

A Renewed Call to Action: The Need for Systematic Research on Interventions for FASD

Heather Carmichael Olson

IT IS NOW generally accepted that fetal alcohol spectrum disorders (FASD) comprise a set of lifelong neurodevelopmental disabilities, and that prenatal alcohol exposure (PAE) can lead to wide-ranging, pervasive deficits. Despite many individual strengths and positive characteristics, a major concern is that those with FASD commonly display clinically significant behavior problems in childhood and adolescence. Over the life span, individuals with FASD also show high rates of many “secondary disabilities,” such as psychiatric conditions, substance use, and other troubling problems in daily function. In addition, their families have important unmet needs, and there are many barriers to accessing care and resources through service systems.

There is hope. Around the world, public and professional awareness has grown regarding the negative impact of PAE and FASD. This is partly because epidemiologic studies have revealed FASD prevalence rates estimated as high as 2 to 5% from in-school studies (May et al., 2009), and efforts are now under way by entities such as the World Health Organization to assess FASD prevalence. In many countries, the work of parent support and advocacy organizations, governmental organizations, scientists, and international research collaborations have, together, brought increased understanding. But the need for action has truly grown because of the voices of families who face the challenge of FASD, every day, in their own homes, around the world. Simply put, these families want to know what to do.

FASD AND THE EFFECTS OF PAE: AN INTERVENTION RESEARCH GAP

Over the years, research has been carried out internationally to describe teratogenic mechanisms of alcohol,

From the Department of Psychiatry and Behavioral Sciences (HCO), University of Washington School of Medicine, Seattle, Washington; and Seattle Children's Research Institute (HCO), Seattle, Washington.

Received for publication June 1, 2016; accepted June 6, 2016.

Reprint requests: Heather Carmichael Olson, PhD, Department of Psychiatry and Behavioral Sciences, University of Washington School of Medicine and Seattle Children's Research Institute, West 8th Building, 2001 Eighth Avenue, Suite 400, Seattle, WA 98121; Tel.: 206-987-3810; Fax: 206-987-2246; E-mail: heather.carmichaelolson@seattlechildrens.org

Copyright © 2016 by the Research Society on Alcoholism.

DOI: 10.1111/acer.13150

characterize the biobehavioral impact of PAE, and refine clinical understanding of FASD. Several diagnostic systems have been created, and are evolving and being systematically compared. Proposed criteria now in the DSM-5, although in need of empirical validation, make identification possible within the mental health system, using the new term “neurobehavioral disorder associated with prenatal alcohol exposure (ND-PAE)” (American Psychiatric Association, 2013, p. 798). But identifying mechanisms, describing and diagnosing affected individuals are not enough. As noted in a 2009 monograph titled “A Call to Action: Advancing Essential Services and Research on Fetal Alcohol Spectrum Disorders,” written by members of a national task force in the United States (Olson et al., 2009), it is also vital to take steps to “establish effective interventions,” “translate interventions to the community,” and “improve the quality and utilization of interventions in all service systems for those with FASDs” (p. 2).

INTERVENTION RESEARCH SO FAR

There have certainly been intervention-related research advances not focused on behavioral intervention, such as the study of neuroprotective agents, designed to alter or ameliorate teratogenic alcohol effects. There have been efforts to find biomarkers and methods of early identification in the hope that the benefits of neuroplasticity can be maximized. There are thought-provoking papers discussing this research (e.g., Kodituwakku and Kodituwakku, 2011; Warren et al., 2011).

But the focus of this commentary is on progress in clinical-level, human behavioral intervention studies with affected individuals, families, and systems. Starting in the 1990s, “lived experience” and other qualitative data have been gathered as a basis for behavioral intervention research. In a far-seeing move in 2001, the Centers for Disease Control and Prevention (CDC) in the United States funded an early cooperative research group of intervention trials for preschool and school-aged children with FASD, and their caregivers. These well-designed randomized controlled trials (RCTs) were followed by a second round of community efficacy trials. Findings were that these tailored interventions, monitored for fidelity, and targeted to the deficits of children with FASD, resulted in measurable outcome improvements. Findings also revealed these interventions could be implemented as value-added

treatments within the context of other community services. Study results are being published over time and have led to organized dissemination efforts and new intervention research trials (see summary of early findings in Bertrand, 2009).

