have complete information on my child's thinking problems.
55.8%	Be reassured that it is usual to have negative feelings about changes in my child's behavior.
51.0%	Be shown what to do when my child is upset or acting strange.
48.1%	Be told why my child acts in ways that are different, difficult or strange.
47.1%	Have different professionals agree on the best way to help my child.
47.1%	Pay attention to my own needs.

"Important" is defined as parent report that a need was "important" or "very important" (where there were two other levels indicating less importance). "Unmet" was defined as parent report that a need was met "not at all" or "a little" (where there were two other levels indicating that a need was met more completely). Items shown here were the most frequently endorsed items; the remaining items (of 20) received far less frequent endorsements.

Mash, 1989] and to our knowledge not previously studied in FASD. Scores gathered before treatment began were compared to reference data for mothers of children with typical development available in the literature (aged 4–9 years). Surprisingly, primary caregivers of these children with FASD felt just as satisfied with parenting as the reference group (an affective dimension reflecting the degree to which the parent feels frustrated, anxious, and poorly motivated in the parent role). Also of note, these parents tended to feel slightly more efficacious than the reference group (an instrumental dimension, reflecting the degree to which the parent feels competent, capable of problem solving, and familiar with parenting). For intervention planning, further enhancing these attitudes is of interest, but other parenting attitudes may also need to be targeted.

Before treatment began, parents in this sample expressed a number of important but unmet family needs. Results are presented in Table 2, and offer specific directions for targeted FASD intervention. Frequently endorsed unmet needs included a call for adequate resources and the support of other parents. But reassurance and support for negative emotions and the need for self-care were also reported as frequently unmet needs. Another unmet set of needs were for obtaining the kind of input that can be offered in collaborative behavioral consultation by a clinician with special expertise in FASD. Examples of such input are obtaining complete information on the child's thinking problems, or being shown what to do when the child is upset or

acting in an unusual manner. The Families Moving Forward Program [Olson et al., 2005; ICFRC, 2009] is one such behavioral consultation intervention, specialized for families raising children with FASD or other forms of neurological impairment.

Parenting Behavior

Self-reported parenting behavior was assessed, among other measures, by the Alabama Parenting Questionnaire (APQ) [Shelton et al., 1996], and has not previously been studied in FASD. The APQ is an instrument used to assess empirically-identified positive and negative parenting styles important to the emergence of conduct problems in children. Reference data were available from literature for children with typical development in a community sample [caregivers of children aged 4–9 years; Dadds et al., 2003], and children with disruptive behavior disorders assessed after treatment [aged 4–8 years; Finefield and Baker, 2004]. Even though they were raising challenging children with FASD, and data were collected before receiving treatment, parents in the current study on average reported parenting behavior similar in many ways to reference parents. Before intervention, parents raising children with FASD reported being just as involved with their children, engaged in positive parenting, and consistent in discipline as were reference parents. They did report monitoring less fully than parents raising children with typical development. Based on self-report, then, these parents would not seem to need assistance on parenting skills typically targeted in intervention for children who have be-

havioral concerns, such as relationship-building or training on compliance strategies. However, instruction on monitoring and supervision might still be useful, and these parents may need other types of parent training.

These new data extend what is already known about FASD and the family. Data highlight the diversity of families raising children with FASD, and some exploratory differences between family types, highlighting why flexible, individualized interventions are imperative. The extent of the children's learning and behavioral deficits, creating functional impairment, and their frequent cooccurring mental health conditions coupled with psychosocial risk, are very clear. This helps to explain the high level of caregiver burden, stress, and distress reported by their parents. Counterpoint to this are the positive child characteristics reported by parents which serve a protective function. Data on self-reported parenting attitudes and parenting behavior suggest that interventions should aim to further enhance parenting self-efficacy and satisfaction, link parents to resources and parent support, and provide collaborative behavioral consultation and emotional support that takes a different approach than standard parent training.

