

# Spina Bifida Community-Centered Research Agenda Update

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# Building the Spina Bifida Community-Centered Research Agenda

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**2016**

Update  
Healthcare  
Guidelines



**100**

**2017**

Held  
Consensus  
Workshop  
and  
Identified  
Gaps



**80**

**2018**

Launched  
*Guidelines  
for the Care  
of People  
with Spina  
Bifida*

**2019**

Surveyed  
Community  
on Issues  
that  
Impacted  
Daily Life



**2000**

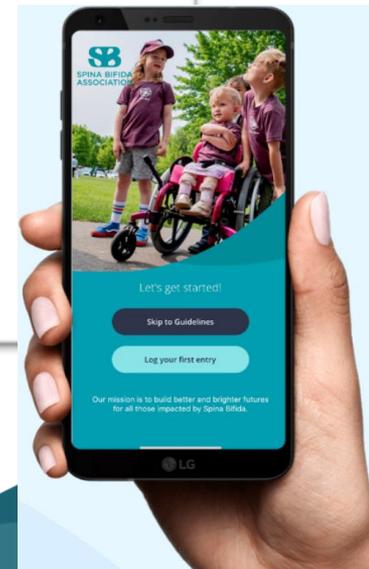
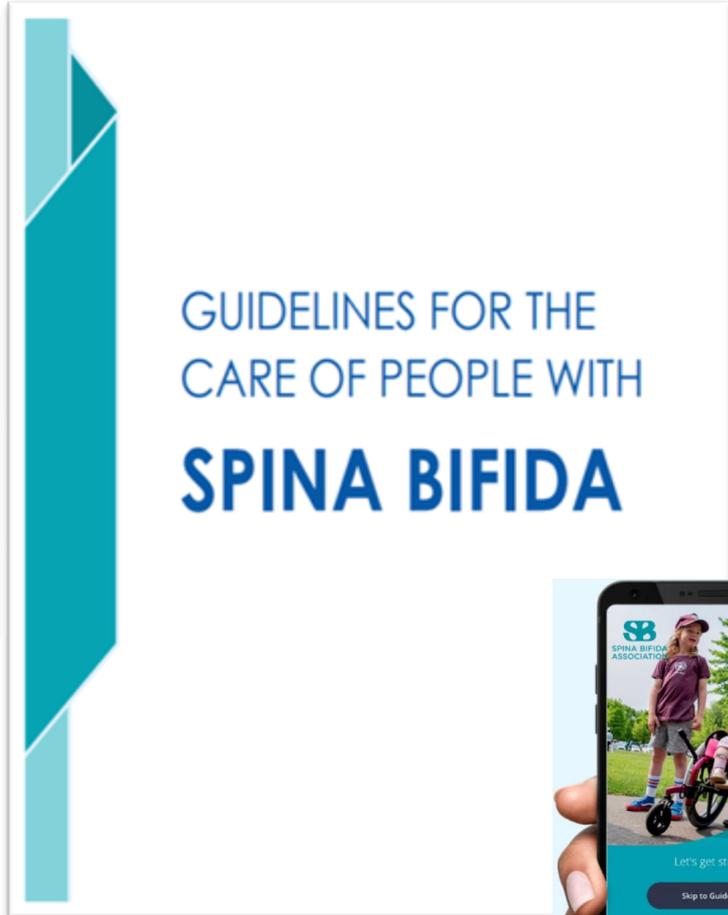
**2020**

Convened  
RAC and  
held  
Discussion  
Groups



**100**

# Healthcare Guidelines



<https://www.spinabifidaassociation.org/resource/guidelinespdfull/>

<https://www.spinabifidaassociation.org/mobile-application/>

<https://www.sciencedirect.com/science/article/abs/pii/S1936657419301062?via%3Dihub>

# Healthcare Guidelines

## System of Care

- Care Coordination
- Health Promotion and Preventive Health Care Services
- Prenatal Counseling
- Transition

## Psychosocial Context for Self-Management

- Family Functioning
- Mental Health
- Self-Management and Independence
- Quality of Life

## Neuropsychology and Neurosurgery

- Neuropsychology
- Neurosurgery

## Mobility, Orthopedics, and Physical Activity

- Mobility
- Orthopedics
- Physical Activity

## Urology and Sexual Health

- Men's Health
- Sexual Health and Education
- Urology
- Women's Health

## Specific Health Issues

- Bowel Function and Care
- Endocrine: Puberty and Precocious Puberty
- Endocrine and the Use of Human Growth Hormone
- Integument (Skin)
- Latex and Latex Allergy in Spina Bifida
- Nutrition, Metabolic Syndrome, and Obesity
- Sleep-Related Breathing Disorders

Over 250 research gaps!

