Information Collection Request
New

The Natural History of Spina Bifida in Children Pilot Project

Supporting Statement Part B

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B. Statistical Methods

Because this pilot project relies in part on a convenience sample of participants, statistical sampling methods will not be used to draw a study sample or analyze the project data. Additional information regarding participant recruitment is included in section B1.

B1. Respondent Universe and Sampling Methods

Participants will be drawn from families with a child diagnosed with SB who is residing in the state of Georgia. Families with a child who is 3-, 4-, or 5-years of age at the time of recruitment will be eligible to enroll in the project. Depending on the start date of data collection, it is most likely that the eligible children will have been born between the years 2003 and 2006. Reliable estimates of how many eligible children with SB were born and currently live in the state of Georgia are not available. Therefore, we cannot know a priori the size of the potential respondent universe, or where in Georgia potential participants reside. One of the aims of this pilot project is to explore the feasibility of locating and recruiting participants using and comparing different sources. For this pilot project we propose to identify participants from three major sources (Metropolitan Atlanta Congenital Defects Program (i.e. MACDP), SB clinic, and “Other”) resulting in a population based-sample (MACDP) and a convenience sample (SB clinic and “Other”).

The MACDP covers five Georgia counties (Fulton, DeKalb, Cobb, Gwinnett, and Clayton). The majority of the eligible families identified in the MACDP are expected to still reside in this five-county region; however, some of these families may have relocated outside of the MACDP catchment area. Based on current information supplied by MACDP personnel, 57 children were born with SB in the MACDP catchment area between 2003 and 2006 (this number may change, as the data for 2006 were incomplete at the time of this submission). No comparable estimates exist for the rest of the state. To use the MACDP for recruitment purposes provide strengths, such as an established sampling frame of eligible children, a physician-confirmed diagnosis of SB, and limited contact information that is periodically updated. A drawback of solely recruiting from the MACDP is that it is limited to five Georgia counties. The experience of growing up with SB may be quite different for families and children with SB who are not living in metropolitan Atlanta. Recruitment strategies to reach these families will be different as well. Therefore, a convenience sample was added. In addition to recruiting from the MACDP, eligible families will be recruited from the Children’s Healthcare of Atlanta at Scottish Rite SB clinic, through flyers posted in pediatric sub specialists’ offices located throughout Georgia, and through advertisements posted in newsletters and websites likely to be seen by parents of children with SB (convenience sample). The inclusion of a convenience sample will provide information on how to create a sampling frame in the absence of a surveillance program such as MACDP.

The estimated sample size of approximately 40 families is likely too small for inferential statistical analyses. Moreover, this is a pilot project and the results will inform the planning and implementation of a larger, prospective multi-state project in the future. Thus, specific hypothesis testing is not the main purpose of the project. Therefore, no statistical power estimations were calculated. Nonetheless, as the MACDP is a population-based, active surveillance system, inferences could possibly be made to families with a young child with SB residing in metropolitan Atlanta. The remainder of the project sample will constitute a convenience sample of families with a young child born with SB residing in the State of Georgia. Any inferences made will have to be contingent on that.

B2. Procedures for the Collection of Information

As described in B1, participants will be recruited from a variety of sources. The recruitment procedures are described below.

Recruitment Sources

MACDP

MACDP personnel will identify the eligible families in their database and mail the families a letter (see Attachment P) asking for permission to share their contact information with the researchers at the CDC's Disability and Health Branch. The MACDP data are protected with an Assurance of Confidentiality under Section 308(d) of the Public Health Service Act. Therefore, personnel from the Disability and Health Branch cannot access any information from the MACDP before the eligible families have agreed to let them do so. Mr. Jim Kucik will identify the eligible families and mail them a letter asking for permission to share their contact information with other CDC researchers (see Attachment P). Project personnel will then telephone parents who grant permission to MACDP to share their contact information. The recruitment script can be found in Attachment Q. Prior to the telephone call, an advance letter will be mailed to the family (Attachment R).

