



# Reframing QoL assessment in persons with neurodevelopmental disorders

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## Abstract

The paper reviews the international literature on quality of life (QoL) for persons with neurodevelopmental disorders (NDD) in order to define the theoretical frame for optimal assessment. The application of the QoL approach to assessment procedures should be based on three main aspects: shared QoL, personal QoL and family QoL. The first aspect refers to characteristics of individual life that are shared with other people. The second aspect proceeds from the fact that each individual has a changing set of personal attributes that determine the subjective experience of life. In the third aspect the previous two are applied to the family that includes a person with NDD. Disability impacts the whole family and the determination of appropriate conceptualization of family outcomes requires an understanding of the impact of members with a disability on family QoL. At any level, it seems best to take a comprehensive approach to assessing QoL, integrating subjective and objective aspects, self-reports and hetero-evaluations. The QoL approach is above all a way to explore the rich intricacies of personal quality of life. Such assessment may be used effectively with people with NDD, independently from the severity of their functioning impairment. Individuals with profound ID may express their inner states through consistent behavioural repertoires, which can be discerned by persons closest to them and validated by more independent others. Attention must be paid in using non-generic instruments, such as those that measure health-related QoL. Although they do focus on the individual person, they still support a theoretical perspective of QoL that has not departed significantly from the traditional medical approach. Currently available generic tools, although they have some common conceptual and evaluation characteristics, still show considerable differences in the areas to be included in “shared QoL”, the dimensions used to evaluate “Individual QoL”, and the role attributed to indicators of QoL. QoL assessment should not represent a classification of individuals, services or systems, but it should help provide, within service systems and organizations, a value system that is consistent with those values held by people with NDD.

## Key words

- quality of life
- intellectual disability
- autism-spectrum
- assessment
- measurement

## INTRODUCTION

Person-centred care originated in the area of disability and is now widely used within the areas of mental health, aged care services, education, general health-care, and the criminal justice system. Its focus within disability support organisations is to ensure that services provided concentrate on what matters most to the people receiving them and their families. Person-centred approaches ensure that service users are seen as unique individuals with valued strengths and contributions [1].

Among person-centred approaches, Quality of Life (QoL) has raised particular interest in the scientific community across the last 30 years. Hundreds of research and conceptual articles have been published,

and numerous books have focused directly on QoL. At present, the QoL concept seems to be at the crossroads of all intervention strategies in the various branches of social care and medicine, especially in psychiatry.

The meaning of the term QoL may appear easy to understand and, in its general sense, it is. But because it is so frequently used to refer to the “goodness” of life only in general, it runs the risk of becoming a banality. When we dig deeper and apply the term QoL to real life, it can take on multiple meanings, so much so that it is almost impossible to talk about it without a clarification of contextualized its meaning. When QoL is used in the public media, especially for promoting new ideas or products, it typically refers quite clearly to high level



of life quality or the “best” of life. This popularization of the term QoL has resulted in it being understood and used in ways that are considerably less substantial than the conceptualizations that have been described and applied by academics and researchers. For this reason, it is sometimes considered that QoL is a concept that lacks sufficient precision to have specific usefulness in the field of intellectual disability (ID), and that it is more advantageous to focus on clearer concepts or simply on providing practical supports.

Yet, a considerable amount of substantive work has been done in the area of QoL and ID. Numerous thoughtful books, book chapters, and academic articles have been published that not only advanced our understanding of QoL, but have made ID the leading field for substantive work in QoL. In the last ten years, the research activity in the area of QoL and ID has been extended to autism spectrum disorder (ASD), especially to persons with low functioning. ID and Low-Functioning ASD (LF-ASD) often co-occur and are difficult to distinguish, especially when cognitive impairment is more severe [2].

It is clear from this work that achieving high quality of living is not the main purpose. Rather, it provides an approach for how professionals view the patient-person, and for how they view the relationship between the patient-person and the system of care. The QoL approach has to be interpreted in qualitative rather than quantitative terms, as it consists more in choosing a course than in achieving a goal. It provides both a viable and an alternative approach to the traditional medical approach. In fact, it tries, overall, to help people feel satisfied with their own lives in ways that are customary to them and valued by them, while the latter focuses primarily on restoring morphological and functional integrity to aspects of the person that have been affected by the morbid condition. The QoL approach begins by professionals understanding what is important to and valued by each individual, and what aspects of life or the environment contribute positively to life quality or detract from it. This approach makes every effort to respect the right of the individual to help choose the course of action that best suits him or her, and endeavours to provide whatever positive supports are required to assist the individual to live an effective life that is uniquely shaped by individual characteristics and circumstances [3]. One of the fundamental principles of the QoL approach is that QoL is important for all people, and that all people are thus entitled to a life of quality [4]. This principle applies equally to people with ID and to people who do not have ID.

The question of how we can ensure entitlement to a life of quality for people with ID who have mental health problems is an interesting and challenging one. Yet, the emphasis within the conceptualization of QoL on finding uniquely individual solutions to presenting problems suggests that it may be a particularly apt approach to take with this population.

