Journal of the American Heart Association

SPECIAL REPORT

Operational and Ethical Considerations for a National Adult Congenital Heart Disease Database

Elisa A. Bradley, MD; Abigail Khan, MD; Demetria M. McNeal, PhD, MBA; Katia Bravo-Jaimes, MD; Amber Khanna, MD; Stephen Cook, MD; Alexander R. Opotowsky, MD, MMSc; Anitha John, MD, PhD; Marc Lee, MD; Sara Pasquali, MD; Curt J. Daniels, MD; Michael Pernick, JD; James N. Kirkpatrick, MD; Michelle Gurvitz, MD

ABSTRACT: As more adults survive with congenital heart disease, the need to better understand the long-term complications, and comorbid disease will become increasingly important. Improved care and survival into the early and late adult years for all patients equitably requires accurate, timely, and comprehensive data to support research and quality-based initiatives. National data collection in adult congenital heart disease will require a sound foundation emphasizing core ethical principles that acknowledge patient and clinician perspectives and promote national collaboration. In this document we examine these foundational principles and offer suggestions for developing an ethically responsible and inclusive framework for national ACHD data collection.

Key Words: ACHD ■ Big Data ■ ethics ■ quality

The global birth prevalence of congenital heart disease (CHD) is almost 1% of live births.[1] Cardiac surgery, transcatheter intervention, and advanced medical care have improved survival, and as a result, adults with CHD have been estimated to outnumber children with CHD since approximately the year 2000.[2,3] As of 2010, the number of adults with CHD in the United States was estimated to be 1.4 million[4]; however, this is extrapolated from administrative data sets. These data sets are influenced by discrepancies in access to specialized care, correct coding practice, resource availability, health insurance coverage, and other limitations inherent to such data sets (Figure 1).[5–7] However, as this population continues to grow, we must understand better the long-term complications and comorbid diseases faced by patients with adult congenital heart disease (ACHD). In so doing we will be able to identify opportunities to improve care and extend survival. Accurate and timely comprehensive data must be available to support both research and quality-based initiatives.[4] However, the ACHD population comprises patients with heterogenous anatomy and physiology, representing a diverse set of uncommon diagnoses. Therefore, data accrual must take place from multicenter, preferably national, collaborations.

Large-scale clinical and/or administrative data collection will require a solid foundation and infrastructure based on strong ethical principles and consideration of both patient and clinician perspectives. As the field of CHD seeks to broaden the understanding of late congenital heart disease, adult outcomes, and quality-based initiatives, meaningful understanding of the ethical principles in data sharing is required. We recognize there are many ethical constructs, but for the purposes of this work, we are focusing on widely accepted principles of research ethics: respect for people, beneficence, and justice.[8] To begin organizing the conceptual and ethical infrastructure critical

The opinions expressed in this article are not necessarily those of the editors or of the American Heart Association.

Correspondence to: Elisa A. Bradley, MD, Division Cardiovascular Medicine, Milton S. Hershey Medical Center, 500 University Drive, Hershey, PA 17033.
E-mail: ebradley3@pennstatehealth.psu.edu

For Sources of Funding and Disclosures, see page 11.

JAHA is available at: www.ahajournals.org/journal/jaha

Nonstandard Abbreviations and Acronyms

ACHD	adult congenital heart disease
PCOR	patient-centered outcomes research

to understanding ACHD-based data and clinical outcomes, and to provide a platform for future data collection, benchmarks and initiatives, we will (1) evaluate ethical principles in large-scale data collection for the population of patients with ACHD,[9] and (2) discuss strategies that promote sound governance, avoid bias, and promote national collaboration.

CURRENT ACHD DATA SETS IN THE UNITED STATES

Administrative and Clinical Databases

In the United States, there is no comprehensive ACHD data collection tool or database that collates outcome-based data. This presents a challenge for observational research and assessment of quality of care, because these activities depend on longitudinal surveillance programs and comprehensive cohorts.[10] Currently, patients with ACHD in the United States are identified through 2 primary sources: administrative databases and clinical databases or registries (Table 1). Administrative data bases typically rely on health care claims-based information and only include patients who are able to access health care. The benefit of this type of Big Data[11] is that it has the capacity to aggregate and cross-reference a large population,[12] in addition to offering the opportunity to identify small patterns or connections within a subset of the larger data set.[13]

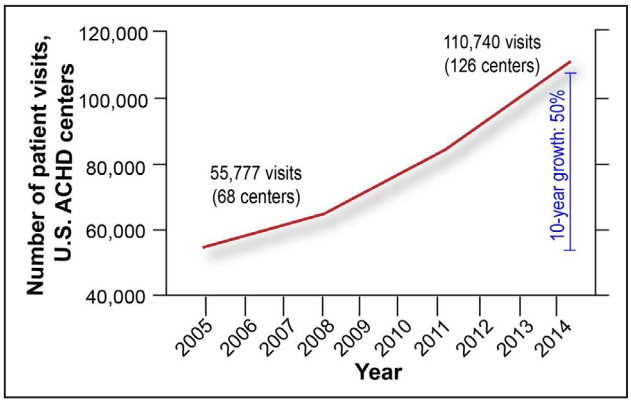

Figure 1. ACHD visits at North American centers.
The number of annual visits to US adult congenital heart centers has increased steadily in the decade between 2005 and 2014. Shown here are self-reported center data documenting an almost 100% increase in the annual number of visits of patients with ACHD during this timeframe, demonstrating the growing need for a national database that can support data-driven research and quality programs that span across multiple US centers. Data derived from Krasuski and Bashore[7] and Avila et al.[6] ACHD indicates adult congenital heart disease.

Big Data in the area of ACHD may advance medical knowledge in disease diagnosis, treatment, and prevention.[14] The validity of these data sets remain dependent on the accuracy of the diagnostic codes, which perform best in those with moderate and complex forms of CHD, in younger patients, and in those seen by an ACHD specialist.[15] Disease heterogeneity remains an important limitation, because administrative data sets often lack detailed historical and clinical data, and thus fail to account for the high degree of variability in surgical repair, interventional procedures, physiologic stage, and disease complexity.[16] The main strength of administrative data sets is the large size and potential to integrate data across health care systems and payers. Administrative data sets are often best at measuring the magnitude of health care use and cost, but they struggle to measure detailed clinical information. Recent efforts to combine and/or synthesize data from administrative sources to perform population-level surveillance of ACHD are an initial step forward, but significant challenges remain,[5] for instance addressing health disparities in this type of database. Although incorporation of demographics and social determinants in the electronic health record is recommended as a potential tool to reduce disparities in future studies, these recommendations are not yet widely integrated in the electronic health record.[17]

Historically, most ACHD research in the United States has been performed using single-center clinical data. The main strength of clinical data is the ability to store more detailed patient-level data, which may allow for improved disease-specific characterization and a nuanced understanding of outcomes. However, the primary limitations in single-center ACHD clinical data sets are the relatively small size, lack of appropriate identification across health systems, and the challenge of data harmonization (both input and output). Importantly, this is a theoretically surmountable obstacle, because participation in national registries is now near universal for pediatric congenital heart surgery and interventional procedures.[18] However, crossover to the adult cohort has not taken place to a significant degree (Table 1).

