

Hearing Care and Management Priority Among Parents of Children with Down Syndrome: A
Grounded Theory

John J. Whicker^{1*}, Karen Muñoz^{2,3}, Nicole J. Pearson², Trenton J. Landon⁴, Lauri H. Nelson²,
Karl R. White^{3,5}, and Michael P. Twohig⁵

¹Department of Rehabilitation, Primary Children's Hospital, Salt Lake City, UT

²Department of Communicative Disorders and Deaf Education, Utah State University, Logan, UT

³National Center for Hearing Assessment and Management, Utah State University, Logan, UT

⁴Department of Special Education and Rehabilitation, Utah State University, Logan, UT

⁵Department of Psychology, Utah State University, Logan, UT

Corresponding author: John J. Whicker; jj.whicker@imail.org; 3845 West 4700 South, Taylorsville, UT 84129; 801-840-4390.

Abstract

Objective: This study qualitatively explored the factors that influence how parents of children who are Deaf or hard-of-hearing with Down syndrome prioritize hearing care and management and developed an associated theory to explain that priority.

Design: Grounded theory was used for the purposes of this qualitative study. Data were collected using in-depth interviews which were analyzed using a three-tiered qualitative coding process.

Study Sample: Eighteen mothers of children who are Deaf or hard-of-hearing with Down syndrome participated in this study.

Results: The higher the extent of engaged professional support, perception of benefit for child, parent activation, and family engagement, the higher the priority for hearing care and management will likely be among parents of children who are Deaf or hard-of-hearing with Down syndrome.

Conclusions: Understanding how parents of children who are Deaf or hard-of-hearing with Down syndrome decide to prioritize hearing care and management has implications for how hearing health providers and others provide care to parents to enhance priority for hearing-related needs.

Hearing Care and Management Priority Among Parents of Children with Down Syndrome: A Grounded Theory

An estimated 75% of children with Down syndrome (DS) have hearing loss in addition to a variety of other health concerns, including congenital heart defects, vision impairments, respiratory concerns, and feeding difficulties (Center for Disease Control and Prevention [CDC], 2019). While it is assumed and expected that having DS in addition to deafness/hearing loss will compound challenges, a scant amount of research has been conducted to understand the impact of co-occurring disabilities on the prioritization and management of hearing health by parents/guardians. In a review of literature, Whicker, Muñoz, and Nelson (2019) synthesized a variety of reported parent challenges and experiences caring for children who are deaf or hard-of-hearing (DHH) with other disabilities as impacting personal and family factors (e.g., decision-making, planning, family support, burnout), medical and educational factors (e.g., professional attitudes, interprofessional collaboration, education placement and services), and child communication (e.g., communication barriers, challenging behaviors). While the findings from this review were insightful, understanding how priority for hearing health changes when children have multiple disabilities remains unclear. In addition, few of the participants sampled in the studies reviewed had DS.

Understanding how parents prioritize hearing health when children are DHH with DS can have implications for how audiologists and other professionals communicate and partner with parents to enhance outcomes for language and auditory development. Early parent-child interactions are critical for language development (Blaiser, 2012). For children who are DHH, interactions may be intermittent, given parents' lower sense of involvement or self-efficacy in managing the child's hearing-related needs (DesJardins & Eisenberg, 2007). In addition, parents

may experience strong emotions, such as depression, that limit how they enforce hearing aid wear-time (Muñoz, Olson, Twohig, Preston, Blaiser, & White, 2015). For children who are DHH with DS, these challenges may be compounded, and parents and professionals alike may easily neglect any hearing-related concerns until a later time, ultimately jeopardizing children's communicative development. However, communicative development should remain a priority despite competing complications (Whicker, Muñoz, & Pearson, 2020), as it can have long-term implications for social (e.g., Aro, Eklund, Nurmi, & Poikkeus, 2012; Botting & Conti-Ramsden, 2008; Malesa, Foss-Feig, Yoder, Warren, Walden, & Stone, 2013), and academic (e.g., Johnson, Beitchman, & Brownlie, 2010) outcomes.

Early identification of deafness/hearing loss with timely intervention to address the developmental needs of the child optimizes children's access to language (Joint Committee on Infant Hearing, 2019). This emphasizes the need to maintain hearing care and management as a priority, despite the complexities that accompany children who are DHH with DS. To address the question regarding priority of hearing health among parents of children who are DHH with DS, the present study sought to develop a theory explaining parental prioritization of hearing care and management. To accomplish this, the current qualitative study sought to answer the following research question: What factors influence parents to place high, moderate, or low priority on hearing care and management for children who are DHH with DS?

Methods

Design

This study used a qualitative design with Grounded Theory as the analytical strategy and tool to theorize how events, actions, or interactions might explain an outcome (Corbin & Strauss, 1990; Creswell & Poth, 2018). For the present study, the outcome in question is the extent to

which parents prioritize hearing care and management for their children who are DHH with DS. This study received ethical approval from the Institutional Review Board at the author's academic institution.

Instrument

A simple demographic form was used to gather information regarding parents' role, age, education level, and income. Demographic information was also obtained to understand the child's age and gender. In addition, this study used in-depth interviews. In-depth interviews allow the researcher to adjust questioning based on individual responses, which is a critical piece of grounded theory (Corbin & Strauss, 1990). For a full list of the stem questions used to guide the interview flow, please see the Appendix. Questions were designed to gauge how parents ascribe importance to what the researchers consider elements of hearing care and management, including general hearing health, hearing aids, communicative development, speech-language therapy, and visual language. Given the in-depth nature of the interviews, additional questions to probe deeper into participant responses were often unique to the particular interview. However, general questions (e.g., "Tell me more about . . .") were most often used to illicit more detail in regard to experiences or phenomena reported.

Participants

Eligible participants for this study were parents of at least one child who is DHH with DS. Sample size was based on data saturation (Creswell & Poth, 2018). Saturation was determined when no new events, actions, or interactions were being reported that warranted a new label or category during the concurrent data analysis (see below). For this study, saturation was confirmed by completing three additional interviews once saturation was indicated.

Procedures

Information regarding the study and eligibility criteria were posted onto Facebook group pages dedicated to parents of children with DS. Shortly after beginning recruitment, one state and one regional organization that provide resources for parents of children with DS emailed the first author and offered to send the study information as part of their monthly newsletter. Interested parents were informed to email the first author, who then sent a link to a secure online survey that included an informed consent statement and the demographic questionnaire. Once enrolled, parents scheduled a one-time phone interview with the first author. Phone call interviews were anticipated to last approximately 40-60 minutes. Interviews were schedule two days apart from each other to accommodate for analysis time (e.g., interview with parent two occurred at a minimum of two days after the interview with parent one). Audio of the interviews were recorded for subsequent transcription and analysis.

Analysis

Each interview was transcribed by the first author verbatim. Transcripts were not reviewed by other team members prior to analyses. Following the transcription, the first and third authors independently completed a three-tiered coding process, consistent with a systematic grounded theory (Corbin & Strauss, 1990, 2008). With grounded theory, analyses happen concurrently with data collection. To enhance analytical depth with this process, memoing was used to track researchers' ongoing inferences being made regarding various observations, which were used to fuel further iterative inquiry with subsequent interviews. At the end of each interview, the first author completed a member check by summarizing the interview conversation and the initial inferences made based on that conversation. This allowed the participants the opportunity to identify any disagreements or provide clarification on the summary and ensure accurate representation of the parents' message and experience.

