Supporting Statement Part A

Congenital Heart Survey To Recognize Outcomes, Needs, and well-beinG (CH STRONG)

OMB number 0920-15BHD

Reinstatement with Change

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- **Goal of the study:** The purpose of CH STRONG is to assess barriers to care, quality of life, social and educational outcomes, and transition of care among adults born with congenital heart defects (CHD).

- **Intended use of the resulting data:** Data from CH STRONG will enable federal, state, and local governments and organizations to understand the needs of adults with CHD, allocate resources, and establish programs accordingly.

- **Methods to be used to collect:** CH STRONG is a cross-sectional surveillance project. Data will be collected once from a participant, via paper or online survey, depending on how the participant chooses to complete the survey.

- **The subpopulation to be studied:** CH STRONG will survey adults aged 18 to 45 years of age and born with a congenital heart defect as identified through the birth defects surveillance system in three participating sites in the United States. One site will be Metropolitan Atlanta Congenital Defects Program (MACDP). The two additional sites, The University of Arizona and University of Arkansas, were determined through an objective and competitive funding process.

- **How data will be analyzed:** When analyzing the data, prevalence estimates with 95% confidence intervals (CI) will be calculated for demographic and health characteristics, as well as barriers to care, quality of life, social and educational outcomes, and transition of care. Estimates will be calculated overall and stratified by site, by type of CHD, and by important demographic characteristics, such as age and sex. Prevalence estimates and associated 95% CIs will be compared to those from national, state, or local publicly available population-based surveys, such as the Behavioral Risk Factor Surveillance System. Amongst adults born with CHD, univariate and multivariable log-binomial regression may be used to determine risk factors for given outcomes, such as poor quality of life. Risk factors may include type of CHD, demographic characteristics, and healthcare use and access.

Supporting Statement Part A.

A. Justification

Section A.1. Circumstances Making the Collection of Information Necessary

This Information Collection Request is submitted under the classification "Reinstatement with change" request. The length of data collection requested for Office of Management and Budget (OMB) approval is 2 years. The National Center on Birth Defects and Developmental Disabilities (NCBDDD) at the Centers for Disease Control and Prevention (CDC) is making this request as authorized by Section 301 of the Public Health Service Act (42 U.S.C. 241) **(Attachment 1)**.

Congenital heart defects (CHDs) are the most common type of structural birth defects, affecting approximately 1 in 110 live-born children [1]. Prior to the 1970s, many CHDs were considered fatal during infancy or childhood, but with tremendous advances in pediatric cardiology and cardiac surgery, at least 85% of patients now survive to adulthood [2, 3]. There are approximately 1.5 million adults with CHD in the United States today [4], and adults with CHD now outnumber children [5, 6]. With vast declines in mortality from pediatric heart disease over the past 30 years, it is vital to assess long term outcomes and quality of life issues.

With improved survival to adulthood has come increasing co-morbidities. Studies show that 61% of 22 year-olds with CHD had not received any specialized cardiac care since turning 18 years old, and 42% of adults with CHD had at least a three year gap in care [7, 8]. However, U.S. data are lacking in regard to long term outcomes and quality of life issues of those born with CHD. Lack of knowledge concerning their CHD and lack of appropriate medical follow-up are modifiable issues that could be addressed to improve outcomes in this at-risk population [9]. As a result of their CHD, adults with CHD are at increased likelihood of having long-term problems with other organ systems, such as lung, liver, kidney, and brain [10, 11].

Beyond survival and medical outcomes, only limited data are available to address other social and quality of life issues of adults born with CHD. The majority of data come from Europe and South America and offer conflicting results with regard to educational attainment [12, 13], employment [12, 14-16], and

social relationships [12, 17]. Some studies have shown worse outcomes in comparison to the general population, but others have shown comparable or better outcomes. The scant data, mostly from other counties, is insufficient to provide insight into the public health questions that remain for adults with CHD or to develop services and allocate resources designed to improve long-term health and wellbeing.

