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Perspective

Consent recommendations for research and international data sharing involving persons with dementia

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Abstract Consent is generally required for research and sharing rich individual-level data but presents additional ethical and legal challenges where participants have diminished decision-making capacity. We formed a multi-disciplinary team to develop best practices for consent in data-intensive dementia research. We recommend that consent processes for research and data sharing support decision-making by persons with dementia, protect them from exploitation, and promote the common good. Broad consent designed to endure beyond a loss of capacity and combined with ongoing oversight can best achieve these goals. Persons with dementia should be supported to make decisions and enabled to express their will and preferences about participation in advance of a loss of capacity. Regulatory frameworks should clarify who can act as a representative for research decisions. By promoting harmonization of consent practices across institutions, sectors, and countries, we hope to facilitate data sharing to accelerate progress in dementia research, care, and prevention.

Keywords: Alzheimer's disease; Dementia; Consent process; Broad consent; International data sharing; Research participation; Decision-making support; Will and preferences; Advance directive; Representative

1. Introduction

Progress toward understanding and treating dementia has been painfully slow. In all biomedical research, advances in research techniques (e.g., genomic sequencing, brain imaging) and information technology have led to a marked trend for gathering, linking, reusing, and sharing rich health-related data over long periods. It is now widely recognized that maximizing the societal benefit of health research almost always entails the timely release of data to the international research community. Data sharing honors the contributions of research participants, improves the transparency of research, and facilitates targeted recruitment for clinical studies. Increasingly, clinicians and health-care organizations are also expected to share data with

[1]https://www.ga4gh.org/ga4ghtoolkit/regulatoryandethics/.

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Box 1
Literature search strategy and selection criteria

We carried out a scoping review of the literature for our research question: What consent and capacity issues impact dementia research and data sharing? We performed database searches using the terms (Dementia OR Alzheimer) AND Research AND Consent published since 2007 on Web of Science (filters: Topic), PubMed (filters: MeSH and free text; Title/abstract), Google Scholar, and SSRN, until July 21st, 2017. French language searches were carried out on SCOPUS and Google Scholar for the terms (Alzheimer OR démence) AND recherche AND (consentement OR éthique). We experimented with numerous spelling variations, synonyms, and additional terms (e.g., capacity, competence, and data sharing), but these did not produce additional findings. Eight hundred fourteen results were reduced to 585 after removing duplicates. References were excluded when purely scientific articles (166), sources not available in English or French (8), incomplete or inaccessible sources (13) as well as literature not addressing or only superficially addressing populations with dementia (95), health research (166), or consent issues (31). The remaining articles were grouped according to key themes established iteratively through article review and consensus deliberation of the larger task team, which included consent, decision-making authority and support, planning in advance, representation, and capacity assessment. Two researchers (A.T., G.D.) reviewed the titles and abstracts from all references. Full texts were screened, where application of exclusion criteria or key theme grouping was unclear. An additional 34 articles addressing the key themes were found in reference lists or contributed by task team members for a total of 116. The literature included systematic reviews, empirical studies of stakeholder perspectives and practices, and regulatory and ethical analyses. A complete reference list can be found in Supplementary Materials. A limitation is that we did not search for literature about consent and capacity issues concerning other neurodegenerative conditions (e.g., stroke) or decision-making contexts (e.g., treatment, organ donation, and assisted dying). Such literature may provide important indirect insights but was too expansive to include, not to mention that some considerations are condition specific.

researchers to support "learning health systems" [1]. Data sharing presents opportunities for the dementia research community to pool high-quality data sets, attain larger sample sizes, maximize the value drawn from data already collected, and reduce wasteful—and sometimes harmful—duplication and delays in research.

Demonstrating the effectiveness of interventions for dementia requires participation of healthy persons and persons with dementia in clinical studies and biobanks and sharing of their genomic- and health-related data with many researchers. Where research involves physical, psychological, or privacy risks, researchers are generally required to seek consent [2,3]. New data types such as whole genome sequences present risks of participant re-identification, disclosure of sensitive information about disease risk or biological relationships, and misuse (discrimination in the workplace and insurance, stigmatization) [4]. It is therefore best practice for researchers to seek consent for sharing rich individual-level data (or the samples they are derived from) and also to protect these data using encryption, robust access control and ethical oversight, and network technologies that maintain secure, local storage while enabling federated analyses [5]. In general, data or sample sharing between institutions and across borders over long periods raises important consent challenges [6]. When is consent required? What form should consent take? Can adequate privacy protections be ensured between countries and institutions?

Dementia and other disorders of cognitive impairment are characterized by a progressive diminishment of cognitive skills (e.g., memory, reasoning, and language) that can impact on decision-making capacity. Researchers seeking consent from persons with dementia confront ethical and legal uncertainty, such as when and how to assess capacity to consent [7]. Regulatory frameworks governing decision-making involving persons with dementia are relatively clear for treatment, but not for research. Where rules are supplied for research, they often fail to accommodate the data sharing practices and digital interconnectivity of modern research. Guidance developed for invasive clinical studies tends to be disproportionately restrictive when applied to observational research or data sharing. Most international or national research guidelines rarely delve into the consent issues for adults with diminished capacity, deferring instead to unclear or restrictive local laws. Other laws and ethical guidelines tend to lump persons with dementia together with other vulnerable populations, overlooking ethical concerns specific to adults with diminishing capacity [8].

Well-meaning safeguards to protect persons with limited capacity from abuse and exploitation, such as the requirement that researchers seek consent from a legally authorized representative (LAR), can function to exclude persons with dementia from research and data sharing activities [2,3]. Disproportionate safeguards hinder improvements in dementia research, care, and prevention and undermine the right of persons with dementia to full and effective participation and inclusion in society [9]. Legal variation across jurisdictions or sectors can