

Understanding the Needs of Families of Children Who Are Deaf/Hard of Hearing with an Autism Spectrum Disorder

DOI: 10.3766/jaaa.16139

Susan Wiley*
Jareen Meizen-Derr†
Lisa Hunter‡
Rebekah Hudock§
Dora Murphy¶
Katie Bentley||
Tanishia Williams***

Abstract

Background: There is a significant lack of evidence guiding our understanding of the needs of families of children who are deaf/hard of hearing (Deaf/HH) with an autism spectrum disorder (ASD). Much of our current knowledge is founded in case report studies with very small numbers of children with the dual diagnosis.

Purpose: The purpose of this study was to gain an understanding of the factors relating to caregiver stress and needs (i.e., supports and interventions) in families of children who are Deaf/HH with ASD.

Research Design: Comparison groups of families of children who were Deaf/HH, families with a hearing child with ASD, and families of children who were Deaf/HH with ASD were administered standardized questionnaires of stress with brief qualitative questionnaires focusing on family-identified needs.

Study Sample: Six families of children with the dual diagnosis, four families of children who were Deaf/HH, and three families of children with ASD.

Data Collection and Analysis: Surveys included demographic and support questionnaires, the Parenting Stress Index (PSI), the Pediatric Hearing Impairment Caregiver Experience, and a qualitative questionnaire.

Results: Families of children who were Deaf/HH with ASD had a higher median total stress score on the PSI as compared to families of children who were Deaf/HH only (58.5 versus 41.5, respectively; $p = 0.02$) and higher Child Domain scores (60 versus 43, respectively; $p = 0.02$), indicating higher levels of stress in families of children with the dual diagnosis. The families of children who were Deaf/HH with ASD reported similar levels of stress as families of children with ASD.

Conclusions: Families of children who are Deaf/HH with an ASD experience stress and describe similar needs and priorities as families of hearing children with ASD. This suggests the needs related to having an autism spectrum disorder are of high priority in families of children with the dual diagnosis.

Key Words: autism spectrum disorder, dual diagnosis, pediatric hearing loss

*Division of Developmental and Behavioral Pediatrics, Cincinnati Children's Hospital Medical Center, University of Cincinnati, Cincinnati, OH; †Division of Biostatistics and Epidemiology, Cincinnati Children's Hospital Medical Center, University of Cincinnati, Cincinnati, OH; ‡Division of Audiology, Cincinnati Children's Hospital Medical Center, University of Cincinnati, Cincinnati, OH; §Department of Pediatrics, University of Minnesota, Minneapolis, MN; ¶Ear, Nose and Throat Specialty, The Christ Hospital Physicians, Cincinnati, OH; ||Human Development Institute, University of Kentucky, Kentucky Family Advocacy Network, Lexington, KY; ***Division of Developmental-Behavioral Pediatrics, Children's Specialized Hospital Medical Department, Hamilton, NJ

Corresponding author: Susan Wiley, Division of Developmental and Behavioral Pediatrics, Cincinnati Children's Hospital Medical Center, University of Cincinnati, Cincinnati, OH 45229; E-mail: susan.wiley@cchmc.org

Presented at 2014 Early Hearing Detection and Intervention Annual Conference in Jacksonville, FL, April 15th, 2014.

Abbreviations: ADOS = Autism Diagnostic Observation Schedule; ASD = autism spectrum disorder; Deaf/HH = Deaf/Hard of Hearing; GARS = Gilliam Autism Rating Scale; PHICE = Pediatric Hearing Impairment Caregiver Experience; PSI = Parenting Stress Index

BACKGROUND

Relatively little is known about children who are Deaf/Hard of Hearing (Deaf/HH) with an autism spectrum disorder (ASD). What is understood is often based on case reports or small series of children with the dual diagnosis. This paucity of knowledge is in striking contrast to the availability of studies about hearing children with ASD and their families. These studies span numerous facets, including assessment, evidence-based interventions, and family support.

In the areas of evidence-based treatment for children with ASD, there has been enough momentum in the field to develop a National Standards Report in 2008 with an update in 2015 (Wong et al, 2015). Having available evidence-based treatment allows parents a level of support and confidence that they can positively impact their child's developmental needs. However, even with effective interventions, parents of children with ASD can still experience a high level of stress.

In broad conceptual models of parental stress, stress has been described as involving factors attributed to the child, factors attributed to the parent, and factors attributed to the interaction between the parent and child (Abidin et al, 1992). When considering children with disabilities or those who behaviorally or temperamentally are more challenging, this framework helps begin to consider various strategies to address the most salient contributors to stress levels.

It is evident that parents of children with ASD experience large amounts of stress that may potentially persist throughout a lifetime of caring for their child. Parents of children with ASD experience levels of stress that are higher than levels experienced by parents of other children (Donovan, 1988; Dumas et al, 1991; Hastings and Johnson, 2001; Weiss, 2002; Tomanik et al, 2004). This stress can often be related directly to the parenting role. Stress may result from child characteristics such as behavior problems (Konstantareas and Homatidis, 1989; Lecavalier et al, 2006) or indirect sources and outcomes related to this stress such as marital strain, or increased anxiety, depression, or social isolation experienced by families (Baker-Ericzen et al, 2005). Examining the experience of stress among parents of children with ASD is important to determine factors related to stress and to identify interventions to help alleviate this stress.