People with ID are characterized by lower levels of overall intelligence, and lower potential for learning and developing logical-deductive skills. For this reason, it is not a realistic goal for therapeutical interventions

to attempt to provide “normal” intellectual functioning, but rather such interventions need to work within the confines of each individual’s adaptive functioning, which, by definition, shows deficits that significantly hamper for the individual’s independence and ability to meet their social responsibility [5]. Similarly, it is not a reasonable therapeutic goal to have people with ID live their lives as close as possible to people without ID, because ultimately they have to find satisfaction in those aspects of life that are important to them and that they can understand and experience. The more traditional medical approach to therapeutic interventions with people who have ID and mental health problems have tended not to consider the uniqueness of people with ID fully and have been more likely to use the general population norm as their point of reference. This has resulted in interventions that have dealt primarily with containing troublesome behaviours and helping to alleviate symptoms, rather than to address the root causes of the problems. Thus, assuming an overall QoL approach to intervention goes a long way to ensuring entitlement to a life of quality.

A second step to begin to ensure a life of quality is to promote changes in the way mental health professionals perceive people with ID through professional training. Too often, such professionals still perceive people with ID as having intellectual and skill incompetence that is so different from “normal” functioning that the intelligence and skills they do possess are not valued or seen as possible to enhance. As a result, intervention very often focuses on simply changing behaviour so that it is more manageable and less troublesome to the individuals and to those around them. However, it has been recognized for a number of years that people with ID are considerably over-represented among those who present with mental health problems, and this may be increasing in recent years due to people with ID living much longer than was the case in the past. At the same time, it is increasingly recognized that a number of therapeutic interventions are effective with this population, and that they have positive impact on the lives of individuals. These changes are bringing new challenges to mental health services, and they are suggesting the use of positive interventions that view the health – and indeed the whole life – of people with ID in more holistic and integrated ways. The QoL approach is ideally suited to respond to the demands of such changes.

In recent years, scientific interest has already moved from theoretical considerations to measurement methods. The reasons are manifold, but the main ones seem to relate primarily to the growing use of QoL measurement as a crucial aspect of the individualized evaluation and programming, as a comprehensive outcome measure, and as a key to access the application and dissemination of the concept [6-8]. There is also growing support for evidence-based practice, that is, the view that there should be clear and reliable evidence from research that our service practices are effective [9, 10]. In the QoL context, evidence-based practice infers that there should be clear evidence that our practices contribute positively to a person’s QoL. The focus on evidence-based practice is placing additional impor-



tance on the QoL measures that have been developed since about 1990, and it is encouraging their refinement. Some authors suggested that abandoning highly complex models in favour of simpler and empirically testable propositions could represent the most effective way to support this progression [11]. Already by 2002 the International Association for the Scientific Study of Intellectual and Developmental Disabilities (IAS-SIDD) argued that QoL should represent the reference frame of all therapeutic and rehabilitative interventions and assume a prominent place in education and training targeted at professionals [12].

Despite its widespread implementation, QoL measurement is still associated with numerous problems, related to the definition of the QoL concept, the way it can be effectively measured, for whom, by whom, and to what purpose. Evaluation tools produced to date are numerous and the fields of application vary, from social support to health, employment, statistics or economics.

### **Study Objective**

The aim of this study was to investigate the state of knowledge and current issues in QoL assessment for people with ID and/or LF-ASD, through a systematic mapping of the scientific literature.

### **METHODS**

A systematic mapping (SM) of the literature differs from a systematic review (SR) by the breadth of the research topic, the type of questions posed, and the precision of the extracted data [13]. In SM research, topic and data extraction activity are broader than in SR, and the analysis does not include depth techniques, but rather summaries.

The reference questions for the present mapping were: 1) "What is the purpose of the QoL measures available so far?"; 2) "What is actually measured by QoL measures for persons with ID/LF-ASD?"; 3) "For which persons with ID/ASD is measurement possible?" and 4) "By whom should a QoL measurement be made?".

The authors examined the international literature of the past 23 years, from January 1996 to March 2019. The article search was carried out using the engines of Medline, Medmatrix, NHS Evidence, and Cochrane Library. The keywords used were: "quality of life", "subjective well-being" or "person-centered outcome (measures)", combined with "assessment" or "measure (ment)", further combined with "intellectual disability" (or "learning disability", or "mental retardation"), and/or ["autism" (or autism spectrum disorder) and "low functioning"].

Articles identified by this procedure were integrated with those previously collected by the authors of the present paper through direct contacts with members of the international scientific community.

Articles published only in languages other than English and Italian have been excluded. Further additional papers were identified by screening the bibliographic references included in the articles already selected, and by searching the world wide web through using the above mentioned keywords in two of the main generalist search engines, namely Google and Yahoo, and in

two of the main directories, namely Dmoz and Digital Librarian.

Articles and chapters that matched the above listed keywords were screened by titles and abstracts, before being read in full.

### **RESULTS**

The search through Medline, Medmatrix, NHS Evidence and Cochrane Library identified 1536 articles. After title and abstract reading, 221 were selected as relevant. Of these, only 52 were judged to be useful for answering the mapping reference questions.

The search through the generalist engines of the world wide web identified 184 links of potential interest. After reading the full web pages, 24 were found to be relevant. After reading all the documents contained therein, including books and book chapters, and after excluding overlaps with the texts already identified through the engines for scientific literature, only 13 were considered useful to answer the questions of the mapping.

### **Purpose of QoL measures**

QoL measures can be grouped as referring to three main aspects: shared QoL, personal QoL, and family QoL.

