



Article

Co-Design of Social Impact Domains with the Huntington's Disease Community

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Abstract: *Background:* For people living with or affected by Huntington's Disease (HD) to experience a good quality of life, tailored support is required to meet physical, cognitive-behavioral, psychological, and social support needs. Substantial service and knowledge gaps regarding HD exist across support providers and service systems. Measuring unmet needs and what quality of life looks like is a fundamental step required to determine the social impact of service investment and provision. The objectives of this study were to validate and map a draft set of HD Social Impact Domains (HD-SID) against existing national and international outcome frameworks; and evaluate and finalize the HD-SID set using a co-design approach with people with lived experience of, and expertise in, HD. *Methods:* This research used a qualitative co-design process, with 39 participants across four stakeholder groups (people who were HD gene-positive, gene-negative family members, academics, peak organizations, and service providers) to: (i) map and verify the social life areas impacted by HD; (ii) undertake a rigorous three-phased, qualitative process to critically evaluate the draft HD-SID; and (iii) seek feedback on and endorsement of the HD-SID through this co-design process, with a final set of HD-SID identified. *Results:* Endorsed HD-SID comprised risks and safety (including housing stability, and economic sustainability) and social inclusion (including health and symptom management, physical wellbeing, emotional wellbeing, and building resilient relationships). *Conclusions:* Effective measurement of the impacts and outcomes for people with HD is informed by both extant measures and an understanding of the specific population needs. This qualitative co-design research demonstrates that HD-SID resonate with the HD community.



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1. Introduction

Huntington's Disease (HD) is a neurodegenerative disorder of the central nervous system characterized by involuntary choreatic movements, cognitive and behavioral disturbance, psychiatric disorders, and, for some, dementia [1,2]. HD results in brain cell death and affects the regions of the brain that are responsible for motor movement control and coordination, cognition, personality and emotions, leading to significant impairments in one's ability to think, feel and move. HD onset predominantly occurs in young–middle adulthood [3]. As HD is an autosomally dominant genetic disease, each child of a parent with HD has a 50% chance of inheriting the defective Huntington's gene [3].

There is evidence that in Australia, North America and Western Europe (including the United Kingdom), HD prevalence has increased over the past 50-plus years [4]. In Western countries, it is estimated that about five to seven people per 100,000 are affected by HD. In Australia, it was previously estimated that over 1800 people have Huntington's Disease, with a reported prevalence rate in Australia that ranged from 4.5 per 100,000 to

6.5 per 100,000 [5]. However, these estimates were based on incomplete and dated data, indicating that the current HD prevalence in Australia is not fully known. Most recently, the prevalence in Australia was estimated at approximately 8.4 people per 100,000 [6]. With a current population of 25.7 million people in Australia, this data suggests that there are presently around 2160 people with a diagnosis of HD.

The needs of people living with HD, and their communities, are multiple and vary depending upon the stage of the disease, increasing over time as it progresses [7]. It has long been held that HD has a significant impact on both the person's physical and psychosocial wellbeing, with the latter frequently found to be more severely affected [8]. Given the range of complex physical, cognitive, behavioral, neurological and neuropsychiatric outcomes that can occur, HD leads to the requirement for a high level of support in community living, impacts the broader family, with gene-negative family members often being required to move into caregiving roles, and for some people with HD, it leads to early entry to institutional settings, including residential aged care [9,10]. Access to accurate and coordinated information and support services is therefore vital for people impacted by HD, including both the person with HD and their family and social network [11,12]. In light of these issues, it is important to understand and measure the domains of social impact deemed important by people with HD and their families, to inform service provision.

Social impact is defined as "a logic chain of results in which organizational inputs and activities lead to a series of outputs, outcomes and ultimately to a set of societal impacts" [13] (p. 3). Social impact measurement is "the processes of analyzing, monitoring and managing the intended and unintended social consequences, both positive and negative, of planned interventions (policies, programs, plans, projects) and any social change processes invoked by those interventions" [14] (p. 5). Limited evidence exists on the domains of social impact relevant to HD. A small body of research over the past two decades has however examined the needs and wants of HD communities. This research has identified unique perspectives of—and complex disease processes and system interfaces for—individuals and their families, which have implications for the management and development of interventions, and consideration of social impact, across the spectrum of HD stages [8,15]. Research has also identified that both coping mechanisms and illness perceptions have been found to make a major contribution to the explanation of variance in people's psychosocial wellbeing [16]. Active seeking of up-to-date, relevant and quality information across all areas of life is an identified need, and a range of barriers to accessing information has been noted [11].

In Australia, there is no national body for HD, but there are a number of state-based organizations [3]. Concerns have long been raised regarding fragmented, underfunded and complex support systems that exist for people with disabilities in Australia, including people with HD [17,18]. For people with HD and their families, the breadth and complexity of government service systems are well documented, as is the substantial informal care sector [7,19]. As part of the National Disability Strategy, in 2013 Australia began to introduce a new no-fault national disability insurance scheme (NDIS) for people who experience significant and permanent disability, who are aged under 65 years at the time of scheme entry; national scheme implementation was achieved in 2020 and includes independent regulation of the NDIS-funded services [20,21].