There are some studies that help understand what families of children with ASD seek out when looking for resources about ASD. Families seek information that is concise and comprehensive and provides hope

for the future (Muligan et al, 2010). They value practical strategies to build language and interactive play in their children (Whitaker, 2002). Parents often need to take on an advocacy role to ensure their children receive supports for a variety of needs. Families have used advocacy as an effective coping strategy as well as to allow the opportunity to positively impact their child's future (Boshoff et al, 2016).

Similarly, there is a literature base to support our understanding of children who are Deaf/HH. There have been a number of articles recognizing the positive impact of early identification and intervention for hearing loss. Hearing technology and communication skills have also been addressed in the existing literature. There is some knowledge about parental stress and coping among parents of children who are Deaf/HH.

In parents of children who are Deaf/HH, studies on parental coping have focused on family reaction to the diagnosis of hearing loss, parent-child interaction, and stress related to the specific needs around medical care, technology needs, and communication (Meadow-Orlans and Steinberg, 1993; Calderon and Greenberg, 1999; Kurtzer-White and Luterman, 2003; Meinzen-Derr et al, 2008).

Based on the presence of hearing loss in a child, it seems intuitive to think that parents may experience high levels of stress. However, this is not uniformly supported in the literature. Among a group of parents of toddlers with moderate hearing loss in the Netherlands, parental stress was comparable to parental stress in parents of hearing toddlers (Dirks et al, 2016). When children performed more poorly in the areas of social-emotional functioning and language development, their parents noted higher levels of stress. This finding is similar to a study in the United States on a larger group of mothers of children who were Deaf/HH ($n = 184$) (Pipp-Segal et al, 2002). In this study, mothers of children who were Deaf/HH had similar levels of stress as measured by the Parental Stress Index. Having a child with a disability in addition to hearing loss or having a child with more substantial language delay predicted higher levels of parental stress. Among mothers of children who were Deaf/HH, 13% had clinically significant levels of stress. This was slightly lower than other studies (Meadows-Orlans and Steinberg, 1993). Various authors have also found differing levels of stress associated with various ages of children (those parents with younger children exhibiting more levels of stress).

Levels of stress have been evaluated in relation to aspects of care across ages and duration since identification of hearing loss. In parents of children with a

longer duration of hearing loss, stresses around education and supports were more likely to be reported as compared to stress related to health care and emotional well-being among parents of children with a recent diagnosis of hearing loss. Stress related to communication has been found among parents of children who are Deaf/HH across all ages (Meinzen-Derr et al, 2008).

In our clinical experience, families of children who are Deaf/HH with an ASD have indicated not feeling connected to supports and family networks related to their child's hearing loss but also convey feelings of not quite fitting in with the networks of families of children with ASD. This has prompted a sense of isolation in some families. This isolation may also be due to the limited evidence supporting ASD-specific strategies in children who are Deaf/HH. There is case report level of support for ASD-specific information including the use of the Picture Exchange Communication System (Malandraki and Okalidou, 2007) and a parent-training curriculum (Garcia and Turk, 2007). These studies showed promise that these may be appropriate interventions for children who are Deaf/HH with an ASD. It is unclear whether these findings are generalizable, yet they also mirror clinician experience in which a focus on the core social and communication deficits are appropriate to prioritize intervention strategies for children with the dual diagnosis.

An understanding about family needs is also embedded in small studies. Beals (2004) wrote about her personal experiences in attaining services for her young child with a dual diagnosis. She described frustrations about the lack of knowledge in early intervention providers about both deafness and autism spectrum. Other small case series have also noted a number of challenges and needs in accessing appropriate and effective supports for children who are Deaf/HH with ASD (Myck-Wayne et al, 2011; Wiley et al, 2014).

In our clinical experience, it has also been recognized that the priorities of families of children who are Deaf/HH with ASD mirror those of families of children with ASD. Therefore, we felt it was important to consider how parents of children who are Deaf/HH with ASD are similar or different as compared to parents of children who are Deaf/HH and as compared to parents of hearing children with ASD. We sought to understand the stress level as well as the priorities and needs of families of children who are Deaf/HH with ASD using a mixed-methods approach that included small groupings of parents of children who were Deaf/HH and parents of children with ASD for comparison. By examining parenting stress in the lives of families with children who are Deaf/HH with ASD, we hoped to gain insight into the specific stressors and supports these families have and how professionals can support families.

PURPOSE

The purpose of this study was to gain an understanding of the factors relating to caregiver stress and needs in families of children who are Deaf/HH with ASD. By administering standardized questionnaires of stress with brief qualitative questionnaires focusing on family-identified needs and including comparison groups of families of children who were Deaf/HH and families with a hearing child with ASD, we hoped to better understand the needs and experiences of families of children who were Deaf/HH with ASD.

This study was approved by the institutional review board at Cincinnati Children's Hospital Medical Center.

RESEARCH DESIGN

This study used a mixed-methods approach with quantitative and qualitative parent report measures to understand the stress and needs of families of children who were Deaf/HH, families of hearing children with ASD, and families of children who were Deaf/HH with an ASD. Families of children from three groups were identified through clinical and research registries at a large urban pediatric tertiary care center for recruitment. This institution houses a strong interdisciplinary program for children who are Deaf/HH (Wiley et al, 2011) as well as a program focusing on children with developmental disabilities and serves as a site for the Autism Treatment Network (<https://www.autismspeaks.org/science/resources-programs/autism-treatment-network>).

