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European Platform for Neurodegenerative Diseases

WP6 – Stakeholder involvement, external communication and dissemination

D6.3 – Roadmap and tools for stakeholder engagement

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Abstract

The two parts of this deliverable reflect the importance of working together with a broad range of stakeholders, such as researchers, clinicians, technicians, current and future patients, and members of the public, who have an interest in or may be affected by the outcome of the EPND project. EPND is about sharing clinical and research data related to neurodegenerative diseases. It is therefore important to ensure that their insight and perspectives are included throughout this research. People with neurodegenerative diseases are, of course, not only members of the general public, but also citizens, service users, stakeholders in many IMI projects and care professionals and patients in the clinical setting. The separation of this deliverable into two parts does not reflect an “us” versus “them” division but rather different approaches to involving different stakeholders in EPND.

Part 1 reports the activities and the action plan of Task 6.2, in enabling and supporting the active participation of EPND stakeholders. EPND stakeholders are heterogeneous. Therefore, we need to address challenges specific to transdisciplinary collaboration (TDC). We have reviewed TDC literature and selected, among the main theoretical frameworks, those most useful to EPND. Based on the selected reference frameworks, we performed internal consultations in close collaboration with other EPND WPs (WP7 in particular, but also WPs 1, 2, 3, 5, 8). We have examined the main hurdles and gaps, spontaneous solutions to be leveraged with our action, and potential tools to enable or support effective participation.

Clarifying and harmonizing language emerged as a priority; adapting methods across disciplines appeared as both a need and a unique EPND potential. We developed a Glossary consisting of 8 modules clarifying specialist terms to other professionals. We started an extensive review to extract common structure and best practices in using the target product profile (TPP), a tool mostly used to facilitate communication and co-development among different stakeholders in industry. This tool is being used to plan the EPND development systematically, and our review will support this effort as well as its uptake outside EPND, especially in academic contexts.

Since citizens are also stakeholders in IMI projects, we have performed a preliminary assessment of the feasibility of their participation in EPND, focusing so far mainly on the legal and ethical aspects. To assess the level of TDC and the impact of our action, we have extracted preliminary indicators based on the selected reference frameworks. Next developments include completing the Glossary with the wider participation of EPND experts, completing the assessment of feasibility of citizens’ participation, and fine-tuning, collecting and comparing the indicators of stakeholder engagement. Results are co-developed with, and in turn being fed to other WPs, especially WP7.

Part 2 of this deliverable draws on some of the Public Involvement (PI) work being conducted in this project by Alzheimer Europe in collaboration with the Spanish Parkinson’s Federation. PI is a concept and particular approach to involving people in research other than as research participants. In this project, it is about drawing on the unique insight that people with a neurodegenerative disease have, which they can share with researchers to improve the research process. The focus of the PI contribution to this EPND deliverable is on respectful and inclusive communication and about interaction with people with neurodegenerative diseases in the context of PI work.

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Acronyms

ADDI	Alzheimer's Disease Data Initiative
CRO	Contract Research Organization
CS	Citizen Science
CS4H	Citizen Science for Health
DOA	Description of Actions
DPIA	Data Protection Impact Assessment
ECSA	European Citizen Science Association
EPND	European Platform for Neurodegenerative Diseases
EU	European Union
FAIR	Findable Accessible Interoperable Reusable
FEP	Federación Española Parkinson (Spanish Parkinson's Federation)
GDPR	General Data Protection Regulation
GV	Gates Ventures
HTA	Health Technology Assessment
IMI	Innovative Medicine Initiative
M&E	Monitoring and Evaluation
NDD	Neurodegenerative Diseases
PI	Public Involvement
PIPA	Participatory Impact Pathways Analysis
PPP	Private Public Partnership
QC	Quality Control
R&D	Research and (technological) Development
SME	Small-Medium Enterprise
TDC	Transdisciplinary collaboration

ToC	Theory of Change
TPP	Target Product Profile
UNAIDS	The Joint United Nations Programme on HIV/AIDS
WG	Working Group
WHO	World Health Organization
WP	Work package

Part 1: Stakeholders and transdisciplinary collaboration (TDC)

To advantage clarity, we present our content in two sections, the first covering stakeholders in general, the second focused on patients. This separation is artificial, and should not be interpreted as a separation between the people with the disease and those without. Still, the methods used are different, and this content organization may facilitate reading. People with neurodegenerative diseases are, of course, not only members of the general public but also citizens and service users. In both sections, we use the concept of Public Involvement, to denote a particular approach to involving people in research other than as research participants. The separation of this deliverable into two parts does not, therefore, reflect an "us" versus "them" division but rather different approaches to involvement. This will be explained in each part of the deliverable.

1. Background and objectives

The main objective of Task 6.2, defining tools and roadmap for stakeholder involvement, consists of enabling and supporting effective collaboration among the heterogeneous professionals and stakeholders contributing to EPND, as well as those who will use EPND and will therefore determine its success. Collaboration across heterogeneous stakeholders cannot be taken for granted, and the challenges it poses should not be underestimated. Different stakeholders have different motivations, mandates, objectives, tools, procedures, and languages. As a public-private partnership involving 29 organizations and institutions from across Europe, EPND includes a wide range of stakeholders and requires specific support to a) achieve shared goals in a cost- and time-effective manner, and b) to make sure that our platform will meet concrete needs and will be used in practice. Such support consists of specific tools and methods that are not always required in a unitary discipline, where individual members are already aligned on vocabulary, methods, background technical knowledge, and a whole set of more or less implicitly shared context including values, beliefs, mandates or objectives.

In this deliverable, and especially in section I, we use the term "trans-disciplinary collaboration" (TDC) to reference multi-stakeholder engagement in projects such as EPND. "Trans-disciplinary" is the current term used in the field of Innovation Science to denote contexts where different stakeholders converge (Aboelela et al. 2007). TDC generally involves professionals from a range of specialist backgrounds, as well as societal stakeholders including patients and citizens as the ultimate beneficiaries of our research. The TDC concept highlights the value and importance of engaging stakeholders into efficient synergies to achieve concrete results through effective co-development where the participation of all necessary stakeholders is well-balanced.

This deliverable leverages the most innovative contributions in TDC and innovation science, and use systematic methods to identify and develop tools to support EPND activities, where the interaction across stakeholders can be difficult due to the mentioned heterogeneity. Effective collaboration between diverse stakeholders from different sectors, backgrounds and research

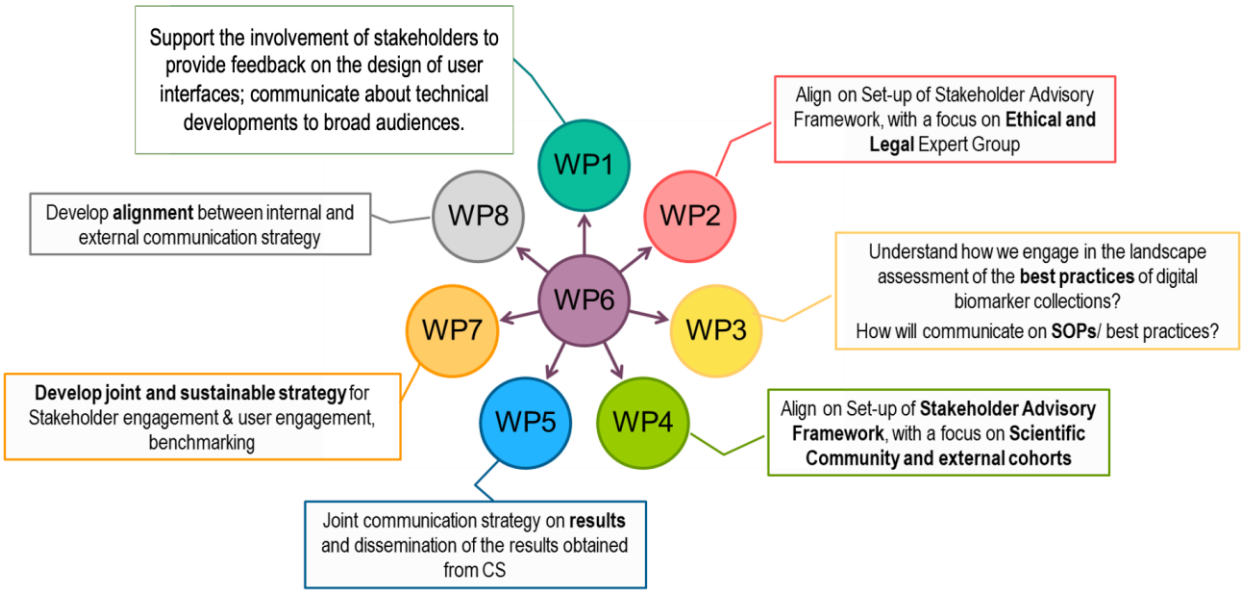
areas will allow EPND to deliver a sustainable and scalable platform for sharing of NDD samples and data in Europe, effectively answering users' needs.

In this deliverable, we summarize existing strategies to promote multi-stakeholder collaboration, that can be leveraged for best efficacy, and identify potential needs. We then aim to devise tools to meet these needs, and outline a roadmap to deploy and use such tools in EPND. We will also describe our effort to transfer methods that, typically used in a specific discipline, may also be useful to others after clarification and adaptation.

1.1 The challenge of transdisciplinary collaboration within EPND

The EPND ambition to build a platform making data and samples FAIR requires solving a variety of issues ranging from setting the digital tool enabling data and samples sharing, to overcoming legal hurdles to sharing, as well as to harmonizing the heterogeneous data from different cohorts to enable appropriate pooling and processing. This requires collaboration among partners whose competence is specific to very different fields: IT, legal, neuroscience, regulatory experts, etc. Indeed, to achieve its goals, many EPND tasks may primarily rely on their own specific language and methods (e.g., WP1 is mainly focused on the technical building of the platform; WP2 on legal aspects, etc.). However, this work has to be coordinated and integrated with wider perspectives from EPND partners in other areas and work-packages, and co-development is specifically required for some tasks (See Figure 1). A flexible mix of methods for collaborative engagement (see Part I, Section 1.3.1) are therefore required, in particular for work in cross-cutting WPs such as WP6 (communication), WP7 (sustainability) as well as in the overall coordination of EPND.

Figure 1. Critical dependencies of WP6 with the other WPs. Analogous interactions exist especially for WP7, but a high degree of interdependency is also required to all other WPs.



In addition to this fundamental challenge of effective collaboration with internal stakeholders, EPND aims to be useful to a large variety of *external* users and beneficiaries in the long run. Users may be academic researchers, or researchers from SMEs or big pharma companies. EPND also aims to engage a diverse range of data and samples depositors, who may be academic researchers, private companies, or hospitals who own large amounts of data they cannot steward or exploit. Understanding the motivations, interests, rewards, and constraints for these stakeholders in using EPND requires particular stakeholder engagement efforts. This exemplifies how successful TDC is critical to EPND's ambition of providing the platform of choice and being sustainable by design in the long term.

Considering that EPND aims to serve a whole ecosystem pointing at efficient and impactful translational research, relevant stakeholders may be even broader than those listed in Table 1 below (adapted from D6.1). The EPND stakeholder composition reflects in large amount this complexity. Supporting internal collaboration will also be useful to engaging with external stakeholders.

Table 1: The EPND stakeholder panorama (see also EPND Deliverable 6.1).

Primary audiences: core contributors and users of the EPND platform	
Data and sample contributors	Cohort leads (academic & industry)
	Neurodegeneration research projects
	Biobanks
	SMEs & CROs
Data and sample users	Clinical & biomedical researchers (academic & industry)
	Data scientists (academic & industry)
	SMEs & CROs
Landscape peers	Data and sample sharing initiatives
	Partner projects
	Professional networks & bodies (e.g., EADC)
Secondary audiences: key recipients and beneficiaries of outputs from EPND; groups that could drive adoption of EPND	
Research beneficiaries	Neurodegeneration research participants

	People with neurodegenerative diseases and caregivers
	Patient organizations
	General public
Research funders	EU & international funding agencies
	National funding agencies
	Charity, philanthropic and private sector funders
Decision-makers & amplifiers	EU & national policymakers
	Regulators & payers
	HTA
	Academic publishers
	Science media & journalism

Following are a few concrete examples depicting the complexity of collaboration across such different stakeholders:

- Building a hub supporting a platform that can be most useful for users requires a user friendly interface, providing the key services that motivate users and data and samples providers to engage increasingly with EPND, and assessing costs and benefits to guarantee effective engagement and sustainability. This involves WP1, WP2, WP4, WP6 and WP7

- Getting the target information from different potential users requires a good understanding of the pattern of rewards and motivation specific to their field. This requires EPND researchers, for example, to have both scientific and business expertise, and perform tasks of competitive intelligence. The challenge that arises here is in involving and collaborating with partners from other backgrounds with different types of expertise, alongside partners that are scientific or business experts. Communicating the tasks, objectives, and selecting and transmitting the relevant information and know-how requires a robust transdisciplinary approach to collaboration, enabling to transmit, acquire and use such new expertise effectively within the project time-line.

- Providing data obtained from different sources for being pooled and processed within single studies requires harmonization. Harmonizing data entails methodological expertise, as well as a complex range of information ranging from the features of specific biomarkers to quality assurance and regulatory requirements for the validity of studies based on such biomarkers. Again, the basic and advanced concepts of these disciplines may not all reside in single individuals and, even in this case, the work to be performed

may require additional collaborators who should quickly get and integrate the essential terms, logics and methods from such different field.

- Citizens, including patients, the ultimate target of our research, are also the ultimate owners of the data to be shared. Beyond their rights, already guaranteed by the law, they may have additional wishes or requirements on the use of their data for research purposes and extended re-use. Their attitudes are not always known to researchers, and may vary based on their own knowledge and understanding of research practices, aims and requirements. Importantly, such attitudes may even be much less restrictive than required by the laws aimed to guarantee citizens' own rights (see Part I, Section 2.3). The quality of the communication between citizens, patients and researchers can therefore affect trust and collaboration in a fundamental way. This requires to pay attention that technical terms be made accessible to any kind of stakeholder connected to the platform, and that specific channels and methods be devised to communicate and collaborate effectively with the wider community. In particular in the case of patients, specific attention should be paid to empowering and overcoming typical ways of communicating that make research efficient but are possibly experienced as discriminatory by our ultimate stakeholders, thereby hindering meaningful and truly effective collaboration (see Part II).

- Finally, all of the components developed through TDC must be developed in sync across WPs. The level of inter-disciplinarity required for such collaboration is of course highest, and tasks are performed by very experienced leading teams. Still, having the whole project working requires that all of the members involved at any level pick quickly new needs and methods and be able to comply with transdisciplinary interactions and tasks.

TDC is necessary not only to enable delivering the promised platform, but also to inform a business plan making it self-sustainable in the long run. EPND can thrive beyond the end of the funded project only if such different stakeholders are effectively involved to produce what is actually needed.

In particular in hyper-specialized contexts, integrating this heterogeneous set of information and competences requires a specific effort, tailored to each project. If this is well-known in innovation science (Le Dantec 2016; Latour 1987; Kiwanuka 2015) and in the technological field, this approach is not necessarily acquired in the field of neuroscience. The tasks defined in this Deliverable may therefore be of interest also outside EPND.

1.2 Objectives

The objectives outlined in Part I of this deliverable consist of assessing the kind of spontaneous functioning in support of stakeholder collaboration, identifying the primary needs to enable, support or boost stakeholder collaboration within -and consequently outside- EPND. We then aim to build the methodological tools deemed necessary to answer such needs, and devise a strategy for their use, that leverage what is already done spontaneously to overcome specialist

boundaries. Finally, previous research in innovation science and internal consultations will help to identify indicators allowing to assess the impact of such action and, if possible, to export the devised tools to a wider audience of users beyond EPND. All this eventually serves the definition and actualization of a Roadmap for stakeholder involvement.

1.3 Reference frameworks

As mentioned, TDC is still a relatively new concept in biomedical research. Participatory methods and initiatives are frequent in the field of cancer research, and also increasingly frequent in the field of neurodegenerative diseases (NDD). These usually involve patients, whose involvement poses specific challenges in the field of NDD. Part II of this Deliverable will detail such methods thoroughly, based on specific consolidated frameworks (Gove et al. 2018) and methods. On the other hand, as outlined in the previous section, EPND involves many more stakeholders, consistent with the transdisciplinary approach consolidated in the field of research and technological development (R&D) (Le Dantec 2016; Latour 1987; Kiwanuka 2015).

We identified three main reference frameworks which may help support collaboration among different stakeholders in EPND. These frameworks are:

1. the Quadruple Helix Framework, which describes systematic involvement of academia, industry, governmental institutions and civil society, a concept that is embedded in IMI projects,
2. the TDC Framework, which identifies increasing degrees of integration across heterogeneous stakeholders, and can support collaboration within and beyond EPND, and
3. the Citizen Science (CS) Framework, where citizens directly perform scientific activities, but which is under-developed in the NDD field.

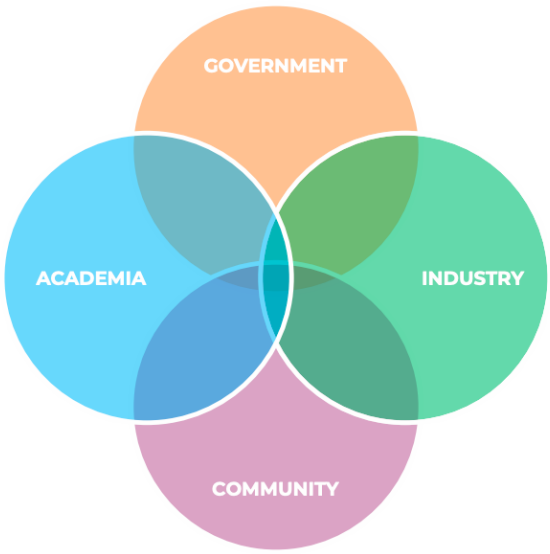
1.3.1 The quadruple-helix framework

The overarching approach, that incorporates other more specific approaches like TDC and CS, is the “Quadruple Helix” (Carayannis and Campbell 2009) (Figure 2). It is meant to enable building an effective innovation ecosystem, and consists of planning the systematic involvement of four major classes of stakeholders all along the development of innovative products. The four “helices” include Academia, Industry, Governmental Organizations and Civil Society. The approach does not differ in substance from what EU and IMI projects already require: indeed these stakeholder categories take part in the typical consortia of major European projects. However, the approach is still little known as a formal framework in biomedical research. Therefore, it may thus not be implemented systematically or effectively at all levels. Bringing it to explicit attention may clarify needs and methods to structure TDC more effectively: making sure that the parties of the Quadruple Helix be consistently represented at all EPND development steps and processes would help to achieve aims with wider societal interest, like

reducing research costs and increasing its efficiency by making data FAIR. Keeping the Quadruple Helix in mind would then help define advisory boards in a more balanced way, guaranteeing that all needs be taken care of and all perspective leveraged. This would also help compile key documents like the TPP, devise services to be provided, or networking that EPND may facilitate.

Within this wider framework, more specific approaches (see sections 1.3.2-3; (Aboelela et al. 2007; Gebbie et al. 2008 (Shirk et al. 2012)) aim to support concrete cooperation among stakeholders from such different "helices".

Figure 2. The Quadruple Helix approach (Carayannis and Campbell 2009) involves stakeholders coming from the pillars of academia, industry, governmental institutions (eg, policy makers, regulators) and civil society.



1.3.2 The trans-disciplinary collaboration (TDC) framework

Aboelela's approach (Table 2) frames the different kinds of collaboration across heterogeneous parties along a continuum ranging from a *multi*- to *inter*- to *trans*-disciplinary mode. These levels are characterized by an increasing integration of *language*, *methods* and *products*, going from the parallel work of groups, each using their own specific modalities, to levels of increasing translation and integration of languages and methods. The highest level of integration entails the generation of *new* languages and methods, answering the needs and incorporating the culture and know-how of contributors. Thanks to Aboelela's framework, we can "map" the current *modus operandi* of specific working groups including different professionals and stakeholders (see Section 2.4 and Table 6), identify desirable endpoints, for those processes whose efficiency

may be improved by greater trans-disciplinarity, plan the production of tools facilitating the achievement of those endpoints, and extract indicators summarizing progress towards such endpoints, highlighting any further needs to achieve them, and monitoring the impact of our action to enable adjustments.

As mentioned, different work processes in EPND cover, appropriately, any of these modalities, and not all need to be pushed to increased levels of integration. However, Aboelela's framework allows us to assess multi-stakeholder integration and identify tools and enablers that can support increasing integration whenever needed. For example, Table 2 shows language as a core dimension of TDC. If specialist language can be a hurdle, glossaries clarifying the meaning of terms required within the project can enable integration and support collaboration.