As HD progresses, people with the diagnosis can apply for NDIS access and plan for supports linked to their goals and needs. However, the NDIS was not designed to fill the gap in other mainstream systems that exist, and delineation of funding responsibilities of the Scheme and other service systems has been provided [22]. Other government systems that are frequently accessed or required by people with HD and their families include health, mental health, aged care, early childhood development, child protection and family support, education (primary, secondary, higher and vocational systems), employment, housing and community infrastructure, transport, and justice [22]. Given the complex and multiple needs experienced in the later stages of HD, a person and their formal and informal supporters may engage with all these service systems, often concurrently and at

multiple times over the lifespan [6]. This can lead to significant issues with coordination of services and information [19], and difficulty understanding the features of service delivery that can achieve positive impacts and outcomes experienced by people with HD and their supporters.

With this in mind, it is important to note that the impacts and outcomes of any human service or support can be viewed and reported in many different ways. It is critical to “measure what matters” to the individual and to select frameworks that capture the “complex open system” that is life for people living with health conditions such as HD [23–25]. A number of frameworks relevant to social impact have been published. For example, in the arena of sport, social impact was conceptualized as including the dimensions of social capital, collective identities, health literacy, wellbeing and human capital [26]. In contrast, research to establish a conceptual framework for understanding the social impact of burn injuries in adults identified the primary construct as social participation, which contained two concepts: societal role and personal relationships. Subdomains included work, recreation and leisure, relating with strangers, and romantic, sexual, family, and informal relationships [27]. However, to date, there has been only limited work exploring social impact across the entire disease spectrum, focused on health-related quality of life concepts [15]. It is therefore necessary to look to other existing outcome frameworks to holistically consider the social impact of HD.

The World Health Organization offers the International Classification of Functioning, Disability and Health (WHO ICF) [28], which identifies the aspects of a person, their environments, and their chosen tasks, and enables any person to describe their functional status, and the barriers and facilitators to functioning. As well as body functions, body structures (a person’s capabilities) and environmental factors (the barriers and facilitators surrounding the person), the WHO ICF identifies activities and participations. These are identified as learning and applying knowledge, general tasks and demands, communication, mobility, self-care, domestic life, interpersonal interactions and relationships, major life areas (education, economic), and community, social and civic life (culture, recreation, spiritual, political). The WHO ICF framework is extensive, and many communities have elected to identify subsets of factors that apply specifically to them (termed “ICF core sets”).

Another potentially relevant framework that was recently developed is provided by Australia’s National Disability Insurance Agency (NDIA), the agency that administers the NDIS. The NDIA has identified eight outcome domains comprising daily living, home, health and wellbeing, lifelong learning, work, social & community participation, relationships, and choice and control [29]. These outcome domains are deemed relevant for Australians living with permanent and significant disability, including the potential to be applicable for use with and by the HD community. However, people living with HD are at a significant intersect between disability, aged care and health systems, and the Council of Australian Governments previously identified the funding responsibilities of these mainstream systems in contrast to the NDIS; the NDIA is now working with state and territory governments to clarify these applied principles [30]. The relevance of NDIA outcome domains therefore may not offer a holistic perspective for, and have not been adequately tested with, the HD population, or been considered in relation to social impact. Service users such as the HD community have dynamic, complex and long-term needs that require coordinated inputs and activities to achieve desired outcomes. To document and evidence these, a framework is required which fully captures both the interrelationships and complexities, whilst recognizing the diversity of formal and informal supports and foregrounding any gaps in that support.

Given the range of complexities and gaps identified, drawn from the lived experience of the HD community and clinicians within its network, Huntington’s Victoria has previously worked to document key domains of social impact (see Table 1). For each of the eight domains, Huntington’s Victoria and the Huntington’s community defined a positive outcome and provided examples and evidence points. The positive outcome is framed

as an overarching aspirational statement. The domains are not listed in order of priority, rather in order of identification during the co-design process. The first three domains are health-focused and include health/symptom management, as well as physical and emotional wellbeing. The following four domains have a community/sustainability focus and include social inclusion and resilient relationships, as well a housing and economic stability. The final domain is focused on risks and safety. This includes the absence of “behavior” by the individual or toward the individual that places them at risk of harm, or of not achieving a positive outcome in the other seven domains.

Table 1. Social Impact Domains of HD (HD-SID).