The initial request for this project was one year, but there were delays in recruitment due to challenges with tracking and tracing individuals for correct addresses. The three sites, Metro-Atlanta Congenital Defect Program (MACDP), University of Arizona, and University of Arkansas, decided to conduct more intensive and time-consuming tracking and tracing to identify more accurate contact information for all eligible individuals and for those individuals whose materials were returned as undeliverable. At MACDP, this required modifying a contract to include the task of tracking and tracing 2,313 individuals. While the large majority of tracking and tracing at all three sites took place in the first year of the project, including that for the 2,313 individuals above, an additional 1,115 mothers of eligible individuals need to be sent a contact information form to assist to locating their child. Due to these delays and changes in the recruitment process, CH STRONG data collection is expected to last an additional 24 months and conclude two years after receiving OMB approval.

Since July 2016, the three CH STRONG sites identified 9,228 individuals with CHD through their respective birth defects registries. The CH STRONG project has successfully tracked and traced 6,417 individuals for current contact information. To date, the three sites have sent recruitment materials to 3,651 individuals (40% of all individuals).

Section A.2. Purpose and Use of Information Collection

I. How this information will be used and for what purpose:

The purpose of this survey is to collect information on barriers to health care, quality of life, social and educational outcomes, and transition of care from childhood to adulthood among adults born with CHD. Currently, Congress has appropriated approximately $4 million per year to CDC to conduct surveillance among adults with CHD.

CH STRONG will survey adults aged 18 to 45 years of age and born with a CHD as identified through the birth defects surveillance system in three participating sites in the United States. The information collected from this cohort will be used to identify the healthcare, educational, and social service needs of adults with CHDs **(Attachment 3)**. Findings will be reported through peer-reviewed publications, presentations at state and national conferences, and webinars and reports to partners who work on CHD. The findings will be used by national, state and local organizations to allocate resources and develop services and programs for adults with CHD.

II. Justification for data collection in terms of positive needs and the negative consequences of not having the information:

With the information collected in this survey, the CDC, along with its partners, will have information on healthcare needs and quality of life among a U.S. population-based group of adults with CHD. This information will inform local, state, and federal resource allocation for services targeting U.S. adults with CHD, a group that is increasing in size and currently totals over 1.5 million. Additionally, clinicians will have information to counsel families of children with CHD on how to prepare for their child's future. Without the information, needed resource allocation and services for adults and information on long-term outcomes for children with CHD are unknown.

Section A.3. Use of Improved Information Technology and Burden Reduction

All data (100%) can be collected online through a user-friendly internet-based survey using IBM SPSS software. The participant will be sent an introductory letter **(Attachment 4)**, consent document **(Attachment 5)**, and paper survey **(Attachment 3)** with additional information on how to access the survey online **(Attachment 6)**, if desired. The participant may choose their desired mode of survey completion—paper or online. The online survey has skip patterns built into the program, whereby participants are asked only questions that pertain to them, based on their sex and answers to previous questions, thus, reducing the total amount of time and effort needed to complete the survey. Additionally, the online survey saves the information from each completed page of the survey when the participant clicks to the subsequent page. Therefore, the participant has the option of completing the survey in stages, rather than all at

once. In addition, information from the birth defects surveillance system will be electronically linked by use of a unique identification number to the respondents' survey information, eliminating the need to ask additional questions already captured in electronic surveillance databases.

Section A.4. Efforts to Identify Duplication and Use of Similar Information

In 2012, the CDC convened a panel of CHD experts to discuss how to use limited resources to address major gaps in information among individuals of all ages born with CHD. One of the gaps the group identified was the need for information on access to healthcare, continuation of care from adolescence to adulthood, and quality of life among adults with CHD, since U.S. data does not exist. Additionally, the CDC holds regular calls with its partner organizations (e.g. National Heart Lung and Blood Institute (NHLBI), March of Dimes, CHD advocacy organizations) focused on CHD to inform them of our current and future work on the topic, including this surveillance project.

A study from Canada found that 61% of 22 year-olds with CHD had not received cardiology care since their 18th birthday [7]. Additionally, a clinic-based study found that 42% of adults returning to care had not been seen for care for over three years [8]. However, findings from clinic-based populations and from outside of the United States are not generalizable to the larger population of U.S. adults with CHD who may not be accessing care.