Children's Healthcare of Atlanta at Scottish Rite SB Clinic

A clinic coordinator at the Children's Healthcare of Atlanta at Scottish Rite SB clinic will identify eligible patients in their files and mail these families a letter about the project (See Attachment S). The pilot project staff will not have access to the clinic's files. Additionally, project flyers and pamphlets will be posted in the clinic waiting room. These project flyers and pamphlets are the same as in Supporting Statement A and to avoid redundancy they are therefore not attached in Supporting Statement B. Interested parents can contact the project recruiter to learn more about the project and enroll. The project will be subject to Children's Healthcare of Atlanta internal IRB review.

Pediatric Sub Specialists in the State of Georgia

Pediatric sub specialists (e.g., neurosurgeons and urologists) in Georgia who treat patients with SB will be contacted and informed about the project. With their permission, we will initiate recruitment of eligible families from their practices by placing recruitment flyers and pamphlets in the waiting rooms.

Spina Bifida Association of Georgia
A notice about the project will be posted in the SB Association of Georgia's newsletter and on their webpage (see Attachment T).

NBDPS
The NBDPS distributes a newsletter to their study participants. A brief ad containing the same text as attachment T about the project will be placed in the newsletter to inform eligible families of the current project. Although the families in the NBDPS will also appear in the MACDP, an ad in the NBDPS newsletter may communicate the study's legitimacy and it may prompt families who did not receive a letter from MACDP (because the letter was undeliverable or MACDP did not have a current address for the family) to contact us to learn more about the pilot study.

Recruitment Process
Families identified through MACDP (who have authorized in writing the MACDP to share their contact information with the pilot project researchers) or the SB clinic at the Children's Healthcare of Atlanta at Scottish Rite will be mailed a letter that describes the project (attachments R and S). A NORC bilingual interviewer will then attempt to contact the family by telephone to answer any questions that they might have about the project and ask whether the family would like to participate in the pilot project. If a family responds to flyers, notices, or advertisements, they will make initial contact with NORC via a toll-free project telephone number. At that time, the NORC interviewer will answer any questions the family might have about the project and ask if they would like to participate. The recruitment script is included in Attachment Q.

During the recruitment process, parents will be asked to verbally confirm that their child has a diagnosis of SB and that their child is between 3 and 5 years of age. An explanation of the project will follow during which the parents will be encouraged to ask any project related questions they may have. If they are interested in participating, updated contact information will be obtained. Next, the parent will choose which project component they want to participate in. At this time, only the telephone survey component will be available in Spanish, as we do not have the resources needed to offer the in-person component in a language other than English. If the in-person component is chosen, the parents will be given the option of participating in the "research-only" (shorter time commitment) or the "in-depth" evaluation (the neuropsychologist will provide individualized feedback based on the child’s performance and the parent’s interview). Next, an appointment will be scheduled. If the parent chooses to participate in the telephone survey component, the interviewer will proceed by either administering the survey or by scheduling an appointment to administer the telephone survey during a time that is convenient for the parent. Both the parent and the child will participate in the in-person component whereas only the parent participates in the telephone survey component.

If parents decide not to participate in the pilot project, the interviewer will attempt to determine why the parent is no longer interested (“May I ask why you are no longer interested?”) and then thank the parent for taking the time to learn more about the project. A separate recruitment log will be used to record the reasons stated for deciding not to participate (Attachment U).

We have developed an answering machine script to be used by interviewers in the event that they are unable to reach the parent for a recruitment call or a scheduled interview (Attachment V). Interviewers will leave a message every third call when attempting to recruit participants. If a parent misses a scheduled appointment, interviewers will leave the message when they reach the family at the scheduled time.

Procedure and Measures

Telephone Survey Component

Parents who choose to participate in the telephone-survey component will complete a 35-45 minute telephone interview (same survey as included in Supporting Statement A and therefore not included herein). The survey contains 201 items and addresses the following six domains: medical concerns; development and learning; nutrition and physical growth; mobility and functioning; general health; and family demographics. Although most of the items were created for this specific project, many of the more generic items have previously been used in large national surveys such as the Youth Risk Behavior Survey and the National Early Intervention Longitudinal Study. After completion of the project, the survey will be revised based on participant and interviewer feedback. A NORC telephone interviewer will administer the interview at a time that is most convenient to the parent. Parents who complete the telephone survey will indicate their verbal consent to participate in the interview. The verbal consent will also inform parents that the project will seek a separate authorization to access the child’s medical and EI records (where applicable). Parents will be told that they can still participate in the survey even if they do not want to authorize access to their child’s records. At the conclusion of the interview, parents will be asked to provide their address so that we can mail the $25.00 token of appreciation payment to them. In addition, parents will be reminded that authorization forms to retrieve the child’s medical and EI records will be included with the mailing. A letter will be included with the incentives (Attachment X).