DOMAIN 1 Health and Symptom Management	
Definition of a positive outcome	Achievement of HD symptom stability and overall ongoing maintenance of these symptoms Achievement of overall health separate from HD, that when not attained can negatively impact on the individual
Examples	Ongoing active participation in allied health intervention (diet or physical activity) Link to HD specialist for symptom management Continue the management of the health care plan (GP, HD specialist) Managing client progression throughout the various stages of the disease Maintaining physical and cognitive stimulation
DOMAIN 2 Physical Wellbeing	
Definition of a positive outcome	Achievement and maintenance of the highest possible level of physical independence for stage of disease
Examples	Mobility appropriate at the stage of HD Safety in home and community (environment) Physically capable of completing ADLs
Evidence points	Equipment accessed to support the individual at home and in the community Allied Health review embedded in care plan
DOMAIN 3 Emotional Wellbeing	
Definition of a positive outcome	To achieve emotional wellbeing and quality of life when living with HD
Examples	Improved mental health Mental health maintenance Improved coping skills and resilience Confidence building Maintenance of self-identity Increased hope Life satisfaction
Evidence points	Access to therapeutic intervention (medical and non-medical) Engagement in activities/routines that promote self-worth and identity
DOMAIN 4 Social Inclusion	
Definition of a positive outcome	To identify as a valued member of their local community. To maintain social connections and networks throughout the disease progression
Examples	Strengthening social skills (awareness of HD, self in the HD context) Reduced social isolation/contact/community connections Inclusive and accessible communities Access to venues (dining, entertainment, sporting, etc.) without discrimination
Evidence points	Engagement in age-appropriate social activities Engagement in regular community access Capacity building of local venues to enhance community access experiences
DOMAIN 5 Housing Stability	
Definition of a positive outcome	To either obtain and/or maintain stable housing that meets the support needs at any given point during disease progression
Examples	Housing security/safety Housing that is accessible and structured to maximize ongoing support needs (minimized risks of falls, capacity for in-home modification if needed) Cost of rent or mortgage that can be sustained long-term Cost of utilities and other household-related expenses are affordable In-home staff are skilled to meet the care needs of the individual

Table 1. Cont.

Evidence points	Secured permanent disability accommodation Access to in-home modifications Center-pay or other financial institutions implemented to pay bills and manage funds, as needed Services and supports implemented
DOMAIN 6 Economic Sustainability	
Definition of a positive outcome	To achieve and/or maintain financial security. To live without financial hardship and be able to afford basic needs.
Examples	Maintaining appropriate employment/supporting opportunities for appropriate employment Education/skills development Obtainment of appropriate income stream (Centrelink pension, superannuation, paid employment)
Evidence points	In receipt of disability support package (DSP), superannuation, total and permanent disability (TPD) pay Capacity building of workplace for reduced/modified employment Completed training/skill development
DOMAIN 7 Building Resilient Relationships	
Definition of a positive outcome	To build and/or maintain resilient relationships with partners, family members, friends, carers, neighbors, etc.
Examples	Family resilience Reconnecting families/siblings Preventing carer burnout
Evidence points	Regular respite opportunities Participated in meaningful activities/quality time together Capacity building of family members
DOMAIN 8 Risks and Safety	
Definition of a positive outcome	The absence of "behavior" by the individual or toward the individual that places them at risk of harm, or of not achieving the above measures.
Examples	Reduced incidents of risks (vulnerable to financial, emotional, sexual, physical abuse) Maintaining service delivery through funded packages Competent and supported decision-making Reduced incidences of "challenging behavior" that places the individual at risk of losing current accommodation, criminal/civil law proceedings, removal/ceasing of critical care need supports, isolation Reduced incidences of industrial relations issues and other acts of discrimination
Evidence points	Enduring power of attorney (EPOA) financial, guardianship appointed Behavior management plan implemented Behavioral management services engaged Advocacy within the justice system Advocacy within the legal setting (court, VCAT, tenancy)

Consistent with the emergence of communities owning and articulating the impacts and outcomes that are important to them [31], and acknowledging that stakeholder-based approaches are the most appropriate solution for the selection of social impact measurements [32], the research objectives are to utilize a qualitative co-design methodology to:

1. validate or refute, and map a draft set of HD social impact domains (HD-SID) against existing national and international outcome frameworks; and
2. evaluate and finalize the HD-SID set using a co-design approach with people with lived experience of, and expertise in, HD.

This research into an HD-SID framework is the first phase of a larger, multi-year study researching ways to provide nationwide access to up-to-date, relevant and quality information, including peer support, for the Huntington's community [33].

2. Materials and Methods

This project received Monash University Human Research Ethics Committee approval prior to commencing data collection (Project ID: 24641).

A method for critical evaluation of the draft HD-SID framework was developed using a two-stage process of stakeholder engagement [33]. This included (i) investigation of the face validity of the HD-SID via a steering committee, and mapping of the draft domains against existing national and international outcome frameworks (Phase 1a–c); and (ii) a

series of qualitative focus groups undertaken with people with HD, their families and clinical experts in the field of HD and the broader HD community (via a state-based conference on HD) (Phase 2a,b) (see Figure 1).