After each interview, the first (JJW) and third (NJP) authors met to compare findings and inferences. Lexical differences for labelling and categorizing themes existed between the researchers; however, differences were assessed for overall meaning and researchers developed mutual wording for how themes and categories were labelled. For example, JJW may have labelled a theme as “Professional decisions/competencies” while NJP labelled the same theme as “Professional interactions and recommendations”. After discourse, it was clear both labels were attempting to capture the extent to which professionals were engaged in a supportive way. Thus, the resulting wording for the theme was “Engaged Professional Support”. No substantial differences in how the content was interpreted by the researchers existed.

Given the scope of the interviews regarding parents’ perspective of importance associated with different elements of hearing care, parents’ experiences seemed to naturally integrate into sub-themes of low, moderate, or high priority (open coding). Greater themes were generated based on the context of the event, action, or interaction surrounding the experience (axial coding). Finally, the resulting Grounded Theory was generated from the core themes that unified all other themes and emerged consistently across experiences (selective coding). For the purpose of this study, a theme was considered consistent if it emerged in at least three of the five elements of hearing care and management.

Results

A total of 18 mothers of children who are DHH with DS participated in this study. Data saturation was indicated after interview 15 and confirmed following the three subsequent interviews. Table 1 shows the spread of demographic variables obtained regarding both the participating mothers and their represented children. Interviews lasted, on average, 36.4 minutes (SD=7.5 minutes; Range=21-51 minutes).

A variable number of themes were coded in response to questions regarding elements of hearing care. The selective coding revealed four themes that consistently emerged across experiences and unified the others to explain how mothers determined to place a high, moderate, or low priority for each element. Themes included mothers' perceptions of (a) the extent of professional engagement regarding their child's hearing related needs, (b) benefit for child outcomes, (c) their own activation, and (d) the extent of family engagement. Thus, Figure 1 attempts to capture the following theory:

The higher the extent of engaged professional support, perception of benefit for child, parent activation, and family engagement, the higher the priority for hearing care and management will likely be among parents of children who are DHH with DS.

While these four key elements were found to emerge consistently across experiences to explain priority, it should be noted that other lesser themes emerged that influenced decision making for some mothers, including precedence of medical concerns, parent fatigue, goal-setting, child behaviors, and child preferences for communication. However, given they were not consistent across experiences, they were not included in the final theory, yet they are no less important to consider. Thus, visual schematics of the inferred decision-making process regarding priority for each element of hearing care can be seen in Figures 2-6.

Engaged Professional Support

Low Priority. Communication style and messaging from healthcare professionals influenced some mothers to hold a low priority for hearing health, hearing aids, and speech-language therapy. For hearing health, one mother (Mom 2) described how her child failed her first newborn hearing screen attempt, yet the hospital staff told Mom 2 that she did not need to

return for follow-up testing until three months later, and then again at six months. Both appointments yielded different results, and Mom 2 reported not knowing what to believe.

For hearing aids, four mothers (Moms 2, 5, 6, 14) described how poor professional engagement resulted in their low priority. For example, Mom 14 reported she did not pursue hearing aids based on a recommendation from the child's audiologist, who felt that because of the child's age and limited spoken language, the hearing aids would not be helpful. Thus, Mom 14 expressed: *So, I mean we thought about it quite seriously, and we finally decided not to do it.*

For speech-language therapy, two mothers (Moms 10 and 17) described their feelings regarding the services they received from speech-language pathologists that were not catered toward their children's unique needs, thus resulting in a low priority for speech-language interventions. For example, Mom 10 explained:

The difficulty is, there are a lot of great SLPs out there, and they know a lot, but it's working with the Down syndrome that's the hard part, because they are just hard to work with. And so, it's hard to find an SLP who knows how to make them work and doesn't just sit down and wait for them to cooperate. So, it's finding that person, and that's really hard.

Moderate Priority. One mother (Mom 13) reported a moderate level of priority regarding speech-language therapy that was dependent on the therapist's level of expertise and experience. She explained:

Honestly, it depends on the individual who is providing. . . . it's important for me, that AAC [augmentative and alternative communication] should be their area of expertise. But I know the speech pathologist that's working this summer at school does not have knowledge of AAC and has very limited sign language knowledge.

High Priority. Engaged professional support influenced a high priority for hearing health, hearing aids, and visual language. For hearing health seven mothers (Moms 7, 9, 10, 11, 13, 17, and 18) described how engaged professionals support was crucial in their ability to follow through on diagnostic testing following a failed newborn hearing screen and subsequent follow-up appointments, and for enrollment in early intervention.

For hearing aids, three mothers (Moms 3, 7, and 9) described how engaged professional support influenced them to consistently value and use hearing aids. For example, Mom 7 described how her son's early interventionist pressed the "importance of his needing hearing aids". Similarly, Mom 9 reported:

[Hearing aids] are very important. Because what they told us was, have [my son] wear them as much as he can. Especially because it'll help with clarity of sound, and also for safety. Because he doesn't hear the soft sounds as well. And to keep the . . . for him to have as much awareness over time, the more he wears them.

For visual language, two mothers (Moms 6 and 18) described how their early interventionists played a role in helping to facilitate a high priority. Mom 18 described:

We have our [early intervention] team, and they really set me up right off the bat with Deaf culture, and she's been really great. She now comes once a week. And she's been definitely encouraging me to . . . different kinds of way to use sign language or has taught me different signs that I need to be using. And has definitely also told me just of like a variety of different programs and whatnot, of what to expect.

Shifting Priority. Finally, a change in professional engagement influenced a shift in how parents prioritized both hearing health and hearing aids. For hearing health, one mother (Mom 12) described how, initially, her daughter's failed hearing screen was not important due to

misconceptions provided by the hospital staff. Later, however, the child's pediatrician urged for a diagnostic hearing assessment that the mother took seriously. She explained:

My daughter was not diagnosed with a hearing loss until she was a year old. And that was after multiple failures at those early newborn hearing screens. The nurses and the hospitals and everyone just gave an excuse for failing. Like, oh she's congested, or oh she was fussy, or oh she was sucking on a pacifier. . . . We finally realized at age one that we couldn't make excuses for it anymore, and my pediatrician pushed to do a sedated ABR. And that was when they discovered that she was born with a mild to moderate bilateral hearing loss.

For hearing aids, two mothers (Moms 15 and 17) described how their priority for hearing aids shifted from low to high due to a change in how engaged and supportive different providers were. For example, Mom 17 explained:

. . . when we went to the audiologist, they told me he definitely had some sort of hearing loss. However, due to our insurance, they didn't really play it off like it was a really big deal, or that he would ever be a candidate for hearing aids, so unfortunately he did go unaided for a very long time, because I was actually told that he was not a candidate for hearing aids. Had I known that he could have benefitted from hearing aids, I absolutely would have moved the mountains to make sure he received what he needed.

Perception of Benefit

Low Priority. Three mothers (Moms 10, 16, and 18) indicated a low priority for hearing aids because they did not consider the hearing aids to be beneficial for their child with Down syndrome. For example, Mom 10 described her experience:

I guess I feel like she can hear well enough, and the bother of [hearing aids] was such a pain, that it's just not worth it to me. And as far as her speech goes, I don't feel like her speech is that great, and I don't think it has much to do with her hearing . . . I think it's more of a Down syndrome thing than a hearing thing.

High Priority. Conversely, how parents perceived benefit explained mothers attributed high priority for hearing health, hearing aids, communicative development, and visual language. For hearing health, one mother (Mom 8) described her recognition of the role of hearing in general development. She stated:

From my perspective, the hearing loss was a really big hit, because I knew that [my child] was already at risk for having trouble learning, because of the developmental delays that come with Down syndrome. And then knowing that he wasn't hearing on top of that, just had me really freaked out that my child wasn't going to be able to learn.

