STUDY PROTOCOL

REVISED COVID-19 IDD: A global survey exploring family members' and paid staff's perceptions of the impact of COVID-19 on individuals with intellectual and developmental disabilities and their caregivers. [version 2; peer review: 2 approved]

Previously titled: COVID-19 IDD: A global survey exploring the impact of COVID-19 on individuals with intellectual and developmental disabilities and their caregivers

Christine Linehan[1], Tal Araten-Bergam[2], Jennifer Baumbusch[3], Julie Beadle-Brown[4], Christine Bigby[2], Gail Birkbeck[5], Valerie Bradley[6], Michael Brown[7], Femmianne Bredewold[8], Masauso Chirwa[9], Jialiang Cui[10], Marta Godoy Gimenez[11], Tiziano Gomiero[12], Sarka Kanova[13], Thilo Kroll[14], Mac MacLachlan[15], Brigit Mirfin-Veitch[16], Jayanthi Narayan[17], Finiki Nearchou[18], Adam Nolan[1], Mary-Ann O'Donovan[19], Flavia H. Santos[18], Jan Siska[20], Tim Stainton[3], Magnus Tideman[21,22], Jan Tossebro[23]

[1]UCD Centre for Disability Studies, University College Dublin, Belfield, Dublin, Dublin 4, Ireland
[2]Living with Disability Research Centre, School of Allied Health, Human Services & Sport, La Trobe University, Bundoora, Victoria, 3086, Australia
[3]Canadian Institute for Inclusion and Citizenship, University of British Columbia, 2080 West Mall, Vancouver, BC Canada, V6T 1Z2, Canada
[4]Tizard Centre, University of Kent, Canterbury, Kent, CT2 7NZ, UK
[5]Business Information Systems, University College Cork, O'Rahilly Building, Cork, Ireland
[6]Human Services Research Institute, 2336 Massachusetts Ave, Cambridge, MA 02140, USA
[7]School of Nursing and Midwifery, Queen's University, 97 Lisburn Road, Medical Biology Centre, Belfast, BT9 7BL, UK
[8]University of Humanistic Studies, Kromme Nieuwegracht 29, Utrecht, 3512 HD, The Netherlands
[9]School of Humanities and Social Sciences, Department of Social Work and Sociology, University of Zambia, Great East Road Campus P.O Box 32379, Lusaka, 10101, Zambia
[10]Department of Social Work, The Chinese University of Hong Kong, Shatin, New Territories, Hong Kong, China
[11]Department of Psychology, University of Almeria, La Cañada de San Urbano, 04120, Almería, Spain
[12]ANFFAS Trentino Onlus DAD© Project Group, via Giambattista Unterveger, 38121 Trento Trentino,, Italy
[13]Department of Education, University of West Bohemia, Univerzitní 2732/8, Plzeň 3, 301 00, Czech Republic
[14]School of Nursing, Midwifery and Health Systems, University College Dublin, Belfield, Dublin, 4, Ireland
[15]School of Psychology, Maynooth University, Maynooth, Ireland
[16]Donald Beasley Institute, 248 Cumberland Street, Dunedin Central, Dunedin 9016, New Zealand
[17]Inclusive Education at Faculty of Health, Education and Society, University of Northampton, Northampton, UK
[18]UCD School of Psychology, University College Dublin, Belfield, Dublin, 4, Ireland
[19]Centre for Disability Studies, Sydney Medical School, Faculty of Medicine and Health, University of Sydney, 92-94 Parramatta Rd, Camperdown, Sydney, NSW 2050, Australia
[20]Department of Special Education, Charles University, Magdalény Rettigové 4, Praha 1, 116 39, Czech Republic
[21]Department of Social Sciences, Ersta Sköndal Bräcke University, Box 441, Sköndal, SE-128 06, Sweden

[22]School of Health and Welfare, Halmstad University, Box 823, Halmstad, SE 301 18, Sweden
[23]Department of Social Work, Norwegian University of Science and Technology, Trondheim, NO-7491, Norway

v2 **First published:** 23 Jun 2020, **3**:39
https://doi.org/10.12688/hrbopenres.13077.1
Latest published: 03 Dec 2020, **3**:39
https://doi.org/10.12688/hrbopenres.13077.2

Abstract

Background: This protocol outlines research to explore family members' and paid staff's perceptions of the impact of COVID-19 on individuals with intellectual and developmental disabilities and their caregivers. Evidence suggests that people with intellectual and developmental disabilities experience disparities in healthcare access and utilisation. This disparity was evident early in the pandemic when discussions arose regarding the potential exclusion of this population to critical care.

Methods: An anonymous online survey will be conducted with caregivers, both family members and paid staff, to explore their perceptions of the impact of COVID-19 in terms of demographics, living arrangements, access to services, social distancing, and carer wellbeing. The survey will be developed by the research team, many of whom are experts in intellectual disability within their own jurisdictions. Using back-translation our team will translate the survey for distribution in 18 countries worldwide for international comparison. The survey team have extensive personal and professional networks and will promote the survey widely on social media with the support of local disability and advocacy agencies. Statistical descriptive and comparative analyses will be conducted. Ethical approval has been obtained for this study from University College Dublin's Human Research Ethics Committee (HS-20-28-Linehan).

Dissemination: Study findings will be prepared in a number of formats in order to meet the needs of different audiences. Outputs will include academic papers, lessons learned paper, practice guidelines, reports, infographics and video content. These outputs will be directed to families, frontline and management delivering disability services, national-level policy makers, healthcare quality and delivery authorities, national pandemic organisations and international bodies.

Keywords

Caregivers Carers, Coronavirus, COVID-19, Health Disparity, Intellectual and Developmental Disability, Intellectual Disability, Pandemic

This article is included in the Coronavirus (COVID-19) collection.

Open Peer Review

Reviewer Status ✓ ✓

	Invited Reviewers	
	1	**2**
version 2 (revision) 03 Dec 2020		✓ report
version 1 23 Jun 2020	✓ report	? report

1. **Gloria L. Krahn**, Oregon State University, Corvallis, USA

2. **Germain Weber**, University of Vienna, Vienna, Austria
Elisabeth Zeilinger, University of Vienna, Vienna, Austria

Any reports and responses or comments on the article can be found at the end of the article.