To guide further development of research on FASD and the family, this article now turns to selected, recent research addressing developmental disabilities in general (with an emphasis on the pediatric literature), a more mature literature with much to offer. This literature was selected because the authors view FASD as a developmental disability (arising from a neurobehavioral teratogen) that typically presents with comorbid mental health conditions. Brief reference is also made to selected studies from research on traumatic brain injury and disruptive behavior disorders, which offer additional insight into directions for study of FASD and the family.

DEVELOPMENTAL DISABILITIES AND THE FAMILY

Developmental disabilities are known to have a profound impact on children's health and functioning. Recent literature clearly reveals that "the experience of having a child with an intellectual or developmental disability almost inevitably has a significant impact on the family" [p 291; Bailey, 2007]. In turn, the family environment, assessed in a variety of ways, influences the development and behavior of a

child with developmental disabilities. For intervention planning in the field of FASD, this means that a focus on the family is imperative.

Bailey, a leader in developmental disabilities research, underscores the potential positive influence of the family environment on the development and behavior of a child with intellectual and developmental disabilities. This literature finds that direct interactional variables, such as positive maternal “expressed emotion” or the responsiveness within the parent–child interaction, play an important role. But indirect variables, such as a positive family climate or availability of financial resources, are also crucial influences on children’s development and behavior.

Bailey also notes the research consensus that mental health and behavior problems of individuals with developmental disabilities are central factors associated with problems in family adaptation and parental well-being, including measures of negative impact and parenting stress, caregiver burden and parental depression. There is also consensus that both mental health concerns and the negative impact on parental well-being can be lasting. For young children, Webster et al. [2008] found that a child’s socialization skills were the strongest functional and developmental predictors of the level of parenting stress. Blacher and Baker [2007] found that behavioral challenges account for more variance in negative indicators of parental well-being than disability status, in samples of both preschoolers and young adults with developmental disabilities.

Caregiver Stress and Cognitive Appraisal

It is well-known that families raising children with developmental disabilities experience stress. But one interesting new approach is to use longitudinal data to examine stress trajectories, which allows for examination of the level and rate of change in stress over time. Most et al. [2006] studied mothers raising children with Down syndrome in comparison to a group of mothers raising children with developmental disabilities of mixed etiologies. Their findings revealed a surprising pattern of rising maternal stress over the first few years of life in those raising children with Down syndrome. This pattern may be linked to emergence of the phenotypic characteristics of cognitive and linguistic deficits, and behavioral issues, among children with Down syndrome. These data provide a window

into family development over time, modifying previous ideas that there was a “Down syndrome advantage” in family functioning. These interesting findings suggest that caregiver stress may change across the lifespan, and that interventions for different disabilities (such as FASD) may need to be time-sensitive, targeted to important “turning points” in development.

Moderators and mediators of parenting stress have been studied in developmental disabilities. Plant and Sanders [2007] focused on generally well-educated, higher SES mothers of younger children with generally mild disabilities but clinically concerning behavior problems. Among other findings, parents’ cognitive appraisal (way of judging, ranging from positive to negative) of their child’s level of disability directly influenced their parenting stress. In addition, parents’ cognitive appraisal of caregiving responsibilities was a significant independent predictor of parenting stress. Varying types of social support buffered different sources of parenting stress. For example, high levels of support from external agencies influenced parenting stress related to overall levels of child problem behavior. In contrast, partner/family support buffered parenting stress related to the child’s level of disability.

Cognitive appraisal is an important construct that has been examined in more detail. Parents’ interpretation of the family impact of childhood disability appears to include both positive and negative appraisals, which can cooccur and follow different trajectories over time, and can be different across mothers and fathers [Trute et al., 2007].