For hearing aids, three mothers (Moms 7, 12, and 13) attributed a high priority to hearing aids in part due to their perception of how the hearing aids benefit their children. Mom 13 described hearing aid use as “non-negotiable”. Similarly, Mom 12 explained:

The hearing aids help her to hear clearly and help her to be more attentive to what's going on around her. . . in groups, at parties, in social settings, things like that, it is vital that she has those hearing aids in.

For communicative development, six mothers (Moms 13, 14, 15, 16, 17, and 18) described the different benefits of communication they perceive for their child that fuel their high priority for facilitating communicative development. These benefits included minimized frustration for the child and the family, and overall improved child behaviors.

For visual language, five mothers (Moms 6, 10, 13, 17, and 18) described the benefit for using a visual language with their child that inferred a high priority. For example, Mom 10 simply stated that visual language is so important, because it is the only way her child communicates. Mom 13 described how visual language taps into both her daughter's visual and communication needs:

I mean, I can sign something, and [my daughter] will follow through with it. You know, some of the basic things: stand and sit, and you know even doing a directed point. So, to give her that visual attention, even if she doesn't hear it, she understands some of those simple things.

Parent Activation

Low Priority. This explanation for low priority was expressed regarding hearing health and speech-language therapy. For hearing health, Mom 6 described a low motivation to monitor for changes in her child's hearing acuity, given his overall demeanor of happiness and ability to express himself.

For speech-language therapy, Mom 15 described her activation in terms of how little she knew about her daughter's special education services and whether they included speech-language intervention. She described:

I'm not sure about [my daughter]. I know she gets pulled out of normal classes when she doesn't pay attention, or if she needs a break, they'll pull her out. But I don't know if they do anything else after that. I know that they just call it 'specials', but I don't know what 'specials' are.

Moderate Priority. Parent activation was also an inferred reason for when moderate priority was attributed by mothers regarding communicative development, speech-language

therapy, and visual language. For communicative development, one mother (Mom 9) described a high value but recognized she does not spend as much time facilitating her child's communicative development as she feels she could. She explained:

[Communication is] important to me because I want [my son] to be able to fend for himself and communicate, and ask for what he wants, and advocate, all those things. But, you know, you do what you can. I might leave stuff where I know he knows where to find it. So, he doesn't have to ask for it, he can just get up and get it himself. . . . So, I probably need to have more strategies. I probably do need to do more . . .

For speech-language therapy, three mothers (Moms 2, 4, and 5) described a limited activation regarding speech intervention services in that they valued such services and consented they be provided in school; however, beyond that, the mothers were not as involved. For example, Mom 2 simply stated: *You know, I actually don't speak with the speech therapist. They email me if they have any questions. [My daughter] enjoys going.*

Regarding visual language, Mom 15 indicated she highly valued sign language for her daughter's ability to communicate her needs; however, mom reported not learning sign language herself. Thus, despite the high appreciation for a manual language, the limited effort to learn it and efficiently use it indicated the priority for manual language to be moderate.

High Priority. Parent activation could also explain high priority for hearing health, communicative development, speech-language therapy, and visual language for some mothers. For hearing health, one mother (Mom 4) explained how she and her husband took intentional action to learn as much as they could about Down syndrome and its complexities, which prompted them to pay close attention to their child's hearing-related needs.

For communicative development, six mothers (Moms 1, 6, 7, 11, 12, and 16) described specific actions they routinely took to facilitate communicative development for their children.

For example, Mom 12 described:

It's really hard to say, because [facilitating communication is] just built into what we do. I mean, we've been living with this for 16 years, so I just do it and I don't even realize I'm doing it. You know, working things like making her look at us. When I really want her to pay attention, I say look at me. Look at me. And that to me is a strategy. Repeating things for her, having her repeat back to me. Having her agree – so, when I ask her to do something, I don't walk away until she says 'okay, mom'. And I think that that's a communication strategy. So, I don't know . . . it's just all day constantly.

For speech-language therapy, six mothers (Moms 1, 3, 9, 12, 14, and 18) reported a high level of involvement in their children's speech-language interventions. For example, Mom 1 is herself a speech-language pathologist and was able to use her training to consistently work on her son's speech therapy goals. For Mom 3, she considered her involvement with her daughter's speech therapy as one of her primary roles. She explained:

I mean, it's fun for me. And I see the, I see how applicable it is and how we can use it in every day. So, yeah, it's very important for me to be involved. And it's my job – I see it as being my job, not theirs.

For four mothers (Moms 6, 10, 13, and 17), learning a visual language was both important and something they incorporated into their routines. For example, Mom 17 stated:

I mean I'm always reaching out and trying to learn more. I'm not fluent in sign language by any means. . . . But I have taken college courses. I have taken online courses. I have

reached out to Deaf culture people with Deaf events. I have spent pretty much [my son's] whole life trying to learn more.

Shifting Priority. Finally, a change in parent activation was found to influence a shift in how Mom 7 prioritized her son's hearing health. She described how hearing loss was "put on hold" due to other medical issues; however, over time, mom took action to ensure that his hearing was re-evaluated.

Family Engagement

Low Priority. One mother (Mom 6) attributed a low priority for hearing health to her husband's lack of involvement. She explained:

I feel that he could have been a lot more . . . Well, he's been a workaholic because I think he shies away from [helping]. He knew that what I was doing was correct. But he didn't want to get involved, because it just took so much of his time.

Moderate Priority. Moms 1 and 5 reported that the minimal family support they receive influenced their decision to make hearing aids a moderate priority. For Mom 1, she reported that she felt she is the only one working to manage her child's difficult behaviors related to his consistent hearing aid use. She explained:

Like, my husband is really good to kind of help and support on that, but he's not usually here, so he's not super available for that. Like, I just don't have as much support at home as I do like as he would at school.

High Priority. High priority was inferred surrounding family engagement for hearing aids, communicative development, and speech-language therapy. For hearing aids, two mothers (Moms 3 and 9) reported engaged family support played a role in helping them to place and maintain a high priority for hearing aids. For example, Mom 3 explained:

When my husband gets home – he, you know, I’ll even tell him: okay, it’s your turn to get after her, because I’ve been doing it all day. And he does, he’s really good about that. I know that he is there, and as far as when she acts out and pulls the hearing aid out, when he’s around, he definitely steps up, steps in.

For communicative development, two mothers (Moms 2 and 15) ascribed their high priority to family support. For example, Mom 2 explained:

We have a Bi-Bi language home, like I said. My husband’s family is completely Deaf . . . And my family is primarily hearing. So, we have both. And, as far as her grandparents, you know they speak, and they’re very supportive if we sign, or [if] we speak. And then when we go to my husband’s parents, and they’re very supportive as well.

For speech-language therapy, one mother, (Mom 14) reported how her children and husband have played roles in supporting a high priority for speech-language therapy. She explained:

So, the [siblings] were fantastic helpers, especially the girls, doing therapy with [my son] and trying to get him to do the things that the therapists . . . You have all these things that you’re working with at the same time. And my husband’s fantastic. He would take off for doctor’s appointments. We’re a good team. He still works really well with [my son]. So, I’ve had great family support.

Discussion

The purpose of this study was to develop a grounded theory to describe the factors that influence the decision-making process parents experience when determining how they prioritize hearing care and management while caring for children who are DHH with DS. The findings

from this study present the first developed theory to explain when and how parents choose to highly prioritize hearing care and management. It is hoped such a theory may lay the groundwork for future research to further define and explain how priority for hearing health may change for parents of any child who is DHH with other disabilities.

