Children with Disabilities and Impacts on Families

by

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Amy Houtrow

Abstract

Children with disabilities and their families face substantial health and social challenges. Health service inequities exist in multiple arenas for children with disabilities that can profoundly influence health, function and well-being. The impacts of disability in childhood also extend to their families who may experience psychological and physical stress, social restrictions and financial burdens. Using social ecological modeling to contextualize children with disabilities, the experience of children and their families can be studied in multiple settings. Using the behavioral model of health services use, health inequities for children with disabilities can be elucidated. The 3 papers included in this dissertation utilize these frameworks to profile the health and health services of children with disabilities, to evaluate the family impacts of childhood mental health problems and to rate the impacts of proton radiation therapy on the families of children with brain tumors.

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Introduction

I have devoted my academic career to improving the health, function and well-being of children with disabilities. Clinically, I care exclusively for children with disabilities. It is this work that guides my research interests. I am constantly reminded of stark health care inequities, challenges to accessing care and the negative impacts families experience. Because I felt (and feel) that systems level change was needed and that I was ill-equipped to join the dialogue, I pursued an MPH in Health Management and Policy at the University of Michigan School of Public Health during my residencies. Once in my faculty position at UCSF, I recognized how much more I needed to learn about research methods and the theoretical foundations for understanding the intersections between health and social factors. My PhD training in Sociology imbedded in my research career development award coupled with fantastic mentorship has poised me well for a career in health services research for children with disabilities.

For the past few years, I have been engaged in a variety of research projects focusing on children with disabilities. These projects are listed below.

Research conducted during my PhD training

1. Okumura MJ, Van Cleave J, Gnanasekaran S, Houtrow A. Understanding factors associated with work loss for families caring for CSHCN. *Pediatrics.* Dec 2009;124 Suppl 4:S392-398.
2. Edwards J, Davidson, E, Houtrow AJ, Graham R. Pediatric Resident Attitudes toward Caring for Children with Severe Disabilities. American

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7. Kuo DZ, Houtrow AJ, Arango P, Kuhlthau KA, Simmons J, Neff JM. Family-Centered Care: Current Applications and Future Directions in Pediatric Health Care. Maternal and Child Health Journal (2012) Vol 16 (2) 297-305.
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9. Halfon N, Houtrow A, Larson K, Newacheck P. The changing landscape of disability in childhood. Future of Children Spring 2012 Volume 22 (1) 13-42.
10. Houtrow AJ, Bhandal M, Pratini NR, Davidson L. The Rehabilitation of Children with Anti-NMDA-Receptor Encephalitis: A Case Series. The American Journal of Physical Medicine and Rehabilitation. May 2012. 91 (5): 435-441.
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12. Houtrow A, Jones J, Ghandour R, Strickland B, Newacheck P. Participation of Children with Special Health Care Needs in School and the Community. Academic Pediatrics (in press August 2012)
13. Houtrow AJ, Kang T, Newcomer R. In-Home Supportive Services for individuals with cerebral palsy in California. Journal of Pediatric Rehabilitation Medicine. 2012 (in press)

14. Houtrow AJ, Okumura MJ, Inpatient Care for Children with Spina Bifida in the United States. (planned submission to special issue of the Journal of Pediatric Rehabilitation Medicine).
15. Houtrow AJ, Newacheck PW, Larson K, Halfon N. Trends in Childhood Disability 2000-2010. (in process)
16. Houtrow AJ, Informal caregiving for Children with Special Health Care Needs with Physical Disabilities (in process)

With the exception of the clinical research projects, the research included in this dissertation and listed above is guided by the conceptual framework that children with disabilities have characteristics particular to them and function and live within a family structure, embedded in an environment that includes the community, school and larger social influences. Using this type of framework pulls the child away from the purely medical model of disability (disability located within and inherent to the individual) and places the child in a framework of interactions and influences on their health and function. This framework also highlights the importance of the family.

Social Ecology Model

In this section I will briefly review social ecology and will present Bronferbrenner's ecological theory of development as it relates to children with disabilities because his work provides a classic example of social ecological modeling. Central to his theory are the concepts of personal characteristics, hierarchical environmental influences and time (Sontag, 1996). Essentially, social ecology is the study of the relationship between a developing human being (in

my research, a child) and the contexts/settings in which the human being is engaged (Kazak, 1986). It is well established that well-being of a child with disabilities is influenced not only by the presence of health conditions, but also by socio-cultural influences. The International Classification of Function (ICF) is an example of a social ecology model for disability in which disability is understood as an interaction between an individual and contextual factors (personal and environmental) (Lollar & Simeonsson, 2005). In social ecological models, there are multiple factors that can influence individual experiences. The social ecological model attends to feedback loops and the reciprocal nature of interactions (Sontag, 1996). Interactions are understood as the exchanges an individual makes with their environment that may be simple or complex and reciprocal (U. Bronfenbrenner, 1994). Within an individual's world are interrelationships between hierarchically ordered systems (U. Bronfenbrenner, 1994).

Bronfenbrenner's model has four nested concentric structures: the microsystem, the mesosystem, the exosystem and the macrosystem (Kazak, 1986). The microsystem includes the daily relations and roles a child has which can occur at home, at school or with peers; the mesosystems are interrelated microsystems; the exosystem are the environments that the child is not directly in contact with but influenced by; and the macrosystem is the culture and sociopolitical structures in which the child lives (Urie Bronfenbrenner, 1979). For example, the child lives in a family, has daily experiences the community through attending school, is impacted by their family's support network and is influenced

by the higher order cultural milieu. How a child functions depends on the child-environmental interactions in the immediate and remote spheres of influence (Sontag, 1996). Bronfenbrenner sees personal attributes as influential to future development such that a child's orientation to the environment will influence developmental progression (Sontag, 1996). Thus there is a bidirectional influence from the child to the environment and from the environment to the child. Bronfenbrenner also highlights the importance of how the individual perceives and experiences influences (Sontag, 1996).

Other social ecologists, including Powell Lawton, focus on how the environment is perceived and experienced, and how the individual functions within various environments (Lawton, 1974). Lawton found that environments that foster participation and performance yield better outcomes for elderly persons and than environments that are either too demanding or too limited and thus are associated with deprivation (Lawton, 1974). There is a clear connection to children with disabilities who can be effectively encouraged or discouraged to participate with adaptations in their environments at home, at school and in the community (Sontag, 1996). Much of the caregiving literature can be framed using a social ecological perspective. For example, Kazak reports on studies that found that families of children with disabilities experience social isolation (exosystem influences), that families experience stress and strain (microsystem) and do not receive adequate supports from the health care system (macrosystem) (Kazak, 1986).

In addition to the use of standard social ecology models, other

researchers have developed more specific models that focus on person-environment interactions. For example, the Model of Competence was developed specifically to address individuals with motor disabilities (Rousseau, Potvin, Dutil, & Falta, 2002). This model includes six concepts: 1) the person, 2) the environment, 3) the activity, 4) the role, 5) competence, and 6) the 'handicap situation' (Rousseau et al., 2002). The interactional nature of this model highlights the differences between activity (engaging with non-human object such as climbing stairs) and roles (engaging with other people in certain ways) and also focuses on the evaluation of competence in roles and activities or when not competent, the 'handicap situation' (Rousseau et al., 2002). It is clear that some environments pose more challenges than others and describing the interaction of the person in the environment can elucidate solutions to environmental challenges. Accessibility is determined not just by the environment and its modifications but the individual competence of the person engaging with the environment (Iwarsson & Stahl, 2003). To be able to use something in the environment, it needs to be functional. Often times accessible and usable are discussed interchangeably. Despite having different definitions, it is the person interacting with the environment that determines its usability or accessibility. Fit is determined by the interaction not by the individual or the environment alone (Iwarsson & Stahl, 2003). Universal design attempts to address accessibility and usability on a population level. All of these concepts (accessibility, usability and universal design) are relational. (Iwarsson & Stahl, 2003) are best understood using a social ecological model of person-environment interactions, and fit well

within the ICF framework for understanding disability.

My own work can be easily framed using the social ecology model. I consider children to be nested in 'worlds' of ever increasing size: family, community and society. In general, I hypothesize that factors at multiple levels (some easily measured and most not) influence the lives of children with disabilities and their families. So factors are inherent and others mutable. In my opinion, attending to both the mutable and immutable factors is important. Future work that focuses on the altering the mutable and mitigating or circumventing the immutable should positively impact children with disabilities and their families. At the level of the child, I consider demographics to potentially impact health experiences because health disparities and inequities are so common. Other factors such as health status and the presence of certain types of health conditions or behavioral problems likely also influence families' experiences. Family characteristics such as poverty status, insurance coverage, educational attainment in the household and martial status are factors that might positively or negatively influence child health and well-being and family life. Within the community, such factors as the school, community programs and the presence of health services likely influence families and their children with disabilities. On a macro or societal level, how health care is organized, social values and belief systems are likely influential to the experiences of children with disabilities and their families.

Behavioral Model of Health Services Use

When considering research that focuses on health care access and the inequities associated with differential services, it important to consider the sentinel work in this area. The behavioral model of health services use was first described in the 1960s and was revisited and revised in the subsequent three decades (Andersen, 1995). Initially the model intended to explain or predict health services use based on a function of need, the individual's predisposition to seek services and factors that could enable or impede access. In this model, need alone does not adequately explain health service use because other factors, both personal and environmental, influence use of services (Andersen, 1995). One of the major goals of the model is to attend to the issue of access to health services. This is particularly relevant to my research because children with disabilities have more need for services but also have more unmet need (Benedict, 2006; Hill, Freeman, Yucel, & Kuhlthau, 2007; Nageswaran, Silver, & Stein, 2008). Realized access is actual use of health services and should be equitable based on need (Andersen & Aday, 1978). Potential access is the presence of resources that would enable an individual to receive services. Access is considered inequitable when social structures, beliefs and resources differ in such a way as to limit access when services are needed (Andersen, 1995). This framework for understanding health services inequities, coupled with the social ecology model of understanding the impacts of childhood disability, provides me with the tools to conceptualize health services inequities for children with disabilities and also helps me relate child health services to family

experiences. Just as I discussed in the previous section, some factors that impact health and health services are alterable and some are not. In the behavioral model of health services use, demographic characteristics and the social structure are considered to have low mutability, need has variable mutability, health beliefs have medium mutability and enabling factors have high mutability (Andersen, 1995). Advancement and enhancements to the behavioral model of health services use occurred in the 1970's through the 1990s. Consumer satisfaction was added to the model in 1970s (Aday & Andersen, 1981; Andersen, 1995; Andersen & Aday, 1978). Of note, family satisfaction with care and family centered care are key aspects of quality care for children with disabilities (Kuo et al., 2011; Strickland, Jones, Ghandour, Kogan, & Newacheck, 2011; B. B. Strickland et al., 2011). Later, the concepts of efficient and effective access were added, as were the external environmental factors of physical, political and economic worlds (Andersen, 1995). By adding more contextual variables such as the environmental factors just listed, the model is well-aligned with social ecological models that frame disability in childhood.

The International Classification of Functioning

Any detailed discussion of disability should include the World Health Organization's International Classification of Functioning, Disability and Health. Briefly, I will review this model for understanding disability. The model of disability embraced by the creators of the International Classification of Functioning,

Disability and Health (ICF) is an amalgam of the medical model of disability and the social model. In the medical model, disability is caused by a disease, trauma or other health condition and requires medical intervention to ameliorate it. In contrast, the social model identifies the problem at the societal instead of individual level. In a biopsychosocial model of disability, such as in the ICF, disability is understood as an interaction between the individual and the context in which they live. In addition, the ICF acknowledges that individuals can experience decrements in health and experience disability. Instead of focusing on disease states, the ICF focuses on impact and has neutral language to describe health and related states (World Health Organization, 2002). The emphasis has shifted from disability to level of health and functioning. The International Classification of Functioning, Disability and Health for Children and Youth (ICF-CY) is the version created especially for children to account for the specific and unique aspects of disability in childhood. For children, disability must be explained in the context of delays, deviations and variations in growth or development (Ibragimova, 2005). Disability is not static, especially in childhood, and therefore requires a method to account for developmental factors (Simeonsson et al., 2003). One of the major critiques of the ICF model is the blurring of distinctions between activities and participation. Activity can be understood as occurring on an individual level and participation on a societal level. The experiences of activity limitations are more closely correlated with impairments where participation is more complex with more interacting factors (Whiteneck, 2006). This has particular relevance in childhood because children

are social actors within the context of their families and depend on their families to address their participation needs (Colver, 2005). This framework for understanding disability aligns well with the CSHCN Screener which identifies children with difficulty functioning.