Corresponding author: Christine Linehan (christine.linehan@ucd.ie)

Author roles: Linehan C: Conceptualization, Funding Acquisition, Methodology, Project Administration, Supervision, Visualization, Writing – Original Draft Preparation, Writing – Review & Editing; **Araten-Bergam T**: Conceptualization, Methodology, Writing – Review & Editing; **Baumbusch J**: Writing – Review & Editing; **Beadle-Brown J**: Conceptualization, Methodology, Writing – Review & Editing; **Bigby C** : Conceptualization, Methodology, Writing – Review & Editing; **Birkbeck G**: Conceptualization, Data Curation, Methodology, Visualization, Writing – Original Draft Preparation, Writing – Review & Editing; **Bradley V**: Conceptualization, Methodology, Writing – Review & Editing; **Brown M**: Conceptualization, Methodology, Writing – Review & Editing; **Bredewold F**: Conceptualization, Methodology, Writing – Review & Editing; **Chirwa M**: Conceptualization, Methodology, Writing – Review & Editing; **Cui J**: Writing – Review & Editing; **Godoy Gimenez M**: Writing – Review & Editing; **Gomiero T**: Writing – Review & Editing; **Kanova S**: Conceptualization, Methodology, Writing – Review & Editing; **Kroll T**: Conceptualization, Methodology, Writing – Original Draft Preparation, Writing – Review & Editing; **MacLachlan M**: Conceptualization, Methodology, Writing – Review & Editing; **Mirfin-Veitch B**: Writing – Review & Editing; **Narayan J**: Conceptualization, Methodology, Writing – Review & Editing; **Nearchou F**: Conceptualization, Methodology, Writing – Review & Editing; **Nolan A**: Conceptualization, Methodology, Writing – Review & Editing; **O'Donovan MA**: Conceptualization, Methodology, Writing – Review & Editing; **Santos FH**: Conceptualization, Methodology, Writing – Review & Editing; **Siska J**: Conceptualization, Methodology, Writing – Review & Editing; **Stainton T**: Writing – Review & Editing; **Tideman M**: Conceptualization, Methodology, Writing – Review & Editing; **Tossebro J**: Conceptualization, Methodology, Writing – Review & Editing

Competing interests: No competing interests were disclosed.

Grant information: Health Research Board, Ireland [COV19-2020-028]
The funders had no role in study design, data collection and analysis, decision to publish, or preparation of the manuscript.

How to cite this article: Linehan C, Araten-Bergam T, Baumbusch J *et al.* **COVID-19 IDD: A global survey exploring family members' and paid staff's perceptions of the impact of COVID-19 on individuals with intellectual and developmental disabilities and their caregivers. [version 2; peer review: 2 approved]** HRB Open Research 2020, **3**:39 https://doi.org/10.12688/hrbopenres.13077.2

First published: 23 Jun 2020, **3**:39 https://doi.org/10.12688/hrbopenres.13077.1

REVISED **Amendments from Version 1**

This is a revised version of the original study protocol which addresses the commentary of the reviewers. The following editorial changes have been applied. The number of participating countries expanded from the original study protocol and this is reflected in the authorship and listed countries. The title of the study has been amended to more appropriately reflect the fact that data are reported by caregivers. The most substantial edit relates to the rationale for limiting respondents to family members and paid staff which is outlined in detail. A clarification is made to the dissemination strategy. These issues are further detailed in the authors' response to each of the reviewers which is published with this revised manuscript.

Any further responses from the reviewers can be found at the end of the article

Introduction

Intellectual and developmental disability is a term, growing in usage, which acknowledges that intellectual disability is often accompanied by other disabilities including, but not limited to, sensory disability, speech and language difficulties, seizures, behavioural disorder, or difficulties with movement[1]. Intellectual disability is diagnosed as deficits in intellectual functioning, deficits in adaptive behaviour, and onset during the developmental period[2–4]. An estimated 1% of the world's population has intellectual disability with higher proportions living in low income countries[5]. Using a global population of 7.7 billion[6], it can be estimated that approximately 77 million persons worldwide live with intellectual disability, many of whom will present with additional disabilities such as those listed above.

A paradigm shift in models of disability emphasises the critical need for appropriate support to be available to people with intellectual disability. This position advances previous understandings of disability as either a medical condition requiring a 'treatment' or 'cure'[7,8] or as a social concept requiring attitudinal and environmental change[9]. Developed by the American Association on Intellectual and Developmental Disabilities, the support needs model argues that good quality outcomes for people with intellectual disabilities are a function of the support they receive, or *'put another way, if supports were removed, people with ID (intellectual disability) would not be able to function as successfully in typical activities and settings'*[4]. The COVID-19 pandemic has disrupted access to supports typically received by people with intellectual and developmental disability and has placed additional challenges on mainstream systems to make adjustments to accommodate need. The impact of these challenges has yet to be empirically assessed.

The United Nation's Convention on the Rights of Persons with Disabilities affirms the right of persons with disabilities to full inclusion and participation in all aspects of life, charging signatories to the Convention with organising, strengthening and extending support services. Addressing the need for full inclusion, global efforts in deinstitutionalisation have resulted in growing numbers of individuals with intellectual and developmental disability being moved from institutional settings to the community to live in their own home, or in small dispersed community housing often owned by agents such as disability services or mainstream social services[10,11]. Paid staff provide various levels of support from drop-in to 24/7. Across all settings, including the family home, there is evidence that appropriate support, drawing on practices such as Active Support, are necessary to promote good quality of life outcomes[12,13]. Caregivers therefore play a critical role in the current pandemic. Among many concerns are the impact of caregivers contracting the virus, the challenge of ensuring continuity of care for those who live in community settings, and the situations of those who live and work within large institutional settings.