Parents will be informed that it is their choice to allow the project access to their child’s records and that access to the records will allow researchers to better understand the issues children and families with SB experience. Parents will be asked to return the signed authorization forms to the PI using a self-addressed, stamped envelope that will be included in the mailing.

In-person Component

Parents who choose to participate in the in-person component will be required to travel to the neuropsychology clinic at the Children’s Healthcare of Atlanta at Scottish Rite. A map of where the clinic is located will be included in the reminder appointment letter that will be mailed by the recruiter once the appointment has been made (See Attachment Y). When the family arrives at the study site, the CDC PI or other trained project professional will greet the family and obtain the parent’s written consent for himself/herself and the child to participate in the in-person component. There are two separate consent forms for the in-person component: one for the “research only” and one for the “in-depth” evaluation (consent forms attached in Supporting Statement A) and the parents will be given the appropriate consent form based on which evaluation they said they wanted during the recruitment. During the consent process, the parent will also be reminded about the separate authorization forms to access the child’s medical and EI records. Parents will be informed that it is their choice to allow the project access to the child’s records and that they can still participate even if they do not authorize access to their child’s records.

The CDC PI, or other trained project professional, will administer the survey (same survey as used in the telephone component) and, when the parent has chosen the “in-depth” evaluation, the Children’s Healthcare of Atlanta Patient History Questionnaire (Attachment AG) to the parent. Then the project professional will ask the parent to complete a few additional self-administered measures. In case the parent has difficulty reading, the CDC PI will read the measures and assist the parent to complete them. While the parent completes his/her portion of the project, a licensed child neuropsychologist will assess the child in a separate room. If a child or parent is uncomfortable with being separated from each other, parents will have the option to sit in the room with the child and silently observe the assessments. If the family chooses this option, parents will be asked to complete their self-administered measures while seated in the room with the child and asked specifically to not assist the child with the assessments. If the parent chooses to be in the room with the child, the parent’s interview will be completed prior to beginning the child’s assessments. At the conclusion of the in-person component, parents will receive a $50.00 token of appreciation payment and be reimbursed for their travel expenses based on the federal mileage rate.

The other measures included in the In-Person Component are: the Behavior Rating Inventory of Executive Function-Preschool Version (attachment AD); the Pediatric Evaluation of Disability Inventory (attachment AC); the McMaster

Family Assessment Device (attachment AB); the Behavior Assessment System for Children, 2nd Edition ("in-depth" evaluation only: attachment AE); and the Adaptive Behavior Assessment System-2nd Edition ("in-depth" evaluation only: attachment AF).These measures are described in more detail below:

Parent Measures:

Behavior Rating Inventory of Executive Function - Preschool Version (BRIEF-P). The parent version of the BRIEF-P will be used as one measure of child executive functions. The BRIEF-P consists of 63 items and is comprised of five statistically and theoretically derived subscales of executive functions (Emotional Control, Shift, Inhibit, Working Memory, and Plan/Organize), three broader indexes (Inhibitory Self-Control, Flexibility, and Emergent Metacognition), and an overall composite score (Global Executive Composite; Gioia, Andrews Espy, & Isquith, 2003). In addition, two validity scales can be calculated (Inconsistency and Negativity). A 3-point Likert response scale (Never, Sometimes, or Often) is used to indicate how often the child has had a problem with a specific behavior over the past six months. Raw scores are converted to age and sex specific *T*-scores, percentile scores, and confidence intervals. *T*-scores above 65 are considered potential areas of concern (e.g., Mahone & Hoffman, 2007). The BRIEF-P was standardized using a sample of 460 parents of children with children as young as 2 years of age and as old as 5 years and 11 months from different geographical regions, socio-economic backgrounds, and races (Gioia et al., 2003). Internal consistency, as measured by Cronbach's alpha coefficients, ranged from .80 (Plan/Organize) to .95 (Global Executive Composite). Test-retest reliability coefficient ranged from .78 to .90 over an average interval of 4.5 weeks (Gioia et al., 2003). The BRIEF-P has been used with children with a wide range of neurological, psychiatric, and medical conditions as well as pervasive developmental disabilities, traumatic brain injuries, learning disabilities, lead exposure, language disorders, and attention deficit hyperactivity disorder (Gioia et al., 2003). The BRIEF-P is included in Attachment AD.