Family Impacts of Childhood Disability

Family, and mothers in particular, experience tremendous impacts of raising children with disabilities. Mothers report stress, strain, loss of employment, fatigue, and physical and mental health problems related to caring for their children with disabilities (Anderson & Eifert, 1989; Banks, 2003; Dodgson et al., 2000; Eddy & Engel, 2008; Fleming et al., 1994; Hassall, Rose, & McDonald, 2005; Reichman, Corman, & Noonan, 2008; Witt, Riley, & Coiro, 2003). Despite the policy mandates for community-based supports, the actual services that are available for children with disabilities vary considerably (Benedict, 2006). For the most part, the extra care responsibilities fall to mothers (Anderson & Eifert, 1989; Hassall et al., 2005; Loebig, 1990; Traustadottir, 1991). The extra work that mothers (or other caregivers) do as care providers for their children with disabilities is often highly specialized and technical, (Traustadottir, 1991) yet very rarely is this work paid. Women who care for their children with severe disabilities often refer to this caring as ‘my life’s work’ and devote considerable time and energy to becoming skilled at managing the health and well-being of their children (Kazak, 1986; Traustadottir, 1991; Viner-Brown & Kim, 2005). In ethnographic studies of caregivers for children with disabilities, mothers almost

always assume the caring role (Rehm & Bradley, 2005). While nearly 60% of mothers of typical children maintain employment or re-enter the workforce outside the home after the birth of their children, very few mothers of children with disabilities work outside of the home (Okumura, Van Cleave, Gnanasekaran, & Houtrow, 2009; Traustadottir, 1991). Caring for a child with a disability can be exceptionally demanding and time-consuming which seriously limits a mother's opportunities for employment outside the home and financial independence (Traustadottir, 1991). Mothering always includes providing for, protecting, nourishing, teaching and caring for children to optimize their development and well-being. How this work is done varies considerably when a child has a disability. The work is altered, exaggerated and extends well past the typical timeframe and expectations of motherhood (Green, 2007; McKeever & Miller, 2004). It is with this knowledge regarding the family impacts of disability that I participated in the Family Impacts of Brain Tumors Treated with Proton Radiation Therapy. While the ability to assess the impacts on families was limited within the structure of the research project, we gained valuable information about what families experience.

The three papers presented in this dissertation focus on health care inequities for children with disabilities, the experiences families have when caring for a child undergoing proton radiation therapy for brain tumors and the negative impacts (burdens) of caring for a child with mental health problems. These three papers

are part of a larger body of work (as listed above) that attempts to expand the existing knowledges regarding children with disabilities and their families.

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Profiling Health and Health-Related Services for Children With Special Health Care Needs With and Without Disabilities

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ABSTRACT

OBJECTIVE: The aims of this study were to profile and compare the health and health services characteristics for children with special health care needs (CSHCN), with and without disabilities, and to determine factors associated with unmet need.

METHODS: Secondary data analysis of the 2005–2006 National Survey of Children with Special Health Care Needs was conducted. The sociodemographics, health, and health services of CSHCN with and without disabilities were compared. Multivariable logistic regression was employed to examine factors associated with unmet need for health services.

RESULTS: Children from minority racial and ethnic groups and children living in or near poverty were over-represented among CSHCN with disabilities, compared with other CSHCN. Statistically higher percentages of CSHCN with disabilities had behavioral problems (39.6% vs 25.2%), anxiety/depressed mood (46.1% vs 24.0%), and trouble making/keeping friends (38.1% vs 15.6%) compared with other CSHCN. Thirty-two percent of CSHCN with disabilities received care in a medical home compared with 51% of other CSHCN. CSHCN with disabilities had higher rates of need and unmet need than other CSHCN for specialty care, therapy services, mental health services, home health, assistive devices, medical supplies, and durable medical equipment. The adjusted odds of unmet need for CSHCN with disabilities were 71% higher than for other CSHCN.

CONCLUSION: CSHCN with disabilities had more severe health conditions and more health services need, but they less commonly received care within a medical home and had more unmet need. These health care inequities should be amenable to policy and health service delivery interventions to improve outcomes for CSHCN with disabilities.

KEYWORDS: children with special health care needs; disabilities; medical home; unmet need

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WHAT'S NEW

Children with special health care needs (CSHCN) with disabilities are identified as a distinct group of CSHCN because of their sociodemographic, health, and health services characteristics. CSHCN with disabilities had more psychosocial problems and health service needs than other CSHCN. We also document inequities in health services by highlighting the rates of unmet need and low percentages of care within a medical home for CSHCN with disabilities.

CHILDREN WITH SPECIAL health care needs (CSHCN) are those children with chronic physical, developmental, emotional, or behavioral conditions who need or use health and related services of a type or amount beyond that typically required by children.[1] A child can qualify as having special health care needs if he/she has a chronic condition that has lasted or is expected to last at least 1 year and is associated with at least 1 of the following 5 consequences: needing or using prescription medication; needing or using more medical care, mental health, or educational services than is usual for most children of the same age; being limited or prevented in any way in his/her ability to do the things most children of the same age can do; needing or receiving special therapy; and/or needing or receiving treatment or counseling for any emotional, developmental, or behavioral problem.[2] Over 20% of CSHCN qualify because they are limited or prevented in their abilities to do things that most children of the same age can do[3] and, thus, are considered to be disabled based on the International Classification of Functioning, Disability and Health (ICF) framework for understanding disability.[4]

CSHCN with disabilities are a special and vulnerable subset of CSHCN because the consequences of having a disabling health condition can be profound.[5] Children with disabilities are reported to have extensive health care needs, high rates of health services utilization and costs, and poorer access to needed health services.[6–8] Furthermore, the consequences related to disability in childhood extend beyond experiences with the health care system and can include difficulties with school and participation in life events.[3,9] These consequences can have long-term impacts on health outcomes, life opportunities, and participation in adulthood.[10] Because of the negative

impacts of disability, it is important for pediatric health providers to understand the population of children with disabilities to provide optimal health care and advocate for the services and assistance they need to be successful in life.

Since the operationalization of the new definitional framework for CSHCN over a decade ago, fewer health services research studies specifically focus on children with disabilities, and only a handful of recent studies have focused on the subgroup of CSHCN with disabilities.[2,8,9,11,12] Instead, most studies have focused on the general population of CSHCN and have identified issues around access, health insurance, quality of medical care, financial impacts on families, and health disparities.[12,13–16] Few studies have looked at services specifically related to children with disabilities/functional limitations, such as durable medical equipment and assistive aids, although some studies identify the presence of functional limitations as a risk factor for unmet need and difficulty with health care access.[8,9,17,18] Therefore, a gap in the health services literature exists for CSHCN with disabilities. The purpose of this project is to fill the gap in the literature by profiling and comparing CSCHN with disabilities to other CSHCN to identify sociodemographic, health, and health services differences and to determine factors associated with unmet need. We hypothesize that CSHCN with disabilities have more severe and less stable health conditions than other CSHCN and have more extensive health services needs, but have higher rates of unmet needs and less commonly receive care within a medical home than other CSCHN. We further hypothesize that after controlling for health condition severity and sociodemographic characteristics often associated with health care inequities, that CSHCN with disabilities have increased odds of unmet service need.

METHODS

DATASET

The 2005–2006 National Survey of Children with Special Health Care Needs (NS-CSHCN) is a nationally representative sample of CSHCN that was conducted by the National Center for Health Statistics (NCHS), the Maternal and Child Health Bureau, and the Centers for Disease Control and Prevention between April 2005 and February 2007.[19] The NS-CSHCN offers a special opportunity to evaluate CSHCN with disabilities because it is the most extensive and up-to-date version of these periodic surveys of CSHCN.[19] The State and Local Area Integrated Telephone Survey mechanism was used to randomly identify 4 million household phone numbers. A computer-assisted telephone interview system was used to screen households for eligible children and to administer the CSHCN survey. From the 192 083 households with children, 364 481 children were screened for having special health care needs via the CSHCN Screener and 42 332 (11.6%) qualified. If a household had more than 1 identified child with special health care needs, 1 child was randomly chosen to be included in the sample.[3] For these children, full interviews were conducted with the adult in the household most familiar with the child's special health care needs (usually the mother), with a completion rate of 96.2%.[3]

CONCEPTUALIZING DISABILITY AND HEALTH SERVICES

To frame our research we used 2 conceptual models. The first model, the ICF, provided a framework for classifying CSHCN as having disabilities or not. Individuals who are limited in their ability to do what people are typically able to do can be considered to have disabilities at 1 or more of the following levels: bodily impairments, activity limitations, or participation restrictions.[20] This robust framework is well aligned with the CSHCN Screener, which identifies CSHCN who are considered by their caregiver to be limited in their ability to do the things that most children of the same age can do because of a medical, behavioral, emotional, or developmental condition that has lasted or is expected to last at least 1 year.[2] Based on these CSHCN Screener questions, we dichotomized CSHCN into those with disabilities and those without. We note that the ICF framework for disability relates health conditions to functioning but does not require the identification of a specific etiology nor does it require a minimum amount of time for the condition to be present.[20] Because disability among CSHCN is more narrowly defined than in the ICF framework, the NS-CSHCN population estimates may subsequently be lower than other reports.

The second model, the behavioral model of health services use, framed our analyses of health utilization and unmet need. This model frames health service use and access to health services based on predisposing characteristics and enabling resources.[21] Individuals with the need for health services may have those needs met through realized access or might have unmet needs and experience health care inequities.[22] For example, the presence of a mobility limitation might predispose an individual to need durable medical equipment, and their insurance might act as an enabling factor. There are certainly other factors that hinder or enable access. Therefore, we used this model to guide us in determining which factors should be included in our multivariable logistic regression analysis of presence of unmet need. Using these 2 frameworks, the ICF and the behavioral model of health services use, we examined factors that relate to the experience of disability in childhood.

SOCIODEMOGRAPHIC, HEALTH, AND PSYCHOSOCIAL CHARACTERISTICS

The sociodemographic variables of interest for this study included gender, age, race/ethnicity, income, insurance status, household composition, and highest educational attainment in the household. Age was categorized into the following groups: 0 to 4 years (preschool age), 5 to 13 years (school age), and 14 to 17 years (high school age). Race/ethnicity was categorized into 4 groups: white non-Hispanic, black non-Hispanic, Hispanic, and other. Income was divided into 3 categories by using federal poverty level (FPL) criteria: less than 200% FPL, 200% to 399% FPL, and 400% or greater of the FPL. Insurance

status was categorized into the following categories: full year private insurance, full year public insurance, full year private and public coverage, full year other comprehensive insurance, and uninsured at the time of the interview. Household composition included the following categories: single mother, 2 parent, and other type of household composition. The highest educational attainment in the household was defined as less than high school, graduated from high school, and more than high school.

We identified 2 questions from the survey to describe the health status of CSHCN with and without disabilities. Parents/caregivers reported the severity of their child's health conditions/problems (no severity, mild, moderate, and severe) and how stable the child's health was (changed all the time, changed once in a while, or was usually stable). In addition, we compared the percentages of CSHCN with and without disabilities whose parents reported that their child felt anxious or depressed, had behavior problems, and/or had trouble keeping and making friends. As a measure of health impact, we report the percentage of CSHCN with and without disabilities who missed 20 or more days of school because of their health problems.

Health Services: The Medical Home, Service Need, and Unmet Need

To evaluate health services, we measured care within a medical home, and need and unmet need for a variety of services. The presence of a medical home was operationalized using the following 5 Maternal and Child Health Bureau criteria: having a personal doctor or nurse, having a usual source of care, receiving family-centered care, having no problem with getting referrals when needed, and receiving effective care coordination when needed.[23,24] For our analysis, having a usual source of care, having a personal doctor or nurse, and having family-centered care were dichotomized as present or not. The criterion of having no problems with referrals was measured as yes, no, and did not require; therefore, the percentage of children getting the service when needed was calculated as a fraction of those who reported needing it. The criterion for care coordination was calculated in the same way as having no problems with referrals. Therefore, both the referral outcome and care coordination outcome were considered met if individuals did not have a need, or when need was reported, it was also reported met. We also created a composite measure to classify children as receiving care in a medical home when all 5 criteria were met. To address need, we identified whether the sample child used specialty care; prescription medication; physical therapy, occupational therapy, and/or speech therapy; mental health care; home health care; mobility aids; communication aids; medical supplies; durable medical equipment; and/or respite care in the 12 months preceding the survey. When one of the aforementioned items/services were needed but not received in the 12 months preceding the survey, the need was considered unmet. Additionally, we created a composite measure of unmet need as the presence of 1 or more of the aforementioned types of unmet need for our multivariable analysis.