Table 2. Main dimensions reflecting increasing integration of professionals and stakeholders within the TDC framework (modified from (Aboelela et al. 2007)).

A	B	C	D	E
	Modus operandi	**Language**	**Methods**	**Publications**
Multi-disciplinary	Parallel	Specific	Specific	Team-specific
Inter-disciplinary	Coordinated	Translated	Mixed	Complementary sections merged
Trans-disciplinary	Integrated	Merged/ generated (project-specific)	Integrated/ adapted/ generated	Fully shared data generation and presentation

Indeed, research on language within TDC confirmed how the same term can assume different meanings in different contexts, with inconsistencies going unnoticed for long time and hurdling collaboration without participants even being aware of the problem until late in the project life (Hesketh et al. 2018). But beyond this basic issue, Aboelela illustrates how the highest levels of trans-disciplinarity potentially lead to the emergence of entirely new languages and methods, co-defined and shared by all members and specific to that project or context. Although apparently obvious and repeatedly advocated in the trans-disciplinary literature, this is not systematically accounted for in many contexts requiring transdisciplinary collaboration. Only a few previous examples show considerable degree of commitment in the definition of Glossaries (Kelly et al. 2022; Gainforth et al. 2021; Hesketh et al. 2018; Dopp et al. 2019), e.g.:

- Kelly et al. defined a consensus glossary to get aligned on food and physical activity policies (Kelly et al. 2022)
- Gainforth et al. harmonized language to integrate knowledge translation and improve conduction and dissemination of research in partnership, in the field of spinal cord injury (Gainforth et al. 2021)
- Hesketh et al. had to align on existing terms with entirely different meanings in different field and define the new language needed to the innovative field of storing digital information on DNA support (Hesketh et al. 2018)
- Dopp et al. defined a glossary to address user-centered design strategies for implementation experts (Dopp et al. 2019)
- Zhu et al. focused on the role of language and communication in group identity and activism to reduce stigma in people with rare diseases (Zhu et al. 2017).

The above efforts aimed to enable understanding within and outside specific projects or contexts. For projects, this includes enabling reviewers to properly assess deliverables or scientific contributions, increasing the impact and accessibility by non-specialists, and extracting a controlled vocabulary to make communication clear and effective (see Part I, paragraph 2.2.1). With the exception of some recent Public Involvement work (please see Part II), to our knowledge, such examples did not entail multidisciplinary professionals within multi-stakeholder projects in the field of NDD research so far.

Additional work in the transdisciplinary literature identified which attitudes, behaviors or competencies are most essential to TDC (Gebbie et al. 2008; Knapp et al. 2015). “Competencies”, as opposed to attitudes or personality features, can be trained, therefore this work highlights space for possible action to foster TDC. The competencies identified by Gebbie and colleagues (Gebbie et al. 2008) include:

- integrate concepts, theories and methods from different disciplines

- reading journals/attending lectures from other disciplines

- communicating regularly with scholars from other disciplines

- use a language meaningful to interdisciplinary teams

- modify own work based on interdisciplinary influx

- present at venues with interdisciplinary participants

- Engage colleagues from other disciplines to gain their perspectives

(See Table 3 in Gebbie et al, 2007 for the original detailed exhaustive list).

Feeding or soliciting such opportunities is indeed a feasible way to support TDC within EPND.

1.3.3 The Citizen Science framework

Citizen Science (CS) is variably defined by different organizations and professional bodies, but generally involves “scientific work undertaken by members of the public, often in collaboration with or under the direction of professional scientists and scientific institutions.” (Oxford English Dictionary). CS aims to boost the involvement of citizens in science, in a democratization effort, but also to increase the reach of research, that only with citizens’ help can obtain some kind of information, or obtain it in a timely manner. CS also aims at leveraging the computational power that citizens can provide, while enabling them to learn, qualify as researchers and even coauthor publications, depending on the kind of contribution they provided. CS includes but does not coincide with participatory research (e.g., research involving patient collaboration). Like participatory research, it is highly valued in EU projects. The ECSA, European Association for Citizen Science, defines the principles qualifying projects as proper CS, has specific thematic working groups to support CS development, and an EU-funded platform where citizens can find and connect to qualified CS projects.

EPND does not aim at providing a platform for CS. However, here we consider the possibility to enable citizens’ participation for multiple reasons. People with neurodegenerative diseases are first and foremost citizens, and therefore CS should be considered as a more general and fundamental framework. If citizens do not understand or do not feel involved in our work, people with neurodegenerative diseases may feel even more stranger and disconnected, and our efforts to involve them in participatory collaborations may be less effective. Even in projects that, like EPND, make a focused effort to involve patients directly, the CS framework may anyway support a more inclusive and natural involvement of patients, by creating an environment where citizen’s connection to scientific activities is easy and accessible. For example, patients may recognize their participation as similar to that of other citizens who may be contributing to data processing, while learning about biomarker validation. Within such framework, citizens, as well as patients and caregivers may all have direct contact and experience with research, exerting specific roles in scientific development. Targeting citizens rather than patients only and engaging more of them in research developments may also help address issues potentially leading to stigma or other prejudices that currently contribute to the low impact of research on the general community (see for example the low rate of dementia detection even in developed countries).

Citizens in general may be interested in accessing and getting to know more about neuroscience, neurodegenerative disorders, biomarkers, diagnostic procedures, and their development, and to know what is practically or theoretically possible for patients to improve their health. Enabling citizens to access and contribute to process some of the EPND data may give them access the research world, enabling to understand more of our efforts to cure neurodegenerative disorders, increasing proper information, trust and participation in science at some level. Such participation may also turn useful to EPND: as in other CS projects (see www.eu.citizen-science for ongoing examples) simple and time-consuming tasks that do not necessarily require specialist knowledge may well be performed by lay citizens after training. If

successful, such effort may eventually contribute to some extent to the EPND sustainability plan. We aim therefore to assess its feasibility, and to identify what is needed to make it happen.

The main reference framework for this kind of participation is provided by Shirk et al. (Shirk et al. 2012) (Table 3). It frames citizens' involvement within 5 levels of increasing integration. The first just entails citizens asking researchers to perform research on a topic of interest (Contract level), the second (Contribution) includes citizens' data collection. From the third level (Collaboration) does more concrete co-development occur, considering that here citizens contribute to study design or data processing. At higher levels, citizens are assisted by scientists or even develop scientific research in full autonomy, as multiple examples in physics, astronomy, meteorology and environment show (see for example activities by Vincenzo Galilei, Mary Anning, Fred Whipple, Rick Grocke, etc.).

Based on this framework, the kind of citizens' participation we may envisage for EPND would be at a relatively high level of interaction, involving Collaboration. The kind of activities that may presumably be performed range from data cleaning (e.g., spotting values out of the proper range) to simple processing (e.g., checking the appropriateness of results of brain scans segmented with automated algorithms, after some training). This level entails a considerable complexity. A feasibility analysis is therefore useful before launching any concrete action. From the EPND side, the level of commitment required to enable citizens understand and process data properly may not be negligible (preparing tutorial; monitoring performance; providing feedback; availability to answer questions and take care of relations). Moreover, the exact range of possible activities and the level of effort required should be assessed, in light of the possible return in terms of sustainability. Still, the most upstream challenge consists of the legal requirements to enable citizens to access patient data as those treated by EPND. Any further investigation on feasibility depends on the output of this preliminary assessment on legal/privacy feasibility, which is the work presented in this deliverable.

Table 3. Main reference framework on Citizen Science (modified from (Shirk et al. 2012))

Type of bond-collaboration	Corresponding action
Contract	Citizens ask scientists to conduct a scientific investigation and report on results
Contribution	Scientists ask citizens to collect and contribute data and/or samples
Collaboration	Citizens assist scientists to develop a study, and collect and analyze data for shared research goals

Co-development	Citizens develop a study with input from scientists
Qualified co-researchers	Citizens conduct independent research that advances knowledge in a scientific discipline

2. Trans-disciplinary collaboration in EPND

Based on the dimension selected from these reference frameworks, as a first step, we observed how EPND partners already support cooperation across different specialists. The structure of the interconnected EPND tasks is complex and inhomogeneous: not all WPs or tasks require the same operational modalities; ongoing activities may already work in an optimal manner, others may be supported or boosted by targeted action based on what we learned from the recent research on TDC. Here, we report some ongoing activities already adopting some degree of transdisciplinary modality thanks to the unsolicited initiatives of individuals with practical expertise in TDC contexts and based on the observation of some needs due to the complexity and the multi-stakeholder component of the project. We will then describe how targeted action is being proposed in a possibly ecological manner, building on such initiatives and leveraging already proposed solutions whenever possible. Such leverage is useful to optimize results, as well as to minimize friction possibly due to proposing changes to consolidated routines (Siedlok and Hibbert 2013).

We underline here that the same elements are considered in this deliverable under different points of view. For example, a first action in providing a rough Glossary is reported among the *ongoing activities* already happening in EPND, and selected for being leveraged in focused coordinated intervention. Then, a later section (see Section 2.2.1) will describe the methods used in active interventions to foster TDC through the *systematic development* of an EPND Glossary. Finally, analyzing the terms included in the glossary can provide an *indicator* of the degree of integration of heterogeneous stakeholders. Therefore, the apparent redundancy of recurrent topics (like the Glossary) serves these different features and roles in our analysis, development, and deployment of tools fostering TDC.

2.1 Previous EPND activities in support of TDC

- *From multi-/inter- to transdisciplinary collaboration through language harmonization*. The building of the digital infrastructure hosting the EPND services, performed within WP1, can be performed in a relatively independent manner, leveraging know-how, language and methods specific to the IT field. The main output will consist of digital tools that, although coordinated, will be produced in parallel to the output of other WPs. As well, publications covering such specific topics may mostly include authors with IT competence. At the same time, this work also requires

input from different WPs. This relates for example to how the hub should be structured, to enable discovery and use of data and samples by researchers. Similarly, communications within and outside EPND need some level of harmonization to get aligned and pass consistent messages. Indeed, consistently, WP1 started to harmonize the terms used to refer to different elements of EPND. This effort occurred simultaneously with other WPs realizing that harmonization on terms was needed, for example to refer consistently to cohort owners (or data donors), or to disambiguate when talking about the EPND "Platform" (therefore including the community and services offered by EPND) or the hub, meaning the digital infrastructure supporting the platform. WP1 also solicited a more precise use of terms, for example the term "metadata" may have different meanings for different persons. A further consistent, but entirely independent work on language was proposed from the beginning of the project, through the collection of acronyms. Due to the initial uncoordinated action on language, this effort has been repeated twice (first time during the DOA writing, and a second time after the project started). Based on the Aboelela framework, we proposed to curate a systematic Glossary, leveraging these previous documents and efforts, to support multi-WP transdisciplinary cooperation in a soundly structured way. Within this task, we included the Modules already created by WP1, as well as the acronym lists, using the definition provided by the most experienced contributor in case of duplicates, and added to the multi-module Glossary (see section 2.2.1).

- *Previously ongoing import and adaptation of methods across fields*. WP7 is pursuing the goal of devising an operational model enabling EPND to be a self-sustainable platform. This operation leverages methods imported from heterogeneous fields, like technological development and business. The participation of experts from pharmaceutical industry allowed to import and adapt a method typically used in drug development: the target product profile (TPP) (U.S. Department of Health and Human Services, Food and Drug Administration, Center for Drug Evaluation and Research (CDER), Center for Biologics Evaluation and Research (CBER), Center for Veterinary Medicine (CVM) 2011) (see section 2.2.2). During its second workshop on Sept 29, 2022, WP6 started this task from a basic TPP, incorporating a general definition of what EPND is supposed to be as a final product. Professionals from different fields were then invited to feed a first revision of such model, based on their own background and on the input from interviews and questionnaires deployed as a marketing search (See EPND Deliverable 7.1). Subsequent revisions of the EPND TPP are being processed with different participants, and include the advice by the Research Community Expert Group, a part of the EPND advisory framework. On January 31, 2023, experts from Academia, Industry, non-profit biomarker platforms and GV discussed the first version of the TPP and provided feedback for the next revision. Such integration of business, marketing, and R&D methods from pharmaceutical companies (originally developed in collaboration with regulators) (U.S. Department of Health and Human Services, Food and Drug Administration, Center for Drug Evaluation and Research (CDER), Center for Biologics Evaluation and Research (CBER), Center for Veterinary Medicine (CVM) 2011) is allowing to define the target features that EPND should have, to guarantee its fitness for purpose and long-term self-sustainability.

Leveraging this effort, such integrated action is being systematically examined, to propose adaptations and tools maximizing the benefit to the EPND aims, as well as facilitating the uptake and use of TPP in other contexts, especially academic research (Abstract submitted to AAIC, 2023) (Ibnidris et al. July, 2023) (see section 2.2.2). Indeed, this tool is mostly used in industry, but it is increasingly recommended also in academic contexts, not least by WHO, (Chowdhary et al. 6/9/2022; Cataldi et al. 2022). However, tools enabling its practical uptake in academia are not sufficient to date; developing them would be precious particularly in the NDD and in the biomarker field (Cocco et al. 2020).

2.2 Targeted action to support integration

As already underlined, there is no need to systematically try push all WPs or their tasks towards higher levels of integration: it is perfectly functional to EPND that some WPs or tasks work in a traditional intra- or multi-disciplinary fashion. However, characterizing, mapping and operationalizing different possible degrees of integration allows to better detect misalignments, gaps and needs, and promote higher integration whenever useful to improve efficiency, within a transdisciplinary framework. Outlining unsolicited efforts in support of transdisciplinary integration provides the natural leverage to generate tailored tools and propose them in a natural and “ecological” way within the EPND environment, thus holding the greatest promise to usefully support ongoing efforts. In the following paragraphs, we detail the focused action taken in this sense.

2.2.1 Glossary

As said, a shared language is both a basic requirement allowing transdisciplinary collaboration and a parameter to assess the degree of integration within transdisciplinary teams (Table 2, column C). Independent efforts to simplify and harmonize language have already been undertaken in different phases of EPND. We are now leveraging such efforts, to build a comprehensive Glossary that covers all terms necessary to work at EPND effectively. This entails enabling all participants to *understand* the used terms, even when they are not a specialist in a given domain, and getting *alignment* on the semantic meaning in the whole EPND context. For example, terms like “Consent” can be intended at the light of informed consent to participate in a research study, or to the consent to handle sensitive information based on the GDPR regulation. Participants may understand the word according to these different definitions based on their own background and experience of the term. Moreover, the concept of “Legal readiness” may not be clear at all to those outside WP2. Indeed, the term refers to the *degree* to which data can be shared, rather than to a dichotomic feature opposed to something as “non-readiness”. Importantly, features like “regulatory readiness” are often used to characterize the target EPND data, but achieving this common endpoint requires the same definition and understanding across participants.

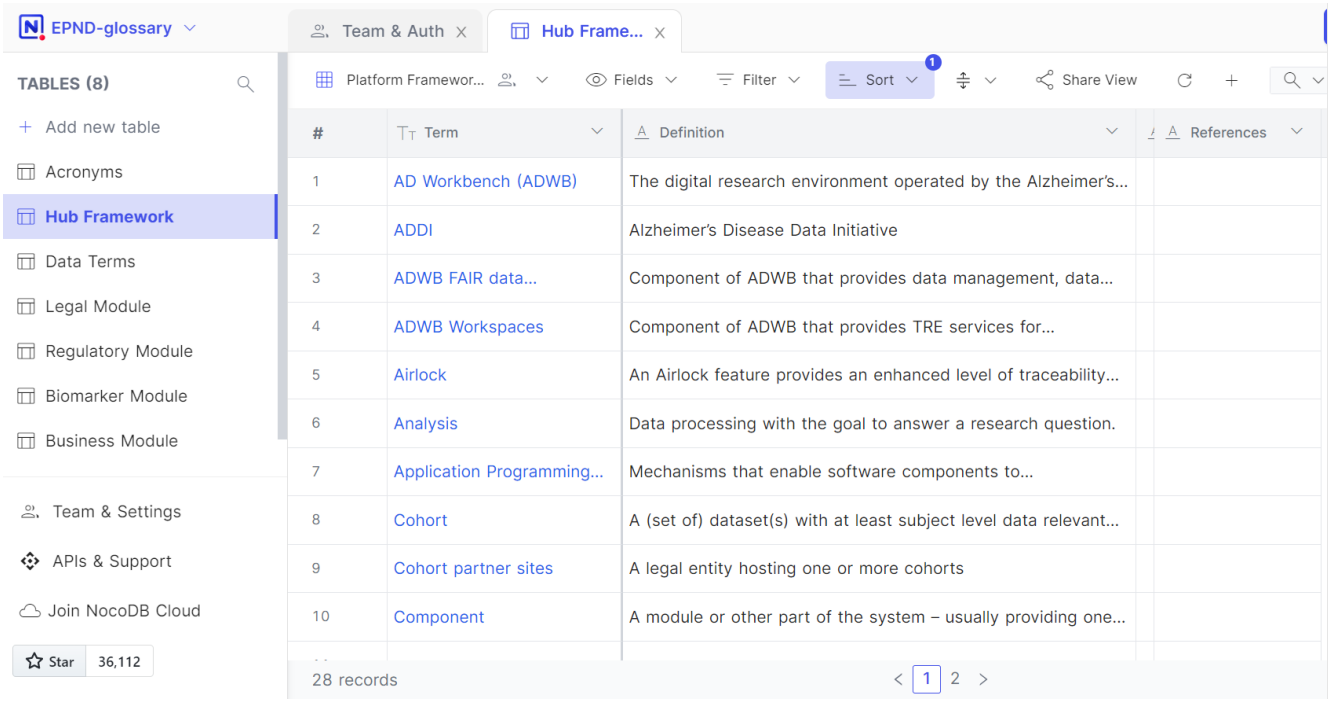
As of M18, a first full version of the EPND Glossary has been developed, with the main aim of increasing understanding and alignment on semantic meaning of specialist terms *within* the project. Further development will consist in harmonizing communication within and outside EPND, using this source to extract a "controlled vocabulary" (Hesketh et al. 2018). Besides reducing ambiguities and misunderstanding within EPND, the definitions in the Glossary will facilitate understanding of readers external to EPND; this may include reviewers of EPND scientific outputs. Finally, improved accessibility is expected to increase the impact of EPND assets (Hesketh et al. 2018). The target audience of the Glossary consists therefore primarily of EPND members but should ideally include any potential users of EPND. The EPND Glossary is meant as a living tool, to be edited, ideally, by any EPND member with the competence to do so, or wishing to request the definition of additional terms. As in the Aboelela's conceptualization, such definitions may consist merely of the explanation of technical terms specific to a single discipline (e.g., legal or regulatory science), but may also consist of terms that may have a new and specific meaning to EPND (higher level of TDC based on the Aboelela's framework; Table 2, Column C).

So far, the methods used to build the EPND Glossary are analogous, although more restrictive, than those used for instance for Wikipedia, which can be "edited by anyone at any time with an inclusion threshold of verifiability". At the moment, only selected EPND members can edit the Glossary, and we are discussing to what extent we should enable editing. In order to implement such Wiki-like editing, the data harmonization team from Maastricht University, responsible for task 1.2 of WP1, set a platform where multiple modules of the Glossary can be uploaded, and editing and comments can be tracked. The system allows multiple sources of information. So far, forms include item number, the target terms, their definitions, and additional columns reporting, if available, the reference for the provided definition, comments, and description of tasks to be completed (Figure 3). A last column enables to upload documents, which can be accessed online and downloaded. This tool can be accessed by EPND users using private credentials (links for external viewers are provided below). So far, more than ten EPND members contributed to its definition, from the platform internal release on February 2023

The Glossary has been created by identifying the main Modules of interest (i.e., legal, regulatory, business, biomarkers, etc.) based on the different kinds of tasks to be performed across the EPND WPs and the involved stakeholders. The coordinator then filled a starting page with basic terms encountered in the DOA or other internal material or meetings, being specific to that field and choosing terms that are not straightforwardly clear to non-specialists. This starting list was meant to simplify the work to the expert referent and was not binding as to the final term selection. The coordinator then contacted referent experts for each field from the specific EPND WPs/tasks, and asked to enrich, integrate and edit the starting version. The request was to cover only the terms that EPND partners need to deal with while completing their tasks, without trying to be exhaustive on any matter. Experts were asked to include references wherever possible, and were free to attach supplementary material (e.g., papers, tables). In this way, the planned

tool would be as small, easy to consult, transparent and useful as possible. WP6 meetings then allowed to discuss how to extend access for editing or using the Glossary. A tutorial has been developed to facilitate the editors' task (available on the EPND SharePoint).

