Introduction to the Special Section: Psychological Outcomes of Pediatric Conditions That Affect the Central Nervous System

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Large numbers of children and adolescents have conditions that affect the CNS and disrupt normal brain development due to genetic and environmental factors (e.g., fetal alcohol syndrome), trauma, or chronic illness (Yeates, Ris, Taylor, & Pennington, 2010). Such problems have multifaceted effects on the child’s cognitive, emotional, and social development, health-related quality of life (HRQOL), and on family functioning. Pediatric psychologists provide assessment and interventions to children and adolescents with these conditions and their families and conduct research that characterizes the impact of these problems. Such research provides important data that describe the impact of various neurological conditions, including the role of risk and protective factors on psychological outcomes. These data are critical in targeting interventions to reduce the risk for a wide range of problematic psychological outcomes. Despite the importance of such research for enhancing the psychological outcomes of children and adolescents with conditions that involve the CNS, this work has been underrepresented in published work in the Journal of Pediatric Psychology. This special section was invited to address this need. Taken together, the contributions to this special section have advanced scientific knowledge concerning the psychological outcomes of children and adolescents with conditions that involve the CNS. The purpose of this introduction is to summarize key points and consider the implications for future research.

Socioemotional Functioning of Children with Neurofibromatosis

Martin et al. (2012) described the relationship of cognitive, illness, and environmental variables to the socioemotional functioning of children with neurofibromatosis type 1 (NF-1), a genetic disorder characterized by neurofibromas that result in frequent CNS impairments and multiple cognitive deficits (Tonsgard, 2006). Although children and adolescents with NF-1 are at risk for a range of socioemotional problems such as Attention Deficit Hyperactivity Disorder that affect their social and emotional functioning, relatively little is known about this outcome (Graf, Landolt, Mori, & Bolshhauser, 2006).

To address this need, Martin and colleagues (2012) studied 53 children (mean age of 12 years) with the Behavioral Assessment System for Children-Second Edition. Based on parent ratings, almost one-third of the sample had internalizing problems in the at-risk or clinically significant range, but only 11% for externalizing problems. Teacher ratings based on the Behavioral Assessment System for Children-Second Edition yielded comparable frequencies of risk across symptom domains: 22% for internalizing problems and 15% for externalizing problems. There was substantial individual variation in the socioemotional functioning of children with NF-1. Based on a socioecological predictive model (Kazak, 1989), lower cognitive functioning, greater disease severity, and more frequent stressful life events were associated with less adequate socioemotional functioning. Findings have potential implications for targeting interventions for this population.

Methodological limitations involve the relatively small sample size and uncontrolled, cross-sectional study design. Future studies will need to address important but as yet unanswered questions such as: (1) what specific neurological findings or neuropsychological variables affect various domains of socioemotional functioning in NF-1? (2) what are the mediators of the relationship among cognitive,
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Impact of Fetal Alcohol Spectrum Disorders on Cognitive Functioning

Rasmussen, Kully-Marten, Pei, & Job (2012) described the results of a controlled study on the cognitive functioning of children with histories of fetal alcohol syndrome spectrum disorders (FASD). Although FASD is a leading preventable cause of intellectual disability and associated cognitive, educational, and behavioral problems that necessitate costly services (Stade, Unjar, Stevens, Beyene, & Koren, 2006), this condition has received very little attention in published research in pediatric psychology.

To address this need, Rasmussen and colleagues (2012) used experimental tasks to assess recognition memory and source monitoring, defined as the ability to distinguish between internal versus external sources of memory. Children with FASD (average age of 9 years) demonstrated generalized impairments in both recognition and source memory when compared with their peers. One important implication of this study is that children with FASD demonstrated deficits not only in the quantity, but in the quality of their information processing. Thus, the central cognitive deficit in FASD appears to be much broader than general memory in that it involves integration and organization of contextual information that surrounds verbal events.

These findings have potentially useful clinical implications: The cognitive deficits identified in this study may disrupt the functional capacities of children with histories of FASD by contributing to oppositional responses to parents’ and teachers’ directions and problems in processing social communications with peers (Yeates et al., 2009). Limitations of the study that might be addressed in future research include the small sample size, use of a clinic-based sample, and failure to measure contextual variables such as the home environment that could affect the children’s recognition memory and source monitoring.

