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Development of a Decision Support Tool in Pediatric Differences/ Disorders of Sex Development

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Abstract

Decisions about how best to clinically care for young children born with Disorders of Sex Development (DSD) can be challenging because some decisions are irreversible, have lasting physical and mental health effects, and are frequently made before the affected person is able to participate in decision-making. This multi-stage study involved (1) the development of a web-based decision support tool (DST) for parents of infants or young children and the clinicians caring for them; (2) the assessment of communications and decision making between DSD specialists and parents both before and after introduction of the DST; and (3) interviews with a broad range of stakeholders regarding optimizing the DST and integrating it into usual care. Experience over the course of the 3 stages of this research suggests the need for further refinement of the DST to increase acceptability to all stakeholder groups, the necessity to address misperceptions by providers that they are already accomplishing all aspects of SDM in regular care without a DST and misunderstandings by parents that decisions are unnecessary because only a single option is apparent, and to better incorporate the tool into regular clinic workflow.

Keywords

Decision support tool; Disorders of sex development; Shared decision-making

Introduction

Shared decision-making (SDM) has been promoted in healthcare by a number of professional societies and advocacy groups.[1,2] The objective of SDM is to help patients (or their surrogates) make informed, preference-based clinical management choices among several relevant options.[3] SDM does not imply that providers and patients must have equal responsibility for the final decision.[4] Rather, SDM combines healthcare providers' expert knowledge and patients' (or surrogates') rights to make healthcare decisions with full information; it requires the involvement of both healthcare providers and the patients/caregivers, with bidirectional information exchange, mutual deliberation on treatment options, and agreement on treatment plans.[5,6]

In the context of SDM, decision support tools (DSTs) have evolved as a widely accepted approach to improve patients' and surrogate decision-makers' understanding of treatment-related information, promote informed decision making consistent with patient values and preferences, and minimize decision-related stress.[7,8] However, few DSTs have been developed to assist parents and clinicians navigate complex decisions, in particular elective surgical decisions, about a child's care.

Disorders of sex development (DSD)

DSD is an umbrella term encompassing congenital conditions in which chromosomal, gonadal, or anatomic sex development is atypical.[9] They are most commonly diagnosed at birth because of visible genital differences or because of discordance between prenatal

genetic testing and the child's genital phenotype. Dependent on the definition of DSD applied, the estimated incidence is as high as 1% of all live births.[10]

Decision-making In DSD

Although conditions encompassed by the term DSD are diverse in terms of pathophysiology, they share overlapping features shown to exert profound effects on family adjustment, parenting, and emerging self-concept of the affected person. Decisions faced by families and healthcare providers on behalf of newborns and young children with DSD may include gender of rearing; if, how, and when to talk to others (e.g., extended family, close friends, the child him/herself, and siblings) about the DSD; contacting peer support and/or advocacy organizations; pros and cons of next-generation genetic sequencing; surgery (genital and/or gonadal) and its timing; and others.[11,12]

Clinical management decisions (some, largely irreversible) are commonly made in the first year of life, a time when parents can feel intense emotional distress and struggle with learning about their child's medical condition and its implications. Anxiety-driven decisions often reflect perceptions of a limited range of options with inadequately-weighed risk and benefit potentials.[13-18] Often, the "right" decisions about best courses of action are not always obvious. While major advances in diagnostic assessment (e.g., genetic testing) have occurred, results do not always reveal the cause of the DSD. Even when the tests uncover the responsible genetic variation, that knowledge does not commonly lead to a single "correct" treatment plan as a diagnosis (even a molecular genetic diagnosis) may suggest a number of different clinical management options -- each of which may be associated with a positive outcome.[19,20] Additionally, many treatment options are considered not only elective, but also controversial.[21-24]

With the assistance of the child's healthcare providers and others, parents need to make decisions based on their knowledge of all options and associated risks and benefits of each and modulated by personal and family preferences, values, and religion/culture.[25,26] However, parents often do not recognize that there are decisions to be made, nor that they play important roles in the SDM process.For example, a qualitative study of parental experiences revealed that many parents viewed genital surgery to "normalize" their child's sexual anatomy as an obvious and necessary clinical management component; i.e., for them, there was no point at which any decisions needed to be made regarding genital surgery.[27] Additionally, former patients and healthcare advocates complain that parents are provided inadequate information with which to make decisions for their child. Research corroborates the claim that parents have often not heard of DSD conditions before their child's birth and typically do not perceive that treatment choices exist; instead, many assumed the diagnosis implied a single, specific treatment path.[27]