Figure 3. A screenshot from the EPND Glossary



Overall, the Glossary is conceived as a living document, to be dynamically edited as EPND evolves and possibly to be opened for future EPND users. Moreover, a GitHub repository describing how the platform that supports the Glossary was deployed is being created, to facilitate other projects to use the same method to build their own glossaries.

At M18, the starting version of the Glossary includes 8 Modules: 1) Acronyms; 2) General terms; 3) Data terms; 4) Legal module; 5) Regulatory module; 6) Biomarker module; 7) Business module; and 8) Lay participant module (to be developed later by AE and FEP in the framework on the ongoing Public Involvement work). Most modules already included one or more rounds of editing among EPND partners. Below we report summary information on each module, and a link enabling to access them as viewers. Please note, the Glossary is a living document, and the number of items are expected to change over time.

1) The Acronym module is an inclusive list of Acronyms specific to or frequently used in EPND. Its original version has been curated by WP6 and 8; currently, it contains 351 entries.
https://epnd-glossary.azurewebsites.net/dashboard/#/nc/view/97085ad1-330a-448b-9c00-f80d44d1a48f
2) The Hub Framework consists of general terms regarding the EPND data initiative and its main components. It is aimed at harmonizing communications within and outside EPND. It was launched and primarily curated by WP1 and currently contains 28 entries
https://epnd-glossary.azurewebsites.net/dashboard/#/nc/view/a8ada7c8-1f20-429b-857a-5f21730e6fe0
3) The Data Terms module details the different levels of data expected in EPND and aims at making the use of terms as efficient and harmonized as possible, e.g. by avoiding the use of "metadata" as a broad and non-specific term by providing more precise definitions instead. It was launched and curated by WP1 and contains 9 entries.
https://epnd-glossary.azurewebsites.net/dashboard/#/nc/view/e4b0c4fe-1f8a-496d-9e12-e442d8813dd6
4) The Legal module has been curated by WP2 and contains 23 entries.
https://epnd-glossary.azurewebsites.net/dashboard/#/nc/view/6533ba8f-7fe8-4d9b-bb23-8c0fac41d56f
5) The Regulatory module has been curated by WP6 and contains 17 entries.
https://epnd-glossary.azurewebsites.net/dashboard/#/nc/view/3a86f9d9-852e-47d6-8e14-26bbc64c7fde
6) The Biomarker module is being curated by WP6 and 5, and contains 39 entries.
https://epnd-glossary.azurewebsites.net/dashboard/#/nc/view/5222977c-873e-4888-aaef-1415fe0e3f3c
7) The Business module has been curated by WP7 and contains 8 entries.
https://epnd-glossary.azurewebsites.net/dashboard/#/nc/view/66ef0330-994c-4950-ad71-b7485362ea12
8) The Lay Participant module is being curated by WP6.
https://epnd-glossary.azurewebsites.net/dashboard/#/nc/view/1d26413f-f55a-4520-a582-b041e81cbc29

The method we used to create the EPND Glossary may be less systematic than other Glossaries developed in implementation science, which involved for example Delphi rounds, which requires multiple participants converge on the final meaning of each term (Hesketh et al. 2018; Dopp et al. 2019; Kelly et al. 2022). The possibility to share and edit across EPND participants is a way to enable such a shared refinement of definitions, and has the advantage of feasibility, within a project where such Glossary is only meant to be a tool. In any case, we believe that this procedure is, at the moment, sufficient to serve EPND needs.

We are finalizing an abstract to be submitted to the Innovation section of the 2024 congress of Alzheimer's Disease International. The availability of the GitHub repository enabling to easily

replicate the structure of the platform to build other such glossaries will contribute not only to efficient EPND functioning, but also to its dissemination and promotion to future users.

2.2.2 Methods adaptation and integration: the Target Product Profile (TPP)

The level of integration between partners and stakeholders from different fields can be also assessed by the extent of interdisciplinary cross-fertilization that occurs, for example in cases where common methods and tools from a given field are modified and adopted by another (Table 2, Column D). The decision to design a modified version of the TPP framework, a commonly used tool used in industry to communicate the desired characteristics of a drug in development, to collate and identify differentiated characteristics of EPND as a "product" is a good example.

The first version of EPND's TPP has been drafted by WP7 using information collated from an initial survey of users and contributors of data and samples within Academia and Industry (October 2022, please refer to D7.1 for more details). The team of Marina Boccardi at DZNE has extended the remit of this initiative for wider benefit further highlighting the impact of cross-fertilization within PPPs. Taking advantage of the learnings from EPND, DZNE has undertaken an initiative to expand the possibility to adapt TPPs beyond EPND by performing a systematic review to extract common features to develop a method that can be used for different types of research including biomarker and NDD, but not exclusively. This is particularly relevant and timely as WHO has identified that TPPs are useful but not very common in academia (Cataldi et al. 2022; Chowdhary et al. 6/9/2022). This research will in turn benefit EPND not only at project stage but also beyond; it will be used as a guidance tool to support scalability, as currently available support is limited, and does not always facilitate practical uptake in academic contexts. For example, a recent review specific to the field of NDD biomarkers showed inconsistencies in their use and large gaps, especially related to the demonstration of clinical utility (Cocco et al. 2020). To extract the general features of TPPs, as they are currently used so far, we are performing: a) interviews with experts and opinion leaders on TPP (TPPs are usually private documents for industry), coming from within or outside EPND, b) a systematic review of published material, c) data extraction, and d) classifications of data based on reference methodological frameworks; this will include instructions and checklists to disseminate the method and simplify adoption. Providing a formal connection between TPPs and the current formal methodology for biomarker validation in the NDD field (Boccardi et al. 2021) will nicely complement the remit of EPND's aims of supporting high quality large scale biomarker research.

As of M18, we have searched multiple sources (PubMed, Medline, CINHAL and Scopus) and identified 1297 papers, of which 124 eligible for inclusion in a systematic review. Papers include different kinds of products (therapeutics, diagnostics, devices, other) and diseases (infectious diseases, cancer, with only one TPP in the NDD field). Noticeably, only one study involved NDDs (Alzheimer's disease). We are extracting the general features of these TPPs, and we are performing stratified analyses targeting in particular studies regarding biomarker development, that is of primary interest to EPND. As a preliminary exercise, data on the use of TPP in

biomarker development have been extracted from a sub-sample of 20 of the eligible papers. We have then mapped their features onto the phases of biomarker development as defined by current frameworks (Boccardi et al. 2021; Lijmer et al. 2009). Our preliminary results show that such features do not cover biomarker development consistently. This is consistent with findings from a previous review (Cocco et al. 2020). In addition to the findings of Cocco et al., (2020) we have also found that clear guidance on defining TPP features is lacking. This highlights the value of a systematic approach to produce a set of rules and principles to guide researches wishing to adopt a TPP as part of their strategy to improve research outcomes. The benefit of our research will therefore be manifold as it will: contribute to the efforts to streamline the biomarker validation process to reduce attrition rates (EMA high unmet need (Bakker et al. 2022)); support the development and expansion of EPND TPP during its lifetime; and add value to other research projects and IMI initiatives by developing a methodology that can be implemented in a variety of contexts.

The outcomes of this research, leveraging the transdisciplinary potential of a PPP will be presented at the Alzheimer's Association International Conference (AAIC) 2023 (poster #70964). This contributes to raising awareness about EPND in the NDD community and has the potential to support the development of EPND TPP throughout its lifetime.

2.3 Citizen Science (CS) feasibility

For CS, the task coordinator drafted starting issues and a plan for interviews across EPND participants, and then discussed it within WP6 and WP7. Discussions outlined the legal aspects as the most upstream issues. We then performed consultations within and beyond EPND, focusing on the possibility to access health data for CS within a FAIR context.

Consultations within EPND. In an internal assessment, our legal expert highlighted a degree of ambiguity around the definition of "anonymous/anonymized data" in the GDPR. The assessment therefore concluded that organizations managing neurodegenerative disease cohorts, who act as controllers under the GDPR and are therefore responsible for demonstrating their compliance, will likely diverge in their interpretation of what constitutes personal and/or anonymous data. However, our legal expert noted that in practice, most European biomedical research organizations tend to interpret anonymization in a strict way. The strictest level of anonymization means that no party, even the cohort owner, must be able to trace back the identity of the research participant using reasonable effort. This may not be feasible in biomedical research contexts, where cohort owners are also responsible for providing ongoing medical care to research participants; guaranteeing the possibility to add prospective data collected at subsequent follow-up visits is also relevant for both clinical and research purposes. Consultations within WP6 and WP7 highlighted possible inconsistencies between this assessment and activities performed in biobanks or other projects, leading to consider a more shaded scenario at the light of risk attenuation and trying to respond to the spirit of the law rather

than to the strictest literal interpretation. A solution to this issue is still not achieved and further work on this topic is ongoing.

Consultations outside EPND. We have also started to investigate how the issue is handled in CS contexts, specifically to health-related studies. Assessing the projects published on the eu.citizen-science platform was not informative (from only two projects using health data, one did not answer our request, the other one confirmed that their data are completely anonymized according to the strictest criteria). We have then contacted Claudia Fabo-Cartas, project officer of the European Citizen Science Association (ECSA), who addressed us to its working group "Citizen Science for Health" (CS4H, https://ecsa.citizen-science.net/working-groups/citizen-science-for-health/). The CS4H group maintains that citizen science in health has a big potential to deliver innovation to society. They underline that engaging patients in health research has occurred for many years, although their impact on research questions, methodology, ethics, analysis and data management has usually been limited. Health is therefore still widely under-represented in citizen science. Ongoing projects are very heterogeneous in type (models of collaboration and benefits of CS in health are described in a reference paper from 2019 (Wiggins and Wilbanks 2019)). The ECSA CS4H working group has therefore the objective to promote and increase the impact of citizen participation in health research. Its main objectives consist of: creating a community of stakeholders dedicated to promoting and developing Citizen Science for health by fostering dialogue and collaboration; developing and disseminating tools, methods, ethical frameworks and training material enabling and supporting CS4H; enhancing the visibility and potential of CS projects in the health domain.

Interviews were then conducted with Gaston Remmers and Lieke Heesink, in their capacities of chair and member of the CS4H group. They confirmed that our question is among the topics treated in the working group, as CS studies on health entail not just donating data and contributing to their processing, but also enabling citizens to access the data collected within the whole study, and therefore accessing also the sensitive data of other participants. Moreover, also the CS4H studies consider making data reusable by other citizens, with exactly the same issues encountered within EPND (See Table 4 for a comparison of CS4H and potential EPND features relative to CS).

The CS4H colleagues underlined how such ethical issues have been receiving attention only very recently. As the first contribution, Wiggins and Wilbanks highlighted the "ethic gaps" left in the evolving CS only in 2019. Legal protection for citizens taking part in health CS research has not kept pace with the transition of the science, and its connected risks, out of the clinic and into the hands of individuals (Wiggins and Wilbanks 2019; Patrick-Lake and Goldsack 2019). Patrick-Lake and Goldsack conclude that solving such ethical gap is urgent, and solutions should be devised with citizens themselves, in the same democratic context as CS studies. To do this, the authors underline that swift action is needed, although it should leverage existing laws (Patrick-Lake and Goldsack 2019)). While Patrick-Lake and Goldsack summarize US regulations, others also take into explicit account EU and GDPR rules (Evans 2020; Ficorilli et al. 2021). Some underline that these very regulations should be extended to include the empowerment of citizens

as citizen scientists (Ficorilli et al. 2021), but not all agree that ethical and privacy regulations should be context-dependent (Evans 2020). Currently, publications focusing on the topic are less than 10, and there is yet no convergence on consensus or institutional guidelines or rules. Interestingly for our next investigation steps, resources and consultancy opportunities are available from investigators with specific expertise (see, for example the websites: https://jcom.sissa.it/archive/20/06/JCOM_2006_2021_A04; https://ois.lbg.ac.at/ois-support-services/research-ethics/).

Table 4: Project features and differences between EPND and current CS-Health projects.

Feature	CS-Health	EPND	Notes
Data ownership/access	Data owners=data viewers	Data owners differ from data viewers	Ideally, patients should provide informed consent. Principles may be consistent in general
FAIR	CS also considering the utility and complexity of making CS data FAIR	FAIR as a requirement for open science; however, legal uncertainties under the GDPR may discourage FAIRification practices*	The tensions between FAIR requirements and controller's GDPR obligations may need to be solved at a higher level and may cover both contexts. Enabling CS in EPND may contribute to trust and to co-defining solutions.
Citizens to be recognized as researchers: competence and training	Citizens recognized as researchers, after receiving proper training	A CS section in EPND should entail tutorials, training, monitoring and QC	EPND may also need to decide how to acknowledge publicly citizens' contribution
Citizens to be recognized as researchers: conduct and ethics	Citizens recognized as researchers, after signing code of conduct	Can be provided	

*In spirit, the GDPR supports data availability and reusability, particularly for scientific research purposes. However, it may be challenging for controllers to demonstrate compliance with certain GDPR requirements and safeguards. This may make such processing risky from a controller's point of view.

Based on this background, operative solutions have been developed for 5 projects performed within the CitieS-Health initiative on urban pollution and health (citieshealth.eu) (Ficorilli et al. 2021). Their key features, and the possibility for EPND to comply with them in a similar manner are listed in Table 5.

Table 5: Requirements and potential EPND compliance, based on consultations with the CS4H working group and literature to date.

Requirement	EPND (possibility of) compliance	Reference
Data available only upon reasonable request	YES. Access would entail requests by EPND researchers and not by any citizen	Wolkorte et al, 2022 (Wolkorte et al.)
Easy to find info re data management upfront	Yes (to be provided)	Wolkorte et al, 2022
Permission and informed consent to make data accessible	Not for data available so far (for making data accessible through the EPND specifically). May be requested for subsequent collection, or retrospectively for subsamples. Relevant not only for CS but for the whole platform/FAIR initiative	Wolkorte et al, 2022
Development of a data management guide for citizen science for health	Yes (to be provided)	Wolkorte et al, 2022
Study protocol to include training on ethical standards, regulations and rights related to use, re-use, storage, sharing of data, research integrity	Yes in theory, it may require additional dedicated budget	Ficorilli et al, 2021
Enable a participatory approach where citizens can propose and receive answer on issues including use/re-use of data	Yes in theory, it may require additional dedicated budget	Ficorilli et al, 2021

Citizens to disclose financial or non-financial conflict of interest	Yes (to be provided)	Ficorilli et al, 2021
Whatever the compliance, criteria for authorship to be stated upfront	Yes (to be provided)	CS4H consultation
Modalities for data handling to be defined (possibly avoid download on private laptop and mandate processing on protected cloud)	Yes, possible in theory. For example, the ADDI workbench enables online processing with only final results from federated analyses transferred to users	CS4H consultation

Specific points for more detailed consideration are:

- Informed consent to re-use the data for additional research should be obtained whenever possible. Such consent is not yet available within EPND; however, cohort owners making their data available to EPND may collect it from next patients or during the next follow-up assessment of already recruited patients. This consent, primarily required also for the general aim of making data FAIR, may be devised to expressly include the possibility to enable CS.

- An analysis of the ethical issues raised by the processing of personal data involved in the study and an explanation of how these issues will be mitigated in practice must be provided. Data protection impact assessment (DPIA) in line with Article 35 GDPR [European Parliament and the Council, 2016] and supplementary guidance on DPIAs may also be provided [European Commission, 2018]. Complete anonymization being difficult to achieve, procedures should be defined to limit as much as possible the risk that those accessing the data may track the study participants. These procedures should be described in detail and provided upfront.

- *Per se*, the issue of making data FAIR is conceptually the same for EPND and for CS. What protects *researchers* from accessing such “FAIRified” data? Or, in other words, once we solve the issue for researchers, what is the difference between researchers and citizens, based on which citizens should not have the same access rights? Training-qualification, compliance with ethical standards, approval by committee to use the data for relevant/adequate research aim, and any other such defining feature should be explicitly defined. If complied with, the accessibility to data by citizens has no reason to be questioned.

In summary, for ECSA4H, the key-point consists of: a) defining all the above features, and b) upgrading citizens to the role of researchers, although “informal”. This is relevant under many respects, well beyond the mere enabling of data access.

Considerations for EPND to understand whether to invest and enable CS should therefore entail an estimate of the investment required to enable such upgrading, and the return for the project. Indeed, the investment may require additional dedicated funding, possibly within a CS context. Training-qualification should entail formal training, and the creation of specific tutorial, availability to answer questions, and results QC at different levels. On the other hand, it would be relevant to increase the research capacity of society. Enabling to understand anonymization and other procedures minimizing risk and protecting privacy, ethics and requirements in data handling can increase trust and potentially foster further participation. Being acknowledged as a qualified informal researcher may be experienced as a prestigious reward by citizens, who can in addition learn more about research in general and specific scientific issues in neuroscience. Feeling included in scientific processes may bring a step forward in the democratization of research.

2.4 Definition of indicators

Indicators are an essential component of monitoring and evaluation (M&E) procedures, enabling to collect information conveying strategic insight for effective planning and management of an action or program (UNAIDS) (https://www.unaids.org/sites/default/files/sub_landing/files/8_2-Intro-to-IndicatorsFMEF.pdf). Indicators summarize the level of a target feature of interest, able to inform whether the action is achieving the meant result or whether, and in which direction, this may need to be adjusted. Their collection also allows to compare directly the effectiveness of different interventions. For this reason, it is important that indicators be meaningful, easy to collect, and consistent across projects. Therefore, reuse of already defined indicators should be preferred whenever possible (UNAIDS). On the other hand, literature on transdisciplinary collaboration (TDC) acknowledges that TDC is so complex and variable that this may not always be possible, or even desirable (European Commission and Directorate-General for Research and Innovation 2015). With variable consortia, tasks and aims, one measure may not fit all, and indicators for the relevant dimensions of TDC functioning, output and impact are still to be defined. Very few are the publications on this regard, the latest and most authoritative in the field dating 2022 (van Drooge and Spaapen 2022). In any case, evaluation in this field has been evolving towards a constructivist approach, valuing qualitative information and considering indicators as tools supporting interactions rather than assessing, measuring or enabling accountability (Lay and Papadopoulos 2007).

Consistently, also for the EPND WP6, the definition of indicators turned out to be a challenging task. Some specific criteria to choose indicators for public involvement had been defined in previous projects, but regarded mostly the involvement of patients in drug development (Vat et al. 2021). These entail for example the feeling of being taken into account and having an impact on project development. Indeed, these are not straightforward relevant to EPND, and may not easily adapted to professionals and stakeholders other than patients. Moreover, analogous modalities to collect indicators of this kind may not be perceived as ecological or useful within the project, indeed another acknowledged challenge of defining indicators in TDC (van Drooge and Spaapen 2022).

This very recent contribution examined the kinds of indicators available in contexts close to TDC or Responsible Research (European Commission and Directorate-General for Research and Innovation 2015), and distinguished two types of indicators, summative (meant to account for the activity) and formative (aimed at mutual learning and improving the process itself), and concluded that only the second is of interest to TDC. The authors outline how most indicators so far are based on the "new public management" model (Scriven 1996, 1991), and new indicators, as well as concepts of governance, are needed for challenge-oriented research. Among the very few pioneer attempts in the field, Drooge and Spaapen select the participatory impact pathways analysis (PIPA), a method that combines evaluation with planning, based on the Theory of Change (Silva et al. 2014).

A theory of change is a causal narrative outlining how –i.e., through which impact pathway- a result can be achieved. This narrative is defined by the different stakeholders involved, rather than based on linear logical reasoning defined within a research procedure disconnected from stakeholders (Bush 1945). Within this context, evaluation and monitoring are focused on evidence about whether or not the impacts have been or are likely to be achieved, and on highlighting potential hurdles from the specific narrative that is built in the study. The PIPA entails the definition of the pathway leading to impact, i.e., the articulation of elements within a logical framework, and the causal assumptions, i.e. why and how, this impact would be achieved, which are the elements constituting the theory of change. Consequently, the "indicators" highlighted by van Drooge and Spaapen are essentially qualitative and specific to each project (for example, indicators emerged from their analysis included the difficulty to have all needed stakeholders to their workshop, the observed attrition between the logics of different stakeholders and the imbalance of their views (van Drooge and Spaapen 2022)).