Pediatric Evaluation of Disability Inventory (PEDI). The PEDI was developed as a tool to measure functional abilities of children with different types of disabilities by either observation, structured interview, or parent report (Haley, Coster, Ludlow, Haltiwanger, & Andrellos, 1992). The parent report version will be used in the current project. The PEDI measures both capability and performance of functional activities in three domains: Self-care, Mobility, and Social Function. The inventory is organized into three distinct parts. Part I measures Functional Skills (197 items); Part II measures Caregiver Assistance; and Part III measures Modifications (Part II and III 20 items in total). The response anchor for Functional Skills is "0 = Unable, or Limited in Capability, to Perform Item in Most Situations", or "1 = Capable of Performing Item in Most Situations, or Item has been Previously Mastered and Functional Skills have Progressed Beyond this Level". Each of the 20 items requires one score for Caregiver Assistance and one for Modifications (Parts II and III). The response anchor for Caregiver Assistance is "5 = Independent", "4 = Supervise/Prompt/Monitor", "3 = Minimal

Assistance", "2 = Moderate Assistance", "1 = Maximal Assistance", or "0 = Total Assistance." Modifications is coded as "N = No Modifications", "C = Child-oriented (non-specialized) Modifications", "R = Rehabilitation Equipment", "E = Extensive Modifications". Summary scores ($M = 50$, $SD = 10$) will be calculated for Functional Skills and Caregiver Assistance in each of the three content domains (Social Function, Mobility, and Self-Care), resulting in six separate scores. A total score cannot be calculated.

The normative sample for the PEDI consisted of 412 children between the ages of 6 months and 7.5 years from the Northeast of the United States. Cronbach's alpha coefficients range between .95 and .99 (Haley et al., 1992). As there are different methods of gathering the PEDI data, intraclass correlation coefficients were calculated between responses from parents and members of the child's rehabilitation team. The intraclass correlation scores ranged from .74 to .96 for most of the scale summary scores (Haley et al., 1992). Expected relationships with other measures of similar constructs support the validity of the PEDI (Haley et al., 1992). The PEDI is included in Attachment AC.

McMaster Family Assessment Device (FAD). McMaster Family Assessment Device (FAD). The FAD will be used as a parent report of family functioning. The measure has been developed for use with adolescent, geriatric, clinical or non-clinical populations. The FAD consists of 60 items that load on 7 scales (Problem Solving; Communication; Roles; Affective Responsiveness; Affective Involvement; Behavior Control; and General Functioning; Ryan, Epstein, Keitner, Miller, & Bishop, 2005). Both unhealthy family functioning (negative) and healthy family functioning (positive) items are included. Negative items are transformed before final scores are computed. A 4-point Likert response scale (Strongly Agree, Agree, Disagree, or Strongly Disagree) is used to indicate how well each of the 60 statements describes the family. Raw scores will be transformed to scaled scores (Ryan et al., 2005). The scale scores range from 1.00 (healthy) to 4.00 (unhealthy). Cronbach's alpha coefficients for the different subscales range from .57 to .86. One-week test-retest reliability coefficients range from .67 to .76 (Sawin & Harrigan, 1995). The FAS is included in Attachment AB.