Objectives

As some former patients, patient advocacy, and human rights organizations complain that families are not provided adequate information, nor sufficient professional and peer support necessary to make informed decisions, the overall objective of this project was to develop a DST for healthcare specialists and parents to navigate complex and contentious clinical

management decisions on behalf of their young children. More specifically, this research set out to: (1) determine the content and delivery format for a DST desired by key stakeholders (i.e., parents of affected children, affected adults, patient advocates, pediatric endocrinologists, geneticists, gynecologists, psychologists, urologists/surgeons, ethicists, counselors, and others); (2) assess the quality of communications between providers and parents, both before and after introducing the DST into regular care; and (3) determine what is needed to fully integrate the DST into routine clinical management from the perspective of all stakeholders. Also evaluated where what specific characteristics of the DST and the healthcare system affect its likelihood for implementation.

Methods

Aligned with the three objectives, this project proceeded in three phases: DST development, examining provider-family communication during clinic visits, and identifying opportunities and barriers to DST implementation.

DST development

Guiding principles.—DST development was guided by the 2006 DSD consensus statement[9] and the International Patient Decision Aid Standards (IPDAS) effectiveness criteria.[28-30] The 2006 consensus statement recommends a multidisciplinary team approach (including families and specialists who form a core, including endocrinology, surgery and/or urology, and behavioral health); transparent communication of diagnostic and procedural information with parents, and emotional and informational support (via professional or peer support group approaches.) These elements were recognized to be integral to the care and ongoing management of patients.[9,31] Compared to usual care in the absence of a DST, or care using DSTs that are inadequately constructed, DSTs that apply IPDAS criteria have been found to improve knowledge, reduce decisional conflict, and help align participants' personal values with the decision.[32,33] Finally, the DST component addressing expanded genetic testing was informed by the 2013 Academy of Pediatrics' policy statement regarding genetic testing in minors and disclosure of incidental findings.[34]

Development.—This process of constructing the DST began by creating a "content document" (i.e., guidelines that specify the principles and goals of the DST). A draft content document based on peer-reviewed research literature and cumulative clinical experiences of the investigators was shared for comment with stakeholders who were recruited via "snowball sampling".[35] Collated feedback from recruited stakeholders (patient advocacy, n=2; clinical research, n=1; healthcare providers in endocrinology, genetic counseling, genetics, psychology, urology/surgery, n=5; public health, n=1; sociology, n=1;) led to content document revisions which were subsequently shared with the entire research team and external stakeholders in an iterative fashion and until a consensus-based final version was produced. Stakeholders identified the following as essential ingredients for a DSD-specific DST: (1) presenting information regarding common diagnostic procedures (biochemical and genetic, imaging studies, exploration under anesthesia) and their timing; (2) interpretation of "incidental" genetic findings and carrier status; (3) interpretation of test results and their relationships to gender assignment decisions; (4) providing balanced

information about treatment options and associated risks and benefits; (5) eliciting caregiver values and preferences specific to treatment options; (6) using personal stories/testimonials; (7) coaching caregivers in effective communication with the healthcare team; (8) disclosing potential conflicts of interest; (9) addressing health literacy; and (10) delivering decision support on the internet.

Following the methods and standards used in developing the content document, the web-based DST was similarly created via an iterative process of drafting materials and receiving feedback from stakeholders (parents of affected children recruited through multispecialty DSD clinics and patient advocacy organizations, specialty providers, and patient advocates). All feedback was qualitatively coded and sorted into themes. Informal "member-checking"[36] of these themes occurred via email or phone exchanges with more formal checks occurring as edits, based upon the feedback. Edits were subsequently shared and further revised until an online DST was established. Qualitative analysis of the feedback led to identification of several themes – accessibility, language, structure, and navigation – commonly emphasized in human factors engineering; i.e., the field that focuses on creating systems based on user-centered designs.[37,38]