One theoretically-guided approach to the study of caregiver stress among parents raising children with intellectual disability and behavioral difficulties was carried out by Hassall et al. [2005]. Among mothers in a sample from the UK, stress was found to be mediated largely by caregiver attitudes (parenting sense of competence (especially satisfaction) and parental locus of control), and by the child’s behavior problems. Stress was also related to the larger environmental variable of perceived helpfulness of social support available to the family, though the parent variable of locus of control seemed to be a mediating factor in this relationship. The basic idea is that parents employ a variety of coping strategies to successfully adapt to the challenge of a child with disabilities. For intervention planning in the field of FASD, this means that treatments

addressing parental cognitions/attitudes and problem-focused management may be a useful adjunct to helping parents learn skills through behavioral parent training.

Positive Views of the Individual with Disabilities

Of recent interest is the evolving idea of the positive family impact of the individual with disabilities, as separate from, and not necessarily associated with, negative impact [Perry, 2005]. Studying two large samples quite different in age and level of disability, Blacher and Baker [2007] proposed several types of positive perspectives on raising a child with a disability, including a “low negative” view, a “common benefits” view, and a “special benefits” view. They further proposed that these various perspectives on “positive” merge in more generic coping models, where positive perceptions serve therapeutically as an adaptive coping mechanism. Among other findings, Blacher and Baker noted that when childrearing challenges were lower, there was little relationship between positive views of parenting and experienced stress. But with increased challenges, those caregivers who held the least positive views of parenting experienced the highest levels of stress. Findings further revealed that even though raising a child with a disability is more stressful than raising a child who is typically developing, parents of both types of children do experience the “common benefits” of parenting.

Blacher and Baker note that models of stress and coping suggest the relationship between child stressors and parental well-being is affected by parental cognitions and attitudes, but also by family resources (e.g., income and social support). In addition, they suggest that personality characteristics of parents (such as an optimistic disposition) can buffer childrearing challenges. For intervention planning in the field of FASD, this means that positive views are of importance.

Informal Support Systems and the Many Roles of Parents

According to Bailey [2007], the nature and quality of social support available to families has been repeatedly demonstrated as critical to positive family adaptation. Social support comes from many sources, but multiple analyses show that positive family adaptation is more likely to occur for individuals or families with strong informal support

systems. This is vital to intervention planning in the field of FASD.

In developmental disabilities, there are many roles parents can play, not only as an advocate for their own child but as also a peer support to other families, a formal member on health care committees, someone involved in the education of clinicians, and a person actively involved in systems change in health care [Dokken and Ahman, 2006; Landis, 2007; Williams, 2007]. Parent support has grown to become sustainable and formalized in many ways. There are statewide networks of general parent-to-parent support, fathers' networks, and parent-to-parent support specialized for different disabilities (such as FASD). Parent support networks are the platform for many services and efforts at self-help.

Cultural and Sociocultural Views

A wide range of studies have shown that cultural and sociocultural contexts shape the meanings families place on intellectual and developmental disabilities, which in turn affect their experiences, responses, accommodations, adaptations, and the practices they choose [Nehring, 2007; Skinner and Weisner, 2007]. Indeed, data reveal that predictors of the family impact of disability differed markedly for three ethnic groups (White/Non-Hispanic, Hispanic, African-American) in analysis of national interview survey data. This suggests that generalizing across ethnic groups may limit interpretability of data [Neely-Barnes and Marcenko, 2004]. A recent study of perceived negative impact among mothers of children with autism also found differences across ethnic groups [Bishop et al., 2007]. Results indicated that African-American mothers reported significantly less perceived negative impact than Caucasian mothers.

But ethnic or cultural group comparisons are only one simple way to examine this issue. Skinner and Weisner [2007] suggest a sociocultural approach to research on family adaptation that envisions "culture" as a system of meanings and practices that evolves between families, the medical and service community, and the larger world. To understand the complexities of the disability experience, they suggest use of different types of measurement, including ethnography and narrative analysis, such as an "eco-cultural interview" in which parents of children with disabilities are engaged in a conversation to walk the interviewer through their day. This approach examines meaningful issues

such as well-being and quality of life, and family "accommodations" (or the functional responses to having a child with developmental delays). With the dramatic diversity that characterizes families raising children with FASD, suggestions to consider cultural and sociocultural views are important to intervention planning.