Behavior Assessment System for Children 2nd Edition (BASC-2; included in in-depth evaluation only) The BASC-2 is a multimethod, multidimensional system that is used to evaluate the behavior and self-perceptions of children and young adults ages 2-25 years (Reynolds & Kamphouse, 2004). It is multimethod because it contains five components (teacher and parent rating scales, self-report, structured developmental history, and classroom observations) that can be used individually or in any combination. The BASC-2 is multidimensional because it measures multiple aspects of personality and behavior, including adaptive as well as clinical dimensions. Only the Parent Rating Scales (PRS) for 2-5 year olds will be included in the proposed project. The parent responds to descriptions of observable positive and negative behaviors using a 4-point scale to indicate whether each item "Never", "Sometimes", "Often", or "Almost Always" applies. Responses are then summed to create separate primary and content raw scale scores and a composite score, which are then converted to T-scores. Cronbach's alpha coefficients for primary and content scale scores on the PRS for 2-5 year olds range from .70 to .93. Test-retest reliability ranges between .72 and .86 (Reynolds & Kamphouse, 2004). The BASC-2 is included in Attachment AE.

Adaptive Behavior Assessment System 2nd Edition (ABAS-2; included in in-depth evaluation only) The ABAS-2 is an assessment of the adaptive behaviors and skills of individuals from birth through age 89. This system is a comprehensive and norm-referenced assessment that measures daily living skills, which are defined as what people actually do, or can do, without the assistance of others. This assessment covers the ten adaptive skills specified in the Diagnostic and Statistical Manual of Mental Disorders, Fourth Edition, Text Revision (DSM-IV-TR), which are grouped into the following three domains: social, practical and conceptual. Scores are generated for all ten skill areas, the three domains, as well as an overall composite score. The ABAS-2 is designed to gather different perspectives as it contains five rating forms for specific age ranges and specific respondents who are familiar with the daily activities of the evaluated individual. The parent/caregiver form will be used for the proposed project. The internal consistency coefficient for the proposed age group and for all forms ranges from .98 to .99 (Harrison & Oakland, 2003). The test-retest reliability coefficient is .90 for all forms including the parent/caregiver form. The ABAS-2 is included in Attachment AF.

Children's Healthcare of Atlanta Patient History Questionnaire (included in in-depth evaluation only) This is a non-standardized background measure that the neuropsychologists of the Department of Neuropsychology at the Children's Healthcare of Atlanta at Scottish Rite have developed to provide additional information deemed important when evaluating a child's neuropsychological function. The questionnaire is divided into 6 sections: Identifying Information; Pregnancy and Newborn History; Developmental History; Medical History; Educational Background; and Social History. Some of the information, or very similar information, included in this particular measure is already asked in other measures that are included in the project. In order to avoid redundancy, the items or sections that are asked in other portions of the project have been put in parentheses and will not be asked of the parents. After the family has finished and left the clinic, the CDC PI or other project staff will transfer the relevant information to the Children's Healthcare of Atlanta Patient History Questionnaire in order that the neuropsychologist will have the complete information they need. The Children's Healthcare of Atlanta Patient History is included in Attachment AG.

Child Measures:

Differential Abilities Scale 2nd Edition (DAS-2)

The DAS-2 measures a wide range of cognitive abilities in children aged 21/2 to 17 years and 11 months (Elliott, 2007). The DAS-2 is divided into 2 batteries (Early Years and School-Age) and is comprised of 20 subtests, which can be divided into core and diagnostic subtests. In the proposed project, we will use the 6 core subtests of the Early Years Battery, which are: Verbal Comprehension, Picture Similarities, Naming Vocabulary, Recall of Objects, Pattern Construction, Matrices, and Copying. The scores from these subtests provide the basis for the computation of a General Conceptual Ability score as well as for cluster scores. T-scores, standard scores, percentiles, and confidence intervals can be computed (Elliott, 2007). The DAS-2 was standardized on a normative sample that is representative to the general population. In addition, children and adolescents with identified developmental risks, attention-deficit-hyperactivity disorder, and certain learning disabilities were also included. The DAS-2 is included in Attachment AH.

Peabody Picture Vocabulary Test 4th Edition (PPVT-4) The PPVT-4 measures receptive vocabulary and can be administered to children as young as 2 years and 6 months. The PPVT-4 includes 4 training items and 228 test items each consisting of four full-color pictures as response options on a page. Children are presented with these 4 pictures and asked to indicate which picture represents the word spoken by the administrator. The 228 items sample words represent 20 content areas and parts of speech across all levels of difficulty. The one-week test-retest reliability coefficient for the PPVT-4 was .93 (Dunn & Dunn, 2007). The internal consistency reliability within the proposed age group is .94 (Dunn & Dunn, 2007). The PPVT-4 is included in Attachment AI.