Four reasons were uncovered to theorize how and when parents highly prioritize hearing care and management. These include engaged professional support, a high perception of benefit for the child involved, high parent activation, and engaged family support. In healthcare, how providers interact with patients has shown to influence patient outcomes (Brown, Stewart, & Ryan, 2003; Zolnierek & DiMatteo, 2009). For mothers in this study, comments regarding professional interactions were often about how information was shared and recommendations made. This has implications for how mothers can establish a relationship of trust with their providers to ensure adequate follow-up care (Gabay, 2015). For example, some mothers reported being misinformed regarding the need to follow-up on failed newborn hearing screens, and some were provided inconsistent recommendations for treatment. As revealed in the results section, an improvement in how professionals engage with families or changing to providers who are more engaged can influence parents to shift how they prioritize hearing care and management.

How mothers perceived elements of hearing care as beneficial to their children influenced how highly they prioritized that element. This phenomenon is consistent with the Health Belief Model (Rosenstock, Strecher, & Becker, 1988), a widely recognized model for health-related action that emphasizes the role of the patient's belief in the condition and the belief in treatment in order to adhere to provider recommendations. For some mothers in this study, the mild nature of their child's degree of hearing loss, or the unilaterality of their child's hearing loss, impeded how they perceived hearing loss to be a threat to their child's development. This, incidentally,

seemed to bar their perception for how helpful hearing aids can be. Conversely, most mothers easily understood the threat to their children's wellbeing when communicative development was jeopardized, and this perceived threat to their children's wellbeing seemed to enhance how mothers acted to facilitate communicative development.

Related to perception of benefit and its link to the Health Belief Model is the extent to which parents take action to maintain a priority for hearing care. Understanding the influence behind parent activation is an important element in identifying and addressing potential barriers parents may face that impede their motivation to pursue and maintain quality care in the home environment. Evidence has indicated the extent of patient engagement can enhance health outcomes and experiences, as well as make health care more cost efficient (Hibbard & Greene, 2013). Interestingly, findings from this study suggest that parents' perception of belief and their level of activation do not seem to be totally connected. For example, while perception of a belief played a role in how parents prioritized hearing aids, parent activation did not emerge as a theme for this element of hearing care. While Grounded Theory is designed to probe into why certain themes do not emerge in some cases when they emerged in others, understanding the altogether lack of parent activation as an influencing factor for hearing aid priority could have been missed and would be of interest for future studies.

Family support has been shown to enhance how patients adhere to treatment regimens or recommendations. In a meta-analysis of 122 studies, DiMatteo (2004) found that those individuals who received family support to manage their diabetes showed 27% higher adherence to treatments. In a more recent review of empirical literature looking at the effects of family support while managing diabetes, Miller and DiMatteo (2013) indicated that, while the benefits of family support are evident, the mechanism for how family support is effective is yet unclear.

For example, family support can have negative outcomes when family members are critical or nagging (Carter-Edwards, Skelly, Cagle, & Appel, 2004), or when family members are particularly demanding (Gallant, Spitze, & Prohaska, 2007).

Some findings were surprising; although with Grounded Theory the researcher's assumptions for how the theory in question may develop are inappropriate (which is why a systematic approach was used in this study to mitigate researcher bias through constant and iterative comparisons of events, actions, or interactions described by mothers, and confirm whether observations are consistent or variable across conditions through data saturation), some clinical assumptions were made that prompted the development of this study. One such assumption was that the role of medical complexities would highly influence how parents prioritized hearing care and management; however, this theme hardly emerged, and when it did it was not consistent across the elements of hearing care, even though many children did have considerable medical complexities

Understanding the factors that explain how parents decide to prioritize effective hearing care and management has important implications for clinical practice. The foremost implication relates to how audiologists and other providers can develop partnerships with parents of children who are DHH with other disabilities in order to understand the challenges and barriers associated with a minimal priority for hearing care. For example, while for some mothers other complexities existed and took precedent over hearing-related needs, it is clear from this study that most mothers valued communicative development and desired for their child to become efficient in their spoken or manual language. Thus, the value for hearing care and management was there, yet the priority varied dependent on the factors described.

By engaging in person-centered care through counseling, audiologists can illuminate these incongruencies (i.e. value versus practice) and bring them to parents' attention. This can open opportunities for clinicians and families to directly talk about challenges that impede adherence to their own values and preferences related to their child's hearing-related needs and set goals to help parents better align their care to those values. Values-based conversations are consistent with evidence-based strategies for helping individuals make behavior-based changes, such as Acceptance and Commitment Therapy (Hayes, Luoma, Bond, Masuda, & Lillis, 2006) and Motivational Interviewing (Rollnick, Miller, & Butler, 2008).

Because priority likely changes from day to day and is the result of factors not illuminated by the present study, making clinical assumptions about how parents prioritize hearing care and management based on whether professional engagement, perception of benefit, parent activation, and family support are in place could be over-simplified. Clinicians should be careful to understand the full scope of parent experiences, needs, and routines. Open-ended questioning and reflection can aid the clinician in comprehending parents' true feelings and perception for how they are prioritizing hearing care and management. Because of the possibility for changing or fluctuating priority, clinicians should evaluate how parents are prioritizing hearing care and management at every step of the journey (e.g., diagnosis and all follow-up). Evaluating how parents are prioritizing hearing care and management can be asking such open-ended questions as: "How important is your child's hearing to you right now?", "What are you doing at home to manage your child's hearing related needs?", or "What challenges are you facing that you did not anticipate that seem to hinder how much you can take care of your child's hearing-related needs?". When asked in this way, parents may feel invited to share their

challenges without fear of judgement which can open opportunities for the clinician to set goals for increasing hearing care priority with parents.

The goal of a grounded theory design is to establish a conceptual framework for future research. This study was prompted by the need to illuminate challenges that impede how parents prioritize and manage hearing care when children who are DHH have other disabilities. This study focused solely on children who are DHH with Down syndrome. Future studies may expand on the findings reported in this paper by evaluating and theorizing how parents of children who DHH with other types of disabilities determine to prioritize hearing care and management priority. For example, a variety of other disabilities or diagnoses have strong implications for overall child health and wellbeing that may similarly influence whether hearing-related needs are given appropriate attention.

Other limitations exist that should be considered when interpreting this data. For one, the codes and themes generated from the interviews were only reviewed by JJW and NJP. Having the remaining authors on this paper review the codes and themes as they were generated may have increased the rigour of the data analysis. Because a grounded theory design is qualitative, it is not possible to generalize the findings revealed in this study (Creswell & Poth, 2018). Even if this was not the case, generalization would be limited given the homogenous nature of the sample used. For example, unintentionally, all participants in this study were mothers and almost half of participants reported a high annual income and education level. Therefore, it could be that several considerations for how mothers determine priority for effective hearing care and management are left undiscovered as a result of lacking diverse backgrounds.

Conclusion

Parents of children who are DHH with Down syndrome have a wide variety of challenges and experiences that influence what level of priority they attribute to hearing-related needs. This study theorized that the higher the extent of engaged professional support, the extent of how parents perceive benefit, the level of parent activation, and the quality of family engagement, the higher the priority parents place on hearing care and management. Audiologists and other providers may consider ways to approach their service delivery to create partnerships with parents to maintain a high priority for hearing care despite children's additional complex needs.