Impact of Traumatic Brain Injury on HRQOL and Intellectual Outcome Effects of Mild TBI on HRQOL

Traumatic brain injury (TBI) is a leading cause of morbidity, mortality, and deficits in cognitive, behavioral, and social functioning and HRQOL in pediatric populations (Schneier, Shields, Hostetter, Xiang, & Smith, 2006). Despite the higher prevalence of mild TBI, the outcomes of this population are not as well described as they are for moderate to severe TBI in published research in pediatric psychology. In this section, Moran et al. (2012) described HRQOL in children with mild TBI at 3 and 12 months postinjury and the prediction of postconcussive symptoms (PCS) for this outcome. Children (mean age of 12 years at point of injury) demonstrated deficits in physical, but not psychosocial HRQOL as measured by the Child Health Questionnaire at 12-month follow-up. Somatic PCS (e.g., headaches, dizziness) predicted physical HRQOL (e.g., pain and physical function).

Limitations of the study include the use of parent reports to assess PCS and HRQOL, which could inflate the effects that were identified, and low recruitment rates (less than 50%), which affects generalizability of findings. Future research priorities include studies of larger, more representative samples of children and adolescents with TBI studies over longer periods and evaluation of interventions designed to reduce the negative impact of mild TBI on HRQOL.

Impact of Timing of TBI on Intellectual Abilities

Crowe, Catroppa, Babi, Rosenfeld, & Anderson (2012) studied the intellectual abilities of children of varying TBI severity (N = 181) 2 years postinjury who were classified into four ages at injury groups (infant, preschool, middle childhood, and late childhood). This research is noteworthy for a test of the early vulnerability versus critical period hypothesis on subsequent intellectual abilities. Findings suggested that cognitive outcomes were influenced by age of injury as well as specific stage of cognitive maturation. Children with severe TBI demonstrated poorer outcomes, but age at injury also showed significant effects. Children who were injured in middle childhood demonstrated the poorest outcomes. Consistent with the double hazard hypothesis, children with more severe injuries and greater social disadvantage had the greatest deficit in nonverbal abilities.

Limitations included small samples for specific age groups, wide variation in age range and TBI severity, and absence of measures of specific cognitive domains (e.g., executive functioning). Salient recommendations for future research included examination of a wider range of abilities to clarify the range of impairments associated with TBI and assessment of MRI to identify specific TBI-related neuropathology associated with specific cognitive deficits.

disease, and environmental variables and socioemotional functioning among children and adolescents with NF-1? and (3) what specific interventions can enhance the socioemotional development of children and adolescents with NF-1?
Developmental and Cultural Influences on the Psychological and Family Outcomes of Spina Bifida

Spina bifida is a birth defect that results in malformations of CNS structures, a wide range of cognitive deficits and physical impairments (Fletcher & Dennis, 2009), and has a significant impact on parent–child relationships (Coakley, Holmbeck, Friedman, Greenley, & Thill, 2002). In this section, Wasserman, Holmbeck, Lennon, & Amaro (2012) examined the effect of pubertal timing on parent–adolescent conflict, emotional distancing, and depressive symptoms in a prospective study of children and adolescents (age 8–17 years) with spina bifida. Findings partially supported the hypothesis that the relationship between early pubertal timing and higher rates of depressive symptoms, conflict, and emotional distancing would be stronger for typically developing youth. Early pubertal timing also predicted child reported conflict, emotional distancing, and mother reported depressive symptoms for children with spina bifida and their physically healthy counterparts. One of the most important implications of the findings is the need for interventions to enhance communication and parental involvement for children with spina bifida as they transition to puberty.

Limitations included a small and racially homogenous Caucasian sample. Moreover, generalization of these findings to other pediatric conditions with CNS-related conditions, physical handicaps, or cognitive difficulties need to be established.

Devine, Holbein, Psihogios, Amaro, & Holmbeck (2012) described the individual psychological adjustment, parental functioning, and perceived social support of families of children with spina bifida (mean age of 11 years) among Hispanic and non-Hispanic mothers. Hispanic mothers and fathers reported lower levels of parental satisfaction and sense of competence, and greater perceptions of child vulnerability compared with non-Hispanic white mothers. Mothers of Hispanic children with spina bifida also reported lower levels of social support. Contrary to hypotheses, no differences were found in parents’ individual adjustment or parental protectiveness.