The vast majority of existing population-based national surveys (BRFSS, National Health and Nutrition Examination Survey (NHANES)) do not inquire about whether the respondent was born with CHD. Though the most common structural birth defect, CHD is a rare condition, affecting less than 1% of infants [1]. Therefore, adding a question on CHD to a current population-based survey would not provide sufficient sample size to generate precise prevalence estimates for healthcare access, quality of life, and other issues among adults with CHD. For example, from an exploratory analysis we conducted of the population-based national Medical Expenditure Panel Survey, which surveyed 8,293 individuals aged 18 to 40 years in 2013, only 8 individuals self-reported having CHD. This low number prohibits using the data to generate precise prevalence estimates and findings for the U.S. adults with CHD.

In order to have a comparison group of individuals without CHD, yet reduce the number of people surveyed, the CH STRONG survey includes validated questions from nationally-representative and state-representative population-based surveys, such as NHANES and BRFSS. During data analysis, the data from individuals aged 18 to 45 years who participated in these surveys will be compared to data collected from the CH STRONG survey.

In formulating the surveillance project and developing the survey, the CDC reached out to its partner organizations and individual experts—NHLBI, CHD advocacy organizations, March of Dimes, and pediatric cardiologists within academia—to provide feedback on the survey. All reviewers, along with the expert panel convened in 2012, supported our efforts to collect such information, since none exists from a U.S. population-based sample.

Section A.5. Impact on Small Businesses or Other Small Entities

This data collection will not involve small businesses.

Section A.6. Consequences of Collecting the Information Less Frequently

The information will only be collected once and has not been collected previously. The consequence of not collecting the information would be to have no information from U.S. population-based data sources to inform the public health needs of and resource allocation for services targeting U.S. adults with CHD, a group that is increasing in size and currently totals over 1.5 million [4].

Each respondent will be asked to respond once.

There are no legal obstacles to reduce the burden.

Section A.7. Special Circumstances Relating to the Guidelines of 5 CFR 1320.5

This request fully complies with the guidelines of 5 CFR 1320.5.

Section A.8. Comments in Response to the Federal Register Notice and Efforts to Consult Outside the Agency

A. A 60-day Federal Register Notice was published in the Federal Register/Vol. 82, No.181/Wednesday, September 20, 2017, pp 43991-43992 **(Attachment 2).** CDC did not receive public comments related to this notice.

B. From January 2014 to April 2015, representatives from several organizations **(Attachment 7)** outside of CDC were consulted and asked to review the data collection instruments for this study. Based on their review, the CDC shortened the survey and revised questions for clarity.

Section A.9. Explanation of Any Payment or Gift to Respondents

Respondents will receive a $5 gift card when sent the survey materials in the initial mailing and will receive an additional $10 gift card once their survey has been returned as a token of appreciation. Research suggests that providing tokens of appreciation to eligible participants when they receive the survey materials will increase response rates and prevent bias, making findings generalizable to U.S. adults with CHD [18, 19]. Literature examining the benefit of tokens of appreciation for participation was summarized by Yu J, et al. in their paper "A quantitative review of research design effects on response rates to questionnaires" [18]. It reviewed 497 response rates found in 93 journal articles and found that response rates increased with monetary and non-monetary gifts to participants.

Section A.10. Protection of the Privacy and Confidentiality of Information Provided by Respondents

This submission has been reviewed by the NCBDDD Privacy Officer, who has determined that the Privacy Act does apply.

This data collection effort is subject to the CDC Privacy Act System in accordance with CDC's System of Records Notice (SORN) #09-20-0136 Epidemiologic Studies and Surveillance of Disease Problems, Department of Health and Human Services/CDC/National Center for Infectious Diseases.

An Informed Consent Document will be provided to all individuals eligible to participate in the surveillance project **(Attachment 5)**. Completion of the survey will be taken as consent to

participate. Because this work presents no more than minimal risk of harm to subjects and involves no procedures for which written consent is normally required outside of the research context, we will not request written documentation of informed consent. The informed consent informs the participant about the purpose and procedures of the project. Additionally, the informed consent states that there are no known risks to the participant and all personal information will be kept private to the extent allowed under federal laws. The informed consent also states there is no benefit to completing the survey, but answers may help identify unmet needs of adults with CHD.