Article 25 of the Convention specifically affirms the right of persons with disabilities to enjoy the highest attainable standard of health without discrimination on the basis of disability. Despite this protection, many individuals with intellectual and developmental disability experience significant disparities in the prevalence of adverse health conditions and behavioural disorder, attention to healthcare needs, preventative care/health promotion activities, and access to health care[14]. These disparities were brought into sharp focus by a UK confidential inquiry into mortality which revealed that avoidable deaths from causes amenable to change by good quality healthcare are twice as likely among this population when compared to the general population[15]. As the COVID-19 pandemic evolves, questions have arisen in the US regarding the legality of specifically withholding COVID-19 treatment from individuals with severe intellectual disability[16]. Questions have also arisen in the UK where COVID-19 guidelines produced by the National Institute for Health & Care Excellence were deemed to reduce access to critical care for those with intellectual and developmental disabilities, forcing an immediate modification[17].

The present study aims to collect survey data on family members' and paid staff's perceptions of the impact of COVID-19 on individuals with intellectual and developmental disabilities and their caregivers throughout 18 international jurisdictions. To the authors' knowledge this will be the first international dataset on this topic.

Protocol

This research study comprises an international survey of family members and paid staff who provide support to individuals with intellectual and developmental disability. The survey will be conducted by an international network of academics and practitioners. The research team will develop and disseminate an anonymous online survey for completion by family members and paid staff addressing two core questions. Firstly, what is their perception of the impact of COVID-19 pandemic on individuals with intellectual and developmental disability and their caregivers, for example, in access to healthcare and impact of restrictive practices? Secondly, do differences exist in the self-reported experiences of those living in different living arrangements and in different international jurisdictions?

The Principal Investigator is Chair of the Comparative Policy and Practice (CPP) special interest group of IASSIDD, the International Association for the Scientific Study of Intellectual and Developmental Disabilities, the leading professional association in the intellectual and developmental disability field. The CPP membership includes some of the foremost intellectual disability researchers in their respective countries who have previous experience collaborating on research. This group has been supplemented by non-CPP members who bring other expertise to the project including data management, data analysis and translation skills.

To promote clear communication and understanding among the study team, a logic model has been developed outlining the study's inputs, activities, outputs, short-term outcomes and long-term outcomes. The model provides a useful graphic to concisely encapsulate key areas of the study. The logic model is presented in Figure 1 below.

Design

This study is a cross-sectional, anonymous, online survey of adult caregivers, comprising family members and paid staff, who support individuals with intellectual and developmental disabilities. This open survey will be hosted online using the platform Qualtrics Core XM™. The study team, representing 18 countries - Australia, Brazil, Canada, China, Czech Republic, Germany, Greece, India, Ireland, Israel, Italy, Netherlands, Norway, Spain, Sweden, UK, US, and Zambia - will play a key role in creating awareness of the survey in their countries. It is hoped that additional countries may participate if representatives with expertise in this field can be sourced. Additional ethical approval will be sought, as required, for their inclusion.

An advisory group, comprised of a number of study team members, will be established to ensure standardisation in the promotion of the survey. A promotional pack will be developed to include a note outlining the background of study, its purpose and global reach, survey inclusion criteria, ethical approval as well as the study team and funder. A link and a QR code to the survey will also be provided.

Recruitment of study participants

The participants in this study will self-select to complete the online survey. To facilitate recruitment the study team will engage in a number of activities to promote awareness of the survey and consequently notify caregivers about the survey. Firstly, members of the study team will compile a listing of relevant disability and advocacy organisations within their jurisdiction. The target organisations include those that provide services and support to individuals who have intellectual and developmental disabilities or their family members, ranging from formal state services to informal social media support groups. Promotional information will be shared by the team members in their respective countries and following this, organisations will be invited to disseminate a survey link to staff and more widely through their communication and social media channels. As the study

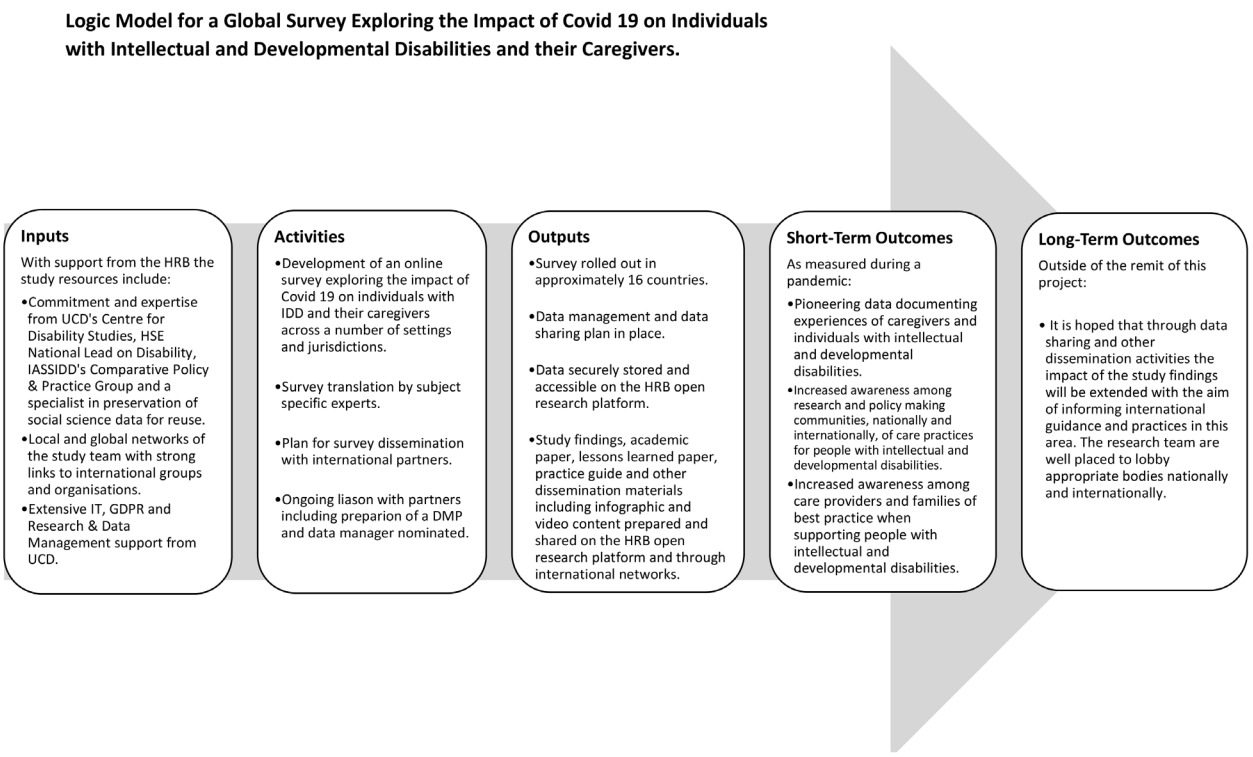

Figure 1. Logic model for coronavirus disease 2019 (COVID-19) intellectual and developmental disabilities (IDD) research project.