NEPSY-II The NEPSY–II is a measure that allows clinicians to create a tailored cognitive assessment specific to a child's situation in order to answer referral questions or diagnostic concerns. The NEPSY-II is designed for children ages 3 years to 16 years and 11 months. This measure assesses across the following six domains: Attention and Executive functioning, Language, Memory and Learning, Sensorimotor, Visuospatial Processing and Social Perception. These six domains are divided into 32 subtests that allow for precise application. The Comprehension of Instructions and the Word Generation subtests from the Language domain and the Sentence Repetition from the Memory and Learning domain will be the only subtests administered for the proposed project. A scaled score will be obtained from each of the proposed subtests. The test-retest reliability ranges from .72 to .89 for the proposed subtests. The consistency reliability coefficient within the proposed age group for the Language domain is .90 and .91 for the Memory and Learning domain (Korkman, Kirk, & Kemp, 2007). The NEPSY–II are included in Attachment AJ and AK.

Wide Range Assessment of Visual Motor Abilities (WRAVMA) The WRAVMA provides a Visual-Motor Integration Composite for children aged 3 to 17 years of age from three separate subtests of Fine-Motor, Visual-Spatial, and Visual-Motor abilities. The three areas can be assessed individually or in combination to yield a comparison of a child's integrated visual-motor ability with the skill areas of visual-spatial and fine motor abilities. The proposed project will utilize the WRAVMA Matching Visual-Spatial subtest as well as the WRAVMA Pegboard Fine-Motor subtest. A standard score will be obtained for each of these subtests. Reliability measures of the three subtests of the WRAVMA show internal consistency coefficients exceeding .90 and test-retest reliability coefficients ranging from .81-.91. Construct validity is supported by item separations of .99 (Adams & Sheslow, 1995). The WRAVMA is included in Attachment AL.

Bracken Basic Concept Scale (BBCS-R) The BBCS-R is used to assess basic concept acquisition and receptive language skills of children ages 2 years 6 months through 7 years 11 months. The BBCS-R measures comprehension of 308 foundational and functionally relevant educational concepts in 11 subtests or concept categories: colors, letters, numbers/counting, sizes, comparisons, shapes, direction/position, self/social awareness, texture/material, quantity, and time/sequence. Test items are largely pictorial, and the concepts are presented orally within the context of complete sentences and visually in a multiple-choice format.

Of the 11 subtests, the first six compose the School Readiness Composite (SRC). The SRC is used to assess children's knowledge of those readiness concepts that parents and preschool and kindergarten teachers traditionally teach children in preparation for formal education. The SRC will be the only assessment from the BBCS-R used for the proposed project. Standard scores will be calculated for the six subtests and the for the SRC. Test-retest reliabilities have been reported at .86 and split-half internal consistency coefficient has been reported at .95 (Bracken, 2006). The BBCS-R is included in Attachment AM.

Measures Included but not Filled Out by Participants:
We will schedule collection of medical and EI records for the children whose parents have authorized that we may do so at a time that is convenient for officials working at the relevant medical clinic or EI site. More information can be found in A5. In addition, a recruitment summary form will be used to collect data on recruitment

Recruitment Summary Form: The recruitment summary form will be filled out bi-weekly by NORC personnel in charge of participant recruitment. The form consists of 9 items and was created specifically for this project to facilitate tracking of the recruitment process (attachment H).

Medical Records Data Abstraction Form: The medical records form was created to facilitate and structure the data collection of SB specific medical information. The form consists of four overall sections A) Neurosurgery, B) Urology, C) Orthopedics, and D) Hospitalization. In creating this form, leading experts and practicing physicians who treat children with SB (pediatric neurosurgeon, pediatric urologist, and pediatric orthopedic surgeon), thus record medical information on a daily basis, were consulted. The form will be revised as needed after completion of the project (attachment F).

EI Records Abstraction Form. The EI records abstraction form will be used to abstract information from the early intervention records. It will be revised as needed after the completion of the project (attachment G).