References

- Aro, T., Eklund, K., Nurmi, J., & Poikkeus, A. (2012). Early language and behavioral regulation skills as predictors of social outcomes. *Journal of Speech, Language, and Hearing Research*, 55, 395-408. DOI: 10.1044/1092-4388(2011/10-0245).
- Bauer, A.M., Thiekle, S.M., Katon, W., Unützer, J., and Areán, P. (2015). Aligning health information technologies with effective service delivery models to improve chronic disease care. *Preventative Medicine*, (0), 167-172. DOI: 10.1016/j.ypmed.2014.06.017.
- Blaiser, K. (2012). Supporting communicative development of infants and toddlers with hearing loss. *Seminars in Speech and Language*, 33(4), 273-279. DOI: 10.1055/s-0032-1326911.
- Botting, N., & Conti-Ramsden, G. (2008). The role of language, social cognition, and social skill in the functional social outcomes of young adolescents with and without a history of SLI. *British Journal of Developmental Psychology*, 25, 281-300. DOI: 10.1348/026151007X235891.
- Brown, J.B., Stewart, M., and Ryan, B.L. (2003). Outcomes of patient-provider interaction. In, Thompson, T.L., Dorsey, A., Parrott, R., and Miller, K. (2003). *The Routledge Handbook of Health Communication* (1st Edition). Routledge: NY.
- Carter-Edwards, L., Skelly, A.H., Cagle, C.S., and Appel, S.J. (2004). They care but don't understand: Family support of African American women with type 2 diabetes. *Diabetes Education*, 30, 493-501.

- Center for Disease Control and Prevention. (2019). *Data and Statistics on Down Syndrome*.
<https://www.cdc.gov/ncbddd/birthdefects/downsyndrome/data.html>. [Access 28 Jan 2020].
- Convery, E., Hickson, L., Keidser, G., and Meyer, C. (2019). The Chronic Care Model and chronic condition self-management: An introduction for audiologists. *Seminars in Hearing*, 40(1), 7-25.
- Corbin, J., & Strauss, A. (1990). Grounded theory research: Procedures, canons, and evaluative criteria. *Qualitative Sociology*, 13(1), 3-21.
- Corbin, J., & Strauss, A. (2008). *Basics of Qualitative Research: Techniques and Procedures for Developing Grounded Theory* (3rd ed.). Thousand Oaks, CA: SAGE Publication, Inc.
- Creswell, J., & Poth, C. (2018). *Qualitative Inquiry and Research Design: Choosing Among Five Approaches* (4th ed.). Thousand Oaks, CA: SAGE Publication, Inc.
- DesJardin, J.L., Eisenberg, L.S. (2007) Maternal contributions: Supporting language development in young children with cochlear implants. *Ear and Hearing*, 28(4), 456–469. DOI: 10.1097/AUD.0b013e31806dc1ab.
- DiMatteo, M.R. (2004). Social support and patient adherence to medical treatment: A meta-analysis. *Health Psychology*, 23, 207-208.
- Gabay, G. (2015). Perceived control over health, communication and patient–physician trust. *Patient Education and Counseling*, 98(12), 1550-1557.

Gallant, M.P., Spitze, G.D., and Prohaska, T.R. (2007). Help or hindrance? How family and friends influence chronic illness self-management among older adults. *Research on Aging*, 29(5), 375-409. DOI: 10.1177/0164027507303169.

Gallaudet Research Institute (2011). Regional and national summary report of data from the 2009-10 annual survey of deaf and hard of hearing children and youth. Washington, D.C.: GRI, Gallaudet University.

Hayes, S.C., Luoma, J.B., Bond, F.W., Masuda, A., and Lillis, J. (2006). Acceptance and Commitment Therapy: Model, processes and outcomes. *Behaviour Research and Therapy*, 44(1), 1-25. DOI: 10.1016/j.brat.2005.06.006.

Hibbard, J.H., and Greene, J. (2013). What the evidence shows about patient activation: Better health outcomes and care experiences; fewer data on costs. *Health Affairs*, 2, 207-214.

Johnson, C.J., Beitchman, J.H., & Brownlie, E.B. (2010). Twenty-year follow-up of children with and without speech-language impairments: Family, educational, occupational, and quality of life outcomes. *American Journal of Speech-Language Pathology*, 19, 51-65.

Joint Committee on Infant Hearing. (2019). Year 2019 position statement: Principles and guidelines for early hearing detection and intervention programs. *Journal of Early Hearing Detection and Intervention*, 4(2), 1-44.

Malesa, E., Foss-Feig, J., Yoder, P., Warren, Z., Walden, T., & Stone, W.L. (2013). Predicting language and social outcomes at age 5 for later-born siblings of children with autism spectrum disorders. *Autism*, 17(5), 558-570. DOI: 10.1177/1362361312444628.

- Miller, T.A., and DiMatteo, M.R. (2013). Importance of family/social support and impact on adherence to diabetic therapy. *Diabetes, Metabolic Syndrome and Obesity: Targets and Therapy*, 6, 421-426.
- Muñoz, K., Olson, W., Twohig, M., Preston, E., Blaiser, K., & White, K. (2015). Pediatric hearing aid use: Parent-reported challenges. *Ear and Hearing*, 36(2), 279-287.
- Muñoz, K., Rusk, E., Nelson, L., Preston, E., White, K., Barrett, T., & Twohig, M. (2016). Pediatric hearing aid management: Parent-reported needs for learning support. *Ear and Hearing*, 37, 703-709.
- Pennarola, B.W., Rodday, A.M., Bingen, K . . . and Parsons, S.K. (2015). Changing factors associated with parent activation after pediatric hematopoietic stem cell transplant. *Support Care Cancer*, 23, 1997-2006. DOI: 10.1007/s00520-014-2544-1
- Reynolds, B., Yoho, S.E., Muñoz, K., and Pitt, C. (2019). Family involvement in adult hearing evaluation appointments: Patient perspectives. *American Journal of Audiology*, DOI: 10.1044/2019_AJA-19-0013.
- Rollnick, S., Miller, M.R., and Butler, C.C. (2008). *Motivational Interviewing in Health Care*. New York, NY: Guilford Publication, Inc.
- Rosenstock, I.M., Strecher, V.J., and Becker, M.H. (1988). Social Learning Theory and the Health Belief Model. *Health Education Quarterly*, 15(2), 175-183.
- Walker, E., Spratford, M., Moeller, M., Oleson, J., Ou, H., Roush, P., & Jacobs, S. (2013). Predictors of hearing aid use time in children with mild-severe hearing loss. *Language, Speech, and Hearing Services in Schools*, 44, 73–88.

Whicker, J., Muñoz, K., & Nelson, L. (2019). Parent challenges, perspectives, and experiences caring for children who are deaf or hard-of-hearing with other disabilities. *International Journal of Audiology*, 58(1), 5-11. DOI: 10.1080/14992027.2018.1534011

Whicker, J.J., Muñoz, K., & Pearson, N. (2020). Audiology-parent engagement enhances hearing care of children with Down syndrome. *The Hearing Journal*, 73(4), 8-9. DOI: 10.1097/01.HJ.0000661576.11834.2a.

Tables and Figures

Table 1.