# Online Survey & 2 Focus Groups



- Spring, 2019
- 1607 Survey Participants:
  - Adults with Spina Bifida
  - Parents of children with Spina Bifida
- Two Focus Groups
- Explore research gaps identified in Guidelines

# 27 Items/Topics

Accessing Coordinated Care b/w PCPs & Specialists	Leg/Foot Deformities	Difficulty Breathing due to side effects of SB: Chiari II or Scoliosis
Finding Doctors who care for Adults/Children with SB	Skin Breakdown	Managing Preventable Health Conditions to Avoid ER Visits
Obstructive Sleep Disorders /Sleep Apnea	Urinary Incontinence	Managing Chiari II Symptoms/Consideration Surgical Options
Encourage Self-Management Behaviors in Children	Urinary Tract Infections	Shunt Insertion/Complications
Independence/Ability to Care for Oneself	Bowel Incontinence	Management/Care related to Tethered Cord
Ensuring Daily Physical Activity	Menopause/Women's Health Issues	Learning Disabilities
Mobility	Prenatal Counseling for Women	Depression/Mental Health
Pain Related to Mobility	Prostate Cancer Screening/ Treatment	Managing Weight/Nutrition
Use of HGH/Human Growth Hormone	Early Onset of Puberty	Having an Active Sex Life

# Top 5 Biggest Issues/Problems Overall

## % Citing Each Item as one of their 5 Biggest Issues: Top Mentions\*

Parents of Child <18 with SB	%	Parents of Child 18+ with SB	%	Adults with SB	%
Bowel Incontinence	64%	Finding Doctors	50%	Finding Doctors	55%
Urinary Incontinence	51%	Bowel Incontinence	49%	Bowel Incontinence	51%
Independence	50%	Independence	44%	Managing Weight	44%
Encourage Self Mgt	48%	Managing Weight	40%	Urinary Incontinence	43%
Learning Disabilities/Attention Disorders	41%	Learning Disabilities/Attention Disorders	32%	Depression/Mental Health	42%

\*Multiple responses

# Spina Bifida Community- Centered Research Agenda

# SBA Professional Advisory Council (PAC)

- Research Advisory Council
- Spina Bifida Community-Centered Research Agenda
- Science and Research Subcommittee
  - review and update Guidelines
  - review research proposals
  - inform committee of important/new research

# Science and Research Committee



- Members Partial List
  - Tim Brei,
  - John Foley,
  - Konrad Szymanski,
  - Kathryn Smith,
  - Amy Houtrow,
  - Alexander Van Speybroeck,
  - Michael Partington,
  - Ellen Fremion,
  - Robin Bowman,
  - Sara Struwe,
  - Jonathan Routh,
  - Jerome Chelliah,
  - Judy Thibadeau
- Guidelines Updates either complete and approved by PAC or in preparation and review
  - Neurosurgery
  - Urology
  - Orthopedics
  - Osteoporosis (NEW)
  - Transition
  - Mental Health
  - Care Coordination
  - Family Functioning
  - Weight Management
  - Neonatal (NEW)

# Research Advisory Council (RAC)



- Spring, 2020
- Parents of Children with SB, adults with SB, health care professionals with SB expertise
- Address 6 topic areas of importance that emerged:

