

Meeting Minutes

National Institute of Diabetes and Digestive and Kidney Diseases (NIDDK)

Research Needs for Effective Transition in Lifelong Care of Congenital Genitourinary Conditions

National Institutes of Health (NIH) Natcher Conference Center
Bethesda, MD
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Final Meeting Summary

WELCOME

Robert Star, M.D., Director, Division of Kidney, Urologic, and Hematologic Diseases, NIDDK, NIH, Bethesda, MD

Dr. Robert Star welcomed the meeting participants. He brought additional greetings from Dr. Griffin P. Rodgers, Director of NIDDK, who was unable to attend. The NIDDK supports research in internal medicine and surgery for abdominal organs below the diaphragm plus the esophagus. The NIDDK sponsors a large amount of basic science, applied science, and clinical research, increasingly understands that research results must be translated to clinicians and the general public to improve health care in the United States.

Dr. Star began by recognizing that the topic of the meeting—improving the transition of urologic patients from childhood to adolescence to adulthood—is not straightforward to tackle. The meeting was designed to address the unmet research needs that will improve transition care for urologic patients, in a way that enhances the intersection between the urology and nephrology communities. Relevant conditions include urinary tract infections (UTIs), reflux, urinary incontinence, sexual function, and infertility. Dr. Star noted that to achieve effective transition for lifelong care of myriad urological conditions, many stakeholders will need to be engaged. These stakeholders, in addition to the NIH, include multiple federal organizations, professional schools, training programs, and professional organizations.

He then said that NIDDK is expanding its urology research agenda. NIDDK is supporting research investigating how to improve adherence to treatment in adolescents with chronic kidney disease. It is initiating the Prevention of Lower Urinary tract Symptoms (PLUS) Consortium to investigate behaviors that support bladder health and risk factors for bladder conditions in adolescent girls and women, laying the groundwork for future prevention studies. This group will consider incontinence, UTIs, and other bladder issues. Another new NIDDK effort will consider kidney stones across the lifespan, including in the pediatric population.

Dr. Star explained that the audience was assembled not to discuss genitourinary conditions per se, but to think about how most effectively to support the children, adolescents, and young adults that they serve in their clinical practices, health services, and research projects. These patients contend with issues affecting their body image, interactions with others, and relationships with friends. Patients in transition ask such questions as, How do others view me? Can I drink water? If so, how much? Can I exercise, and if I do will I leak? How do I date? Can I engage in sexual activity? Will childbirth worsen my condition? Through the meeting presentations and discussions, the participants would consider how best to provide care and support for transition patients.

Dr. Star concluded his remarks by thanking Dr. Bavendam for organizing the meeting, and to Dr. Marva Moxey-Mims, NIH colleagues from other Institutes, and the outstanding scientists and clinicians who were part of the planning committee. He expressed enthusiasm for the stimulating discussions that would be generated by the participants during the meeting, both within the conference room and in the hallways.

BACKGROUND

Tamara Bavendam, M.D., M.S., NIDDK, NIH, Bethesda, MD

Dr. Tamara Bavendam, a former urologic surgeon, shared that organizing the meeting required collaboration among several Institutes at the NIH and across multiple government agencies. She extended her appreciation to all those involved. The organizers assembled a stellar faculty with varied backgrounds who understand the current state of transition care and would be able to help facilitate discussions. Dr. Bavendam also thanked those who submitted poster presentations, noting that this is the first meeting at which NIDDK attempted an electronic format for posters.

Dr. Bavendam explained that the agenda will begin with a set of presentations that will frame the breadth and depth of the transition care theme. Presentations would provide insights and perspectives that surgeons might not think about on a regular basis. Breakout sessions following the presentations would identify research needs according to condition and then prioritize the most critical research questions. The breakout groups have been organized with the intent to cover many aspects of transition care.

Dr. Bavendam concluded by acknowledging that the goal of the meeting is challenging. She thanked the participants for their attendance and expressed appreciation for colleagues working behind the scenes, especially Ms. Jenna Norton, who was invaluable in helping to organize the meeting, and the Spina Bifida Association (SBA), which helped recruit the two women with spina bifida to provide a patient perspective.

INVITED PRESENTATIONS: SETTING THE STAGE

Transitions and Adult Health Care: Perspective of Individuals with Spina Bifida

Twila Hunt and Meezan Ford

Twila Hunt

Ms. Twila Hunt presented her experience with the health care system during her transition from pediatric to adult care from her perspective as a 40-year-old woman with spina bifida. She stated her central concern as a pediatric patient was that her many individual doctors were not located in one central place.

At age 23, when she moved out of her parents' home, a friend suggested she visit the Spina Bifida Clinic in Baltimore, Maryland, to help ease the transition to living on her own. This clinic accepts adults, and she was able to visit all her doctors within a 5- to 6-hour time frame. She greatly preferred this arrangement over having a series of individual appointments in different locations, which would be much more logistically challenging. Ms. Hunt remembered that these days felt long, but the appointments were convenient because she worked full time and needed to take only a single day off work. She visited the Clinic between the ages of 23 and 25.

When Ms. Hunt moved to the Washington, DC, area, she began to look for another clinic and was directed to attend the Spina Bifida Clinic at Children's Hospital in Washington, DC, which also accepts adults. Again, Ms. Hunt mentioned the benefit to being able to see all her doctors in one day.

From there, Ms. Hunt searched for a primary care physician (PCP) located closer to her home. Through her health insurance, she was able to meet with several candidates prior to making a decision. She ultimately chose a PCP based on the physician's knowledge of spina bifida, the physician's relationships with doctors who had performed her previous surgeries, and the proximity of the physician to her home. Ms. Hunt concluded that she has not experienced any issues related to spina bifida in a long time and is pleased with her health care so far.

Discussion

A participant asked Ms. Hunt what characteristics she looked for when choosing a PCP. Ms. Hunt recounted how she knew she had found the wrong physician when he or she had no knowledge of spina bifida. Ms. Hunt stated that the key characteristics of a potential PCP were knowledge of her type of spina bifida, knowledge of UTIs, the ability to manage an emergency situation, and proximity to her home in case of such an emergency.

A meeting attendee asked about the biggest barriers that Ms. Hunt encountered when transitioning into adulthood. The attendee wondered what Ms. Hunt's physicians could have done better to help her integrate into activities outside of medicine. Ms. Hunt replied that expressing hope and being positive were among the most important, along with informing her of future challenges while at the same time helping her find a workaround to those challenges.

An audience member asked Ms. Hunt how she identified clinics that would accept her. When she progressed from pediatrics to adult care, did a care coordinator help her, or did she receive assistance from her insurance company? Ms. Hunt stated that peers she met through the Spina Bifida Association recommended that she visit the clinics in Baltimore, Maryland, and Washington, DC. She also did much of the research on her own, especially when seeking care closer to her home.

A participant asked whether the multidisciplinary care Ms. Hunt received as a pediatric patient was necessary during her transition into adult care. The participant wondered whether it would suffice to instead have a "gatekeeper" who understands her situation and could send her to the right specialists. Ms. Hunt acknowledged the value in receiving preventative care by having all the doctors examine her once per year. At the same time, if a specific problem were to arise, she would rather simply address this problem.

Another participant wondered what skills and abilities Ms. Hunt would suggest that a young person in transition develop. What advice would she give to him or her? Ms. Hunt emphasized the value in being independent, particularly in wanting to be independent and capable of taking care of oneself. She suggested that the clinics are a good place to begin interacting with people and social groups.

An attendee inquired about Ms. Hunt's understanding of the state of her kidneys. Ms. Hunt responded that the extent of her knowledge is that there is a risk of kidney infections through UTIs.

A participant, in considering how to connect health care to quality of life, asked Ms. Hunt how her condition could be made less of a bother in everyday life. The participant shared how his patients often voice concerns that their condition interferes with going to school, finding a job, social relationships, and so forth. For example, some children encounter conflict when a school nurse tells them to use a catheter in a particular way, though they have been instructed otherwise. In other cases, children learn a tremendous amount from a school nurse. Ms. Hunt replied that she has not had, or does not remember having, any of these kinds of experiences. She knew that she needed to use a catheter every 3 hours like clockwork, but she does not recall any related conflicts.

An audience member asked whether Ms. Hunt was ever concerned about going to a children's hospital as an adult. Ms. Hunt responded that she was not concerned at all. She was merely glad that she had found the clinic, since her parents had never before been able to take her to a central location for her health care. She added that there were other adult patients visiting the clinic, as well as the pediatric patients.

A meeting participant recognized that large multidisciplinary clinics are not always conveniently located for many patients. Some of his patients need to make a 2- to 3-day trip in order to visit the clinic. What can specialists do to help patients or assist PCPs with delivering health care more locally? Ms. Hunt stated that more doctors are needed who are familiar with spina bifida. She clarified that her PCP refers her to specialists as the need arises.

In response to a question, Ms. Hunt remarked that she felt ready to transition from pediatrics to the adult world when she moved out of her parents' home at age 23. At that point, she was forced not only to do things on her own, but also to figure out how to do them. She mentioned that mentally, she may have been ready earlier, but that the transition itself did not happen for her until she began living on her own.

A participant inquired as to whether Ms. Hunt carries around information about her health or medical care. Ms. Hunt replied that she does have x-rays, and that she has given her current physician reports from her previous spina bifida clinic.

Meezan Ford

Ms. Meezan Ford expressed gratitude for being invited to the meeting and thanked the participants for the opportunity to

share her story. She emphasized that the transition from pediatric to adult care is important and said that the participants' desire to learn more fills her with hope.

Ms. Ford was born in 1973 with spina bifida myelomeningocele. She received pediatric medical care at Children's Hospital in Washington, DC, nearly an hour away from where she lived at the time. She says the distance was worth it, as she and her parents knew she was receiving great care. However, Ms. Ford did not see her doctors through the hospital's Spina Bifida Clinic. Instead, she saw them individually on different days. She recalls having a PCP, an orthopedic surgeon, a neurosurgeon, and a urologist. Although she says it is difficult not to associate her childhood with countless surgeries and hospital stays, she realizes that the surgeries, the doctors who performed them, and the nurses who aided in her recoveries helped her get to where she is.