STUDY SAMPLE

The three target groups included (a) families of children who are Deaf/HH, (b) families of children with ASD, and (c) families of children who are Deaf/HH with ASD.

Children who were Deaf/HH were identified through a registry on children with documented permanent hearing loss. Children with any degree (mild to profound) of permanent hearing loss were included. These children had all received a standardized pediatric audiometric assessment. The severity of hearing loss was clinically classified between mild and profound using a combination of frequency-specific decibel loss.

Children with a diagnosis of ASD (and no hearing loss) were identified through an autism research registry. Children in the research registry had previously completed a comprehensive multidisciplinary assessment including autism spectrum-specific assessment tools. Evaluations included the Gilliam Autism Rating Scale (GARS) (Gilliam, 1995), Autism Diagnostic Observation Schedule (ADOS) (Lord et al, 1999), cognitive testing, test of adaptive behavior, and language testing.

Normal hearing was confirmed through formal audiological evaluation by pediatric audiologists. Testing included behavioral testing when possible and auditory brainstem response testing if a child could not provide reliable results in the sound booth.

Children who were Deaf/HH with ASD were identified through a registry of children who are Deaf/HH who had completed comprehensive developmental evaluations. The additional diagnosis of ASD had been clinically made prior to entry into the study. All children who were Deaf/HH with the diagnosis of ASD (dual diagnosis) had completed a comprehensive evaluation using standardized autism assessments which included the GARS (Gilliam, 1995), ADOS (Lord et al, 2000), cognitive testing, evaluation of adaptive behavior, comprehensive speech/language evaluation, and an evaluation by a developmental pediatrician with expertise in evaluating children who are Deaf/HH. As autism diagnostic tools (e.g., GARS and ADOS) have not been validated for use in children who are Deaf/HH, both qualitative and quantitative information from the assessments were used in conjunction with *Diagnostic and Statistical Manual of Mental Disorders-IV* criteria (APA, 2000). Thus, a team of professionals representing expertise in ASD and expertise in hearing loss arrived at a consensus opinion for the diagnosis of ASD. The evaluations were used to clarify developmental patterns and were interpreted with the impact of hearing loss on scoring considered.

Once the target families of children who were Deaf/HH with ASD were identified for potential recruitment, children with ASD and children who were Deaf/HH were matched on age and gender. Hearing children with ASD were matched on child severity of ASD and children who were Deaf/HH were matched on the child's degree of hearing loss. Once these children were identified, their families were then recruited for participation.

DATA COLLECTION

Three groups of participants ($n = 24$ per group) were identified using these criteria, and were invited to participate by mail with return postage provided. Families received demographic and support questionnaires, the Parenting Stress Index (PSI), the Pediatric Hearing Impairment Caregiver Experience (PHICE) (for children who were Deaf/HH and Deaf/HH with ASD), and a Qualitative Questionnaire (short-answer format). Phone calls to encourage participation and answer questions were made to all families after the mailing was sent.

Child-specific information was gathered from families including child's age, family size, education level of person filling out the questionnaires, and income level categories. The child's medical record was reviewed for degree of hearing loss, age of identification of hearing

loss, age of identification of autism spectrum disorder, and specific autism spectrum diagnosis (autism, pervasive developmental disorder, not otherwise specified, or Asperger's syndrome).

Questionnaires

PSI

The PSI (Abidin, 1995) is a 120-item self-report measure that assesses parenting stress as a result of the parenting role and the parent-child relationship. Items are scored on a 5-point Likert scale, with a response of 5 indicating "strongly agree" and 1 indicating "strongly disagree." The measure contains a Child Characteristics Domain and Parent Characteristics Domain that were derived from factor analysis. High scores in the Child Domain indicate parenting stress stems from child characteristics or child behavior problems that make parenting difficult. High scores in the Parent Domain indicate stress stems from parent functioning or factors that affect the parent's ability to parent competently. The Child Domain consists of six subscales: Adaptability, Acceptability, Demandingness, Mood, Distractibility/Hyperactivity, and Reinforces Parent. The Parent Domain consists of seven subscales: Depression, Attachment, Restriction of Role, Sense of Competence, Social Isolation, Relationship with Spouse, and Parent Health. This tool was given to all three groups of families.

PHICE

PHICE is a measure that was developed and validated specifically on families of children who are Deaf/HH (Meinzen-Derr et al, 2008). The tool examines various potential stressors families may experience related to their child's diagnosis of hearing loss. The PHICE is a 68-item questionnaire that covers eight domains of stress. These domains include communication (10 questions), education (7 questions), emotional well-being (11 questions), equipment (3 questions), financial issues (4 questions), health care (14 questions), social needs (8 questions), and support systems (11 questions). Parents rate stress on an 8-point Likert scale ranging from no stress (score of 1) to extremely high stress (score of 8). The PHICE was found to have an overall reliability of 0.96. The PHICE has previously been used to assess caregiver stress in families of children who are Deaf/HH (Meinzen-Derr et al, 2008; Lim et al, 2014). This tool was administered to the groups of families including a child who was Deaf/HH and families of children who were Deaf/HH with ASD.