Statistical Analysis

We performed univariate and bivariate analyses to evaluate the differences between CSHCN with and without disabilities. Survey weights provided by the NCHS[19,25] were used to obtain population level estimates. Multivariable logistic regression was conducted to identify factors associated with unmet need for services based on the behavioral model of health services use. We used the multiple imputation files available from the NCHS to account for the 9% missing income values[26] and did not otherwise impute values of missing covariates. Instead, we compared the fit of models that included and excluded missing covariate values, and we found negligible differences between parameter estimates or confidence intervals for any covariate. We performed a Hosmer-Lemeshow goodness of fit test designed to take into account the complex survey design and found that our model had a good fit, with $P = .44$. The adjusted estimated prevalences of unmet need among CSHCN were calculated from the regression model. All analyses were conducted using STATA 11 (StataCorp, College Station, TX) to account for the complex nature of the survey design and to appropriately weight the estimates. The Committee on Human Research at the University of California, San Francisco, approved this study in the exempt category.

Results

Sociodemographics, Health, and Psychosocial Characteristics

We estimate that in 2005 to 2006, 13.9% of children in the United States had special health care needs. Of these children, 21.5% qualified as having disabilities for this study because they had at least 1 functional limitation, as shown in Table 1. This equates to 2.2 million children with disabilities associated with chronic conditions and 8 million other CSHCN. Boys, minority children, children living near or in poverty, uninsured and publicly insured children, children living in households headed by a single mother, and children living in homes in which the highest educational attainment was high school or less were overrepresented in the sample of CSHCN with disabilities compared with CSHCN without disabilities. For example, 52.2% (95% confidence interval [CI], 50.3–54.1) of CSHCN with disabilities live in homes with incomes below 200% of the FPL compared with 37.9% (95% CI, 36.9–38.9) of CSHCN without disabilities.

CSHCN with disabilities had conditions that "changed all the time" 4 times as frequently as other CSHCN, and their conditions were rated as "severe" 7 times as frequently (Table 1). Feeling anxious and/or depressed was twice as commonly reported for CSHCN with disabilities than other CSHCN (46% vs 24%; $P < .001$). Additionally, nearly 40% of CSHCN with disabilities had behavioral problems compared with only 25% of other CSHCN; $P < .001$. Similarly, 38% of CSHCN with disabilities had trouble making or keeping friends compared with only 16% of other CSHCN; $P < .001$. Of CSHCN with disabilities, 12% missed more than 3 weeks

Table 1. Distributions of Sociodemographic and Child Health and Related Characteristics of CSHCN by Disability Status*

Characteristic	CSHCN With Disabilities		CSHCN Without Disabilities	
	Sample Distribution n = 8739 Percentage (95% CI†)	Estimated Population (In Millions)	Sample Distribution n = 31 984 Percentage (95% CI)	Estimated Population (In Millions)
All	21.5 (20.8–22.1)	2.2	78.5 (77.9–79.2)	8.0
Gender‡				
Boys	61.8 (60.0–63.5)	1.4	58.7 (57.8–59.6)	4.7
Girls	38.2 (36.5–40.0)	0.8	41.3 (40.4–42.2)	3.3
Age, y				
0–4	15.5 (14.3–16.9)	0.3	16.3 (15.5–17.0)	1.3
5–13	55.4 (53.7–57.2)	1.2	56.1 (55.1–57.0)	4.5
14–17	29.0 (27.4–30.7)	0.6	27.7 (26.9–28.5)	2.2
Race/ethnicity‡				
White non-Hispanic	62.2 (60.4–64.0)	1.4	66.2 (65.2–67.1)	5.3
Black non-Hispanic	17.9 (16.5–19.4)	0.4	15.8 (15.1–16.6)	1.3
Hispanic	12.7 (11.4–14.1)	0.3	11.5 (10.9–12.2)	0.9
Other	7.2 (6.4–8.2)	0.2	6.5 (6.1–7.0)	0.5
Income‡				
<200% FPL§	52.2 (50.3–54.1)	1.1	37.9 (36.9–38.9)	2.8
200%–399% FPL	27.0 (25.4–28.7)	0.54	31.0 (30.1–31.9)	2.3
≥400% FPL	20.8 (19.4–22.3)	0.42	31.1 (30.3–32.0)	2.3
Insurance status‡				
Private	45.5 (43.7–47.2)	1.0	62.9 (61.9–63.8)	5.0
Public	36.7 (35.0–38.5)	0.8	25.7 (24.8–26.6)	2.1
Private and public	11.7 (10.7–12.9)	0.3	6.2 (5.7–6.6)	0.5
Other insurance	1.9 (1.5–2.3)	0.04	2.0 (1.8–2.3)	0.2
Uninsured	4.2 (3.9–5.0)	0.09	3.3 (3.0–3.6)	0.3
Household composition‡				
Two parent	59.5 (57.7–61.3)	1.3	66.4 (65.5–67.3)	5.1
Single mother	35.3 (33.5–37.1)	0.7	28.4 (27.5–29.3)	2.2
Other	5.2 (4.6–6.0)	0.1	5.2 (4.8–5.6)	0.4
Highest educational attainment in the home‡				
Less than high school	8.7 (7.6–9.8)	0.2	6.3 (5.8–6.9)	0.5
High school	27.7 (26.0–29.4)	0.6	21.8 (21.0–22.7)	1.8
Greater than high school	63.7 (61.9–65.4)	1.4	71.8 (70.9–72.7)	5.8
Condition severity‡				
None/not applicable	2.4 (2.0–3.0)	0.05	18.5 (17.8–19.2)	1.5
Minor	18.7 (17.4–20.0)	0.4	50.1 (49.2–51.1)	4.0
Moderate	53.2 (51.4–55.0)	1.2	27.6 (26.7–28.4)	2.2
Severe	25.7 (24.2–27.3)	0.6	3.9 (3.5–4.3)	0.3
Condition stability, health care needs‡				
Were usually stable	48.9 (47.1–50.7)	1.1	70.5 (69.6–71.4)	5.6
Changed once in a while	35.6 (34.0–37.3)	0.8	25.8 (25.0–26.7)	2.1
Changed all the time	15.5 (14.2–16.8)	0.3	3.7 (3.2–4.1)	0.3
Missed ≥20 days of school‡	12.0 (10.9–13.3)	0.2	3.3 (3.0–3.8)	0.2
Feels anxious or depressed‡	46.1 (44.3–47.9)	1.0	24.0 (23.2–24.8)	1.9
Has behavioral problems‡	39.6 (37.8–41.4)	0.8	25.2 (24.4–26.0)	2.0
Has trouble making or keeping friends‡	38.1 (36.3–39.9)	0.8	15.6 (14.9–16.3)	1.2

*CSHCN = children with special health care needs.
†CI = confidence interval.
‡Chi-squared and *t* tests were used to identify statistically significant differences ($P < .01$) between CSHCN with and without disabilities.
§FPL = federal poverty level.

of school compared with 3.3% of CSHCN without disabilities; $P < .001$.

HEALTH SERVICES: THE MEDICAL HOME, SERVICE NEED, AND UNMET NEED

When the medical home was measured as a composite, only 32.2% of CSCHN with disabilities were receiving care within a medical home, compared with over half of other CSHCN (Figure 1). Over 93% of CSHCN reported having a personal doctor or nurse, regardless of disability status. On all other components of the medical home, statistically significant differences were noted such that CSHCN with disabilities less commonly reported meeting the component criteria. Most notably, only 48.8% of CSHCN with disabilities reported adequate care coordination compared with 73.5% of other CSHCN.

As shown in Table 2, CSHCN with disabilities needed fewer prescription medications but a statistically significant quantity of more of every other item/service studied. Overall, 94.9% of CSHCN without disabilities and 96.7% of CSHCN with disabilities had an identified need for at least 1 item/service. As a composite measure of

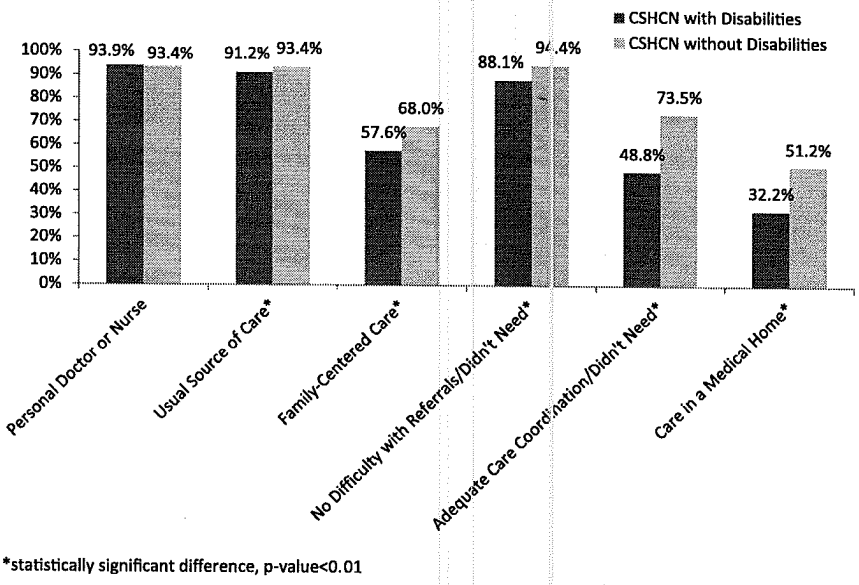

Figure 1. *Percentages of CSHCN with and without disabilities who meet medical home criteria. *Statistically significant difference; P < .01. CSHCN indicates children with special health care needs.*

unmet need, 22.8% (95% CI, 21.3–24.3) of CSHCN with disabilities had an unmet need for at least 1 of the aforementioned items/services compared with 7.4% (95% CI, 6.9–7.9) of other CSHCN. The unadjusted odds of having at least 1 unmet need for the aforementioned items/services was 3.71 (95% CI, 3.30–4.16) for CSHCN with disabilities compared with other CSHCN. CSHCN with disabilities has statistically higher odds of unmet need for the following services: prescription medication; specialty care; mental health services; physical therapy, occupational therapy, and/or speech therapy; medical supplies; durable medical equipment; and communication aids.

The adjusted odds of having at least 1 unmet need was 1.68 (95% CI, 1.45–1.94) for CSHCN with disabilities compared with other CSHCN (Table 3). Other child level predictors of unmet need included the "other" race designation, living below 400% of the FPL, increased condition severity levels, and having health care needs that were not usually stable. The family level predictors of unmet need were living in a home headed by a single mother and living in a home where the highest educational attainment level was less than high school. The health systems factors of being uninsured and not receiving care within a medical home were associated with increased adjusted odds of unmet need, 3.03 (95% CI, 2.33–3.95) and 3.40 (95% CI, 2.87–4.03), respectively. The adjusted estimated prevalences of having at least 1 unmet need were highest among CSHCN with severe health conditions (32.0%), CSHCN with conditions that were unstable (24.3%), CSHCN who were uninsured (25.4%), and CSCHN with disabilities (18.6%); as shown in Table 3.

DISCUSSION

Our analysis demonstrates that CSHCN are a distinct subset of CSHCN. Because of their higher rates of severe health conditions, psychosocial issues, and unmet need, CSHCN with disabilities could benefit from focused attention to address their needs in the health and social realms. We found that CSCHN with disabilities differ from other CSHCN in many ways. Among CSHCN with disabilities, there is an over-representation of boys, blacks, children covered by public insurance, uninsured children, and those living in relative poverty. These differences are even more alarming considering the known sociodemographic disparities between CSHCN and children without special health care needs.[6,25–28] We also observed that CSHCN with disabilities had more severe and less stable health conditions than other CSHCN. Bramlett and colleagues[8] categorized CSHCN by functional status and also found differences between CSHCN with functional limitations and those without in terms of health status and health complexity. This is not unexpected, because as conditions such as asthma or cystic fibrosis become more severe, they more likely will limit children's activities. Conversely, though, a child with mild cerebral palsy might be considered to be very healthy and stable but have disabilities in multiple functional domains. Therefore, practitioners should consider how factors that lead to disability can be mitigated and if stabilizing the child's health condition might improve functional outcomes.