When Ms. Ford was 1 year old, she underwent a urinary diversion. She experienced many UTIs until 1991, when she had surgery to tighten her stoma. At that point Ms. Ford started using a catheter. She has not been diagnosed with a UTI since then.

Moving to adult care was difficult for Ms. Ford because she was very happy with the doctors at Children's Hospital. She described the feeling to be like moving to a new school and having all new teachers. She remembers Children's Hospital as being a very colorful and happy place and the doctors and nurses being very caring. Ms. Ford was nervous about the change, but she remarked that her father—who is a physician—assured her that she would be fine with new doctors. Ms. Ford switched to Howard University Hospital, where she saw a PCP and a urologist who both took great care of her.

Ms. Ford moved out of her parents' home in 1998 and began seeing doctors in Howard County, Maryland, close to where she lived. She currently sees a PCP every 3 months, a podiatrist every 9 weeks, a cardiologist once per year (unrelated to her spina bifida), and a urologist once per year. Ms. Ford's PCP performs routine bloodwork every 3 months. Because her baseline white blood cell count is normally high, a high count will not be treated as an infection unless coupled with symptoms. As a preventative measure, her urologist has advised her to take cranberry supplements twice per day.

While Ms. Ford is very happy with the care she is receiving, she would love to see an increasing number of doctors become more familiar with spina bifida, specifically for adults. With spina bifida patients now living longer, Ms. Ford considers this a necessity.

Discussion

A meeting participant noted that both Ms. Hunt and Ms. Ford described seeking a balance between meeting people with knowledge about their specific conditions and local accessibility to such people. The participant mentioned that his patients often worry about what to do and whom to talk to if a problem arises at, for example, 5:05 p.m. on a Friday night. He asked the women if this has happened to them or those they know and, if so, how this has been managed. Ms. Hunt replied that she has not experienced an urgent matter relating to spina bifida in a very long time. As an asthmatic, she visits an urgent care facility if necessary. Ms. Ford agreed and said that the last time she needed to go to the emergency room for care related to spina bifida was when she was a teenager. Most of the health issues that she must deal with—such as high cholesterol, diabetes, and tachycardia—are not related to spina bifida.

An attendee asked what the top health worries or concerns are of adults with spina bifida and whether any specific preventative measures might be useful. Ms. Ford requested that health care providers be familiar with shunts, including being able to identify problems with the shunt. She also mentioned that UTIs are common among people she knows who have spina bifida.

State of Transition Care for Genitourinary Patients in the United States

Hadley Wood, M.D., FACS, Cleveland Clinic, Cleveland, OH

Dr. Hadley Wood provided an overview of the current state of transition care. She began by sharing her excitement in seeing at the meeting many people from her past who have shared the vision of transforming care of transition patients.

She thanked Dr. Star, Dr. Moxey-Mims, Ms. Norton, the NIDDK team, and especially Dr. Bavendam, noting her energy, enthusiasm, and ability to organize the meeting.

Dr. Wood pointed out that many patients with urologic conditions walk and look like otherwise healthy people. She showed a slide of two of her patients with bladder exstrophy, emphasizing that they, like any American couple, are striving to build their dream together.

When thinking about transition care, Dr. Wood categorizes conditions into three groups: (1) primary urologic conditions (e.g., bladder exstrophy, hypospadias, disorders of sexual differentiation, cloacal anomalies, valve disorders, vesicoureteral reflux, ureteral anomalies), (2) primary neurological conditions (e.g., spina bifida, cerebral palsy, mitochondrial disorders, neuromuscular diseases), and (3) other conditions (e.g., nephrologic conditions, renal transplant, pediatric cancer survivors).

Dr. Wood defined “transition” as a process for ensuring that high-quality, developmentally appropriate health care services are available in an uninterrupted manner as a patient moves from childhood to adulthood. She highlighted the two key elements of this definition: that care is developmentally appropriate and that it is uninterrupted.

Around 4.4 million children in the United States are in the age range for transition (12–18 years), but multiple studies have shown that less than half (about 40%) meet the national standard for transition. Dr. Wood noted that patients without transition care are more likely to be admitted to hospitals, to have more costly admissions, and to visit the emergency room for needs that could have been handled in an outpatient setting. The reality many providers face is that patients disappear into adulthood and later reappear in an emergency room having developed conditions that are often fatal. Dr. Wood highlighted a submitted abstract from researchers at Duke University suggesting that about one-third of all emergency room visits for patients with spina bifida are related to urologic issues.

The transition process can be divided into two main phases: preparation and handoff. Dr. Wood explained that during childhood, the parent and pediatric provider tend to drive decision-making. As the child moves into adolescence, he or she has more of a role in decision-making. Ideally, the adult health care provider is also brought in during this phase to help in or share the decision-making. The goal is that by young adulthood the patient is making decisions with the provider, with the parent serving as a consultant.

The preparation phase includes psychological and emotional preparation, health education, self-care, and a focus on functional independence and shared goal-setting. It may require social work and nursing expertise, the need to interface with the child’s school, and the development of readiness tools. The patient’s urologist and nephrologist are also involved, but the majority of the responsibility falls onto the parent and medical support structure surrounding the patient.

Dr. Wood suggested that urologists and nephrologists could assume a more direct role during the handoff phase. She was reminded of Ms. Meezan Ford, who described a happy and colorful children’s hospital in comparison with a less colorful, scarier adult hospital. Dr. Wood acknowledged that this transition, including needing to visit doctors in various buildings within a medical center, is frightening for many patients. Those with mobility limitations are especially affected. Geography, too, is an obstacle for patients who do not live near a major medical center, where most providers who specialize in care of patients with chronic conditions are found. One way this could be overcome is with increased access to electronic medical records.

Dr. Wood urged providers to seek to overcome other obstacles. This might include forming contracting agreements with children’s hospitals, considering appropriate reimbursement schema, or accepting that medical infrastructure is an important variable in the ability to provide appropriate care. Sufficient knowledge and experience remains one of the greatest challenges in urology. The American Board of Urology does not have any education requirements for residents, and no formal fellowship programs exist in transition or adult congenitalism.

Dr. Wood described two examples of models of transition care: one at Riley Hospital for Children in Indianapolis and the other at the University of Oklahoma. Under the model at Riley Hospital for Children, 40 percent of pediatric patients make it from transition to the adult clinic within 2 years. At the University of Oklahoma, patients are rated based on their readiness for transition (i.e., T1–T4). As they progress through the stages, they become increasingly prepared to

handle decision-making.

Dr. Wood claimed that urologic transition care in the United States is currently in its embryonic stage. Urologists now must critically examine the barriers to appropriate transition into adult life, think about the needs of patients and parents, and study the different models of care that have already been implemented. The needs of providers also are important, particularly as they relate to adult support services and provider education. Urologists should develop guidelines to help PCPs and regional providers, who are closer to the patients, provide care. Both health outcomes and patient-reported outcomes are needed.

Physicians should also consider how to approach transition care at the levels of policy advocacy and interfacing with the American Board of Urology or Nephrology as well as society organizations.

Dr. Wood encouraged participants to read through the meeting abstracts and initiate conversations with their authors to generate a synergy of effort on transitional urology.

Historical Perspective of Transitional Urology

Dan Wood, Ph.D., FRCS, University College London Hospitals, London, UK

Mr. Dan Wood provided a historical perspective on urologic transition care. He began by thanking Dr. Bavendam and the team at NIDDK for supporting the meeting.

Mr. Wood acknowledged that the amount of history on transitional urology is limited, and it is not well understood how history has affected the current state of transition care. He revealed that the word “adolescence” is rooted in the Latin word *adolescere*, which means “to grow.” Transitional urology not only considers a group of patients who are growing, but also it is itself a growing specialty and a growing interest.

The opportunity exists to significantly improve care for transition patients. Mr. Wood stated that to succeed the urology community will need to harness some of the energy of its forbearers, particularly Sir David Innis Williams, Dr. Philip Ransley, and Dr. Christopher Woodhouse. Dr. Woodhouse is synonymous with adolescent urology and is credited with inventing the specialty. He did so by understanding that providing pediatric urologic care—or care of any type of major congenital anomaly—represents a contract of care. Patients are dependent for the remainder of their lives on the delivery of health care to ensure their survival, well-being, and satisfactory quality of life.

Dr. Woodhouse determined several years ago that 21 centers within Britain had some degree of an interest in adolescent urology. Unfortunately, in a recent survey assessing the need for transition care across pediatric urologists, one person wrote in their freehand comments that they do not understand the need for adolescent urology. Most respondents, however, did agree there is a need for a group of people with a special interest in adolescent urology. Mr. Wood described how pediatric patients and teams form very close bonds and an enormous amount of trust over 16–18 years, and that phasing patients into the adult care model can be very difficult.

Mr. Wood stated that geography can play a role in health care, recognizing that the proximity of University College Hospital to a tube station is an advantage it has over Great Ormand Street Hospital. The University College London Hospitals has an adolescent ward set up so that transition-age patients are not thrust into the adult world. Instead, they are able to familiarize themselves with the team and the adult care system before fully engaging. There has been some backing for transition care in the United Kingdom.

Mr. Wood explained that preparation (e.g., readiness questionnaires, carrying around health information) is important in helping improve delivery of care. A large problem remains, however, in knowing what outcome measures need to be assessed. Moreover, inappropriate delivery or support of health care generates conflict, a loss of medical adherence, and a lack of engagement. People who lose engagement with health care tend to experience medical emergencies. Some researchers have developed suggestions for how to deliver transition service. For instance, one group has proposed the following six core elements of health care transition: transition policy, transition tracking and monitoring, transition readiness, transition planning, transfer of care, and transfer completion.

Mr. Wood discussed how urologic patients aspire to have a normal quality of life, including becoming pregnant and having children. He stressed the importance in providing care for patients with these needs, as infant death rates can be affected quite markedly. Patients who attempt to deliver babies outside an environment where appropriate urologic support is available often require an emergency caesarean section with a catastrophic outcome.