While the elements of a Theory of Change or PIPA are planned to be defined in one workshop, it would be impossible to define such elements and narrative for EPND in one session (such narrative/Theory of Change entails how we can make EPND the platform of choice for FAIR data in Europe and beyond). Such "narrative" is however built along the many regular meetings, and is expressed and operationalized through the TPP (see Section 2.2.2). All steps between current the current status and the final target are the object of recursive discussion in the regular meetings of the different WPs involved in tackling each of the involved mechanism. To make an example: to make data "interoperable" (the "I" in FAIR), these must be harmonized. The whole WP3 works at such harmonization. To make data Findable, both WP1 and WP4 contribute at building a digital system and engaging cohort owners, along with the support of others like WP6 facilitating connection and communication. The processing described in the theory of change procedure, to detail the causal mechanisms leading to the desired impact, is extensively performed in such meetings (e.g., identifying the incentives able to motivate cohort owners to lend their data; defining the features of the EPND data or procedures that would convince users to use EPND rather than other channels to perform their experiments, etc.). In addition to discussion within the pertinent WPs, such discussions benefit of more or less structured cross-WP interactions, as well as expert advice through the Advisory Framework set within WP6. The process involves therefore a large variety of stakeholders within EPND. Through extensive

interview processes, also the view of many more potential end users has been collected, and used to define the TPP and by WP7.

Based on this recent literature on indicators in TDC, we can state that EPND has started and is continuously evolving a PIPA, or theory of change, that allows the project multi-dimensional progress along a network of causal paths that lead from purpose to outcome based on the causal mechanisms known and detailed by a variety of stakeholders, through such continuous integrated work that is summarized in the TPP. *The target of formative indicators in EPND, therefore, consist of assessing how smooth and balanced is the communication and contribution by the required stakeholders in defining the EPND path from plan to endpoint, and the TPP*. To make such evaluation as simple, objective, systematic and useful as possible, we leveraged also the literature on TDC (see Table 2) outlined in the general frameworks section. From these, we have extracted the most relevant indicators serving stakeholder contribution.

One such indicator consists of characterizing the kind of multi-stakeholder collaboration itself, as from Aboelela's framework (Table 2). As said, the kind of collaboration (multi-, inter- and trans-disciplinary) is characterized by specific parameters (kind of language, kind of methods, composition of authors in publications, competencies of participants, etc.) that can be observed. So far, we focused our attention mostly on Language, Stakeholder involvement, Competencies and Publications.

- *Language*. At the starting point, language could be considered at the level of multi-disciplinary collaboration: despite the spontaneous starting initiatives to clarify acronyms and agree on a common dictionary for defining terms on data and EPND, we had to launch a dedicated effort to make specialist terms transparent to all EPND members (see Glossary section and Table 6). We have no way at present to assess whether this is actually increasing the level of integration within EPND, however this is a necessary tool to facilitate communication and support integration. Spontaneous initiatives occurred, defining terms in a way that is meaningful only within EPND (e.g., see the Hub Framework and the Data terms modules in the Appendix). On the other hand, terms that are uniquely clear to specialists, and that are now explained in the glossary, are not necessarily understood or used consistently throughout the project without additional effort. At present, we are soliciting active involvement in editing the Glossary. We have also defined a questionnaire to be administered in the next months, to collect suggestions for improvement and assess actual use. We will also monitor its evolution along time, its impact on the efficiency of the collaboration along the project timeline, and will explore strategies to use it with focused intervention to increase its use and impact.
- *Involvement*. We have assessed the input of interviews collected by WP7 to inform the EPND TPP at the light of the variability of the stakeholders required to make EPND the platform of choice for FAIR biomarker data. The collected information mostly came from potential users. Despite involving SMEs and academia, most responders were however researchers, either processing the data and/or owning them and in the position to make them available to EPND. We explored whether collecting information to feed the TPP from

more heterogeneous stakeholders (e.g., regulators) could be considered useful, and indeed this is planned for future WP7 activities. The number of stakeholder categories involved in the WP7 interviews aimed and in defining the TPP is an easy to collect, objective, and is an informative indicator able to support better coverage and stakeholder involvement along the project.

- *Methods*. Based on Aboelela's framework (Aboelela 2007), methods are increasingly imported and adapted from other fields with increasing integration among different professionals and stakeholders. From the EPND start, efforts to import methods have been done, although in an empirical way. These include for example using the TPP to define a priori the final EPND features and stick to the plan through the project life, and imported from drug development to the development of the platform. We are now structuring the adaptation of the TPP for wider, more systematic and useful adoption, within and outside EPND.
- *Competencies*. As from Gebbie's framework (Gebbie et al. 2008), competencies are objective abilities that can be modified with dedicated action or training. These involve for example attending meetings or conferences from different disciplines or reading journals from other specialties. So far, we can report that spontaneous offers for such opportunities occur regularly within WP6 and WP7, i.e., the most trans-disciplinary WPs. At the moment, specific actions to increase and promote more of such spontaneous proposals do not seem necessary. While it would not be difficult to ask participants to provide more resources, an overload of information may not necessarily help the project proceedings. However, we will take care that a good balance be maintained, within the context of ongoing work. To this avail, the recently set newsletter is a privileged communication tool.
- *Publications*. Based on Table 2, the heterogeneity of professionals and stakeholders among the authors produced within the project indicates higher levels of transdisciplinary integration (Aboelela et al. 2007). At this early time of the project, only one full paper is published (Bose et al. 2022) and one is drafted (Ibnidris et al, abstract submitted; see section on TPP). The first represents 3 stakeholder categories (philantropy, academia, industry) within a set of four authors, already in line with the IMI-2 multistakeholder effort and, interestingly, including both an EU and US point of view. The second includes academy, industry, for-profit and non-profit consultancy companies. The composition of authorships, as well as the integration of methods from different disciplines (Aboelela et al. 2007) can be assessed objectively in the next publications produced during the project. Although expected to reflect the multi-stakeholder nature of the initiative in a natural and spontaneous way, explicit attention to these parameters will help balance participation and extend attention to any neglected stakeholder as appropriate based on each task.

In summary, we have identified meaningful indicators useful to EPND in line with the most advanced literature on TDC (Table 6). These belong to two categories: a) acknowledged requirements enabling transdisciplinary collaboration (Aboelela et al. 2007) and b) participatory impact pathways analysis (PIPA) (van Drooge and Spaapen 2022).

Indicators in a), include:

- Language
- Involvement
- Methods
- Competencies
- Publications.

Indicators in b) relate to the qualitative information that dynamically emerges during the EPND "PIPA", i.e. the Theory of Change formulated in the many meetings aimed to achieve the EPND target. This is not liable of reporting in this context, but is relevant for moving forward within EPND activities. Indicators in a), instead, are more general and straightforward to extract and compare.

Particularly the indicators in a) can reasonably be collected during EPND development and compared at different time points. Both a) and b) indicators are useful to support its proceedings in the contexts requiring closer interaction among heterogeneous stakeholders. We underline that, in any case, these indicators mostly belong to the category of "formative" rather than "summative" indicators, i.e. they are functional to serve mutual learning and help develop a strategy leading to an impactful endpoint for the project, rather than providing mere measurements of performance. The advantage of indicators in a) is that they also allow "summative" functions, by comparing change in different time points (Table 6). Since such comparison enables to understand where greater support should be provided, the ultimate aim of all these indicators consists of supporting EPND activities, rather than assessing them.

With this framework and starting assessment in mind, we are ready to serve different needs and aspects of transdisciplinary collaboration along the project (see Roadmap section).

Table 6. Using the dimensions defining TDC (modified from Table 2 and from (Aboelela et al. 2007)) as indicators in support of increasing multistakeholder integration in EPND. The use of indicators helps to foster (rather than assess) integration (van Drooge and Spaapen 2022); it is useful to identify gaps for focused action and monitor whether change occurs in the meant direction. Location in the table represents degree of integration achieved so far. Color reflects whether such achievement was spontaneous, whether focused systematic action was launched, whether provided tools are occasionally used by some, or systematically used by many

A	B	C	D	E	F
	Modus operandi	Language	Methods	Publications	Competencies

Multi-disciplinary	Parallel	Specific	Specific	Team-specific	Discipline specific
Inter-disciplinary	Coordinated	Translated	Mixed	Complementary sections merged	Occasional access to sources from other disciplines by some participants
Trans-disciplinary	Integrated	Merged/ generated (project-specific)	Integrated/ adapted/ generated	Fully shared data generation and presentation	Frequent access by many participants

3 Roadmap: future development and deployment

In the previous sections, we have identified stakeholder involvement frameworks of relevance to EPND, presenting an analysis that highlights the value of approaches to enhance transdisciplinary collaboration. Development of the EPND Glossary and target product profile (TPP) is described, showing how these tools, themselves a product of transdisciplinary collaboration, can enable more effective engagement, involvement and collaboration between internal EPND stakeholders. In this section, we identify further EPND milestones that involve a broad range of stakeholders, to anticipate where the Glossary and other tools could be leveraged to enhance collaboration (Table 7).

Table 7. EPND milestones: timeline, verification and required stakeholder contribution.

EPND Milestone	Timeframe (project month/date)	Means of verification/ description	EPND partners involved	Stakeholder groups
M1.5 All separate components ready for the resource-level discovery service	M24/November 2023	EPND cohorts can be discovered via the ADWB	BMD, UMCG, ULEIC, UM, Aridhia, GV	SME (software & infrastructure development), Academic (bioinformatics & data science), Non-profit (venture philanthropy)

D3.6 Published report on SOP collections	M24/November 2023	Publication of report on generated SOPs, including body materials, biomarker validation, biobanking best practices	LIH-IBBL, UNILU, BBMRI, UGOT, UCB, SVAR	Biobanks, Biobanking platform, Academic biomarker experts, Industry
D7.2 Benchmarking report	M24/November 2023	Report with benchmarking of previous and ongoing initiatives of similar scope and context to EPND; identifying potential operational models	EATRIS, AE, BBMRI, GV, Sanofi, Novartis, Janssen	Biobanking platform, translational science platform, patient organisation, Industry
M4.3 SOP-presenting webinars completed (1 of 2)	M24/November 2023	Participation in webinar	VUmc, BBMRI, all partners	Academic biomarker experts, biobanking platform; all partners
M1.7 Implemented MVP of the EPND Platform node of ADWB as a biobank and cohort data platform	M30/May 2024	Cohort & sample data shown to be discoverable at record and resource level, with options for workspace-based analysis	UNILU, Aridhia, UM, BMD, UMCG, CHUV, GV, Roche, Takeda	SME (software development), Academic (bioinformatics & data science), Public sector (medical informatics platform), Non-profit (venture philanthropy), Industry
D6.7 Regulatory advice on complement biomarkers for use in trials	M36/November 2024	Report on briefing meeting with the innovation taskforce of EMA on the application of EPND to support biomarker use in trials, or qualification	CBG-MEB, LYG, EATRIS, AE, GV, UCB	Regulatory body, project management company, translational science platform, patient organisation, non-profit (venture philanthropy), Industry

D7.4 Final sustainability plan	M48/November 2025	Report detailing operations, funding plan, policies and templates for access requests, policies to manage IP, confidentiality, publications.	EATRIS, AE, DZNE, LYG, GV, UCB	translational science platform, patient organisation, academic research institution, project management company, non-profit (venture philanthropy), Industry
M1.8 All elements of the final EPND platform fully integrated	M48/November 2025	All EPND cohorts and biorepositories are connected and discoverable, and datasets available for analysis by central or federated methods	UNILU, Aridhia, UM, BMD, UMCG, CHUV, GV, Roche, Takeda	SME (software & infrastructure development), Academic (bioinformatics & data science), Public sector (medical informatics platform), Non-profit (venture philanthropy), Industry
D2.3 ELSI White Paper v2.0	M48/November 2025	Final Govenance and Data Protection Framework	BBMRI, UNILU, EATRIS, AE, Sanofi, UCB	Data protection experts, Biobanking platform, translational science platform, patient organisation, Industry
D4.6 Report on case studies started in Phase 2	M54/May 2026	Report on cohort identification, contractual and financial issues, support for logistics of data and sample transfer	UM, UNILU, UOXF, UCB, KCL, DZNE	Academic research experts on AD, PD & DLB, Academic biomarker experts
M4.5 Two rounds of BBMRI self-assessment surveys completed	M54/May 2026	Completion of biobanking self-assessment surveys by 30 cohorts, based on international standards for pre-analytical sample processing or	VUmc, BBMRI, all partners	Academic biomarker experts, biobanking platform; all partners

		applicable EPND standards/SOPs		
D5.7 Report on case studies started in Phase 2	M54/May 2026	Report on CS5, pilot studies to test the full suite of functionalities of the EPND platform.	UNIGE; all partners	Academic research experts on AD; all partners

3.1 Further development

The content described so far entails research on available theoretical frameworks and tools. We extracted elements able to support EPND and detailed actions already started to specifically support transdisciplinary collaboration in different tasks of the project.

More research is warranted to improve such tools. Next action will mostly include drafting questionnaires and collecting information from EPND participants, to understand how exactly we should adapt the above elements and better serve EPND.

Table 8 summarizes the content described in the previous sections and the next planned actions, specifically to the assessment of spontaneous support of TDC, collection of indicators, active and planned actions in support of next activities, and criteria for the final assessment of results.

Table 8. Activities specific to the detection of spontaneous support of TDC, collection of indicators, active or planned support of next activities, and criteria for the final assessment of results.

Problem or dimension	**Indicator: (A) Active intervention; (S) spontaneous action**	**Planned action**	**Assessment of impact**
Understanding how to achieve impact	(S) “distributed” definition of Theory of Change (ToC); (S) TPP	Involvement of more diverse stakeholders; Help making the currently implicit ToC explicit, and clarify its transfer into the TPP	Achieving the target defined in the ToC and summarized in the TPP; Use cases; Users access

Inconsistent understanding or use of terms	(S/A) Set up of Glossary	Promotion of consultation and editing; selection of controlled vocabulary; collect input to increase utility and impact through questionnaires	Metrics of access, edits, contributors
Supporting the improvement or development of transdisciplinary competences	(S) opportunities of education, information or transdisciplinary contribution are regularly offered	Repository collecting online materials spontaneously provided	-
Engagement of diverse stakeholders	(A) assessment of contributors to TPP and ongoing publications	Extend interviews to other stakeholders Encourage participation of different stakeholders in own publications	Completeness of TPP in addressing a whole ecosystem rather than selected users
Methods	(S) TPP adopted (A) TPP methodology investigated to facilitate adoption in different contexts	Systematic review to support transfer of TPP to different fields	Assess TPP coverage of the domains identified by the systematic review, as compared from its first formulation

The subsequent paragraphs detail specific actions planned for selected areas described in this deliverable. Due to the novelty and the complexity of this field, some of these planned actions may not be entirely worked out in detail, or they may not cover all of the mentioned (or needed) categories. However, we aim to further process all of this material, to provide increasingly effective action in support of TDC within EPND, as well as a model to be easily translated to other projects of this kind, and in NDD research in general.

3.1.1 Glossary

Starting from the current Glossary (see Appendix), we will invite WP-6 and WP-7 members (i.e., the most multi-disciplinary) to access, edit, comment available definitions, and flag all terms that may be required for a controlled dictionary, to be used for communications within and outside EPND. We will then ask them to fill a short questionnaire highlighting how the tool may be modified or proposed, to maximize its utility. Finally, we will work on simplifying the visualization of edits or comments proposed by users: these can be used as indicators of access, use and

co-definition, e.g., of transdisciplinary generation of a shared language (Table 2, Column C) (Aboelela et al. 2007).

3.1.2 Competencies (Gebbie et al. 2008)

Based on our assessment of needs and spontaneous *modus operandi*, we believe that no further specific action should be taken, also to avoid overload of information. However, partners will be made available of this content, and this will enable them to spontaneously see even more possibilities of proposing relevant input in specific circumstances. Moreover, they will be invited to upload material and information on a dedicated section of the EPND sharepoint, to make this aspect of TDC-support explicit and accessible to all, and enable more people (i.e., beyond WP6-7) willing to expand their transdisciplinary competencies to find such information, already selected for being relevant to the EPND tasks. Indicators of increasing transdisciplinary competencies within the project will then consist of the number and variety of documents/materials and, if possible, number of accesses and downloads along time.

3.1.3 Citizen Science

We will extend and update our research on CS in health research and FAIR data, leveraging more of the available resources, e.g.: (Aungst et al. 2017; Ficorilli et al. 2021; Lambertson et al. 2015), consultation of the “Citizen science theory and practice” journal; participation to the ECSA CS4H), to provide a more exhaustive definition of requirements and constraints to enable CS within EPND. Further literature search may expand how to acknowledge credit to citizens. On the other hand, internal questionnaires may allow to identify the most feasible tasks that may be assigned to citizens, and to quantify the amount of work and commitment required to EPND researchers.

While a CS section may not be feasible within the project timeline, requirements may be taken into account for further developments. For example, if the informed consent of new subjects collected for cohorts adhering to EPND have to include a new option on data re-usability beyond the specific project, we may recommend that such integration also include access for CS processing.

Additional information on feasibility entails which tasks can be envisioned as feasible by EPND researchers and cohort owners (typically, time-consuming tasks requiring only minimum amount of training and supervision) and estimates of the amount of effort requested to produce educational material for such tasks, check the quality of processed material, etc. This is planned to be investigated with dedicated questionnaires in the next months.

Finally, a comparison will be performed between the decisions/preferences expressed by citizens participating to CS4H studies, and those expressed by EPND patient expert groups

regarding data and sample sharing. This will inform even more specifically the kind of requirements for a specific EPND-CS context.

3.1.4 Indicators and other parameters

The collection of indicators, as well as any other parameter described in this deliverable will be further refined and flexibly followed up based on project proceedings. If possible and useful to the international community, we will also produce publications presenting our processing and results from the EPND experience.

3.2 Deployment

We will systematically use the know-how and tools built by the EPND transdisciplinary community to support internal cooperation, to achieve specific milestone as well as with the outreach of heterogeneous stakeholders outside EPND.

4. Conclusions

Intervening with targeted action to improve transdisciplinary collaboration in well consolidated consortia like EPND is challenging. The baseline level of integration is already considerably high. Supporting further integration with new methods may interfere with previous working habits and be difficult to implement.

While a dedicated theoretical effort in support of TDC activities is relatively new in IMI projects, it is not new in innovation science. Awareness of such research and available techniques is disproportionately greater in the technological and industry environment than in translational neuroscience academic contexts. However, the transdisciplinary context of IMI consortia is a privileged environment to import, adapt, and transfer the most advanced R&D and innovation frameworks, strategies and tools improving translational neuroscience.

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PART 2. Clear and respectful language use, communication and interaction towards people with dementia and Parkinson's disease

Section 1: Introduction

What is Part 2 of this deliverable about?

Neurodegenerative diseases primarily affect neurons and involve a degeneration of nerve cells. Examples include amyotrophic lateral sclerosis, multiple sclerosis, Parkinson's disease, Alzheimer's disease, Huntington's disease, multiple system atrophy and prion diseases. Part 2 of this deliverable provides guidance for researchers on respectful communication and interaction with people with neurodegenerative diseases, aimed at promoting meaningful and mutually beneficial Public Involvement (PI) work. It provides valuable insight from people with dementia, Parkinson's disease and parkinsonism, as well their carers/supporter (please see Section 2 on the methodology) and touches on the issue of promoting diversity, reciprocity and avoiding stigmatization. It also includes guidance on planning and conducting PI.

After this introduction and the methodology, we will explain what PI is and then provide some guidance on planning and conducting PI work in the context of research into neurodegenerative diseases. This will be followed by three sections on language, communication and interaction. The first is about communicating and interacting with people with neurodegenerative diseases in the PI process, the second is about communicating about and portraying them in the context of research but also to the wider community, and the third is about language used in the context of research about neurodegenerative diseases.

Whilst EPND is about the development of a platform to share research data about neurodegenerative diseases and this deliverable is about communication and how to conduct meaningful PI, the goals and expectations surrounding this report are very much linked to real people. In the next subsection, Marcelino, Chris and Petri share their experience of how having dementia or Parkinson's disease impacts on their daily lives.

Personal testimonies from people with neurodegenerative diseases

Marcelino Morcuende speaks about his experience of having Parkinson's disease

Having an older brother with Parkinson's disease, the diagnosis did not surprise me at all. The first symptoms that alerted me were that my arms weren't swinging like usual when I was walking and pain in my shoulder, my right arm and my wrist. My fine motor skills had "gone to hell" and I had clear signs of micrographia (small, cramped handwriting that is typical of Parkinson's disease). I had lost a lot of my sense of smell, which, as I discovered later, is a symptom, like others, that can occur years before diagnosis.

After the ordeal preceding the diagnosis (I went through traumatology, which is the branch of medicine that look at wounds and injuries, as well as internal medicine, and did different tests to rule out other possible conditions), we came to the conclusion that it was "possible Parkinson's disease", which was also reflected in successive reports from the neurologists.

Chris Roberts speaks about his experience of having mixed dementia

My name is Chris Roberts and I live in North Wales (United Kingdom). I was diagnosed when I was 50, firstly with vascular dementia and a bit later with Alzheimer's too, which is more commonly named mixed dementia. Before and during my dementia diagnosis, my memory was getting worse and word finding was sometimes quite difficult. Sorting money and loose change was also becoming problematic. I compensated by paying with notes instead. My GP was very good and supportive. She referred me to our local memory clinic for testing. It took 13 months. During this time, we noticed my driving wasn't so good. I was looking down to see where my feet were supposed to be, forgetting where the controls were and kept getting lost in familiar places.