Qualitative Questionnaire: A short, open-ended qualitative questionnaire was developed based on prior work with a small focus group of families of children who are Deaf/HH with an ASD (Wiley et al, 2014). All families were

asked to answer the following questions: (a) How has the diagnosis impacted your family? (b) How has the diagnosis impacted communication strategies and decisions? (c) Where do you find support? (d) What resources have you found helpful? (e) What is on your wish list?

DATA ANALYSIS

Descriptive statistics (medians, ranges) were used to depict the sample. Differences in parenting stress as measured by the PSI and PHICE were examined by comparing total and domain-specific scores across the three groups (dual diagnosis, ASD, and Deaf/HH) using the Wilcoxon rank sum test (Wilcoxon, 1945). This test was chosen due to the small sample size and to allow comparison of nonnormally distributed data.

Because of the small sample size, information from qualitative results was consolidated based on question rather than through a thematic approach.

RESULTS

Twenty-four families within each category received questionnaires through the mail. We had

challenges with response rate despite multiple attempts to contact families to encourage participation. The overall response rate was 18%. The families of children with the dual diagnosis had the highest response rate with six families of children who were Deaf/HH with ASD (25% response rate) completing the study. In the families of children who were Deaf/HH, four families responded (16% response rate), and among families of children with ASD, three families responded (12.5% response rate).

Table 1 describes characteristics of the participating families and their children. Among children who were Deaf/HH with ASD, the age range was fairly wide (7–20 yr). The children had varying degrees of hearing loss and used a variety of communication strategies. Educational placements included being fully mainstreamed, partially mainstreamed, and placed within self-contained classrooms. The children who were Deaf/HH were all described as using solely spoken communication and all participated in mainstream settings. Children with ASD used a variety of communication strategies, including sign language, and participated in either mainstreamed or partial mainstreamed classroom settings.

Table 1. Characteristics of Participating Families and Their Children

	Dual Diagnosis (N = 6)	Deaf/HH Only (N = 4)	ASD Only (N = 3)
Family characteristics			
Number of children in family (median/range)	2 (1–4)	3 (2–4)	1 (1–2)
Level of parent education			
High school	0	1	0
Some college	1	1	0
Completed college	3	0	2
Graduate school	2	2	1
Household income			
<20,000	1	0	0
20–29,000	1	1	0
30–39,000	0	0	2
50–59,000	1	0	0
70–79,000	2	2	1
>80,000	1	1	0
Child characteristics			
Age at time of study in years (range)	7–20	11–17	9–12
Degree of hearing loss			
Unilateral/mild	2	1	N/A
Moderate-severe	0	1	N/A
Profound	4	2	N/A
Communication mode (family reported)			
Oral	2	4	2
Oral/behavior	1	0	0
Sign	0	0	1
Sign/behavior	2	0	0
Educational setting			
Fully mainstreamed	1	4	1
Partially mainstreamed	4	0	2
Self-contained classroom	1	0	0

Note: N/A = not applicable.

PSI (Administered to All Three Groups of Families)

Families of children who were Deaf/HH with ASD had a higher median total stress score as compared to families of children who were Deaf/HH only (58.5 versus 41.5, respectively; $p = 0.02$), indicating higher levels of overall stress in families of children with the dual diagnosis. Families of children who were Deaf/HH with ASD reported higher Child Domain scores (Figure 1A) as compared to families of children who were Deaf/HH only (60 versus 43, $p = 0.02$). The families of children who were Deaf/HH with ASD reported similar levels of stress as families of children with ASD on the PSI. Within the entire group of participating families, five families (38%) had a total PSI score above the 85th percentile, indicating high levels of stress (Figure 1B). Among the families reporting higher levels of stress, they were either parents of children with ASD or parents of children who were Deaf/HH with ASD. No family of a child who was Deaf/HH reached the clinical criteria for high levels of stress on the total PSI, nor were there clinically significant elevations in any subdomain of the PSI. One family of a child who was Deaf/HH with ASD reported clinically significant stress across all domains of the PSI (Child Domain, Parent Domain, and Total Stress Index). One family of a child with ASD reported clinically significant stress within the Child Domain.

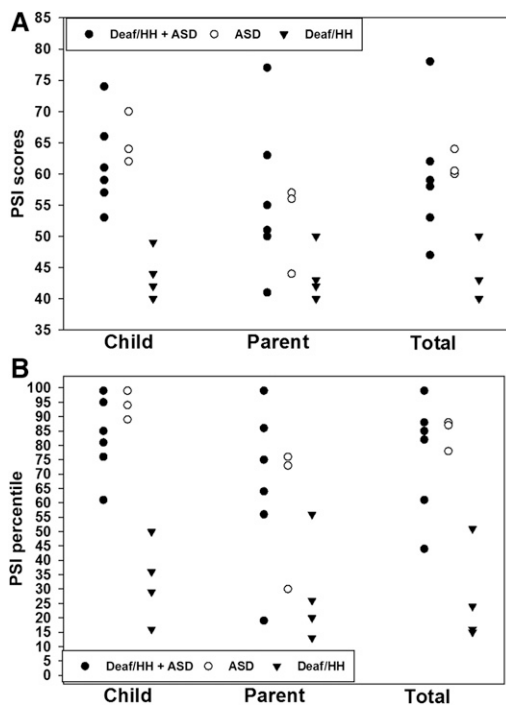


Figure 1. (A) Distribution of PSI scores for the Child Domain and Parent Domain, and total scores for Deaf/HH + ASD, ASD only, and Deaf/HH only. (B) Distribution of PSI percentiles for the Child Domain and Parent Domain, and total scores for Deaf/HH + ASD, ASD only, and Deaf/HH only.