Mr. Wood observed that the United Kingdom trains pediatric urologists, adolescent urologists, and adult urologists, but he maintained that this care needs to become one continuous phase. Mr. Wood acknowledged that models of care appropriate in the United States would not necessarily be appropriate for the United Kingdom. He expressed hope in Europe's establishment of the Congenital Lifelong Urology Working Group, a multidisciplinary group that is beginning to hold meetings to build a framework across Europe. Mr. Wood hopes that the working groups in the United States and Europe will be able to inform each other and share ideas.

Mr. Wood concluded by remarking that urologic care merely 50 or 70 years ago emphasized patient survival. Now urologists are able to focus specifically on continence and renal preservation, safe follow-up, and achieving a satisfactory quality of life for patients. An enormous amount of progress has been made in congenital lifelong urologic care over a relatively short period of time.

Engaging the Unengageable: Male Adolescent Health

David Bell, M.D., M.P.H., Columbia University Medical Center, New York, NY

Dr. David Bell shared his perspective as the medical director of the Young Men's Clinic, which is part of the Family Planning Clinic. He hoped to communicate to attendees the health concerns of young men without congenital urologic conditions, with the idea that for patients with congenital urologic conditions these concerns are likely to be augmented many times. He admitted that he had not thought of himself as working in the arena of transition care and thanked Mr. Dan Wood for inviting him to speak.

Dr. Bell explained that one of his main responsibilities has been to inspire young men to continue seeking health care. After completing his undergraduate and medical education in Texas and his medical training in New York City, he relocated to California for a fellowship in adolescent medicine. While there, a longtime nurse recognized that Dr. Bell, of all of the fellows at the clinic, had the largest number of young men arriving to his clinical hours on a daily basis. The nurse's observation compelled Dr. Bell to think about how best to engage adolescent and young adult males in health care services. He began investigating the current state of adolescent male services in the United States.

Three questions Dr. Bell hoped to broach in his presentation are (1) Can we engage adolescent and young adult males? (2) What are their concerns? and (3) Where do we go from here? The literature has shown that health care visits by young adult males decrease after age 15, whereas in young adult females they increase (primarily to obstetricians and gynecologists). Though society often blames males for not seeking health care and attributes the drop to concerns about masculinity, Dr. Bell maintains that systematic factors may influence this trend. First, male adolescents are poorly informed about where to access reproductive and sexual health services. Second, the insurance status of males often shifts during young adulthood. Finally, health care visits servicing young adult males typically do not require scheduling follow-up visits, as they often do for females.

Dr. Bell described his experience serving in the role of medical director of the Young Men's Clinic for the last 15 years and as medical director of the Family Planning Clinic for the last 4 years. The Family Planning Clinic sees 130–150 women per day and 20–60 males per day. In 1999, it operated on a 4-hour week (Monday nights only) and provided assistance to 740 unique patients per year. Currently, it operates 5 days per week and provides assistance to 5,400 unique patients per year, with around 6,800 visits per year. Insurance coverage has increased from around 23 percent to 30 percent since implementation of the Affordable Care Act. Dr. Bell also noted that the Young Men's Clinic is truly a transitional clinic, serving men ages 14–35 years. He has repeatedly needed to increase the upper age limit over time, as the older men who typically continue to not have insurance do not know where to access services.

Dr. Bell reported that the most common first question he is asked by patients at the Young Men's Clinic is, Am I normal? Young men are concerned about the appearance and function of their body and penis, citing premature

ejaculation, erectile dysfunction, and fertility as concerns. Dr. Bell has been particularly struck by the number of men concerned about fertility. For example, he explained that many men become worried when they engage in unprotected sex that does not result in pregnancy. Patients also wonder about relating intimately to a partner. Will my partner be accepting of my body?

Dr. Bell stressed that adolescent young adult males are willing to discuss topics surrounding reproductive and sexual health, but that they prefer to have clinicians begin the discussion. He suggested clinicians create a space for them to feel comfortable and that they initiate the conversation.

Dr. Bell noted that inherent in the culture of his profession is engaging with patients. This could take the form of wearing relevant, attention-grabbing neckties, for instance. He emphasized that the Young Men's Clinic strives to ease each patient's burden, allowing him to seamlessly progress from one stage of life to the next. He contrasted this with adult medical care, which often centers on instructing patients how to care for themselves and blaming the patients if they do not follow instructions.

Dr. Bell shared the story of a young man with sickle cell disease. The patient could no longer visit his pediatric hematologist because his mother had developed a mental illness and the system would not allow him to be seen without his parents. A nurse practitioner in the pediatric hematology clinic reached out to Dr. Bell and asked if he could support the patient with the hematology clinic's assistance. Dr. Bell agreed, and the patient was assigned to an adult hematologist who had a 4-hour clinic on Wednesdays only. However, the patient often missed his scheduled appointments due to his chaotic life, which meant he needed to wait a few weeks until another opening was available. In the interim, the Young Men's Clinic provided supplementary services, such as medication refills, which kept the patient out of the emergency room. Dr. Bell emphasized that flexibility within the pediatric and adult systems of care are important to work through these types of difficult situations.

Dr. Bell shared another story about a longtime patient with surgical correction of hypospadias who was arrested for striking a police officer. While preparing for incarceration he and his mother visited Dr. Bell to ask for help on how to care for his urologic condition (including his pump) while in prison. They also requested that Dr. Bell write letters to inform the warden and judge of his condition. Dr. Bell worked on these issues with the man, who is now pursuing a successful career and is engaged to be married.

The ultimate goal is for every individual to be comfortable with him- or herself and to live a healthy life. Dr. Bell concluded by assuring the audience that young men can be engaged, that most of their concerns are related to normality and intimacy, and that the need is to determine and document the specific concerns of young men with congenital urologic conditions.

Intellectual Disability/Developmental Disability and Transition

Nienke Dosa, M.D., M.P.H., SUNY Upstate Medical University, Syracuse, NY

Dr. Nienke Dosa, a developmental pediatrician, provided her perspective on transition care of patients with intellectual and developmental disabilities. She works at the Center for Health Care Transition Improvement (Got Transition) and also contributes to Healthy Transitions New York, which focuses specifically on health care transition for young adults with developmental disabilities. Dr. Dosa runs the regional spina bifida clinic in Syracuse, New York, and is on the Professional Advisory Council for the Spina Bifida Association.

Dr. Dosa began by stating that physicians see a decline in both health (e.g., renal transplant loss, sickle cell disease complications, increasing obesity) and health care (e.g., worsened adherence, decreased follow-up, lapses in insurance coverage) during the transition to young adulthood. Discussions about quality improvement often focus on the 500,000 youth with special needs transitioning each year. However, especially at a national level, these discussions must keep in mind the estimated 18 million adolescents ages 18–21 years, about a quarter of whom have chronic conditions, who are transitioning from pediatric to adult care. Millions more could be included in this number when considering those in the age range of 12–26 years, which is when transition happens. Dr. Dosa reminded the audience that transition is a process, not an event.

The majority of youth are ill-prepared for transition. Surveys of health care providers consistently show that they lack a systematic way to provide transition support. An important clinical report published in Pediatrics in 2011 targets all youth, integrates transition into the medical home (thereby addressing the role of the PCP and team), extends to adult health care providers, and provides guidance for financial support of health care transition. It is very useful because it is a practical algorithm for how to better plan and implement health care transitions, provides branching for youth with special health care needs, applies to both primary and specialty practices, and provides a structure for thinking about education and research around this topic.

The National Center for Health Care Transition Improvement, funded by the Maternal and Child Health Bureau, is a national resource that is responsible for developing transition tools that are aligned with the clinical report. Dr. Dosa encouraged participants to remember the website www.GotTransition.org. She stressed that it is an excellent resource with a large amount of ready-to-use materials. The site contains portals for health care providers, youth, and families, as well as for researchers and policymakers.

The www.GotTransition.org website is organized around the six core elements of transition: (1) transition policy (recommended at ages 12–14), (2) transition tracking and monitoring (ages 14–18), (3) transition readiness (ages 14–18), (4) transition planning (ages 14–18), (5) transfer of care (ages 18–21), and (6) transfer completion (3–6 months after transfer). Dr. Dosa explained that transition policy refers to discussions about the age at which transition will occur. Transition readiness involves assessing patient skills. Transition planning includes writing a concrete plan with endpoints, specific needs, and specific dates, and collecting key information for the transition process, including baseline physical exam findings and instructions for patient-specific emergency scenarios. The website also has quality improvement measures that a medical practice can use to assess its progress on implementing the six core elements.

Dr. Dosa shared that transition readiness at the spina bifida clinic in Syracuse includes using an SBA resource called Preparations, which is an interactive web tool that patients can use to assess self-care skills. The clinic also evaluates navigation skills, such as calling and scheduling an appointment, as well as decision-making.

Dr. Dosa detailed the contents of her clinic's transfer packet. The packet can include a checklist of concrete skills the patient has acquired, self-assessment tool reports the patient has printed out, and materials that adult providers might find useful. The transition information form contains a transfer letter with date of transfer, a transition readiness assessment, a plan of care with transition goals, an updated medical summary, and emergency care plans. Other forms and documents may include guardianship or health care proxy documents, fact sheets, and additional provider records.

The transfer packet also includes documentation of any learning disability. Dr. Dosa emphasized that such documentation is very important for accessing rehabilitation services. When patients transfer out of education-based rehabilitation services, the documents need to be distributed to other state organizations, and it can be hard to navigate the paperwork involved. Another service the clinic provides is letters requesting disability support services, such as accommodations that would be helpful in college dormitory rooms. The clinic has also begun producing video introductions for adult providers.

Dr. Dosa transitioned into a discussion about defining intellectual and developmental disabilities. An “intellectual disability” (ID) is characterized by scoring two standard deviations below the mean score on IQ tests and adaptive behavior tests. Intellectual disability includes deficits in concept skills, social skills, and/or practical skills, and it originates before the age of 18. “Developmental disabilities” (DD) is an umbrella term that includes intellectual disability and also other childhood-onset chronic disabilities that can be cognitive, physical, or both. Onset occurs before the age of 22.

Patients with spina bifida, though they have typical IQ scores, commonly suffer from a learning disability called executive dysfunction. Executive functions work to help a person achieve goals. They include managing time and attention, switching focus, planning and organizing, remembering details, curbing inappropriate speech or behavior, and integrating past experience with present action. Dr. Dosa explained that non-verbal learning disability is a more subtle disability. These patients are able to retrieve information from semantic memory (e.g., vocabulary), but they have difficulty with the construction or integration of information (e.g., math). Dr. Dosa cautioned physicians not to assume that spina bifida patients with typical IQ scores are able to make decisions. Conversely, physicians should not assume

that patients with low IQ scores are not able to make appropriate decisions.