In addition to the relationships between disability status and condition severity and stability, we found that CSHCN with disabilities more commonly had psychosocial issues compared with other CSHCN. CSHCN with disabilities had more problems with behavior, feeling anxious or depressed, and trouble making or keeping friends. These findings have important practice implications. With the knowledge that CSHCN with disabilities more commonly experience psychosocial problems, health care providers can screen those with disabilities more closely to identify and make recommendations to address psychosocial issues as needed. Addressing psychosocial issues early may help lessen the long-term effects on mental health and

Table 2. Reported Need and Unmet Need for Services for CSHCN, With and Without Disabilities*

Type of Service or Item	Percentage With Service Need Present			Percentage With Unmet Need for Services		
	CSHCN With Disabilities n = 8719	CSHCN Without Disabilities n = 32 004	CSHCN With Disabilities Compared to Other CSHCN Unadjusted OR (95% CI)†	CSHCN With Disabilities	CSHCN Without Disabilities	CSHCN With Disabilities Compared to Other CSHCN Unadjusted OR (95% CI)
Prescription medication n = 35 179	84.5	86.9	0.82 (0.74–0.92)	3.6	1.4	2.62 (1.97–3.47)
Specialty care n = 21 064	67.6	47.4	2.32 (2.13–2.52)	8.6	4.1	2.25 (1.81–2.79)
Mental health n = 10 171	33.7	22.6	1.74 (1.59–1.90)	17.9	13.8	1.36 (1.11–1.68)
PT/OT/speech therapy** n = 9305	51.3	15.1	5.92 (5.43–6.48)	16.6	10.9	1.63 (1.30–2.05)
Medical supplies n = 7588	29.4	15.7	2.24 (2.04–2.46)	3.9	1.8	2.26 (1.39–3.67)
Durable medical equipment n = 4662	20.8	8.9	2.70 (2.41–3.02)	6.5	2.2	3.03 (1.71–5.38)
Respite care n = 1855	14.3	1.9	8.69 (7.19–10.50)	50.5	43.0	1.35 (0.93–1.95)
Home health n = 1826	11.4	2.6	4.82 (4.06–5.73)	12.9	8.0	1.71 (1.00–2.91)
Mobility aids n = 1823	11.0	2.7	4.53 (3.82–5.37)	9.8	4.2	2.48 (1.00–6.15)
Communication aids n = 898	8.1	0.6	14.99 (11.18–20.09)	26.4	14.6	2.09 (1.08–4.03)
At least 1 of the listed identified n = 39 020	96.7	94.9	1.60 (1.31–1.95)	22.8	7.4	3.71 (3.30–4.16)

CSHCN = children with special health care needs.
*If need was identified as present, the survey asked if that need had been met. Unmet need represents when the service was identified as needed and not met.
**PT = physical therapy; OT = occupational therapy
†OR = odds ratio; CI = confidence interval.

Table 3. Adjusted Estimated Prevalences and Adjusted Odds Ratios of Unmet Need for at Least 1 Health Service/Item

Characteristic	Adjusted Estimated Prevalences* of Unmet Need Percentage (95% CI†)	Adjusted Odds* of Unmet Need (95% CI)
Presence of disability		
No	4.5 (4.0–5.2)	REF‡
Yes	18.6 (17.0–20.3)	1.68 (1.45–1.94)
Gender		
Girls	5.7 (5.0–6.7)	REF
Boys	6.5 (5.8–7.4)	0.98 (0.85–1.13)
Age, y		
0–4	4.8 (3.9–6.0)	REF
5–13	6.3 (5.6–7.2)	1.14 (0.92–1.41)
14–17	6.7 (5.8–7.8)	1.26 (1.00–1.58)
Race/ethnicity		
White non-Hispanic	5.5 (4.9–6.3)	REF
Black non-Hispanic	7.2 (6.0–8.7)	0.73 (0.60–0.91)
Hispanic	8.8 (7.3–10.6)	0.92 (0.75–1.14)
Other	9.8 (7.8–12.2)	1.34 (1.05–1.72)
Income		
<200% FPL§	12.0 (10.8–13.2)	2.19 (1.73–2.77)
200%–399% FPL	5.6 (4.8–6.5)	1.43 (1.15–1.77)
≥400% FPL	3.3 (2.7–4.0)	REF
Insurance status		
Private	4.4 (3.8–5.1)	REF
Public	10.7 (9.6–12.1)	0.98 (0.80–1.20)
Private and public	12.7 (10.5–15.4)	1.10 (0.85–1.41)
Other insurance	5.4 (3.6–8.1)	1.01 (0.66–1.55)
Uninsured	25.4 (21.3–30.0)	3.03 (2.33–3.95)
Household composition		
Two parent	5.0 (4.4–5.8)	REF
Single mother	10.4 (9.2–11.6)	1.23 (1.05–1.44)
Other	7.6 (5.9–9.6)	1.08 (0.83–1.42)
Highest educational attainment in the home		
Less than high school	10.5 (8.2–13.5)	1.47 (1.09–1.98)
High school	7.6 (6.5–8.8)	1.05 (0.77–1.44)
More than high school	5.8 (5.1–6.6)	REF
Condition severity		
None/not applicable	1.5 (0.9–2.5)	REF
Minor	4.2 (3.6–4.9)	2.12 (1.28–3.50)
Moderate	13.6 (12.5–14.8)	4.35 (2.67–7.08)
Severe	32.0 (28.9–35.3)	8.37 (5.01–13.97)
Condition stability-health care needs		
Were usually stable	4.4 (3.8–5.1)	REF
Changed once in a while	10.9 (9.7–12.1)	1.42 (1.22–1.65)
Changed all the time	24.3 (20.7–28.3)	1.62 (1.27–2.07)
Care within the medical home		
Presence	2.6 (2.2–3.2)	REF
Absence	13.6 (12.5–14.8)	3.40 (2.87–4.03)

*Adjusted for all other variables in the model.
†CI = confidence interval.
‡REF = referent group.
§FPL = federal poverty level.

well-being.[29] There is also a need to study more closely the factors that contribute to the differential experience of psychosocial problems between CSHCN with disabilities and other CSHCN in order to intervene on behalf of children in terms of their psychosocial well-being.

We also found that CSHCN with disabilities also experience health care inequities when compared with other CSHCN. Despite having increased need for health services, CSHCN with disabilities had more unmet need and were less commonly receiving care within a medical home. The lack of assistance with care coordination was especially notable. Bramlett and colleagues[8] also found that CSHCN with functional limitations experienced health care inequities in terms of insurance adequacy. According to the behavioral model of health service use, a multitude of factors may contribute to health care inequities.[21,22,30] Our multivariable model points to condition severity as being the most strongly associated with unmet need. But even when controlling for condition severity, disability status was a predictor of unmet need, and a significantly higher percentage of CSHCN with disabilities had at least 1 unmet need than other CSHCN. This indicates that although attending to severity is important, examining health factors beyond the condition

itself is important for understanding health inequities. Both excess needs and excess unmet needs should be considered when tailoring programs and interventions to maximize the health and well-being of CSHCN with disabilities. Policies and practices that address unmet need are particularly relevant to CSHCN with disabilities because of the long-term potential impacts of unrealized access to care that could negatively impact health outcomes and participation in life events. Additionally, we found that the family characteristics of living in or near poverty, having lower educational attainment levels in the home, and living in single mother households increased the odds of unmet need. Practitioners should be cognizant of these risk factors because these families are often disadvantaged in a multitude of ways that may limit their success in their interactions with the health care system. Conversely, the enabling factors of having care within a medical home and having health insurance were associated with decreased odds of unmet need. These associations were expected and have been shown in the literature previously for CSHCN.[31,32] Thus, our findings add credence to the national call to address health insurance adequacy and care within a medical home for CSHCN with and without disabilities.

LIMITATIONS

In this study we used a screening tool through which parents/guardians identified children who had limitations in their ability to do the things that other children of the same age can do. The CSHCN Screener may not capture all children with disabilities, especially children with relatively mild functional limitations. Furthermore, by limiting our sample to CSHCN, we might have underestimated the number of children with disabilities. We note that the national estimates of disability in childhood vary substantially. Using 2000 Medical Expenditures Panel Survey (MEPS) data, Newacheck and colleagues[33] estimated that 7.3% of children have disabilities when *disability* is defined as a social role limitation or based on the receipt of special services. Nageswaran and colleagues[9] found that 60% of the estimated 12.8% CSHCN in the United States have functional limitations using the NS-CSHCN 2001. These differences indicate that defining and measuring disability in childhood likely requires refinement and consensus building for improved uniformity. And lastly, we used a cross-sectional survey for our analyses, thus we are limited in our ability to draw conclusions from the data because we are only able to identify associations. Further research is necessary to identify causal relationships between child, family and health systems factors and health care inequities for CSHCN with disabilities.

CONCLUSION

Our study highlights that the health and social challenges faced by CSHCN are more problematic for those with disabilities than those without. Furthermore, despite having more health services needs, CSHCN with disabilities have more unmet need and are not commonly receiving care within a medical home. Pediatric health providers should be cognizant of these findings and work to address the differences in health and health care delivery in their practices and community settings. Based on the differences noted between CSHCN with and without disabilities, we conclude that special attention needs to be given to those with disabilities to ensure that their health is maximized and the negative impacts of disability are minimized. Our findings also point to the need for continued research on this population to evaluate disparities and identify areas of intervention that successfully ameliorate the negative health and social consequences associated with disabilities and improve health services delivery and access.

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Pediatric mental health problems and associated burden on families

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Approximately 20% of children in the United States have mental health problems. The factors associated with childhood mental health problems and the associated burdens on families are not well understood. Therefore, our goals were to profile mental health problems in children to identify disparities, and to quantify and identify correlates of family burden. We used the National Survey of Children's Health, 2003 ($n = 85{,}116$ children aged 3–17 years) for this analysis. The prevalence, unadjusted and adjusted odds ratios (AOR) of mental health problems and family burden were calculated for children by child-, family- and health systems-level characteristics. The prevalence of mental health problems among children aged 3–17 years was 18%. The odds of mental health problems were higher for boys, older children, children living in or near relative poverty, those covered by public insurance, children of mothers with fair or poor mental health, children living in homes without two parents, children without a personal doctor or nurse and children with unmet health care needs. Among families with children with mental health problems, 28% reported family burden. Correlates of family burden included white race, severity, older age, higher income, non-two-parent family structure and having a mother with mental health problems. In conclusion, childhood mental health problems are common, and disproportionally affect children with fewer family and health care resources. Families frequently report burden, especially if the mental health problem is moderate to severe, but the correlates of family burden are not the same correlates associated with mental health problems. Understanding those highest at risk for mental health problems and family burden will help assist clinicians and policy makers to ensure appropriate support systems for children and families.

Keywords: children; disparities; family burden; mental health

Introduction

Mental health is defined as "how a person feels and acts when faced with life's situations" (National Mental Health Information Center & Center for Mental Health Services, 2003). In general, mental health problems in childhood refer to the broad range of emotional, behavioral and mental disorders that can affect children (Foy & Perrin, 2010). Approximately one in five children in the United States have an identified mental health problem, and mental health problems disproportionately affect certain minorities and the poor (Mark & Buck, 2006; Shaffer et al., 1996). Mental health problems among children and adolescents are recognized as a public health crisis in the United States

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(Huang & Mayberg, 2003). An additional area of concern is the impact of childhood mental problems on families (Huang & Mayberg, 2003). Previous research has demonstrated that childhood mental health problems are associated with social and economic burden on families (World Health Organization [WHO], 1999). Families of children with mental health problems experience higher rates of parental stress and grief with disruption to their families' lives (Doornbos, 1997). It is readily apparent that the negative impacts extend beyond home life to affect schools, employment and the greater community (Simpson, Bloom, Cohen, Blumberg, & Bourdon, 2005).

Unfortunately, little is known about the child-level, family-level and health systems-level factors associated with the experience of burden for families raising children with mental health problems. To our knowledge, no studies address both the prevalence of mental health problems in childhood and the associated impact on families. Tying together the experiences of children and their families is critical when planning for holistic programs that target both children and families in the context of their communities (Brauner & Stephens, 2006; Foy & Perrin, 2010; Kim, Viner-Brown, & Garcia, 2007; Waxman, 2006). Therefore, the objectives of this study are threefold: (1) to describe the prevalence of mental health problems among children aged 3–17 years by various child-, family- and health systems-level characteristics, (2) to analyze the factors that are related to having mental health problems to identify possible sociodemographic disparities, and then (3) to describe and determine correlates of family burden.