Gaps in the Currently Available Data

Despite the progress in use of cohorts and administrative data, there remain significant gaps in our ability to study and track CHD across the lifespan. For instance, the current estimate of US ACHD disease burden is derived from international data,[4] emphasizing one of the glaring deficiencies in understanding the needs

Table 1. Selected List of Large Databases Including Patients With Adult Congenital Heart Disease in the United States

Name	Brief description	Major strengths	Major weaknesses
Administrative databases			
National Inpatient Sample	Stratified ≈20% sample of all discharges from US hospitals, excluding rehabilitation and long-term acute care and federal institutions (ie, Veterans Association and Indian Health Service)	Geographic diversity; multicenter; publicly available; large size: >7 million hospital stays/year; payer independent (all nonfederal)	Inpatient only; visit based not patient based; inability to identify states and/or hospitals; codes not validated
IBM MarketScan Database	Employers and health plan, including inpatient, outpatient, and pharmacy care	Includes inpatient and outpatient care; large size: >245 million unique patients; longitudinal; includes multiple payers	Convenience sample; insured patients only; claims based; codes not validated
State All Payer All Claims database	Vary among states; usually includes medical and pharmacy claims from private and public payers; reported directly from insurers to states and administered by the state	Include inpatient and outpatient care; regional specificity; longitudinal within state; relatively comprehensive of care delivered	State-by-state variability in administration and management; claims based; not available in all states; codes not validated
Nationwide Emergency Department Sample	National sample of hospital-owned emergency department visits	Large size: 30 million visits annually (unweighted)	Emergency only visits; claims based; codes not validated: unknown accuracy of codes in ED setting
Transformed Medicaid Statistical Information System	Centers for Medicare and Medicaid Services administered multistate database included claims and managed care data from state Medicaid programs	Large site; multistate (nearly all states participating); inpatient and outpatient care	Claims based; Medicaid specific; codes not validated
Registries and multicenter clinical databases			
Improving Pediatric and Adult Congenital Treatments Registry	Includes demographic and clinical information on a subset of interventional catheterization procedures	Multicenter; allows benchmarking on outcomes and quality of care	Biased sample of specific participating sites; procedure specific; limited data collected, variable accuracy; benchmarking for quality limited by ability to risk adjust; many variables unvalidated
Organ Procurement Transplant Network Database	All national data on waiting list, donation, and transplantation outcomes	Includes all transplant centers in the United States; allows benchmarking on outcomes and other metrics	Limited to organ transplant; minimal information on CHD anatomy and complexity; codes not validated
Society of Thoracic Surgeons Congenital Heart Surgery Database and Adult Cardiac Surgery Database	Procedural database specific to congenital cardiac malformations	Granular clinical data; allows benchmarking on outcomes and quality of care; manual data entry and audits	Short-term outcomes only
National Cardiovascular Data Registry ICD registry	National database of ICD and CRT-D procedures	Allows benchmarking on outcomes and quality of care	Lacks detailed information on CHD disease complexity; short-term outcomes only; unclear how well CHD is coded
Pediatric Cardiac Critical Care Consortium	Primarily North American centers, data on patients (medical and surgical) hospitalized in the cardiac ICU	Supports research, benchmarking, and quality initiatives	Focused on ICU care and outcomes; limited to those admitted at participating pediatric cardiology centers
Pediatric Acute Care Cardiology Collaborative	Multicenter data on patients hospitalized in a participating pediatric cardiology center general cardiology/stepdown unit	Supports research, benchmarking, and quality initiatives	Limited to patients with CHD admitted at participating pediatric cardiology centers
Cardiac Networks United	Collaboration across several networks and registries (PC4, PAC3, NPCQIC, ACTION, CNOC, Pediperform, CCRC)	Facilitates data sharing across participating registries to support analyses across phases of care, shared resources to support QI activities	Limited to those cared for at a participating pediatric cardiology center (mostly larger, academic)

ACTION indicates acute coronary treatment and intervention outcomes network; CCRC, Congenital Cardiac Research Collaborative; CHD, congenital heart disease; CNOC, Cardiac Neurodevelopment Outcome Collaborative; CRT-D, cardiac resynchronization therapy-defibrillator; ED, emergency department; ICD, implanted cardiac defibrillator; ICU, intensive care unit; NPCQIC, National Pediatric Cardiology Quality Improvement Initiative; PAC3, pediatric acute care cardiology collaborative; PC4, Pediatric Cardiac Critical Care Consortium; and QI, quality initiative.

of US-based patients with ACHD. Current US clinical data collection is not equitable or equal, because it is often directed to patients receiving care at regional ACHD centers, and it comprises a self-selected group impacted by geography, socioeconomics, insurance access, health literacy, and other factors that propagate disparities in health care.[19,20] It is therefore critically important that strategic and targeted efforts be made to ensure universal access/representation for all patients with ACHD in the United States. Therefore, this important point becomes the cornerstone upon which US-based ACHD investigators have begun to consider the building blocks for a comprehensive US ACHD database. Importantly, this initiative will aim to include groups that may have been historically underrepresented in ACHD research, including, but not limited to rural populations, the elderly, poor, non-English speakers, the uninsured, undocumented immigrants, and people of color.[19,21]

Diversity, Equity, and Inclusion in a National ACHD Database

Historically, large-scale data collection has not been inclusive in part because of segregation and racism within the medical profession and the greater health care system, which has particularly impacted the Black community.[22,23] Thus, it is not surprising that >50% to 75% of contemporary randomized controlled trials of heart failure and acute coronary syndromes, respectively, did not report racial and ethnic distributions of their enrolled participants.[24,25] When they are reported, disease-to-prevalence ratios demonstrate the disproportionately low participation of underrepresented racial and ethnic minority groups.[26] Reasons for this include narrow inclusion criteria, selective site recruitment, as well as participant mistrust, time, and resource constraints, and lack of comfort, information, or awareness.[27] Clinical research under these conditions contributes to the denial of effective treatment to groups who might benefit, raises questions about the external validity of study results, and widens health care disparities.

These disparities are largely driven by social determinants of health, including economic stability, neighborhood and physical environment, education, food, community, structural racism, and health care system.[28] Racial disparities in cardiovascular care in the United States have been well documented in multiple domains. Specifically, Black individuals have a higher prevalence of less-than-ideal cardiovascular health (especially in blood pressure and healthy diet score domains in both children >12 years and adults),[29] experience overall increased readmission rates when treated for myocardial infarction[30] or heart failure,[31,32] and are less likely to receive cardiac resynchronization[33] or advanced heart failure therapies.[34] Additionally, Black mothers are 3 times more likely to die from pregnancy complications, of which 30% are related to cardiovascular disease.[35]

Although existing national pediatric databases report racial disparities in CHD more broadly,[36,37] analyses often focus on the first years of life, and to date, only 1 study has highlighted the existing racial and ethnic disparities in CHD-related mortality among children and adults in the United States.[38] This study demonstrated that despite the steady decrease in CHD-attributed mortality in the United States among all races and ethnicities and both sexes, over the past 20 years, pediatric and adult Black individuals have continued to experience higher age-adjusted mortality compared with their White counterparts.[38] Similarly, a sex disparity disproportionately affecting men is also present. Gaps in care attributed to lack of or insufficient health insurance among patients with ACHD may influence these outcomes as they do with morbidity.[20,39] Barriers to insurance access caused by defining CHD as a preexisting condition and/or total lifetime dollar benefit caps were diminished with the Affordable Care Act, and this was reflected in the increasing insurance coverage among hospitalized patients with ACHD after full Affordable Care Act implementation.[40] Although this represents a start toward more equitable care, persistent disparities involving transition ages (18–25 years) and Hispanic individuals remain.[40]

The current landscape of disparities in ACHD is still unfolding, and future studies linking sociodemographics, specific congenital heart lesions, and clinical outcomes require investigation. Nevertheless, from the outset, the goal should be to increase participation and representation from a diverse group, highlighting equity and inclusivity. This can be supported with community engagement to help form partnerships and collaborations that will help enhance inclusivity and participation. By engaging diverse groups, new expertise and recruitment strategies may be used to help narrow the gap, such that accessibility and the applicability of data extends to all groups, including those traditionally underrepresented.

Practically, we need to create a broad national ACHD database that could integrate data coming not only from accredited ACHD centers but also from community, rural, and safety-net hospitals. This is supported by the ethical principles of beneficence and justice as well as the fact that the most vulnerable populations (uninsured adults, those belonging to households below the federal poverty level, and adults with less than college education) are more likely to live >6 hours away from a mid- to high-volume ACHD center.[19] As we continue to uncover disparities in ACHD care and outcomes, methods to reduce the impact of disparities will become clearer, including ways to ensure

participation of diverse patients in clinical studies, to leverage technology to reach rural communities, to enhance patient navigation, to enact policy changes, and to amplify and diversify the ACHD workforce.