The CDC network used for the online data collection is a secure network. Data from the three sites' completed survey questionnaires will be stored at the CDC. The survey instrument asks for the participant's name, current residence and email address, and, for individuals who state they would like to be re-contacted for similar surveys, the name and contact information for a friend or family member, in case the participant is unreachable in the future. If the participant does not want to be re-contacted or does not answer the question, they will not be re-contacted regarding any future projects related to this survey. For the participants who are willing to be re-contacted in the future, the CDC would only re-contact them after submitting and receiving IRB approval on a separate protocol. The respondent is not required to provide information in identifiable form (IIF) to participate. If the respondent opts to provide IIF, that information will be stored separately from other survey information. All data will be kept on password-protected systems only accessible by CDC project staff. The CDC project officer, study coordinator, MACDP site lead, data managers, and data entry staff will be the only project staff with access to the IIF, which will be kept in a locked filing cabinet in a locked room or on a password protected server, if collected electronically.

The NCBDDD data manager will clean survey data, link it to de-identified birth defects surveillance data via a participant identification number, and create a de-identified dataset for use by project staff. Only the data managers, CDC project officer, study coordinator, MACDP site lead, and data entry staff will have access to the link between participant identification number and participant IIF. This information is kept on a password-protected CDC server. Project staff at funded sites may access de-identified data stored at CDC through secure data-transfer systems. Project staff will not use these data in any way except for statistical reporting and analysis; they will not share the

individual-level data with anyone; they will not attempt to use the dataset to learn the identity of any person or establishment, and they will use reasonable measures to protect all individual-level data from eye observation, theft, or accidental loss or misplacement.

Section A. 11 Institutional Review Board Approval and Justification for Sensitive Questions

Original IRB approval was granted on 8/19/2015, and the most recent IRB continuation was approved on 8/10/2017 and will expire on 8/09/2018. The current IRB approval letter is included as **Attachment 8**.

The CH STRONG survey asks questions about topics that may be considered sensitive: name, address, and contact person if participant is not reachable in the future, pregnancy history, discussions about contraception with a clinician, depressive symptoms, use of special education services, and disabilities. These topics are included in the survey because several reports indicate they are important issues for adults with CHD. As mentioned, the consent document **(Attachment 5)** states three times that participation is voluntary, nothing will happen if the person decides not to participate, that the participant may skip any question he/she does not wish to answer, and that all information collected will be kept secure. Additionally, the survey instrument also states that the participant may skip any question he/she does not wish to answer **(Attachment 3)**. There is also a statement in the introductory letter that reads: "None of your answers will be linked to your name, nor will your name ever be released as having a heart condition, having completed the survey, or having been asked to participate" **(Attachment 4).**

Section A.12. Estimates of Annualized Burden Hours and Costs

Across the three sites, there are 2,766 individuals that were tracked and traced in the first year of the project, but have not yet been recruited to participate in the survey. Additionally, mothers of 1,115 individuals will be sent a letter and contact information form to assist in reaching their child **(Attachments 16-19)**. It is estimated that half of these mothers will complete the form (n=556); 85% (n=474) in English and 15% (n=83) in Spanish. Therefore, with the 2,766 yet to be recruited, and the approximately 556 individuals that will be successfully tracked and traced through the mother's contact form, approximately 3,322 potential respondents will be mailed an introductory letter **(Attachment 4),** consent document **(Attachment 5)**, and survey **(Attachment 3).** It is expected that approximately 70%, or 2,325 respondents, will participate.

The survey will be conducted 1 time only and survey completion will take 20 minutes. It is expected that the Spanish and English contact information forms will take 2 minutes to complete and will only be completed once. Therefore, based on the combined two years of data collection for this extension, the estimated total annual burden hours for all individuals and mothers is 564 hours.

There are no costs to respondents other than their time.

BURDEN TABLE:

A.12.A. Estimated Annualized Burden Hours

Type of Respondents	Form Name	No. of Respondents	No. Responses per Respondent	Average Burden per Response (in hours)	Total Burden Hours

Individuals aged 18-45 years who were born with a congenital heart defect	Survey questionnaire	1,661	1	20/60	554
English-speaking mothers of respondents	Contact Information Form – English	237	1	2/60	8
Spanish - speaking mothers of respondents	Contact Information Form - Spanish	42	1	2/60	2
TOTAL		**1,940**	**—**	**—**	**564**

The annualized cost burden is shown in Table A.12.B. The median hourly wage rate is based on the most recent (May 2011) National Occupational Employment and Wage Estimates for all occupations, published on the Bureau of Labor Statistics website which is $17.81. See http://www.bls.gov/oes/current/oes_nat.htm.