#### *Shared QoL*

Much of the QoL work that has emerged in the field of ID/LF-ASD and elsewhere is based on characteristics of human life and humans' environments that are common to, or shared with, other people. For example, humans are social beings by nature and thus the support of other people, especially family, friends, and co-workers, is typically a relevant aspect of life to all people. In some QoL theoretical models and associated measures [14, 15], aspects of QoL based on shared human characteristics are confounded or overlapped with human rights, which are represented by a set of rights and freedoms that belong to every person in the world, from birth until death. However, shared QoL and human rights represent two different concepts, although they are interconnected. Rights are based on shared values like dignity, fairness, equality, respect and independence, while shared QoL refers to areas of life that have qualitative implications for everybody's life. Rights as well as life itself can be considered as presuppositions or antecedents of QoL and not as aspects of shared QoL.

Shared QoL is relatively easy to assess, even in persons with severe ID/LF-ASD, although no consensus has been reached yet within the scientific community on exactly what aspects should be assessed. This assessment is appropriate for groups of people, for services and supports that address the needs of large numbers of individuals, and for making policies that affect populations.

#### *Personal QoL*

In QoL assessment, it is also widely recognized that there are individual differences, and that sometimes these are quite substantial. First, for individuals, shared characteristics take on increased or decreased personal

value in response to personal genetic makeup, individual personality, or personal environmental conditions. Second, all individuals have unique characteristics and interests that are sometimes very meaningful in their lives, and thus add to their life quality, but mean little or nothing to another person [16]. For example, a musician may spend hours every day practising her instrument, performing, and seeking out new music and new music-related opportunities. Most of her friends and close acquaintances may be musicians. Music is a dominant and driving force in her life, and one that is highly valued and gives strong meaning and direction to her life. Yet to her neighbour, who knows almost nothing about music and does not care to learn or even listen to it, music is not valued and it means almost nothing to him. Unique talents and interests need to be central to quality of life evaluation, as they can sometimes be central to a person's sense of self and enjoyment of life. Third, QoL is a personal concept in that it is, to a high degree, based on a set of personal perceptions, attitudes, values, and skills, and on the physical and social environment. These factors determine not only how a person's life is experienced, but also how it is interpreted. Finally, the changing nature of QoL has been described for many years (e.g., Raphael, Brown, Renwick, and Rootman [17]), but Reinders [18] made an important elaboration: "...the concept of QoL is necessarily open-ended. There is on any account of the matter a dimension of incompleteness in assessments of QoL in the sense that we may discover things about ourselves that in due time will change our judgments. Precisely as a comprehensive concept QoL must entail an element of the unknown future of our existence".

Good QoL assessment, then, allows for the concept to be explored at a personal level as well as at the shared level [3]. When QoL assessment is used to improve life quality at the individual level, it is essential to place a strong emphasis on personal QoL.

#### *Family QoL*

Due to policy changes in most developed countries, families that include a person with ID/LF-ASD are increasingly being required to accept a larger burden or responsibility than was the case in the past, not always with entitlement to services [4]. As a result, families are often overburdened and marginalized in society, and they often need help, or support, to achieve lives of quality [19]. Disability impacts the whole family [20] and the determination of appropriate conceptualization of family outcomes requires an understanding of the impact of members with a disability on family QoL. According to Summers and collaborators [21], this involves three main issues that research has explored in recent years: stress and caregiving burden, impact on family functioning, and eco-cultural adaptation.

QoL assessment should guide personal, service, or policy enhancement rather than representing a classification of individuals, services or systems [8].

It should help provide, within service systems and organizations, a value system that is consistent with those values held by people with ID. The *Ask Me! Project* in Maryland, USA, is an example of one of many organi-

zations that now use QoL values and information can usefully direct services to develop goals and to monitor achievements. The project puts particular emphasis on predictors of personal development, relationships between QoL domains, and consistency with mission statement [22].

At the front-line staff level, QoL can represent an integral and multidimensional view of the health of the person with ID that allows the multidisciplinary team to identify needs and wishes, and to plan interventions in the most useful way. In Baum's opinion, this is a particularly useful approach in managing challenging behaviours [23].

Generally, a QoL approach should identify the degree to which individuals attribute importance to various areas of their lives, and the degree to which they perceive satisfaction. In doing so, global QoL measures will be increased because we give more strength to those aspects of life that are more important and less strength to those aspects that are of lesser importance.

The goal here is to assess QoL with a view to more accurately reflect overall life satisfaction in the life areas that are deemed to be most important. The QoL approach should also be seen as an effort to mobilize and revalue resources that can help individuals (and the holistic systems that they represent) to embark on or to continue developing personal skills over their lifespans.

#### ***What current QoL measures actually measure***

Measuring QoL has been somewhat challenging, especially because it is a social construct (a concept based on ideas that do not have a physical presence), and thus evidence for or against it must be inferred from indicators that of our own construction. Achieving consensus on what the best indicators of QoL are for people with ID/ASD has been a goal of the QoL and ID work (see especially Schalock and colleagues[12]), but a healthy debate continues. There seems to be little doubt that conceptual and measurement knowledge will be enhanced as more of our professional work with individuals takes a QoL approach. While attempting to help solve their real-life problems, we will become better able to understand the essence of their QoL, and how it can be most realistically and effectively measured.

Since QoL is a somewhat complex concept involving the whole life of the person, most theorists and researchers agree that QoL can be best measured by obtaining and combining scores from several life domains. The point to be made here is that precise units of measurement based on a clear, concise definition are not currently available, and, as is the case with other complex social constructs (e.g., health, learning), such measurement may not be available in the future. But there some well-developed measures for QoL for people with ID/LF-ASD that share some agreed-upon structural elements [24].