Training for All Study Personnel

NORC will be responsible for participant recruitment and the telephone survey data collection. All interviewers and supervisors at NORC undergo extensive training in telephone survey operations and receive training on security and confidentiality.

Training Related to the Phone Survey Component

All NORC interviewers undergo training to instruct them on basic interviewing skills needed to conduct interviews successfully; education on policies and procedures, with special emphasis on respondent confidentiality; ongoing evaluation on the quality of the interviewing process; general overview of the interviewer function in survey research; introduction of the study, topic, and sponsor to the respondent; ways to gain cooperation and assure respondent confidentiality; methods to avert refusal; how to conduct a model interview; mechanics of a questionnaire; coding and probing techniques; confidentiality of data; and record keeping. A variety of learning situations and teaching goals are employed, including demonstrations of the equipment to be used, overhead projection of graphic materials, frequent reference to the interview's question-by-question specifications, mock interviews, and special presentations by the research staff to address the other aspects of the study. The interviewers are tested for mastery of the material repeatedly during the training process. In addition, the training focuses on security and confidentiality. The legal and ethical obligations of interviewers and staff are thoroughly covered. All employees sign and Affidavit of Nondisclosure as a condition of employment.

Training related to the In-Person Component

A Georgia licensed clinical neuropsychologist will administer and interpret the child assessments included in the in-person component. The neuropsychologists at the Children's Healthcare of Atlanta at Scottish Rite all have experience working with children with certain special needs, including children with SB. Project personnel involved with the informed consent process and/or data collection will participate in "mock sessions" prior to the actual start of the project. These mock sessions will familiarize project personnel with the protocol and prepare them to answer potential concerns or questions that may arise. These mock sessions are also intended to improve the flow of the protocol administration and to detect avoidable problems prior to assessing actual project participants.

Training related to Medical Records and EI Records Abstraction

Dr. Alriksson-Schmidt and Ms. Thibadeau will independently abstract the relevant information and record it on the appropriate data collection forms. The data will be compared to ensure consistency. Any case of inconsistency in the data recording will be resolved before the data is entered into the database. Dr. Alriksson-Schmidt has abstracted information from medical records in the past, and Ms. Thibadeau has more than 20 years experience of recording clinical information in medical records.

B3. Methods to Maximize Response Rates and Deal with Nonresponse

SB is a relatively rare medical condition, and with any rare condition or inclusion criteria, there is a limited pool of eligible participants. In this case, the size of the limited pool of eligible participants is unknown (i.e., the number of 3-, 4-, and 5-year-old children born with SB in the State of Georgia is unknown). We do know the number of eligible families identified through the MACDP and Children's Healthcare of Atlanta at Scottish Rite SB Clinic, but these two sources do not cover the entire State of Georgia. To supplement these two sources, we will also recruit a convenience sample from specialty doctors' offices. Thus, it is not possible to draw a statistical sample of known families with a child with SB within the entire State of Georgia and we are not proposing to draw such a sample.

Researchers are encouraged to calculate response rates based on one of the American Association for Public Opinion Research's definitions. The recruitment process in the current pilot project makes it difficult to calculate these types of response rates, however. Although it is possible to identify (in the MACDP and the Children's Healthcare of Atlanta at Scottish Rite SB Clinic databases) the number of eligible families (i.e., denominator) and how many families complete either of the components (i.e., the "eligible and interviewed" group), the major interest for this particular project, in terms of response rates, is the information captured in the "eligible and not interviewed group". Clearly, any calculation related to recruitment from the MACDP will be very much dependent on how many of the eligible families actually receive (and return) the original letter that will be mailed by the MACDP as we will not be able to contact any of the families before they have authorized the MACDP to share contact information with us. The diversity captured in the "eligible and not interviewed" group will possibly include: eligible families who did not receive the letter from the MACDP who would, or would not have enrolled; eligible families who received the letter, returned it, and did not authorize MACDP to share contact information; eligible families who received the letter, authorized the sharing of contact information but did not enroll. We will tabulate this information to the extent possible. Although it is *not* the goal of this pilot project to generate generalizable results, we are nevertheless interested to learn about the reasons why some parents choose not to participate in the project, as this information will have important implications when preparing for a future prospective study on the natural history of SB. We will attempt to ascertain the reasons why (to the extent possible) during the recruitment phase by posing this question to parents who decide not to enroll after contacting the project.