Family Involvement

Parents of children with FASD are often required to be their child's advocate and are directly involved in their treatment. Many times, this is because professionals lack knowledge of FASD, so parents assume responsibility for their education. This is true more generally in the field of developmental disabilities. Indeed, family involvement in planning, choosing, and even shaping services for their children is now a key intervention concept in the field of developmental disabilities, and more highly involved families have been found to show better outcomes [Neely-Barnes et al., 2008]. Family-centered care has become recognized as the standard of practice for children with special needs in health care, early intervention, and education [Dokken and Ahman, 2006].

Person-centered and family-centered care is also strongly advocated for individuals with comorbid mental health problems and developmental disabilities [McGinty et al., 2008], a common situation among those with FASD. Important aspects of care include ensuring access, coordination and continuity of care, and helping parents (and professionals) learn about guardianship and financial issues, advocacy, and major systems of care. Also important are understanding and taking into account the diversity of family structure, and issues related to minority and immigrant status. McGinty et al. acknowledge the many ways a family, including the family as a whole as well as parents and siblings individually, can be stressed when the presence of a family member with comorbid mental health problems and developmental disabilities creates renegotiation of family roles, marital problems, financial stress, career concessions, social support limitations, and a reverberating impact on the larger extended family.

Interventions for Families Raising Children with Developmental Disabilities

Bailey [2007] notes that disabilities research has produced mixed findings regarding the benefits of a wide variety of formal programs of family

support and intervention. However, data (largely with mothers) do show that appropriate training, support, and help-giving practices can improve parental interaction styles, decrease depressive symptoms, and other forms of psychological distress, and improve self-efficacy beliefs and other important family outcomes. Data from multiple studies reveal that families who participate actively in problem-solving and stress management have more positive outcomes [e.g., Kim et al., 2003].

Singer et al. [2007] conducted a meta-analysis of established parenting and stress management interventions for parents of children with developmental disabilities. This informative study revealed, among other findings, that multiple component interventions addressing both parental well-being and behavioral parent training were clearly more effective than either parent training or cognitive behavioral training alone. There was support for the assertion that there are established evidence-based interventions that reduce psychological distress for parents raising children with developmental disabilities, at least in middle class mothers and over the short-term. Parent behavioral training was found to have small collateral benefits in reducing depressive symptoms in mothers. Singer et al. called for future research that measures family-level variables and also parenting self-efficacy, and assesses positive adaptation (rather than simply reduction in negative factors) as an outcome. Singer et al. also called for replications with more diverse groups of parents and longer-term follow-up. These are important points for intervention planning in the field of FASD.

A CONCEPTUAL FRAMEWORK: JOINING A DEVELOPMENTAL SYSTEMS PERSPECTIVE WITH A FAMILY SYSTEMS APPROACH

A conceptual framework can help to make sense of what is known so far about FASD and the family from informal literature and systematic research, and ideas sparked by data from the more general field of developmental disabilities. A framework can guide future research and intervention planning. Presented here is a conceptual framework that joins a developmental systems perspective with a family systems approach. This framework was based originally on a developmental systems model created by Guralnick [2001] for early intervention with children who have special cognitive and physical needs, and then applied to early intervention for FASD

by Olson et al. [2007]. Here this developmental systems perspective is augmented through integration with a family systems approach, and extended to apply through the lifespan for those with FASD who remain in close contact with their families. Bailey [2007] encourages thinking at the level of family systems when trying to understand how families face the extraordinary challenge of raising an individual with disabilities, such as FASD. Other researchers in FASD have seen the utility of using a developmental systems approach, and examining the role of risk and protective factors on child outcomes [Rasmussen et al., 2008].