PHICE (Administered to the Families of Children Who Were Deaf/HH and Deaf/HH with ASD)

Families of children with the dual diagnosis had a higher median total PHICE score compared to families of children who were Deaf/HH (total score 179.6 versus 150.5, $p = 0.08$), indicating higher levels of caregiver concern specifically focusing on hearing loss. Figure 2 depicts the distribution of score across the groups. Two families, one from each group (Deaf/HH and Deaf/HH with ASD), reported high levels of stress in the communication domain. Seven families (five of six Deaf/HH with ASD and two of four Deaf/HH) reported high levels of stress among questions in the social domain. Table 2 illustrates items on the PHICE on which parents reported high levels of stress. Half of the families within the dual diagnosis group identified high levels of stress about their child's future and 33% worried about not doing enough for their child, not attending to other family needs, and high levels of stress related to childcare.

Qualitative Questionnaires by Group

Answers by families from all three groups on the qualitative questionnaire exhibited overlapping characteristics in responses (Table 3). There were also family responses that were unique to the experience of having ASD (dual diagnosis or ASD only). All families described that they were *accessing supports* from a network of families, friends, therapists, and schools as well as described limitations within these support networks. "I want a better understanding within the school system, teachers with more education about hearing loss." "People at school don't think he can do things. It's frustrating because they just assume so many things and don't expect achievement, but we do at home."

All families desired *independence and happiness* for their children. Specific comments included: "I hope my son can grow up to be self-sufficient." "I wish that she

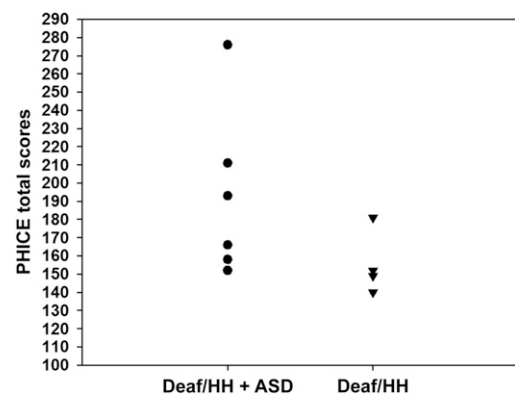


Figure 2. Distribution of PHICE total scores for Deaf/HH + ASD and Deaf/HH only.

Table 2. Items on the PHICE on Which Parents Reported High Levels of Stress

Domains	Items Reported as High Stress	ASD + HL	HL Only
Communication	Temper tantrums related to hearing loss		1
	Distinguishing between behavior and communication	1	
Social	Feeling uncertain about my child's future	3	
	Worries about others taking advantage of my child	1	
	Worries about my child's safety	2	1
	Worries about my child's ability to make friends	3	1
	Worries about how my child fits into the hearing community	2	1
	Seeing my child frustrated because of his/her hearing loss	1	1
Emotional and supports	Worries about not doing enough for child	2	
	Not being able to attend to the needs of other family members	2	
	Tired due to caring for my child with hearing loss	1	
	Having no time for myself	1	1
	Inadequate support or understanding from friends and relatives	1	
	Lack of childcare or babysitters for child	2	
Health care and finances	Worries about my child losing or damaging his/her equipment	2	1
	Inadequate insurance coverage for treatment of hearing loss	1	1
	Traveling to medical appointments	1	
	Worrying about success of my child's therapy or rehabilitation	1	
	Problems dealing with insurance company	1	1
	Costly medical equipment and expenses	1	1
Education	Worries about decisions I have made for my child's education	1	
	Lack of educational opportunities for my child	1	
	Child's reading ability	2	

could live in the world and get a job and live healthy and happy." "I want my child to have an increased ability to socialize and make friends."

Families in all three groups made specific comments related to the *identification of their child's needs*. "The diagnosis was heart-breaking." "We don't take things for granted." "It has had an emotional and physical toll."

Acceptance and expanded social interactions were also notable among all groups with comments such as a desire for "increased ability to socialize," to "increase his circle of friends," "to be able to function in a regular class," and "to become more social, develop social skills." Families of children with ASD only and families with children with a dual diagnosis described a *desire for broader acceptance* and understanding of their child's needs by others. "When my child goes out into the community, I want others to accept him as he is and be compassionate towards his needs and his frustrations. I will not always be able to run interference." "I would like for people not to judge."

Families of children with the dual diagnosis tended to describe more challenges with *feelings of isolation* and the *need for personal time*. "I don't know any other parents with a dual diagnosis like this." "I want a good break for myself from time to time."

Communication challenges were cited by families in all groups. This was quite prominent in the dual diagnosis group with the *development of effective communication skills* being notably complex for children. Families made specific comments such as "We are always looking

for a communication strategy," "The dual diagnosis complicates sign language," and "My child doesn't remember much of what has been taught." "Autism complicates sign language so we give him all the tools and follow his lead" "Dual diagnosis makes it more frustrating. You ask, "Did he hear this? Ignore it? Etc."