Technological and Other Barriers to Developing a National ACHD Database

There are foreseeable challenges that will arise in the development of a national ACHD database that include funding, engagement of health systems, and technological support (Table 2). Broadly, the initial priority when building the foundation for a national ACHD database will be to ensure that patients are correctly identified and that disease is classified accurately. At the outset, disease heterogeneity and complex coding algorithms have limited the ability to improve input data harmonization. For instance, the current absence of a universal patient identifier in the United States makes it difficult to identify, merge, and track patient data between systems. The *International Classification of Diseases, Ninth Revision and Tenth Revision* (*ICD-9* and *ICD-10*) coding systems are often nonspecific or erroneous when used to identify adults with CHD.[15,41–43] The International Society for Nomenclature of Pediatric and Congenital Heart Disease (IPCCC.net) has mapped codes across algorithms, with the anticipation that new universal coding will be available in the *International Classification of Diseases, Eleventh Revision* (*ICD-11*). In the interim, additional methods such as the *Systematized Nomenclature of Medicine Clinical Terms* (*SNOMED-CT*) will likely provide greater ACHD diagnostic specificity. In the initial ACHD database consideration, complementary search strategies will be required to correctly identify disease and data points for collection, and therefore the development of a data dictionary will be needed to facilitate data linking across sites.[44] It is likely that artificial intelligence will prove to be important in data harmonization, including standardizing input harmonization, but also in mapping and unifying output (output harmonization).

ETHICAL CONCEPTS FOR DATA SCIENCE IN LOW-PREVALENCE DISEASE

With respect to ethics in data science, 11 themes have been identified as critical to a framework to guide ethical assessment and governance of Big Data practice (Figure 2).[9] Several of these can directly inform the development of an ACHD database in the United States, including[9] informed consent, privacy, objectivity, ownership, and epistemology as it refers to the source of knowledge (distinguishing opinion from justified belief). These principles particularly impact the custodianship and participant levels, and so we review them in this context here.

Research using data registries is, in most cases, considered human subjects research and thus is governed by regulations including those determined by the Common Rule of the Department of Health and Human Services.[45] In addition, privacy rules are further governed by the Health Insurance Portability and Accountability Act of 1996[46] and modified by the Health Information Technology for Economic and Clinical Health Act in 2009.[47] These federal privacy rules help define a foundation with which entities such as health

Table 2. Main Barriers to Establishing a National ACHD Database in the United States

Barrier type	Current status	Potential solutions
Funding	• Limited funding sources to support database/registry development	• Identify a consistent funding source to develop and maintain a national database
Engagement	• Focus on clinician productivity limits ability of ACHD practitioners to dedicate time to registry development/enrollment • Patient advocacy organizations such as the Adult Congenital Heart Association provide an ideal platform to increase patient awareness of efforts	• Identification of a funding source to support time and effort for registry work as well as the time and effort of associated staff • Harness existing organizations to promote initiatives to patients • Creation of national incentives to improve and maintain quality in ACHD care
Technical or logistical	• No universal patient identifier upon which to link existing data sets • Privacy concerns limit ability to share data across health systems • Differing methods of aggregating/storing data across health systems (systems do not “talk” to one another) • Limited accuracy of administrative data • Accuracy in community-based samples is unknown • Heterogeneity of CHD phenotypes limits comparison without detailed clinical data • Historical focus on academic medical center populations limits understanding of diverse patient populations	• Development of a universal patient identifier • Patient-initiated registry participation to increase the breadth of participation • Develop methods of merging data from disparate electronic medical record systems • Support for continued research into accuracy of administrative data • Merging of clinically oriented and administrative data sets to increase level of available detail • Patient and clinician outreach to aim to capture patients not receiving care at tertiary centers (inclusion of community and safety-net hospitals)

ACHD indicates adult congenital heart disease; and CHD, congenital heart disease.

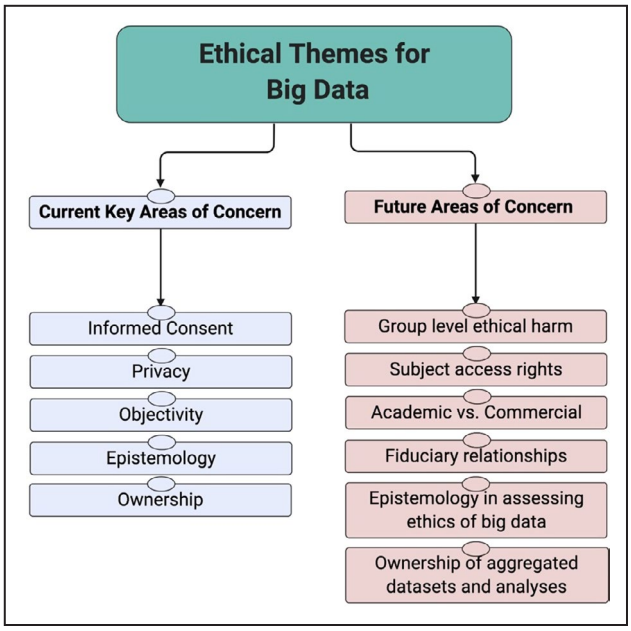

Figure 2. Ethical themes in data science.
Ethical themes suggested as current and future areas of concern as they apply to biomedical data sets. The current key areas of concern have been well described and debated in the existing literature. Potential future areas of concern have not yet attracted extensive debate in the existing literature, but are likely to require careful examination in the near future.[9] Created with BioRender.com.

Table 3. Principles of Sound Governance and Potential Application in Big Data

Principles of sound governance	Description (as it may apply to Big Data)
Tell the truth	Be honest and transparent
Be faithful and fair	Maintain ethical standards and remain consistent in adherence to the principles of governance set forth for the data set, participants, and custodians
Insist on accountability	Participate and encourage evaluation and reevaluation of the governing structure, policies, and outcomes, with an emphasis on resolving problems
Respect the governed and the government	Be open, honest, and transparent to the needs of participants and the governing body
Govern with humility	Acknowledge the limits and boundaries of the governing body, evaluating effects, and not necessarily intentions
Serve the governed, not the systems and people of government	Principles should come before custodial members and individual participants so as to reduce disproportionate influence of stakeholder groups (ie, the effect on all people must be considered)
Acknowledge the nature of government	Governing bodies seek to accomplish an agenda but are managed by individuals who have their own goals and incentives. Therefore, a careful balance must be acknowledged between the custodianship and participant groups

Modified[50] and expanded upon as the principles of sound governance may apply to Big Data

plans and health practitioners and their associates can use, disclose, and share patient information for appropriate purposes including research. These privacy rules can be further subject to more stringent rules from states or other entities.[48] However, not all database activities will be covered by these regulations. As a result, it is critical that any ACHD database include its own commitments to ethics to promote universal access and representation.

Custodianship Level

Governance

Governance refers to processes and oversight, and mechanisms of governance in the case of an ACHD database may include review councils, committees, and/or an elected board that act as custodians entrusted to manage the data and operations. The concept of sound governance[49] implies that the societal norms in a diverse population should favor balance and minimize bias, thereby promoting value to all groups. The bulk of research and writing in the topic area of sound governance applies to governmental function.[49,50] However, we can learn and apply similar principles to quality and research initiatives as they apply to an ACHD database (Table 3).

To simplify sound governance as it applies to data science, at least 2 major overarching ethical goals have a direct impact, usefulness and justice. Usefulness refers to maximizing societal benefit, considering benefit versus harm. Justice implies that individuals should be treated fairly, with equitable allocation of resources. Justice in data science means equal access to participation as well as fair access to the resulting data and application of the data in a way that does not cause unfair consequences. Therefore, we propose a set of principles applicable to future ACHD databases (Figure 3).