Identifying and handling controversy.—DSD comprise numerous medical conditions for which the "right" decisions about the course of action are not always obvious and can, at times, become controversial. At all stages of development, diverse groups of stakeholders were sampled to help identify components, and provide feedback on DST development. Stakeholders included parents of affected children, patient resource and advocacy leaders and members, specialty (endocrinology, genetic counseling, genetics, gynecology, psychology, nursing, and urology/surgery) and primary care providers, bioethicists, and others. It was intended, and was borne out in his project, that sampling this variety of stakeholders should result in a wide range of perspectives. Differences observed in views included: geneticists wanting to include highly detailed information on genetic testing and causes of DSD as well as carrier testing; patient advocates encouraged limiting information to "just the basics," so that parents can identify occasions when decision-making opportunities exist, pros and cons of each option, and known risks and benefits without becoming overwhelmed. Specialist healthcare providers urged that the DST be a stand-alone product that families could use largely on their own, while patient advocacy representatives called for its use in the clinical setting alongside the providers. Additionally, some patient advocates have equated all elective surgical interventions on genital or reproductive structures to “torture.” Some of these advocates have worked with the UN High Commissioner for Human Rights and the UN Special Rapporteur on Torture and Other Cruel, Inhuman or Degrading Treatment or Punishment to call for the “prohibition of surgery and treatment on the sex characteristics of minors without informed consent."[39] Further, some of these advocates called for the inclusion of this perspective in the DST. In contrast, other patient advocates and healthcare providers opposed inclusion of this material – concerned that it would no longer allow for parental choice and decision-making on behalf of their children with regard to surgery. As such, shared / cross-cutting perspectives across groups were included in the provisional DST (e.g., educating families, supporting families, providing known pros and cons about procedures). In contrast, positions stated by any one

stakeholder group which were in conflict with all other stakeholder group perspectives were noted, but not included in the final design.

Family-provider communication during clinic visits

Sixty-three parents/caregivers of 31 young children who were seen at one of three multidisciplinary DSD clinics agreed to allow the research team to audio-record their interactions with providers during their clinic appointments. Providers (n=60) included specialists in endocrinology, genetic counseling, genetics, gynecology, nursing, psychology, social work, and urology. Index cases were identified via a review of medical charts for those with upcoming appointments at three medical centers. All potentially eligible participants (i.e., adult caregivers of young children 0-5.9 years being seen for the assessment and/or management of a DSD in which at least one major clinical management decision needed to be made on behalf of the child) were informed about the study and offered participation. Caregivers included biological, adoptive, and step-parents; grandparents; and other family members who were regularly involved with the child's care.

Thirty-six caregivers participated prior to DST creation; 25 different caregivers participated after its creation. Recordings took place before and after clinical management decisions had been made. Decisions differed by index case (e.g., whether or not to purse genetic testing, whether or not to pursue a surgical intervention). After sharing the DST with families and providers in clinic, audio-recorded consultations provided clues regarding uptake and use in the context of regular care.

Opportunities and barriers to DST implementation

In addition to feedback gleaned from caregivers who were provided access to the DST and whose clinic appointments were audio-recorded, 40 additional stakeholders (parents of affected children, n=11; resource and advocacy organization leaders, n=4; healthcare providers, n=25 –two of whom were the primary care physicians of chart-selected patients whose parents participated in the recorded clinic visits) provided feedback on the provisional DST that was developed for use in clinic. All participants were provided access to the on-line DST and provided feedback via web, email, and/or telephone interview. Content analysis of transcribed interviews and written feedback of healthcare provider and patient advocate stakeholders was conducted. Content was categorized into discrete domains and topics of concern: positive/negative features of DST, changes needed, relevance, format, timing for use as well as an identification of the factors influencing use of the DST, decision support and impact on the shared decision-making process.

Results

The DST

The web-based DST (“DSD Support Tool: A Guide for Parents”; no longer online) presented educational content and supported interactive involvement and data collection. The DST opened with an introductory module, seen by all users, which conveyed basic information about DSD and social support, including considerations for sharing information about the child’s medical condition with family and close friends (see Figure 1 for a list of

all modules). Basic information was presented to all users, with the option to dive deeper into topics via hyperlinks. Based on the child's karyotype and genetic diagnosis (or anatomic phenotype in cases where genetic diagnosis was unavailable), caregivers were directed to follow tailored paths that provided more specific information about their child's DSD, its evaluation and treatment options, and were assisted with identifying the near-term decisions they may face regarding gender of rearing, gonadal management and genital management. Information graphics accompanied the text as visual representations of the information and these could be printed out. Families were asked to consider other issues important to their particular decision and clarify their family values using a "weigh scale" (eg, see Figure 1, Interactive Decision Support Tool: Components: Values) to rate the perceived importance of particular decisions and associated values. They were also asked to identify their preference for participation in decision making[40] and to indicate their predisposition or "leaning" toward certain decisions.[41] A summary of their value and decision preferences could be printed out or electronically transmitted to their child's DSD team for further discussion. Parents were also asked to list their questions (via a pre-identified question prompt list) to be discussed in follow-up visits with healthcare providers.[42]