Brief Overview of the Conceptual Model

Developmental systems thinking suggests that, over time, characteristics of an individual transact with those of the caregivers (and characteristics of the family and larger ecological context). The focus is on developmental outcome as revealed in life trajectories, and on developmental influences that differ or change in importance at various stages in the lifespan. Developmental systems thinking suggests further that risk and protective factors can be identified in general and for particular populations, and that interventions should be aimed to concurrently reduce disabling individual and environmental risks while at the same time enhancing protective factors. Interventions should aim to alter systems so as to support the life path of an individual with disabilities in a positive direction over time. This can be joined with a family systems approach that suggests trajectories, influences and outcomes be measured not only at the level of the individual but also at the level of the family. Further, family systems thinking suggests that treatment should be directed toward all family members as needed (so as to impact the entire family system), and that family culture (variously defined) should be considered in intervention planning. Interventions should aim to alter systems so as to support the life path of family adaptation in a positive direction over time.

Implications of this Conceptual Framework for the Field of FASD

Using this framework, prenatal alcohol exposure can be seen as placing an individual at risk for (or creating) biological vulnerability and, therefore, biologically-based “disabling characteristics,” viewed as functional impairments such as behavior regulation problems or a general difficulty in processing more

complex information. These deficits are individually variable, can range in severity and (more importantly) the degree of adaptive support needed from the caregiver, and their trajectory can become more debilitating over the lifespan. Of course, gender, age, psychiatric comorbidities, or other individual differences, can influence how disabling characteristics are manifested. These disabling characteristics are linked in a reciprocal, transactional process with key direct or indirect “environmental risks,” such negative maternal expressed emotion or caregiver stress, and with key direct or indirect protective factors, such as a high level of emotional support for the child or positive caregiver cognitive appraisals of parenting self-efficacy. Environmental risk and protective factors can occur at the level of the individual. A family systems approach suggests that these factors can also occur at the level of the family, with risks such as limited family resources, or protective factors such as high levels of family involvement in treatment planning.

Environmental risks can be thought of in many ways, which in turn can be studied and those found to be important incorporated into effective interventions. Data from the field of FASD so far has focused on caregiver and interactional risks, and suggests that the difficulties of raising a child with FASD can disrupt parent-child interaction patterns, and can create information and resource needs for parents, threats to parent confidence, and directly lead to caregiver stress. Qualitative data in the field of FASD clearly point out that risks can vary by family type (e.g., birth vs. adoptive vs. foster).

Environmental risk factors are likely to be those caregiver, interactional, and family variables identified from study of normative development at different developmental phases, such as insecure attachment quality in infancy, lack of caregiver emotional support for the child in early childhood, inadequate parental monitoring in childhood and beyond, and unsuccessful family communication patterns in adolescence. Environmental risk factors are also likely to be those caregiver and family variables identified as especially important in more general study of developmental disabilities, such as the adequacy of family resources, especially for those with lower socioeconomic status.

An especially important caregiver-level “environmental” factor in FASD may be found in the parent’s cognitive

appraisals of their child’s level of disability and of their own caregiving responsibilities. Cognitive appraisal may be especially complicated in FASD, and include both positive and negative appraisals which change over the lifespan. Negative appraisals may occur because of confusing discrepancies between functional and chronological age, difficult-to-understand learning deficits, extended dependent living, emotional overlay because of the preventable nature of the birth defect, and more. Positive appraisals may arise from the special benefits in raising those with FASD, such as enjoyment of the many humorous situations and remarkably fresh perspective of affected individuals mentioned in the informal literature. Another important caregiver-level “environmental” risk factor identified through clinical experience with FASD is tied to the parent’s cognitive appraisals of the disabling characteristics of the individual with FASD. Caregivers may not easily recognize the affected individual’s actions as the result of neurodevelopmental disabilities, but instead appraise these actions negatively as the result of willful disobedience. Therefore caregiver reactions may be mismatched to the actual situation.