Methods

Data source

We used the National Survey of Children's Health (NSCH) 2003 for our analyses because the key variables of interest for this study were omitted from the 2007 version of the survey. The NSCH 2003 was a nationally representative cross-sectional telephone survey of US households sponsored by the Maternal and Child Health Bureau in partnership with the National Center for Health Statistics and the Centers for Disease Control and Prevention. The purpose of the NSCH was to produce prevalence estimates of health indicators and experiences with the health-care system for children younger than 18 years of age. One child in each household was randomly selected to be the subject of the survey, and the respondent for the interview was the parent or guardian who was most familiar with the child's health and health care. The survey included questions about demographics, health status, health insurance and access to and use of health-care services (van Dyck et al., 2004). Estimates reported here are based on 102,353 interviews completed from January 2003 to July 2004 (Blumberg et al., 2005; Kogan & Newacheck, 2007). For this analysis, the potential sample included 85,389 children between the ages of 3 and 17 years, as the question about mental health problems was not asked to caregivers of children under 3 years. Our final sample, $n = 85{,}116$, excluded 273 children for whom mental health data was missing.

Conceptual model

To determine which variables to utilize from the NSCH data set to address our hypotheses we developed a conceptual framework based a model of risk factors for special health-care needs put forth by Newacheck and colleagues (Newacheck, Kim, Blumberg, & Rising, 2008). Our conceptual framework places the child in the center of a series of concentric

and overlapping circles indicating progressively larger social spheres. In this model, child-level, family-level factors and health systems-level factors potentially impact the mental health of the child. We also note that how the family experiences the impact of caring for a child with mental health problems does not depend solely on the characteristics or severity of child's problems, but probably relates to a plethora of other factors, including availability and quality of health services. In this model, we are attempting to relate the sociodemographic and health-care characteristics of children to the experience of mental health problems and their impacts on families to identify and quantify potential disparities.

Study variables

Defining mental health problems and family burden

Children were identified as having a mental health problem if the survey responder answered affirmatively to the following question: "Overall, do you think that *sample child* has difficulties with one or more of the following areas: emotions, concentration, behavior, or being able to get along with other people?". This question is derived from the Strength and Difficulties Questionnaire (Blumberg et al., 2005) and the Child Behavior Checklist, both of which are used frequently in research to assess for emotional health and psychosocial problems in childhood (Achenbach & Ruffle, 2000; Goodman, 2001). Once the sample child was identified as having a mental health problem, the survey respondent was asked to rate the mental health difficulties as: "minor", "moderate" or "severe". For our analysis, we dichotomized the severity of mental health difficulties into two groups: mild and moderate/severe. To determine family burden, the survey respondents who affirmatively reported that their child had a mental health problem were asked: "Overall, would you say *sample child's* mental and emotional health puts a burden on your family a great deal, a medium amount, a little or not at all?". For this study, we considered responses "a great deal" or "a medium amount" to indicate the experience of family burden.

Child-, family- and health systems-level factors

Based on our conceptual model, we identified multiple independent variables of interest at the level of the child, family and health system. The child- and family-level characteristics of interest were age (3–11 years old and 12–17 years old), gender, race/ethnicity (white, black, Hispanic and non-Hispanic other), income categories based on Federal Poverty Level (FPL) designation (>200% FPL, 200–400% FPL and >400% FPL), insurance type and status (private coverage, public coverage, disrupted insurance during the past year or uninsured), region of the country (Northeast, Midwest, South and West), family structure (two-parent, single-parent or other) and the mental health status of mothers (excellent/very good, good or fair/poor). We also report on the systems-level factors of having an unmet need for health services and the presence of a personal doctor or nurse.

Statistical analysis

Estimates presented in the text and tables have been statistically weighted to reflect national population totals. For our data analysis, we first calculated descriptive statistics to generate the population estimates of having a mental health problem. Unadjusted odds (bivariate statistics) were calculated for the presence of mental health problems and experience of family burden for each of the independent variables (data not shown in the results).

We performed a multiple logistic regression to predict the adjusted odds for having a mental health-care problem. We then performed logistic regression for the experience of family burden, stratified by mental health problem severity. We stratified by severity because we felt that the burden experienced by families of children with more severe mental health problems would probably be different to the burden experienced by families of children with mild mental health problems. All estimates and analyses were performed using STATA 11 (STATA Corp., College Station, TX, USA) to account for the complex sample design of the survey. Our secondary data analysis falls under the exempt category by the University of California San Francisco Committee on Human Research.

Results

Prevalence of mental health problems

Nearly 11 million, or 18%, of children in the United States had mental health problems identified by their caregivers/parents. Table 1 summarizes the prevalence of mental health problems by child-, family- and health systems-level characteristics. There were disparities in the prevalence of mental health problems. Mental health problems were more commonly identified among blacks, children living below 200% of the FPL and children covered by public insurance. The disparities became more pronounced when evaluating children with moderate to severe mental health problems. Twice the proportion of children living below 200% FPL had moderate to severe mental health problems compared to children living above 400% FPL (12.6% vs. 6.1%, respectively). Similarly, the prevalence of severe mental health problems was twice as high for those covered by public insurance than those covered by private insurance (14.9% vs. 7.1%).

Prevalence of family burden

Among families of children with mental health problems, 28% reported family burden. The experience of family burden was more frequently identified in families of children with moderate–severe mental health problems compared to those with mild mental health problems, 45.1% vs. 9.6%, $p<0.0001$. Table 2 summarizes the prevalence of family burden stratified by severity. Among families of children with mild mental health problems, the highest prevalence of family burden was reported by caregivers of children with unmet health-care needs (16.0%). Conversely, caregivers of children with mild mental health problems without insurance reported the least burden (3.0%). Similar to the response pattern of caregivers of children with mild mental health problems, among families with children with moderate to severe mental health problems, those without insurance reported the least amount of family burden (30.9%) and those with unmet health-care needs reported the most burden (65.6%).

Adjusted odds of mental health problems by sociodemographic characteristics

The adjusted odds ratios (AOR) of having an identified mental health problem are presented in Table 3. After adjustment, the following characteristics were associated with higher odds of having mental health problems: male gender, older age, living below 400% FPL, having public insurance coverage, living in a home not headed by two parents, having a mother with fair or poor mental health, having unmet health-care needs and not having a personal doctor or nurse. Maternal mental health status was the strongest predictor of child

Table 1. Prevalence of mental health problems in children aged 3–17 years by child-, family- and health systems-level characteristics.

Population	Weighted prevalence of mental health problems % (SE)	Weighted estimate of mental health problems	Weighted prevalence of mild mental health problems % (SE)	Weighted prevalence of moderate/severe mental health problems % (SE)
Children 3–17 years	17.80% (0.24)	10,900,000	8.59% (0.18)	9.16% (0.17)
Age				
3–11 years	16.50% (0.31)*	6,018,000	8.49% (0.24)	7.96% (0.21)*
12–17 years	19.70% (0.38)	4,903,000	8.72% (0.26)	10.91% (0.30)
Sex				
Female	14.21% (0.32)*	4,252,000	7.29% (0.25)*	6.89% (0.22)*
Male	21.22% (0.35)	6,661,000	9.82% (0.26)	11.33% (0.27)
Race				
W-NH	17.10% (0.26)*	6,335,000	8.06% (0.19)*	9.00% (0.20)*
B-NH	24.68% (0.80)	2,165,000	13.03% (0.65)	11.55% (0.59)
Hispanic	15.08% (0.64)	1,541,000	6.70% (0.45)	8.32% (0.49)
O-NH	17.44% (1.08)	752,000	9.38% (0.93)	8.05% (0.63)
Income				
>400% FPL	13.33% (0.39)*	1,971,000	7.21% (0.31)*	6.12% (0.25)*
200–400% FPL	15.98% (0.37)	2,949,000	8.04% (0.28)	7.92% (0.26)
<200% FPL	23.18% (0.48)	5,091,000	10.47% (0.36)	12.62% (0.37)
Insurance				
Private	14.66% (0.25)*	5,817,000	7.57% (0.19)*	7.05% (0.18)*
Public	26.25% (0.59)	4,175,000	11.31% (0.44)	14.89% (0.46)
Disrupted	16.95% (1.16)	440,000	8.87% (0.91)	7.86% (0.77)
Uninsured	15.39% (1.10)	475,000	7.28% (0.84)	8.04% (0.77)
Region				
Northeast	17.75% (0.54)*	1,916,000	8.01% (0.37)*	9.68% (0.43)*
Midwest	17.37% (0.39)	2,394,000	8.13% (0.28)	9.14% (0.31)
South	19.15% (0.39)	4,247,000	9.42% (0.29)	9.69% (0.29)
West	16.19% (0.60)	2,364,000	8.18% (0.47)	8.00% (0.41)
Family structure				
Two-parent	14.62% (0.25)*	6,195,000	7.31% (0.19)*	7.27% (0.18)*
Single-parent	24.61% (0.59)	3,544,000	10.92% (0.43)	13.62% (0.47)
Other	23.21% (1.22)	670,000	11.64% (1.04)	11.41% (0.79)
Maternal mental health				
Excellent/very good/good	15.69% (0.24)*	8,329,000	7.93% (0.18)*	7.73% (0.17)*
Fair/poor	39.13% (1.31)	1,527,000	12.93% (0.94)	26.03% (1.14)
Personal doctor or nurse				
Yes	17.77% (0.26)	9,040,000	8.61% (0.19)	9.11% (0.19)
No	17.88% (0.64)	1,834,000	8.44% (0.45)	9.35% (0.49)
Unmet need for health services				
No	17.53% (0.24)*	10,600,000	8.51% (0.18)*	8.98% (0.18)*
Yes	38.98% (3.54)	208,000	4.65% (2.66)	23.46% (2.61)

Notes: *t-test or chi-squared test (for within sociodemographic group differences) significant at $p<0.001$. SE, standard error; FPL, Federal Poverty Level; W-NH, white non-Hispanic; B-WH, black non-Hispanic; O-NH, other non-Hispanic.

Table 2. Prevalence of family burden among children with mental health problems by child-, family- and health systems-level characteristics and stratified by severity.

	Percentage of families who experience family burden from their child's mental health problem % (SE)	Percentage of families who experience family burden when the child's mental health problem was mild % (SE)	Percentage of families who experience family burden when the child's mental health problem was moderate/severe % (SE)
All children	27.97% (0.66)	9.58% (0.72)	45.09% (1.00)
Age			
3–11 years	24.05% (0.87)*	8.48% (1.02)	40.50% (1.35)*
12–17 years	32.79% (1.00)	11.13% (0.96)	50.00% (1.45)
Sex			
Female	26.90% (1.07)*	10.81% (1.33)	43.83% (1.61)
Male	28.69% (0.84)	8.72% (0.77)	45.83% (1.27)
Race			
W-NH	30.11% (0.76)*	9.02% (0.67)	48.89% (1.17)*
B-NH	23.78% (1.65)	9.86% (1.98)	39.20% (2.61)
Hispanic	24.28% (1.98)	8.50% (1.76)	37.00% (3.00)
O-NH	29.54% (3.21)	14.88% (4.62)	46.56% (4.03)
Income			
>400% FPL	29.16% (1.42)	9.62% (1.53)	52.17% (2.09)*
200–400% FPL	27.05% (1.09)	8.96% (1.04)	45.41% (1.73)
<200% FPL	27.96% (1.05)	9.60% (1.24)	43.08% (1.54)
Insurance			
Private	26.93% (0.83)*	8.73% (0.76)*	46.30% (1.32)*
Public	31.05% (1.19)	12.07% (1.61)	45.39% (1.65)
Disrupted	24.20% (2.89)	7.94% (2.21)	42.37% (4.91)
Uninsured	17.55% (2.57)	2.98% (0.96)	30.91% (4.43)
Region			
Northeast	30.63% (1.57)*	9.01% (1.45)	48.31% (2.36)*
Midwest	29.73% (1.12)	9.33% (1.04)	47.70% (1.75)
South	24.95% (0.95)	8.75% (0.93)	40.68% (1.53)
West	29.45% (1.82)	11.66% (2.28)	47.62% (2.61)
Family structure			
Two-parent	25.77% (0.79)*	7.49% (0.67)*	44.05% (1.27)
Single-parent	30.34% (1.26)	11.56% (1.52)	45.44% (1.83)
Other	30.55% (2.67)	14.23% (3.42)	46.41% (3.61)
Maternal mental health			
Excellent/very good/good	25.54% (2.71)*	8.49% (0.71)*	42.91% (1.19)*
Fair/poor	39.50% (2.01)	14.75% (2.85)	51.86% (2.50)
Personal doctor or nurse			
Yes	28.63% (0.72)*	9.77% (0.81)	46.28% (1.07)*
No	24.79% (1.68)	7.25% (0.69)	39.34% (2.69)
Unmet need for health services			
Yes	46.27% (4.82)*	16.02% (5.82)	65.63% (5.70)
No	27.52% (0.67)	9.52% (0.73)	44.45% (1.02)

Notes: *t-test or chi-squared test to determine within sociodemographic group differences, $p<0.05$. SE, standard error; FPL, Federal Poverty Level; W-NH, white non-Hispanic; B-WH, black non-Hispanic; O-NH, other non-Hispanic.

mental health problems. Children of mothers with fair/poor mental health had nearly three times the odds of mental health problems compared to other children. Hispanic children had statistically lower odds (AOR = 0.60) of reporting mental health problems than white non-Hispanics.