Bowel incontinence  
Mental health  
Self-management

Transition  
Urinary incontinence  
Weight mgt/nutrition

# Research Advisory Council



## Members

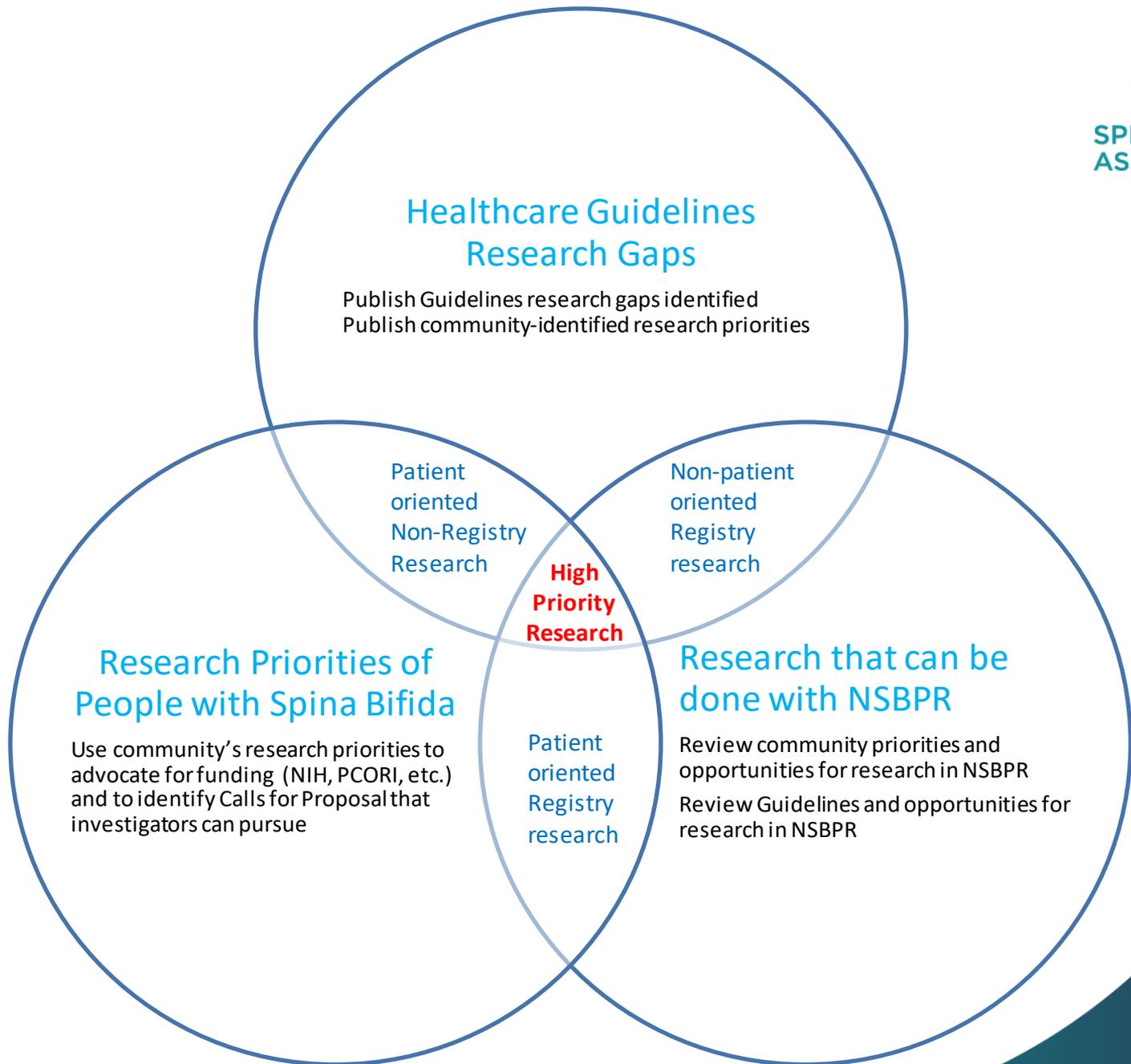
- Linda Thunn
- Maryellen Kelly
- Maya Evans
- Jenni Zimlich
- Sara Berghoefer
- Robert Trierweiler
- Ginny Briggs
- Kerry Wallace
- Jennifer Tudor
- Linda Krach
- Hetty Mollert
- Denise Garza
- Monique Ridosh
- Hubert Swana
- Erin Kelly
- Katie Schmidt
- Rebecca Nieters
- Andrea Fairman
- Sara Beck
- Chairs Alex Van Speybroeck
- Judy Thibadeau