Demographic description (n=18)

<i>Demographic</i>	<i>%(n)</i>	<i>M</i>	<i>SD</i>	<i>Range</i>
Relationship				
Mother	100(18)			
Parent Age (years)				
<20	0(0)			
21-30	11(2)			
31-40	33(6)			
41-50	33(6)			
>50	23(4)			
PNA	0(0)			
Income				
<\$20,000	11(2)			
\$21-40,000	23(4)			
\$41-60,000	0(0)			
\$61-80,000	11(2)			
>\$80,000	44(8)			
PNA	11(2)			
Education Level				
High School/GED diploma	5(1)			
Some College	17(3)			
Baccalaureate degree	23(4)			
Graduate degree	44(8)			
Vocational training	11(2)			
PNA	0(0)			
Child Age (years)		8.3	5.5	16 months – 20 years
Child Gender				
Male	44(8)			
Female	56(10)			
Interview Length (minutes)		36.4	7.5	21-51 minutes

PNA=Preferred not to answer

Figure 1

Model for The Hearing Care and Management Priority Theory

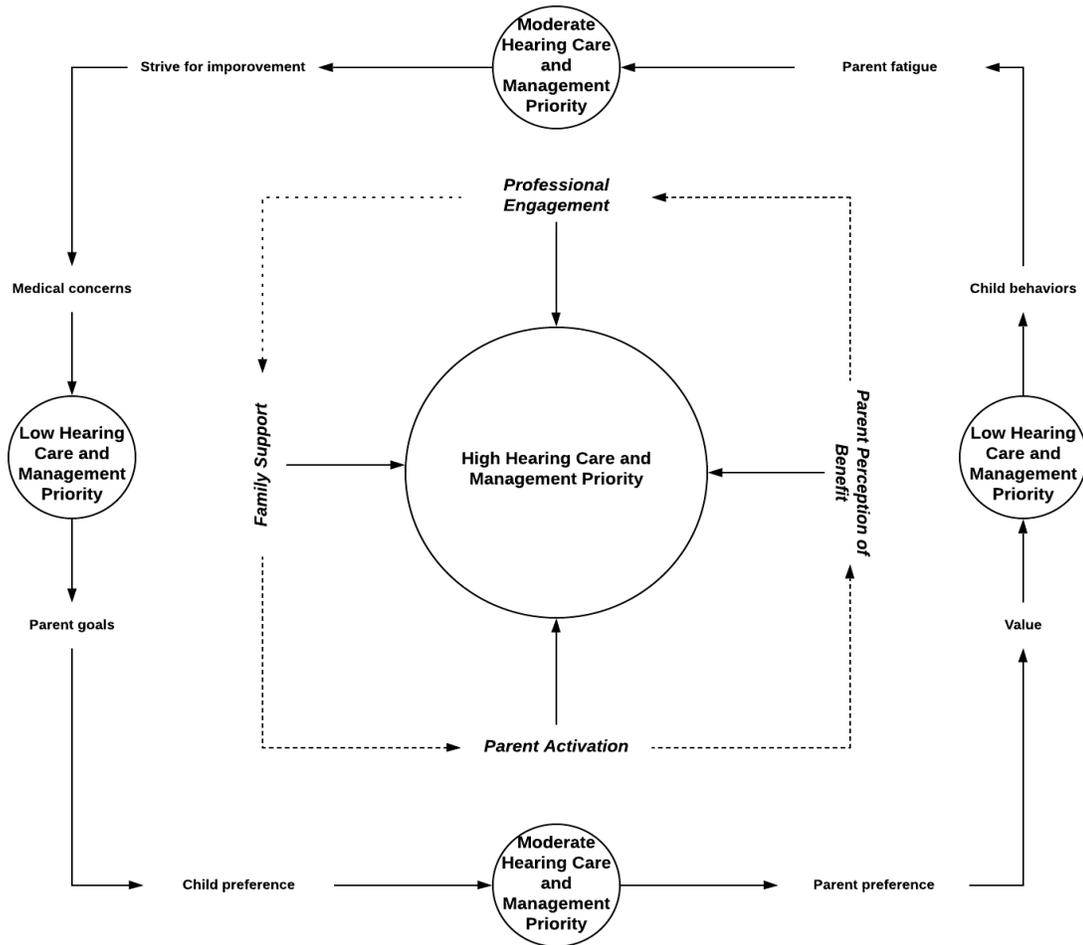
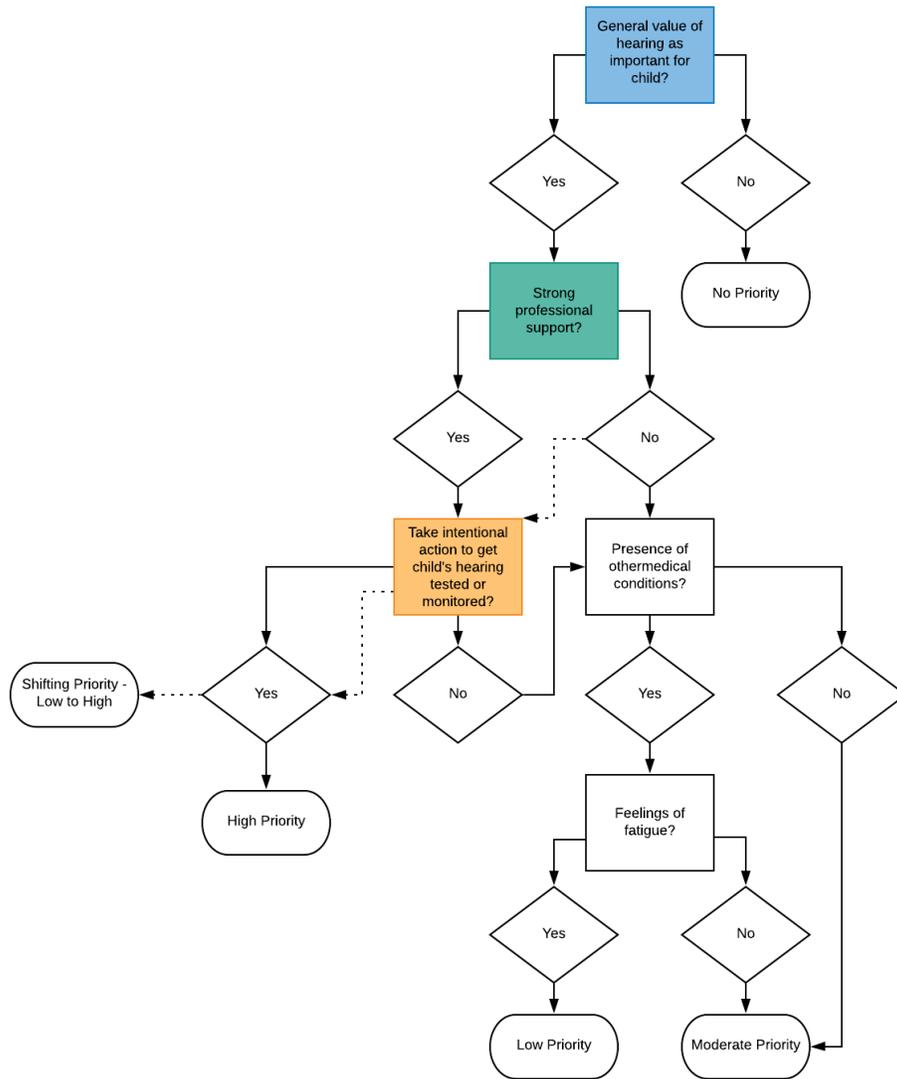