#### ***Elements of QoL measurement***

There is high agreement that QoL measurement should be based on both qualitative and quantitative information from both subjective and objective perspectives [11, 6], with some flexibility depending on



the purpose of the measurement. Perry and Felce [25] explored the relationship between objective and subjective measures of three of the most frequently assessed QoL outcomes in British deinstitutionalisation research: choice, participation in activities, and community and social integration. This research was conducted within a randomly-selected sample of small community housing services. The results showed that residents' satisfaction with choice, as measured by ComQoL [26] did not correlate with any objective measures. Satisfaction with friends, free time, recreation, and leisure scores of the *Lifestyle Satisfaction Survey* [27] were significantly correlated with engagement in non-social activity but not with participation in domestic life. Again, within the area of social and community affiliation, none of the nine correlations between objective and subjective measures was significant. The study was not designed to give participants an opportunity to rate the degree to which they considered each indicator important, and thus the critical relationship between *importance* to the person and *satisfaction*, often considered a key element of accurate QoL measurement, could not be explored. Nevertheless, the results support previous findings that assessment of objective life conditions and perceptions of personal satisfaction appear to be distinct. It may well be that they simply measure different things. One plausible explanation for this emerges from the extensive work by Cummins and colleagues on the homeostatic effects on subjective well-being [28, 29]. In this view, individuals have genetically-programmed "set-points" to which their moods have a strong tendency to return, despite external (objective) circumstances. Times of joy and times of stress are temporary emotional states for most healthy individuals, but the set-point is the stable mood regulator. In short, we have an innate mechanism for finding contentment in our various circumstances, as long they are not unduly stressful or unduly prolonged. It would be useful in the future to continue these lines of thinking and to undertake new work that would help us understand if objective and subjective measures should be considered as distinct measures, or if each contributes to a broader measure of QoL.

It would also be useful to determine if either objective or subjective measurement should dominate the other in QoL measurement. Measurement based primarily on objective indicators has the advantage of being easier to validate and perhaps to gather information, but measurement based primarily on subjective information honours the person-centred nature of the QoL approach.

The question of whether respondents' perceptions should be considered objective or subjective is an interesting one. An example of this is a study on the difference between the universal (etic) and cross-cultural (emic) properties of QoL reported by Schallock and collaborators [30] and Jenaro [31]. These researchers surveyed three respondent groups (consumers, professionals, and parents) from six geographical areas (South and Central Europe, Latin America, Canada, China, and USA) on the importance and use of the 24 core QoL indicators most commonly reported in the international literature. Results seemed to confirm etic and

emic characteristics of the QoL construct by indicating that: (1) there are similar profiles across respondent and geographical groups; and (2) there are significant differences in mean QoL, importance, and use scores. But it is the measurement method that is of interest here. Respondents gave their perceptions of importance and use on the 24 indicators that were themselves QoL domain-specific perceptions, behaviours or conditions that give an indication of the individual's actual well-being. This measurement method, then, uses respondents' perceptions of both others' subjective experiences and objective indicators of life experience as data for QoL measurement [8].

There is general agreement in the QoL measurement literature that the concept quality of life is multi-dimensional (e.g., Schallock and colleagues) [12]. Most instruments set out QoL indicators under a number of logical domains. Schallock and his colleagues identified eight core domains that are most common in the QoL literature: emotional well-being, interpersonal relations, material well-being, personal development, physical well-being, self-determination, social inclusion and rights [32, 15, 33]. Two things are not clear at this point: first, whether the domains identified to date accurately capture QoL, and second, the extent to which the domains make unique contributions to overall QoL.

There is emerging evidence from analyses of the study referred to above [30, 31], that these two points merit some future exploration. The factor scores on importance and use generally did not fit into the eight core domains proposed by Schallock to describe the set of cross-cultural factors that can impact personal well-being. Furthermore, QoL domains as defined by Schallock et al. have been empirically investigated across the world with different results. In Poland, Otrebski [34] found that professionals' age, education, and place of work can strongly affect the evaluation of the importance and the use of certain domains. In the component analysis of the importance ratings conducted by Xu and colleagues [35] in China, two (personal development and physical well-being) of the original eight domains were highly correlated and loaded onto one component. Aznar and Castanon [36] gathered 111 statements from the pool of answers given by 50 Latin-American families to the question: "What does living a life of quality means for the disabled member of your family?". They found a very high variability of the relevance of these statements to the eight QoL domains. The percentage of statements by domains ranged from 27 in interpersonal relations to 4 in physical well-being.

According to Reinders, an authentic comprehensive evaluation of QoL must necessarily explore the anticipation of a different future and this exploration has to be extended to each domain of QoL investigated. In a critical review of the Schallock and Verdugo's conceptualization, Reinders observed that, without further qualification, objective (shared QoL) measures do not actually measure QoL, but only separate domains of life. People do not typically experience their QoL as divided among separate domains, but rather evaluate how their experiences within these various domains interact with one another. Furthermore, some QoL domains of the

Schalock and Verdugo’s model, such as “self-determination” or “personal development” should be attributed to “individual QoL” and assessed as qualifiers of all QoL domains.

A good combination of “shared” and “individual” QoL assessment is provided by the tool produced by the Quality of Life Research Unit, Centre for Health Promotion, University of Toronto, called Quality of Life Instrument Package (QoL-IP) [37]. This tool and the model upon which it is based foresee that there are areas of life with emic transcultural value, which have qualitative implications for anybody’s life, independently from the presence of disability and its specific features. The tool also assumes that the best way to assess the individual gradation of quality in these areas is represented by the following four dimensions: 1) importance, attributed by the person to every area of life; 2) satisfaction perceived by the person in the same areas; 3) opportunities that the person had/has to develop importance and to perceive satisfaction in the different areas of life; and 4) choices that the person could/can make in the same areas.