In 2007, the research organization called the FASD Study Group focused their annual meeting on the topic of intervention, signaling growing international professional scientific interest. In 2009, in the United States, the National Institute on Alcohol Abuse and Alcoholism funded several innovative intervention studies, with findings just now being published. During these years, through the FASD Center for Excellence in the United States, the Substance Abuse and Mental Health Services Administration (SAMHSA) funded “service to science” efforts to operationalize existing clinical interventions. Over time, innovative intervention studies have also been initiated through international consortia and elsewhere in the world, especially in Canada, and as far afield as South Africa and Australia.

In 2009, Peadon and colleagues carried out a systematic FASD intervention review, finding very limited data, methodological limitations, often small samples, and short-term follow-up (Peadon et al., 2009). Paley and O’Connor (2011) reviewed FASD behavioral interventions, finding some promising evidence but arguing for larger sample sizes, examining factors modifying treatment response, and follow-up to detect sustained effects. In 2015, Reid and colleagues carried out another systematic review of FASD interventions, across the life span (Reid et al., 2015). They found a heartening increase in the number of intervention studies and use of rigorous methods and growing evidence for interventions that measurably improved outcomes in early to middle childhood. But there were still many research gaps. The authors expressed concern about limited research focusing on young children with PAE, given the promise of early intervention. They noted a distinct lack of interventions for adolescents and adults with FASD, who show a widening developmental gap and increased risk for accumulating secondary impairment. They found wide variability in methodological rigor and follow-up time frame. The authors advocated use of intervention frameworks (discussed below) to create future interventions that address multiple domains of function for those with FASD.

Starting in the late 1990s, varied publications on recommended best practices for what has been called “FASD-informed care,” and policy perspectives, have been produced by experts in the field. A pivotal contribution in the United States, containing a consensus of expert advice, is a “Treatment Improvement Protocol (TIP #58)” called “Addressing Fetal Alcohol Spectrum Disorders” (SAMHSA, 2014). This free, downloadable monograph aims to show substance abuse and mental health agencies and clinicians both how to provide and (importantly) effectively implement FASD-informed care.

A RENEWED AND VIGOROUS CALL TO ACTION

Clearly, FASD intervention research has advanced. What is important now is to build on existing data and consensus of expert opinion, and generate a body of evidence-based practices (EBPs) for FASD-informed care that are effectively carried out in real-world settings. This commentary is a renewed and spirited call to action—a call to focus intensively and strategically on intervention research and implementation. What can help propel research forward is to understand the important research questions to be explored,

Table 1. Important Questions for Intervention Research on FASD and the Effects of PAE

Defining the scope of the problem at different developmental stages	
	<ul style="list-style-type: none"> • What are the adverse outcomes of individuals with FASD or the effects of PAE, and of their families and service systems? • What do needs assessments reveal? • What are universal and population-specific risk factors? • What are population-specific protective factors that lead to more positive outcomes? • What is the lived experience of individuals and families with FASD across multiple domains (from Reid et al., 2015)? • What mechanisms appear to underlie adverse outcomes?
Defining an efficacious response to the problem at different developmental stages	
	<ul style="list-style-type: none"> • How can adverse outcomes be prevented? • What interventions measurably improve outcomes for individuals affected by FASD, or the effects of PAE, and their families? • How can existing evidence-based interventions (designed for other clinical populations) be adapted to be efficacious? • What innovative, tailored interventions must be developed? • Are interventions feasible? • How should interventions be targeted, and who needs less intensive or more intensive intervention? • Are multicomponent interventions necessary, especially given the complexity of life circumstances (and ecological context) for many with FASD, or the effects of PAE? If so, what components should be linked together?
Ensuring interventions become evidence-based practices	
	<ul style="list-style-type: none"> • How can interventions be carried out with fidelity, and how can fidelity be monitored? • How can promising or efficacious interventions be replicated and validated using high-quality designs to generate a convincing evidence base? • How can a broad and generalizable evidence base be generated?
Ensuring translation to the community and improving implementation, quality, and utilization	
	<ul style="list-style-type: none"> • How can interventions be implemented appropriately (according to the model and with fidelity) in the community? • How can interventions be implemented to actually change and improve provider practice (e.g., transfer and maintain interventions in “real-world” settings)? • What interventions are effective in “real-world” practice, and how does this compare to their efficacy in “ideal” practice? • How much can interventions be streamlined and remain effective? • What adaptations are needed to suit interventions to the varying characteristics of setting, community, culture, and subgroup of the population to be treated?
Adapting and creating service systems across the life span	
	<ul style="list-style-type: none"> • How can existing service systems be adapted, or linked together, to better address the needs of those with FASD, or the effects of PAE, and their caregivers? • What new service systems must be created? • How should individual communities organize their network of services?

and to learn from exemplary research programs and proposed frameworks for intervention development.