Family-provider communication during clinic visits

Actual use of the DST among caregivers who were provided access to the DST varied across and within families. Use was tracked online with a software plug-in that tracked when users completed each of the decision-making exercises. Few caregivers used the web-based DST as designed: 16% of caregivers (representing 15% of children) reviewed all content and completed decision-making exercises; 44% (representing 54% of children) used it at least partially, and 40% (representing 31% of children) did not use it past initial log-ins at clinic or home. Usage and patient age were related [$F(2,22) = 25.1$ $p < .001$] such that those who used it fully had older children ($m = 4.2$ years, $p < .001$) than those who used it partially ($m = 0.9$y) and those who did not use it past initial login ($m = 0.2$y, $p < .001$); the difference between the latter two groups did not reach statistical significance ($p < .13$). Three caregivers, who reported experiencing technological issues, requested and received printed-out/paper copies of relevant DST elements and are included in the counts above. With regard to those who neither used the DST beyond initial log in nor requested a paper copy after experiencing technical difficulties, clinical and research coordinators asked caregivers if they would like printed copies; all declined.

In-clinic recordings showed that, aside from brief mentions when caregivers were first provided log in information to the DST, few clinicians explicitly mentioned its use during subsequent encounters. A comparison of transcribed audio-recordings, prior to introduction of the DST with those in which providers and caregivers were provided access to the DST, did not reveal systematic differences in the qualities of the clinician-caregiver communication.[43]

Opportunities and barriers to DST implementation

Feedback from caregivers in clinic.—Qualitative assessments of caregivers' user experience with the DST, conducted via semi-structured interview, revealed that satisfaction

with specific aspects of the DST varied. A majority were positive as to how and by whom the DST was introduced, reporting that the DST was introduced in the DSD clinic by either a member of the research team or by a team provider. Most also reported the DST was introduced during their first appointment at the DSD clinic; however, a little under half experienced this as overwhelming, in combination with other information received during the first appointment. One caregiver remarked that it would have been helpful to have access to the DST prior to the first clinic appointment (Table 1, panel A). Among those who accessed the online DST, most reported positive feelings regarding the aesthetics, functionality, and ease of navigation (Table 1, panel B); however, several reported having technical difficulties with logging onto the website, with some indicating they accessed the DST via paper copies.

Caregivers had conflicting feelings regarding the content of the tool. Some felt that the tool did not provide enough of the specific information that they sought; yet others felt that the tool provided too much information. Some caregivers wished they had accessed the online DST with their partner or spouse, or together with a provider (Table 1, panel C). Most also reported that the information available in the DST was consistent with the information provided to them by their healthcare providers. Global caregiver experience in using the DST was described as positive (Table 1, panel D).

Finally, responses regarding the DST's influence on DSD treatment decision-making suggest the DST was most useful in helping caregivers think about how involved personally they wanted to be in the decisions as well as helping them to identify questions to ask of the healthcare team. Detracting from its utility, some caregivers reported that the DST had been introduced after they believed a decision had already been made or that no decision needed to be made in their specific case.

Feedback from stakeholders outside clinic.—Participants identified the following needs: (1) refine the DST in order for it to be viewed as acceptable to all stakeholder groups; (2) convince providers to alter their usual model of care by integrating the DST in their practice; and (3) implement effective systems for delivering decision support and adequate follow-up. More specifically, with regard to DST refinement, stakeholders noted some technological glitches and a general reading level that was considered too high for some parents. However, these were not universal critiques; e.g., while advocates mentioned reading level concerns, parents of DSD-affected children did not. Additionally, some disagreement about terminology (e.g., "intersex" vs "DSD") used in the DST remained unresolved as different stakeholder groups held contrasting and mutually exclusive opinions about appropriate terminology. Concerned about overwhelming families at already lengthy follow-up visits, healthcare providers preferred introducing the tool in clinic and encouraging families to further use it at home on the condition that they had proper instructions for using the tool and wrote down their questions and concerns. In contrast, representatives of DSD support and advocacy organizations underscored the importance for healthcare providers to review DST content together with families.