Objectivity and Data Ownership and Use

Among the custodianship-level guiding precepts for governing data registries for ACHD are objectivity and data ownership and use. The definition of objectivity in data collection is to strive to avoid bias in all aspects of research such that a method of data collection must always come to the same result, regardless of who ascertains the data.[9] Data ownership and use are not only about property law and rights of the researcher, but must take into consideration respect for people, beneficence, and justice, particularly impacting privacy and rights of those who contribute data but also the ethical implications of restricted access to data and the impact of data use (or misuse).[8]

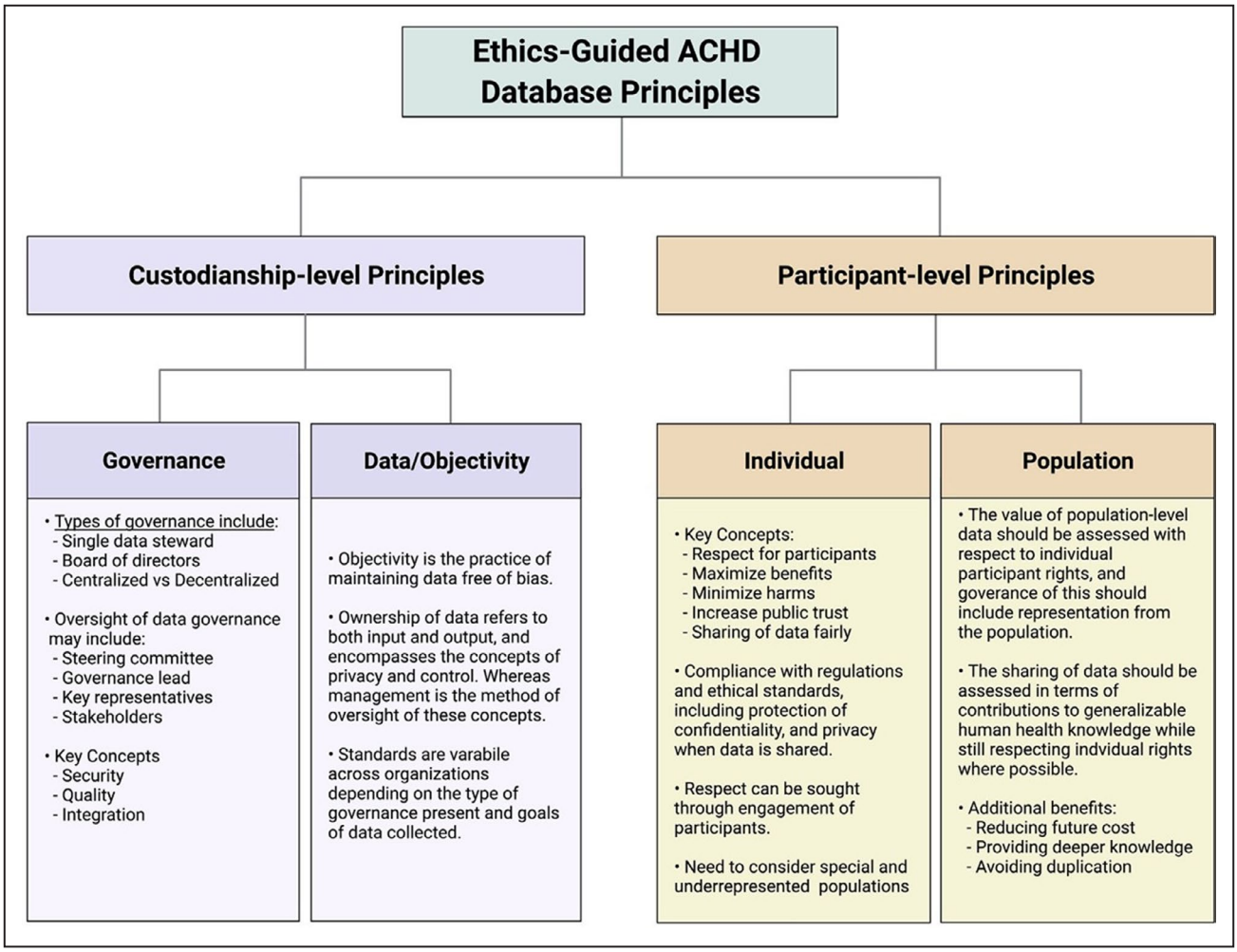

Figure 3. Principles to consider in developing a shared national ACHD database.
Considering current and future ethical themes in data science,[9] and honoring the principles of usefulness and justice, the figure depicts custodianship-level principles and participant-level principles for evaluating the initial ethical framework for a national ACHD database. ACHD indicates adult congenital heart disease. Created with BioRender.com.

Big Data research does not typically benefit individuals but may provide societal benefit. Respect for people may obligate informed consent of the participant, frequently including consent to the original use as well as future projects. A waiver of such consent may, however, be reasonable if the data are not identifiable, and results are expected to provide societal good (beneficence) but do not obviate the need for data security, particularly protection of potential identifiers. Furthermore, data collected should be limited, and there should be a justifiable reason for each item of information collected, but particularly in relation to identifiers and sensitive information. This can present difficulties in data sets that aim to be comprehensive and to anticipate future research needs.

As discussed above, participant inclusion should be fair and equitable, with every attempt made to avoid systemic bias and improper exclusion. This can present difficulties, because respect for people requires that individuals may opt out of participations, even when participation may be beneficial to society and may promote justice. Challenges to recruiting/including underrepresented groups, however, does not justify the a priori exclusion of individuals from these groups. Rather, they highlight the importance of transparency, particularly in educating individuals (where possible) and the public as to privacy protections, data usage, and benefits of research findings from registries.

Ownership of patient information in data registries has not been consistently addressed in the research literature, and there are no consensus policies on application and legal regulations. Control over the use of the health information pertains not only to data storage but also analysis and dissemination/publication.[51] Multiple views have been promulgated in the literature, ranging from the idea that all health data should be in the public domain[52] to the perspective that health data should be the exclusive property of the individuals who

contributed the data.[53] It has been suggested that data ownership considerations should be separated from considerations of data privacy.[54] In relation to health information in data sets, ownership and management are particularly complicated because individual records currently may be owned by governments, health plans, clinician entities, and/or others. For larger, multistakeholder data sets, there are variable state laws and regulations, depending on where the data were originally collected. This underscores the importance of data-use agreements and addressing ownership issues proactively. There is little research in ownership of personal data for large data sets and registries, and these areas are yet to be legally tested. Practically, the focus remains on privacy. Given the lack of consensus standards surrounding property rights in collection of individual health information, it is critical to abide by the Common Rule and pertinent local privacy rules.

Health equity and researcher diversity in data science are not new concepts, evidenced by the National Institutes of Health's Artificial Intelligence/Machine Learning Consortium to Advance Health Equity and Researcher Diversity program, which targets to increase participation and representation of both researchers and communities underrepresented in electronic health record–driven data projects. In the design of a national ACHD database, we propose a diverse and inclusive governing board, including patients, clinicians, researchers, ethicists, and underrepresented groups, to oversee data and monitor objectivity (Figure 3). This group will be tasked with vetting decisions about data points to be collected, addressing privacy concerns, ensuring data integrity, defining data ownership, and adjudicating decisions about data reporting.

Participant-Level

Individual Rights

Ethical principles are subject to individual interpretation, and seemingly clear-cut principles are often ambiguous when applied in practice. Fundamentally, in data science, individual rights are balanced by the value of generalizable knowledge for societal benefit. An individual's participation in a data set may impact their own privacy but also the privacy and autonomy of relatives, with or without awareness. A prime example is genomic-based research. Given how strongly autonomy is valued in the United States and many other nations, one might expect this consideration to provide a major obstacle to large-scale research. Newer frameworks are needed to balance the potential power of evolving technology (including Big Data) to improve health outcomes against harms, including privacy breaches and other threats to autonomy. If we do not consider individual versus societal rights, we risk making arguments based on an overly optimistic expectation of gain and a lack of imagination about harms, a pattern that has been replayed throughout history.[55]

Respect for individuals (or respect for people) is a fundamental bioethical principle that implies asking permission to perform research after an understandable explanation. The investigator cannot know what will be important to each participant, and an overly long informed consent document does not advance true understanding. The informed consent process explicitly encourages questioning and open-ended dialogue between investigators and potential participants. Individuals with idiosyncratic or otherwise unanticipated viewpoints and concerns are thereby able to understand whether participation will align with their personal beliefs. Conversely, one may argue that a person consenting to medical care is ostensibly benefiting from the consent of others and should be assumed to subscribe to a pact to provide benefit to future patients, at least when there is little or no risk. This provides a potential for coercion or overreach, and this approach must be coupled with strong regulatory structures and sound governance to minimize harm.

That said, the structure of certain Big Data research endeavors may run counter to the right of an individual to choose whether to participate. A subset of considerations that differs between the use of Big Data and standard hypothesis-driven research projects are outlined in Table 4.