Self-determination is a characteristic of a person that leads them to (1) make choices and decisions based on their own preferences and interests, (2) monitor and regulate their own actions, and (3) be goal-oriented and self-directing. Both the abilities of the person and the opportunities presented by the environment contribute to the degree of self-determination that can be expressed. Fostering self-determination will help improve the transition process.

Dr. Dosa shared that the prevalence of developmental disability in childhood is currently approaching 14 percent, mostly due to attention deficit activity disorder (ADHD) (6.7%). The prevalence of intellectual disability is 0.7 percent. Trends in prevalence of disability over the last 15 years show an increase in ADHD and learning disability and a dramatic increase in autism.

The prevalence of ID and DD in adulthood is difficult to determine, and this reflects the fragmented adult care system. A conservative estimate is between 1.5 and 2.5 percent. A recent compelling article on the front page of the New York Times reported that these patients are more likely to report fair or poor health, inadequate emotional support, greater risk of having four or five chronic health conditions, and significant medical care utilization disparities (e.g., access to dental care). One challenge is that the health care that people with ID receive requires not only the medical health care system, but also the rehabilitation services system, including assistive technologies and behavioral health specialists.

National organizations, including the American Association on Intellectual and Developmental Disabilities and The Arc, have made recommendations to develop standards of care for primary care teams. The recommendation include (1) making a special effort to recognize and foster self-determination as a human rights issue for these patients, (2) collaborating across systems of care (e.g., education, housing, employment, transportation), and (3) linking with community-based behavioral or mental health services. This will be critical with the influx of patients with autism now entering the adult health care system.

Dr. Dosa suggested increasing professional training in the field of adult ID/DD care and potentially creating a new fellowship program or board certification comparable to that for developmental pediatrics. Emphasis has been placed on researching the relationships between health status and quality of life, the systems of healthcare delivery and health status, and the benefits of health promotion on health status. Research should examine interventions at both the clinical treatment level and the policy developmental level.

Dr. Dosa described how the SBA has supported much work around transition care. This work includes a cluster of longitudinal research studies on transition; the Adolescent Self-Management and Independence Scale developed by Dr. Kathleen Sawin, who is the next presenter at the meeting; the Kennedy Independence Scales; iMobile Health and Rehabilitation (iMHere), a collection of innovative technologies developed by attendee Dr. Brad Dicianno and his group; Spina Bifida Preparations; SBA's educational programs SB University, Jon Krainak Program, and Journeys; updated Health Care Guidelines for the 2017 World Congress; and the National Spina Bifida Patient Registry.

Self-Management: Key to Successful Transition

Kathleen Sawin, Ph.D., CPNP-PC, FAAN, University of Wisconsin–Milwaukee and Children's Hospital of Wisconsin, Milwaukee, WI

Dr. Kathleen Sawin, a pediatric nurse practitioner and a nurse researcher, presented her work on assessing self-management during transition care. She stressed that transition requires behaviors and skills in many domains and that self-management is central to any transition.

Dr. Sawin began by addressing points raised by preceding presenters. As a researcher, she expressed her appreciation for qualitative methods as a way of listening to the unfiltered voices of adolescents and families. A study she performed has shown that when transition is based on best practices, individuals and families have a very positive experience. Nearly all of the parents and young adults indicated that the difference they noticed the most when going to the adult setting is that the patients were treated like adults.

Dr. Sawin stated that failure to establish or maintain effective self-management behavior is a pervasive, and costly, public health problem. She defined self-management as a process by which individuals and families use knowledge and beliefs, self-regulation skills and abilities, and social facilitations to establish self-management behaviors and achieve health-related outcomes. Self-management takes place in the context of risk and protective factors specific to the condition, physical and social environment, and individual and family factors.

Dr. Sawin is involved primarily in the measurement and theory of self-management, though she will focus her presentation on theory. She believes theory is important as a pragmatic tool because it makes her a smarter, more articulate, and more effective clinician. Dr. Sawin claimed that use of a theory results in better outcomes for individuals and enhances communication. Furthermore, testing of theories facilitates research and clinical excellence. One such theory developed from a synthesis of the literature is the Individual and Family Self-Management Theory (IFSMT).

Dr. Sawin underscored that only 10 percent of health behavior is influenced by health professionals and that over one-half of written prescriptions go unfilled. She emphasized working collaboratively with families and individuals to establish self-management behaviors that will lead to positive health outcomes.

IFSMT is a person-centered model. The concepts of adherence (i.e., following instructions given by a health care professional), alliance, and compliance are perceived as contrary to self-management, as they dismiss the notion that the primary responsibility and control lies with the individual and family. In the IFSMT model, the health care professional is very important in helping to develop skills and abilities, but he or she is not the driver of self-management. The assumptions of the model include a willingness on the patient's part to assume responsibility. This personal interest and control results in better outcomes than does following external directions. The goal for personal engagement is activity, not perfection. The exception is in a very narrow medical situation in which there is a very narrow range of behaviors needed to survive.

Self-management involves dynamic, interactive processes that require time, repetition, and reflection. Executive functioning, a major issue for individuals with spina bifida, is an important component of self-management. Person- or family-centered interventions are the most effective in fostering engagement in self-management behaviors and achieving outcomes. Self-management behaviors can be influenced by preferences, culture, social norms, and family.

The IFSMT is a context-process-outcome theory, with a focus on process. The context includes the condition (i.e., its complexity, trajectory, treatment, and transitions), the physical and social environment, and individual and family factors. The process has three components: knowledge and beliefs, self-regulation skills and abilities, and social facilitation.

Dr. Sawin emphasized that years of research in a variety of behaviors have shown that knowledge is not related to behavior change. Self-efficacy, the confidence to participate in a behavior, however, is related to behavior change. Interestingly, knowledge is necessary, but not sufficient, for self-efficacy. Providers must help individuals make the connection between self-efficacy and behavior.

Dr. Sawin recognized the existence of barriers to self-management, for example, when two goals are not in congruence. Dr. Sawin offered the example of women with osteoporosis. They often strive to eat foods that facilitate osteoporosis health and also have a low-calorie intake. The foods that help with osteoporosis, however, are often high-calorie. Unless these two competing goals can be addressed, the behavior will not change. Dr. Sawin stressed the importance of implementing an action plan.

Health care professionals are facilitators. They can direct, encourage, and support engagement in self-management behaviors. Many providers assume that if a person is engaged in activities and treatment regimens that are appropriate and effective, then he or she will achieve a better health care outcome. It is not known, however, whether this is the case.

Dr. Sawin shared as an example a recent study of adolescents with diabetes, whose involvement in self-management behaviors using a valid and reliable tool has a very small relationship to the results of their A1C blood glucose test. She suggests that hormonal changes or other physiological markers may be playing a role, but additional testing is needed.

Dr. Sawin highlighted that the IFSMT3 model comes from clinical practice and emerged from clinicians, and that clinicians and researchers can intervene at both the context and process levels. Dr. Sawin noted that if a young person has depression, the ability to intervene on process skills is limited. Dr. Sawin concluded that the IFSMT model offers a different way of thinking about self-management, and she urged the participants to consider its person- and family-centered approach.

BREAKOUT SESSION 1: CONDITION ORIENTATION

The goal of Breakout Session 1 was to brainstorm research needs according to urologic condition. Breakout groups (listed below) were organized with the intent to cover many aspects of transition care. Identified research needs for each group are included in the Appendix. The ideas were then sorted according to research domain and used to pre-populate spreadsheets for use in Breakout Session 2

Spina Bifida: Reproductive Health Group

Moderators: Melissa Kaufman, M.D., Ph.D., Vanderbilt Medical Center, Nashville, TN; Naida Kalloo, M.D., Children's National Medical Center, Washington, DC

Spina Bifida: Nonurologic Contributors to Transition Group

Moderators: Brad Dicianno, M.D., University of Pittsburgh Medical Center and VA Pittsburgh Healthcare Center, Pittsburgh, PA; Peter Scal, M.D., M.P.H., University of Minnesota, Minneapolis, MN

Neuropathic Bladder and Exstrophy: Lower Urinary Tract and Bowel Dysfunction Group

Moderators: Rosalia Misseri, M.D., Indiana University School of Medicine, Indianapolis, IN; Dan Wood, Ph.D., FRCS, University College London Hospitals, London, UK

Neuropathic Bladder and Exstrophy: Nephrologic/Metabolic Issues Group

Moderators: Tej Mattoo, M.D., DCH, FRCP, FAAP, Children's Hospital of Michigan/Wayne State University School of Medicine; Clare Close, M.D., Close Pediatric Urology, Henderson, NV

Male Genital Issues and Reproductive Health Group

Moderators: Hadley Wood, M.D., FACS, Cleveland Clinic, Cleveland, OH; Michael Hsieh, M.D., Children's National Health Center System and the George Washington University, Washington, DC

Female Genital Issues and Reproductive Health Group

Moderators: Veronica Gomez-Lobo, M.D., Medstar Washington Hospital Center/Children's National Medical Center, Washington, DC; E. James Wright, M.D., Johns Hopkins University, Baltimore, MD

BREAKOUT SESSION 2: RESEARCH DOMAIN ORIENTATION

The goal of Breakout Session 2 was to prioritize research needs according to research domain. Breakout groups consolidated redundancies/overlap in the research needs generated by Breakout Session 1. Ideas were fine-tuned into concrete research needs and questions. Afterward, a representative from each group shared the "high-priority" research needs identified by his or her group. Collectively, meeting participants would attempt to organize these into a priority list and establish a research agenda.

Epidemiology Group

Moderators: Michael Hsieh, M.D., Children's National Health Center System and the George Washington University, Washington, DC; Enrique Schisterman, Ph.D., Eunice Kennedy Shriver National Institute of Child Health and Human Development, NIH, Bethesda, MD

The Epidemiology Group was tasked with prioritizing epidemiological research needs related to transitional urology from both cross-sectional and longitudinal perspectives. Dr. Michael Hsieh reported that the group reviewed 36 needs collected during the first breakout sessions. In some cases, research needs were modified to enhance their relevance.