Figure 2

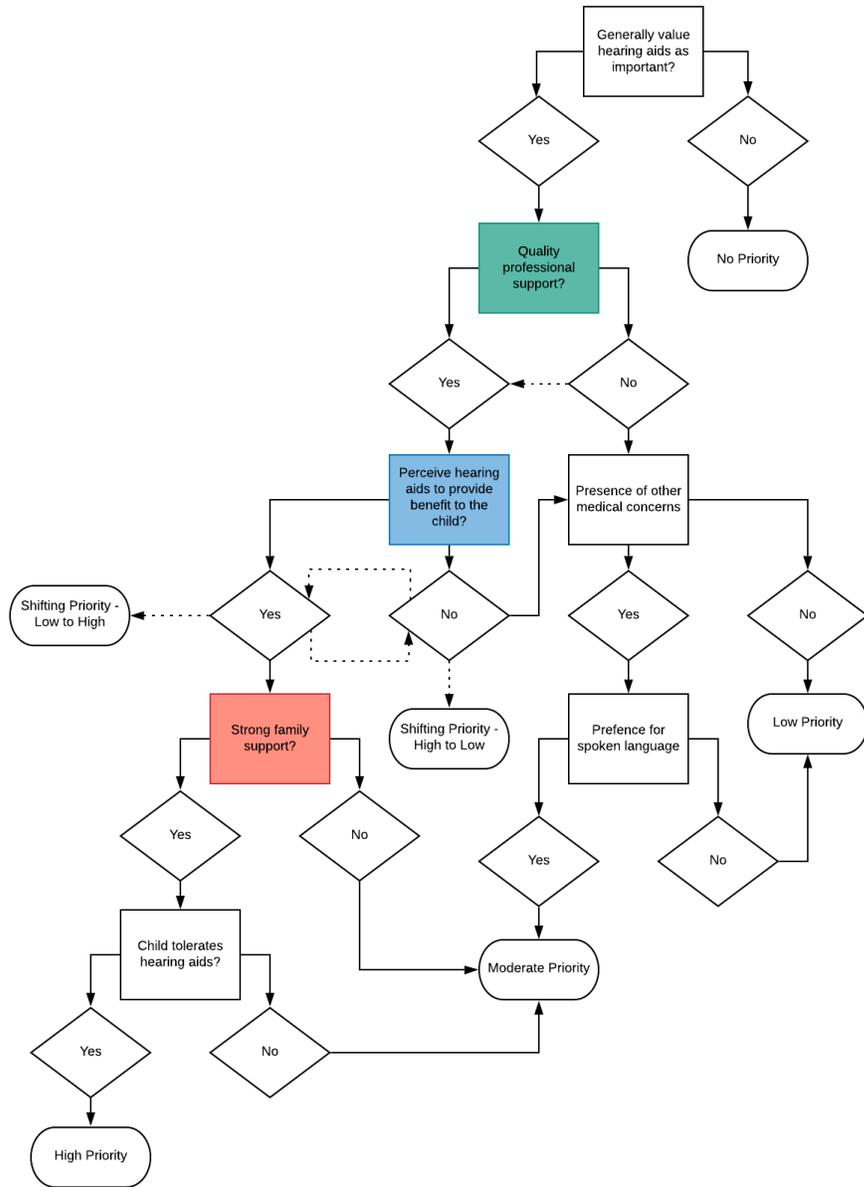
Hearing Health Priority Decision Tree



Note: Dotted line infers secondary decision making to influence shifting priority

Figure 3

Hearing Aid Priority Decision Tree



Note: Dotted line infers secondary decision making to influence shifting priority