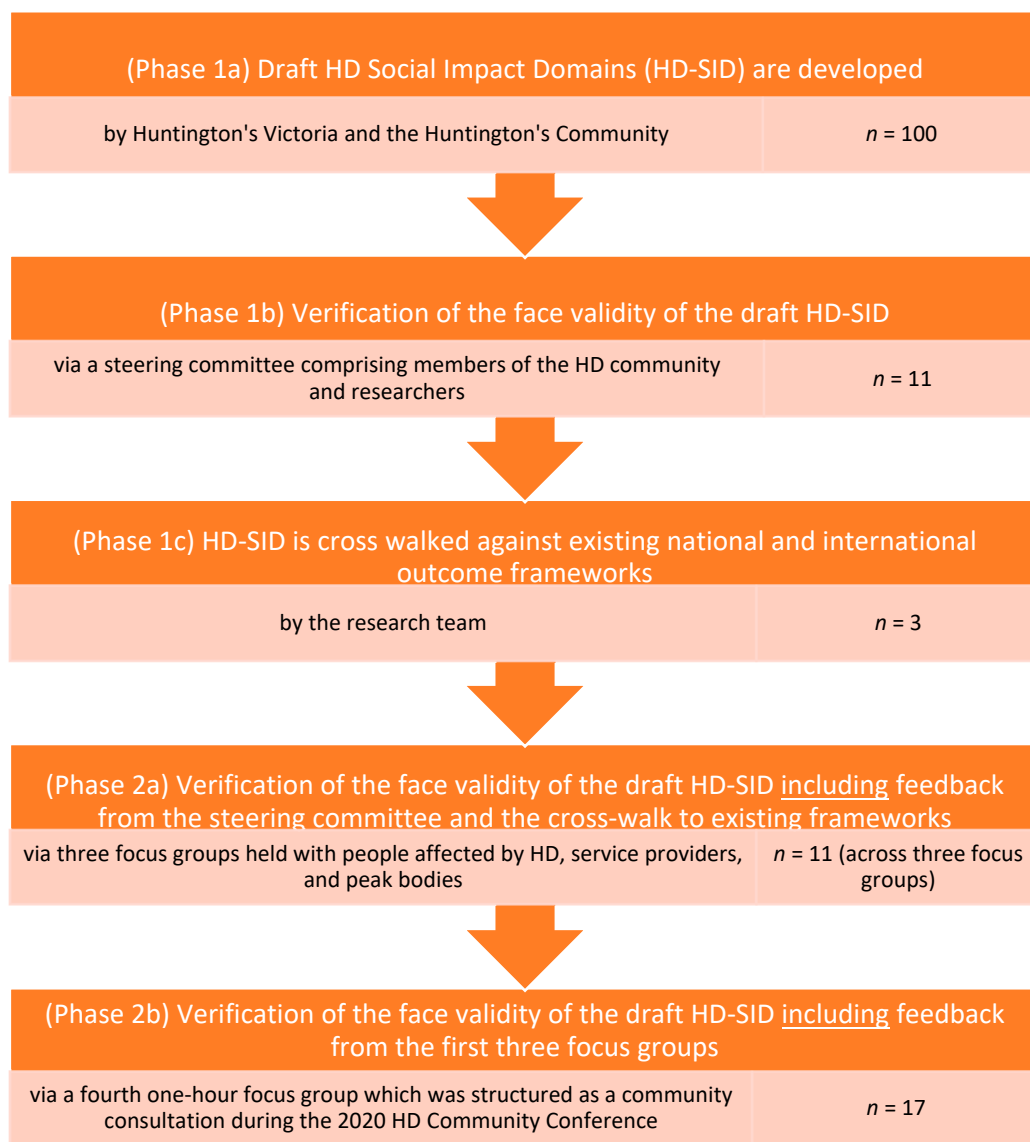


Figure 1. Flow of the qualitative research process used in the HD-SID verification process.

Regarding participant consent, while Phase 2a, with the focus group attendees, required written informed consent prior to participation, all other parts of the study used implied consent for participation. Implied consent involved informing potential participants about the project underway prior to participation, with a clear understanding that participation was voluntary and the act of participating implied consent (e.g., Phase 2b community consultation during the 2020 HD Community Conference).

The method for each of these is outlined below:

2.1. Verification or Refutation of the Draft HD Social Impact Domains

A Steering Committee was established for the project duration, comprising people who were HD gene-positive (symptomatic and non-symptomatic) (*n* = 2), gene-negative and family members (*n* = 2), academics (*n* = 4), and HD peak organization representatives (*n* = 3). Based on the principles of co-design, these lived experience and content experts reviewed the draft HD-SID framework and discussed face validity. A mapping process was

then used by the researchers to triangulate and investigate the content validity of the draft HD-SID domains against three key national and international policy frameworks: World Health Organization international classification of functioning (WHO ICF) [28]; the national disability insurance scheme outcome framework [29]; and the COAG system interface domains [22] endorsed by the Committee. The domains were then used to structure the focus group schedule (see Appendix A for the focus group schedule).