Discussion

Decision aids and support tools are commonly found in adult healthcare and have been shown, in systematic reviews, to reduce decisional conflict stemming from perceptions of inadequate information and lack of clarity about personal values, to increase the proportion of individuals who assume an active role in decision making, and to reduce the proportion of those who remain undecided after their use.[33] The DST for DSD is novel in that it was designed specifically for the surrogate-decision makers for infants and young children with a condition for which there is often no clear "best treatment." The goal of the resulting DST was to assist caregivers (and the child's healthcare providers) in navigating complex diagnostic and interventional aspects of DSD decision-making, as well as the associated psychological, social, and ethical aspects potentially driving decisions. This study represents an essential first step in understanding the unique decisional support needs of caregivers grappling with decisions associated with controversy and uncertainty in outcomes.

The finding that the availability of a DST does not automatically translate into its effective use[44-46] has been reported; results, here, are in-line with other research showing that fewer than expected parents were directed to use the web-based DST.[44] Potential barriers to full uptake noted in the literature include: technical problems, clinicians' limited understanding of how patient DSTs could be helpful, clinician perception that shared decision-making for DSD treatment was already commonplace, and external factors, such as efficiency targets and best practice recommendations.[44,47-49] Adoption of strategies from implementation science will be critical to advancing the use of DSTs to support a robust SDM process.[50]

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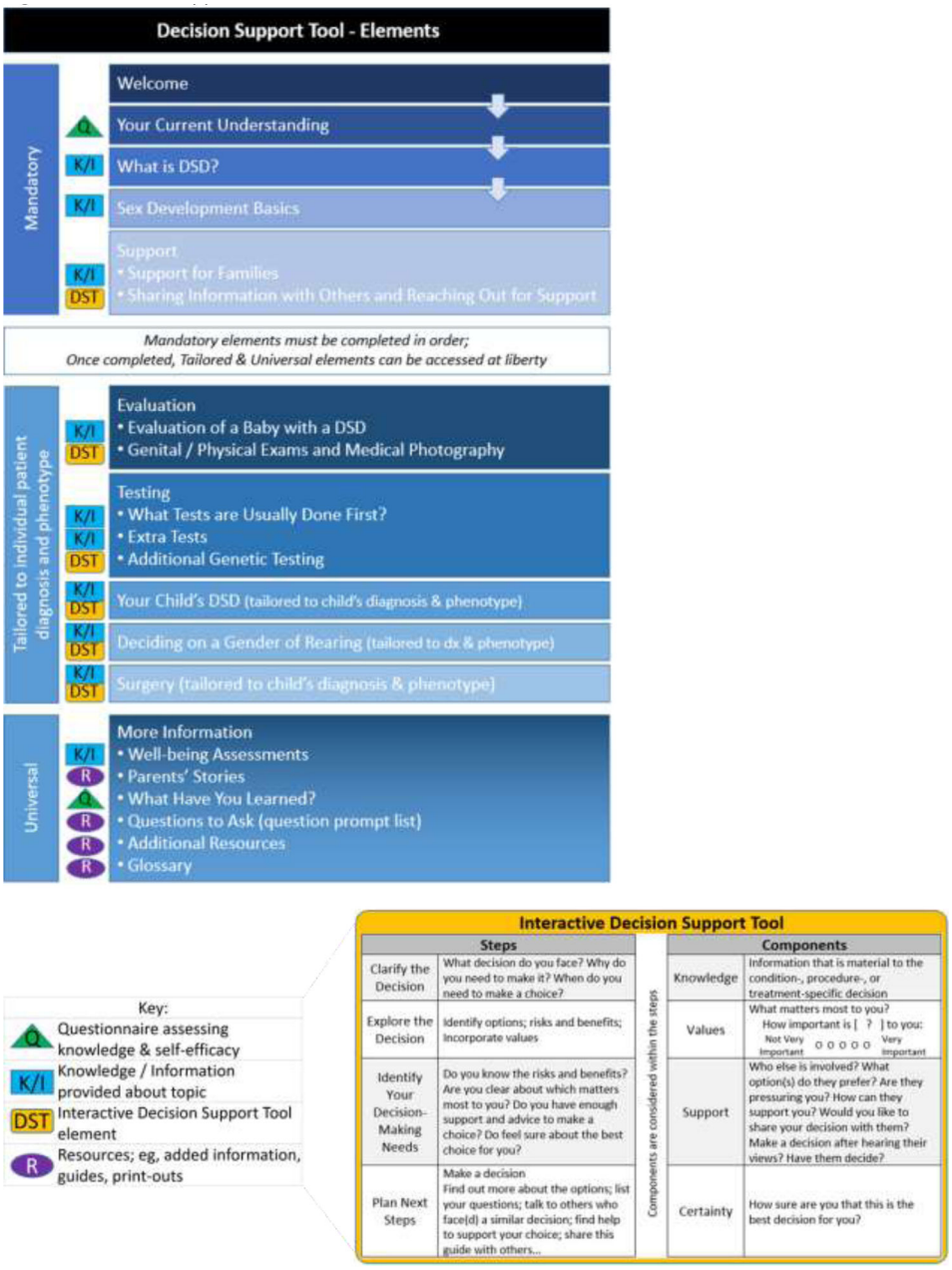