Adjusted odds of family burden

As shown in Table 3, when controlling for all other factors in the multivariable model, only non-two-parent family structures and fair/poor maternal mental health were statistically associated with increased odds of family burden for families of children with mild mental health problems. Those without insurance and those living below 200% of the FPL had significantly decreased odds of family burden. For families of children with moderate to severe mental health problems, families of older children, children with unmet health-care needs and mothers with fair/poor mental health had higher odds of experiencing burden from their child's mental health problems. The adjusted odds of experiencing family burden among families of children with moderate to severe mental health problems were found to be lower among blacks, Hispanics and those living below 400% of the FPL.

Discussion

Mental health problems

This is the first study to simultaneously describe the prevalence of mental health problems in children and associated burdens on families, and to delineate factors associated with both mental health disparities in children and burden on families. Similar to previous studies that document mental health problems in children, we found that nearly 20% of children aged 3–17 years had mental health problems (Carter et al., 2010; Kim et al., 2007; Mark & Buck, 2006). We also found a prevalence of moderate to severe mental health problems of nearly 10%, which is similar to the results from the National Health Interview Survey (Mark & Buck, 2006). Our analysis identified important sociodemographic disparities in mental health, but not necessarily in the patterns identified in other studies (Costello et al., 1996; Ghandour, Kogan, Blumberg & Perry, 2010; Mark & Buck, 2006; Simpson et al., 2005). We found that children with mental health problems were disproportionately living in or near poverty, in non-two-parent homes and in families already affected by mental health problems. Similar to Mark and Buck (2006), we found that the prevalence of mental health problems was statistically higher for black children, but in our study, when other factors were controlled for in the multivariable model, minority racial status was no longer statistically significant. In our analysis, Hispanic race was actually associated with decreased odds of having a mental health problem. Lower reports of mental health problems among Hispanic families may be due to true differences in prevalence, differences in cultural understandings of mental health problems, the "immigrant paradox" in which recent immigrants experience fewer psychiatric symptoms despite the stress of immigration compared to acculturated immigrant groups, or over-reporting by other groups (Alegria et al., 2008; Crijnen, Achenbach, & Verhulst, 1999; Mendoza, 2009; Teagle, 2002).

In addition to elucidating income disparities, this study also identified important associations between mental health problems and health-systems factors. Children with public insurance, children with unmet health-care needs and children without a personal doctor or

Table 3. Adjusted odds ratios of having mental health problems and experiencing family burden stratified by severity.

	Adjusted odds of having a mental health problem (CI)	Adjusted odds of family burden among families of children with mild mental health problems (CI)	Adjusted odds of family burden among families of children with moderate to severe mental health problems (CI)
Age			
3–11 years	Ref.	Ref.	Ref.
12–17 years	**1.20 (1.12–1.29)**	1.39 (1.00–1.93)	**1.32 (1.10–1.59)**
Sex			
Female	Ref.	Ref.	Ref.
Male	**1.71 (1.59–1.83)**	0.97 (0.68–1.37)	1.18 (0.98–1.42)
Race			
W-NH	Ref.	Ref.	Ref.
B-NH	1.02 (0.91–1.15)	0.73 (0.45–1.18)	**0.63 (0.48–0.83)**
Hispanic	**0.60 (0.53–0.68)**	0.97 (0.55–1.70)	**0.65 (0.47–0.89)**
O-NH	0.92 (0.79–1.08)	1.78 (0.83–3.56)	0.81 (0.56–1.19)
Income			
>400% FPL	Ref.	Ref.	Ref.
200–400% FPL	**1.15 (1.06–1.26)**	**0.51 (0.28–0.93)**	**0.64 (0.49–0.83)**
<200% FPL	**1.35 (1.21–1.51)**	0.82 (0.55–1.23)	**0.74 (0.59–0.92)**
Insurance			
Private	Ref.	Ref.	Ref.
Public	**1.60 (1.44–1.78)**	1.58 (0.93–2.53)	1.20 (0.95–1.52)
Disrupted	1.03 (0.85–1.25)	0.72 (0.33–1.38)	0.96 (0.60–1.63)
Uninsured	1.04 (0.84–1.29)	**0.29 (0.12–0.72)**	0.67 (0.39–1.14)
Region			
Northeast	Ref.	Ref.	Ref.
Midwest	0.96 (0.87–1.07)	1.21 (0.79–1.88)	1.05 (0.82–1.38)
South	1.01 (0.91–1.12)	1.28 (0.84–1.97)	0.81 (0.63–1.04)
West	0.99 (0.87–1.12)	1.36 (0.84–2.21)	1.06 (0.78–1.44)
Family structure			
Two-parent	Ref.	Ref.	Ref.
Single-parent/other	**1.47 (1.35–1.61)**	**1.82 (1.25–2.65)**	1.23 (1.00–1.51)
Maternal mental health			
Excellent/very good/good	Ref.	Ref.	Ref.
Fair/poor	**2.80 (2.46–3.18)**	**2.03 (1.23–3.35)**	**1.53 (1.21–1.94)**
Personal doctor or nurse			
Yes	Ref.	Ref.	Ref.
No	**1.18 (1.06–1.32)**	1.05 (0.65–1.72)	0.86 (0.65–1.15)
Unmet need for medical care			
No	Ref.	Ref.	Ref.
Yes	**2.32 (1.67–3.22)**	1.13 (0.40–3.18)	**2.40 (1.37–4.21)**

Notes: Ref., referent group; CI, confidence interval; FPL, Federal Poverty Level; W-NH, white non-Hispanic; B-WH, black non-Hispanic; O-NH, other non-Hispanic. Adjusted odds are adjusted for all other variables in the model. Odds ratios shown in bold type are statistically significant.

nurse had higher odds of mental health problems. The relationship between health insurance type and the presence of mental health problems is most certainly complex, because of issues regarding mental health parity and out-of-pocket expenses for treatments that may effectively manage mental health problems (Barry & Busch, 2007). Regardless of insurance type, reports of unmet need for mental health services are substantial. Among children covered by private insurance, 36.8% [confidence interval (CI) 34.0–39.6] reported unmet need for mental health services compared to 41.4% (CI 37.7–45.1) of publicly insured children (Child and Adolescent Health Measurement Initiative, 2003). These high rates of unmet need for children covered by either public or private insurance highlight the inadequacies of the current payment structures to meet the needs of children with mental health problems. The issues of limited access to needed services are further compounded by the experience of living in or near poverty for many children covered by public insurance (Ganz & Tendulkar, 2006). The findings in our study, taken together with the existing literature that identifies the limitations of mental health-care delivery for children, indicate the need for advancing care delivery and policies. This points to the potential role that a community-based system of care could play in the mitigation of mental health problems through family-centered comprehensive care within a medical home (Perrin et al., 2007), coupled with strategies to apply a population perspective to mental health needs and service delivery (Foy, 2010).

Family burden

The experience of family burden was common. As identified in other studies we found that caregivers of children with more severe mental health problems report more burden than those with children with mild mental health problems (Doornbos, 1997; Kim et al., 2007). Our results are also similar to the results found by Teagle (2002), who reported family impacts for 32% of families with children with more than one psychiatric diagnosis. An unexpected finding in our analysis is the sociodemographic distribution of family burden. We anticipated that the pattern of factors associated with mental health problems would be the same for family burden. Under that expectation, the families of children who were more likely to experience family burden would be demographically similar to those with mental health problems, that is, living in or near poverty. This was not what we found in our analysis. For families of children with moderate to severe mental health problems, the odds of burden were actually lower for families living below 400% of the FPL. The odds of family burden were also lower for blacks and Hispanics compared to whites. Among families of children with mild mental health problems, the odds of family burden were lower when the child was without health insurance. While there is no clear explanation for these differences, we consider the possibility that minority families and those living in or near poverty may experience other competing social and economic stressors that could lessen the perceived impact that mental health problems have on families. The experience of poverty is exceptionally complex, and includes a conglomerate of stressful situations and conditions (McLoyd, 1990) which may diminish the impact of mental health problems on families. Similarly, the context in which a child is uninsured is probably quite stressful for families, and a mild mental health problem might not add substantially more family burden. In contrast, non-minority families and families living well above the FPL are less likely to experience chronic economic and social stressors, and therefore the burden of childhood mental health problems may be more acutely felt. Another possible explanation is that cultural differences in the ways families deal with stressors, such as the reliance on extended family support, account for the differences in the experience of family burden.

Unfortunately, these possibilities cannot be studied using the NSCH and therefore further research is necessary to determine how different levels of social and economic stress and informal support systems impact perceived family burden. Additionally, research is warranted to correlate our findings with disparities in access to care, out-of-pocket expenses and other types of family burden, such as job loss. Furthermore, studying whether the medical home and other models of service delivery provide some protection from unmet health care needs would help to guide public policies and promote community-based supports.

Limitations

Although the NSCH is the largest comprehensive study of children's health, there are several limitations. The NSCH is a cross-sectional study, and therefore we are able to present associations and cannot determine directionality of the association in terms of causality. In addition, our definition of mental health problems (problems with emotions, concentration, behavior or being able to get along with other people) is an accepted but not a standardized definition which makes comparisons across different studies difficult. We also cannot address issues related to specific mental health conditions. Furthermore, this study uses parental/caregiver report to determine the presence of mental health problems which were not verified by a health-care professional and may be reported differently by parents from different cultures (Crijnen et al., 1999). This may lead to reporting bias, although we note that parental report of mental health problems is highly correlated with practitioner verification (Glascoe, 2003). The experience of family burden is also subjective and may be mediated by families' cultural backgrounds, especially as burden was not specifically defined in the survey question. Lastly, we assessed only a small number of factors that might relate to families' experiences of burden.

Conclusions

Mental health problems impact a substantial number of children and their families. Addressing the mental health needs of children is a priority area for the American Academy of Pediatrics (AAP, 2008) and reducing mental health disparities is a major goal of Healthy People 2010 (US Department of Health and Human Services, 2005). As the AAP Task Force on the Family points out, our current social and public policies are not meeting the needs of families, leaving them stressed to meet their responsibilities (Schor, 2003). An important mechanism to address existing mental health disparities would be through the reallocation of preventative and mental health services to ensure that those lacking health-care resources can receive adequate preventative services and mental health care (Foy & Perrin, 2010; Inkelas, Raghavan, Larson, Kuo, & Ortega, 2007; Sturm, Ringel, & Andreyeva, 2003). By identifying factors associated with mental health problems and family burden, we have set the stage for the development of targeted health service delivery interventions that could help to minimize the experience of mental health problems, maximize the care for these children and reduce the burden experienced by families.

Disclaimer

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The Family Impacts of Proton Radiation Therapy for Children With Brain Tumors

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Abstract

Children with brain tumors experience significant alterations to their health and well-being due to the tumors themselves and oncologic treatment. Caring for children with brain tumors can have significant impacts on families, especially during and shortly after treatment. In this study of the impacts on families caring for children undergoing proton radiation therapy for brain tumors, the authors found that families experienced a broad array of negative impacts. Families reported feeling like they were living on a roller coaster, feeling that others treated them differently, and having to give up things as a family. In the multivariable linear regression model, older age of the child and higher reported child health–related quality of life were associated with less family impact. The presence of concurrent chemotherapy was associated with increased family impact. This is the first study to specifically evaluate the families of children being treated with proton radiation therapy. The findings in this study are consistent with the findings in other studies of children treated with standard therapy that show that families experience a variety of stressors and negative impacts while their children are receiving treatment. Health care providers should be aware of the potential impacts on families of children with brain tumors and their treatment to provide robust services to meet the health, psychological, and social needs of such children and their families.