According to our literature mapping, the QoL-IP is the only tool that explicitly applies the relationship between importance and satisfaction to QoL measurement, an idea that was put forward by a number of scholars in the 1990s, including Becker, Diamond, and Sainfort (1993) [38]. According to this idea, a thing that is highly important and gives high satisfaction has a considerable positive impact on a person’s QoL, but a thing that does not interest a person or is not valued will never add satisfaction or quality to the person’s life [6, 3].

The QoL-IP includes nine areas of emic transcultural value, which are gathered in the three macro-areas of being, belonging, and becoming [39] (see Table 1).

The dynamic QoL model that underlies the QoL-IP is linked in a straightforward way with the well-known capability theory proposed by Amartya Sen [40]. According to Reinders [18], a dynamic approach to QoL “is oriented toward the goal of human flourishing. Human beings flourish to the extent that they are enabled to develop their own capabilities ... when they receive sufficient opportunities to develop their own gifts and talents”. This claim can be rephrased by saying that QoL in a capabilities approach is agent relative, and that

they are typically owned by agents. In Sen’s perspective, agency goals are those objectives that people set themselves, even if they do not benefit directly from them, or indeed even if they undermine other capabilities. Sen saw that people often have goals that are not in their interests, such as sacrificing their health or even their life itself for religious, political, or aesthetic achievement. Adding agency goals to the account serves Sen’s theoretical objective of criticizing welfarist conceptions of the good human life, such as utilitarianism. “The general idea of the freedom to achieve well-being can be called well-being freedom... The existence of genuine choice may actually affect the nature and significance of the functioning achieved” [41].

Reinders suggests that a QoL measure is effective only if conceived as addressed to a human being and his life story, since the latter “provides the hermeneutical context within which agent-relative capabilities are discovered and realized” [18]. A version of the QoL-IP adapted to the Italian language and culture has been recently validated by Bertelli and collaborators [42].

Another commonly used QoL measure is the one developed by the World Health Organisation (WHO), called WHOQOL-DIS (disabilities module), which is an adaptation (supplementary module) of the WHOQOL-BREF for the general population. The WHO developed two main generic QoL tools for the general population, the WHOQOL-100 and the WHOQOL-BREF which is a shorter 26-item version. The WHOQOL-100 has a hierarchical structure that includes overall quality of life, a next level of domains, then facets within each domain, and then four specific items for each facet. The shorter WHOQOL-BREF has a similar hierarchical organisation, except that each facet is represented by a single item.

This tool includes a smaller number of domains and item in comparison to the previously examined measures. It takes into account some aspects of Individual QoL, but in a very simple and partial way. Nevertheless, the WHOQOL methodology provides for cross-cultural validity for QoL assessment across the adult lifespan and across a range of physical and intellectual disabilities [43].

In healthcare services, a large number of measurement instruments have been developed that purport to measure health-related QoL (HR-QoL). These mea-

**Table 1**  
QoL areas/domains in the main QoL assessment tools for person with ID/LF-ASD

Tool	WHOQOL-BREF and disabilities module [44]	Quality of Life questionnaire [15]	Quality of Life instrument package [37]
Number of QoL areas/ domains	5	8	9
List of QoL areas/ domains	<ul style="list-style-type: none"> <li>- Physical</li> <li>- Psychological</li> <li>- Social</li> <li>- Personal environment</li> <li>- Disability</li> </ul>	<ul style="list-style-type: none"> <li>- Personal development</li> <li>- Self-determination</li> <li>- Interpersonal relationships</li> <li>- Social inclusion</li> <li>- Rights</li> <li>- Emotional well-being</li> <li>- Physical well-being</li> <li>- Material well-being</li> </ul>	<ul style="list-style-type: none"> <li>- Physical being</li> <li>- Psychological being</li> <li>- Spiritual being</li> <li>- Physical belonging</li> <li>- Social belonging</li> <li>- Community belonging</li> <li>- Practical becoming</li> <li>- Leisure becoming</li> <li>- Growth becoming</li> </ul>

asures focus on the effects of overall personal health and medical treatment, the presence of physical or mental health conditions and disorders, and also on the effects on the person of specific diseases and treatments (e.g., cancer, HIV, diabetes, arthritis); (See the Quality of Life Instrument Database of the Mapi Research Institute for a list of available instruments). In fact, for people with NDD, there is international agreement that we must be cautious in interpreting scores from all non-generic (specifically-focused) instruments, such as those that measure health-related QoL [11]. HR-QoL instruments typically try to measure both the degree to which symptoms are present and their effects on daily functioning. Thus, although they do focus on the individual person in a way that is of some benefit, they still support a theoretical perspective of QoL that has not departed significantly from the traditional medical approach of only identifying and effectively treating diseases and conditions [44]. From the time QoL began to emerge as an important focus in the field of NDD in the late 1980s, it has focused more broadly on the whole person's life as well as on key processes that enable people to achieve lives of quality (e.g., identifying what is valued by the person, honouring each person's unique interpretation of satisfaction and happiness, providing environmental opportunities, permitting and enabling personal choice, and promoting the concepts of empowerment and self-determination) [3].