The Epidemiology Group identified eight high-priority research needs:

1. Rates and epidemiology of adult sexual dysfunction in spina bifida and other urologic diagnoses affecting sexuality. What is the basis for normal? What are the expectations of patients and parents? Longitudinal body image and sexual function assessment controls are needed for comparison with spina bifida. Questionnaire-based studies in adult clinics could be used.
2. Pregnancy outcomes in patients with spina bifida and other relevant urologic diagnoses. Fertility rates for males and females, including outcomes for the large-scale spina bifida population. Semen analysis and hormonal panels could be performed. If feasibility is a concern, testing could be scaled down.
3. Support networks for patients, including predictors of good outcome, the effect of health care disparities on outcomes, provider factors, and patient factors. What are the underlying mechanisms that contribute to issues of self-management (i.e., biologic, environment)? How does the influence of parents, grandparents, friends, and peers with the same diagnosis (or not) affect outcome?
4. Addition of adults to the current spina bifida registry.
5. Bladder infections, including diagnoses, antibiotic resistance, asymptomatic bacteriuria, impact of age, and bladder and bowel dysfunction (BBD). Bladder colonization. The potential for pyelonephritis, renal insufficiency, cancer risk, and stone formation.
6. Acute kidney injury (AKI), including medications and dehydration. Epidemiology of acute and chronic kidney disease. Diagnostics to identify risk factors, including the importance of bladder pressure, imaging, and BBD.
7. Epidemiology and surveillance of carcinoma, including bladder cancer following augmentation cystoplasty, gastrocystoplasty, exstrophy, UTIs and associated bladder cancers, and catheter-associated cancers.
8. Genetics and heritability. What genes interact with which environmental factors to cause certain diagnoses? Better prognostic tools are needed to predict genetic risk of male genital issues.

Discussion

No discussion points were raised.

Outcome Measures Group

Moderators: Rosalia Misseri, M.D., Indiana University School of Medicine, Indianapolis, IN; Tamara Bavendam, M.D., M.S., NIDDK, NIH, Bethesda, MD

The Outcome Measures Group considered research needs related to outcome measures that currently exist and to those that are still needed. Dr. Rosalia Misseri reported the group's five high-priority research needs:

1. Renal and metabolic outcomes
 - a. Best method to monitor renal function (serum and imaging)
 - b. Definition of AKI. Best method to monitor for vesicoureteral reflux
2. Bladder outcomes
 - a. Define urinary incontinence/continence and bladder emptying outcomes appropriate for patients with neuropathic impairments.
 - b. What is the role of urodynamics and bladder pressures in bladder and renal outcomes?
 - c. What is acceptable to patients who are in the process of attaining independence?
 - d. Define bladder colonization, asymptomatic bacteriuria, and UTI requiring antibiotic treatment.
3. Bowel outcomes
 - a. Define fecal incontinence/continence and bowel emptying outcomes appropriate for patients with neuropathic impairments.
 - b. What is acceptable to patients who are in the process of attaining independence?
4. Sexual function outcomes
 - a. Sexual knowledge
 - b. Sexual functioning
 - c. Sexual behavior
5. Cognitive-behavioral-mental outcomes

- a. Anxiety
- b. Depression
- c. Executive function
- d. Adaptive function

Dr. Misseri elaborated on the last category, noting that cognitive-behavioral-mental outcomes are not typically thought about from a urologic perspective. For patients to transition successfully, however, physicians need to know how to measure these issues. The ability to do this quickly in a clinical setting would help busy clinicians understand what support their patients need.

Dr. Misseri added that medium-priority research needs include systemic issues and broadening the definition of independence and self-management:

1. Systemic concerns
 - a. Optimal bone health (bone density and structure)
 - b. Optimal assessment of pain.
 - c. Defining obesity
2. Broadened definitions of independence and interdependence
 - a. Assess family readiness to give up control
 - b. Assess patient readiness to assume control
3. An appropriate measure for adequate hormonal therapy from puberty to adulthood

Discussion

Dr. Star asked whether the definitions used to describe outcome measures are considered static or whether they are adjusted based on patient age. An attendee replied that the definitions are dynamic. For example, bladder function changes both as a person grows and with changes in neuropathy. Researchers still poorly understand why some people, and not others, develop a hostile bladder or experience renal insult. The attendee added that creatinine level (which changes with body mass) and bowel function measures (which are complicated by obesity and chronic stretching of the colon) are dynamic as well.

Dr. Star followed up by considering the preferences of patients, families, and caregivers at different life stages. Dr. Misseri stated that patient-reported outcomes have been integrated into the cognitive-behavioral-mental outcome measures category identified. The Outcome Measures Group had intended for the cognitive-behavioral-mental category to permeate the other four categories (i.e., renal and metabolic outcomes, bladder outcomes, bowel outcomes, and sexual function outcomes). She acknowledged that the needs of a patient with bladder or bowel issues at age 3 might be very different from their needs at age 10 or 20.

Dr. Bavendam added that the appropriate application of outcome measures, even one as seemingly straightforward as creatinine level, is not well understood.

Clinical Care Group

Moderators: E. James Wright, M.D., Johns Hopkins University, Baltimore, MD; Hadley Wood, M.D., FACS, Cleveland Clinic, Cleveland, OH; Clare Close, M.D., Close Pediatric Urology, Henderson, NV

The Clinical Care Group was tasked with determining the roles of health care professionals during transition care. The group attempted to distill the research needs collected during the first breakout sessions into four top priorities. Dr. E. James Wright reported the group's four major research needs, each consisting of several components:

1. UTIs/colonization
 - a. Definition
 - b. Patient education
 - c. Provider integration—assessments of clinicians outside of transitional arena, dissemination of information

- d. Development of clinical pathways—formulating an algorithm that could guide clinical practice
- e. Patient impact—economic burden, un/misdiagnosis, colonization vs. infection
2. Nephrologic care
 - a. Glomerular filtrate rate (GFR) estimation—guideline or algorithm for tracking/assessing upper-tract status/injury
 - b. Metabolic assessment
 - c. Role of AKI
3. Bladder/reservoir management into adulthood
 - a. Provider education
 - b. Patient education
 - c. Surgical intervention/timing
 - d. Surveillance of reconstructed patients—e.g., composite lower-tract reconstructions for valve diversions, developing an algorithm/process for screening into/through transition into adulthood
4. Sexual and reproductive health
 - a. Clinical screening (practices, abuse) —normative/descriptive data on sexual practices and rates of abuse
 - b. Patient and family education (sexually transmitted infections, genetics/contraception)
 - c. Divergent priorities (patient, provider, family)
 - d. Surgical intervention/timing
 - e. Psychological impact—assessment tools to influence the process in transition
 - f. Functional outcomes
 - g. Fertility impact—effects of choices made during transitional period

Discussion

Dr. Bavendam, after remarking that the four categories identified by the Clinical Care Group resemble those described by the Outcome Measures Group, asked the audience to reflect on the tools available for assessing sexual function in an adolescent population. Dr. Wright proposed that the meeting participants also consider the cultural and legal obstacles to even being able to ask such questions. Varying state and Institutional Review Board (IRB) regulations hinder the ability to ask adolescents questions about sexual health and reproductive issues. A particularly ripe area for inquiry is considering both how and what questions to ask adolescents.

A participant stated that non-urologists with specific areas of expertise need to be recruited as patients transition into the adult provider (or lack of provider) environment. The participant suggested that psychologists or other mental health professionals might be better equipped than urologists to assess sexual function in teenagers. He envisioned intercalating such professionals and areas of expertise into the development of research and solutions, even in the planning and priority-setting phases, since ultimately these individuals will be involved in the research and care of these patients. Dr. Bavendam agreed, noting that the meeting was composed of primarily urologists and gynecologists.

A meeting participant remarked that adequate questionnaires for assessing sexual function do exist, but they are intended for adults. Of major concern is the ability to ask these questions in a population of minors. Minors may legally ask and advocate for health services, but often they are not able to provide consent for a research study due to IRB regulations and group protocols. Parental consent may be required. Discussion ensued as participants tried to determine whether sexual and reproductive health confidentiality laws are consistent across states and IRBs. A consensus was reached that this varies by IRB. Dr. Bell added that there should be a way to galvanize around this issue and work with the various IRBs.

An audience member mentioned that the American College of Obstetricians and Gynecologists (ACOG) has issued a statement on research in adolescents. The statement allows a fair amount of research in this population due to ACOG's focus on reproductive health, which is protected. Many IRBs, however, do not share ACOG's views. The audience member would like to see other organizations issue their own statements. Dr. Bell responded that the Society for Adolescent Health and Medicine (SAHM), on whose Board of Directors he serves, also has issued statements around research in adolescents, especially concerning sexual and reproductive health issues. SAHM members are confident that they can work with the American Urological Association (AUA), SAHM, and ACOG to develop additional

recommendations. Dr. Bell offered to serve as a spokesman and initiate the discussions.

Dr. Sawin pointed out that many researchers have successfully retrieved sensitive material from adolescents by using computer-assisted interviews with earphones. She is aware, however, of at least two states with specific consent legislation that does not allow an adolescent to consent to a research study. In some cases, consent from her parents is required for a pregnant adolescent to enroll her own child in a study. Dr. Sawin expressed confidence that working together on this front could spur progress.