Bridging Qualitative and Quantitative Responses

In attempt to bridge the information gained through qualitative and quantitative responses, we took an individualized approach, pairing PSI Domain subscore elevations from individual parents and connecting their responses to the qualitative answer on the question, "How has the diagnosis impacted your family?" (Table 4). The types of information provided in the qualitative survey were not necessarily in alignment with reporting on quantitative methods. Additionally, those items endorsed on the PHICE with high levels of stress were not necessarily indicated in qualitative responses.

DISCUSSION

While recognizing a low participation rate, the families in our study reported similar challenges and stress as described in the literature. Among families of children who are Deaf/HH, there have been reports that families feel a pressure to have their child reach achievements on a "timetable" (Young and Tattersall,

Table 3. Abbreviated Answers from Qualitative Questionnaire by Group

Questions	Dual Diagnosis (n = 6)	Deaf/HH Only (n = 4)	ASD Only (n = 3)
Impact of diagnosis on family	It has had a physical and emotional toll	The diagnosis was heartbreaking	We do not take things for granted
	Challenges in getting school to recognize both issues	Major reorganization of family was needed, needed to understand navigating therapy, billing, sign language	We have an increased reliance on routines
	Parental feeling of social isolation from friends	Diagnosis was difficult for some members of our family	There is an impact due to therapy appointments
	Frustration with lack of progress	We needed a safety plan, including where our child sat in the car	Change in relationship with spouse
Impact on communication decisions, strategies	The family moved to be closer to effective services		
	Signing (in a child with a CI)	We sent our child to an oral school	Not asked in this group
	We are always looking for a communication strategy (i.e., iPad)	The hearing loss turns a nondecision situation into a situation	
	My child doesn't remember much of what has been taught	Our child has difficulties in loud settings (i.e., cafeteria) and making friends	
Support	The dual diagnosis complicates sign language, we try all tools and follow his lead		
	Family (n = 4)	Family (n = 2)	Family (n = 2)
	Friends (n = 3)	School (n = 2)	Therapists (n = 2)
	School	Friends (n = 1)	Friends (n = 1)
Helpful resources	Challenger League	Internet (n = 1)	Therapists (n = 1)
	School (n = 2)	Books (n = 1)	
	Trial and error	Early intervention (n = 2)	Speech/OT
	I don't know anyone else with a child with the dual diagnosis	FM system	Use of picture schedules
Wish list	Developmental Disabilities Services	Supplemental State Insurance	Accessing biomedical/DAN supports
	Acceptance and tolerance of others	I have gotten my wish, for him to talk	For people to not judge and be more understanding
	Independence, healthy, happy, have a job	Transition successfully to high school and increase his circle of friends	For my child to become more social, develop social skills, and independence
	To be able to communicate and function in a regular class	Increased ability to socialize, make friends	For my child to be independent and happy
Wish list	Finding an educational setting which can meet all of his needs	Hear sounds for spelling words	
	Finding a cause of the issue and fixing it	Waterproof hearing aid	
		Better understanding of school systems and professionals regarding needs related to hearing loss	

Notes: CI = cochlear implant; DAN = defeat autism now; FM system = frequency modulation system; OT = occupational therapy.

2007). Families have also reported dissatisfaction if they did not have a clear understanding on how to best intervene for their child's needs (McCracken et al, 2008). There is also evidence to suggest that families of children who are Deaf/HH have stress that varies over their child's lifespan and is linked with different

needs, such as stress related to accessing health care at young ages and stress related to educational needs at older ages (Meinzen-Derr et al, 2008). Stress related to communication is more stable through various ages of childhood for parents of children who are Deaf/HH (Meinzen-Derr et al, 2008).

Table 4. Bridging Qualitative and Quantitative Data

	PSI Subdomain Elevations	Qualitative Elements
ASD only	AD	"Relationship with wife is not the same."
	AD, DI, AC	"Need for regular routines and therapy appointments."
	AC, IS	"We don't take things for granted, milestones are important."
D/HH + ASD	MO	"It is hard to understand him and help him understand things. The frustration of his progress in learning."
	DI, AD, AC	"Not having any time to myself or any good friends gets to me. Sometimes others lack the understanding that I need to get away."
	CO, IS, AT, RO	"Pushed us to move to a new place for services, leaving behind a network of friends and family. Pushed us to advocate, to earn more money, to learn a new language. Stressful but enormous growth involved."
	DI	

Notes: Child Domains: AC = acceptability, or the extent to which child characteristics meet expectations in the child; AD = adaptability, or the child's ability to adjust to social or physical environment; DI = distractibility/hyperactivity, or symptoms that reflect features of attention-deficit hyperactivity disorder; MO = mood, or the child's affective state. Parent Domains: AT = attachment, the parent's sense of closeness with the child and his or her ability to observe and effectively respond to the child's needs; CO = competence, extent to which the parent feels comfortable and actually is capable in the parenting role; IS = isolation, or parent's degree of social support; RO = role restriction, the parent's sense of limited freedom and constrained personal identify as a result of the parenting role.