2.2. Focus Groups

A total of four focus groups across two phases were held with various HD stakeholders. Focus groups tested the HD-SID domains by (a) enquiring as to service gaps for the community, and (b) clarifying the performance of the domains in capturing this data for participants. Focus group 1 comprised health professionals and included 6 participants (physician, counselor, advocate, speech pathologist, occupational therapy manager, genetic counselor). This sample was drawn purposively from Huntington's Victoria's existing networks, and participants held a range of roles across private and public systems, progressive neurological or HD-specific services, including acute, community and residential care. Focus group 2 included two gene-positive individuals, one of whom identified as being symptomatic. Focus group 3 included three family members involved with supporting relatives (children, parents, spouses) with Huntington's disease, one of whom identified as gene-negative (see Appendix A for the focus group schedule used). There was a final one-hour focus group which was structured as a community consultation (focus group 4). Members of the HD community, including people who were gene-positive, those who were gene-negative, as well as other HD stakeholders, were invited to an online presentation of results from the first three focus groups as well as mapping the HD-SID against existing national and international outcome frameworks; this was held during the 2020 HD Community Conference [34]. Focus group members ($n = 50$) were invited to provide feedback on specific multi-choice questions via an online poll embedded throughout the presentation. A smaller number ($n = 17$) of the total focus group members chose to participate in one or more of the online poll questions. The aim of this activity was to finalize and seek stakeholder endorsement of the HD-SID. A plain language document featuring the HD-SID framework was available on the conference website prior to, during, and following the presentation.

2.3. Data Collection

The initial draft HD-SID validation and consensus work (Phase 1b) was undertaken within a total of 3 meetings, held online using a video-link platform (Zoom.us), and coupled with the framework mapping task. Each focus group (Phase 2a) was also held online via video link. The consensus work and focus groups lasted between 75 and 90 min and were held between October and December 2020. For the Phase 2a focus groups, a representative of Huntington's Victoria was invited online to open each focus group and explain the project's context, and then left the session.

Two of the research team members attended all meetings, and the primary author (N.L.) facilitated each discussion, while the second author (N.B.) acted as note-taker. Both are health professionals (an occupational therapist and physiotherapist, respectively) experienced in working with people with neurological disability and their supporters. Focus group attendees provided HREC-approved signed consent forms prior to participation, and the sessions were recorded via Zoom with participants' permission. The Phase 2b HD community consultation data were captured using Zoom Polls and the Zoom chat function for qualitative responses ($n = 17$ participant responses received).

2.4. Data Management and Analysis

The audio files (for Phase 2a) and exported poll data and chat content (community consultation in Phase 2b) were collected and securely lodged on the researchers' password-protected computers, along with the researchers' reflexive notes. Audio files

were transcribed verbatim, with pseudonyms applied at that time in preparation for thematic analysis. Thematic analysis was used for qualitative data analysis because it can be applied across a range of theoretical and epistemological approaches, whilst offering a rich and detailed account of qualitative data. It also offers a contextualized method where exploration of participant experiences and perspectives are examined and understood within the context of environmental and societal impacts [35].

Using Braun and Clark's 15-point checklist for thematic analysis, a process of the qualitative comparative method of inductive thematic analysis was used for the analysis of the transcribed data and associated reflexive notes [35,36]. This approach acknowledges that analysis is shaped to some extent by the researchers' disciplinary knowledge and standpoint [35]. Data were initially analyzed by, and then across, the focus groups. Inductive content analysis was used to analyze data collected from open-ended written information provided via the Zoom chat function in the Phase 2b consultation [37]. The results were synthesized into a tabulated summary of themes, aligned with the research aims.

To ensure qualitative rigor, both researchers undertaking the data collection kept reflective field notes during the two research stages, which included the researchers' subjective thoughts and feelings as well as observations within, and reflections upon, data collection [38].

In addition, regular meetings between the researchers involved in data collection and analysis and Huntington's Victoria were held during the data collection period. These meetings provided an opportunity to discuss the two-stage qualitative processes, emerging findings and reflections. The reflective notes kept by the two researchers were examined prior to, and during, thematic analysis, acknowledging the personal reflexivity the researchers brought into the research [35,38].

3. Results

3.1. Verification of Domains

The verification work of the steering committee (Phase 1b) firstly considered fidelity to the HV population and, to principles of consumer co-design, resulted in an affirmation of the social impact domain approach. An examination of the domains themselves, in light of lived experience, confirmed the validity of the draft HD-SID, but noted the dynamic and overlapping nature of, in particular, "health and symptom management" with "physical wellbeing". Therefore, the areas "health and symptom management" and "physical wellbeing" were clustered under the broader area of "health and wellbeing" within the focus group schedule.

Table 2 contains mapping against the World Health Organization international classification of functioning (WHO ICF), the national disability insurance scheme outcome framework, and the COAG system interface domains. Researchers identified a high degree of congruence between HD-SID domains and these taxonomies. Specificity varied, as depicted in Table 2, with a number of domains from one framework applying to multiple domains across other frameworks. The only clear outliers identified were the concept of "aged care" (COAG domains), and the domain "products and technology" was an enabler across a high number of HV domains.