There can also be general and unique protective factors. Building on the work of Perry [2005], who created a model of stress in families with developmental disabilities, protective factors might be seen as “resources” and “supports.” More general caregiver protective factors might include the personal resources of the parent’s level of optimism and sense of parenting efficacy, and linkage to informal or formal supports. Protective factors unique to FASD might include the personal resources of the parent’s knowledge of FASD, use of specialized parenting practices appropriate to FASD, and use of advocacy skills particular to this disability condition.

Family-level variables may influence outcome, and intervention can be tailored according to different family characteristics. In the field of FASD, family “type” is significant, because birth, foster, kinship, and nonkinship adoptive families appear to respond differently to the presence of a child with FASD. Family size and composition (e.g., families raising only one affected child vs. those with multiple affected family members) are factors that also seem to be important. For instance, attention to a child with FASD may negatively impact the function of non-

exposed siblings. As another example, a large family size (typically thought of as a risk factor in developmental research for those with typical development) may actually be protective if an affected child is being raised with other affected siblings so that peer support and accumulated family resources are available. Family adaptation over time is also important. There are different considerations when a family first discovers they have a child with FASD versus when parents are growing older and must consider whether their young adult with FASD can be emancipated. In addition, family disruption caused by past or current parental substance abuse leads to changes in family adaptation over time. Socio-cultural interpretations of disability and perceived impact of an affected individual's deficits, and what families see as culturally-relevant treatments, are factors that may also be important.

INFORMING FUTURE RESEARCH IN THE FIELD OF FASD

Systematic Data-Gathering

At this early stage in empirical study of FASD, systematic data-gathering has focused on the negative impact of individuals with FASD. Research has only begun to touch on the positive characteristics of these individuals and "protective" influences of caregivers and the family environment, and to study those over the lifespan. Future research should specifically examine both the "common benefits" and "special benefits" of parenting someone with FASD, and how these can be enhanced. Also important to investigate are the potentially protective influence of caregivers' positive cognitive appraisals.

Systematic analysis of qualitative data, and use of anthropological research methods, seem a productive source of information to guide intervention development. So far parents have been interviewed and their answers subjected to qualitative thematic analysis. But there is also need to understand the lived experiences of the affected individuals themselves, and of non-exposed siblings, and extended family members such as foster or adoptive grandparents, all of whom find their lives permanently changed by a family member with FASD, and who may identify benefits that can be enhanced or what they need for support.

Developmental systems research is much further advanced in the more general study of developmental disabilities, and in a related population termed

"children of alcoholics" which may provide further ideas for research in the field of FASD [Leonard and Eiden, 2007]. But studies of FASD (and children born prenatally alcohol exposed) are beginning to trace the transactional influences between the affected individual and his or her environment. The pioneering infancy and early childhood studies of O'Connor et al., including Paley et al., are leading the way. Developmental systems research that also measures family-level variables, defines stress and life trajectories, and involves longer-term follow-up, is sorely needed. This type of research can lay the foundation for discovering how to target interventions in a time-sensitive manner, and identify pivotal "turning points" in development.

Only two studies so far have described the longer-term life trajectories of individuals with FASD [Streissguth et al., 2004; Spohr et al., 2007]. Both studies have limitations and lack a family focus. These longer-term natural history and clinical follow-up studies require replication, with both a broader representation of the FASD population and comparison to other disability groups. Comparison studies will set in context the alarming data documenting the high prevalence of secondary disabilities among those with FASD. Comparison studies are also needed because different developmental disabilities likely have varying life trajectories, and thus correspondingly different trajectories for the stress and support needs of caregivers and affected individuals.

What data reflect so far is the chronic nature of functional limitations among individuals with FASD, including the high degree of adaptive behavior support needed by this group and common secondary disabilities. Qualitative data so far reveal that parents raising persons with FASD must contend with the possibility that they will be "parenting" for the rest of their lives, and must plan for the future even past their own old age. Future research on long-term function is needed to replicate what seems to be especially diminished family functioning in this disability group. Future research must study what appears to be the crucial importance to positive adaptation of the qualities of parent and family flexibility and resilience.