Societal Rights: Specific Risks With Big Data

Arguments for more lax rules governing the use of Big Data often point to a low absolute risk of harm. However, this is not a certainty, and although there are likely many benefits to the use of Big Data in ACHD, there may also be drawbacks in relation to society as well as to individuals. There is a high risk of bias, unmeasured confounders, and inaccurate classification. Big Data interpretation often involves the analysis of large volumes of data initially collected by clinicians or others without consideration for the question under study and performed by scientists who do not necessarily understand the relevant validity of underlying data. These issues are compounded by the fact that statistical significance may not reflect clinical significance. Therefore, it is important to address the issue of epistemology (to truly understand data and how data contribute to knowledge), whereby to maximize benefit and minimize harm, there should be a broad and deep review of underlying data and consultation with content experts and those involved in the process of collecting the data (ie, depending on the project, patients, clinicians, billing specialists, insurance providers, electronic medical record [EMR] software engineers) before

Table 4. Considerations Unique to Big Data

Important concerns	Considerations
Minimize/eliminate interaction with participants and limit the capacity to decline participation	• Truly informed consent requires enormous resources, yet may be impractical in Big Data registries • Consider collecting data without identifying information to avoid ethical/privacy concerns • Consider avoiding linkage of patient information within the data set • Inclusion of patients with rare disease in decision-making about individual vs societal benefit, as the population may be the most at risk for privacy concerns, and discretionary judgement will be required
Linkage between data sources	• Recognize that the value of data is enhanced when coupled with additional information • Linked data may pose little risk to individual rights, such as fixed geographic information (such as zip code) linked to environmental variables • Recognize that in other cases, identifiable information may be shared and improve accuracy (avoid double counting a patient receiving care at >1 center) • Linking data sources may expose decisions a patient wishes to keep confidential (ie, receiving care at >1 center)
Linkage between individuals	• Big Data may involve connecting data from a single individual to others, such as family members • Any linked participants may have no vested interest in the question/disease under study
Respect for people	• Respect for people implies that using Big Data does not mislead those that make decisions based on the findings • For quality initiatives, it may be the clinician rather than the patient who constitutes the individual under study • Practitioners or health systems may reasonably be opposed to be included in such research even if their identity is confidential • Historical performance may not reflect changes made in response to negative outcomes and may not adequately adjust for risk • Data may reflect risk attributable not only to quality of care, but to forces outside of the control of individual health systems (ie, sociodemographic characteristics) • Data may not be accessible to patients/families in a way that provides useful assistance with decision making, but rather, may mislead end users into thinking they are making a more informed choice • Considerate review of how to best communicate data to health systems, patients/families, and policymakers will be required to ensure that accurate data are available

project initiation. Importantly, token participation by patients and clinicians may prove inadequate. A scientific process should be applied to whether a proposed method of review and quality control is adequate to identify underlying problems, a process that requires the explicit definition of anticipated threats to validity. Independent review should be part of the iterative process of ensuring Big Data research accurately reflects the issues in question. Additionally, system-level processes should be closely examined to mitigate factors that promote structural racism and influence data quality and broad applicability.[56]

The burden of evidence rests heaviest upon those hoping to use data, with the most prominent concepts among them being the respect for people. For data collected and intended for research purposes, this involves consideration of the level of protected health information collected and how the data are stored, most often requiring informed consent. The same may or may not be true for quality data, where the purpose is internal evaluation, and use of data to make changes, with the goal of improving quality of care for all patients, and the resulting informed consent process.

ACHD DATA SETS: THE PATIENT PERSPECTIVE

In considering the patient perspective, perhaps one of the most important ethical principles is respect for people. Many patients with chronic disease, such as CHD, must transition into adult-centered care, and in the process move from being passive recipients, to actively managing their own health care.[3] As unprecedented numbers of patients with CHD survive into adulthood, health care decision makers are seeking information to help guide their health care choices. In recent years, patient registries have been used to provide this information for other chronic disease states. As patients and caregivers consider participation in registries and data sets, it is important to maintain respect for the individual in the context of seeking answers that apply to and can advance care for all. The focus on availability of real-world evidence with a patient-centered approach has become the basis of improving clinical care.[57] Patient-centered outcomes research (PCOR) is defined as "research that addresses the questions and concerns most relevant to patients."[58] With the growing demand for PCOR, there have been several supportive federal initiatives, including the establishment a Patient Engagement Advisory Committee by the Food and Drug Administration, the creation of Patient Centered Outcomes Research Initiative in 2010, and the development of valid and reliable patient-recorded outcomes measures such as the Patient Reported Outcomes Measurement Information System initiative by the National Institutes of Health.[59,60] Patient advocacy organizations have also served as active voices to influence policy and research agendas. Funding grants, facilitating collaboration, and encouraging patients to participate in research are ways that advocacy organizations have helped to promote patient-centered research.

Registries can support PCOR, because they offer an opportunity to collect health information and outcomes while incorporating multiple perspectives including those of patients, practitioners, and health systems. If designed with patient input and a patient-centered focus, registries offer a way to address research questions and concerns most relevant to patients and caregivers longitudinally. In addition to providing input on research questions, patients can also help support continued patient engagement and participation that promotes sustainability of the mission. Although including PCOR is ideal, it is important to understand that there are challenges that need to be considered when designing infrastructures including survey fatigue, sustainability, provisions for continued engagement, and plans for data dissemination. Engagement with patient advocacy organizations, education, and iterative construction with stakeholders is important for aspects of data that facilitate PCOR and will need to be considered as the platform for a national ACHD database is built. Inclusion of patients at the governance and oversight levels will be critically important as this process proceeds.

ACHD DATA SETS: THE CLINICIAN PERSPECTIVE

Although respect for people is important to individuals participating in large data sets, ethical consideration must also be given in the context of respect for clinicians and health systems, acknowledging the potential conflicts that may exist around participation, performance, transparency, and viability of health care as a business. Patients, health care practitioners, and health systems remain large stakeholders in a national ACHD database initiative. Health care practitioners play a unique role in the development of this infrastructure, because they often serve as a bridge between patients and health care systems. Uniquely, many have clinical, research, and administrative roles that lend themselves to the various aspects of data curation. From a purely clinical standpoint, the primary goal of an ACHD database might be to develop the framework for collecting quality metrics or data on patient experience. Some of these quality metrics will be well defined at the onset of the database, such as vital signs including blood pressure or documentation of resting oxygen saturation; however, others may be based on current research and expert opinion,[61] and yet there will also be future metrics as clinical understanding and research progress over time. Understandably, the act of measuring consistent outcomes and processes does not necessarily result in improved quality of care but provides a foundation to begin to accurately measure and compare results across conditions, centers, and age groups, among others. Once outcomes are well understood, disparities between groups can be identified, and further examination of contributors can be analyzed, with the goal of setting benchmarks and initiatives, using quality improvement projects that rely upon standard and innovative quality improvement methodology.[62–64] One goal of a national ACHD database is to build an infrastructure that eventually supports quality improvement projects that span geographic borders, including diverse patients and investigators, and one that can be managed seamlessly through a common unbiased sound government. Initial research priorities include understanding epidemiology, natural history, definition of prevalence, and comorbid disease,[65,66] in addition to identifying new quality-based and research priorities. Beyond immediate patient-contact quality measures, the data may be used to help understand health care use and fostering support for facility development and workforce planning.[67–69]

Critical to the success of gathering the outlined measures is the construction of a universal platform for data coding, plans for secure transfer of data, and data delivery practice that respects privacy measures. In and of itself, this is an onerous task, but it is further complicated by the heterogeneous way that health-related data are currently stored in both private and public US health care systems. If the data being analyzed are inaccurate or incomplete, the results cannot fully be trusted, and the current state of US databases reflects a high probability of inaccuracy and inadequacy. However, a focus on complete and comprehensive data must be balanced with efficiency and value. For instance, manual patient identification and data abstraction is associated with higher accuracy,[70] but it requires substantial resources, making it impractical to apply across large systems. Although the EMR is dedicated to patient care, the data collected are becoming easier to extract and more cost-efficient. Leveraging the automation of EMR systems will be important to scale a national-level ACHD data set and can assist in improving efficient and accurate data collection.