Keywords

family impact, brain tumors, proton radiation, quality of life

Introduction

Malignant tumors of the brain account for approximately 20% of all pediatric cancers, second only to leukemia in incidence (Fangusaro & Chi, 2009; National Cancer Institute, 2009). Every year, approximately 2500 children are diagnosed with brain tumors (Li, Thompson, Miller, Pollack, & Stewart, 2008; National Cancer Institute., 2009). Low-grade astrocytomas make up about half of all childhood brain tumors (Baldwin & Preston-Martin, 2004; Fangusaro & Chi, 2009; Levy, 2005), whereas medulloblastomas are the most common high-grade brain tumor and the second most common brain tumor in children (Dhall, 2009; Fangusaro & Chi, 2009; Rickert & Paulus, 2001). Treatment of pediatric brain tumors has dramatically improved over the past few decades with improvements in the survival rates as well. Survival rates for pediatric brain tumors are approaching 70% at 5 years (Jemal et al., 2008). Many pediatric brain tumors require aggressive intervention consisting of one or more of the following: surgery, chemotherapy, or radiotherapy. Because the various treatments can cause a variety of side effects that manifest years later, great emphasis has been placed in the past decade on decreasing the side effects associated with the treatment.

Surviving children can be left with substantial deficits and alterations to their quality of life (Mulhern & Butler, 2004; Mulhern, Merchant, Gajjar, Reddick, & Kun, 2004). Neurological sequelae result from the direct and indirect effects of the tumors themselves and the effects of their treatment (Ullrich, 2009). The treatments themselves can also lead to neurological deficits, and in

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particular, radiotherapy has been implicated in causing cognitive dysfunction, endocrine abnormalities, impaired hearing, behavior and adaptation difficulties, and secondary tumors (Fangusaro & Chi, 2009; Levy, 2005; Mulhern & Butler, 2004; Mulhern et al., 2004; Ullrich, 2009). Newer treatment options, including proton radiation therapy, attempt to mitigate the negative consequences by providing more targeted, tissue-sparing therapy. However, there are only a few centers offering this promising therapy in the United States; patients and their families usually need to travel to a proton center and stay for approximately 2 months in order to avail of it. How this treatment and the displacement from home affects families is an area for clinical research inquiry. To date, no studies have evaluated the family impacts for children with brain tumors treated with proton radiation therapy.

It is well documented that the diagnosis, treatment, and sequelae of pediatric brain tumor treatment can have profound effects on families (Hutchinson, Willard, Hardy, & Bonner, 2009). The experiences of parents of children with brain tumors are unique do to the complexities and uncertainties associated with the treatment, the constellation of long-term consequences, and the risk of relapse or secondary malignancy (Hutchinson et al., 2009). Much of the existing literature focuses on specific psychiatric and psychological consequences (Barakat et al., 1997; Fuemmeler, Mullins, & Marx, 2001; Hardy et al., 2008; Kazak, Boeving, Alderfer, Hwang, & Reilly, 2005; Patino-Fernandez et al., 2008; Vrijmoet-Wiersma et al., 2008). Caregivers of children with chronic needs frequently report impairments in family functioning, time constraints, poor health, chronic sorrow, depressive symptoms, and anxiety (Aitken et al., 2009; Brannan & Heflinger, 2006; Dodgson et al., 2000; Fleming et al., 1994; Knafl, Breitmayer, Gallo, & Zoeller, 1996; Reichman, Corman, & Noonan, 2008). Medical uncertainty, an inherent part of brain tumor treatment, is often cited as a major stressor for families (Dodgson et al., 2000; Garwick, Patterson, Meschke, Bennett, & Blum, 2002; Hutchinson et al., 2009; Stewart & Mishel, 2000).

In this article, we seek to examine the negative family impacts as measured by the Impact on Family Scale (IOFS) for parents of children with brain tumors who have sought out, and in many cases, traveled to a proton center to receive this special form of treatment. This work intends to complement existing research examining the quality of life for children treated with proton radiation therapy for brain tumors. Additionally, as this promising therapy gains traction and more pediatric patients requiring therapy will need to travel to receive it, this article will uniquely add to the literature on caregiver stress and strain. We hypothesize that families of children with tumors that require additional treatment beyond proton radiation therapy and families of children with lower quality-of-life scores will experience more substantial negative family impacts.

Methods

Study Description

As part of a longitudinal quality-of-life study, we assessed the impact of the tumor and treatment on families. We approached all English- or Spanish-speaking pediatric patients and their parents between the ages of 2 and 18 years treated with proton radiation at Massachusetts General Hospital (MGH), during the first 2 weeks of the start of radiation, at the end of radiation, and annually thereafter. We collected data for patients treated from March 2004 to March 2010. A total of 285 children treated with protons were offered enrollment, of whom 242 (85%) agreed to participate. Children who had suffered relapse or died were excluded from this cohort. Of the 242 children in the sample, 142 had a brain tumor, and among those, 96 had complete data for the family impact measure and the key independent variables. We recruited parents as proxy respondents for their assessment of their children's quality of life; this assessment was completed at the end of the radiation treatment. When both parents were available, we asked the parent who spent the most time with the child to complete the survey. Study staff approached families in the radiation waiting room and offered to meet with them in a private space. After giving consent, parents filled out the parent proxy forms, and children filled out the self-report forms. The research assistant was available to read the questions to the children. Assessments were conducted in English and Spanish as appropriate. This study was approved by the University of California San Francisco Committee on Human Subjects and by the MGH's internal review board.

Impact on Families

The main outcome of interest is the impact on families. The IOFS was administered to families at the end of proton radiation therapy for all enrollees. The original IOFS was a 33-item questionnaire designed by Drs Stein and Risessman (1980) to measure the impacts of chronic childhood illnesses on families. Families self-report level of agreement from *strongly disagree* to *strongly agree* with each of the survey statements. These statements are scored 1 to 4 to determine the amount of impact parents experience in 4 domains: financial burden, family/social impacts, personal strain, and mastery (Stein & Riessman, 1980). Since the development of the original scale, additional validation studies support the use of 15 of the 33 questions

to assess the overall negative personal, social, and familial impacts of childhood illness (Stein & Jessop, 2003; Williams, Piamjariyakul, Williams, Bruggeman, & Cabanela, 2006). Therefore, our outcome is the total score on the 15 items. The maximum obtainable score is 60, with a higher score indicating more negative impacts on the family.

Child Characteristics

Demographic characteristics, disease-specific and treatment-specific data, as well as quality-of-life data for the child subject were collected on all subjects in the study. Demographic characteristics and disease-specific data were collected on enrollment in the study. Treatment data were gathered from the radiation clinical information system. Quality of life was assessed using parent proxy for the PedsQL (Pediatric Quality of Life Inventory) Core module, Brain Tumor module, and Cancer module.

The PedsQL measures health-related quality of life in children and adolescents and consists of generic brief core scales suitable for use with both healthy populations and populations with acute and chronic health conditions. Physical (8 items), emotional (5 items), social (5 items), and school (3-5 items) functioning are measured. Parents respond based on a 5-point Likert-type scale from 0 (*never*) to 4 (*almost always*). The PedsQL has strong psychometric properties and has been used on a wide variety of healthy and chronically ill populations (Palmer, Meeske, Katz, Burwinkle, & Varni, 2007; Varni, Burwinkle, Katz, Meeske, & Dickinson, 2002; Varni, Seid, & Kurtin, 2001). Subscales include physical and psychosocial functioning (emotional, social, and school functioning). The PedsQL Brain Tumor module (PedsQL-BT) is similar to the PedsQL, but it consists of the following 6 scales: (a) pain and hurt, (b) nausea, (c) procedural anxiety, (d) worry, (e) cognitive problems, and (f) movement and balance (Palmer et al., 2007). We also administered 3 scales from the PedsQL Cancer module that were not part of the PedsQL-BT. They are (a) treatment anxiety, (b) perceived physical appearance, and (c) communication. All quality-of-life scores are scaled from 0 to 100, with higher scores indicating better health-related quality of life.

Statistical Analysis

Descriptive statistics were calculated to characterize the study population, their disease parameters and treatment, and their quality of life. The mean family impact score was calculated, as was the percentage of families reporting agreement with the impact statements. The bivariate relationships between the parental proxy report of the child's quality of life, demographics and child disease characteristics, and the outcome of family impact were calculated. Factors with statistically significant bivariate relationships were included in the multivariate regression model of family impact. All analyses were conducted using SAS (version 9.1; SAS Institute Inc, Cary, NC).

Results

Disease and Treatment Characteristics

Table 1 describes the study population. More than three quarters of the children were white, and more than 50% were male. The age distribution ranged from 2 to 17 years. For the 96 children for whom full clinical data were available from the records, the disease and treatment characteristics are also reported in Table 1. Among the children enrolled in this study, the most frequently reported brain tumor type was medulloblastoma. Approximately half of the children had tumors located in the posterior fossa. All but 3 children had operative intervention, with 63.5% achieving gross total resection. Approximately two thirds of the children were treated with chemotherapy during the study, and 94.8% were treated with high-dose radiation. Only 13.4% required ventriculoperitoneal shunts or third ventriculostomies for hydrocephalus management.

Quality of Life

Parent proxy reports of quality-of-life scores ranged from 60.3 to 83.2, as shown in Table 2. The procedural subscore was notably lower than all other quality-of-life measures (60.3). The highest rated quality-of-life subscore was the Core Social score, 83.2. The average School score was 70, the lowest of the subscores within the Core module. Parent proxies reported high quality-of-life subscores for movement and balance (81.1), pain/hurt (82.8), treatment anxiety (82.4), cognitive problems (80.5), and perceived physical appearance (81.8).

Family Impact

The average IOFS score was 35.3 (SD = 7.8), indicating that families experienced negative social and familial impacts from their children's tumors and treatment, as shown in Table 3. A majority of parents reported fatigue, having to give things up, living on a roller coaster, feeling that they are treated differently, and wondering if they should treat their child specially. Although 85% of families traveled to the center for treatment, only 50% reported strain from traveling to the hospital. Less than one third of families endorsed little desire to go out, difficulty finding reliable child care, thinking about not

Table 1. Demographic, Disease and Treatment Characteristics of Children With Brain Tumors Treated With Proton Radiation Therapy

Demographic Characteristics	n	Percentage
Total	106	100
Age at beginning of treatment (years)		
2-4	21	19.8
5-7	32	30.2
8-12	34	32.1
13-17	19	17.9
Gender		
Female	47	44.3
Male	59	55.7
Race		
White	83	78.3
Other	19	17.9
Not recorded	4	3.8
Disease and Treatment Characteristics	**n**	**Percentage**
Total	106	100
Tumor type		
Astrocytoma/glioma/ neurocytoma/ craniopharyngiomas	27	28.1
Ependymoma	20	20.8
Germ cell tumors	10	10.4
Medulloblastoma/PNET	39	40.6
Tumor location		
Posterior fossa	50	52.1
Other	46	47.9
Tumor management surgery type		
Gross total resection	61	63.5
Subtotal or near resection/ biopsy/no surgery	35	36.5
Shunt for hydrocephalus		
Yes	5	5.2
No	91	94.8
Chemotherapy		
Yes	62	64.6
No	34	35.4
Radiation dose		
Low (<45)	5	5.2
High (45+)	91	94.8
Radiation type		
Craniospinal	41	42.7
Noncraniospinal	55	57.3

having more children, and having no one who understands their burden. Notably, although 76.7% of families reported living on a roller coaster, only 42.2% reported living from day to day, and 49.6% reported needing to change plans at the last minute.