In our study, the parents of children who were Deaf/HH did not exhibit significant levels of stress on quantitative measures. Based on qualitative questions and item level responses on the PHICE, families noted ongoing concerns related to aspects of communication, especially social integration with peers. The children in this group were somewhat older, used spoken communication, and were served within mainstream settings. These child characteristics may have impacted their family's responses and priorities. It would also suggest that the families of children who were Deaf/HH had children who were highly integrated into the hearing society and thus may be less representative of the broader experiences of families of children who are Deaf/HH.

Among families of children with ASD, there are reports of persistent and substantial stress based on caregiving needs of their child (Baker-Ericzen et al, 2005), and families of children with ASD experience higher levels of stress than the general population of parents (Konstantareas and Homatidis, 1989; Dumas et al, 1991). In our small sample of families, the families of children with ASD or the dual diagnosis described high levels of stress as measured by the total score of the PSI (38% with high levels of stress).

While less is known about families of children with the dual diagnosis, Beals (2004) described her personal feelings of being pulled between professionals related to communication modality (spoken versus sign language). She also described a desire for better focus and concrete guidance from professionals knowledgeable in ASD as it related to her child's needs. Her experience would suggest a need for improvements in both areas, those professionals with expertise in the education and intervention of Deaf/HH as well as professionals with expertise working with children with ASD. Our families also described similar challenges in their qualitative responses.

Myck-Wayne et al (2011) completed targeted interviews of families of children with the dual diagnosis. The families in the Myck-Wayne study described feeling shuffled between providers from the two professional streams (Deaf/HH and ASD). They also reported the need for more intensive behavioral supports as well as a lack of sign language environment within ASD programming supports, suggesting communication needs were not fully addressed within these environments. The families participating in our study indicated a sense of isolation. They also noted challenges in getting the systems (educational, clinical, and medical) serving their children to recognize the impact of both areas of need (hearing loss and ASD). Families of children with the dual diagnosis seemed to seek support targeted within the field of developmental disabilities.

To recap, conceptual models of parental stress may involve factors attributed to the child, factors attributed to the parent, and factors attributed to the interaction between the parent and child (Abidin et al, 1992). In our study, child factors varied across subgroups related to the presence or absence of an ASD as well as whether a child was Deaf/HH. Parents of children with the dual diagnosis had a higher rate of elevations in the Child Domain subtest of the PSI, suggesting stress involving factors attributed to the child. This suggests that the presence of an ASD in a child who is Deaf/HH adds a level of parenting stress based on the child's additional complexity.

Although this study provides information that seems consistent with clinical experience, we recognize there can be a selection bias in families' willingness to take the time to respond to this survey. The participation rate was low, limiting the power to generate strong conclusions from the responses. Despite these limitations, we received a very diverse array of participants in age and other demographic characteristics, providing an

overview of issues that families face. The content within the qualitative responses as well as item-level responses on quantitative tools were powerful in better understanding a specific family's needs. By conveying the granular data on specific items families rated and comments from the qualitative questionnaire within tables, the reader is provided the opportunity to process this data as it was relayed by families.

When providing care within clinical, therapeutic, and educational settings, it is important to understand specific stressors families face. It is helpful to use a combination of questions and standardized measures when assessing family needs and stress. Interpretation of elevated scores on standardized measures must be linked back to the specific area of stress reported by families. This will ensure identification of resources and strategies to address unique needs. It is also helpful to recognize resilience factors in families as well as realize that stress can be formative toward action. As one parent in our study indicated, "(their journey was) stressful but (there was) enormous growth involved." Many families indicated a strong support network from their extended family, clinicians, and educators. Although understanding stress and needs is helpful in supporting families, when examining topics such as parental stress, providers should also consider approaches to identify strengths and resilience factors.

The information shared by families should serve as an encouragement to professionals to strive for improved integration of education, interventions, and supports across the fields of Deaf Education and ASD. It is important for professionals in the field of audiology to support families of children who are Deaf/HH with ASD in understanding their children's communication and behavioral needs and guide them to resources to meet their child's needs beyond those from hearing loss alone.

REFERENCES

- Abidin RR. (1995) *Parenting Stress Index: Professional Manual*. Odessa, Ukraine: Psychological Assessment Resources, Inc.
- Abidin RR, Jenkins CL, McGaughey MC. (1992) The relationship of early family variables to children's subsequent behavioral adjustment. *J Clin Child Psychol* 21:60–69.
- American Psychiatric Association (APA). (2000) *Diagnostic and Statistical Manual of Mental Disorders: DSM-IV-TR*. 4th ed. Washington, DC: APA.
- Baker-Ericzen MJ, Brookman-Frazee L, Stahmer A. (2005) Stress levels and adaptability in parents of toddlers with and without autism spectrum disorders. *Res Pract Persons Severe Disabl* 30(4):194–204.
- Beals K. (2004) Early intervention in deafness and autism: one family's experiences, reflections, recommendations. *Infants Young Child* 17:284–290.
- Boshoff K, Gibbs D, Phillips RL, Wiles L, Porter L. (2016) Parents voices: why and how we advocate. A meta-synthesis of parent's ex-