How to measure a whole person's life, and how to weigh in the effects of these key processes, is an ongoing challenge. Brown and colleagues [45] asserted that satisfaction scores weighted by importance scores, by way of a mathematical formula, constituted what they called basic quality of life scores. When an item is important to a respondent, the satisfaction score is weighted more heavily than if the item is not important. These authors also noted, however, that basic quality of life scores do not tell the whole story. To illustrate, they also collected scores for both decision-making, and the availability of opportunities from which decisions can be made, which, they claimed, act as moderators to basic quality of life scores. Thus, we come to the important measurement question of whether specific measures are true outcome indicators of QoL, or if they help to explain current QoL scores. For example, and to expand on the concepts of decision-making and opportunities, a woman who lived in a restricted environment for 40 years valued the recreational activities provided for her and felt very satisfied with them (indicating that she was enjoying recreation of high quality), but she had never been exposed to the many opportunities for recreation offered by her broader community and had never had a voice in making decisions about whether or not to pursue other options. Although her current QoL scores in this area of life were high, it is difficult to assert that her recreational activities were the best ones she could have had. Another example is a man who lived in a group home and was very dissatisfied with his social activities, although they were numerous. His dissatisfaction emerged from the fact that everyone in the home had to attend the same events because there was only one staff on duty at any one time. He had many

opportunities, but his decision-making power was low. Variables such as decision-making and opportunities can be useful in explaining QoL scores, but they are not themselves QoL outcome measures [11]. The relationship between making decisions, choices, or other ways to exercise self-determination is a QoL issue that is the subject of ongoing work [46-48], but there are strong indications that these are universal concepts that hold up in cross-cultural contexts (e.g., Zhang and collaborators) [49].

In recent years, the impact on QoL of the participation of the person with ID/LF-ASD in decision-making processes, and the level of self-determination have represented a particularly lively area of research. According to Lachapelle and collaborators [46], self-determination and the possibility of choice co-vary in an almost linear way with QoL scores. Other authors consider these aspects so important as to include their implementation in the guidelines for good practices [50].

In Cummins' opinion [11], QoL measures might be characterized by either indicator or causal variables, or both, and further research should be conducted to make clearer distinctions. In addition, Cummins suggested that work should be carried out to organize QoL indicator variables for life as a whole in a hierarchical way, a process that would help us better define a minimum set of domains with equal variance.

QoL measurement often relies – sometimes quite heavily – on people's own perceptions and expressions of their well-being or general satisfaction with life, referred to in the measurement literature as subjective well-being (SWB). Since life is experienced by individuals through their unique sets of perceptions, SWB may be an essential pathway to recording “true” personal QoL. However, when recording SWB, it is often noted that QoL scores of people with severe disorders or disabilities do not significantly differ from those of the general population. This so-called “disability paradox” originally introduced by the widely-known article by Albrecht and Devlieger [51], has been explained in various ways, such as methodological bias, or the psychological mechanism of adaptation, coping, or resilience.

For many scholars the “disability paradox” is just the result of a poor conceptualization. For example, Koch [52] highlighted the circularity involved in the notion of “disability paradox”: “(a) physical normalcy equals acceptable life quality; (b) physical differences result in diminished life quality; ergo, an acceptable life quality cannot exist in the face of physical differences. Paradox demands only that a person (a) exhibits conditions that negatively diverge from the norm and (b) reports a positive life quality”.

Considering a population with intellectual disabilities whose cultural marginality, limited opportunities for personal development and social inclusion are widely documented, Reinders does not want to ignore the fact that “...people with disabilities may be content with their lives because they are unfamiliar with other possibilities” [18].

Cummins theory, referred to above, has presented a compelling case for SWB as the result of an affective/cognitive homeostatic system that is characteristic

of humans. Simply put, we have a built-in tendency to assess the world around us, and our place within that world, in normative ways by perceiving that “life is okay”. Only in extreme cases, where there is a strong overload, does the homeostatic system fail. Thus, SWB may not be a particularly sensitive measure, but, when it is low, it may be cause for serious concern [11].

### **QoL measurement summary and future directions**

QoL conceptualization and measurement have advanced very considerably over the past 20 years, but work in this area is ongoing and requires further development. The tools used for the evaluation of QoL in people with DNS – and the theoretical models from which they derive – present many commonalities but also important differences. The latter represent a considerable obstacle for the implementation of research on practical consequences of using this type of outcome measures.

It will be important to clarify the relationship between QoL based on shared human characteristics and QoL as experienced in unique ways by individuals. It will be essential to address emerging measurement issues, such as those referred to above and others. One expressed goal is to have, in the near future, a generic instrument that can be validly applied to anybody, irrespective of health condition, disability, culture or socio-economical status [11]. Finally, it will be critical to focus carefully on how QoL as a concept and measurement tool are applied in clinical settings, in order to promote practices that enhance QoL of individuals with ID and to avoid practices that (purposely or inadvertently) detract from QoL. For the field of psychiatry and mental health, in particular, understanding that the QoL concept can be applied to everyday practice in a way that is systematic, reliable, and helpful, is the challenge for the near future. To address this challenge, we will need methodologically rigorous research, carried out by professionals with expertise in research and application, that evaluates over time the effectiveness of taking a QoL approach in mental health care practice.