After a diagnosis of dementia your whole family also receive the diagnosis and it becomes a team effort. It has to. With any life changing illness, you have to change your life. Only then can you embrace, accept and live with it better. But, mainly due to the misconceptions, myths and all of what you see in the media, you are left thinking there is nothing that can be done. It becomes and is a very lonely place to be in.

As well as dementia, it also comes hand in hand with depression, guilt and further future disabilities. I have problems with my balance and mobility, I can't always see steps and inclines, I don't see same colours on same colours, I don't always recognise friends and family, and I can even get lost in my own house. I have difficulties with any multi-tasking too. I find writing problematic as I can't always see the letters in my head. I need support to cross roads as sometimes I fail to distinguish which way the traffic is going, struggle with reasoning and fact, getting mixed up with dreams and reality.

We realised how little not just ourselves, but also a lot of the general public, understand what it means to live with my diagnosis, and it becomes easier not to talk about your diagnosis because of the stigma and misconceptions added to the general lack of understanding. But getting involved with the Working Group and contributing towards research projects have given me lots of hope and I have gained a lot more confidence to live better with my dementia. People are very good when they know about my dementia and are very supportive in general but more education and awareness is still needed.

Petri Lampinen speaks about his experience of having frontotemporal dementia

The most stressful period for me lasted for two years until I received a memory disorder diagnosis.

First, let's go back a little in time to when I experienced the first signs of the disease. When I was still working, I couldn't stand still and I was always moving about. I couldn't wait to take a lunch break that lasted more than 15 minutes. I didn't finish my work, moving hastily from one task to the other. I forgot about scheduled meetings and deliveries. I had been a parish gardener for a very long time, and I noticed that something was seriously wrong with me by then. At the time, I was very keen to contact occupational health care but the threshold for eligibility was very high. I feared losing my job and financial difficulties were a real worry. At that point, I sought help, but unfortunately in the wrong direction. I started self-medicating with alcohol. In retrospect, it was the worst thing I could do to myself at the time. I haven't consumed alcohol in eight years now, having stopped when I got up the courage to seek real help for myself.

I had been diagnosed with moderate to severe depression. I had also been diagnosed with memory disorder and psychosis. The main problem during that period was the lack of available appointments and insufficient communication between certain service units. One unit didn't always know what the other was doing. After a few months, neuropsychological tests and medical examinations/interviews were conducted. It wasn't that long before I finally got my diagnosis of FTD, which was a great relief.

I maintain my ability to function with diet, social contact, exercise and even the smallest things in everyday life. I do things that I like for myself, as much as I can do independently. Safety is important and also the art of forgiveness. I lose a lot of things, and of course, it's going to cause me a lot of difficulties. On the train or bus, I hold the strap of my bag, otherwise, I will forget it. I have problems with attention, concentration and short-term memory. I'm also very accident-prone, I stumble easily and my wife sometimes has to patch me up. However, I keep moving forward, with curiosity and courage. Despite my illness, life has given me things over these past years. I continue to move on and believe life still has more to give. I am prepared for the progression of the disease, for any new treatment that is developed and have prepared an advance directive. I have also told my wife and children about my wish to be separated from my family if my behaviour becomes aggressive or too difficult for them. I've prepared a playlist and picture galleries for me to use in case that dreaded moment arises in the hope that maybe they'll calm me down. I hope that time doesn't come, but life is full of surprises and so am I.

The focus on language, communication and interaction

Communication is a process involving the exchange of information, thoughts and feelings by and between individuals and groups. It is not a one-way street. Moreover, pieces of information are not necessarily understood or interpreted by everyone in the same way. Sometimes, the message received is quite different from the message that someone thought they had sent. It is not just a matter of understanding but also of respect. The manner in which information is communicated can, irrespective of how effective the message was communicated, contribute towards people feeling either valued and accepted or devalued and stigmatized.

Researchers are often involved in communicating information and interacting with people who have neurodegenerative diseases and with their carers (or supporters) in the context of research participation or Public Involvement work. They may also at some point be involved in communicating about or to people who have neurodegenerative diseases, as well as other members of the general public, who are simply getting on with their lives and not involved in research in any way (e.g. they might just hear about a recent study on the news). Whilst researchers may strive for clarity and respectful communication, it can sometimes be difficult to know how to go about this. This is not generally part of their training and many have had little or no prior contact with people with neurodegenerative diseases in their everyday lives. Others may have had contact with people from these groups as patients but this is not the same thing as in the research context (where people with neurodegenerative diseases may be research participants or contributing towards PI work).

Many researchers have had some experience of involving people with neurodegenerative diseases as research participants in their research but some may have been focusing primarily on existing data sets, carrying out experiments on laboratory animals, manipulating biological materials or involving computer simulations. Therefore, depending on the nature of the research and of the required contribution to the research, some researchers may have had little actual contact with people with neurodegenerative diseases, especially in terms of understanding their thoughts, experience and expectations about research and about living with a neurodegenerative condition. Researchers employing qualitative research methods are likely to have had more experience interacting with people with neurodegenerative disease, often exploring their personal experience of a relevant aspect of their experience of living with a particular condition.

Irrespective of researchers' level of familiarity with people with neurodegenerative diseases, there are several times before, during and after a particular research study when it is necessary to communicate to or about people with neurodegenerative diseases e.g. in the project proposal, in applications for ethics

approval and funding, in relation to participant-facing materials, for recruitment and retention measures, when conducting various tests and instruments, to raise awareness about the research and to disseminate findings at different events and levels (e.g. in an academic conference, public event or peer-reviewed article). The information and guidance provided in Part 2 of this deliverable is the result of PI work and is aimed at promoting meaningful and effective PI which will hopefully have a positive impact on the above-mentioned research-related activities.

Section 2: Methodology

The guiding framework and overall topic

The work currently being carried out in the EPND project and reported in Part 2 of this deliverable constitutes Public Involvement (PI) as defined in the classification and conceptualization developed by INVOLVE (2017)[1] and summarized in Figure 1 below. As there is often some confusion about the above terms, we will start by clarifying what PI is not, namely research participation or Public Engagement (PE).

Figure 1: Clarification of terms

Being a research participant	This consists of people taking part in a research study as a research participant (formerly often referred to as a research subject).
Public Engagement (PE)	This consists of people receiving information and being informed about a research study.
Public Involvement (PI)	This consists of the active involvement of people in research projects and in research organisations other than as research participants. It is about carrying out research and developing policies with or by members of the public and patients rather than on or for them.

Research participation and Public Engagement

Most researchers are likely to be more familiar with the concept of research participation. In the past, people who participated in research were called research subjects but the term research participant is now increasingly used and preferred because it is considered as more respectful, presents people less as passive subjects or material from which to obtain data and more like partners in the process of discovery (Grady 2022).

PE (like PI to some extent) is used in a range of different settings such as policy making, local government, the development of healthcare or community services, education and research. PE in research is about “engaging” with people outside the research setting (e.g. with members of the general public, people with specific health conditions, carers and supporters) with the aim of creating and maintaining their interest in the topic of the research being conducted and, in some cases, their potential willingness to use certain services or products or to accept or trust the results and implications of research findings. It is a two-way process in that it involves interaction and is hopefully mutually beneficial to all involved. Successful PE is likely to contribute towards a more trusting relationship between researchers and the general public and promote a healthy interest in research, which is vital for the research field, where difficulties with recruitment are frequently encountered.

[1] INVOLVE no longer exists. It was taken over by NIHR Centre for Engagement and Dissemination in April 2020.

Public Involvement

PI is about creating a partnership between researchers and the public, whereby all contribute collaboratively in varying degrees towards the research process. The term 'public' is understood as including members of the general public, patients and potential patients, informal (unpaid) carers/supporters, parents, legal guardians, people with disabilities and people who use, or have used, health and social care services. It does not include people who are employed as health or social care professionals, or academics. Our main focus in this report is on people who have neurodegenerative diseases and their carers/supporters.

PI is not a specific method but an approach to involving people in research other than as research participants or in PE activities or events. PI could be considered as an overarching term which groups together a wide range of approaches and methods designed to enable people to share their insight and experience of having a particular condition. The nature and extent of the involvement may differ from one research project to the next. It can occur along a continuum from involvement in an isolated task, through involvement at several or all stages of the research process up to full involvement as a core member of the research team. Furthermore, different people may be involved at different stages of the research or in different tasks. The rationale for PI is broadly based on two key arguments (Ives, Damery and Redwood 2013, Gradinger et al. 2015).

- The first, based on normative arguments, is sometimes described as ideological or process orientated. It emphasises ethical, social and political concerns. PI is portrayed as "an end in itself", linking involvement to democracy (e.g. democratic decision making, public accountability, legitimisation and transparency), people having rights (e.g. a right to voice, a right to be involved in research relevant to one's own condition) and to ethical principles of justice and fairness.
- The second, based on substantive arguments, is sometimes described as pragmatic, portraying PI as a "means to an end", linked to attempts to improve the quality, validity, relevance and/or utility of research. It can also be considered as instrumental in providing knowledge that might otherwise be missing. This includes, for example, highlighting issues and asking questions about things that researchers have perhaps not considered. Tritter and McCallum (2006) suggest that key contributions often arise from personal experience and a non-medical/non-technical frame of reference.

In the past, it was largely assumed that people with dementia were unable to express their opinions or share their experience. This was perhaps because dementia used to be diagnosed at a much later stage when people tended to have less capacity to express themselves with ease and when less was known about how to communicate effectively with people with cognitive decline. A narrow medical model of dementia was also the norm. This tended to emphasise deficit and did not adequately consider the impact of the environment (i.e. in terms of disability). Consequently, people with dementia were often silenced and represented by informal or professional carers who did not always have the same perspectives or fully understand their views and experience. It is now increasingly accepted that people with dementia have an important contribution to make to research and PI is increasingly required for research funding or ethics approval in some countries. However, whilst the concept is gradually becoming more common in the field of dementia research, the practice is not yet widespread across the whole of Europe.

In the context of mental health research, Patterson, Trite and Weaver (2014) point out that incorporating lived experience into the research process may improve the quality, relevance, acceptability and ethical status of research but is by no means universally accepted. Critics have claimed that conducting PI is time-consuming, challenging and often tokenistic. A key challenge, therefore, is to ensure that PI is

properly planned, with appropriate time allocated to it, with an appropriate budget and with due reflection about potential challenges, and that it is indeed never tokenistic. For more details, please see the next Section of this report.

In PI work, people share information, ideas, perspectives and insight which are unique to them (as a unique person with a neurodegenerative disease). This may or may not be shared by other people with the same condition. These people are not representative (in the statistical sense) of the wider population. The goal in PI is to gain some insight into their experience and its relevance to a particular aspect of the research that might otherwise be missed. It is not the goal of PI to provide generalisable data. Moreover, the feedback provided and insight shared, is not research data and is not analysed.

The two groups contributing to the PI work in EPND

The PI work in EPND helps ensure that the project is informed by the meaningful contribution of people with neurodegenerative diseases, and their carers/supporters. The PI activities have been carried out by the Patient Expert Group (PEG), which is composed of two groups which work in parallel, namely:

- The European Working Group of People with Dementia (EWGPWD): This is a working group of people with dementia which was initially set up by Alzheimer Europe (AE) in 2012. The current group started its 2-year term of office in 2022. It is composed of 15 men and women of different ages, with different types of dementia and from different countries in Europe. Each member of the group was nominated by a national Alzheimer association and is supported by a person of their choice to travel to meetings and for any assistance needed during consultations. Every two years, Alzheimer Europe's member associations are invited to nominate people with dementia to join the group and there is always a good mix of new members and others who continue for a second term of office.
- The Parkinson's Advisory Group (the abbreviation in Spanish is GAP for Grupo Asesor Párkinson): This group was set up by the Federación Española de Párkinson (FEP) specifically for the EPND project. In July 2022, the FEP shared information about the project with its members through its internal newsletter to the 67 local Parkinson's member associations. The FEP then sent emails in August 2022 inviting people with Parkinson's who had been trained through its online course ('How to participate in research studies') to take part in the GAP. Following AE recommendations, the FEP created an online form for people to express their interest in taking part and 34 people signed up. Later on, FEP organised two informative meetings (on 5 and 6 September) to explain the project and provide details about participation. Finally, the FEP sent another online form for people to join the GAP and 17 people confirmed their willingness to become a member of the group.

PI activities/work

The PI work consisted of joint coordinated PI activities, which were planned and facilitated by AE and the FEP. Background documents in plain and accessible language and format were prepared and shared with both groups a week in advance of the meeting. This included background information about the topic and an agenda. AE and the FEP conducted the following consultations with groups of people with dementia and Parkinson's disease or parkinsonism, in English and Spanish respectively. The feedback provided by the members of both groups was written up and discussed by AE and the FEP.

The consultations were organised around key themes, namely:

- clear and accessible information,
- respectful communication and interaction,
- promoting inclusion,
- barriers and facilitators to meaningful and effective PI.

The members of the EWGPWD met virtually and in person several times:

- In each of the three face-to-face meetings of the EWGPWD in Luxembourg and Brussels (in May, June and September 2022) a session was dedicated to EPND. During these sessions, members received information about the project and discussed issues related to respectful, ethical and inclusive communication about and portrayal of dementia and people with dementia.
- In addition, two online meetings were dedicated to this topic. The first session took place on 29 November 2022 and the second on 30 January 2023. The discussions focused on the topics of clarity, reciprocity, and barriers and facilitators to PI work. Existing published guidelines on these topics were reviewed with the aim of revising and elaborating them.
- The idea of the glossary of research-related terms for lay people was discussed and the members of the EWGPWD agreed to review sub-sections of the glossary that AE was in the process of drafting.
- The face-to-face and online meetings were attended by members of the EWGPWD (2022-2022 and 2022-2024 term of office).

The members of the group involved in the various meetings were in their late fifties to eighties and were supported by a person of their choice (i.e. a member of their family[2], a friend and, occasionally, a volunteer from an Alzheimer association). Supporters attending the meetings were also invited to share their views on the topic in their own right and in their capacity as a supporter/carer. In total, 16 people with dementia and 8 supporters contributed toward these discussions.

The members of the GAP met twice online.

- The first meeting was held on 30 November 2022 and was attended by 11 of the 17 members of the GAP. This included four women with Parkinson's disease and seven men, of which six had Parkinson's disease and one was affected by parkinsonism. The average age of the participants was 60 and they came from eight of Spain's 17 Autonomous Communities. Seven of the participants were members of a local Parkinson's association and four were not. The following topics were addressed: clear communication and terminology, ethical communication and respectful communication
- The second meeting was held on 9 February and was attended by 12 of the 17 members of the GAP, including five women affected by Parkinson's disease and seven men (five with Parkinson's disease and two affected by parkinsonism). The average age of the participants was 62 and they came from eight of the 17 Autonomous Communities. Seven of the participants were members of a local Parkinson's association and five were not. In this meeting, the following topics were addressed: the concept of reciprocity and barriers and factors that facilitate the participation of people with PD or parkinsonism in research.

[2] Whenever we use the term "family" in this report we are referring to biological families and families of choice A family of choice, also known as chosen family or found family is a group of non-biologically related people who are important to someone and may, like biological families, provide support and care to a greater or lesser extent. Families of choice are common within the LGBTQ+ community, groups of veterans and within supportive communities overcoming shared trauma.

Section 3: Practical issues related to inclusive and meaningful PI

Introduction

In addition to reflecting on how to communicate about neurodegenerative diseases and how to interact with people with these conditions respectfully (see Sections 4 to 6), inclusive and meaningful PI requires careful attention to the organisation and conduct of this kind of involvement. In 2018, Alzheimer Europe produced a position paper on PI together with the INTERDEM[3] research network and the EWGPWD. This was followed by subsequent work under EU operating grants to explore facilitators and obstacles to meaningful and successful PI. This topic was further discussed by the EWGPWD and the GAP during the virtual and face-to-face meetings mentioned earlier. The key points are presented in the guidelines below.

Guidelines

Reasonable accommodation/making appropriate adaptations

When planning and conducting PI with people with neurodegenerative diseases certain adaptations may need to be made because of various impairments that people with these conditions have and which often result in disability.

Researchers should be sensitive to the needs and abilities of the people carrying out PI work and make any adaptations needed. Dynamic structures and processes are needed, which should be developed by both people with neurodegenerative diseases and researchers. The former must be empowered to help shape the methods and approaches to their involvement (Tritter and McCallum 2006). Their capacity to do so may change over time but is also dependent on efforts being made to facilitate this kind of involvement in constructing and constantly refining these processes.

Such adaptation is linked to the concept of disability and is an example of "reasonable accommodation" (which has nothing to do with having a roof over your head or a room in a hotel). Reasonable accommodation is described in Article 2 of the United Nations Convention of the Rights of Persons with Disabilities (CRPD, 2006)[4] as:

> *"necessary and appropriate modification and adjustments not imposing a disproportionate or undue burden, where needed in a particular case, to ensure to persons with disabilities the enjoyment or exercise on an equal basis with others of all human rights and fundamental freedoms"*[5]

[3] Interdem is a pan-European network of researchers collaborating in research on and dissemination of Early, Timely and Quality Psychosocial Interventions in Dementia aimed at improving the quality of life of people with dementia and their supporters, across Europe.

[4] For a discussion about dementia as a disability, please see Alzheimer Europe's 2017 report on this topic: https://www.alzheimer-europe.org/Publications/Alzheimer-Europe-Reports

[5] Please see: https://www.un.org/development/desa/disabilities/convention-on-the-rights-of-persons-with-disabilities/convention-on-the-rights-of-persons-with-disabilities-2.html

Under the CRPD, people with disabilities include

> *"those who have long-term physical, mental, intellectual or sensory impairments which in interaction with various barriers may hinder their full and effective participation in society on an equal basis with others" (Article 1).*

Reasonable accommodation requires an investment in terms of time, effort and funds. With specific regard to respectful and inclusive communication and interaction, it means, for example:

- providing clear and easily understandable background reading materials,
- avoiding the use of jargon and acronyms,
- explaining terms upon request and/or through a glossary of relevant terms (including acronyms),
- providing any necessary written information about the topic and what is expected in advance,
- getting to know individuals doing PI in order to better understand their habits, customs, values, fears and expectations, including how they feel about the condition they have (e.g. in terms of having concerns about certain symptoms they are experiencing, feeling apprehensive about the future or having a more balanced or positive approach etc.),
- understanding that the desire, strength, ability and motivation to contribute to PI activities of some people with chronic, neurodegenerative conditions (such as PD or a form of dementia) may diminish over time but may also fluctuate,
- researchers having the necessary expertise to understand the needs of the people with neurodegenerative diseases,
- researchers being open to interacting with and willing to adapt to the needs of the people with neurodegenerative diseases,
- making sure that there is a support person if needed before or during the meeting (e.g. to help people keep track of the discussion, find their place in the materials and formulate and express their ideas, and to help out in difficult or unforeseen situations),
- using appropriate support materials to compensate for memory difficulties, disorientation and confusion (e.g. flip charts, written summaries of key issues, an agenda and good signposting),
- using available technology to support PI (e.g. online agendas, digital reminders and links to free AI translation tools for people not working in their mother tongue),
- providing sufficient breaks and limiting the duration of discussions to avoid exhaustion and help people remain focused,
- giving people time for reflection and, where necessary, for translation/interpretation,
- speaking slowly and clearly (but not speaking to people as if they were children or deaf),
- ensuring that non-academic staff at research centres and meeting venues are also respectful and attentive to the needs of people with neurodegenerative diseases (training should be provided).

Rendering research and the research topic accessible

In some cases, people with neurodegenerative diseases engaging in PI have prior or even current experience of conducting research. When this is not the case, some researchers feel that people engaged in PI should be offered training so that they can better understand the topic, the issues and the implications of adopting various methods, as well as what different tests and procedures involve, and thereby contribute more meaningfully to discussions. Brett et al. (2014), for example, argue that training in research methodology would empower people to contribute to discussions surrounding the research design and to ask questions about the study rather than limiting their contribution to accounts of their

lived experience. Such training may be helpful and beneficial if it is the right level for the people involved but if perceived in terms of training people with neurodegenerative diseases 'to bring them up to the level of researchers', it could be construed as failing to value the knowledge that they already bring to the research process. Moreover, a key aim of PI is for researchers to benefit from the experience and insight that people with neurodegenerative diseases have in relation to the research topic and the conduct of their study based on the fact that they have personal experience of a particular condition. In addition, people doing PI have 'outsider' status and the ability to be a critical observer, free from the concerns and pressures of publication, generating income and building up an impressive CV.