Table 2. PedsQL Scores: Parent Proxy Reported During Treatment

	Parent proxy	
	n	Mean (SD)
PedsQL Core module		
PedsQL Core Total score	90	73.5 (16.6)
PedsQL Core Physical Health score	90	70.6 (21.3)
PedsQL Core Psychosocial Health score	89	75.8 (15.8)
PedsQL Core Emotional score	90	72.9 (19.3)
PedsQL Core Social score	89	83.2 (18.0)
PedsQL Core School score	59	70.0 (22.0)
PedsQL Tumor/Cancer module		
PedsQL Tumor Total score	90	78.3 (13.0)
Pain/hurt	90	82.8 (17.9)
Nausea	89	78.9 (21.7)
Procedure anxiety	90	60.3 (33.3)
Movement and balance	56	81.1 (22.3)
Treatment anxiety	89	82.4 (22.0)
Worry	89	79.0 (25.5)
Cognitive problems	55	80.5 (17.8)
Perceived physical appearance	88	81.8 (20.2)
Communication	89	78.1 (25.1)

NOTE: SD = standard deviation; PedsQL = Pediatric Quality of Life Inventory (possible score range = 0-100, with higher score reflecting better quality of life).

Table 4 shows the unadjusted and adjusted relative risks of family impact by demographic, disease, treatment, and child quality-of-life variables. In the unadjusted analysis, higher average IOFS scores were associated with non-white race and treatment with chemotherapy. Lower average IOFS scores (less burden) were associated with older age of child, medulloblastoma, noncraniospinal radiation, and higher reported quality of life on both the Core and Tumor modules. For every additional point on the PedsQL Core module parent proxy, the average IOFS score was 0.14 points lower. Similarly, for every additional point on the PedsQL Tumor/Cancer module parent proxy, the average IOFS score was 0.21 points lower. The multivariate regression model included the independent variables that were significant in the bivariate analyses: age, race, tumor type, tumor location, the need for chemotherapy, radiation

Table 3. Average Family Impact Score by Item

Family Impact Questionnaire[a]	Percentage Agree or Strongly Agree	Mean (SD)
Fatigue is a problem because of illness	55.0	2.5 (0.8)
See family and friends less	43.0	2.2 (0.9)
Need to change plans at the last minute	49.6	2.4 (0.9)
Little desire to go out	28.5	2.0 (0.9)
No time for other family members	47.0	2.3 (0.8)
Live from day to day	42.2	2.3 (0.8)
Hard to find reliable person to care for child	19.4	1.9 (0.8)
Family gives up things	60.0	2.6 (0.8)
Nobody understands the burden	27.9	2.1 (0.8)
Can't travel out of the city	48.5	2.3 (1.0)
Live on a roller coaster	76.7	3.1 (0.8)
People treat us as special	70.4	2.8 (0.8)
Traveling to the hospital is a strain	50.0	2.5 (0.8)
Think about not having more children	26.5	1.9 (1.1)
Wonder whether to treat child specially	50.4	2.5 (0.8)
Total (max = 60)	N/A	35.3 (7.8)

a. Each item has a possible score of 1 to 4, with 4 indicating more family impact.

type, and PedsQL values. In this model, older age remained significantly related to the IOFS, such that for every additional year of age, the IOFS score was 1.79 points lower. This indicates that the parents of older children endorsed less family burden after controlling for other factors in the model. In addition, for every 1-point increase in the PedsQL Tumor/Cancer module, there was a 0.23-point decrease in the IOFS scale, indicating that higher quality of life was significantly associated with lower reports of family burden after controlling for other factors. Conversely, families of children treated with chemotherapy endorsed more family burden. Compared with families of children who did not require chemotherapy, families of children treated with chemotherapy had IOFS scores 4.83 points higher.

Discussion

This study is the first of its kind to detail the family impacts for families of children treated with proton radiation therapy for brain tumors. Parents identified various impacts—personal, familial, and social. Most notably, more than three quarters of parents reported feeling like they live on a roller coaster, although fewer families felt like they were living from day to day. There was also high agreement with feeling that people treat them specially and having to give things up because of their child's condition. On a positive note, a large majority of families felt that they could find reliable child care, still desired to go out, and did not worry about having more children. Similarly, only slightly more than one quarter of parents reported that no one understood their burden.

The level of impact endorsed by families in this study was overall similar to the level reported by families caring for adolescents with cancer (Sawyer, Antoniou, Toogood, & Rice, 1999). Sawyer et al. found that families of adolescents undergoing active treatment for cancer reported an average score of 36.6 on the combined personal strains and family/social subscales. They also reported an average score of 42 for families of children off therapy (Sawyer et al., 1999). Although direct comparisons to our study cannot be made because the populations of children in each study differed substantially, Sawyer et al.'s findings indicate that, in general, families of children with cancer experience negative family impacts.

Because of the negative family impacts experienced by children with cancer, providers should be aware of how families are experiencing the diagnosis, treatment, and follow-up of childhood brain tumors. Much of clinical practice is focused on the problems experienced by patients, but in the case of pediatric chronic disease, it is especially important to attend to the experiences of families (Witt et al., 2010). Addressing this aspect of family-centered care would likely enhance the care experiences of families because there may be areas for intervention to help families adjust. For example, families who report living on a roller coaster might benefit from increased anticipatory guidance and a clear timeline for follow-up. Families that struggle with social limitations, such as seeing family and friends less, having difficulty with child care, and feeling that they cannot travel, might benefit from social worker support or counseling to determine if alternatives exist. Similarly, parents who feel that there is little time for other family members and that the family has to give up certain things may benefit from sibling support groups and designing family plans to incorporate the needs of the whole family. Parents endorsing items that are often associated with depression, such as fatigue and having little desire to go out, may benefit from further evaluation and a referral to a mental health specialist. Clinics might also consider helping families with other aspects of daily life to alleviate some of the stressors families often experience. Concrete

Table 4. Factors Associated With Increased Risk of Family Impact: Relative Risk and Adjusted Relative Risk

Factor	Relative Risk (Confidence Intervals)	Adjusted Relative Risk (Confidence Intervals)[a]
Age at beginning of treatment	−1.48 (−2.96, 0.00)	**−1.79 (−3.44, −0.15)**
Gender		
Male	Reference	—
Female	−1.75 (−4.77, 1.26)	N/A
Race		
White	Reference	Reference
Other racial groups	5.17 (1.31, 9.03)	3.68 (−0.42, 7.79)
Tumor type		
Medulloblastoma/PNET	−0.35 (−7.40, −1.30)	−5.08 (−11.82, 1.67)
Tumor location		
Posterior fossa	−3.66 (−6.67, −0.65)	2.16 (−2.64, −6.96)
Other	Reference	Reference
Tumor management surgery type		
Gross total resection	Reference	—
Near or Subtotal/biopsy/none	−1.07 (−4.29, 2.14)	N/A
Shunt for hydrocephalus		
Yes	1.18 (−5.93, 8.29)	N/A
No	Reference	—
Chemotherapy		
Yes	6.07 (3.07, 9.06)	**4.83 (0.59, 9.08)**
No	Reference	Reference
Radiation dose		
Low (<45)	Reference	N/A
High (45+)	2.14 (−4.83, 9.10)	
Radiation type		
Craniospinal	Reference	Reference
Noncraniospinal	−3.68 (−6.72, −0.63)	2.66 (−2.80, 7.84)
PedsQL Core module	−0.14 (−0.24, −0.04)	.03 (−0.10, 0.17)
PedsQL Tumor/Cancer module	−0.21 (−0.34, −0.09)	**−.24 (−0.40, −0.08)**

NOTE: PedsQL = Pediatric Quality of Life Inventory; N/A = not applicable. Entries in boldface indicate statistical significance in the adjusted model.
a. Adjusted model includes the following variable: age, race, tumor type, tumor location, treatment with chemotherapy, radiation type, and PedsQL Core and PedsQL Tumor/Cancer modules.

resources such as travel vouchers, letters to employers, and even parking passes could help mitigate some of the strain of traveling for treatments and dealing with unexpected problems as they arise. Clinical programs and policies that encourage parental screening, coupled with appropriate referrals and resource assistance, should be incorporated into the care of children with brain tumors, with the intention of improving outcomes for families (Witt et al., 2010).

As one might expect, when the child's quality of life was deemed poorer, the negative impact on families was more substantial. In the regression analysis, after controlling for other factors, the child's quality of life was inversely associated with family impacts such that as quality of life was rated higher, the family impacts were scored as less of a problem. Therefore, mechanisms that might improve the quality of life of children undergoing tumor treatment, such as proton radiation, might attenuate the negative family impacts of childhood brain tumors. Because our study does not have a control population of children treated with standard photon radiation therapy, we are unable to elucidate the relationship between treatment type, quality of life, and impact on families. Therefore, we are unable to comment on the direct impact of proton therapy. Nonetheless, improving childhood outcomes through enhanced therapeutics and addressing the psychosocial issues that arise during and after treatment should be associated with improved family outcomes. We also found that families of children requiring concurrent chemotherapy experience more negative impacts. This is not surprising because these children likely have more severe disease. Therefore,

providers should be aware of the potentially added burden on families when the tumor requires more advanced treatment.

The results of this study support findings from other studies that indicate that families of children with brain tumors and other cancers struggle with family functioning (Lähteenmäki, Sjöblom, Korhonen, & Salmi, 2004; Witt et al., 2010). Our study also adds to the existing literature that focuses on the psychological impacts of childhood cancers on parents. Posttraumatic stress has been identified by several researchers (Barakat et al., 1997; Fuemmeler et al., 2001; Kazak et al., 2005). For example, Fuemmeler and colleagues (2001) found that 42% of parents met the diagnostic criteria for posttraumatic stress disorder. Symptoms of posttraumatic stress were reported by nearly all families in a study by Kazak et al (2005). Although our study does not specifically capture the diagnostic criteria of posttraumatic stress, parents did report familial stresses. Hutchinson and colleagues (2009) found that much of the burden experienced by parents/caregivers was internalized in the form of uncertainty, guilt, and worry. Our study findings of living on a roller coaster and living from day to day indicate that some parents are concerned about uncertainty. Half of the parents in this study also worried about how to treat their children, and more than one fourth think of not having more children, indicating ongoing worry and concern. These findings, taken with the existing literature, indicate that there are likely several areas for successful intervention to help families adapt and adjust to the chronicity and uncertainty of childhood brain tumors.

Limitations

This project has several limitations. It is important to note that although the IOFS is well validated, it does not address all the potential impacts on families. This measure was not intended to capture issues such as posttraumatic stress or parental depression. Furthermore, we cannot determine whether the impacts result from the disease itself or its treatment; nor do we have a comparison group of children treated with standard radiotherapy. Therefore, we cannot attribute the impact on families to proton radiation therapy. Another important limitation is that this study is cross-sectional in design. Family impacts may change over time, especially as the treatment phase ends and families have to deal with the long-term sequelae of brain tumors and their treatment. Evaluating this change is a part of the longitudinal study from which the data for this project were derived. And last, although the clinical site of this study has numerous support systems in place for families, we are unable to decipher which families used these supports and therefore cannot determine the potential benefit of these supportive interventions.

Future Research

This project provides a snapshot of the family impacts of childhood brain tumors after diagnosis and initial treatment. Because there are significant sequelae of brain tumors and their treatment for children, the family impacts are likely to persist and change over time (Van Dongen-Melman, Van Zuuren, & Verhulst, 1998). Therefore, it is important to longitudinally follow children and their families to determine how family impacts change over time. Additional research could also include an evaluation of the relationship of neurocognitive deficits and physical limitations with family impacts, as one might hypothesize that families of children with more substantial impairments would experience more family impacts. Furthermore, studies that link family impact with parental stress, physical and mental health, and quality of life would benefit our understanding of how best to mitigate the negative experiences from pediatric brain tumors, their treatment, and their sequelae.

Conclusion

This study is the first of its kind to evaluate the family impacts of brain tumors and their treatment on a cohort of children treated with proton radiotherapy. We identified several negative consequences endorsed by families, including uncertainty, social restrictions, and concerns for personal and family well-being. These impacts are likely not unique to families of children treated with proton radiation and have been endorsed in other cohorts (Hsieh, Huang, Lin, Wu, & Lee, 2009; Sawyer et al., 1999). Therefore, this study is likely applicable to families of children with brain tumors treated with standard therapies. Our findings indicated that there are several possible areas of intervention at the level of the family to improve their experiences with the diagnosis, treatment, and follow-up of childhood brain tumors. As treatment advances continue, addressing optimal quality of life for children and minimizing the negative impacts on families are increasingly important.

Authors' Note

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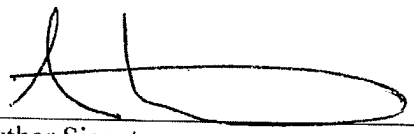

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