In order to maximize respondent cooperation and participation, we will offer flexible scheduling opportunities, Spanish and English versions of the telephone survey, different options for participation (in-person or telephone survey components), and possibly useful feedback (in-depth evaluation in the in-person component). In addition, the survey contains skipping patterns to reduce the time required to complete the interview.

Finally, as we have created and are testing a new survey in this pilot project we are interested in calculating item nonresponse rates. If there are certain items on the survey that are commonly skipped or answered with “don’t know,” we will consider whether the specific item will need revision.

B4. Test of Procedures or Methods to be Undertaken

This new ICR represents a proposed pilot test of procedures and methods. Thus, the procedures for collection of information described in B2 will be tested under this ICR. The proposed pilot test will be used to make significant decisions about data collection methodology and content that might be employed in future, prospective studies of the natural history of SB. Any future studies that are based on the outcomes of this proposed pilot study will be submitted for review and clearance under a separate ICR.

The survey, the medical records abstraction form, and the EI records abstraction form included were created specifically for this pilot project. As stated in B2, we consulted experts in neurosurgery, orthopedics, and urology to develop the medical records abstraction form. We also received input from an experienced EI researcher on the EI records abstraction form. Different professionals at the CDC have provided feedback on the survey. All the other measures included in this ICR are standardized measures with sound psychometric properties (as referenced in B2) that have been used in both research and clinic settings in the past. Some of the measures, such as the PEDI, were designed specifically to be used with individuals with disabilities, whereas other measures, such as the FAD and the PPVT-4, can be used with individuals with or without disabilities. Although the results from this pilot project would not lead us to change the content of any standardized measures, we will evaluate the usefulness and appropriateness of the measures that are included.

B5. Individuals Consulted on Statistical Aspects and Individuals Collecting and/or Analyzing Data

CDC Project Staff and Contractors at the CDC:

- Ann Alriksson-Schmidt, Ph.D., MSPH (Association of University Centers on Disabilities Fellow) is the CDC PI. She is in charge of planning and overseeing the project. She will also be involved with some of the in-person component data collection and will be responsible for analyzing the data generated from this pilot study. Phone: 404.498.3487; E-mail: sax3@cdc.gov

- Judy Thibadeau (McKing Corporation Contractor) RN, MN is in charge of the National SB Program and coordinates all SB related programs and research at the CDC’s Disability and Health Branch. Ms. Thibadeau will assist with the medical records review. Phone: 404.498.3559; E-mail: csn2@cdc.gov

- Mark Swanson, MD, MPH (CDC) has provided valuable input in the planning process of the proposed pilot project and will be involved when preparing and interpreting the results from the project. Phone: 404.498.3076; E-mail: cfu9@cdc.gov

- Vince Campbell, Ph.D. (CDC) has also provided valuable input in the planning stages and his expertise will be sought on an as needed basis when preparing the results of the project. Phone: 404.498.3012; Email: vbc6@cdc.gov

- Melissa Danielson, MPH. (SAIC Contractor) is a biostatistician who will advice on the statistical analyses on an as-needed basis. Phone: 404.498.3016; Email: ekd6@cdc.gov

Staff Outside of CDC:

- Kari Carris, Ph.D is a Senior Survey Director at NORC. She is serving as the NORC Project Director on this study. She has primary responsibility for the development of the data collection protocol, overseeing participant recruitment data collection for the telephone component, and writing the final project reports. Phone: 312.759.4295; E-mail: carris-kari@norc.org

- Keeshawna Brooks, MA is a Survey Director at NORC. She will assist with the development of the data collection protocol and will oversee the data collection process. Phone: 312.325.2529; E-mail: brooks-keeshawna@norc.org.

- David Marcus, Ph.D. is Georgia licensed clinical neuropsychologist with the Department of Neuropsychology at the Children’s Healthcare of Atlanta at Scottish Rite. We have collaborated with Dr. Marcus in deciding what measures would be appropriate to use to measure the constructs of interest in the pilot project. Dr. Marcus will be in charge of the child data collection of the in-person component. Phone: 404.785.2813; Email: DavidJ.Marcus@choa.org

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