Limitations of the study included a relatively small sample size and no analysis of individual differences in the Hispanic sample with respect to country of origin, cultural practices, and acculturation. Opportunities for future research include the need for large sample sizes to help clarify the relationships among SES, culture, and outcomes, studying a comparison group of Hispanic families of typically developing youth, determining whether these findings are unique to parents of children with spina bifida or generalize to other chronic pediatric conditions that involve the CNS, and testing interventions that enhance parental sense of competence and reduce their worry about their children’s vulnerability.

Discussion and Future Directions

Challenges in Conducting Research on Outcomes of Conditions that Affect the CNS

Investigators who contributed to this special section faced a number of significant challenges in designing and implementing their research. The conditions that were studied were heterogeneous in the nature of their effects on the CNS and exposure to relevant family and socioeconomic influences. Such important variables are difficult to operationalize, particularly in a manner that yields samples of sufficient size to allow for well-powered analyses.

The timing of CNS insults such as TBI with respect to the child’s age is also variable not only across different CNS disorders but also within disorders. The manner in which CNS-related conditions affect psychological development reflects an ongoing developmental process that varies with the child’s age and cognitive developmental level. Important background variables such as socioeconomic status may exert different effects depending on the child’s stage of development, thus offering unique opportunities for intervention. Evaluation of influences on psychological outcomes at various ages within the same CNS condition is also necessary because the older the child, the greater the cumulative impact of environmental influences on psychological development. Finally, it should be noted that the multifactorial influences on children’s outcomes reflect a continuum of potential modifiability. For example, some influences such as family relationships are far more modifiable than the traumatic injury to the CNS experienced by the child.

Specific Recommendations for Future Research

Taken together, the contributions to this special section have advanced scientific knowledge of research on the outcomes of children and adolescents with a range of conditions that affect the CNS. The impact of future research on such conditions will be enhanced by considering the following recommendations (also see Taylor, 2010; Yeates, 2010).
Identify the Specific Influence of Neuropathology on Psychological Outcomes

The CNS problems that were represented in this section were heterogeneous in etiology and specific impact. Neurofibromatosis is a specific genetic deficit (Graf et al., 2006), and spina bifida is also influenced by genetic factors (Fletcher & Dennis, 2009). In contrast, FAS reflects the influence of a specific toxin on the developing fetus and CNS, whereas TBI reflects the impact of varying severity and locations of injury that can occur at different ages. None of the studies in this section provided independent measurement of neuropathology and hence could not shed light on the relationship between the neuropathology experienced by individual children and subsequent psychological outcomes.

Scientific understanding of how the specific neural substrate that affects the psychological associated outcomes with various CNS disorders that are studied will be improved by independently measuring brain structure and function. Several options are now available, including a broad range of modern MRI methods including functional neuroimaging (Hunt & Thomas, 2008). Although these methods are costly and not without difficult challenges in documenting sources of within group and between group variability in magnetic resonance signals (Hunt & Thomas, 2008), studies of the links between psychological outcomes and imaging findings are important and welcomed by Journal of Pediatric Psychology.

Identify Modifiable Versus Nonmodifiable Mediators of Psychological Outcomes

Investigators who study the influences of psychological outcomes of CNS disorders need to consider a broad range of influences, including genetic, family, developmental, and intervention influences that interact with the effects of CNS impairment to influence psychological outcomes (Yeates et al., 2007; Taylor, 2010; Yeates, 2010). These factors may be categorized as risk factors that increase the likelihood that the child may experience a problematic psychological outcome or resource or protective factors that increase the likelihood of more adaptive outcomes. Several authors in this special section included potentially modifiable risk factors such as parent–adolescent conflict and maternal depressive symptoms (Wasserman et al., 2012), and resource factors such as parental competence and social support that might be targeted in future intervention studies (Devine et al., 2012).