At this time, data-gathering in the field of FASD has not really been focused at the broader level of the family system. Empirical studies have taken only a beginning look at characteristics and influences within, for example, dif-

ferent family structures, despite professional opinion that different dynamics operate within birth, kinship, and non-kinship adoptive families, so that different help-giving strategies would be important for each. Given the dramatic demographic diversity of families raising children with FASD, analysis at the level of different family variables, and certainly analysis of different cultural and sociocultural contexts, is bound to be complex and will ultimately require much larger samples. But ethnographic and narrative analysis approaches to measurement, and the use of meaningful family-level measures such as those focused on family quality of life, can be used now with the smaller samples at hand. In the field of FASD, it will be important to remember to look not only at ethnicity as a measure of culture, but at the positive and negative impacts on family function of experiencing other "types" of culture. One may be the "culture" of disability in a world that is not always accepting of the need for structure or protection by parents even as children move into adolescence and adulthood. Another may be the "culture" of traditional alcohol use in a world that may challenge the appropriateness of raising abstinent children.

Data-gathering in the field of FASD has also so far been largely limited to caregiver report, rather than direct observation of naturalistic caregiver-child interaction or family interaction patterns which has been so productive in the study of other childhood conditions. The work of Paley et al. [2005], in which a caregiver-child task was observed during a home visit and the important developmental construct of "supportive presence" was coded, is a promising early step in direct observation.

Designing and Refining FASD Interventions that Involve the Family

It is known that good quality caregiving and stability of the home environment are vital to successful outcome of those with FASD. It has been observed clinically that caregivers (and families) are the primary advocate for affected individuals across the lifespan. The question for intervention planning is how to make this happen in the most effective way.

There are already a number of very useful manuals and books, written by parents and professionals, suggesting parenting and family interventions for children with FASD (see FASD Center for Excellence website). Most of these

have been based on expert opinion and clinical experience, since little systematic intervention research exists. There are even general intervention guidelines, and FASD educational curricula including comments about intervention, available from national resources such as the Centers for Disease Control and Prevention [Bertrand et al., 2004; FASD Regional Training Centers Curriculum Development Team, 2009]. Data reviewed here, informed by the conceptual framework in this article, can be instructive in further developing these clinically-based interventions, and advising caution about treatments that may not have a sufficient evidence base.

In more general study of developmental disabilities, it is the affected individual's mental health and behavior problems that have central and lasting effects on family adaptation and parental well-being, and on measures of negative caregiver impact. Available data emphasize that many children with FASD, who present with concerning behavior problems, also meet criteria for multiple psychiatric conditions. As Paley et al. demonstrate, externalizing behavior is associated with caregiver stress among children who are alcohol-exposed, and appears more important than their diagnosis of FAS vs. ARND. In the field of FASD, then, interventions focused on reducing target symptoms and improving externalizing behavior appear essential, and several CDC-funded interventions recently tested appear to accomplish just this.

One direction for design of future interventions may be to build on positive characteristics already shown by children with FASD, such as reinforcing children's existing tendency to seek and willingly receive (and even value) help from their caregivers. Another direction for treatment design is to emphasize for parents the various types of "positive" benefits of raising the individual with FASD, helping to promote positive parenting views, and positive cognitive appraisal especially for those caregivers experiencing the highest levels of stress.