IMPORTANT INTERVENTION RESEARCH QUESTIONS

Table 1 lists research questions vital to advance intervention research on FASD and the effects of PAE. Domains include: (i) defining the scope of the problem at different developmental stages; (ii) defining an efficacious response to the problem at different developmental stages; (iii) ensuring interventions become EBPs; (iv) ensuring translation of interventions to the community and improving implementation, quality, and utilization; and (v) adapting and creating service systems across the life span.

LEARNING FROM AN INSTRUCTIVE RESEARCH PROGRAM

One instructive example of programmatic intervention research addressing several developmental phases is that led by Mary O'Connor, PhD.

Starting in the 1980s, O'Connor and colleagues carried out developmental systems research to explore how the parent–infant relationship was affected by maternal drinking and depression in transaction with the impact of PAE (and genetic predispositions). In longitudinal study, they further examined how development of these children with PAE unfolded into the preschool years. Studying a sample of children born to moderate to heavy drinking birth mothers at low psychosocial risk, findings were of a complex developmental process, with final analyses suggesting that the neurodevelopmental effect of PAE was salient (with other factors) to the emergence of childhood depression. Additional research with a sample at higher psychosocial risk replicated these findings, but revealed an even greater risk of emerging internalizing symptoms. O'Connor and her team also described possible underlying mechanisms, highlighting intervention possibilities, including the utility of helping young children modulate behavior and alter possibly negative self-cognitions (see review in O'Connor, 2001; Olson et al., 2001).

Aware that young children affected by PAE commonly showed behavioral difficulties with social skills deficits and peer interaction problems, O'Connor and her team initiated an RCT of an existing social skills intervention, called the Children's Friendship Training (CFT) program, as part of the CDC cooperative research group. Intervention adaptations were made based on literature describing neuropsychological impairments of children with FASD (Laugeson et al., 2007). This model targeted the developmental phase of elementary school and included both child-focused friendship training and caregiver education components. Compared to a delayed treatment control, for children with PAE the CFT intervention had immediate and sustained positive, measurable effects on social knowledge and skills, and problem behavior (O'Connor et al., 2006). There was also

improvement in some hostile attributions, an aspect of social information processing (Keil et al., 2010). In a subsequent community translation trial, the CFT intervention was found more effective than a community-based social skills intervention and equally effective for children with PAE than those without (O'Connor et al., 2012).

Thinking about the next life phase, and understanding that adolescents with FASD are frequently at high risk for substance use problems for genetic, familial, and/or complex teratogenic reasons, O'Connor initiated treatment development of Project Step Up (O'Connor et al., 2016). This is a manualized intervention for adolescents with FASD using a harm reduction approach and adapted for neurocognitive deficits, with an adjunct caregiver education program. The authors report this intervention was feasible, satisfactory, did not appear to have iatrogenic effects, and, compared to a well-matched control group, resulted in measurable outcome improvements. Via Project Step Up, O'Connor and her team have begun to address the concerning gap in interventions suitable for adolescents with FASD (or affected by PAE).

The intervention models and methods developed by O'Connor and her colleagues, detailed in the publications just discussed, are responsive to many of the questions listed in Table 1. In O'Connor's work, commonly occurring areas of impairment appropriate to each developmental stage are targeted. Theoretically, if these deficits are ameliorated, over the life trajectory outcome improvements can enhance protective influences and reduce risk—with reverberating positive effects on other functional domains. The intervention methods of O'Connor and her team are feasible, include multiple components, and are comprised of adaptations of existing interventions combined in innovative ways. Adaptations they have identified stimulate ideas for how to adapt existing EBPs and create innovative new interventions. In analysis, O'Connor and her team have identified possible mechanisms underlying behavioral change (such as change in hostile attributions), which also lead to new intervention ideas.