- Diverse Group from all over the country
- Adults with Spina Bifida
- Parents of Children with Spina Bifida
- Professionals from a wide variety of disciplines who work with people with Spina Bifida

# RAC Accomplishments



- Identified 10-12 research questions for each topic they perceived as most important or in need of research
- 12 virtual discussion groups:
  - 2 for each of 6 topics
  - Oct 2020 to April 2021
  - clarify the research questions
  - rank/identify the most important questions for research



# Bowel Incontinence Research Questions



- 1.01 How do long-term, large volume bowel irrigations, anterograde and retrograde, effect colorectal health and bowel continence?
- 1.02 What is the association between bowel continence in childhood and early teen years on the transition to adulthood (specifically looking at outcomes of employment, college, intimacy, and independence)?
- 1.03 Does initiation of a bowel program before age 3 (or 5), predict long-term bowel management compliance? Does it predict short or long-term bowel continence outcomes?
- 1.04 Does close follow-up after changing a bowel program predict short or long-term improved bowel continence outcomes?
- 1.05 What clinical factors predict improved bowel continence?
- 1.06 What modifiable factors have the largest effect of closing the health disparity gap of bowel incontinence

# Bowel Incontinence

Maryellen Kelly DNP, MHSc, CPNP  
Research Advisory Council Topic Leader

# Mental Health Research Questions



- 2.01 What is the correlation of depression and degree of involvement in community activities (working, volunteering, playing adaptive sports) for adults with SB?
- 2.02 Specific to children with SB, what are the mental health concerns faced by their parents, and what systems and structures are needed to address those effectively?
- 2.03 What generates mental health inequities faced by individuals with SB (e.g., lack of access to care, bias/discrimination in the healthcare system, lack of education among providers about how to meet the needs of individuals with SB, particularly those who are also part of other minority groups)?
- 2.04 What factors that support mental health are correlated to a better quality of life in those with Spina Bifida (e.g., social support, mentorship, physical activity, transportation, level of education, etc.)?
- 2.05 Do treatments for mental health disorders have significantly different effects in those with Spina Bifida?
- 2.06 What types of system changes are needed to confront mental health inequities experienced by individuals with SB (e.g., more education for healthcare providers, etc.)?

# Mental Health

Ginny Briggs, PhD

Research Advisory Council Topic Leader

# Self-Management Research Questions



- 3.01 How do factors (e.g., cognitive ability, executive functioning, gross motor delays, surgeries, parental readiness, social factors) interact with introduction (i.e., early intervention) of self-management interventions to influence outcomes?
- 3.02 What self-management interventions for Spina Bifida are efficacious and available across various settings for educators, mental health providers, and rehab specialists (i.e., PT, OT, Speech)?
- 3.03 How can we support individuals and families in the development of self-management skills, ability, responsibility, strategies, techniques, behaviors across settings and lifespan (early childhood, middle childhood, adolescence, young adulthood, adulthood, older adult, etc.)?
- 3.04 How do factors that predict self-management behaviors in youth impact outcomes (e.g., health, employment, quality of life) in adults and their family?
- 3.05 How does executive functioning of the individual (adult populations) play a role in their self-management ability?

# Self-Management

Monique Ridosh, PhD, RN  
Research Advisory Council Topic  
Leader

### **3.01 How do factors (e.g., cognitive ability, executive functioning, gross motor delays, surgeries, parental readiness, social factors) interact with introduction (i.e., early intervention) of self-management interventions to influence outcomes?**



#### **Proposed study /aims - Did the individual experience a formal transition from childhood care to adult care?**

- Assumptions are often made for adults still living with their parents. Need good assessments to see if the “child” can be independent.
- Only info in registry is type of health insurance
- Self- Efficacy
- Parental support & what the household looks like
- What type of supports are available for adults?
- AMIS-II has independent living scale.
- Good ongoing assessment – Vineland Adaptive Behavior Scales – assesses functioning in many domains
- IEP – special education/accommodations
- Consider Full scale IQ