- periences of advocating for their child with autism spectrum disorder. *Child Care Health Dev* 42(6):784–797.
- Calderon R, Greenberg MT. (1999) Stress and coping in hearing mothers of children with hearing loss: factors affecting mother and child adjustment. *Am Ann Deaf* 144(1):7–18.
- Dirks E, Uilenburg N, Rieff C. (2016) Parental stress among toddlers with moderate hearing loss. *Res Dev Disabil* 55:27–36.
- Donovan AM. (1988) Family stress and ways of coping with adolescents who have handicaps: maternal perceptions. *Am J Ment Retard* 92(6):502–509.
- Dumas JE, Wolf LC, Fisman SN, Culligan A. (1991) Parenting stress, child behavior problems, and dysphoria in parents of children with autism, Down syndrome, behavior disorders, and normal development. *Exceptionality* 2:97–110.
- Garcia R, Turk J. (2007) The applicability of Webster-Stratton Parenting Programmes to deaf children with emotional and behavioural problems, and autism, and their families: annotation and case report of a child with autistic spectrum disorder. *Clin Child Psychol Psychiatry* 12(1):125–136.
- Gilliam JE. (1995) *Gilliam Autism Rating Scale*. Austin, TX: Pro-Ed.
- Hastings RP, Johnson E. (2001) Stress in UK families conducting intensive home-based behavioral intervention for their young child with autism. *J Autism Dev Disord* 31(3):327–336.
- Konstantareas MM, Homatidis S. (1989) Assessing child symptom severity and stress in parents of autistic children. *J Child Psychol Psychiatry* 30(3):459–470.
- Kurtzer-White E, Luterman D. (2003) Families and children with hearing loss: grief and coping. *Ment Retard Dev Disabil Res Rev* 9(4):232–235.
- Lecavalier L, Leone S, Wiltz J. (2006) The impact of behaviour problems on caregiver stress in young people with autism spectrum disorders. *J Intellect Disabil Res* 50(Pt 3):172–183.
- Lim LH, Xiang L, Wong NL, Yuen KC, Li R. (2014) Validation of the Paediatric Hearing Impairment Caregiver Experience (PHICE) Questionnaire. *Ann Acad Med Singapore* 43(7):362–370.
- Lord C, Risi S, Lambrecht L, Cook EH Jr, Leventhal BL, DiLavore PC, Pickles A, Rutter M. (2000) The autism diagnostic observation schedule-generic: a standard measure of social and communication deficits associated with the spectrum of autism. *J Autism Dev Disord* 30(3):205–223.
- Lord C, Rutter M, DiLavore PC, Risi S. (1999) *Autism Diagnostic Observation Schedule-WPS (ADOS-WPS)*. Los Angeles, CA: Western Psychological Services.
- Malandraki G, Okalidou A. (2007) The application of PECS in a deaf child with autism: a case study. *Focus Autism Other Dev Disabil* 22:23.
- McCracken W, Young A, Tattersall H. (2008) Universal newborn hearing screening: parental reflections on very early audiological management. *Ear Hear* 29(1):54–64.
- Meadow-Orlans KP, Steinberg AG. (1993) Effects of infant hearing loss and maternal support on mother-infant interactions at 18 months. *J Appl Dev Psychol* 14:407–426.
- Meinzen-Derr J, Lim LHY, Choo DI, Buyniski S, Wiley S. (2008) Pediatric hearing impairment caregiver experience: impact of duration of hearing loss on parental stress. *Int J Pediatr Otorhinolaryngol* 72(11):1693–1703.

- Mulligan J, Steel L, Macculloch R, Nicholas D. (2010) Evaluation of an information resource for parents of children with autism spectrum disorder. *Autism* 14(2):113–126.
- Myck-Wayne J, Robinson S, Henson E. (2011) Serving and supporting young children with a dual diagnosis of hearing loss and autism: the stories of four families. *Am Ann Deaf* 156(4):379–390.
- Pipp-Siegel S, Sedey AL, Yoshinaga-Itano C. (2002) Predictors of parental stress in mothers of young children with hearing loss. *J Deaf Stud Deaf Educ* 7(1):1–17.
- Tomanik S, Harris GE, Hawkins J. (2004) The relationship between behaviours exhibited by children with autism and maternal stress. *J Intellect Dev Disabil* 29(1):16–26.
- Weiss MJ. (2002) Hardiness and social support as predictors of stress in mothers of typical children, children with autism, and children with mental retardation. *Autism* 6(1):115–130.
- Whitaker P. (2002) Supporting preschool children with autism: what parents want and what helps. *Autism* 6(4):411–426.
- Wilcoxon F. (1945) Individual comparisons by ranking methods. *Biom Bull* 1(6):80–83.
- Wiley S, Arjmand E, Jareenmeizen-Derr, Dixon M. (2011) Findings from multidisciplinary evaluation of children with permanent hearing loss. *Int J Pediatr Otorhinolaryngol* 75(8):1040–1044.
- Wiley S, Gustafson S, Rozniak J. (2014) Needs of parents of children who are deaf/hard of hearing with autism spectrum disorder. *J Deaf Stud Deaf Educ* 19(1):40–49.
- Wong C, Odom SL, Hume KA, Cox AW, Fettig A, Kucharczyk S, Brock ME, Plavnick JB, Fleury VP, Schultz TR. (2015) Evidence-based practices for children, youth, and young adults with autism spectrum disorder: a comprehensive review. *J Autism Dev Disord* 45(7):1951–1966.
- Young A, Tattersall H. (2007) Universal newborn hearing screening and early identification of deafness: parents' responses to knowing early and their expectations of child communication development. *J Deaf Stud Deaf Educ* 12(2):209–220.