Self-Determination and Self-Management of Chronic Care Group

Moderators: Kathleen Sawin, Ph.D., CPNP-PC, FAAN, University of Wisconsin–Milwaukee and Children’s Hospital of Wisconsin, Milwaukee, WI; Nienke Dosa, M.D., M.P.H., SUNY Upstate Medical University, Syracuse, NY; Naida Kalloo, M.D., Children’s National Medical Center, Washington, DC

The Self-Determination and Self-Management of Chronic Care Group considered a number of issues around sexual function and reproduction, fertility, and the impact on self-management and long-term outcomes. Several categories focused on determining the specific urologic interventions that would be effective in optimizing self-management. Dr. Kathleen Sawin reported the group’s 10 high-priority research needs:

1. Psychosocial issues (e.g., coping, resilience), behavior issues, parental involvement
2. Self-management as it relates to body image, sexuality, and intimacy
3. Future fertility, including expectations and counseling
4. Changes in bladder management during transition
5. Baseline skills and/or socioeconomic factors important for transitioning into self-management, including critical time points for assessments and interventions in self-management
6. Studies of learning style/strategies of individuals with spina bifida
7. Relationship between wellness programs, self-management, and continence
8. Effect of surgical options on self-management, sexual function, reproduction and fertility, and cancer
9. Behavioral interventions effective in enhancing self-management and urologic outcomes
10. Research on mobile-device applications (“apps”) and other assistive technologies to improve self-management

Discussion

Dr. Bavendam asked the group whether tools for self-assessment already exist. Dr. Sawin described the Adolescent/Young Adult Self-Management Scale, a transition-readiness assessment of self-management behaviors that evaluates both condition management and independent living behaviors. She mentioned that additional tools for self-assessment might exist in the literature on intellectual disabilities. Others shared the view that this area needs development.

A meeting participant stated that he strongly supports the use of technology for the transition population. He believes that telemetry, home monitoring, and mobile monitoring of physiologic status through the use of mobile-device applications (“apps”) will be an expanding area of development in medicine in general, even in the coming 5 or 10 years. Dr. Sawin added that such technology will allow individualized interventions. A person’s behavioral patterns can trigger individualized responses that will either reinforce or correct the behaviors through knowledge or activities.

The participant also encouraged the community to educate parents and providers about self-determination and self-management issues before the patient is ready for self-management. He explained that this a general challenge in pediatrics. The participant recommended building in this type of education and awareness when patients are younger than age 10. An emphasis on self-management would allow clinicians to grant patients more independence earlier, which ultimately benefits patients over time. Dr. Sawin agreed, adding that her research has indicated that the focus is on the age of transfer, not on building transition skills prior to adolescence. She recalled that Got Transition provides materials for patients as young as 12 years old.

Health Care Infrastructure and Training Issues Group

Moderators: Brad Dicianno, M.D., University of Pittsburgh Medical Center and VA Pittsburgh Healthcare Center,

Pittsburgh, PA; Peter Scal, M.D., M.P.H., University of Minnesota, Minneapolis, MN; Melissa Kaufman, M.D., Ph.D., Vanderbilt Medical Center, Nashville, TN

Dr. Brad Dicianno reported that the Health Care Infrastructure and Training Issues Group was able to condense the first breakout session's output into four main themes:

1. Population of patients failing to transition. Who are the 60 percent of patients that are not shifting into adult care? Is this a failure of the current system of transition? What methods should be used to engage patients?
2. Education of primary care physicians. How does provider education change outcomes for patients? This can affect access to care for patients with disabilities. Novel strategies could include telemedicine, for example.
3. Innovative payment models. What innovative payment systems (e.g., reimbursement, coding) could improve care?
4. Comparative analysis of models of care. This includes transitional care models and different ways to coordinate care (e.g., multidisciplinary clinics).

Discussion

Dr. Hadley Wood added that the broad field of medicine, including incentives used and metrics followed, might look very different even in 5 years. It will be important to be able to better identify patient groups, their risks, and reasonable benchmarks for outcomes going forward.

CREATING A RESEARCH AGENDA

Dr. Bavendam challenged the meeting participants to prioritize the aforementioned research needs into a condensed list. Which needs, across all areas, have emerged as most important?

An audience member began the discussion by emphasizing that patients and other stakeholders need to be involved in the process of prioritizing needs. Another attendee stated that prioritization is difficult without understanding the future infrastructure. He noted that the community of health professionals managing transition care of cardiological patients is further along in its development than the community managing transition care of urologic patients. Will each medical field require its own group of transition physicians, or should the medical system develop and train transition physicians who will manage them all? These decisions will hinge on the model of care implemented. Large geographical areas, multiple facilities, and different needs must all be accommodated.

A meeting participant offered her perspective from working at the Transition Medicine Clinic, part of Baylor College of Medicine and Texas Children's Hospital, with Dr. Cynthia Peacock, who manages several transition initiatives. The Clinic has implemented a medical home model, which she described as being nearly equivalent to a special needs clinic for adults and teens in transition. The Clinic does not handle transition readiness. Instead, it specializes in transferring patients to adult care. The initial intent of the Clinic had been to develop transition-ready patients and to transfer them to physicians in their local communities. This would be to each patient's advantage, as he or she would receive care closer to home. Unfortunately, the Clinic was unable to identify physicians who felt equipped to undertake this type of care or to perform the complex care coordination that would be required. The Clinic now retains these patients.

The participant finds it valuable that the Clinic provides care for several different types of patients. Generally, the patients at the Clinic have been diagnosed with (1) severe autism or intellectual disabilities and thus are not appropriate for a typical internal medicine clinic for behavioral reasons; (2) spina bifida; (3) cerebral palsy with technology dependence; or (4) Down Syndrome. She finds being able to mentally compare these populations and learn from multiple collections of medical literature to be helpful.

An audience member stated that effective transition mechanisms exist in the cardiac surgery community, with surgeons often well integrated. He suggested studying models of care that have (and have not) succeeded. He then referred to a slide from Dr. Hadley Wood's presentation, which diagrammed two such models. Each model included three circles representing components of the transition team. In one model, the circles were disconnected from one another; in the other, they were overlapping. The participant suggested striving for a model in which the three circles are overlapping.

For context, Dr. Wood noted that cardiac surgery has a medical analogue in cardiology. She emphasized that cardiology has been the driver of transition care for cardiac surgery patients. In contrast, urology does not have a medical analogue. Urologists are both medical and surgical providers.

An attendee advocated that the community's most important research need is outcome measures. He pointed to the use of oxygen saturation as a reliable measure in cardiac transition. Until the urology community can be confident in good outcome measures, how can a transition be considered successful? The attendee suggested concurrently developing medical outcomes and patient-centered outcomes.

Mr. Dan Wood proposed defining a population of doctors or surgeons who will manage patients into adolescence and adult life. These dedicated physicians would understand the patient's diagnosis at birth and the treatments he or she has undergone throughout childhood. Standardizing care in this way would create a mechanism by which physicians can compare themselves, which he believes would generate the very best possible research outcomes in the long term. Mr. Wood recommended dividing outcomes into medical-surgical outcomes and patient-reported outcomes.

Dr. Sawin returned to an earlier discussion of self-management assessments. She pointed out that Dr. Konrad Szymanski, a meeting participant, has developed a quality-of-life measure for adolescents and adults with spina bifida. Dr. Sawin also noted that other, more general quality-of-life measures exist, although they likely do not capture the physical condition and urologic care components that are important to patients. However, these measures do capture mental and social health, as well as work-cognitive-school aspects. Dr. Sawin suggested that these existing quality-of-life and other appropriate measures of concepts, such as self-efficacy, peer relations, or depression, could be of value to the urology community. The measures are short, usable, and available (e.g., the PROMIS measures developed by the NIH).

Dr. Bavendam asked the audience to consider which research needs are most important for care delivery right now. A participant voiced a major concern about identifying who would take on the transition role. How will such care be realized both in large cities and in rural communities? He wondered whether this problem may require a new specialty of transition care.

Mr. Dan Wood reiterated that the two women with spina bifida who earlier shared their stories both sought a balance between access to physicians with knowledge and the availability of care close to home. He proposed a hub-and-spoke model as a way of striking this balance. Medical centers in big cities can deliver complex care, and community physicians or medical groups closer to home can deliver local care. In order for this model to succeed, however, Mr. Wood maintained that standardization is fundamental. A standardized system would ensure that local physicians are delivering adequate care, and it would guide them in deciding when to refer patients to a larger medical center. Mr. Wood recognized that developing standards will create much disagreement and that taking initial steps forward will require courage.

A participant described the United States as "the land of individual choice." He stated that this mentality can be difficult to overcome, as every clinician and individual patient is likely to want to make his or her own decisions. Thus, he agreed with Mr. Wood that standardization is essential. The participant suggested assembling a group of professionals to discuss appropriate models of care, adding that these models of care would be based on the needs and desires of different types of patients, not of providers. Only when the models are articulated can the evaluation of models take place. The participant asserted that models for young patients living in rural Minnesota will be different from those for patients in New York, who have easy access to specialized care.

Mr. Wood interjected a comment about how care for patients needs to be standardized, regardless of whether patients live in rural Minnesota or in New York. The delivery of care may differ, however, and some decisions may need to be shared with larger transition centers.

The participant responded in agreement that all patients should have the opportunity to choose their care. He reiterated that these models should be articulated and evaluated, since a 300-mile drive to a medical center carries its own set of risks. Patients and families should be making these choices based on evidence, not because of the model of care the doctors in their area have implemented. A thorough evaluation of models is needed, and patients and families should

have knowledge of the outcomes that each model generates.

Dr. Hadley Wood informed the audience that the AUA is sponsoring a working group on this topic. The working group will hold a meeting at the AUA national meeting in New Orleans, Louisiana, on Sunday, May 17, 2015, from 2:00 p.m. to 6:00 p.m. Everyone in attendance will receive an invitation to the working group meeting. Discussion will be primarily case-based.

An attendee strongly supported the need to develop clinical pathways. He stated that providers receiving patients in transition need to feel that they are adequately prepared to do so. Too often they lack a fundamental understanding of how to care for these patients. The attendee recognized that the clinical algorithms that will be developed are likely to be imperfect and will need to be adjusted. At the same time, a reasonable consensus on standards of care needs to be achieved soon, as the outcomes of the care provided to babies now will not be known for 20–30 years. The attendee insisted on the need to establish clinical pathways, reevaluate them frequently, and shift them as necessary.