On the surface, participation in national ACHD data collection will result in positive consequences such as improved understanding of disease burden, health care use, and quality of care, to name a few. However, in reporting these data and identifying high value/quality care centers, we must remain aware of the potential for negative consequences. The goal is not to punish or negatively impact any specific clinician or program, but rather to share data to improve overall patient care, regardless of geography. This will be particularly important because the current payment system shifts to reward (recent) quality, as assessed by specific outcomes or process metrics. Health care systems want to avoid negative financial impact with participation, such as payment

penalties based on performance. Rather, the focus needs to swing to improvement in quality and overall care. Lessons learned from the Centers for Medicare and Medicaid Services' Quality Payment Program can be applied to the ACHD data set to facilitate this transition and incentivize participation by minimizing negative program changes, developing streamlined methods of data collection, providing information technology support, and engaging all participants in program management.[71]

From an institutional perspective, participation in a national ACHD data set will offer the opportunity to provide and demonstrate commitment to quality-based care. However, this will require allocation of resources including access to informaticians, data analyses, and institutional review board support. Additionally, data-specific requirements will include identifying local data storage, determining methods for data extraction, performing quality analysis of control data, and coordinating the transfer of deidentified data. Taking a further step back to collate individual center data at the national level, significant resources will be required. A team of biostatisticians, data analysts, and physicians with clinical expertise will be needed to provide the support required to build a national ACHD database. Additional needs will include secure data storage solutions, data maintenance/integration and plans for distribution. At each of these levels sound governance will be crucial to the success of building and growing a national ACHD database.

CALL TO ACTION

As recognized by the National Heart, Lung, and Blood Institute and the Adult Congenital Heart Association, numerous challenges to developing evidenced-based care for ACHD exist, including the heterogeneity of conditions, lack of infrastructure in the United States to track prevalence, fragmented care systems, loss to follow-up, and changes in treatment strategies over time.[72] There is a clear need for accurate, timely, comprehensive large-scale data collection of long-term complications and comorbid diseases faced by patients with ACHD, and in doing so, identifying opportunities to improve care and survival on the investigation of disparities in ACHD. To optimize the health of all patients with ACHD, we must continue to address the pervasive problem of disparities in health and health care, appreciate the unique methodological challenges associated with its investigation, evaluate interventions, and support quality-based initiatives that aim to provide equal care for all patients with ACHD. The adult CHD population is growing rapidly, yet in the absence of a comprehensive national database we fail to fully understand the impact of morbidity and mortality in ACHD in the United States, and more importantly in the identification of disparities that exist in this patient population. To build upon the success of childhood therapies, it is critically important to understand treatments, outcomes, and long-term sequelae in aging patients with CHD. The lack of available data stymies clinical and basic science and public health research, and impedes policy initiatives to improve care quality and efficiency in this medically complex patient population. This statement outlines the general principles and organization of a data collection process to gather high-quality data in a comprehensive, ethical manner. It is a challenging endeavor but necessary to provide the best possible care to all adult patients with CHD and families now and in the future. Inherent to this mission is inclusivity and equity, aiming to ameliorate disparities and improve quality in the care of the entire population of adults living with CHD in the United States and beyond.

ARTICLE INFORMATION

Affiliations

The Ohio State University Wexner Medical Center, Dorothy M. Davis Heart and Lung Research Institute, Columbus, OH (E.A.B.); Division of Cardiovascular Medicine, Heart and Vascular Institute, Penn State University College of Medicine, Hershey, PA (E.A.B.); Adult Congenital Heart Program, Knight Cardiovascular Institute, Oregon Health and Science University, Portland, OR (A.K.); Department of Medicine, University of Colorado Anschutz Medical Campus, Aurora, CO (D.M.M., A.K.); Division of Cardiovascular Medicine, University of California, Los Angeles, CA (K.B.); Indiana University Health and Riley Children's Hospital, Indianapolis, IN (S.C.); Department of Pediatrics, The Heart Institute, Cincinnati Children's Hospital, University of Cincinnati College of Medicine, Cincinnati, OH (A.R.O.); Division of Cardiology, Children's National Health System, Washington, DC (A.J.); The Heart Center, Nationwide Children's Hospital, Columbus, OH (M.L.); Department of Pediatric Cardiology, University of Michigan and Mott Children's Hospital, Ann Arbor, MI (S.P.); Division of Cardiovascular Medicine & Nationwide Children's Hospital, The Ohio State University Department of Internal Medicine, Columbus, OH (C.J.D.); Board of Directors Member, Adult Congenital Heart Association, Media, PA (M.P.); University of Washington Heart Institute and Department of Bioethics and Humanities, Seattle, WA (J.N.K.); and Department of Cardiology, Boston Children's Hospital, Boston, MA (M.G.).

Acknowledgments

The authors thank Dr DeCamp for thoughtful review and suggestions related to bioethical content and philosophy.

Sources of Funding

The authors are supported by National Institutes of Health grants: HL148701 (Dr Bradley), HL151604 (Dr Opotowsky), HL135061 (Dr Gurvitz), HL152740 (Dr Gurvitz). Dr Opotowsky was also supported by the Heart Institute Research Core at Cincinnati Children's Hospital Medical Center.

Disclosures

None.

REFERENCES

1. Liu Y, Chen S, Zuhlke L, Black GC, Choy MK, Li N, Keavney BD. Global birth prevalence of congenital heart defects 1970–2017: updated systematic review and meta-analysis of 260 studies. *Int J Epidemiol*. 2019;48:455–463. doi: 10.1093/ije/dyz009
2. Marelli AJ, Mackie AS, Ionescu-Ittu R, Rahme E, Pilote L. Congenital heart disease in the general population: changing prevalence and age