A.12.B. Estimated Annualized Burden Costs

Type of Respondents	Form Name	Total Burden Hours	Hourly Wage Rate ($)	Total Respondent Costs ($)
Individuals aged 18-45 years who were born with a congenital heart defect	Survey questionnaire	554	17.81	9,866.74

English-speaking mothers of respondents	Contact Information Form – English	8	17.81	142.48
Spanish - speaking mothers of respondents	Contact Information Form - Spanish	2	17.81	35.62
TOTAL		**564**	**—**	**10,044.84**

Section A.13. Estimates of Other Total Annual Cost Burden to Respondents or Record Keepers

There are no costs to respondents associated with either capital and startup efforts or operation and maintenance of services for this project.

Section A.14. Annualized Cost to the Government

The average annualized cost to the Government to collect this information is $78,000. Costs for tracking and tracing in the MACDP sample was paid to a contractor in fiscal year 2016. Government costs paid to the University of Arizona and University of Arkansas occurred in fiscal year 2015. The two universities are currently on no-cost extensions. The government costs paid to the March of Dimes occurred in fiscal years 2015 and 2016. No additional costs to the government are anticipated in the extension period beyond personnel costs detailed below.

		Percent time	**Total ($)**
Federal Government Personnel costs	CDC Project Officer/Project Lead	0.2	22,000
	MACDP/CDC Site Lead	0.1	22,000
	CDC Project Coordinator	0.15	12,000

	CDC Epidemiologist	0.05	5500
	CDC Medical Officer	0.05	5500
	CDC Medical Officer	0.05	5500
	CDC Epidemiologist	0.05	5500
Total			78,000

Section A.15. Explanation for Program Changes or Adjustments

This is a reinstatement with change of an existing data collection. The sites decided to conduct more intensive and time-consuming tracking and tracing to identify more accurate contact information for all eligible individuals. The sites also recruited individuals in batches over the year, rather than at a single time. A large proportion of data collection was accomplished in the first year of OMB approval.

Section A.16. Plans for Tabulation and Publication and Project Time Schedule

Upon OMB approval, survey materials, which include the introductory letter, survey questionnaire, consent document, $5 gift card, and pre-addressed, stamped envelope, will be mailed by the three birth defects surveillance sites to the remaining eligible individuals. Mailings will be done in batches of 50-200 individuals every 2 to 4 weeks and will follow the recruitment protocol including two reminder postcards and survey materials without incentive.

A.16.—Project Time Schedule		
Activity	**Timeframe**	
Identify and Recruit Participants	Send survey materials (letter, questionnaire, consent document, gift card, pre-addressed, stamped envelope)to eligible participants	1–18 months after OMB approval
	Reminders for eligible participants	6-24 months after OMB approval

	who have not completed questionnaire via reminder postcard **(Attachment 9)** and telephone calls **(Attachment 10)**	

When analyzing the data, prevalence estimates with 95% confidence intervals (CI) will be calculated for demographic characteristics, type of CHD, and important outcomes of interest, such as quality of life. Prevalence estimates will be calculated overall, and stratified by site, type of CHD, and important demographic characteristics, such as age and sex. For certain questions, prevalence estimates and associated 95% CIs will be compared to those from national, state, or local publicly available population-based surveys, such as the Behavioral Risk Factor Surveillance System **(Attachment 11).** Amongst adults born with CHD, univariate and multivariable log-binomial regression may be used to determine risk factors for given outcomes, such as poor quality of life. Risk factors may include type of CHD, demographic characteristics, and healthcare use and access.

Section A.17. Reason(s) Display of OMB Expiration Date Is Inappropriate

The display of the OMB expiration date is appropriate, no exception is sought.

Section A.18. Exceptions to Certification for Paperwork Reduction Act Submissions

There are no exceptions to the certification.