At Dr. Bavendam's request, Ms. Judy Thibadeau from the Centers for Disease Control and Prevention (CDC) described two CDC relevant initiatives. The first initiative is a clinic-based National Spina Bifida Patient Registry. Nineteen clinics are currently participating, and nearly 5,000 patients, ages 0–70 years, have contributed data since 2009. The registry is now beginning to inform the CDC about clinical practices and outcomes, particularly those related to continence and skin breakdown. The second initiative is the successful development of a standardized protocol for newborns and young children with spina bifida. This protocol was agreed upon by a group of urologists, nephrologists, nurses, and epidemiologists just this year, though Ms. Thibadeau acknowledged that reaching a consensus did prove challenging. Baylor College of Medicine is the first of nine clinics to pass the IRB process. Soon physicians at Baylor will begin enrolling babies as they are born. They will care for them according to the protocol, making exceptions as the physician or caregivers deem necessary and documenting the reasons for the exceptions. Ms. Thibadeau emphasized that the ability to make individual decisions will be preserved and that the protocol will be modified over time. The goal of the program is to learn the most economic and effective way to care for and preserve babies' urologic and kidney function.

In response to several questions about the types of data captured by the registry, Ms. Thibadeau clarified that patients visiting the clinic are asked 47 questions annually. The questions, which are primarily medical and not psychosocial, examine demographics, interventions (e.g., surgical and bowel-bladder programs), and outcomes. Ms. Thibadeau explained that financial limitations restrict the CDC's ability to ask certain types of questions, for example about sexual health. Nonetheless, the CDC gathers information and measures that they believe clinics can collect reliably. The registry is currently in its second version and continues to undergo development.

An audience member who is part of the Disorders of Sex Development Translational Research Network (DSD-TRN) commented that care should be taken not to duplicate others' efforts. DSD-TRN is developing a registry that strives to improve multidisciplinary care. Toward this end, the registry has a large psychosocial evaluation component that seeks to answer questions about how best to help families create resilience and how best to provide care to these patients. Because the questions posed by the CDC and DSD-TRN registries may overlap, the audience member suggested the possibility of sharing information.

An attendee speculated about how to evaluate what fellow participants are suggesting. He asserted that agreement must first be reached on which outcomes to measure and how to measure these outcomes, citing examples related to renal function, UTIs, and clinical pathways. The attendee recommended considering objective, physician-reported outcomes, as well as patient-reported outcomes.

A program scientist for the Management of Myelomeningocele Study (MOMS) offered to share a follow-up about the study. Its goal was to compare in utero and postnatal repair of myelomeningocele, with patients randomized to one or the other procedure. The children are now ages 5–9 years. The primary outcome used is the Vineland Adaptive Behavior Scale. Secondary outcomes include cognition, motor function, brain morphology and function, urologic outcomes, spina bifida outcomes, quality-of-life outcomes, and maternal reproductive functions (of the mother who delivered the baby). All outcome measures are listed on clinicaltrials.gov. Though the study does not assess adult outcomes, it does systematically collect outcomes in a valuable population of pediatric patients.

A participant wondered about the value of surveying the population of providers who do (or do not) accept patients in transition. What do they fear about this population? The participant explained that understanding their attitudes and needs could help guide the outcomes that physicians emphasize. Knowing why these health care providers do not follow or value guidelines can help target education appropriately. Dr. Bavendam agreed and suggested that a professional organization may be interested in undertaking such a project.

Another attendee deemed models of payment to be a very high research priority. Many groups have been experimenting with various systems, including Medicaid, and have been learning about how the systems affect outcomes of cost and quality of care. The attendee suggested taking into account payment models when establishing clinical pathways and evaluating models of care. Dr. Bavendam responded by acknowledging that the issue of reimbursement for the time necessary to provide adequate care is a high priority in general discussions about how to improve care. Tested, cost-effective models of payment offer hope for tackling these issues.

Dr. Bavendam concluded the discussion by recognizing that pulling together the information gathered during the meeting will be challenging. She thanked everyone for attending and for being actively engaged. Dr. Bavendam extended her appreciation to the group facilitators and to all those involved in organizing the meeting.

MEETING SUMMARY

Marva Moxey-Mims, M.D., NIDDK, NIH, Bethesda, MD

Dr. Marva Moxey-Mims thanked the attendees for their participation in the meeting. She remarked that much work remains to be done and acknowledged that establishing a list of priorities is difficult at this point.

Dr. Moxey-Mims assured the participants that all suggestions will be considered as the NIDDK moves forward with a research agenda. She asked the participants, however, to try to understand the issues that the NIH can and cannot address. The NIH is cognizant that reimbursement structure is important, but this is not something over which the NIH has direct influence. Similarly, the NIH is unable to influence the system of education of patients or providers. She expressed hope that research outcomes will be able to influence these areas.

An overarching theme that emerged from the meeting is the need to establish adequate and useful outcome measures. Also important is developing and implementing a transition care system that is applicable across the spectrum of care—not only in big research hospitals, but also in rural settings. Collaboration is needed with other medical areas in which providers consider patient-reported outcomes, where much of the promise lies.

Dr. Moxey-Mims emphasized the need to push forward on this front to explore how best to provide care and support for transitional urology patients. She once again expressed her appreciation for the valuable input provided by the meeting participants.

APPENDIX

Breakout Session 1 groups brainstormed the following research needs:

Spina Bifida: Reproductive Health Group

- Questionnaire-based survey of urologists/gynecologists/urogynecologists regarding training for and treatment of sexual medicine needs of the spina bifida population.
- Health care cost analysis of using a health care coordinator for spina bifida patients (e.g., Will access to a care coordinator decrease costs that result from the overuse of emergency department/admissions? Will it encourage the use of telemedicine resources?).
- Assess existing knowledge around anatomy/physiology of sexual function and needs/expectations for sexual education of spina bifida patients and parents (e.g., Where do patients obtain information? Do parents seek this information? Who should deliver the information? When are patients and parents ready for sexual function

education as a part of transition?).

Define across age groups.

- Develop and test a multifocal program (based on patient and parental needs assessments) for the delivery of sexual education to spina bifida patients. The program should include standardized data for delivery to patients and parents and should outline who would initiate the conversation.
- Rates and epidemiology of adult sexual dysfunction in spina bifida (e.g., questionnaire-based studies in adult clinics).
- Fertility outcomes for a large-scale spina bifida population (e.g., determine fertility rates for males and females via semen analysis/hormonal panels).
- Pregnancy outcomes in the spina bifida population.
- Assessment of correlation of activities of daily living with sexual function and reproductive health among spina bifida patients. Resources at www.GotTransition.org may be helpful.
- Cross-sectional body image/sexual function assessment among controls for comparison to spina bifida (e.g., What is the basis for normal? What are the expectations of patients/parents?).
- Assess cultural influence on spina bifida sexual function.

Spina Bifida: Nonurologic Contributors to Transition Group

- What are the expected developmental milestones and determinants?
- What are the most cost-effective assessments for executive dysfunctions?
- What are the appropriate screening tools for executive functioning, depression/anxiety, and pain?
- Examine the trajectory of expected executive functioning development and its relationship to urogenital conditions.
- A cross-sectional study of youth and young adult urogenital functioning and executive functions.
- Examine the relationship between self-care/self-management and executive functioning skills.
- Is it possible to categorize executive functioning skills and impact health outcomes by applying interventions targeted to the executive function level?
- What risks and protective factors relating to family and the child/youth/adult contribute to optimal health?
- Examine family/caregiver ability to provide support to spina bifida patients. May require development of an assessment tool.
- Impact of the circle of support on adaptive functioning.
- Develop and test wellness programs that improve outcomes and prevent complications.
- What underlying mechanisms contribute to issues of self-management (i.e., biologic, environmental)?
- What are the critical time points for assessment and interventions in self-management?
- What interventions can improve executive functioning?
- Study of learning styles/strategies among individuals with spina bifida.
- The impact of co-occurring conditions on executive function/ability to assume self-management.
- Improved understanding of structural changes in the brain with respect to executive function of individuals with spina bifida. Is it modifiable?

Neuropathic Bladder and Exstrophy: Lower Urinary Tract and Bowel Dysfunction Group

- Define UTI, urinary incontinence, fecal incontinence, and fecal retention for use clinically and in research.
- Develop guidelines for the treatment of UTI: consistent definition, antibiotic use, use of alternatives to antibiotics.
- Does bladder management independently affect sexual function?
- Are certain reconstructive surgeries better suited to certain phases of life?
- What is the impact of urodynamics on management decisions and treatment outcomes?
- How are current bladder and UTI management strategies impacting the primary goal of protecting the kidneys?
- How can renal function be evaluated when body mass index may not be accurate in spina bifida (e.g., creatinine, cystatin C, urine protein)?
- Does the fear of bladder cancer impact the decision to consider bladder augmentation?
- Methods for monitoring an augmented bladder for cancer (e.g., cystoscopy, cytology, imaging). Are the