Mapping demonstrated a good fit between impact domains. This indicated the HD-SID captured all important concepts, and that research related to the HD-SID could be cross-walked to other relevant frameworks as needed. The HD-SID domains were therefore used in their current form for focus groups and consultations, reported below.

Table 2. Mapping of the HD-SID domains against the NDIS, WHO ICF and COAG domains.

Social Impacts of HD	NDIS Adult Outcome Domains	WHO ICF	COAG Domains
Health and symptom management	Health and wellbeing	Body structures and functions Selfcare Learning and applying knowledge General tasks and demands Communication	Health Aged Care *
Physical well being		Mobility Products and technology	
Emotional wellbeing	Choice and control		Mental Health
Social inclusion	Daily Living Lifelong learning Social, community and civic participation	Community, social and civic life (culture, recreation, spiritual, political) Attitudes Products and technology	Transport
Housing stability	Home	Natural environment and human-made changes to environment	Housing and community infrastructure
Economic sustainability	Work	Domestic life Major life areas (education, economic)	Education Higher education and VET Employment
Building resilient relationships	Relationships	Interpersonal interactions and relationships Support and relationships	Early childhood development
Risks and safety		Services, systems and policies	Justice Child protection and family support

KEY: * outliers.

3.2. Focus Group Perspectives

3.2.1. Findings from Focus Group 1: HD Professionals

- Domain 1 (health and wellbeing) subsumes domain 2 (physical wellbeing): participants discussed how to define health and wellbeing, for example, “Does this [domain] cover more than symptom management and physical health?” (physician). People felt that managing health and wellbeing includes health and symptom management, as well as physical wellbeing. Participants observed that service gaps could occur in psychosocial support services, where there is poor understanding that HD is an organic disease, resulting in the emergence of mental health issues and the need for mental health services.
- The discourse around domain 3 (emotional wellbeing) included domain 4 (social inclusion) and domain 7 (relationships). Civic participation as a desirable outcome for people living with HD mapped to domain 8 (risks and safety). Comments included the need for risk management where “cognitive decline is responsible for social isolation”. Domain 3 (emotional wellbeing), domain 7 (maintaining relationships) and domain 4 (social inclusion) were all deemed at risk if people cannot cognitively manage an “ordered lifestyle”. Domain 5 (housing stability) and domain 6 (economic stability) are foundations for these aspects of emotional wellbeing, with many examples discussed of adverse outcomes for individuals “living in toilet blocks or cars . . . Struggling with finances, disturbing neighbors, and requiring emergency accommodation” which in turn caused adverse impacts in all other domains. Protective factors include the presence of family. No other areas were identified, indicating saturation in social impacts through the domains presented.

3.2.2. Findings from Focus Group 2: HD Gene-Positive Individuals

- Domain 1 (health and symptom management), domain 2 (physical wellbeing) and domain 3 (emotional wellbeing) were discussed together, with key supports which enable these areas, including “HV support services, HV counselor, allied health, massage, yoga, meditation, aromatherapy (mood-lifter to help with depression), crystal healing, reflexology or pressure point therapy, reiki, positive affirmations” (symptomatic participant).
- The concept of social inclusion (domain 4) was highly resonant and also individualized, and participants discussed their “own ways to find comfort and support for social inclusion”, including “Op shopping, having coffee, hang in cafes and sit and read the paper, hang out, do a bit of people watching, walking on the beach with my fur babies . . . Pet therapy is really awesome”.
- Domain 5 (housing stability), domain 6 (economic stability) and domain 8 (risks and safety) generated much discussion, with the foundation supports (such as safe and appropriate housing) linking directly to higher-order impacts, “Where can I live? I feel I could live by myself . . . There is no safe place for time-out” (symptomatic participant).
- When asked if there were other social impacts that had not been captured, participants mentioned the availability of assisted dying/euthanasia, and experiences where professionals failed to “give bad news” appropriately. In contrast, focus group 3 participants below allocated “appropriate professional support” to domain 3 (emotional wellbeing) due to the significant impact of this barrier/facilitator on their overall emotional functioning.

3.2.3. Findings from Focus Group 3: HD Gene-Negative Individuals and Family Members

- Domain 1 (health and wellbeing), domain 2 (physical wellbeing) and domain 3 (emotional wellbeing) were discussed together, as the supports were felt to enable other domains. Examples of professional support were explicitly linked to domain 3 (emotional wellbeing), for example: “(it is) challenging to locate helpful professionals with the skillset and then the knowledge of HD . . . (I) remember feeling quite frustrated and sometimes paid a lot privately, and they weren’t up to scratch . . . we would live for them to come that day and they were not informed, really”.
- Housing stability (domain 5), economic sustainability (domain 6), and risks and safety (domain 8) were bundled together with strong narratives around risk. Comments focused on the “falling away” of support when government services changed. For example, “With NDIS, this source of local and timely support (local councils) has fallen away” (family member).
- When asked about other issues not included in the impact domains, participants raised the issue of genetic testing and reflected on poor experiences with health professionals in regard to genetic testing, and “knowing how to support children, relatives, people not yet tested” (gene-negative individual).