#### **Proposed study /aims - What factors increase/decrease self management interventions on outcomes?**

- Questions in Registry are limited; shunt – background

#### **Proposed study /aims - What is the involvement of parents, e.g., parents of adults?**

- Self advocacy vs. parent involvement

# Barriers - Self Management Studies

- There may be a need for smaller projects that are the background needed to answer some of the components of these questions. The questions are large, complex, many require longitudinal methods and more aligned with program of research rather than one study or project
- Limited because self-management is not in registry, there is no common measure
- To seek funding need lead or co-lead with expertise to gain funding
- Building teams around topics will require time and other activities to be able to conduct research such as critical review of measures; recommend meeting again to establish partnerships for activities that would lead to collaboration

# Pilot Study

- The self-management workgroup meeting discussion led to an idea for a pilot across 3-4 interested spina bifida clinic sites to develop a telehealth/goal setting intervention. This could be a behaviorally focused telehealth visit (bridge between regular clinical visits) to optimize goals. This may be a helpful point of contact, perhaps billable, that could contribute to sustainability of a goal attainment initiative

# Transition

## Research Questions



- 4.01 How can medical professionals help parents to support their children to become adults?
- 4.02 What can health care workers do to best support transitioning adolescents to adult services?
- 4.03 For adults with SB who are or were working, what helped make their move to employment successful?
- 4.04 Identify barriers specific to minority individuals with SB in acquiring the skills for moving into the adult world and ways to overcome them.
- 4.05 What are the most important actions that help or hinder children with SB in learning the skills needed to successfully become an adult?

# Transition

Robert Trierweiler CRC, LCPC  
Research Advisory Council Topic  
Leader

# Urinary Incontinence Research Questions



5.01 What is the change in continence for those who have a bladder neck procedure carried out (1 year, 5 year, 10 years post-op)? What is the change in continence for those who undergo bladder augmentation (1 year, 5-year, 10-year post-op)?

5.02 Is there a change in the number of confirmed UTIs an individual has after bladder surgery?

5.03 What is the association between time and type of training/information provided for intermittent catheterization and compliance?

5.04 What is the rate of frequency that urinary continence is discussed in relation to its effects on sexuality and sex?

5.05 Does proximity to a multi-disciplinary SB clinic affect urinary continence outcomes and renal function? What are modifiable factors that can decrease the health disparity of urinary continence in the SB population?

# Urinary Incontinence

Hubert Swana, MD  
Research Advisory Council Topic  
Leader

Research Topic	Proposed Study Hypotheses/Aims	Relevant Variables in NSBPR	Potential Funding Sources	Interested Collaborators *= possible PI/Leader
<p>What is the change in continence for those who have a bladder neck procedure carried out (1 year, 5 year, 10 years post-op)?</p>	<p>Do bladder procedures decrease the frequency, quantity and bother of incontinence -- BN, augment, or both?</p>	<p>-As listed in NSBPR -Some patients may come off meds. -Gender differences -Race, ethnicity -</p>	<p>Within NSBPR</p>	<p>-Courtney Streur -Propose to NSBPR clinics and ask for interested collaborators -Rebecca Sherlock</p>
<p>What is the change in continence for those who undergo bladder augmentation (1 year, 5-year, 10-year post-op)?</p>	<p>-Modifiable factors -compliance with cathing -age at surgery? -Independence/self-management -weight or BMI</p>	<p>Insert NSBPR variables relevant to proposed study 2</p>	<p>Within NSBPR</p>	<p>Leader* Collaborator 1 Collaborator 2</p>
<p>What is the change in continence for those who undergo bladder augmentation (1 year, 5-year, 10-year post-op)?</p>	<p>Relationship to bowel program</p>	<p>-On a bowel program? -Status of bowel continence -Compliance w program? -Type of bowel pgm</p>	<p>Within NSBPR</p>	<p>Leader* Collaborator 1 Collaborator 2</p>