Figure 4

Communicative Development Priority Decision Tree

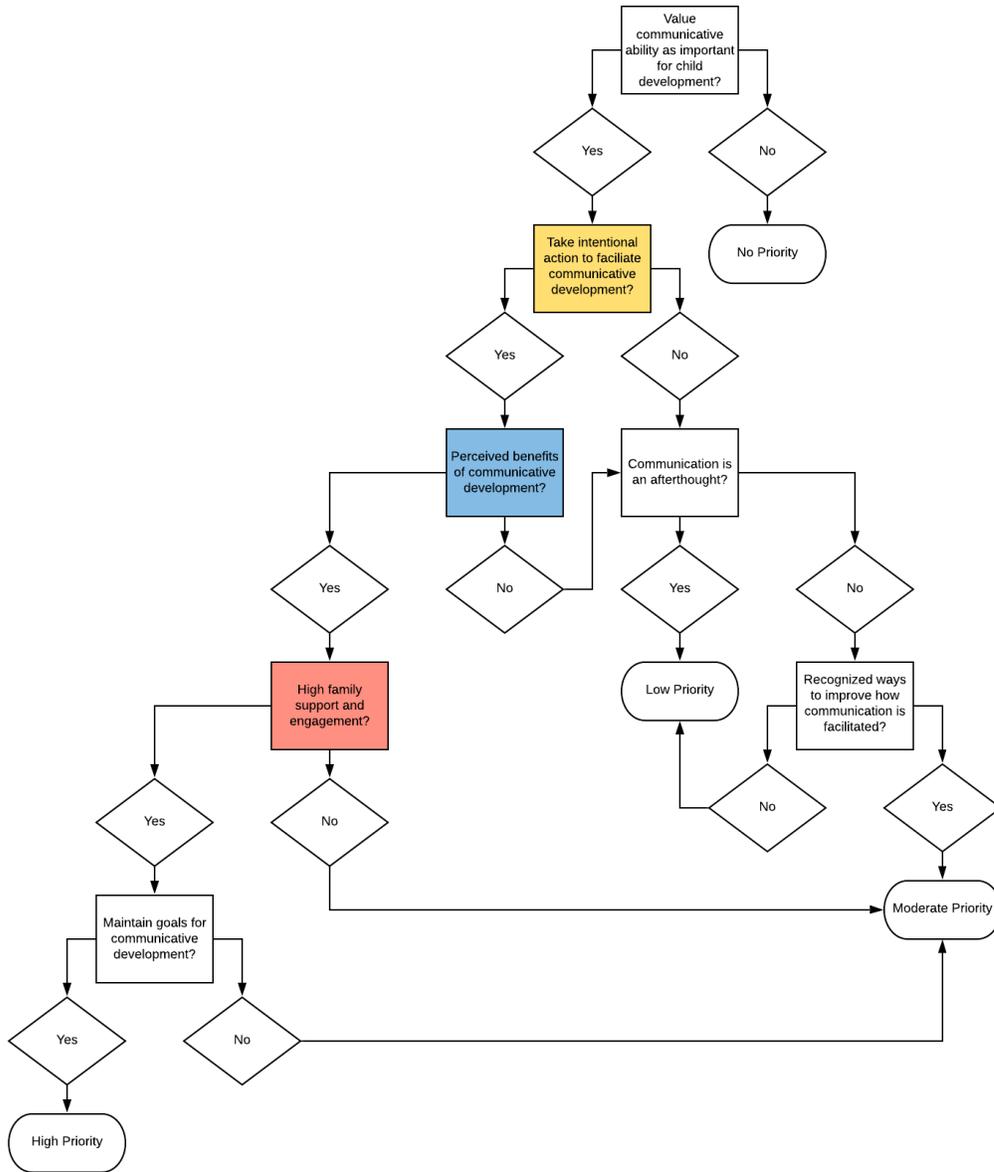


Figure 5

Speech-language Therapy Priority Decision Tree

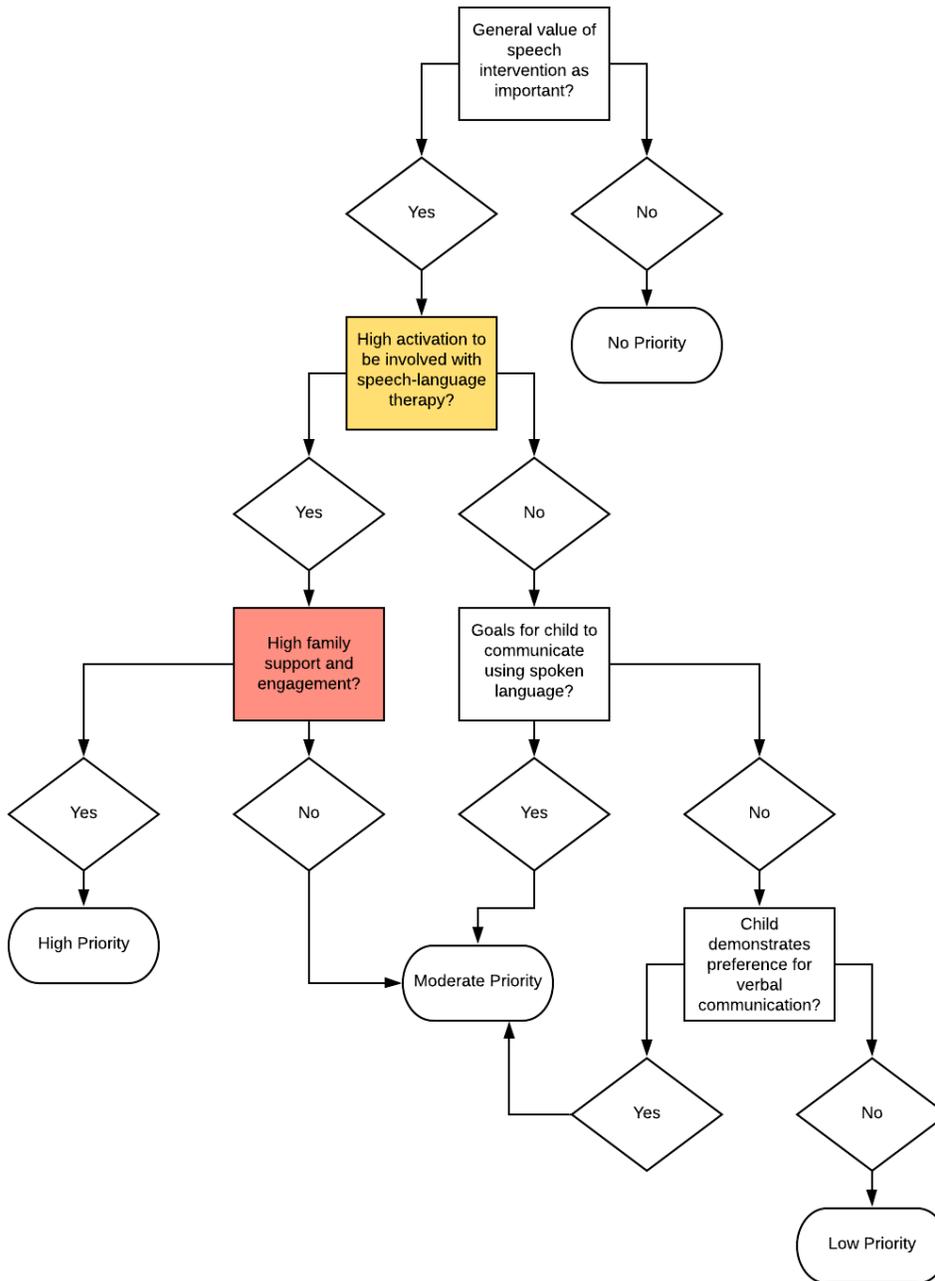
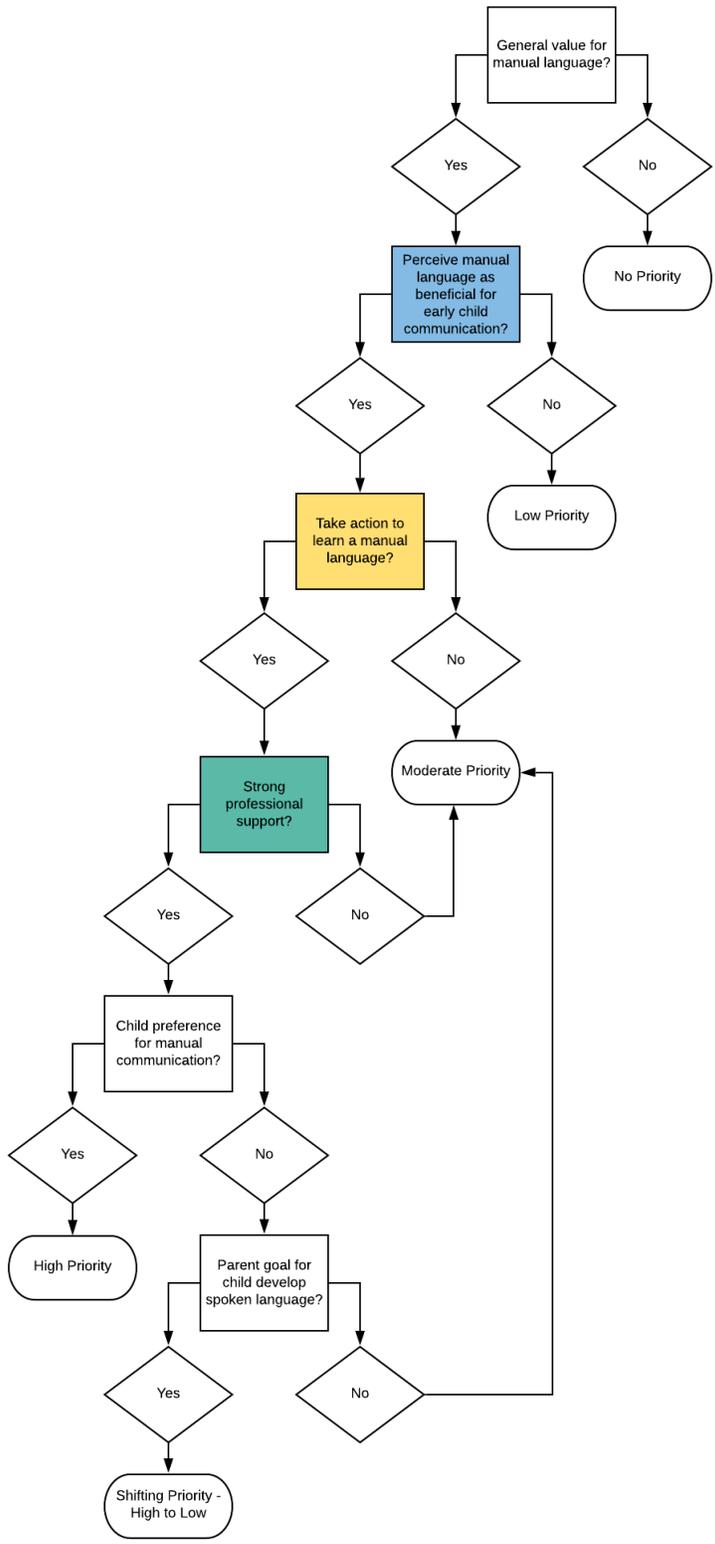


Figure 6

Visual Language Priority Decision Tree



Appendix: Stem Questions

1. Tell me about what it's like caring for a child who has hearing loss and Down Syndrome.
2. One a scale of 1-10 (10 being very important, 1 being little or not important) how important are hearing aids to you? Why?
3. How much does your child use his/her hearing aids?
4. One a scale of 1-10 (10 being very important, 1 being little or not important) how important is learning a manual language to you? Why?
5. How much time do you spend learning a manual language?
6. One a scale of 1-10 (10 being very important, 1 being little or not important) how important is your child's communication to you? Why?
7. How much time do you spend using strategies with your child to facilitate communication?
8. One a scale of 1-10 (10 being very important, 1 being little or not important) how important is your child's speech intervention? Why?
9. How much time do you spend collaborating with your child's speech interventionist?
How much time do you spend transferring skills learned in speech therapy to the home?
10. Tell me about the family support you receive. How has family support influenced your decision for hearing care priority?
11. Tell me about the professional support you receive. How has professional support influenced your decision for hearing care priority?