Based on the current literature examining FASD, in both lower-risk and high-risk samples of children born alcohol-exposed, it is child-related stress (rather than parenting role-related stress or life stress) that is the most salient developmental influence, and related to adverse child behavioral outcome. Interventions in the field of FASD should be directed toward management of this type of stress. The more general study of developmental disability suggests the

potential efficacy of collaborative problem-solving and stress management interventions for caregivers, especially if deployed as a multicomponent intervention in combination with specialized behavioral parent training. Particularly suggested is the use of cognitive behavioral techniques with caregivers of individuals with FASD, to promote understanding of and positive coping with their children's functional impairments and positive cognitive appraisal of their caregiving responsibilities. Data so far in the field of FASD suggest that behavioral parent training may need to focus on different types of parenting practices than typically used with children showing externalizing behavior, such as skills of positive behavior support planning which take into account the neurological impairment that co-occurs with the challenging behavior of children with FASD. Interventions designed with these principles in mind are now being tested with families raising especially challenging children with FASD [Olson et al., 2005; ICFRC, 2009].

Ultimately these types of intervention should be aimed at the central goal of increasing parents' sense of self-efficacy and internal locus of control in handling the often unpredictable or challenging behavior of their children with FASD, which may mediate the impact of perceived social support. The literature on traumatic brain injury, a useful comparative literature if FASD is construed as a "developmental brain injury," also emphasizes positive caregiver coping skills, good psychological well-being, strong family relationships, and a positive caregiver belief system and finds that these have a strong relationship with positive family functioning and reductions in child-related strain [Rivara et al., 1996]. The more general developmental disabilities literature, shows the importance of family involvement and family-centered care, emphasizes the central and active role of the parent (with more highly involved families showing better outcome), and reveals the need for collaborative consultation. Specific topics for caregiver education can be found in the more general developmental disabilities literature. All this is instructive for intervention development in the field of FASD.

Caregiver stress might be reduced by promoting parental self-care, or providing families with respite care (and help in providing the documented, more intensive support children with FASD require for adaptive function)

that is so often lacking. Because families raising children with FASD frequently endorsed an unmet, important need to connect with other parents, promoting the use of informal support networks can be an intervention in itself. Informal support is strongly related to positive family adaptation in the general developmental disabilities literature, and there already exists a vibrant FASD-specific parent support movement, slowly becoming global, that can be accessed. Based on general developmental disabilities and FASD-specific data so far, in higher-risk samples, it is simultaneously important to address the need for family resources, and to minimize other sources of environmental stress.

Even in the lower-risk caregiver sample in new data presented here, a sizable percentage of parents endorsed clinically significant psychological distress or depression. A frequently endorsed unmet, important family need in these new data was for reassurance about negative emotions. Psychological distress should likely be an intervention target and provision of emotional support an intervention method. The more general study of developmental disabilities provides some promise that caregiver distress can be successfully reduced. This is important. In the study of traumatic brain injury, caregiver psychological distress is related to other negative stressors [Wade et al., 2006]. In the study of young children with conduct disorders, who similarly to children with FASD show high levels of externalizing behavior, maternal depression is one important moderator of treatment effects [Beauchaine et al., 2005].

Different family types have been demonstrated to have different needs. Interventions should be geared to provide what foster, adoptive, birth, or other types of families need. Foster parent training, for example, often emphasizes the positive and life-changing experience of fostering and reinforces other motives for being a foster parent, but may also need to provide the supports found in qualitative data that foster parents say are needed to promote successful placements and prevent disruptions. Birth parent intervention, as another example, should likely respond to the issues found in qualitative data to be specific to this population, such as responding to the emotions raised by being blamed or feeling abandoned by health care professionals or the educational system.

Unfortunately, the "double jeopardy" of psychosocial risk coupled with

FASD is quite common, there is a poorer developmental trajectory in higher-risk samples, and longer-term data show especially diminished individual and family functioning in the population of those with FASD. This means that interventions should likely be intensive and lasting, even including help for families who must look forward to providing care for those with FASD who may not be able to be emancipated but require dependent living. A full continuum of services should be available across the lifespan.

Momentum for the future

Data-gathering and intervention development in the field of FASD should be increased. It is a societal imperative to respond to the “invisible epidemic” of fetal alcohol spectrum disorders affecting so many around the world. ■

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