Future studies of the impact of various CNS disorders would be improved by a theoretically driven, multivariate approach that specifies key environmental and developmental influences in predictive models of psychological outcomes (Taylor, 2010 and Yeates, 2010). Potentially modifiable influences that demonstrate the most powerful mediating relationships with key psychological outcomes might be targeted for interventions designed to prevent or ameliorate negative outcomes. Moreover, even less modifiable influences can inform decisions about children and families needing the greatest monitoring and most available support.

Clarify the Role of Developmental Influences on Psychological Outcomes

Developmental influences will be important to consider in future research on the psychological outcomes of children and adolescents who experience CNS-related problems. For example, the age at which the CNS insult occurred (Yeates, 2010) is an important factor. Various theories have asserted periods of developmental vulnerability and resilience, but the empirical literature that have tested them remains small (see Crowe et al., 2012, in this section).

The age at which the child is assessed is another important consideration in research on children and adolescents with CNS-related conditions (Yeates, 2010). The studies represented in this special section included samples of different ages ranging from infancy and preschool to adolescence. One of the most important considerations in understanding psychological outcomes of CNS-related conditions involves the assessment of change in a given outcome across different ages. Prospective studies are necessary to advance scientific understanding of developmental changes in psychological outcomes and potential mechanisms of change among children with conditions that affect the CNS (Taylor, 2010).

In this section, Devine et al.’s (2012) and Wasserman et al.’s (2012) prospective follow-up of adolescents with spina bifida from ages 8 to 17 years represent an exemplar research design. In addition, Moran et al.’s (2012) prospective follow-up of children with TBI 12 months postinjury also provided an opportunity to study predictors of HRQOL. Longer term prospective studies will be relevant in documenting the psychological outcomes of CNS-related problems that have continuing clinical significance in adolescents’ and adults’ lives.

Enhance Sample Sizes and Sampling Procedures

Sampling issues pose difficult challenges in research on CNS disorders. Studies in this special section generally...
sampled from a specific clinical setting and populations. Sample sizes in the studies represented in this section were relatively small (less than 75) in the majority of studies. This poses an important methodological problem especially given the heterogeneity of the CNS-related conditions and associated risk and protective factors. To enhance statistical power, especially for analysis of subgroups, sample sizes and sampling procedures in studies of the outcomes of CNS disorders in some instances need to be increased. Multisite studies will be needed to extend sample size and generalizability of findings.

Clarify Relationships among Key Outcomes in Multiple Domains

Given the multifaceted impact of CNS-related conditions on psychological and family functioning, the selection of measures in studies of psychological outcomes poses a significant challenge. To their credit, investigators in this special section studied a wide range of outcomes, including cognitive outcomes, socioemotional functioning, and family variables, including parental functioning, social support, and parent–child conflict.

The scientific impact of research on the psychological outcomes of CNS-related conditions would be enhanced by greater understanding of the relationships among neuropsychological processes (including strengths and deficits) that underlie functional outcomes in critical domains such as academic achievement, competence in peer relationships, emotion regulation, and psychological symptoms. Comprehensive measurement strategies would also facilitate the identification of profiles of competencies or deficits that need to be targeted in specific interventions.

Design and Evaluate Interventions to Enhance Psychological Outcomes

One of the most important needs for future research is to use new data concerning predictors, including risk and protective factors, of psychological outcomes to design effective interventions to reduce the level of risk experienced by children with conditions that affect the CNS. Although none of the studies published in this special section focused on intervention, it should be noted that each of the studies had relevant implications for intervention. Future research should build on the body of knowledge generated by descriptive studies that describe the outcomes of CNS-related disorders (Yeates, Ris, Taylor, & Pennington, 2010) as well as new intervention strategies (Kanne, O’Kane, Grissom, & Farmer, 2010; Bryck & Fisher, 2012). Exemplars of intervention studies with pediatric populations include Wade and colleagues’ application of family-centered interventions to improve child behavior, social competence, and parental distress following pediatric TBI (Wade, Carey, & Wolfe, 2006a,b) and Butler et al.’s (2008) work on cognitive remediation for the survivors of pediatric malignancy.

Final Note

The contributions to this special section have provided important data on psychological outcomes of pediatric conditions that affect the CNS and have helped to address the gap in published research on this topic in Journal of Pediatric Psychology. Our hope is that other investigators will follow their lead.

Conflicts of interest: None declared.

References