Family QoL is an area of study that has just begun, but it promises to be an important focus in the future. An international group of researchers led by Brown and colleagues from Canada [53] developed a theoretical framework and validated a survey instrument for family QoL. The first version of this instrument was published in 2000 [54], and the second version, available in a number of languages, in 2006 [53]. This instrument includes ratings for six core concepts (importance, opportunities, initiative, stability, attainment and satisfaction) across nine domains: family health, financial well-being, family relationships, support from other people, support from services, influence of values, careers and preparing for careers, leisure and recreation, and community integration [55]. Like individual QoL, a life of quality for a family refers to aspects of family life that are valued by families everywhere, tempered by unique values of an individual family [56].

An instrument for assessing the impact of services on family QoL has been developed by The Beach Center on Disability at the University of Kansas. This instru-

ment, which has strong psychometric properties, includes 5 domains (interactions, parenting, emotional well-being, physical/material well-being, and disability-related support) and 25 items [57, 21]. Another tool was developed by Aznar and Castanon [36] in their investigation on 180 Latin American families. The 42 items of this instrument are grouped in the following 6 areas: emotional well-being, personal strength and development, rules of cohabitation, physical/material well-being, family life, and interpersonal and community relations.

Though on a very small sample, Jokinen and Brown [58] specifically investigated the family QoL of older parents of adults with ID. They used an adaptation of the Brown, Neikrug and Brown [59] *Family Quality of Life Survey* referred to above. Findings indicate, among other things, that older parents have many concerns in common and ongoing concerns about issues related to family caregiving. All respondents reported satisfaction with leisure and life enjoyment, while contribution to community and civic affairs received the lowest satisfaction ratings. Recent work in family quality of life also underscores the importance of exploring the relationship between concepts that are QoL outcome indicators and those that help explain it. The Brown et al., [60] family quality of life survey referred to above provides a way to score six key outcome measures: importance, opportunities, initiative, stability, attainment and satisfaction. Of these, attainment and satisfaction are considered QoL outcome indicators, and importance, opportunities, initiative, and stability are measures that help to describe, explain, and modify the extent to which attainment and satisfaction are relevant.

The relationship between individual and family QoL appear to be quite complex and needs to be clarified in future research [61]. To date, few correlations have been found between the perceptions of people with ID themselves and the perceptions of their family members [62], most of which refers to financial well-being, family relationships, support from services, and support from others [62]. This difference in QoL perception could negatively impact on QoL of people with ID, if their views are not taken into account when planning for family support.

### **For which persons with ID/ASD is measurement possible?**

It is sometimes thought in the field of mental health and in related fields, that people with ID do not possess a sufficient level of cognitive ability to process information and perceive emotions to the degree that they can make accurate judgements about their own life satisfaction. This belief has been strongly discredited in the field of ID as having no demonstrated validity. It is now widely held that people with ID, even those who do not have formal communication systems, are able to express their own likes and dislikes, satisfaction and dissatisfaction, and that such expression is valid because it represents the perception of the person at the time.

Still, other people close to the person with ID have their own perceptions of the person's life. Although there is considerable evidence that proxy data (infor-

mation given by others close to the person) does not correlate well with data from people with ID themselves [63, 64, 8], it may still be important to assess QoL from others' points of view. As Brown, Raphael and Renwick [37] explained, such other people frequently make decisions on behalf of a person with ID, or are influential in helping the person with ID to make decisions, and thus the perspective of those other people are important to note. They are all the more important to note if they differ from the perspective of the person with ID whose life is being influenced. On the other hand, information from proxies can be very helpful to an assessment of QoL, by adding historical information, clarity and depth that might otherwise be missing due to skill limitations. It might even be argued that proxy information is a necessary addition to self-perception.

It seems best to take a comprehensive approach to assessing QoL for use with individuals with ID. Self-report by people with ID should be central, and any means should be mobilized to enable people with ID to express their own views. Information on the background of the person with ID, descriptions of the person's current context and living/working environments, and the perceptions of proxies constitute additional sources of information that are potentially valuable to add to self-perceptions. There is no formula for assessing QoL, nor should there be. Rather, the QoL approach is above all a way to explore the rich intricacies of personal quality of life.

Such assessment may even be used effectively with people with severe ID, as they express their inner states through consistent behavioural repertoires [65].

#### **Additional research findings**

QoL of persons with ID has repeatedly been found to be lower than that of persons without ID. Keith and Bonham [22] identified some main differences in the domains of rights and empowerment/independence, while Bramston and collaborators [66] found that those with ID attribute significantly less importance to all areas of life except material well-being, and report significantly lower satisfaction with intimacy and community involvement, but higher satisfaction with their health. For people with ID, satisfaction with safety, emotional well-being, material well-being, and health are significantly predicted by social support. Health is predicted also by stress in interpersonal relationships [66, 67]. People with ID desire friendship and need support in this area [68, 69], which is often undervalued or presumed to be implicit in the community inclusion [70].

Considerable QoL differences between persons with and without ID have been reported also in respect to employment, with non-disabled workers reporting higher QoL and autonomy. However, Beyer and collaborators [71] found that supported employment can significantly increase satisfaction towards material well-being, productivity, safety, place in the community and emotional well-being despite objective disadvantages. Kober and Eggleton [72] investigated the impact on QoL of open vs sheltered employment, finding no differences for participants with a low Functional Work Ability (FWA), while participants with high FWA in open em-

ployment reported statistically significant higher scores of empowerment/independence, social belonging/community integration, and total QoL, than participants in sheltered employment. Verdugo and collaborators [73] indicate that in supported employment, high levels of job typicalness (the degree to which the characteristics of a job are the same as those of workers without ID) and certain characteristics of support (i.e. hours of direct external support) are associated with a higher QoL.