A possible exception, which is quite rare in large scale research projects, is where people engaged in PI actually 'do' research (e.g. help recruit participants, gain informed consent and collect and analyse data). This is in keeping with the concept of PI occurring along a continuum and with the possibility of people with neurodegenerative diseases being co-researchers but they must have the necessary scientific rigour to conduct high quality research and this poses certain challenges. Researchers may, for example, need to obtain informed consent from research participants for their anonymised data to be shared with people engaged in PI and supervision would be needed (Hoddinott et al. 2018). It may also be necessary to address certain governance issues (e.g. in relation to terms of employment, legal issues and the right to payment (Hoddinott et al. 2018). Such involvement would need to be supervised because a full grasp of all the relevant issues and assimilation of knowledge required to conduct high quality research would normally take several months or years to acquire, not just a couple of hours.

If researchers present PI as something for which training is necessary, this may give the wrong message to people who might otherwise be interested in it. It may be perceived as indicating that PI is only for highly educated people or for people who are good at and willing to study. People who do not fall into that category may feel discouraged or disempowered. Appropriate background materials and support (also involving various researchers with relevant expertise in a particular area) should be the goal, with the aim not of turning people doing PI into researchers but to help them to understand the research process. At the same time, researchers should have opportunities during their studies to learn about PI and to acquire the appropriate skills to make PI accessible to the people involved (Hoddinott et al. 2018).

Planning and conducting PI

People considered doing PI work should receive clear information beforehand about PI and what to expect (e.g. about the overall topic, how often the group would meet or if on an individual basis how often they would be consulted, how long meetings would be, how frequent and at what time of day, who else would be present, how costs would be covered, whether it would be necessary to travel, that it is a voluntary, usually unpaid, activity and what would happen to their input). A first meeting should clarify these issues and allow for any questions and suggestions. Researchers should ensure that they have the necessary experience (or support from others who do) when planning and conducting PI work.

PI work can be valuable at all stages of research. It can be included at the very beginning of a research project (i.e. in discussions about research topics, in the development of research proposals and for ethics approval), throughout the research (e.g. helping develop participant-facing materials and in relation to factors linked to recruitment and retention) and at the end of the research (e.g. for dissemination materials to lay people). People with neurodegenerative diseases are often absent at the stage of development of research proposals. In many cases, their involvement starts when the project starts because funding for PI has not yet been granted and researchers have perhaps not yet established necessary links to people

with neurodegenerative diseases. Once the research is underway, it may be difficult for the people doing the PI work to shape the nature of their involvement because their ideas do not correspond to the details in the proposal and the available budget. If and when it becomes apparent that certain decisions have already been made, people engaged in PI may be reluctant to raise questions or suggest changes, anticipating that their input will be opposed, ignored or not considered a realistic option (Morrow et al., 2010).

Clarifying roles and responsibilities

People with dementia and other neurodegenerative diseases are increasingly being invited by researchers to engage in PI. Whereas co-production and co-design may occur in smaller projects or for certain aspects of research, in large-scale, international research projects, research consortia and clinical trials, involving hundreds of researchers from 10 to 15 different countries on average, it is clearly the researchers who play a leading role in determining the timing, duration and frequency of PI. They also take the lead and have final responsibility for certain tasks (e.g. linked to accounting, safety issues, applications for ethical approval, reporting obligations and statistical analysis).

Respect and recognition of the value of different people's contribution to research should not be interpreted as the need to deny differences in people's roles and responsibilities. At the same time, attention should be paid to how people's roles and responsibilities are portrayed and communicated so as to avoid social positioning and the creation of unnecessary friction between people who are all investing their time, skills, energy and experience in the research. Terms such as 'expert by experience' and 'expert by training' may reflect different types of expertise and reflect the belief that different kinds of knowledge can exist side by side. However, if each type of expertise is truly valued on an equal basis, this has to be demonstrated through actions not just words.

Respecting diversity

The people doing PI work are not supposed to be representative (in the statistical sense) of the population of people with the condition that they have. However, it would be fair and beneficial to research to involve people with all kinds of different characteristics (e.g. people from different ethnic cultures, with different religions, from different socio-economic groups, from rural as well as urban areas, living in the community or in care homes and with different gender identities and sexual orientations).

It is not sufficient to state that everyone is equally welcome to join a PI group. Measures must be taken to attract different people to PI work by ensuring that they feel welcome, that it is relevant to them and their lives and that they would be able to contribute meaningfully. To achieve this, researchers should advertise or reach out to people in places that are not predominantly typical of White, middle class, highly educated populations (e.g. also in barbershops, synagogues, grocery shops, newsagent's shops, GP surgeries and community centres, especially in areas with multicultural populations) and to contact respected gate keepers (i.e. respected and trusted members of different communities). This is clearly most feasible at local level where community groups can be easily identified and where direct contact, also with relevant gatekeepers, can be made.

As older people from some marginalised groups may have more difficulties with language and using technology, and have low incomes, it is important to make it clear what is expected, how costs are covered and what kind of support is possible. The terminology and images used in materials to attract people to PI work also need to reflect the acceptance and promotion of diversity. However, it should not

be assumed that people from marginalised groups (e.g. in the case of people with literacy or hearing problems, from a traveller community or from the LGBTQ+ community etc.) are visibly recognisable. For further information on this topic, please see Alzheimer Europe's reports on the ethics of inclusive research, intercultural care and support, and sex, gender and sexuality in dementia.

Resources and financial issues

There are a few financial issues to consider. The first is to ensure that there is an adequate budget for PI and this needs to be included in the project proposal. It should include funds for person months, travel to meetings (e.g. the general assemblies and steering committee meetings, materials and face-to-face PI meetings.) For PI work involving people from different countries, the costs of such meetings can be fairly high as they need to include flights, accommodation, meals, meeting room hire and equipment for approximately 10 to 15 people. In the case of people with dementia, this cost is doubled because the people doing the PI work need to be accompanied by a support person. The second is to ensure that people are not out of pocket and do not have to comply with complicated reimbursement procedures or wait long periods of time to recuperate any outlay.

In the vast majority of cases, people contribute to PI as volunteers. In the last few years, there has been a movement towards paying 'patient representatives' for their expertise, time and effort based on concerns about fairness, equality, acknowledging the equal value of the expertise they provide and as a sign of respect. However, payment does not automatically convey or guarantee respect for the person receiving it and their contribution. Likewise, voluntary involvement does not preclude respect for a person's contribution or value. On the other hand, people with dementia and other neurodegenerative diseases who have limited opportunities to earn money or limited financial resources might appreciate the opportunity to be paid. Payment might also encourage people from more diverse backgrounds to engage in PI (i.e. people on a low income, people who are perhaps less motivated by an awareness of social and health-related issues and/or people would not normally do volunteer work). For people who are on a low income or receiving benefits from the State, such payment may be problematic in that it could jeopardise their rights to benefits, result in them having to make complicated or costly tax declarations or involve a risk of them being accused of fraud (e.g. based on travelling to and contributing towards meetings and discussions despite having been declared unfit to work on the grounds of disability).

In its position paper on PI, drafted in collaboration with INTERDEM and the EWGPWD, AE argued that if funds are available for the payment of external experts (e.g. fees to attend a meeting or daily allowances), the same money should be offered to people with dementia engaged in PI on an equal basis but that PI conducted on a voluntary basis was not problematic *per se* (Gove et al. 2017).

Keeping people informed

Researchers should keep people doing PI work updated about the progress of the study and the impact of their contribution to the research. Sometimes, people who have done PI in research have found out about the results indirectly (e.g. through the media) and long after completion of the study. This is not appropriate or respectful.

All information about the research should be accessible (please see section on reasonable accommodation) and in plain language (please see guide to plain language in the resources section). When researchers do not share information and use technical terms, this may be perceived by people doing PI as inconsiderate and as the researchers displaying their perceived "superiority".

Reporting on PI and acknowledging the contribution of the people involved

Research funders and ethics committees increasingly ask for evidence of PI in research proposals. Nevertheless, the conduct and outcome of PI work is frequently under or inadequately reported. Even when reported (e.g. in formal reports, official records and/or public project websites), there is often a lack of detail. This is not helpful in promoting the value of PI work and of the contribution of the people involved.

> *"There are often concerns about whether PI work is meaningful or a mere box-ticking exercise to obtain funding or ethics approval. To ensure that PI work truly contributes towards good research and that it is meaningful and well-conducted, it must be reported thoroughly and accurately. It cannot be a "black box" activity (e.g. "we conducted PI work")" (Georges et al., 2022).*

It is important to plan for at least one public deliverable for PI work and to publish comprehensive details of the PI work on project websites and other places that are accessible to the public, as well as in peer-reviewed scientific journals. The people involved in PI activities should be acknowledged for their work (e.g. on public websites, on social media and also in peer-reviewed articles in scientific journals). It should be verified whether they would like to be individually named (e.g. in the acknowledgement section) or acknowledged as a collective group. Usually, members of PI groups prefer to be acknowledged as a group but often this is combined with a link to a project website where their names and short bios can be found. The solution adopted should reflect the wishes of the people involved.

Section 4: Respectful and meaningful communication/interaction in the PI process

Introduction

Respectful communication and interaction to or with people with neurodegenerative diseases and their carers/supporters is very much about what we say and how we treat people. However, it is also closely related to broader concepts and principles such as embracing diversity, striving for inclusivity, respecting and valuing the knowledge of all those involved, sharing power, respecting personhood, ensuring reciprocity, and building and maintaining relationships (INVOLVE 2018, Alzheimer Europe 2021). This should be reflected in all PI work, without any of these concepts necessarily becoming the focal point, the sole measure of involvement or participation, or the criterion for the evaluation of a particular communication or interaction.

Guidelines

Terminology related to the PI concept and to the people involved

Starting, nevertheless, with the words we use, a key issue when involving people with various neurodegenerative diseases in PI work is how to refer to the concept of PI and how to refer to the people doing it. Several terms are used such as a PI group, a working group, a patient expert group or a patient panel. The term 'involvement' is sometimes preceded by the term 'public', 'public and patient' or just 'patient' (e.g. Public Involvement, Patient and Public Involvement or Patient Advisory Group). Public Involvement, and variations of this term, is about involving people either because they have lived experience of a particular condition and/or because they are members of the public (i.e. not health or social care professionals or academics).

The reference to patients is increasingly considered as problematic in that a lot of people with dementia, for example, do not see themselves as patients and are voicing their concerns about being 'positioned' in this way (Alzheimer Europe 2021). Similarly, in the consultation with people with PD and parkinsonism, most people stated that they preferred to be addressed by their name and only as "patients" in the medical setting. One person stated,

> *"I like to say that I am a healthy person with a health problem",* adding *"I have the same needs and desires as everyone else. I don't want to be obsessed with illness" (Person with PD).*

Everyone who consults a medical doctor is at that moment a patient but this is not the sum total of their identity. In some research projects, clinical researchers may have contact with people with research participants who actually are their patients. They should, however, bear in mind that many people with neurodegenerative diseases involved in a particular research study are not their patients (e.g. those involved in PI work or reading about the study in the context of PE activities) and do not identify with this term.

At AE's annual conference in The Hague (in 2019), presenters and members of the audience with dementia emphasised the desire to move away from "patient-focused" language to that of the "public" or the "person", specifying clearly which members of the public and which people are involved in a particular activity. In research involving people with a range of different medical conditions and with a specific focus on biomedical or clinical research, it may sometimes be acceptable to use the terminology of "patients" provided that the people contributing to the PI work find the term acceptable.

People who support someone with a neurodegenerative disease doing PI work are often referred to as "carers". Some find this term acceptable and appropriate whereas others prefer the term "supporter". The reason for this is that "carer" tends to be interpreted as implying a particular relationship (literally of one person providing care to another) but people in the early stages of a neurodegenerative disease do not necessary feel that they need "care". Sometimes, people with neurodegenerative diseases are part of a mutual relationship of care (e.g. between a parent and adult child or between couples who each have a chronic medical condition). The term supporter can be interpreted as relating to the PI work rather than to coping with a neurodegenerative disease.

Promoting meaningful PI and avoiding tokenism

Ensuring meaningful and valuable PI touches on respect for the individual in many ways and, if carried out properly, contributes towards good quality research. If approached in a tokenistic way, it involves deception, contempt and exploitation, perhaps providing a seat at the table, but failing to take into consideration people's contribution or to meaningfully integrate the feedback provided into the actual project or research process. Hopefully, most researchers carry out PI on the basis of ethical, pragmatic and methodological motives. However, as recognition of the importance of involving people with dementia in research increases, so too does the risk of tokenism (Brett et al. 2014, Hardavella et al. 2015). Researchers sometimes conduct PI simply because it is a necessary requirement for funding or in some cases to obtain ethical approval. In such cases, researchers often contact patient organisations with requests to involve people with neurodegenerative diseases in their research at the last minute, shortly before submission of a proposal, without any clear plan or goal, and without appropriate funding. If the rationale or reason for conducting PI is not genuine, then all subsequent communication with the people involved is unethical and disrespectful, wasting people's time and energy, and giving them a false sense of achievement.

Ensuring reciprocity

There are lots of different definitions of reciprocity and this is a term that can be used in different contexts (e.g. for friendships, in close-knit communities and in relation to the provision of care or support). Definitions often emphasize the practice of exchanging things with others for mutual benefit, often implying an element of gratitude and respect. In the context of research, and particularly in relation to PI activities, the concept of reciprocity has received some attention. PI in research is sometimes described as providing an opportunity for reciprocity, with people contributing to something meaningful and useful to society and receiving something in return for that contribution. Some people consider it a moral duty for researchers to ensure that people contributing to research receive something in return. Some link this to financial remuneration but as mentioned in Section 3, payment is not always considered as synonymous with reciprocity. Reciprocity is about giving something freely. It reflects kindness, consideration and genuine recognition for what has already been given freely and out of kindness, not a mere duty-bound or business-like transaction or payment. Comments from people with neurodegenerative diseases highlighted the emotional aspect of reciprocity.

> *"Reciprocity to me is almost an emotion. It is not a payment. It is not a given. It is almost like bartering but then it is not like bartering because I would do this for you and if you do that for me, that would make me feel better. So it is the emotion in the word reciprocity, which is different from gain" (Person with dementia).*

> *"There is no expectation but an offer of what I need. I love it when they think ahead. To me, that is real reciprocity. It is not really what they give you or the gain, it is 'What can I do for you in return for your kindness'" (Person with dementia).*

With regard to symbols or tokens of appreciation of PI work, examples include a simple thank you, a gift or gift voucher or even a medal/award. Some people consider the opportunity to gain insight into and information about their condition as a valuable return. It is important to understand at the beginning of PI work what motivates people to be involved in research in this way so as to help avoid them being disappointed (if this is unrealistic) and to bear in mind when thinking about what they might possibly appreciate.

Providing feedback and sharing the results

Receiving feedback about the study, the results of the research and/or the possible impact of the PI work is important to most people who do PI work. For some, it is a sign of respect, for others an obvious expectation and for yet others, important for their wellbeing linked to wanting to contribute to society and experience a sense of achievement and utility. This should not preclude sharing the results of research they might have contributed to which did not bring the results they perhaps hoped for.

> *"You provide input to researchers that is valuable to them and in return you get a good feeling because you know that you have been productive, have accomplished something and have a sense of ownership, and you don't get much of an opportunity as someone with dementia to really feel that" (Person with dementia).*

However, as another person commented, it might be enjoyable taking part in things, but that should not be the sum total of reciprocity. Moreover, to have that feeling of accomplishment, it is often necessary to see what difference the PI made. This means researchers being totally open and also explaining why certain suggestions were not implemented. In some research projects (e.g. in RADAR-AD)[6], the researchers have been particularly good at doing this and this has been greatly appreciated. Some pharmaceutical companies have also taken on board the importance of providing feedback about PI input provided. Roche, for example, after having carried out a series of consultations with the EWGPWD, recently invited members of the group to a 2-hour zoom webinar entitled "So what on earth happened with your feedback?". One consequence of such thoughtful, well-organised feedback sessions is that it further strengthens the relationship of mutual respect (see below) that has been built up and the reputation of the researchers or company.

Whereas some people who do PI activities appreciate the feeling that that they are still able to be productive (perhaps linked to self-esteem), others appreciate the feeling that they are contributing to society or to the "common good" as some called it. In both cases, feedback from researchers about the value of their feedback is important. In the GAP, a link was made between knowledge and power. It was suggested that a hierarchy is created when researchers, who have information and knowledge, do not share it with people doing PI, as it puts them in a position that may be perceived as superior. Perceived or actual hierarchies may also be communicated through the use of academic titles (e.g. Dr or Prof.), wearing white coats (like those worn in clinical and laboratory settings) and in assumptions that people doing the PI work should adapt to the timetables and availability of the researchers (although many

[6] Add a link to information about PI in RADAR-AD (e.g. article, public deliverable or webpage)

people involved in PI work are willing to adapt to the requirements of researchers which are often understood as being linked to constraints beyond their direct control).

Promoting mutual respect

Mutual respect, in the sense of one human being showing respect for another, is a basic requirement for researchers and people with neurodegenerative diseases to work together in the context of research. Mutual respect is about people valuing each other for who they are and what they bring to the table. It means recognising people's unique contributions whilst also understanding and appreciating differences, which include, amongst other things, the nature of what people bring to the table. As mentioned earlier, this also means recognising that people with neurodegenerative diseases have expertise that is of equal importance to that provided by researchers, and both they and the researchers valuing each other's efforts.

Mutual respect also challenges hierarchies of knowledge and expertise i.e. 'of the expert versus the lay subject', and recognises that communication is not 'a one-way transfer from a knowing subject to a supposedly ignorant one' (Porter, 2010) and that there are 'experts by training' and 'experts by experience' (Cheffey, Hill, McCullough and McCullough 2017).

> *"You might all have a long history of research in the field of dementia but we, who live with dementia, can tell you much better about our demands, needs and wishes. We are the real experts of our condition! Don't use me but involve me!" (Person with dementia).*

The notion of mutual respect should not be interpreted as the need to deny differences in people's roles and responsibilities. Members of the GAP suggested the need for researchers (and any health and social care professionals involved in the PI process) to put themselves in their shoes and communicate in a way that does not suggest a hierarchical relationship towards them.

Another issue related to mutual respect is that of recognizing each other as individuals and being inclusive. This may include being hospitable. Researchers may sometimes be invited into the homes of people with neurodegenerative diseases in the context of research (e.g. for qualitative interviews or in relation to PI work). In some cultures, it would be very impolite to refuse offers of hospitality such as tea and biscuits. It is also important when entering people's homes to be sensitive to people's customs and adapt accordingly (e.g. taking off your shoes at the door, respecting the privacy of others present in the person's home etc.). It could be argued that this is just the kind of respect that should be shown to everyone and in a way, it is. However, It may take on particular importance to people with neurodegenerative diseases and their families who may have experienced stigma or fear being considered as "other", or in some way inferior or incapable of providing what they think the researchers want from them. They may also be anxious about certain symptoms becoming obvious or interfering with the interaction. Welcoming strangers into their home may be stressful and offers of hospitality serve an important social function whereby the accepting is just as important as the giving, involving mutual respect and acceptance, a good starting point for a constructive and meaningful exchange.

Unfortunately, as also explained in the section on stigma (please see page 21), people who are not familiar with neurodegenerative diseases and their impact on people who have them are often influenced by stereotypes which tend to focus on deficits and incapacity. In most cases, no disrespect is intended but people with neurodegenerative diseases may nevertheless feel excluded and offended.

"A researcher came to visit us at home and it was me for to do some survey work and answer some questions, and she started by explaining all about the research to Mary and did not look at me once. So Mary waited until she had finished and said, "Now can you explain it to Barry" (Person with dementia).

"By not learning beforehand a little bit more about the condition of dementia, she didn't show Barry that respect and therefore we didn't show her the respect and asked her to repeat it all again. Had she shown Barry respect, we would have reciprocated by showing her the same respect" (Carer).

"Yes, it is about mutual respect. Respecting each other and also knowing about each other, knowing about each other's needs. Mutual respect is about respecting the researchers for their knowledge and them respecting us for our knowledge" (Person with dementia).