distribution. *Circulation*. 2007;115:163–172. doi: 10.1161/CIRCULATIONAHA.106.627224
3. Marelli AJ, Ionescu-Ittu R, Mackie AS, Guo L, Dendukuri N, Kaouache M. Lifetime prevalence of congenital heart disease in the general population from 2000 to 2010. *Circulation*. 2014;130:749–756. doi: 10.1161/CIRCULATIONAHA.113.008396
4. Gilboa SM, Devine OJ, Kucik JE, Oster ME, Riehle-Colarusso T, Nembhard WN, Xu P, Correa A, Jenkins K, Marelli AJ. Congenital heart defects in the United States: estimating the magnitude of the affected population in 2010. *Circulation*. 2016;134:101–109. doi: 10.1161/CIRCULATIONAHA.115.019307
5. Gurvitz M, Dunn JE, Bhatt A, Book WM, Glidewell J, Hogue C, Lin AE, Lui G, McGarry C, Raskind-Hood C, et al. Characteristics of adults with congenital heart defects in the United States. *J Am Coll Cardiol*. 2020;76:175–182. doi: 10.1016/j.jacc.2020.05.025
6. Ávila P, Mercier L-A, Dore A, Marcotte F, Mongeon F-P, Ibrahim R, Asgar A, Miro J, Andelfinger G, Mondésert B, et al. Adult congenital heart disease: a growing epidemic. *Can J Cardiol*. 2014;30:S410–S419. doi: 10.1016/j.cjca.2014.07.749
7. Krasuski RA, Bashore TM. Congenital heart disease epidemiology in the United States: blindly feeling for the charging elephant. *Circulation*. 2016;134:110–113. doi: 10.1161/CIRCULATIONAHA.116.023370
8. Research. NCftPoHSoBaB. The Belmont report: ethical principles and guidelines for the protection of human subjects of research. 1979.
9. Mittelstadt BD, Floridi L. The ethics of big data: current and foreseeable issues in biomedical contexts. *Sci Eng Ethics*. 2016;22:303–341. doi: 10.1007/s11948-015-9652-2
10. Riehle-Colarusso T, Strickland MJ, Reller MD, Mahle WT, Botto LD, Siffel C, Atkinson M, Correa A. Improving the quality of surveillance data on congenital heart defects in the metropolitan Atlanta congenital defects program. *Birth Defects Res A Clin Mol Teratol*. 2007;79:743–753. doi: 10.1002/bdra.20412
11. McNeely CL, Hahm J-O. The Big (Data) bang: policy, prospects, and challenges. *Rev Policy Res*. 2014;31:304–310. doi: 10.1111/ropr.12082
12. boyd D, Crawford K. Critical questions for big data: provocations for a cultural, technological, and scholarly phenomenon. *Inf Commun Soc*. 2012;15:662–679. doi: 10.1080/1369118X.2012.678878
13. Floridi L. Big data and their epistemological challenge. *Philos Technol*. 2012;25:435–437. doi: 10.1007/s13347-012-0093-4
14. Costa FF. Big data in biomedicine. *Drug Discov Today*. 2014;19:433–440. doi: 10.1016/j.drudis.2013.10.012
15. Khan A, Ramsey K, Ballard C, Armstrong E, Burchill LJ, Menashe V, Pantely G, Broberg CS. Limited accuracy of administrative data for the identification and classification of adult congenital heart disease. *J Am Heart Assoc*. 2018;7:e007378. doi: 10.1161/JAHA.117.007378
16. Stout KK, Daniels CJ, Aboulhosn JA, Bozkurt B, Broberg CS, Colman JM, Crumb SR, Dearani JA, Fuller S, Gurvitz M, et al. 2018 AHA/ACC guideline for the management of adults with congenital heart disease: executive summary: a report of the American College of Cardiology/American Heart Association Task Force on Clinical Practice Guidelines. *Circulation*. 2019;139:e637–e697. doi: 10.1161/CIR.0000000000000602
17. Zhang X, Pérez-Stable EJ, Bourne PE, Peprah E, Duru OK, Breen N, Berrigan D, Wood F, Jackson JS, Wong DWS, et al. Big data science: opportunities and challenges to address minority health and health disparities in the 21st century. *Ethn Dis*. 2017;27:95–106. doi: 10.18865/ed.27.2.95
18. Vener DF, Gaies M, Jacobs JP, Pasquali SK. Clinical databases and registries in congenital and pediatric cardiac surgery, cardiology, critical care, and anesthesiology worldwide. *World J Pediatr Congenit Heart Surg*. 2017;8:77–87. doi: 10.1177/2150135116681730
19. Salciccioli KB, Oluyomi A, Lupo PJ, Ermis PR, Lopez KN. A model for geographic and sociodemographic access to care disparities for adults with congenital heart disease. *Congenit Heart Dis*. 2019;14:752–759. doi: 10.1111/chd.12819
20. Gurvitz M, Valente AM, Broberg C, Cook S, Stout K, Kay J, Ting J, Kuehl K, Earing M, Webb G, et al.; Alliance for Adult Research in Congenital C and Adult Congenital Heart A. Prevalence and predictors of gaps in care among adult congenital heart disease patients: HEART-ACHD (The Health, Education, and Access Research Trial). *J Am Coll Cardiol*. 2013;61:2180–2184. doi: 10.1016/j.jacc.2013.02.048
21. Bhatt AB, Foster E, Kuehl K, Alpert J, Brabeck S, Crumb S, Davidson WR Jr, Earing MG, Ghoshhajra BB, Karamlou T, et al.; American Heart Association Council on Clinical C. Congenital heart disease in the older adult: a scientific statement from the American Heart Association. *Circulation*. 2015;131:1884–1931. doi: 10.1161/CIR.0000000000000204
22. Baker RB, Washington HA, Olakanmi O, Savitt TL, Jacobs EA, Hoover E, Wynia MK. African American physicians and organized medicine, 1846–1968: origins of a racial divide. *JAMA*. 2008;300:306–313. doi: 10.1001/jama.300.3.306
23. Ross PT, Lypson ML, Byington CL, Sánchez JP, Wong BM, Kumagai AK. Learning from the past and working in the present to create an antiracist future for academic medicine. *Acad Med*. 2020;95:1781–1786. doi: 10.1097/ACM.0000000000003756
24. Tahhan AS, Vaduganathan M, Greene SJ, Alrohaibani A, Raad M, Gafeer M, Mehran R, Fonarow GC, Douglas PS, Bhatt DL, et al. Enrollment of older patients, women, and racial/ethnic minority groups in contemporary acute coronary syndrome clinical trials: a systematic review. *JAMA Cardiol*. 2020;5:714–722. doi: 10.1001/jamacardio.2020.0359
25. Tahhan AS, Vaduganathan M, Greene SJ, Fonarow GC, Fiuzat M, Jessup M, Lindenfeld J, O'Connor CM, Butler J. Enrollment of older patients, women, and racial and ethnic minorities in contemporary heart failure clinical trials: a systematic review. *JAMA Cardiol*. 2018;3:1011–1019. doi: 10.1001/jamacardio.2018.2559
26. Preventza O, Critsinelis A, Simpson K, Olive JK, LeMaire SA, Cornwell LD, Jimenez E, Byrne J, Chatterjee S, Rosengart TK, et al. Sex, racial, and ethnic disparities in U.S. cardiovascular trials in more than 230,000 patients. *Ann Thorac Surg*. 2021;112:726–735. doi: 10.1016/j.athoracsur.2020.08.075
27. Clark LT, Watkins L, Piña IL, Elmer M, Akinboboye O, Gorham M, Jamerson B, McCullough C, Pierre C, Polis AB, et al. Increasing diversity in clinical trials: overcoming critical barriers. *Curr Probl Cardiol*. 2019;44:148–172. doi: 10.1016/j.cpcardiol.2018.11.002
28. Secretary's Advisory Committee on Health Promotion and Disease Prevention Objectives for 2020. Healthy People 2020: an opportunity to address the societal determinants of health in the United States. 2010. 2010.
29. Virani SS, Alonso A, Aparicio HJ, Benjamin EJ, Bittencourt MS, Callaway CW, Carson AP, Chamberlain AM, Cheng S, Delling FN, et al.; American Heart Association Council on E, Prevention Statistics C and Stroke Statistics S. Heart disease and stroke statistics-2021 update: a report from the American Heart Association. *Circulation*. 2021;143:e254–e743. doi: 10.1161/CIR.0000000000000950
30. Pandey A, Keshvani N, Khera R, Lu DI, Vaduganathan M, Joynt Maddox KE, Das SR, Kumbhani DJ, Goyal A, Girotra S, et al. Temporal trends in racial differences in 30-day readmission and mortality rates after acute myocardial infarction among Medicare beneficiaries. *JAMA Cardiol*. 2020;5:136–145. doi: 10.1001/jamacardio.2019.4845
31. Durstenfeld MS, Ogedegbe O, Katz SD, Park H, Blecker S. Racial and ethnic differences in heart failure readmissions and mortality in a large municipal healthcare system. *JACC Heart Fail*. 2016;4:885–893.
32. Savitz ST, Leong T, Sung SH, Lee K, Rana JS, Tabada G, Go AS. Contemporary reevaluation of race and ethnicity with outcomes in heart failure. *J Am Heart Assoc*. 2021;10:e016601. doi: 10.1161/JAHA.120.016601 doi: 10.1161/JAHA.120.016601
33. Eapen ZJ, Al-Khatib S, Lopes RD, Wang Y, Bao H, Curtis J, Heidenreich PA, Hernandez AF, Peterson ED, Hammill SC. Are racial/ethnic gaps in the use of cardiac resynchronization therapy narrowing?: an analysis of 107,096 patients from the National Cardiovascular Data Registry's ICD Registry. *J Am Coll Cardiol*. 2012;60:1577–1578. doi: 10.1016/j.jacc.2012.06.024
34. Breathett K, Allen LA, Helmkamp L, Colborn K, Daugherty SL, Khazanie P, Lindrooth R, Peterson PN. The Affordable Care Act Medicaid expansion correlated with increased heart transplant listings in African-Americans but not Hispanics or Caucasians. *JACC Heart Fail*. 2017;5:136–147.
35. Petersen EE, Davis NL, Goodman D, Cox S, Syverson C, Seed K, Shapiro-Mendoza C, Callaghan WM, Barfield W. Racial/ethnic disparities in pregnancy-related deaths—United States, 2007–2016. *MMWR Morb Mortal Wkly Rep*. 2019;68:762–765. doi: 10.15585/mmwr.mm6835a3
36. Soskolne G, Collins JW Jr, Fineman JR, Fuentes-Afflick E. Racial disparities in congenital heart disease: beyond red and blue. *Pediatr Crit Care Med*. 2012;13:218–219. doi: 10.1097/PCC.0b013e31822f112e
37. Nasr VG, Staffa SJ, DiNardo JA, Faraoni D. The association between race and adverse postoperative outcomes in children with congenital heart disease undergoing noncardiac surgery. *Anesth Analg*. 2022;134:357–368. doi: 10.1213/ANE.0000000000005571