team includes intellectual disability experts in their respective countries, it is anticipated that they can encourage disability and advocacy agencies within their jurisdictions to promote the survey. The study team also anticipates the survey will 'go viral' whereby information will snowball beyond the immediate efforts of the team. Similar online surveys of the general population's experiences during the pandemic have successfully employed this methodology, enjoying high response rates[18,19].

Sample size calculation

Attempts to determine sample size are challenging. Firstly, to the authors' knowledge no international online survey of caregivers has previously been undertaken during a pandemic which might provide a reliable estimate. Secondly, in many countries the numbers of persons with intellectual and developmental disabilities and the numbers of caregivers is unknown. Reviews of prevalence estimates of intellectual and developmental disabilities are typically restricted to intellectual disabilities and use administrative rather than population-based data[20] Ireland, as the lead country in this research, has data available on the numbers of persons with intellectual disability and caregivers from the national census, which in 2016 were recorded as 66,611 and 195,263[21] respectively, albeit the latter figure is for all caregivers, not just those supporting people who have intellectual and developmental disabilities. Figures for those employed in disability services are equally challenging as, in Ireland, they are reported as Whole Time Equivalent posts rather than individual staff members. To December of 2019, a total of 18,515 Whole Time Equivalent posts were reported working in disability services in Ireland[22]. Attempting to determine the proportions of caregivers who provide support for persons with intellectual and developmental disabilities, and the actual number of personnel who provide staff support to this population is speculative and therefore has not been determined. Given that sample size calculations are required to determine if sufficient statistical power is available to researchers, it can be argued that if an average of 200 individuals participate in this survey from each of the jurisdictions, the combined sample of 3,200 responses would be more than sufficient to provide the statistical power for any analyses.

Survey instrument

The anonymous online survey will use only closed quantitative items. While the survey is presented to participants in different languages, the survey software, Qualtrics™, enables participants to enter their data onto one global dataset. The survey will comprise seven sections, exploring: characteristics of respondents (e.g. gender, age, status of family member or staff); characteristics of person(s) supported (e.g. level of intellectual disability, presence of additional disability, living arrangements); local practices during the pandemic in family home or workplace (e.g. restrictions to typical activity, introduction of new practices, equipment); access to information and training; experience of symptoms, testing, treatment; impact of social distancing; two standardised scales for caregivers measuring mood and the impact of pandemic. Piloting will determine if the format and length of survey is appropriate. Using a process of back translation, the study team will translate the survey into local languages.

Outcome measures

The primary outcome of interest in this survey is family members' and paid staff's perceptions of the impact of COVID-19 on individuals with intellectual and developmental disabilities and their caregivers. The perception of outcomes for people with intellectual and developmental disability are explored generally throughout the survey, but specifically in questions relating to access to health services and protective equipment, continuity of care, adverse impact of restrictions and questions relating to their experiences of symptoms, testing and treatment. The perception of outcomes for caregivers are also explored generally throughout the survey, but specifically in questions relating to mood and impact, using the DASS 12[23] and Coronavirus Anxiety Scale[24], and questions relating to their experiences of symptoms, testing and treatment.

Data analysis and statistical plan

All data will be analysed using IBM SPSS Version 26 statistical software. Descriptive, comparative, bivariate and multivariate analysis will be conducted to document the circumstances of the respondents and the people they support. Of particular interest are comparative analyses to explore trends by different living arrangement and different jurisdictions.

Ethics

Ethical approval for the study has been obtained from the host institution University College Dublin [HS 20-28-Linehan]. The study team will each assess the requirement for ethical approval within their own jurisdiction. It is not envisaged that the study team will apply for ethical approval from individual disability agencies as to do so would take considerable time and would likely create a circle of amendments which would collectively need to be agreed by all parties. It is also important to note that this research is an anonymous survey. There is no opportunity for participants to identify either themselves or the organisations they work for or engage with.

The study team acknowledge the sensitive nature of this topic and identified a number of ethical issues to the ethical approval body with actions to respond to each issue. Individuals who have intellectual and developmental disability, or support a person or persons with intellectual and developmental disability, may have experienced adverse effects to social distancing, may have contracted the virus or know of family and/or friends who did, and indeed may have experienced the death of family and/or friends to the virus. Reflecting on their experiences, and the experiences of those they support, it is possible that some participants may become distressed when completing the survey. For this reason, participants are directed to national and/or local support services should they wish to avail of support.

The study team is also cognisant of the fact that participants may be aware of cases of abuse, neglect and/or exploitation during this period. Given the anonymous nature of the survey, and the use of closed items, participants will be unable to detail these

experiences to the study team. Instead, participants will be advised to direct any such concerns to the appropriate relevant authorities which will be identified for their attention.