Respect for people and for their contribution to research can be fostered in a number of ways, for example by:

- getting to know about each other (e.g. sharing a dinner before starting the PI work, having a round of introductions before starting work, chatting during coffee breaks, exchanging brief bios and photos before meeting and asking questions etc.),
- building up a friendly working relationship over the duration of the PI work,
- marking the end of the working relationship (not abruptly ceasing all contact with no thank you, feedback or explanation),
- getting the balance right between giving and taking, speaking and listening,
- learning about each other's differences (e.g. finding out about each other's needs and preferences, their ways of working, preferences and constraints etc.),
- promoting good manners (e.g. being polite, courteous and considerate, addressing people in the way they prefer, taking into consideration possible cultural and generational differences, acknowledging gender identities etc.),
- being inclusive (e.g. not talking over or about a person with a neurodegenerative disease who is able, with appropriate support if necessary, to express him or herself),
- having a zero-tolerance approach to disrespect (not only being respectful yourself but ensuring that others are too, challenging derision, rudeness, hostility and negative stereotyping etc.),
- clarifying and recognising boundaries regarding roles and responsibilities so that people know what is expected from them and how they can best share their unique experience. Mutual respect does not mean assuming that everyone has the same abilities, skills, roles and responsibilities,
- Avoiding unnecessary signs of hierarchical position (e.g. wearing white coats like those worn by doctors and laboratory technicians).

Being sensitive to people's emotional and psychological wellbeing

Whilst neurodegenerative diseases may have a considerable impact on people's lives (e.g. in terms of memory, concentration, abstract thinking and orientation etc.), they can also be life-changing diagnoses that can affect people emotionally and psychologically. Some people report feeling angry, frustrated and overwhelmed. Some have feelings of low self-esteem or become depressed linked to the feeling that they and their lives have changed so much. Even when a topic is not particularly challenging or sensitive, people with dementia may sometimes experience frustration, disappointment or sadness linked to the changes that they observe in themselves which may become apparent during a research-related activity

or which were already on their mind. It is important that researchers are non-critical, patient and sensitive, and that they are supportive if and when needed.

> *"I watched my grandfather die with the condition and now I've got it. Dementia has made me another version of myself. I feel that I'm looking down my eyeglass at different aspects of myself" (Person with dementia).*

> *"It took me six months to accept the diagnosis. At first, I felt lost and angry" (Person with dementia).*

In the case of PD and parkinsonism, the person's state of health changes over time, even over the course of a single day, which means that they do not necessarily know how they are going to be at any given time. Motor and non-motor limitations may undermine their confidence in attending research appointments and in their ability to continue participating. This can have a significant psychological impact on them. For example, they may experience feelings of guilt at not fulfilling what they have committed to if unable to attend a consultation because they are not feeling well that day. As a reflection, one participant recalls how important it is to maintain a positive but realistic attitude when dealing with these ups and downs.

Bearing in mind and responding to what motivates people to do PI

There have been several accounts of factors which motivate people with various conditions to take part in research, either as participants or in the context of PI. This includes some debate about financial remuneration for people doing the PI work (e.g. whether this is a necessary sign of respect or somehow missing the point of voluntary work). In the PI work for this project, and as reflected in much of the literature on this topic, the main motivation seems to be altruism with an emphasis on the desire to contribute something to society and this resulting in a feeling of hope, achievement and being part of something bigger. The concept of "the common good" was raised in the consultations and considered by several people as being important.

> *"I just want to do it for the common good" (Person with dementia).*

> *"It gives me hope… hope for the future, hope for the people, for the next generation" (Person with dementia).*

> *"The words for the common good really spoke to me. Because for me it feels like a pool and people put in and they take out. It is much wider than just one person to another and that's very important to me. It really moved me the idea that it is a common good thing rather than a one-to-one basis" (Person with dementia)*

The issue of motivation is linked to several other issues addressed in this report, such as reciprocity, ensuring that people are not financially out of pocket, respect, gratitude and keeping people informed about progress with the study. It is important to consider what people would like to achieve through their participation in PI work and to ensure, in the spirit of reciprocity, that this is possible. Even in the case of altruism and a desire to contribute towards the common good, people need to know that they are accomplishing this, and perhaps some kind of recognition of this.

> *"It is just for the common good and of course we would like to receive some kind of respect and gratitude basically for our work, what we do" (Carer of a person with dementia).*

"They gave us a lot of really good feedback about the impact of our work and that gave us great value, a sense of purpose and also the incentive to carry on" (Person with dementia).

Challenging stigma

It is widely accepted that people with neurodegenerative diseases experience stigma but it is not always clear what different people mean or understand by stigma and how stigma is perpetuated. This is important to consider in the context of research as certain beliefs, attitudes and practices may challenge or reinforce such stigma. This may have an impact on the people involved and their families, but also on their successful and meaningful involvement in research. There are several conceptualisations of stigma. The one below, which was developed by Link and Phelan (2001, 2006), is useful in that it breaks down this complex social phenomenon into a series of components, each of which has implications for respectful and inclusive PI work. Stigma occurs when the various components are present and in the context of unequal power relations.

Figure 2: Conceptualisation of stigma

Identification and labelling	A socially salient attribute, known as a "stigma" or "mark" that is shared by a group of people, is singled out and labelled. This tends to be something that creates unease, concern or some kind of fear (e.g. fear of deterioration of moral standards, of injury, suffering, loss of self, death, social unrest, or a threat to social order or the established *status quo* etc.).
Stereotyping	Other attributes, usually of a negative nature, are associated with the labelled attribute (e.g. people with mental disorders are dangerous).
Cognitive separation	People with the labelled attribute are considered as being "other" (i.e. not like me) in the sense of "us" versus "them".
Emotional response	The people with the attribute elicit an emotional response, usually fear, pity or anger.
Devaluation	The people with the attribute are considered as having less value (being tainted, discredited, inferior, damaged, deficient etc.).
Discrimination	The people with the attribute experience discrimination (e.g. they are denied the same rights and privileges as other people or are socially excluded).

It can be useful to consider the above components when planning or conducting PI work. For example, it is important to reflect on:

- assumptions that you might be making about people with neurodegenerative diseases,
- whether these assumptions are accurate or stereotypes,
- how you react to people with neurodegenerative diseases emotionally (e.g. with pity, anger or fear) and why,
- whether you consider people with neurodegenerative diseases as somehow "other" (i.e. not like you),

- whether you consider people with neurodegenerative diseases as inferior (i.e. having less value than yourself or other people) and
- whether you discriminate against people with neurodegenerative diseases in some way, find certain unequal treatment somehow acceptable or avoid social contact with them.

In the consultations, people with PD explained how researchers and clinicians sometimes make comments about their state of health which they do not think are accurate and reflect stereotypes e.g. "You look well. You don't seem to have Parkinson's". This suggests that they have a stereotypical image of what someone with PD looks like or should look like (which does not correspond to the image of the person in front of them). It should also be noted that when people are not feeling well, they often stay at home.

People with PD and dementia also commented on how researchers and clinicians sometimes look at them when they are experiencing more obvious symptoms (e.g. motor symptoms such as dyskinesias, confusion or memory loss), in a way that they feel is stigmatizing, making them feel "faulty", devalued and "other".

At times, people with neurodegenerative diseases are faced with assumptions about their capacity to consent, their ability to do something or their quality of life, merely on the basis of having a diagnosis. This limits the opportunities that are available to them, sometimes resulting in over-protective measures or influencing researchers' interpretations of their input (leading to false conclusions and thereby invalidating the experience or insight that they have shared).

If some of the behaviour or the attitudes of researchers towards people with neurodegenerative diseases reflect the components of stigma mentioned earlier, researchers may in some way be contributing towards or perpetuating stigma. PI work is sometimes hampered by stigma but at the same time, it provides an opportunity for researchers to better understand the experience of people with neurodegenerative diseases, to relate to them better as valued individuals and together to challenge the stigma of these conditions. Some of the feedback provided in Section 5 below (e.g. in relation to how such conditions are portrayed and the use of imagery and metaphors) is relevant to this issue.

Section 5: Respectful communication about neurodegenerative diseases

Introduction

How we represent neurodegenerative diseases and the people who have them in words and images can influence how we think about and treat people with those conditions. It has implications for the lives of millions of people worldwide and also for their rights (i.e. to full and equal participation in society, and access to healthcare and treatment) and for relationships and emotional wellbeing. It can also significantly influence decisions about research priorities, service development and policy. In this section, we look at communication and portrayal. Communication is a broad term and involves sharing or exchanging information verbally and non-verbally, also including images (e.g. in a picture, film, video, book, speech or report).

Preferences and objections to certain words, whilst important, vary greatly. The choice of words is obviously important but the way in which language is used and the context are also important. People with dementia, for example, sometimes use vivid or "fighting/war" metaphors to convey their personal experience of dementia at a particular moment in time, but the same words and images used by other people risk being interpreted as reflecting the experience of all people with dementia (or people with other neurodegenerative diseases), reducing them as a group to sufferers or patients or portraying every aspect of their condition as a devastating natural catastrophe.

The guidance in this section is aimed at raising awareness of the need to communicate in an ethical and inclusive manner, which means paying attention to messages and images that we use and trying to ensure that they are not harmful (e.g. stigmatising, insulting or degrading) and that they both reflect and promote the inclusion of people with these conditions from all walks of life, including those from marginalised groups, in society. This is what we mean by respectful communication. The focus is on information and images, mainly in the context of research (e.g., in reports, participant-facing materials, deliverables and on website pages) and not one-to-one direct interactions (which we covered in Section 4). In the case of PD and parkinsonism, it is a disease (or syndrome) that is largely unknown and is associated with many myths and stereotypes. For example, many people think that PD only affects older men or that its only symptom is tremor. However, there are some people who are diagnosed before they are 50 years old and 30% of people with PD do not have a tremor. As a consequence, and as with other neurodegenerative diseases, it is common that the language or images used to describe PD and parkinsonism, and the people who have either of these conditions, do not reflect their reality, and therefore often reinforce these stereotypes. Some members of the GAP suggested that researchers would benefit greatly from opportunities to interact more with people with PD and parkinsonism in a relaxed manner so as to develop a more empathic and deeper understanding of their experience of living with this condition.

Guidelines

Avoidance of offensive or stigmatizing language

It is important to use terms that are not offensive or stigmatizing (please see explanation about stigma in Section 4) and that are respectful and inclusive when referring to people with neurodegenerative diseases and/or people providing support or care to people with neurodegenerative diseases. When writing or presenting information about people with neurodegenerative diseases and people providing care or support to them, avoid using terms that make some people feel uncomfortable. For example, in the case

of dementia, in many countries the term “demented” has very negative connotations and many people with dementia from different countries have publicly stated that this is a word they find offensive. Use terms that have more positive connotations, capture a sense of agency and reflect that many people with dementia (and other neurodegenerative diseases) are valued members of society.

There are several national guidelines which include terms to avoid and preferred terms. However, these should not be seen as check lists or recipes for respectful communication as there are many regional differences, as well as differences between language use in different cultural groups. Also, language is constantly evolving. Terms may not have the same connotations in every country and community. The term “dementia”, for example, is considered very negative in some countries or solely used to refer to people at a very advanced stage (e.g., in Finland where the term “memory disorder” is often preferred), whilst in others, it is the preferred or standard term. The context in which terms are used may also influence their perceived appropriateness. The term “patient” (as explained in Section 4) may be considered acceptable in the context of healthcare provision, but not when used in a generic way to refer to people with neurodegenerative diseases going about their daily lives. Members of the GAP felt strongly that researchers should adapt the language they use to the needs of the people with neurodegenerative diseases and their relatives and treat them with empathy, which clearly calls for sensitivity and good interpersonal skills.

Considering how terms and concepts might be interpreted

Researchers should be mindful of the terms and concepts used when communicating and how these might be interpreted by the general public. Terms should be used in a precise and clear way and when necessary with some background information or explanation.

As mentioned in Section 6, the conceptualization of Alzheimer’s disease (AD) has changed over the last few decades and the term “Alzheimer’s disease” is now used to refer to a continuum which includes preclinical (at-risk), prodromal and dementia stages.[7] Many lay people and people with dementia still think of the term Alzheimer’s disease as meaning a type of dementia and do not understand that the term can also be used to refer to pre-dementia stages. Similarly, there is some confusion surrounding the term “parkinsonism”. What professionals and researchers mean may be quite different to what lay people, including people with neurodegenerative diseases, understand. Examples of other complex terms are “risk” and “prevention” of dementia.

Reflecting on dignity, personhood, individuality and citizenship

When writing about or portraying people with neurodegenerative diseases remember that the disease is not a person’s whole identity. It is important to look beyond the disease to the person. John, for example, is not just a man with dementia; he is a retired builder, father of three and Manchester United football fan. Mercedes is not just a woman with PD but a university lecturer, a member of the local town council and an accomplished pianist.

Striving for a balanced/nuanced portrayal of neurodegenerative diseases.

Avoid generalising about the experience and impact of dementia, and other neurodegenerative diseases, as it affects different people in different ways. Focus on terms and positive images such as people with these conditions enjoying life, interacting with others or involving themselves in community, social and

[7] Please see Alzheimer Europe’s report on changing terminology surrounding AD. https://www.alzheimer-europe.org/resources/publications/2016-discussion-paper-ethical-issues-linked-changing-definitions/use-terms

political life (rather than solely focusing on wrinkled hands or on people looking lost, staring into space or trembling). Try to show how some people may be able to carry out their daily activities as they did before and have a sense of meaning in their lives despite the challenges of the condition. Of course, it is not helpful to portray people with neurodegenerative diseases in an overly positive way either. Neurodegenerative diseases can affect all areas of life. People and their experiences of these conditions are quite complex. Don't hide aspects of these conditions that people might find disturbing but put those aspects into perspective and context. Some people with neurodegenerative diseases are keen to emphasise that it is not all about loss.

> *"It is so important for me to emphasise that this disease is not only about loss. It brings change. We can put our remaining capacities to good use and learn new skills. That's where the focus should lie" (Person with dementia).*

> *"Communicate the seriousness of the disease, without denying reality. Emphasise the positive, with examples such as the effectiveness of therapies, advances in research, the importance of contributing towards research and overcoming difficulties" (Person with Parkinson's disease).*

Avoiding deliberately alarmist and frightening language and imagery

Think about whether your portrayal of neurodegenerative diseases reflects or perpetuates negative stereotypes, metaphors or clichés that you have read or heard about. Images of battles and fighting, which used to be common in the field of cancer, suggest courage and strength. They also suggest that there are not only winners but also losers. This also implies being unsuccessful or not having tried hard enough, potentially resulting in pity, and people with dementia feeling guilty and powerless. Referring to natural disasters such as plagues and tsunami may, for example, help capture the extent or scale of dementia within society but also focuses on the negative, implying that dementia is catastrophic and uncontrollable. References to time bombs, explosions and economic burden suggest that people with dementia are dangerous, a threat to society and using up valuable financial resources that could perhaps be better used elsewhere. This kind of terminology and related images are quite common in the press because they capture people's attention. It makes people sit up and take notice, but they come at a price as they are overly negative and lose sight of the individual.

People with PD and parkinsonism in the GAP felt that it is important that information about PD or parkinsonism, whether in the clinical or research context, is communicated in a realistic but positive way with examples of success stories about other people living with PD or parkinsonism. Ethical communication was understood as "honest, logical, positive, according to generally accepted standards, but always practical".

Questioning assumptions about neurodegenerative diseases.

Not everyone has encountered someone with a neurodegenerative disease. This applies especially to young researchers. Those who have may have very different experiences of it. Perhaps when they were a child, they visited a grandparent with dementia in a nursing home or they currently have a relative or friend with PD. However, most of us have heard about dementia on television or in magazines and books. We therefore have images, ideas, beliefs and even fears and concerns that lead to assumptions that may be reflected in what we write and how we portray dementia. It is important to think about this and consider whether, and if so how, you might be unwittingly communicating such assumptions (e.g. people with dementia all being old, in the later stages, passive and their main challenges being just linked to memory).

As mentioned above, there are many myths, stereotypes and misconceptions about PD, parkinsonism and other neurodegenerative diseases. This includes, for example, the belief that PD and dementia only affect older people, that the single and most important symptom of PD is tremor, that both conditions are hereditary and that people with these conditions are all severely cognitively impaired etc. Such misconceptions contribute towards stigmatization and social distancing. This includes self-isolation as people seek to avoid the looks, gestures and attitudes of a society that is totally unaware of their reality. This is one of the main reasons why global awareness campaigns such as World Parkinson's Day (11 April) and World Alzheimer's Day (21 September) are held to raise awareness of the reality of people affected by these conditions and help challenge negative stereotypes and misconceptions.

Avoiding portraying people as "other", fundamentally different or inhuman.

Avoid thinking in terms of "us" and "them". People with and without a neurodegenerative disease are all part of the same society that we live in. You and people you know may at some stage develop dementia or PD. You may have a friend or relative who has or had a neurodegenerative disease. Although such diagnoses can be life changing, people do not change overnight and become a different person or in some way less human. It may be important to specifically mention that people have a particular neurodegenerative disease but try to avoid implying that this makes people fundamentally different than people who don't have one, making negative stereotypes (e.g. people who have dementia or PD are, by definition, X, Y or Z) as this can be dehumanising, threaten their status as valued individuals and fuel stigmatisation. Certain metaphors that are used in everyday speech and visual portrayals, especially in relation to dementia, are particularly dehumanizing in that they reduce people to inanimate objects or monsters. Examples include an empty shell, a shadow, a zombie, a house in which no one is at home, a tree with the leaves blowing away or fallen or a jigsaw puzzle with pieces missing (Alzheimer Europe 2013).

Avoiding reducing people to numbers, objects, medical cases and problems.

Facts and figures about dementia or Parkinson's disease are useful in showing politicians, researchers and service providers that there is a need for services and support for people with those conditions and their carers/supporters, and more research about dementia (e.g. about care and support, but also about prevention and treatment). It is important, however, to avoid reducing people to mere numbers and transforming them into objects (e.g. medical cases or "problems" that need to be addressed). Behind every fact or figure, there is a real person with a unique experience and with individual needs, wishes, hopes, fears and relationships with other people.

Recognising and portraying diversity

Bear in mind that people with neurodegenerative diseases come from a wide range of sub-groups within society and from all walks of life. Neurodegenerative diseases affect people from all ethnic groups. They are not limited to "very old" people or to people who are either straight or from LGBTQ+ communities. Neurodegenerative diseases are not affected by a person's wealth, social position or where they live. Some groups of people are at higher risk than others but literally anyone can develop a neurodegenerative disease. Often, images of people with dementia in Europe show fairly old, White people, surrounded by children and grandchildren and looking fairly well off. This excludes so many people who have dementia and cannot relate to such images and may contribute towards their specific needs being overlooked when planning services or conducting research.

This is similar for people with PD and parkinsonism, as they are often depicted with images of white men or women who are older or in an advanced stage of the disease. This means that society is unaware of

other realities of the disease, such as the fact that it can affect young people who continue to work, or that some women are diagnosed while pregnant.

Seeking feedback from people with neurodegenerative diseases

When writing about or commenting on the experience of neurodegenerative diseases (e.g. in a project proposal for funding, for ethics approval or in a vignette) try to obtain some feedback from an Alzheimer or PD association and if possible from someone who actually has the condition. If the article is about a specific person, it is essential that they see what you have written and give their approval before it is printed. This also applies to any photos or images you wish to use as these can put a whole different slant on the issue and convey a powerful message that contradicts the content of the article (e.g. an article about something extraordinary or important that a person with dementia has achieved being accompanied by a photo of a forlorn-looking individual, staring into space, symbolizing pity, passivity and perhaps a life not worth living). The issue here is not just about accuracy but also about respect, dignity and trust.

Knowing your facts and figures, and putting them into perspective

It is your responsibility to do your homework and ensure that everything you write is correct. If unsure, check with an expert or leave it out. Facts and figures are often included as background information and as a context for the message being communicated. They are not, however, totally neutral because there are usually other facts or figures that could also have been presented but weren't, and for a good reason (e.g. because they do not capture attention in the same way or they show another side of the story that the authors are less interested in showing). Many people do not fully understand statistics and can easily draw false conclusions such as having a high risk of getting a neurodegenerative disease even though their risk is actually very low. For this reason, when presenting statistics about specific risk factors (e.g. of smoking or obesity) provide information about both the relative risk and the absolute risk. When providing facts and figures, put them into context or perspective so that people can make sense of them and see how they relate to them and their lives.

Challenging inaccurate, disrespectful or misleading portrayals of neurodegenerative diseases

You may come across materials, reports and articles produced by colleagues or others in your field of work that portray neurodegenerative diseases inaccurately, are disrespectful to people who have them or are misleading. Have the courage to point this out. The more it is challenged, the less likely it is to be perpetuated.

There are frequently misleading or deliberately ambiguous accounts and headlines in the media such as "Miracle drug halts process of Alzheimer's disease" or "A handful of blueberries a day could keep dementia away". Such headlines certainly attract attention, and some might think they bolster support for research by showing how beneficial and vital it is to society. However, they are unethical in that they are misleading, inaccurate and falsely raise the hopes of people with neurodegenerative diseases. Even if the reports go on to explain the limitations of a particular study, such claims are potentially damaging to people's wellbeing. They may also undermine trust in research and in the medical profession. As researchers, it is not always possible to prevent this kind of sensationalist reporting of research results by the media but it is important to challenge it or correct it whenever possible.