FRAMEWORKS TO GUIDE RESEARCH

Several frameworks have been proposed to guide intervention development research for FASD. Petrenko and colleagues (2014) recently published a "Lived Experience Model" to guide intervention development, based on systematic qualitative research with providers and family members. This model illustrates needs across multiple intervention "targets" (person with FASD, family members, systems of care), and across developmental periods. Five overarching themes useful in shaping interventions are derived, such as the need for life span services and a preventive/proactive intervention stance. Kodituwakku and Kodituwakku (2011) proposed an important integrative "dynamic interactive framework," congruent with recent work on neuroscience, for intervention development. This framework suggests that a successful behavioral intervention can be seen as "planned and guided experiences that give rise to a chain of reciprocal

interactions between neural activities and structures, ultimately resulting in improved performance” (Kodituwakku and Kodituwakku, 2011, p. 217). These authors argue that, for children with FASD, individualizing interventions based on an understanding of children’s individual differences is vital. They further argue for multicomponent, sufficiently comprehensive interventions combining parent support and direct child treatment, emphasizing training on self-regulation (among other skills), adding psychopharmacologic treatment when needed, and perhaps considering developmental therapeutics (such as nutritional supplements) in the future.

REPLICATION, TESTING AVAILABLE EVIDENCE-BASED PRACTICES, AND DEVELOPING INNOVATIVE TREATMENTS

As interventions are developed in the field of FASD, a primary research focus should be independent replication. This is mandatory to acquire the evidence needed to move from the level of what is called a “promising practice,” to the level of a “well-established” EBP. EBPs can be accepted for governmental and funding source registries. Generating an appropriate evidence base for FASD interventions is absolutely necessary to support translation of interventions to the community and ensure their utilization. Another research focus should be exploration of existing EBPs to see whether they are (or are not) as effective as expected for those with PAE, and how they should be adapted. If there are no existing interventions for a particular developmental stage or outcome, a third research focus should be development of innovative, tailored treatments, as in the case of Project Step Up for adolescent substance use prevention.

THE VITAL IMPORTANCE OF IMPLEMENTATION SCIENCE

In carrying out intervention research regarding FASD, and the effects of PAE, it is crucial to learn from the burgeoning field of implementation science (e.g., Bauer et al., 2015). This involves study of the actual process of implementing evidence-based programs and practices, where efficacy has already been established. In some countries, this is referred to as the practice of “knowledge transfer.” The TIP #58 monograph on FASD describes a strategic approach to implementation for FASD-informed care at the agency level.

In FASD research, not only should interventions be translated to real-world practice, but the actual process of intervention adoption and implementation should explicitly be studied through formative evaluation. Implementation aims are generally to encourage intervention adoption, maintain fidelity and effectiveness, adapt interventions to function efficiently in real-world settings, and make interventions sustainable ongoing. Progress on these aims should be assessed. O’Connor’s scientific work, for example, has included

assessment of implementation parameters, such as fidelity, effectiveness, and (vital to sustainability) client/provider satisfaction.

When studying implementation, it is valuable to explicitly ascertain what components are core to the intervention versus what is the “adaptable periphery,” to find out how interventions can be streamlined. Research can explore how best to implement interventions in the settings and existing service delivery systems in which they will be deployed. This means understanding how to accomplish individual provider behavior change within the context of organizational change, and how to successfully carry out the actual implementation process (which involves discernable subprocesses) (Damschroder et al., 2009). The field of implementation science has important study design and methods to offer, such as “hybrid effectiveness-implementation trial designs,” where both health outcomes and implementation strategies are simultaneously examined (Bauer et al., 2015, p. 8).

In conclusion, there is great need to effectively and sustainably treat the effects of PAE, and FASD, across the life span and within the complex ecological context in which these conditions occur (Reid et al., 2015). Documented for FASD are high levels of caregiving stress, frequent life struggles for affected individuals, and high social and economic costs for societies around the world. Resources are limited, so systematic intervention research is required. This renewed call to action provides strategic research guidance and is meant to help the voices of families, asking for treatment, to be heard.

ACKNOWLEDGMENTS

The writing of this commentary was supported by the Seattle Children’s Research Institute and University of Washington through salary support. Sincere appreciation is extended to Christie Petrenko, PhD, for valuable assistance in reviewing and editing this manuscript.

CONFLICT OF INTEREST

The author has no conflict of interest to report.

REFERENCES

- American Psychiatric Association (2013) *Diagnostic and Statistical Manual of Mental Disorders*. 5th ed. American Psychiatric Association, Washington, DC.
- Bauer MS, Damschroder LJ, Hagedorn H, Smith J, Kilbourne AM (2015) An introduction to implementation science for the non-specialist. *BMC Psychol Open Access* 3:32.
- Bertrand J; Interventions for Children with Fetal Alcohol Spectrum Disorders Research Consortium (2009) Interventions for children with fetal alcohol spectrum disorders (FASDs): overview of findings for five innovative research projects. *Res Dev Disabil* 30:986–1006.
- Damschroder LJ, Aron DC, Keith RE, Kirsh SR, Alexander JA, Lowery JC (2009) Fostering implementation of health services research findings into practice: a consolidated framework for advancing implementation science. *Implement Sci* 4:50.