38. Lopez KN, Morris SA, Sexson Tejtel SK, Espaillat A, Salemi JL. US mortality attributable to congenital heart disease across the lifespan from 1999 through 2017 exposes persistent racial/ethnic disparities. *Circulation*. 2020;142:1132–1147. doi: 10.1161/CIRCULATIONAHA.120.046822
39. Yeung E, Kay J, Roosevelt GE, Brandon M, Yetman AT. Lapse of care as a predictor for morbidity in adults with congenital heart disease. *Int J Cardiol*. 2008;125:62–65. doi: 10.1016/j.ijcard.2007.02.023
40. Salciccioli KB, Salemi JL, Broda CR, Lopez KN. Disparities in insurance coverage among hospitalized adult congenital heart disease patients before and after the Affordable Care Act. *Birth Defects Res*. 2021;113:644–659. doi: 10.1002/bdr2.1878
41. Rodriguez FH 3rd, Ephrem G, Gerardin JF, Raskind-Hood C, Hogue C, Book W. The 745.5 issue in code-based, adult congenital heart disease population studies: relevance to current and future ICD-9-CM and ICD-10-CM studies. *Congenit Heart Dis*. 2018;13:59–64. doi: 10.1111/chd.12563
42. Broberg C, McLarry J, Mitchell J, Winter C, Doberne J, Woods P, Burchill L, Weiss J. Accuracy of administrative data for detection and categorization of adult congenital heart disease patients from an electronic medical record. *Pediatr Cardiol*. 2015;36:719–725. doi: 10.1007/s00246-014-1068-2
43. Steiner JM, Kirkpatrick JN, Heckbert SR, Habib A, Sibley J, Lober W, Randall CJ. Identification of adults with congenital heart disease of moderate or great complexity from administrative data. *Congenit Heart Dis*. 2018;13:65–71. doi: 10.1111/chd.12524
44. Mezoff EA, Minneci PC, Hoyt RR, Hoffman JM. Toward an electronic health record leveraged to learn from every complex patient encounter: health informatics considerations with pediatric intestinal rehabilitation as a model. *J Pediatr*. 2019;215:257–263. doi: 10.1016/j.jpeds.2019.07.047
45. Code of Federal Regulations. Title 45 Public Welfare. Department of Health and Human Services. Part 46 protection of human subjects. 2009. Available at: https://www.hhs.gov/ohrp/sites/default/files/ohrp/policy/ohrpregulations.pdf. Accessed January 19, 2021.
46. Part C of Title XI of the Social Security Act, 42 USC §§ 1320d to 1320d-9 , and section 264 of the Health Insurance Portability and Accountability Act of 1996, 42 USC §1320d-2 note; 45 CFR Parts 160 and 164. 1996.
47. Services DoHaH. HITECH Act §13404(a); 45 CFR 164.104(b); 78 Fed. Reg. at 5591. 2013.
48. Gliklich RE, Dreyer NA, Leavy MB. *Chapter 7: Registries for Evaluating Patient Outcomes: A User's Guide*. 3rd ed. Agency for Healthcare Research and Quality (US); 2014.
49. Farazmand A. Sound governance: engaging citizens through collaborative organizations. *Public Organ Rev*. 2012;12:223–241.
50. Lehman J. Seven principles of sound governance. *IMPACT Magazine*. 2016.
51. Mirchev M, Mircheva I, Kerekovska A. The academic viewpoint on patient data ownership in the context of big data: scoping review. *J Med Internet Res*. 2020;22:e22214. doi: 10.2196/22214
52. Rodwin MA. Patient data: property, privacy & the public interest. *Am J Law Med*. 2010;36:586–618.
53. Koskinen JS, Kainu VM, Kimppa KK. The concept of Datenherrschaft of patient information from a Lockean perspective. *J Inf Commun Ethics Soc*. 2016;14:70–86. doi: 10.1108/JICES-06-2014-0029
54. Evans B. Would patient ownership of health data improve confidentiality? *Virtual Mentor*. 2012;14:724–732. doi: 10.1001/virtualmentor.2012.14.9.pfor1-1209
55. Househ MS, Aldosari B, Alanazi A, Kushniruk AW, Borycki EM. Big data, big problems: a healthcare perspective. *Stud Health Technol Inform*. 2017;238:36–39.
56. Knight HE, Deeny SR, Dreyer K, Engmann J, Mackintosh M, Raza S, Stafford M, Tesfaye R, Steventon A. Challenging racism in the use of health data. *Lancet Digit Health*. 2021;3:e144–e146. doi: 10.1016/S2589-7500(21)00019-4
57. Moloney R, Mohr P, Hawe E, Shah K, Garau M, Towse A. Payer perspectives on future acceptability of comparative effectiveness and relative effectiveness research. *Int J Technol Assess Health Care*. 2015;31:90–98.
58. Engagement rubric for applicants. 2021.
59. FDA announces first-ever patient engagement advisory committee. 2015.
60. Cella D, Yount S, Rothrock N, Gershon R, Cook K, Reeve B, Ader D, Fries JF, Bruce B, Rose M; Group PC. The Patient-Reported Outcomes Measurement Information System (PROMIS): progress of an NIH Roadmap cooperative group during its first two years. *Med Care*. 2007;45:S3–S11.
61. Gurvitz M, Marelli A, Mangione-Smith R, Jenkins K. Building quality indicators to improve care for adults with congenital heart disease. *J Am Coll Cardiol*. 2013;62:2244–2253.
62. Hartwell LP, Baker AL, Jenkins KJ, de Ferranti SD, Saleeb SF. Experience participating in the American College of Cardiology Quality NetworkTM: paediatric and adult congenital cardiology collaborative quality improvement. *Cardiol Young*. 2019;29:59–66.
63. Martin GR, Anderson JB, Vincent RN. IMPACT Registry and National Pediatric Cardiology Quality Improvement Collaborative: contributions to quality in congenital heart disease. *World J Pediatr Congenit Heart Surg*. 2019;10:72–80.
64. Winchester DE. Quality improvement in cardiology practice: a fellows' program. *J Am Coll Cardiol*. 2019;74:2533–2537.
65. Cotts T, Khairy P, Opotowsky AR, John AS, Valente AM, Zaidi AN, Cook SC, Aboulhosn J, Ting JG, Gurvitz M, et al.; Alliance for Adult Research in Congenital C. Clinical research priorities in adult congenital heart disease. *Int J Cardiol*. 2014;171:351–360.
66. Helm PC, Korten MA, Abdul-Khaliq H, Asfour B, Baumgartner H, Breithardt G, Kececioglu D, Schlensak C, Diller GP, Bauer UM. Three parties, one direction: research priorities in adults with congenital heart disease. What do professionals, patients and relatives want to know? *Int J Cardiol*. 2016;207:220–229.
67. Ezzeddine FM, Moe T, Ephrem G, Kay WA. Do we have the ACHD physician resources we need to care for the burgeoning ACHD population? *Congenit Heart Dis*. 2019;14:511–516.
68. Thomet C, Moons P, Budts W, De Backer J, Chessa M, Diller G, Eicken A, Gabriel H, Gallego P, Giamberti A, et al.; Disease ESCWGoG-uCH. Staffing, activities, and infrastructure in 96 specialised adult congenital heart disease clinics in Europe. *Int J Cardiol*. 2019;292:100–105.
69. Williams RG, Lu Y. Caring for ACHD in a market-driven society. *Methodist Debakey Cardiovasc J*. 2011;7:33–34.
70. Writing Committee M, Bhatt DL, Drozda JP Jr, Shahian DM, Chan PS, Fonarow GC, Heidenreich PA, Jacobs JP, Masoudi FA, Peterson ED, Welke KF; ACC/AHA Task Force On Performance. ACC/AHA/STS statement on the future of registries and the performance measurement enterprise: a report of the American College of Cardiology/American Heart Association Task Force on Performance Measures and The Society of Thoracic Surgeons. *Circ Cardiovasc Qual Outcomes*. 2015;8:634–648.
71. Peter M, Christine B, Chen PG, Katharine S, Patrick O, Nabeel SQ, Hussey PS. Perspectives of physicians in small rural practices on the medicare quality payment program. *Rand Health Q*. 2020;8:RR-2882-ASPE. eCollection 2020 Jun. Accessed June 2, 2021.
72. Gurvitz M, Burns KM, Brindis R, Broberg CS, Daniels CJ, Fuller SM, Honein MA, Khairy P, Kuehl KS, Landzberg MJ. Emerging research directions in adult congenital heart disease: a report from an NHLBI/ACHA working group. *J Am Coll Cardiol*. 2016;67:1956–1964.