Section 6: Plain and accessible terminology

Introduction

Members of the public (who might one day consider sharing their data or taking part in research) are exposed to a great deal of information about research topics (e.g. about preventive factors, artificial intelligence, big data and drugs being developed). Being informed is closely linked to respect for autonomy and does not begin when a research project starts or when a database is launched. It occurs whenever people hear about neurodegenerative diseases, research in general, data sharing platforms or hopes, expectations and breakthroughs linked to prevention, treatment or care.

Some of the information that members of the general public read was not targeted at them but is accessible to them (particularly on the Internet but also in the media). This will probably increase in the future as more and more research is published on an Open Access basis, as members of the public continue to take a more active role in healthcare decision making and as more people become computer literate. This calls for greater support, especially for people with neurodegenerative diseases, in the form of supported and substituted decision making. Please see Alzheimer Europe's report and guidelines on this topic[8].

Attempts to raise awareness amongst the general public about research, about how they can contribute towards it and what doing so involves must be accompanied by efforts to ensure that such information is understandable to lay people. Research-related terminology can be fairly technical and include terms that are not used in everyday life (e.g. in relation neurodegenerative diseases, the development of platforms for data and sample sharing, data, research methods, ethics, law and inclusion). In this first section, we:

- look at key terms linked to dementia (particularly Alzheimer's disease) and PD,
- describe some of the recent changes and potential confusion surrounding certain terms
- provide some recommendation on the use of such terms in the research context.

Neurodegenerative diseases

A neurodegenerative disease is an umbrella term for a range of conditions primarily affecting the neurons in the human brain, which are incurable and caused by the progressive loss of the structure or function of neurons. This process is known as neurodegeneration and may lead to loss cognitive ability and ultimately cell death. Examples of neurodegenerative diseases include multiple sclerosis, Parkinson's disease, Alzheimer's disease, Huntington's disease, multiple system atrophy, and prion diseases.

In the course of the EPND project the PEG will explore lay people's understanding of this term and the possible implications that this may have for the willingness of people with various neurodegenerative conditions such as dementia and PD to share their data on platforms like the one currently being developed.

Parkinson's disease and parkinsonism

Researchers use a multitude of different terms to talk to other researchers, to people affected by PD, to the general public and to clinicians about PD and parkinsonism. These include, for example:

[8] Alzheimer Europe (2020). *Legal capacity and decision making: The ethical implications of lack of legal capacity on the lives of people with dementia*. Alzheimer Europe

- Idiopathic Parkinson's disease or Parkinson's disease, probable or possible
- Parkinson's Plus
- Parkinsonism
- Atypical or plus parkinsonism
- Early Onset or Advanced Parkinson's disease

Often, the medical and scientific terms that refer to what is commonly known as "Parkinson's disease" or "Parkinson's" are not used accurately or are not understandable to many lay people (including people who have PD or parkinsonism). This is especially the case with regard to the medical terms for various symptoms such as dyskinesias, bradykinesia and dysarthria, which are just not part of everyday language. References to specific stages of the disease can also be confusing to some lay people. People with PD often have no knowledge of the scale(s) used by researchers and neurologists to describe or assess the stages or phases of PD and parkinsonism (e.g. advanced stage or early onset). One person, for example, expressed her confusion about how the term related to her own condition, stating, "Stage 2 of how many?" Although, the Michael J Fox Foundation[9] recently reported the discovery of a new biomarker, there are currently no diagnostic tests for PD. At the moment, there is just a clinical diagnosis based on the observation of symptoms. Neurologists may perform certain tests and use neuroimaging techniques, but these are just to confirm that the symptoms are not caused by another issue. People are therefore sometimes diagnosed with different “things” (like “parkinsonism” or something else) before getting a diagnosis of PD. Members of the GAP felt that the ambiguity in the context of diagnoses of PD and parkinsonism, as well as difficulties understanding terms related to symptoms, could undermine trust in clinicians, which might also be transferred to the research setting.

Failure to explain specialized medical terminology to lay people who are unlikely to be familiar with such terms, whether in the research or clinical setting (i.e. in the context of PI work, informed consent forms to participate in research or diagnosis and treatment) is clearly disrespectful because it reflects a lack of concern for or interest in their wellbeing and right to understand work that they are contributing to out of good will or issues that are relevant to their health and lives. Such practices also reflect a lack of empathy and ability or effort to adapt to the needs of the person with the condition. A clear and simple explanation of terms is therefore important when they are used in the medical and research contexts. This is one of the reasons why we are developing a glossary of research-related terms which will be reported in Deliverable 6.6.

Dementia

Dementia is not a normal part of the ageing process. Nor is dementia a single disease. Rather, dementia is an umbrella term used to describe the loss of memory and thinking ability that is caused by different diseases which damage the brain. Dementia is the leading cause of disability and dependency in older adults, affecting almost 8 million people in the European Union. However, every person with dementia is unique. Dementia affects people in many different ways, depending on the type of dementia they have as well as personal factors such as their social situation and personality.

There are many different types of dementia, and they are all progressive and life-limiting. Dementia symptoms can vary widely from person to person and evolve over time. Typical symptoms of dementia

[9] https://www.michaeljfox.org/news/breaking-news-parkinsons-disease-biomarker-found?pn_cid=pn-a1b1R00000AlQ16

include memory loss and disorientation, as well as problems with thinking, mood and practical activities in daily life. These symptoms are usually relatively mild in the early stages but gradually get worse as dementia progresses. Reactions to the term (could mention France and Finland as examples), a tendency sometimes to use the term to refer solely to AD dementia (as if it were the only form).

AD-related terminology

A key problem with terminology surrounding Alzheimer's disease is that it is often used inconsistently, at times to refer to a form of dementia and at other times to refer to the continuum covering both pre- and post-dementia stages. This lack of precision means that members of the general public may be exposed to potentially conflicting messages from different reliable sources, which in turn may cause distress and undermine trust in healthcare professionals and researchers. This potential issue occurs against the backdrop of confusion amongst many members of the general public about the difference between Alzheimer's disease and dementia and changes in the conceptualization of Alzheimer's disease.

Early AD terminology

In 1906, Dr Alois Alzheimer first described the symptoms and the amyloid plaques and neurobrillary tangles in the brain, which have come to be considered as the hallmarks of Alzheimer's disease (AD). Now, more than a hundred years later, the exact causes of AD are still unknown and a cure is not available. However, significant progress has been made in understanding AD and especially in biomarker research. Signs of abnormal changes in the brain associated with AD can now be detected long before the occurrence of any symptoms of AD dementia. These recent advances have contributed towards a radical change in the way that AD is conceptualised.

In 1984, the "NINCDS-ADRDA" criteria for AD were proposed (McKhann 1984), which permitted a probable clinical diagnosis of AD that could only be confirmed as definite after the person's death when the characteristic plaques and tangles could be observed. This meant a long period of uncertainty and the criteria had a fairly low accuracy rate (Hyman & Trojanowski 1997; Beach et al. 2012) with only 70% of diagnoses being correct, the others being false positive or false negative cases.

- ➔ A key feature of early AD-related terminology was that the term Alzheimer's disease was synonymous with Alzheimer's dementia. In other words, when a person was diagnosed with Alzheimer's disease, this was understood as being a diagnosis of AD dementia. Many people still use the term AD solely to refer to people who have AD dementia even though the meaning of the term has since become much broader.

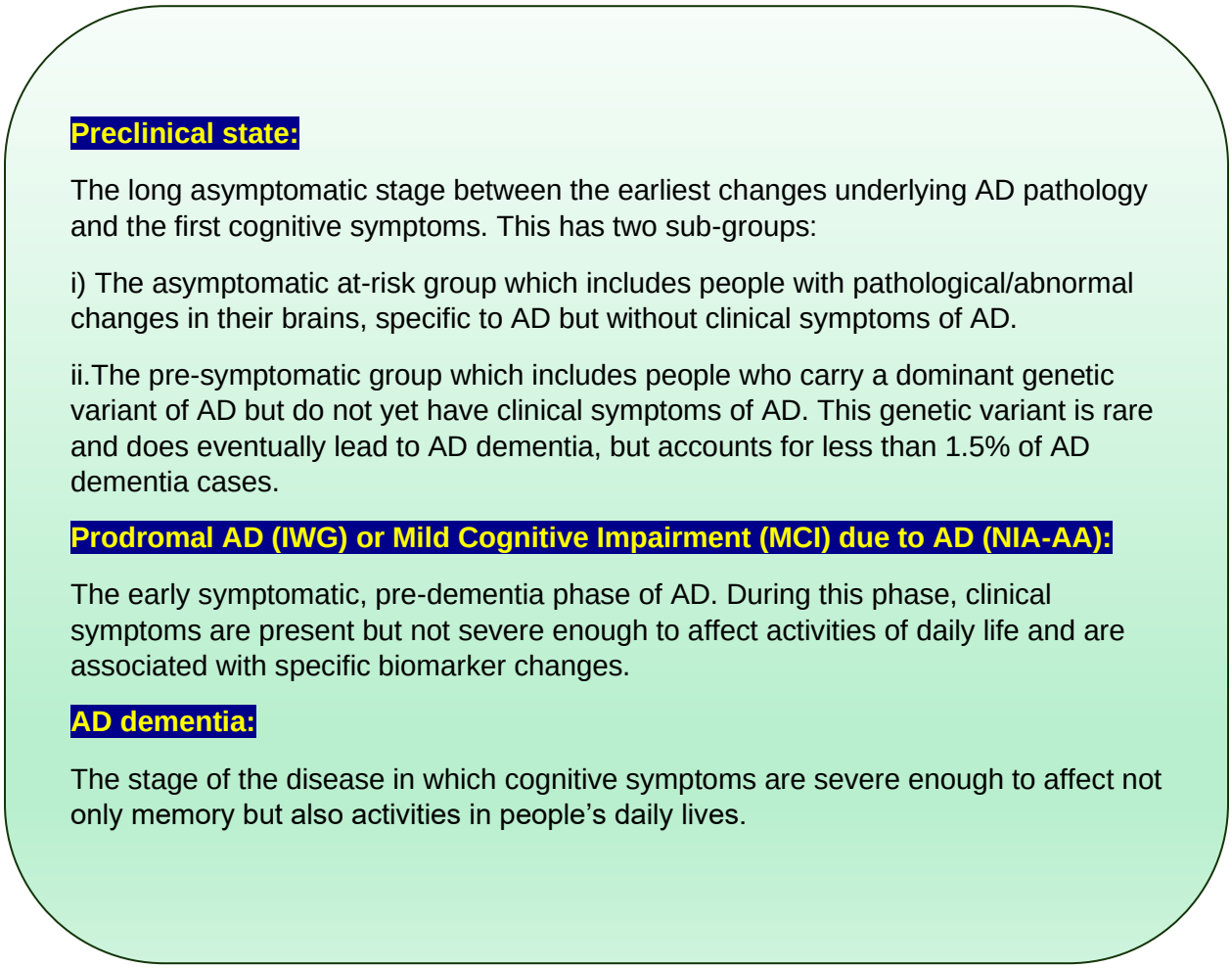
Recent AD terminology – focus on biomarkers and a continuum

A radical reconceptualization of AD occurred from roughly 2006 onwards with the publication of work by the International Working Group (IWG and IWG2) (Dubois et al. 2007, 2014 & 2016) and the National Institute on Aging and the Alzheimer's Association (NIA-AA) (Jack et al., McKhann et al., Albert et al. and Sperling et al., all in 2011). Both groups agreed on the importance of biomarkers in the procedure leading to a potential diagnosis of AD dementia, enabling an *in vivo* diagnosis (i.e. in a living person).

The definition of AD has now been extended to encompass the full spectrum of the disease, including both pre-dementia (preclinical and prodromal AD or MCI due to AD) and dementia phases (Dubois et al. 2010 and 2016). As can be seen in Figure 1 below, the pre-dementia phase includes two preclinical classifications - one being an asymptomatic/at-risk group (for which not all members will eventually develop AD dementia) and the other a pre-symptomatic group (for which all members will eventually go

on to develop AD dementia). Figure 1 also shows two different terms for the next stage preceding dementia which is called prodromal AD by one group and MCI due to AD by the other group.

Figure 1: Combined reconceptualization of Alzheimer's disease



Source: Alzheimer Europe (2016, p. 7)

The different components or stages of AD are considered as occurring along a continuum. Aisen et al. (2017, p.2) describe the AD continuum as follows:

> *"Based on currently available information, AD is best conceptualized as a biological and clinical continuum covering both the preclinical (clinically asymptomatic individuals with evidence of AD pathology) and clinical (symptomatic) phases of AD. In the broadest sense, a continuum is defined as a seamless sequence in which adjacent elements (severities) are not perceptibly different from each other, although the extremes are distinct. In AD, this equates to disease progression from an asymptomatic phase, through a long preclinical period during which pathophysiological changes are reflected by increasing biomarker evidence of disease, to the symptomatic phase, during which biomarker changes continue and symptoms of cognitive and then functional impairment become*

increasingly evident, with the eventual loss of independence and death. These changes in the individual components of the continuum occur in a sequential but overlapping manner."

➔ A key feature of the recent conceptualizations of AD is that people are described as having AD both before and after developing dementia because the term covers the different stages of the whole continuum. People who are not aware of this new extended meaning of the term may misunderstand and think that they or other people have AD dementia when this is not the case.

Mild Cognitive Impairment (due to AD)

Prior to a diagnosis of AD dementia, people with cognitive impairment but with limited impact on their functioning are typically diagnosed with Mild Cognitive Impairment (MCI) (Petersen 2004). Until recently, MCI has been considered as a condition which does not necessarily lead to dementia, with many people reverting back to normal cognitive functioning or remaining stable over time (Petersen et al. 2014). In the new conceptualisation of AD, however, a specific sub-category of MCI, known as MCI due to AD (the equivalent of prodromal AD – see Figure 1) is considered as a stage of AD. According to Dubois et al.:

> *"The proposed conceptual shift is to consider a patient previously diagnosed as having MCI (i.e. with an amnestic syndrome of the hippocampal type and with biomarker evidence positive for brain amyloidosis[10]) to be no longer at risk for developing AD dementia, but to recognise that they already have AD at a prodromal stage with an inevitable progression to AD dementia over time." (Dubois et al. 2010, p.1123)*

The inevitable progression to AD dementia mentioned above is an important piece of information, but is limited to this particular sub-category of MCI (not to MCI in general). There are other forms of MCI that do not inevitably lead to AD dementia or to another form of dementia. Some people with MCI revert back to normal cognitive functioning or simply do not progress to a dementia stage. Lay people may find some of the new terms confusing. "MCI due to AD", for example, could be correctly interpreted as referring to a form of MCI that is due to AD or as implying that MCI (i.e. in general) is due to AD. Similarly, "prodromal" is not an everyday term and most people will not know what it means. Sometimes, the term "MCI/prodromal AD" is used and it is not clear whether the "AD" is linked solely to prodromal or also to MCI (Jessen et al. 2014) and bearing in mind that AD continues to be described in numerous publications as a form of dementia.

➔ A key problem with references to MCI is that researchers do not always specify whether they are referring to MCI in general (covering all different forms and causes) or to the specific sub-category that is part of the AD continuum described earlier.

➔ Some researchers and clinicians describe or frame diagnoses of prodromal AD (IWG) or MCI due to AD (NIA-AA) as a very mild form of AD dementia. This can be confusing and contradicts the portrayal of this form of MCI as a pre-dementia stage.

Recommendations surrounding Parkinson's, AD and/or dementia-related terminology

The lack of attention to detail frequently encountered in the context of research is a problem that must be addressed. Failure to do so is unethical as people have a right to be properly informed about the nature of research that they are contributing towards (through PI) or participating in (as research participants). It is disrespectful towards them to consider potential misunderstanding and confusion as

[10] This is a sub-category of MCI

acceptable, and such misunderstandings may also negatively impact on the quality of the feedback that they provide in the context of PI.

1. When writing or speaking about Parkinson's, AD, dementia and other neurodegenerative conditions, researchers should at all times strive for accurate and consistent use of terminology (even when not directly targeted at the public, patients or research participants).
2. For communications targeted directly at the general public, patients, research participants and people involved in PI work, researchers should:
    a. try to ensure that all medical and technical language used is clearly explained in everyday language
    b. try to ensure that it is clear whether the term AD is being used to refer to the dementia stage or the whole AD continuum/disease pathology
    c. try to ensure that it is clear whether the term MCI is being used to refer to MCI in general or specifically to prodromal AD/MCI due to AD
    d. try to ensure that the difference between Parkinson's and parkinsonism, and any medical terms for symptoms and stages, are clearly explained in everyday language
    e. try to ensure that it is clear what a particular term (e.g. a stage or phase of the condition) means in relation to typical progression and overall stages or phases of the condition.

Section 7: Conclusion

PI is about involving people with lived experience in the design, conduct and dissemination of research; it can make research more meaningful and ethical. EPND is a platform that will connect and bring together researchers from different disciplines, sectors and parts of the world. In Part 2 of this deliverable, drawing on consultations with people with neurodegenerative diseases, we have tried to provide constructive feedback, practical guidance and background information to motivate and enable researchers to communicate respectfully and collaborate meaningfully with people with various neurodegenerative diseases. Much of what we have discussed (e.g. linked to clear language, respectful communication and valuing people) might equally apply to a broad range of people doing PI work. However, it is particularly important in the case of people with neurodegenerative diseases whose difficulties with cognition and possible sensory or physical impairments make them more dependent on others to create the right conditions and atmosphere for such collaboration.

Researchers should therefore reflect on the possible needs and wishes of people with neurodegenerative diseases doing or contemplating doing PI work and ensure that they make reasonable accommodations if and where needed. This will help create a PI process that is more feasible and meaningful which people with neurodegenerative diseases will feel they can realistically contribute towards.

It is equally important that researchers feel equipped and motivated, rather than hesitant or daunted, by the prospect of working together with people with neurodegenerative diseases for the benefit of their research and in turn for the benefit of society.

The information and guidance provided in Part 2 of this deliverable will hopefully be useful but should just be seen as a starting point for a constructive, mutually rewarding collaboration with people with neurodegenerative diseases in the context of research into these conditions.

Section 8: Useful resources

Dementia

Alzheimer Europe (2013). *Perceptions and portrayal of dementia and people with dementia*. Alzheimer Europe

Alzheimer Europe (2018). *The development of intercultural care and support for people with dementia from minority ethnic groups.* Alzheimer Europe

Alzheimer Europe (2019). *Overcoming ethical challenges affecting the involvement of people with dementia in research: recognising diversity and promoting inclusive research*. Alzheimer Europe

Alzheimer Europe (2020). *Legal capacity and decision making: The ethical implications of lack of legal capacity on the lives of people with dementia*. Alzheimer Europe

Alzheimer Europe (2021). *Sex, gender and sexuality in the context of dementia: a discussion paper*. Alzheimer Europe

Alzheimer Europe (2022). *Guidelines for the ethical and inclusive communication about/portrayal of dementia and people with dementia (for the media, researchers, journalists, policy makers and anyone responsible for the portrayal of or communication about dementia)*. Alzheimer Europe

Alzheimer's Society (2018). *Positive language: An Alzheimer's Society guide to talking about dementia.* Positive language guide_0.pdf (alzheimers.org.uk)

Alzheimer's Society of Canada (2017). *Person-centred Language Guidelines.* Person-centred-language-guidelines_Alzheimer-Society.pdf

DEEP (2014). *Dementia words matter: Guidelines on language about dementia.* DEEP-Guide-Language.pdf (dementiavoices.org.uk)

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The Alzheimer's Society of Ireland (2018). *Dementia Friendly Language*. Dementia-Friendly-Language.pdf (alzheimer.ie)

General

Jargon buster: https://www.invo.org.uk/wp-content/uploads/2011/12/PIP44jargonbuster.pdf

Plain English campaign: https://www.plainenglish.co.uk/free-guides.html

Plain language summaries (2021): https://www.nihr.ac.uk/documents/plain-english-summaries/27363

Parkinson's disease

Parkinson's UK

- Supporting your research through involvement and participation:
- Toolkit: clear and simple communication framework to help researchers share updates with participants
- Patient and Public Involvement in research
- Race Equality in Research
- Improving life through research
- Resources for Professionals
- Clinical tools and assessments
- Guidelines
- Research resources for professionals

Parkinson's Foundation

- Women and Parkinson's Research and Care Agenda
- For Researchers

The Michael J. Fox Foundation for Parkinson's Research

- Webinars for Researchers
- Research Tools
- Data Resources
- Study Recruitment

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