3.2.4. Findings from Focus Group 4. Consultation with Members of the HD Community

Approximately 200 HD Community Conference delegates had the opportunity to review the HD-SID Framework through a plain-language summary featured on the HV website, and an advertised one-hour focus group at the virtual Huntington’s Community Conference scheduled for 6 December 2020. Fifty participants attended the session, with live responses from 17 of those delegates to three question sets regarding the HD gap analysis (structured according to the HD-SID framework). Responses indicated the HD-SID framework was found to be appropriate and able to capture issues of importance to the HD community. No alternatives or amendments to the final HD-SID were raised.

3.2.5. Summary of Focus Group Findings

Discourse across the focus groups verified the drafted HD-SID, whilst presenting two key themes, and five subthemes, seen as necessary within the framework, which “clustered” various impact domains together. Broadly, participants grouped domains according to the “safety net” cluster of risks and safety (including housing stability, economic sustainability), or the “social inclusion” cluster (including health and symptom management, physical wellbeing, emotional wellbeing, building resilient relationships) (see Figure 2).

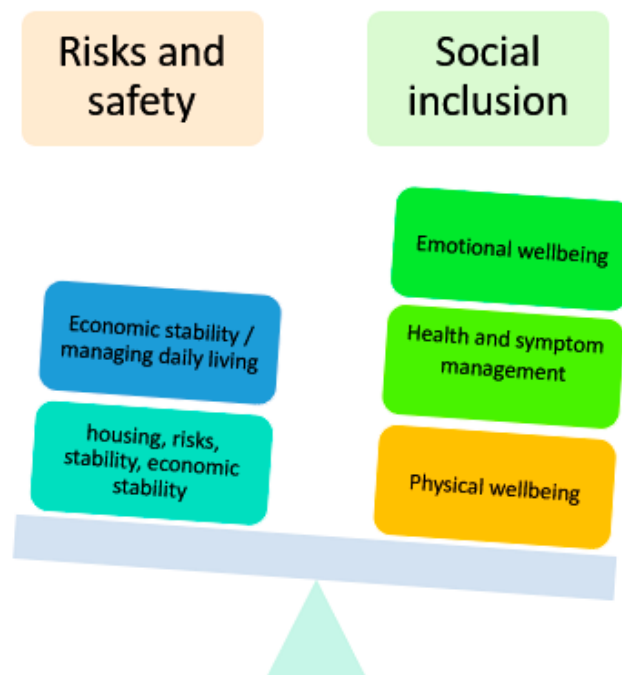


Figure 2. Themes and sub-themes for HD-Social Impact Domains.

4. Discussion

This research used contemporary qualitative co-design methodology with both people with HD and their families, as well as the broader HD community, to verify and map a draft set of Huntington’s Disease Social Impact Domains (HD-SID) against existing outcome frameworks internationally. The HD-SID set was then finalized in preparation for the pilot and evaluation. Evidenced against existing national and international outcome frameworks, this work is the first of its kind to closely consider the exploration and measurement of both the disease itself, as well as personal and environmental factors that may impact the outcomes of people diagnosed with HD.

The social impact of HD can be considerable [2]. To date, however, existing frameworks have not always been feasible for application involving people with HD across the various stages of the disease, including both those who are early post-diagnosis as well as people who experience multiple and complex needs as the disease progresses [15]. Holistic social impact domains that allow both the person, and their key supporters or providers, to use a person-centered approach to the measurement and evaluation of social impact are essential to meet complex and individualized needs.

One of the key areas highlighted in the final set of HD-SID that were endorsed is the balance between choice and control versus service quality and safeguarding for people with HD. As identified previously, HD leads to a range of physical and psychosocial impacts [16]. The HD-SID help to elicit these across multiple domains, aligned with current disability policy and focused on contemporary rights-based considerations [2,21,39,40]. The HD-SID have been closely considered to ensure alignment with rights-based approaches, whilst offering breadth to ensure they are future-proofed to health and disability policy and service changes that may occur internationally, over time. In addition, specific domains

acknowledge the complex service planning and implementation that may be required, and the necessary oversight for both human rights and safeguarding (e.g., in relation to behavior support planning, see domain 8).

Continuing the human rights perspective, existing frameworks outline an imperative to ensure meaningful consumer participation in all aspects of human services work [39,40]. Co-design and coproduction are seen as best-practice approaches in service design, implementation and evaluation; however, the embedding of consumer voices in service planning and provision is arguably less developed. This project used contemporary co-design (both with individuals with active HD, as well as the broader HD community) across all three phases of work, held as central to the development of the final HD-SID. Future implementation of the HD-SID, and evaluation of their utility across the stages of HD, will benefit from this continued co-design approach, with a focus on self-determination and the potential of the HD-SID framework to aid peer-to-peer support [12].