- guidelines for surveillance put forth by Dr. Douglas Husmann and colleagues sufficient?
- Evaluate the risks of bladder cancer in neuropathic bladder (e.g., Is it inherent to disease? Is it related to clean intermittent catheterization [CIC], chronic catheterization, or augmentation?).
 - Assess the effects of patient socioeconomic status on transition (e.g., Is transition affected by reimbursement? What is the impact of reimbursement and transportation challenges on selection and successful implementation of a bladder management option?).
 - Comparison of bladder and bowel outcomes in different transition models.
 - Propose a standard of care with recommended follow-up and evaluate it prospectively. Can certain recommendations be made to establish a “temporary” standard of care, as a reference for future measurement?
 - When should children be transitioned to assume care for bladder emptying (CIC)? How should readiness be assessed?
 - Do different reimbursement resources affect how providers treat patients?
 - What are the bladder and bowel outcomes when parents are no longer able to provide supportive care or when the patient assumes responsibility?
 - Can the CDC’s National Spina Bifida Patient Registry be expanded to include more adults and other types of congenital neurogenic bladders (even all conditions that require lifelong urologic care)?
 - Assess disparities in health care versus outcomes (e.g., impact of socioeconomic status, provider factors, patient factors).
 - What are the differences between transition care in academic versus private practices, or small versus large urology group practices?
 - Does familial environment affect the choice of a management plan for bladder and bowels?
 - Assess the factors contributing to transition failure among those who fail to transition (e.g., Does failure to transition reflect a failure of the current system of transition? Are these patients transitioning for other health care needs, but not genitourinary conditions? Do overall health and bladder/bowel and reproductive health differ in those who transition versus those who do not?).
 - Define and test strategies to entice adolescents to engage in their health care.
 - Develop a conceptual model incorporating all influencers on clinical outcomes to guide research efforts on successful transition.
 - Develop and test telemedicine and mobile-based applications to facilitate engagement and care for the “Millennial” generation.
 - Evaluate the impact of reimbursement for telemedicine on transition and outcomes.
 - Evaluate long-term data on Botox injections in children (e.g., outcomes versus changes in management). Note: Botox is not approved for use in children. Data will take too long to collect.
 - Identify a durable solution for the paucity of adult providers (e.g., Is the creation of an adult–pediatric reconstructive specialist necessary for successful transition care with improved health and economic outcomes?).
 - Who should manage stomas and related procedures/devices (e.g., Malone antegrade enema [MACE]/caecostomy button, Chait tube) among adult patients?
 - Are outcomes affected by primary care providers’ education regarding genitourinary issues that need to be addressed during and after transition?
 - What role do parental satisfaction and parental goals play in pediatric care decisions? Are the management choices made in childhood the best choices for self-management in adulthood?
 - Assess parental ability to give the child independence.
 - Consider collaboration with SBA to develop and refine outcomes measures for neurogenic bladder/bladder exstrophy.
 - Develop guidelines around urodynamics (e.g., Which urodynamic findings drive treatment plans? What are key urodynamic outcomes? Are there standards for measuring urodynamic outcomes? Is there agreement about the values associated with risk? What is the ideal timing of first and subsequent urodynamic studies? This last question should be addressed by the CDC.).
 - Whose quality of life should be measured (e.g., child’s, family unit’s)?
 - Should patient-reported outcomes in transition be assessed through existing models, or are new tools required to measure adolescent and parent perception? How might existing and new tools compare?
 - Develop and validate tools and methods to define and measure successful transition based on patient, family, and clinician needs.
 - How do patients’ perceived needs compare to clinicians’ perceived needs?

- Qualitative research to help determine usefulness of existing tools. Should tools that are judged to bring value be adopted in clinical practice?
- How do patient choices in bladder management change as adolescents transition to adulthood?
- What is the potential for consent among patients with cognitive disability?
- At what age should we transition parents out of the “driver’s seat”?
- What motivates patients to engage in self-care?
- What is the impact of nonphysician support (e.g., social worker, mental health professional) on patient engagement and outcomes?

Neuropathic Bladder and Exstrophy: Nephrologic/Metabolic Issues Group

- Define bladder and renal infection for diagnosis, antibiotic resistance, and asymptomatic bacteriuria/bladder colonization among patients with neuropathic bladder and exstrophy. When should treatment occur for a positive culture in a neuropathic bladder where classic symptoms may not be present?
- What is the effect of age and BBD on prevalence and incidence of bladder/renal infections?
- Develop and test strategies to increase awareness of metabolic alkalosis/acidosis, hypokalemia/hyperkalemia, and total body sodium depletion that may be overlooked in routine clinical care.
- Develop and test strategies to increase awareness of the potential for medications and dehydration to cause AKI in the neuropathic bladder/exstrophy population.
- What is the long-term impact of various bladder management strategies, including bladder neck procedures, on renal function in native and transplanted kidneys? Does aggressive early management of bladder pressures and reflux make a difference? What is the impact of various forms of bladder management on secondary reflux, pyelonephritis, and renal scarring?
- What are the risk factors for bladder stones and carcinomas in various types of bladder management strategies? Define diagnostics to identify risk factors for renal function deterioration (e.g., What is the importance of maintaining low bladder pressures and preventing fecal loading? What is the optimal imaging test and its frequency to identify early renal deterioration?).
- Determine the ideal timing for nephrology involvement in the care of patients with the goal of preserving renal function. Patients with dysplasia may need early nephrology involvement for risk stratification. Identify better ways to measure kidney function/GFR given the issue of commonly decreased muscle mass in the neuropathic bladder/exstrophy population.
- Determine how to integrate primary care into ongoing medical care of the neuropathic bladder/exstrophy population as they age, especially with respect to coordination of care and oversight of overall health needs independent of renal/urologic issues.
- Establish a registry for long-term monitoring of children at risk for deterioration of renal function with common data elements and the ability to follow these patients into adulthood.
- How does renal insufficiency affect growth needs in children and their future bone health as adults? What is the impact of pregnancy on renal function in the neuropathic bladder/exstrophy population?

Male Genital Issues and Reproductive Health Group

- What outcomes should be used to assess fertility?
- Identify indications for varicocelectomy.
- Develop and test provider training to improve genetic counseling.
- Develop better prognostic tools to predict genetic risk.
- Assess the validity of the testicular dysgenesis hypothesis.
- Compare patient- and caregiver-reported outcomes.
- Identify risk factors for and determine the ideal management of UTIs and epididymo-orchitis.
- Determine patient concerns regarding reproductive health.
- Assess the effect of intra-family communication about male genital issues on outcomes/perceived quality of life. assess the psychological effects of male genital diagnoses.
- Conduct an epidemiologic assessment of psychological considerations among male patients with genital/reproductive conditions.
- Conduct an epidemiologic assessment of sexual behaviors among male patients with genital/reproductive

conditions.

- Develop and test assessment and therapy protocols for erectile dysfunction.
- Determine the incidence, prevalence, and natural history of erectile dysfunction in affected adolescents/young adults.
- Conduct an epidemiologic assessment of gender dysphoria.
- Assess the effects of sexual and gender identity and expression on patient self-management.
- Assess reoperative outcomes among males with prior genital surgery.
- Conduct multicenter postoperative outcome studies.
- Harmonize classification systems of genital conditions.
- Develop improved coding within EHR systems to better reflect genital conditions and operations.
- Assess the impact of urethral-affecting disease on bladder function in affected adolescents/young adults.
- Develop and test multidisciplinary training in transition issues.
- Assess the availability and use of (un)reliable sources of patient information/education.
- Determine the efficacy of testicular self-examination in high-risk groups.
- Refine estimates of incidence of primary and secondary gonadal tumors.
- Consider methods of advocacy for male genital conditions.
- Identify ideal longitudinal clinical care pathways.

Female Genital Issues and Reproductive Health Group

- Identify outcomes measures to assess cosmesis following different surgeries.
- Develop and test tools to improve self-image and resilience following different surgeries.
- Develop outcome measures to assess psychosocial issues (e.g., coping, resilience).
- Identify patient factors/characteristics that influence resilience/coping.
- Develop and test health care team training programs that allow clinicians to facilitate resilience and coping.
- Assess patient risk-taking/risk-avoidance behavior and develop ways to address these issues.
- Assess the impact of parental involvement/parenting style on body image, sexuality, and intimacy.
- Identify effective models of care to promote self-management in the transition to adult care across various conditions (e.g., What models are currently used and with what outcomes [epidemiology]? What tools exist/are needed? Is multidisciplinary care best for all conditions, or is there a benefit to having one physician with referrals?).
- Identify the best methods to empower patients and enable system navigation (e.g., tools through electronic media, combined care of adult and pediatric providers during a transition period, assisting the child in beginning to make appointments and taking her own medications).
- Assess the ideal timing and process for discussing fertility issues/expectations/contraception with children (e.g., When is a child ready for fertility counseling/contraception information? What format is best? Should parents be taught how to begin to broach the subject? Should patients have a confidential meeting with the provider?).
- Assess the effect of the home environment (e.g., parents, social aspects, education) on adult outcomes. How is successful transition defined, and what tools/measures are needed to assess transition outcomes?
- Develop and test transition tools for caregivers and patients to support transition planning/decision-making (e.g., patient readiness assessment, tools to assess plan of action, patient/family characteristics to guide care).
- What is the best way to educate adult providers regarding rare conditions that have been predominantly managed in pediatric settings (e.g., materials provided to patients and families, conferences, review articles)?
- Collect normative data on reproductive health among control populations for comparison to women with genitourinary conditions (e.g., compare sexual function and dyspareunia in “normal” controls and women with genitourinary anomalies to determine relative levels of function and identify main issues).
- Assess rates of pregnancy, pregnancy complications (e.g., stillbirth), infertility (e.g., chance of pregnancy after cloacal repair), and other reproductive outcomes across different genitourinary conditions and modes of delivery (e.g., natural, surgical).
- Identify pathways of heritability/genetics across genitourinary conditions and the impact of newer fertility options (e.g., testicular sperm extraction in 46,XY women).
- Assess the potential “overmedicalization” of genitourinary conditions across models of care (e.g., Does a multidisciplinary clinic “overmedicalize” such conditions as Mayer-Rokitansky-Küster-Hauser syndrome?). Assess the clinical impacts and legal issues around parent/guardian consent requirements on

emerging adolescent research (e.g., Do parent/guardian consent requirements limit knowledge obtained from adolescents around personal issues?).

- What interventions/treatment modalities best prevent/manage chronic pain?
- Identify optimal bone health management strategies (e.g., What is the correct amount of estrogen replacement in women post orchiectomy? What are the risks/benefits of estrogen replacement therapy? What are the most effective methods of menstrual suppression for pain control? What risks are involved with hormone suppression in adolescents?).
- What are the most effective protocols for prevention and management of bowel/bladder/uterine prolapse?
- What are the main cancer risk factors and ideal cancer screening protocols for patients with genitourinary conditions/procedures (e.g., bladder augmentation, bowel vaginoplasty)?
- What are the best screening practices and prevention interventions for sexually transmitted diseases (e.g., How can gonorrhea culture/chlamydia be tested for in patients with a urinary diversion who are uncomfortable with a vaginal exam? What are the risks/benefits of the human papillomavirus vaccine in patients with genitourinary conditions?).
- Identify outcome measures to better ascertain sensation in patients with genitourinary conditions with and without reconstructive surgery compared to “normal” control populations.
- What are the risks/benefits for different contraceptive methods in patients with genitourinary conditions? (e.g., How might low risk of pregnancy due to an underlying condition affect the risk/benefit of contraception?).
- Identify and assess new possible methods of fertility preservation (e.g., sperm freezing for androgen insensitivity syndrome).
- Assess intimacy issues unique to genitourinary conditions and identify/test methods to best address them.
- Identify unique best practices for managing specific patient subgroups (e.g., disorders of sex development, congenital adrenal hyperplasia, exstrophy) to improve outcomes.