The study team acknowledge that the study does not involve people with intellectual and developmental disabilities in its design or collection of data instead relying on proxy perspectives and experiences of family members and paid staff. This is a limiting factor as critical experiences and views will be missing from the findings. This study was funded as a 'rapid' grant at a time when many of the researchers were in lockdown and many disability services were severely restricted in the services they could provide, notably as their focus was on reducing risk of infection and maintaining high quality support, both of which were impacted by staff absences caused by the need to self-isolate. A pragmatic decision was required as to what methodology was feasible and an online and anonymous survey of caregivers was deemed most appropriate. It is the opinion of the researchers, that typical methodologies which include the direct participation of individuals with intellectual and developmental disabilities, such as interviews and focus groups, were not feasible options at this time. The availability of the various types of individualized support usually provided by advocates, self-advocacy groups and service providers required to enable people with intellectual and developmental disabilities to participate in surveys or other forms data collection were simply unavailable given the pressures created by the pandemic. It could be argued that individuals with low support needs may not have required additional support to complete a survey tailored to their needs. Strict lock down regimes in many countries, however, meant that even among this group many did not have access to or familiarity with digital devices necessary to complete the survey. It is also important to recognize that even in the best of circumstances there will always be a group of people with more severe and profound intellectual disabilities whose direct experiences cannot be ascertained through spoken or other symbolic forms of communication. For this group, reliance on others who know them well, such as family or support workers is often the only way of gaining insights into their experiences. To address this limitation the researchers have engaged with Inclusion International who will support a group of self-advocates, preferably from the participating countries, to guide the interpretation of findings and recommendations. It is also important to note that since emerging from lockdown, a number of the research teams are actively working on studies that will directly capture the lived experience of individuals with intellectual and developmental disabilities during the COVID-19 pandemic, notably in Ireland, the Netherlands and USA. It is hoped that findings from this survey will inform these studies.

Finally, ethical issues arise regarding the security and anonymity of the online survey, notably as participants are informed that the dataset will be uploaded to an open data portal for use by other researchers. To address these issues, the study team will develop an anonymous survey, without collecting IP addresses that may provide a link to participation, and will use closed response items to ensure that no communication that may be identifiable can be received from participants. The survey is hosted on the Qualtrics platform which has been successfully used in previous large-scale surveys by the lead institution, University College Dublin in Ireland.

Data management and dissemination

A comprehensive data management plan will be developed by the study team and all data gathered will be shared on an approved data repository. The data management plan is due for publication on the HRB Open Research in Month 3 of the project. In preparation for this, the study team used an adapted version of the Data Value Map[25], as a discursive template to facilitate a conversation about the data management requirements for this study. In line with the HRB Data Management Planning template, issues relate to data collection, data storage, data analysis, data sharing and preservation, and ethical and legal requirements, as well as who will be responsible for each stage. This discussion helped to forge an appreciation of the open research lifecycle among the study team and the value that will accrue from this process. A visualisation of this plan, developed by Gail Birkbeck (co-author), is presented in Figure 2.

Study findings will be prepared in a number of formats in order to meet the needs of different audiences. These outputs include academic papers, lessons learned paper, practice guidelines, reports, infographics and video content. These outputs will be directed to the following stakeholders: people with intellectual and developmental disabilities and their families via local advocacy groups; frontline and management delivering disability services; national-level policy makers, healthcare quality and delivery authorities; national pandemic organisations (e.g. the National Public Health Emergency Team, the Health Protection Surveillance Centre); and international bodies responding to the pandemic such as WHO, Fundamental Rights Agency, Amnesty, and EASPD (European Association of Service Providers for Persons with Disabilities). The research team, via IASSIDD and their own personal contacts, also have significant networks to realise extensive dissemination, knowledge exchange and data sharing of the findings and learnings from this survey. While the lead investigator will take responsibility for global dissemination, country leads will be encouraged to undertake dissemination for their own jurisdiction.

When completed, this study will be reported using CHERRIES: Eysenbach G. Improving the quality of Web surveys: the Checklist for Reporting Results of Internet E-Surveys (CHERRIES). J Med Internet Res. 2004; 6(3):e34.

Study status

This study has not yet commenced data collection.

Conclusion

Despite the protections of the UN Convention on the Rights of Persons with Disability, people with intellectual and

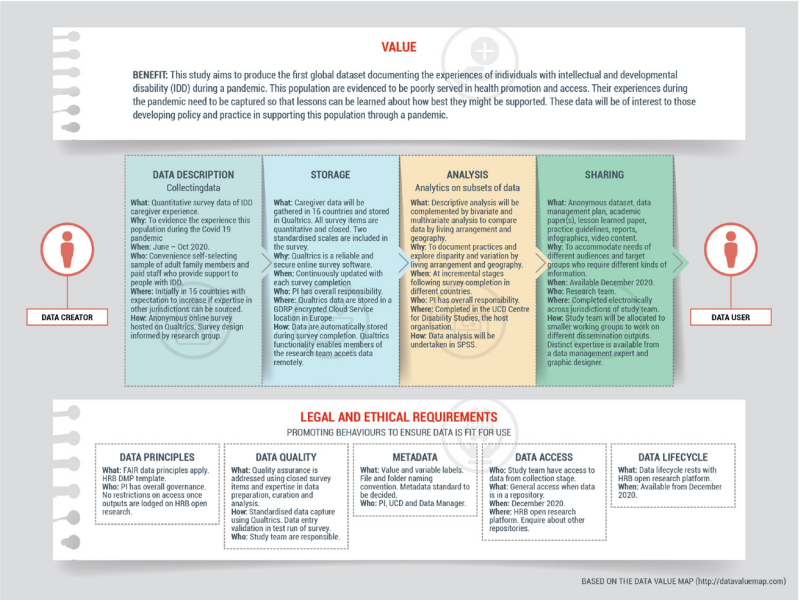

Figure 2. Visualised data management plan for coronavirus disease 2019 (COVID-19) intellectual and developmental disabilities (IDD) research project.

developmental disabilities are at risk of health disparities when compared with the general population. This study aims to gather international data on the experiences of individuals with intellectual and developmental disabilities and their caregivers during the COVID-19 pandemic. These data will provide a first glimpse of the challenges which arose for this population and their caregivers during the pandemic. Of particular interest is whether experiences varied by living arrangement and by country, and whether lessons can be learned to inform policy and practice for future pandemics. By depositing the anonymous dataset on an open forum, other researchers are encouraged to continue the exploration of these data.

Data availability

Underlying data

No data are associated with this article.