Figure 1.
Decision Support Tool Elements Flowchart

Table 1.

User experience: Caregiver feedback

A. Timing: Introducing the DST at the Initial Appointment
Before
• Prior to the appointment would have been fantastic, ‘cause at that point we didn’t have much information, we didn’t have much to go on.
During
• I would say that I was happy. Because at that time we were at a point that we were trying to figure out what was going on. Ya know, how do we, um how do we make all of these steps and decisions in the process, so when you introduced it as a tool that we were going to be able to take away outside of the office because you know the DSD clinic is good, but you’re getting a wealth of information thrown at you.
• The timing was great because… when you’re first learning about diagnosis and things like that, and the first thing you wanna do is get on google and um, research it and find out everything you can even though it’s probably not the best thing to do.
• I like to have something that I can take away and meditate on. Ya know, go through it at my leisure and understand, so it felt good to know that I was going to have an interactive tool that would allow me to um dig deeper into what was going on.
After
• I think just because there’s like a whirlwind of paperwork, and, um, and you’re still kind of reeling from the diagnosis and all of the, everything going on, it just felt a little cumbersome at that first appointment.
• I mean we just had a baby and then a month later, we’re in being told this and that, and then all of a sudden here’s a tool, and then, get on the, that type of a tool where there’s so much information where we don’t even have a diagnosis, is like way overwhelming.
B. Aesthetics, Functionality, and Ease of Navigation
• As far as format, it looked clean. It was very cut and dry and user friendly. There weren’t things all over the place, you know, making issues and that sort of thing, which when you are trying for this type of information, you wouldn’t want all of that type of noise in the way. That I did notice, that it was clean and it kept a professional, there was a professional appeal to it.
• Easy to navigate and kind of self-explanatory I guess you could say.
• I liked how you could go as deep as you wanted with your questions. You know, you could just kind of glaze over it and get very basic stuff, or if you had more questions about a specific thing, you could find that information out. So, it was basically your lead and it was based on your time…how much information you really wanted…
C. Desire for More Healthcare Provider Involvement
• I think what I would’ve preferred is one of them gave us a walk through, because it ended up being like well, this is the link and this and that… so I think whoever it is that shows it to you or that signs you up, it would be good to have like a walk through to show the modules…
• I just kinda got overwhelmed with myself…not knowing the exact diagnosis or anything so, I just, that’s why I kind of backed away…I guess if I looked at it with a member of the healthcare team…maybe they could explain, it would be easier, then you’d get some other explanations of some of the you know…the verbiage and stuff, with some of the terms…
• …with your healthcare provider, also because…they can explain something as you’re going, so that way you can, you know if you have any questions, then you can ask them right then and they can explain a little bit better about what it means.
D. Overall Evaluation
• The best thing about it…comes from the way you were presenting it…I think knowing that it was made with a parent in mind; that it was there to help us. It wasn’t just something else you could google. It wasn’t just another pre-prepared pamphlet or brochure, the fact that it was specialized in the beginning so that the profile matches your child’s situation, to the ability that testing shows. I think the fact that it was, you know, it was for us…I think that was the best thing about it.
• I just think it’s giving me more information, as to, you know, in that in-between visits with the doctors in person, I could get some of my questions answered.