Also for persons with ASD, being employed has been identified as a predictive factor of a better QoL, together with being in a relationship and receiving support, while lower QoL was associated with being female, having a current mental health diagnosis and higher severity of autism symptoms [74].

In general, persons with ASD have been found to have a lower QoL than the general population [74, 75].

#### **By whom should QoL be measured?**

Many researchers believe that assessment tools can detect a person's QoL in a valid way only when used directly with people with good cognitive and introspective abilities. For this reason, their use with people with ID might be discouraged, especially in those with greater impairment. This position is not supported by evidence, which indicates instead that people with ID are not only able to process information, perceive sensations or express emotions in a way suitable to the attribution of importance and the perception of satisfaction in the various areas of the life, but also to manifest these importance and satisfaction to others, at least behaviourally. This applies also to persons with more severe cognitive impairment [76]. In general, it has never been shown that lower IQ, however severe, should be sufficient reason to rule out capacity for self-awareness and self-determination [77, 78, 50].

On the other hand, it has never been shown that individual QoL should be assessed only from the person himself/herself. In fact, assessment in other fields typically emphasizes a comprehensive approach. Practical assessment experiences suggest that evaluations and scores provided by proxies, or other persons less close to the person under assessment, can provide valuable information, adding stability and clarity, and extending the range of assessment information gathered. Such additional information is also an opportunity to understand that assessment information from people themselves is often shaped by various factors, such as specific life events, psychological, or psychopathological states. Some authors have stressed this point of view by claiming that only using individual perception to measure QoL represents a disadvantage to its measurement [11, 8].

Data on the consistency between self-assessments and proxy reports are not homogeneous, even if those indicating a lack of consistency seem to prevail [63]. One argument in support of the low validity of proxy assessment is the presumed existence of a gap in the attribution of quality between people with ID/LF-ASD and those with typical development. Some research findings seem to refute this hypothesis, as they observed a substantial agreement of the assessments performed by proxies with and without ID [79, 80]. Proxy attribution of QoL in the

**Table 2**

State of knowledge and current issues in QoL assessment for people with ID and/or LF-ASD

Mapping question	Key points
Purpose of QoL measures	- shared QoL - personal QoL - family QoL
What QoL measures actually measure	- domains/areas (objective assessment) - dimensions (personal appraisal) - indicators (individual objective assessment)
Persons for which measurement is possible	- every person with ID/LF-ASD (any degree of impairment)
By whom QoL measurement should be made	- individual with ID/LF-ASD - proxies (persons who know the individual well) - external assessors

instrumental assessment does not seem to be influenced even by the degree of disability. Findings suggest that prejudicial attitudes towards the QoL of people with severe ID may be either absent or contained [42].

In general, the literature indicates that the best way to evaluate QoL is to integrate self- and hetero-assessments, which implies in practical terms the merging of data from questionnaires addressed directly to the person whose QoL is under assessment with data from questionnaires filled in by proxies and external assessors. Within this system, the self-assessment must be considered central and it is necessary to provide persons with ID/LF-ASD with every means that can support them in expressing their point of view.

A good example of how this can be implemented even in people with more severe ID is the Lyons *Life Satisfaction Matrix* [65]. This approach assumes that people with severe ID express importance and satisfaction through recurrent behavioural repertoires, identified by those closest to them and validated by other people less involved in the relationship. Lyons' approach also assumes that individual's routine daily activity preferences can be determined from their affective behavioural repertoires [65]. The QoL-IP referred to earlier [37] also keeps self-assessment central, but supplements that with QoL measures both from "other persons" (two people who know the assessed person well) and the external assessor.

To determine subjective components of QoL in persons with severe ID/LF-ASD, other authors used observable expressions and responses, such as facial reaction (smiling, pouting, etc.), sounds or body movements. Various observers have been used, including unfamiliar observers, and confirmation was often provided by supervisory staff [81-83]. Verdugo and his collaborators, during a long validation process of a new questionnaire for proxy assessment of QoL in people with severe ID, confirmed that the QoL areas upon which the assessment has to be based do not differ from the ones for mild

ID and for the general population [84]. This supported earlier assumptions in the use of proxy assessment areas. The World Health Organization has recently completed the validation of an adaptation for people with ID, the WHOQOL-Dis [85, 43] of its QoL assessment tool (WHOQoL) [86] and promoted the evaluation of its applicability to various socio-cultural contexts [87].

## CONCLUSIONS

Key points of the state of knowledge and current issues in QoL assessment for people with ID and / or LF-ASD, identified through our systematic mapping of the scientific literature, are summarised in *Table 2*.

The literature substantially agrees that QoL represents a multidimensional concept, best measured through an integration of qualitative and quantitative methods as well as a through a combination of subjective and objective perspectives. Most authors agree that the assessment should be based on a system of values consistent with that of the person with ID/ASD, and that it should be used to identify the most useful supports and interventions across the life span.

Currently available tools, although they have some common conceptual and evaluation characteristics, still show considerable differences. The most important concerns are: the areas, or domains, that should be included in the "shared QoL"; the dimensions used to evaluate "Individual QoL"; and the role attributed to indicators of QoL.

A highly effective tool should be based on a standardization of all these aspects. The rapid evolution of QoL research in the last decades gives hope that this will occur.

## Conflict of interest statement

The Authors declare no conflict of interest.

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