References

1. **US Department of Health and Human Services**. Accessed 25 May 2020.
Reference Source
2. World Health Organization: **International statistical classification of diseases and related health problems.** World Health Organization. 2004.
Reference Source
3. American Psychiatric Association: **Diagnostic and statistical manual of mental disorders (DSM-5®)**. American Psychiatric Pub. 2013.
Publisher Full Text
4. Schalock RL, Borthwick-Duffy SA, Bradley VJ, *et al.*: **Intellectual disability: Definition, classification, and systems of supports.** American Association on Intellectual and Developmental Disabilities. 444 North Capitol Street NW Suite 846, Washington, DC 20001. 2010.
Reference Source
5. Maulik PK, Mascarenhas MN, Mathers CD, *et al.*: **Prevalence of intellectual disability: a meta-analysis of population-based studies.** *Res Dev Disabil.* 2011; **32**(2): 419–36.
PubMed Abstract | Publisher Full Text
6. United Nations: **World Population Prospects 2019: Highlights**. Accessed 24 May 2020.
Reference Source
7. Drum CE: **Models and Approaches to Disability.** In: CE Drum, GL Krahn, & H Bersani, editors. *Disability and Public Health*. Washington: American Public Health Association. 2009; 27–44.
Publisher Full Text
8. Devlieger P, Rusch F, Pfeiffer D: **Rethinking disability: The emergence of new definitions, concepts and communities**. Garant. 2003.
Reference Source
9. Oliver M: **Understanding disability: From theory to practice**. Macmillan International Higher Education, 1995.
Publisher Full Text
10. Linehan C, O'Doherty S, Tatlow-Golden M, *et al.*: **Mapping the national disability policy landscape**.
Reference Source
11. Linehan C, Carr A: **Personalised Supports.** In: E Braaten, editor. *SAGE Encyclopaedia of intellectual and developmental disorders.* London: SAGE. 2018.
Publisher Full Text
12. Thompson JR, Bradley VJ, Buntinx WH, *et al.*: **Conceptualizing supports and the support needs of people with intellectual disability.** *Intellect Dev Disabil.* 2009; **47**(2): 135–46.
PubMed Abstract | Publisher Full Text
13. Bigby C, Beadle-Brown J: **Improving quality of life outcomes in supported accommodation for people with intellectual disability: What makes a difference?** *J Appl Res Intellect Disabil.* 2018; **31**(2): e182–200.
PubMed Abstract | Publisher Full Text

14. Krahn GL, Hammond L, Turner A: **A cascade of disparities: health and health care access for people with intellectual disabilities.** *Ment Retard Dev Disabil Res Rev.* 2006; **12**(1): 70–82.
PubMed Abstract | **Publisher Full Text**

15. Heslop P, Blair PS, Fleming P, *et al.*: **The Confidential Inquiry into premature deaths of people with intellectual disabilities in the UK: a population-based study.** *Lancet.* 2014; **383**(9920): 889–95.
PubMed Abstract | **Publisher Full Text**

16. Bagenstos SR: **May Hospitals Withhold Ventilators from COVID-19 Patients with Pre-Existing Disabilities? Notes on the Law and Ethics of Disability-Based Medical Rationing. Notes on the Law and Ethics of Disability-Based Medical Rationing (March 24, 2020)**. 2020.
Reference Source

17. **NICE updates rapid COVID-19 guideline on critical care**. 2020.
Reference Source

18. **Coronavirus: Only 37% feel others have changed behaviour in public, survey shows**. Accessed 25 May 2020.
Reference Source

19. **The Big Corona Study.** Accessed 25 May 2020.
Reference Source

20. Friedman DJ, Gibson Parrish R, Fox MH: **A review of global literature on using administrative data to estimate prevalence of intellectual and developmental disabilities**. *J Policy Pract Intellect Disabil.* 2018; **15**(1): 43–62.
PubMed Abstract | **Publisher Full Text** | **Free Full Text**

21. Central Statistics Office: **Census of Population 2016 – Profile 9 Health, Disability and Carers**. 2016; Accessed 25 May 2020.
Reference Source

22. Health Service Executive: **National Service Plan 2020**. Dublin: Health Service Executive. 2019.
Reference Source

23. Osman A, Wong JL, Bagge CL, *et al.*: **The Depression Anxiety Stress Scales-21 (DASS-21): further examination of dimensions, scale reliability, and correlates.** *J Clin Psychol.* 2012; **68**(12): 1322–1338.
PubMed Abstract | **Publisher Full Text**

24. Lee SA: **Coronavirus Anxiety Scale: A brief mental health screener for COVID-19 related anxiety.** *Death Stud.* 2020; **44**(7): 393–401.
PubMed Abstract | **Publisher Full Text**

25. Nagle T, Sammon D: **The data value map: a framework for developing shared understanding on data initiatives.** In: *Proceedings of the 25th European Conference on Information Systems (ECIS).* Guimarães, Portugal. 2017; 1439–1452.
Reference Source

Open Peer Review

Current Peer Review Status: ✔ ✔

Version 2

Reviewer Report 09 December 2020

https://doi.org/10.21956/hrbopenres.14289.r28498

✔ **Germain Weber**

Department of Clinical and Health Psychology, Faculty of Psychology, University of Vienna, Vienna, Austria

The revised version is fine with me, as authors have clearly switched their focus and study aim to the perception of family members and front-line staff on COVID19 related effects for people with IDD! Further authors elaborate on a pragmatic justification for not having included the direct voice of people with IDD in this survey.

Our earlier recommendation to make use of effect values and not p-values in data analysis remains.

With regard to the section “Sample size calculation”, expected responses of 200 per jurisdiction would come up to 3.600 as the revised version includes 16 countries!

Competing Interests: No competing interests were disclosed.

I confirm that I have read this submission and believe that I have an appropriate level of expertise to confirm that it is of an acceptable scientific standard.

Version 1

Reviewer Report 16 September 2020

https://doi.org/10.21956/hrbopenres.14176.r27914

?

Germain Weber
Department of Clinical and Health Psychology, Faculty of Psychology, University of Vienna, Vienna, Austria
Elisabeth Zeilinger
Faculty of Psychology, University of Vienna, Vienna, Austria

The research protocol as described is well planned and organized. The topic is of eminent relevance with regard to health provision and equal opportunities for people with intellectual disability in a period of pandemic. Though the authors do not mention it, the research as planned refers directly to article 11 of the UN-CRPD.