- Keil V, Paley B, Frankel F, O'Connor MJ (2010) Impact of a social skills intervention on the hostile attributions of children with prenatal alcohol exposure. *Alcohol Clin Exp Res* 34:231–241.
- Kodituwakku PW, Kodituwakku EL (2011) From research to practice: an integrative framework for the development of interventions for children with fetal alcohol spectrum disorders. *Neuropsychol Rev* 21:204–223.
- Laugeson EA, Paley B, Schonfeld AM, Carpenter EM, Frankel F, O'Connor MJ (2007) Adaptation of the Children's Friendship Training program for children with fetal alcohol spectrum disorders. *Child Family Behav Therapy* 29:57–69.
- May PA, Gossage JP, Kalberg WO, Robinson LK, Buckley D, Manning M, Hoyme HE (2009) Characteristics of FASD from various research methods with an emphasis on recent in-school studies. *Develop Disabil Rev* 15:176–192.
- O'Connor MJ (2001) Prenatal alcohol exposure and infant negative affect as precursors of depressive features in children. *Infant Mental Health J* 22:291–299.
- O'Connor MJ, Frankel F, Paley B, Schonfeld AM, Carpenter E, Laugeson EA, Marquardt R (2006) A controlled social skills training for children with fetal alcohol spectrum disorders. *J Consult Clin Psychol* 74:639–648.
- O'Connor MJ, Laugeson EA, Mogil C, Lowe E, Welch-Torres K, Keil V, Paley B (2012) Translation of an evidence-based social skills intervention for children with prenatal alcohol exposure in a community mental health setting. *Alcohol Clin Exp Res* 36:141–152.
- O'Connor MJ, Quattlebaum J, Castaneda M, Dipple KM (2016) Alcohol intervention for adolescents with fetal alcohol spectrum disorders: Project Step Up, a treatment development study. *Alcohol Clin Exp Res* doi: 10.1111/acer.13111
- Olson HC, O'Connor MJ, Fitzgerald HE (2001) Lessons learned from study of the developmental impact of parental alcohol use. *Infant Mental Health J* 22:271–290.
- Olson HC, Ohlemiller MM, O'Connor MJ, Brown CW, Morris CA, Damus K; National Task Force on Fetal Alcohol Syndrome and Fetal Alcohol Effect (2009) A Call to Action: Advancing Essential Services and Research on Fetal Alcohol Spectrum Disorders—A report of the National Task Force on Fetal Alcohol Syndrome and Fetal Alcohol Effect. Available at: <http://www.cdc.gov/ncbddd/fasd/documents/calltoaction-textonly.pdf>. Accessed July 5, 2016.
- Paley B, O'Connor MJ (2011) Behavioral interventions for children and adolescents with fetal alcohol spectrum disorders. *Alcohol Res Health* 34:64–75.
- Peadon E, Rhys-Jones B, Bower C, Elliot EJ (2009) Systematic review of interventions for children with fetal alcohol spectrum disorders. *BMC Pediatr* 9:35.
- Petrenko CLM, Tahir N, Mahoney EC, Chin NP (2014) A qualitative assessment of program characteristics for preventing secondary conditions in individuals with fetal alcohol spectrum disorders. *J Populat Therapeut Clin Pharmacol* 21:e246–e259.
- Reid N, Dawe S, Shelton D, Harnett P, Warner J, Armstrong E, LeGros K, O'Callaghan F (2015) Systematic review of fetal alcohol spectrum disorder interventions across the lifespan. *Alcohol Clin Exp Res* 39:2283–2295.
- Substance Abuse and Mental Health Services Administration (SAMHSA) (2014) Addressing Fetal Alcohol Spectrum Disorders (FASD). Treatment Improvement Protocol (TIP) Series 58. HHS Publication No. (SMA) 13-4803. Substance Abuse and Mental Health Services Administration, Rockville, MD.
- Warren KR, Hewitt BG, Thomas JD (2011) Fetal alcohol spectrum disorders: research challenges and opportunities. *Alcohol Res Health* 34:4–14.