Research Topic	Proposed Study Hypotheses/Aims	Relevant Variables in NSBPR	Potential Funding Sources	Interested Collaborators *= possible PI/Leader
<p>What is the rate of frequency that urinary continence is discussed in relation to its effects on sexuality and sex?</p>	<p>Does comfort level of discussing incontinence w partners affect frequency of sex</p>	<ul style="list-style-type: none"> <li>-Age</li> <li>-Relationship status</li> <li>-B/B continence status</li> <li>-Info on sexual activity</li> <li>-Type and level of SB</li> <li>-Gender</li> <li>-Already have “talking points” they share w partner?</li> <li>-coping mechanism</li> </ul>	<p>Insert potential funding sources for Study 3</p>	<p>Leader* Collaborator 1 Collaborator 2</p>
	<p>Are patients avoiding sex and/or intimate relationships because of fear of incont...?</p>	<ul style="list-style-type: none"> <li>-Age</li> <li>-Relationship status</li> <li>-B/B continence status</li> <li>-Info on sexual activity</li> <li>-Type and level of SB</li> <li>-Gender</li> <li>-Already have “talking points” they share w partner?</li> <li>-coping mechanism</li> </ul>	<p>Insert potential funding sources for Study 4</p>	<p>Leader* Collaborator 1 Collaborator 2</p>

Research Topic	Proposed Study Hypotheses/Aims	Relevant Variables in NSBPR	Potential Funding Sources	Interested Collaborators *= possible PI/Leader
<p>What is the rate of frequency that urinary continence is discussed in relation to its effects on sexuality and sex?</p>	<p>-Incontinence occurs during sex and has a negative impact</p>	<p>-b/b incontinence occurring during sex            -Age            -Relationship status            -B/B continence status            -Info on sexual activity            -Type and level of SB            -Gender            -Already have “talking points” they share w partner?            -coping mechanism</p>	<p>Insert potential funding sources for Study 1</p>	<p>Leader*            Collaborator 1            Collaborator 2</p>
	<p>-On demand approaches? ie medication taken in anticipation of sex            Botox, bladder meds, cathing before sex</p>	<p>-Age            -Relationship status            -B/B continence status            -Info on sexual activity            -Type and level of SB            -Gender            -coping mechanism</p>	<p>Insert potential funding sources for Study 2</p>	<p>Leader*            Collaborator 1            Collaborator 2</p>

# Weight Management Research Questions



6.01 For people with Spina Bifida who have been able to maintain and/or improve their health, what factors led to their success? (e.g., nutrition, physical activity, regular sleep, weight management, etc.)

6.02 What things make it difficult for someone with a disability to exercise regularly (e.g., expense of exercise equipment, cost of gym membership, lack of availability of disability-friendly gyms/classes, limitations of handicapped accessibility outdoors, etc.)?

6.03 What difficulties do adults with a disability encounter in healthy eating and independent food preparation (e.g., mobility obstacles in cooking, and grocery shopping, etc.)?

6.04 How do the habits of someone with Spina Bifida early in life affect physical mobility and function across their lifespan?

# Weight Management

Rebecca Nieters

Research Advisory Council Topic  
Leader

# Research Workshop



- Nov, 2021
- Over 200 registrants
- Foster building of grantwriting teams
- Explore potential for NSBPR to be used to answer some questions
- CDC, NIDILRR and NIH attended

# NIH Data Base



[Developing a SMART scaffold for bladder augmentation](#)

[Developmental Mechanisms of Human Meningocele](#)

[Sleep-disordered breathing in infants with myelomeningocele](#)

[Surface ectodermal mechanism and maternal intervention of neural tube defects](#)

[Stem cell-based biomaterials for spinal regeneration in neural tube defects](#)

[IMMUNE RESPONSES IN THE MOTHER-INFANT DYAD INDUCED BY FETAL SURGERY, AND ASSOCIATIONS WITH PREMATUREITY](#)

[Translational profiling of bladder sensory nerves and their cell type identities using dissociation free single nucleus sequencing](#)

[Ecological momentary assessment to understand the impact of daily urinary and fecal incontinence on well-being among adults with spina bifida](#)

[Self-Management in Adolescents and Young Adults with Spina Bifida](#)

[Body Composition and Energy Expenditure in Youth with Spina Bifida](#)

[Examining Epidemiology of Folate Status Attributable to Adolescent Alcohol Use](#)

[Uterine Wall-Membrane Anchor Device for the Prevention of Preterm Premature Rupture of the Membranes Following Fetoscopic Surgery](#)

# Questions?