Having said this, we have to address one major concern and several additional issues listed below:

Major concern:

- Authors state that the main goal of the study is to assess the **experience and the impact** of the pandemic **of and on persons with ID** and their caregivers. However, persons with ID are not directly included in the study. We highly recommend the consortium to respect the requirements of the UN-CRPD and adhere to the principle “nothing about us without us”. We agree that it will take special effort to include persons with ID as direct participants in the study. However, the pandemic is not a good reason to exclude persons with ID from the study. Without the direct experience as expressed by persons with ID the study findings will be of limited impact. We therefor demand the consortium for a change and include people with ID in an acceptable way in the sample. In countries like the UK, Ireland, Australia, the Netherlands, Norway and France with substantial advances and supports for self-advocates this inclusion is more than realistic!

Further aspects:

1. We suggest to reflect on sample representativeness in a differentiate way. Even if in most countries, there is only fragmented knowledge about the characteristics of the sample, data collection should aim at some kind of representativeness. Otherwise, results cannot be generalised, and their full potential will remain unfolded. A representative sample might be randomly selected from a larger dataset after data collection, resulting in a stratified sample. Further, authors are not quite consistent when offering general epidemiological data on people with ID data in the first section of the introduction, and stating later in the section of sample size that the number of people with ID remains unknown in many countries.

2. Considering the statistical analysis, the fact that the total sample in this study will be far beyond 1,000 persons has to be addressed in the study protocol. We highly recommend authors not to use p-values for any interpretation. Using p-values for interpreting results is never advisable with this sample size though unfortunately still frequently done. With such a large sample size, every test will be “statistically significant” with p-values < 0.01. However, this would be an artefact resulting from the large samples size. Thus, we strongly recommend making use of effect sizes for interpretation, as they are independent of sample size.

3. We have some concerns with using only closed questions for data collection. The study design has the potential to explore all kinds of matters arising in relation to the pandemic,

even aspects that may not have been on the authors' minds. Thus, including open-ended questions will provide the opportunity to address aspects not specifically selected primary to data collection.

4. Finally, we advise authors to go more into detail with dissemination. The impact of the study will be highly dependent on a dissemination plan including various stakeholders. Many dissemination frameworks available can guide dissemination activities.

Thank you for offering us the opportunity to review this study protocol. May the consortium understand our concerns and recommendation in the way to transform a good study protocol in an excellent one!

Is the rationale for, and objectives of, the study clearly described?
Partly

Is the study design appropriate for the research question?
Partly

Are sufficient details of the methods provided to allow replication by others?
Yes

Are the datasets clearly presented in a useable and accessible format?
Not applicable

Competing Interests: No competing interests were disclosed.

Reviewer Expertise: Psychology and intellectual Disability. Health promotion and disability. Disability and inclusive research.

We confirm that we have read this submission and believe that we have an appropriate level of expertise to confirm that it is of an acceptable scientific standard, however we have significant reservations, as outlined above.

Author Response 02 Oct 2020

Christine Linehan, University College Dublin, Belfield, Ireland

(1) We thank Reviewers #2 and 3# for their combined review, and for describing the study as being of eminent relevance. We note that through no fault of their own, this review was received when data collection had just one week to run. While the benefits of open publishing of study protocols are many, one challenge is the possibility that rapid research may be near to completion by the time reviews are received. The reviewers may notice that we have added authors who were recruited to gather data in additional countries during this time period.

(2) We understand the reviewers' major concern regarding the fact that individuals with intellectual and developmental disabilities were not invited to complete the survey. We agree with the principle of 'nothing about us without us', however the pandemic did impact

on our choice of methodology. This study was funded as a 'rapid' grant at a time when many of the researchers were in lockdown and many disability services were severely restricted in the services they could provide, including supports to advocacy groups which we agree are present in some of the participating countries. Furthermore, in some countries at the time of the survey, the attention of many service providers was totally focused on reducing risk of infection to people with intellectual and developmental disabilities in their services and maintaining high quality support, and were experiencing extreme pressure in doing so by staff absences caused by the need to self-isolate.

A pragmatic decision was required as to what methodology was feasible and an online and anonymous survey of caregivers was deemed most appropriate. It is the opinion of the researchers, that typical methodologies which include the direct participation of individuals with intellectual and developmental disabilities, such as interviews and focus groups, were not feasible options at this time. The availability of the various types of individualized support usually provided by advocates, self-advocacy groups and service providers required to enable people with IDD to participate in surveys or other forms data collection were simply unavailable given the pressures created by the pandemic.

It could be argued that individuals with low support needs may not have required additional support to complete a survey tailored to their needs. Strict lock down regimes in many countries, however, meant that even among this group many did not have access to or familiarity with digital devices necessary to complete the survey. It is also important to recognize that even in the best of circumstances there will always be a group of people with more severe and profound intellectual disabilities whose direct experiences cannot be ascertained through spoken or other symbolic forms of communication. For this group, reliance on others who know them well, family or support workers is often the only way of gaining insights into their experiences.

We have made the following revisions to address this issue. We have revised the title of the study to highlight that it is caregivers' perceptions which are captured.
The original title was:
COVID-19 IDD: A global survey exploring the impact of COVID-19 on individuals with intellectual and developmental disabilities and their caregivers. The new title is:
COVID-19 IDD: A global survey exploring family members' and paid staff's perceptions of the impact of COVID-19 on individuals with intellectual and developmental disabilities and their caregivers.

We have also revised our protocol to include the fact that our findings will be reviewed in a consultation with an international forum of self-advocates from participating countries, an arrangement that was not formalized at the time the study protocol was published. Finally, it is important to note that a number of partners are conducting qualitative studies involving individuals with intellectual and developmental disabilities to give voice to their experiences during the pandemic, specifically these studies are planned in Ireland and in the US. These opportunities were not possible in the early stages of the pandemic when this study was planned. We hope the findings of this study will inform these studies.