When measuring social impact, it has been acknowledged that a holistic approach that considers the physical, mental and social wellbeing and safety of all involved, with specific attention to and input from more vulnerable groups (including those with disability) is necessary [14]. The system interfaces that a person with HD and their families will experience—including with the health, disability, housing, education and aged care systems—are often multiple and complex to coordinate [19]. These system interfaces have been explicitly represented in the HD-SID to ensure holistic service planning and evaluation, whilst also offering a framework that may build the capacity of systems less experienced with planning for the impacts of HD. The focus on social impact domains that can be considered across, and gathering information to capacity-build within, the service system intersects that a person with HD and their family may experience has therefore been central to the work as developed.

Whilst this research has been designed to begin to bridge an existing evidence gap in relation to social impact measurement for people with HD across all stages of the disease process, as well as their key supporters, some limitations in this work exist. This includes the fact that the research relied on the testing of initial work with a draft set of HD-SID that had previously been developed by Huntington's Victoria (prior to the research commencing). To attempt to manage this, an independent research group and rigorous methodological design were applied to adequately explore and endorse or refute the draft set via multiple perspectives, and triangulate this work with other existing frameworks and across various HD stakeholder groups. Following the very necessary application of co-design, and thus the focus group methodology used, led to some participants inputting more to the qualitative methods than others. For example, of the 50 attendees at the final focus group (hosted as a community consultation), only 17 responded to polling questions. Attempts were made to address this balance in responses by running a series of focus groups, rather than relying on the community consultation alone. Finally, the finalized HD-SID framework developed through this work—although endorsed by people with HD, their families, peak organizations and government bodies—has not yet been formally tested as a social impact measure. The authors also note that this research was not intended to be a systematic review of HD management or best practice, but rather aimed to co-design domains for consideration and evaluation of social impact from the perspective of various stakeholders, including people with HD, their families, academic experts and service providers. As a result, it does not offer a research protocol aligned with the finalized set of HD-SID. Future research that utilizes or applies the HD-SID set will need to develop such protocols. Finally, this research has focused on the Australian context and, therefore, whilst it is expected that the principles apply internationally, there may be nuances/in-country differences which will require future research evaluation. Given these identified limitations, the next important step in this work will be to pilot and evaluate the utility of the HD-SID both with organizations engaged in the provision of services to people with HD and their families, as well as directly with people with HD and/or

their support networks, considering social impacts within service planning or cross-sector coordination of care.

5. Conclusions

To date, there has been limited guidance on relevant and effective social impact domains in the field of HD. This research offers a new contribution of co-designed HD social impact domains comprising risks and safety (including housing stability, and economic sustainability) and social inclusion (including health and symptom management, physical wellbeing, emotional wellbeing, and building resilient relationships). This participatory research has demonstrated that such measurement is informed by both extant measures and a deep and nuanced understanding of the HD population and their support networks. The finalized HD Social Impact Domains have been co-designed, and are demonstrated to resonate, with the HD community. They provide a useful and comprehensive framework for both effective and holistic care coordination and delivery, as well as social impact, and will be used in future research for service evaluation.

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Appendix A

Table A1. Excerpt of Focus Group Schedule based on impact areas.

Gap Analysis Focus Group Guide 2020	
Theme	Prompts
1. Introductions	<ul style="list-style-type: none"> - Researcher introductions & about the project - Tell us your name and a little bit about yourself - How are you involved with the HD community? - How long have you been involved?
2. The idea of a gap analysis	We want to find out about formal and informal support needs. We will be asking questions for each area of life that the HD community, and NDIS, identify as important. We invite you to add any other areas at the end

Table A1. Cont.

Gap Analysis Focus Group Guide 2020	
Theme	Prompts
Supports and gaps across areas of life from a human perspective: We will ask the following questions for areas 3–10 —What is an enabler or a support (what helps) in this area? —What is missing in this area? —What is needed in this area (what would “good” look like?)	
3. Health and wellbeing	<ul style="list-style-type: none"> - Health and symptom management - Physical wellbeing
4. Emotional wellbeing	<ul style="list-style-type: none"> - Choice and control
5. Social inclusion	<ul style="list-style-type: none"> - Lifelong learning - Community participation (political, cultural, spiritual, recreational)
6. Housing stability	<ul style="list-style-type: none"> - Civic participation
7. Economic sustainability	<ul style="list-style-type: none"> - Daily living
8. Relationships	<ul style="list-style-type: none"> - Building resilient relationships - Social participation
9. Risks and safety	
Supports and gaps across areas of life from a government perspective: The Commonwealth Government (COAG) describe 11 areas where their policies “intersect” and where there can be service gaps. Tell us what you think about supports and gaps in the following areas:	
10. Formal supports and gaps	<ul style="list-style-type: none"> - Aged care - Justice - Transport - Housing and community infrastructure - Employment - Higher education and VET - Education - Child protection and family support - Early childhood development - Mental health - Health
11. Other?	
12. Thank you . . .	

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