(3) As noted to our first reviewer, to address the issue of representativeness, where national

data permits, we will weight the samples by level of intellectual ability (mild, moderate, severe/profound). Where national data is not available, we will weight the sample by international distributions of level of ability. We have revised the apparent contradiction where we present general epidemiological data yet state that the number of persons with intellectual and developmental disabilities remains unknown in many countries. A recent review of attempts to estimate the prevalence of IDD (Friedman et al, 2018) specifically excluded the term 'developmental disabilities' in favour of focusing on intellectual disabilities doi: 10.1111/jppi.12220.

(4) Where inferential statistics are conducted, we will present both p values and effect sizes which we agree is a transparent and appropriate reporting of difference.

(5) Where inferential statistics are conducted, we will present both p values and effect sizes which we agree is a transparent and appropriate reporting of difference.

Regarding the proposal to include qualitative questions on the survey, regrettably the reviewers' commentary was received with just one week remaining in data collection and we are therefore unable to change the survey. The rationale for closed questions was to facilitate back-translations of the survey into 15 languages and to avoid having another series of translations of respondents' qualitative data. The survey is substantive, taking 20-25 minutes to complete, and resources were scare with funding of just one research assistant. Finally, the researchers also wanted to ensure that no opportunities arose for respondents to threaten the anonymous nature of the survey through qualitative comments.

(6) We agree that a dissemination plan would be helpful. Our plan is that in each country lead researchers will be take responsibility for writing results into more accessible formats to complement the academic outputs, and where possible will assist in disseminating results to local governments, advocacy groups, and other interested stakeholders using the similar range of media to that used for recruiting participants.

(7) We thank Reviewers #2 and #3 for their helpful suggestions to improve the quality of this study.

Competing Interests: No competing interests were disclosed.

Reviewer Report 14 July 2020

https://doi.org/10.21956/hrbopenres.14176.r27575

Gloria L. Krahn

College of Public Health and Human Sciences, Oregon State University, Corvallis, OR, USA

The proposed study is critically important and timely; the authors/survey planners are commended for their speed in developing an approach to assess the impact of COVID-19 on a population that often gets forgotten. The article is well-written, incorporates many of the concerns that are being voiced now, and proposes to generate data that will provide valuable information.

Questions re: Appropriateness of Design relate to:

1. What efforts will be made to reflect any degree of representativeness in the sample (e.g., comparison with what is known about persons with IDD in each country)? Without some understanding of representativeness, how will findings be interpreted, and interpreted across countries? Will any options be available for responses from persons who do not have internet access?

2. The respondents will necessarily be those who have not become ill or very ill or died due to COVID-19. Will any data be available on hospitalizations or deaths to complement the survey findings?

3. The issue of only carer responses is a concern, as noted by the authors. Was thought given to supplementing with case studies with persons with IDD to capture some of their expressed experiences?

Questions re: Sufficient Details relate to the survey questionnaire still under development.

Because this is a prospective proposal, several comments are added for consideration:

1. Potential impact - in addition to the outcomes to be measured, will there be any questions related to value of training or materials for persons with IDD and carers in learning how to understand and cope with COVID.

2. Any other opportunities for collecting information on “valuable practices”?

3. The public health, medical, and general communities are learning so much about the disease as we gain more experience with it. Will information for each country be “time-stamped” in some way (e.g., date? Or time relative to when the virus became more prevalent in their country?)

4. What information will be collected about their current situation, such as access to clean water in low and middle income countries, or opportunities for social distancing in densely populated urban settings?

Thank you for the opportunity to comment.

Is the rationale for, and objectives of, the study clearly described?

Yes

Is the study design appropriate for the research question?

Partly

Are sufficient details of the methods provided to allow replication by others?

Partly

Are the datasets clearly presented in a useable and accessible format?
Not applicable

Competing Interests: No competing interests were disclosed.

Reviewer Expertise: Disability and public health; disability epidemiology and policy; data for health equity and IDD.

I confirm that I have read this submission and believe that I have an appropriate level of expertise to confirm that it is of an acceptable scientific standard.

Author Response 02 Oct 2020

Christine Linehan, University College Dublin, Belfield, Ireland

(1) We thank the Reviewer #1 for endorsing the critical importance of this study. We would like to inform the reviewer that we were asked to hold on a response until a second review was posted over two months later, hence the delay in responding.

(2) To address the issue of representativeness, where national data permits, we aim to weight the samples by level of intellectual ability (mild, moderate, severe/profound). Where national data is not available, we will weight the sample by international distributions of level of ability.

Only those with internet access can complete the survey, typically on smart phones, tablets or computers. Due to the length of the survey, and the complex branching logic for different respondents, it was not possible to circulate paper copies. Due to social distancing it was not possible for the researchers to lend direct support in completing the survey.

(3) We acknowledge that those who have died from COVID 19 are excluded and that those who are ill or very ill at the time of the survey will be likely excluded or underrepresented. We are hopeful that those who were previously ill or very ill will take part as we ask respondents about their own experience of contracting COVID-19, including possible hospitalisations. We also include questions as to whether the person(s) with intellectual and developmental disability respondents support contracted the virus, were hospitalized, and survived COVID-19.

(4) We agree the issue of caregiver-only responses is a concern. We refer the reviewer to our response below to Reviewers #2 and #3 who also raised this issue.

(5) Regarding the comments for consideration, we agree there is an opportunity to explore potential impact. We included questions on the availability and quality of training and easy to read materials, and we included standardised measures of caregiver mood and anxiety of COVID-19.

(6) In our forthcoming paper reporting on findings, we will ‘time stamp’ each country by

presenting information such as the total number of deaths and current cases in each country at the time of data collection.

(7) Regarding data on participants' current situation, we did not ask about access to clean water or density of population within regions, rather we addressed current situation by including questions on possible reorganization of staff shifts to reduce contacts, reduction in contact with family and friends, use of communications such as phone, internet to keep in touch etc.

(8) We thank the reviewer for taking the time to review this paper and offering some very useful insights to improve the study.

Competing Interests: No competing interests were disclosed.