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Quality of life of persons with profound intellectual and multiple disabilities: A narrative literature review of concepts, assessment methods and assessors

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ABSTRACT

Background: The objective of this study was to provide an overview of concepts, methods of assessment and assessors in empirical studies on quality of life (QoL) of persons with Profound Intellectual and Multiple Disabilities (PIMD).

Method: A narrative literature review was conducted by searching electronic databases until December 2016, and the included studies were then analysed thematically.

Results: Twenty-one articles were included in the analysis. Multidimensional concepts of QoL consisting of both subjective and objective components were found, although the articles used different distinctions for each. QoL was assessed by behavioural observations or interviews/questionnaires. Family members, primary caregivers and more distant observers (or a combination of these three) determined QoL. Triangulation of assessors and methodological pluralism were used.

Conclusions: This review highlighted several components of QoL in persons with PIMD, as well as challenges in researching the QoL of persons who are not able to express themselves verbally. The distinction between objective and subjective QoL was difficult to clarify, with interpretation being both inevitable and necessary. Although triangulation is seen to be good practice in QoL assessment, the role of proxies in assessing the QoL of persons with PIMD needs further investigation.

KEYWORDS

Quality of life; profound intellectual and multiple disabilities; intellectual disability; developmental disabilities; ethics; review


Quality of life (QoL) is often a key factor in the decision-making process for those caring for people with complex care demands (Zaal-Schuller, de Vos, Ewals, van Goudoever, & Willems, 2016). There are decisions to make about major surgeries, tube feeding and withholding or sustaining life support. Clarity in the description and consensus about the assessment of QoL is therefore crucial. Obviously, QoL can best be judged by the individuals themselves, but persons with Profound Intellectual and Multiple Disabilities (PIMD) cannot verbally express how they experience their QoL.

Persons with PIMD are individuals with profound intellectual disabilities (IQ < 20) and profound neuromotor dysfunctions, sometimes with sensory impairments and even medical problems (including seizures, respiratory and feeding problems). Furthermore, these persons seem to have neither an apparent understanding of verbal language nor a symbolic interaction with objects (Nakken & Vlaskamp, 2007). This is in accordance with the working definition developed by the Special Interest Research Group (SIRG)

PIMD of the International Association for the Scientific Study of Intellectual and Developmental Disabilities (IAS-SID). Persons with PIMD are dependent on support from others in all aspects of their daily needs. Communication is mostly pre- or proto-symbolic and consists of movements, sounds, body postures, facial expressions or muscle tensions (Maes, Lambrechts, Hostyn, & Petry, 2007). Persons with PIMD are not officially registered as such, but it is assumed that in the Netherlands approximately 0.06% of the total population have PIMD, although most of them were adults (Vugteveen, van der Putten, & Vlaskamp, 2014).

Since 2000, much academic attention has been given to QoL of persons with intellectual disabilities (ID) (Cummins, 2005; Schalock, 2004; Schalock, Bonham, & Verdugo, 2008; Schalock et al., 2002). Schalock et al. (2002) described the core principles for measuring QoL, stating the following:

- (1) QoL measures the degree to which people have meaningful life experiences that they value.

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- (2) QoL measurement enables people to move towards a meaningful life they enjoy and value.
- (3) QoL measures the degree to which life's domains contribute to a full and interconnected life.
- (4) QoL measurement is undertaken within the context of environments that are important to people: where they live, work and play.
- (5) QoL measurement for individuals is based upon both common human experiences and unique, individual life experiences.

In addition, several books describe the concepts of QoL. In his overview of the literature, Lyons (2010) referred to the Special Interest Group on quality of life (SIRG QoL) of the IASSID and the criteria it describes for research on QoL in persons with ID. These criteria include the following: multidimensionality, guiding operational principles, methodological pluralism, a system perspective and a focus on individual and family QoL. In 2009, Petry and Maes wrote an overview chapter in a book in which they specifically addressed the importance, definitions and measurement of QoL in persons with PIMD. However, they did not provide a review of available studies.

Therefore, no overview of the literature on the assessment of QoL in PIMD has been provided, including articles published after 2009 and supported by a systematic search of electronic databases. Such an overview is necessary to clarify the current state of concepts, methods of assessment and assessors in the research on QoL in persons with PIMD. This can be helpful in future research on QoL in persons with PIMD related to medical decisions. In addition, more information on this subject may help physicians and family members to make adequate decisions when they are confronted with medical care decisions related to the QoL of persons with PIMD. Therefore, the main aim of this study was to answer the following research questions: (1) Which concepts are used for QoL in persons with PIMD? (2) Which instruments or methods are used and which persons determine QoL?

Method

A narrative literature review of the empirical studies describing QoL in persons with PIMD was conducted.

Literature search and selection of articles

The studies were identified through searches in the following databases: MEDLINE, EMBASE, CINAHL, PsychINFO and Philosopher's Index, with the help of an experienced clinical librarian. These searches first occurred in March 2015, and the searches were repeated in May 2016 and December 2016. Two sets of keywords

were used, the first relating to QoL, including but not limited to QoL, Health-related QoL or wellbeing. The second related to the target group, such as persons with PIMD or persons with profound, severe or multiple mental retardation; those with a learning, developmental, intellectual disability; or those with a medical complexity, minimal movement or cerebral palsy. A complete description of search terms and strategies is available from the authors.

We excluded the following sources: articles not written in English, Dutch, German or French, as well as editorials and conference abstracts. In addition, articles in which persons with PIMD were not the target group were excluded. We also excluded articles in which QoL was not the subject but an outcome measure in an interventional study. And finally, articles for those which the subject was QoL of relatives of persons with PIMD, for example, were also excluded.

The inclusion criteria for the target group were a combination of multiple disabilities including both an intellectual and physical disability with a dependence on support from others in all activities in daily life, as well as no apparent understanding of verbal language. Only peer-reviewed articles were included. From these, 3182 articles were found including 790 duplicates, leaving 2392 usable articles. For the search strategy, see [Figure 1](#) (flow chart).

Procedure

In order to apply the inclusion criteria, the first and the last author independently screened the titles of the 2392 articles and then compared their findings until they reached consensus. The articles that clearly did not fulfil the inclusion criteria were excluded. When there was doubt, the article was included for the next round. The abstracts were then screened, following the same procedure. The two researchers had doubt on 18% of the articles, and after discussing together there remained doubt on 6%. This 6% was screened by the second author of this paper, who made a final decision. Of the remaining articles, the full text was screened by the first author and articles that raised doubt (21%) were also screened by the last author. The cross-references of the included articles were screened as well by the first and the last researcher. Both researchers discussed including or excluding these articles until they reached consensus.

Analysis

First, the articles were screened thematically on the research questions (concepts, instruments/methods and assessors). Then, differences and similarities between descriptions of concepts were compared. Subsequently,

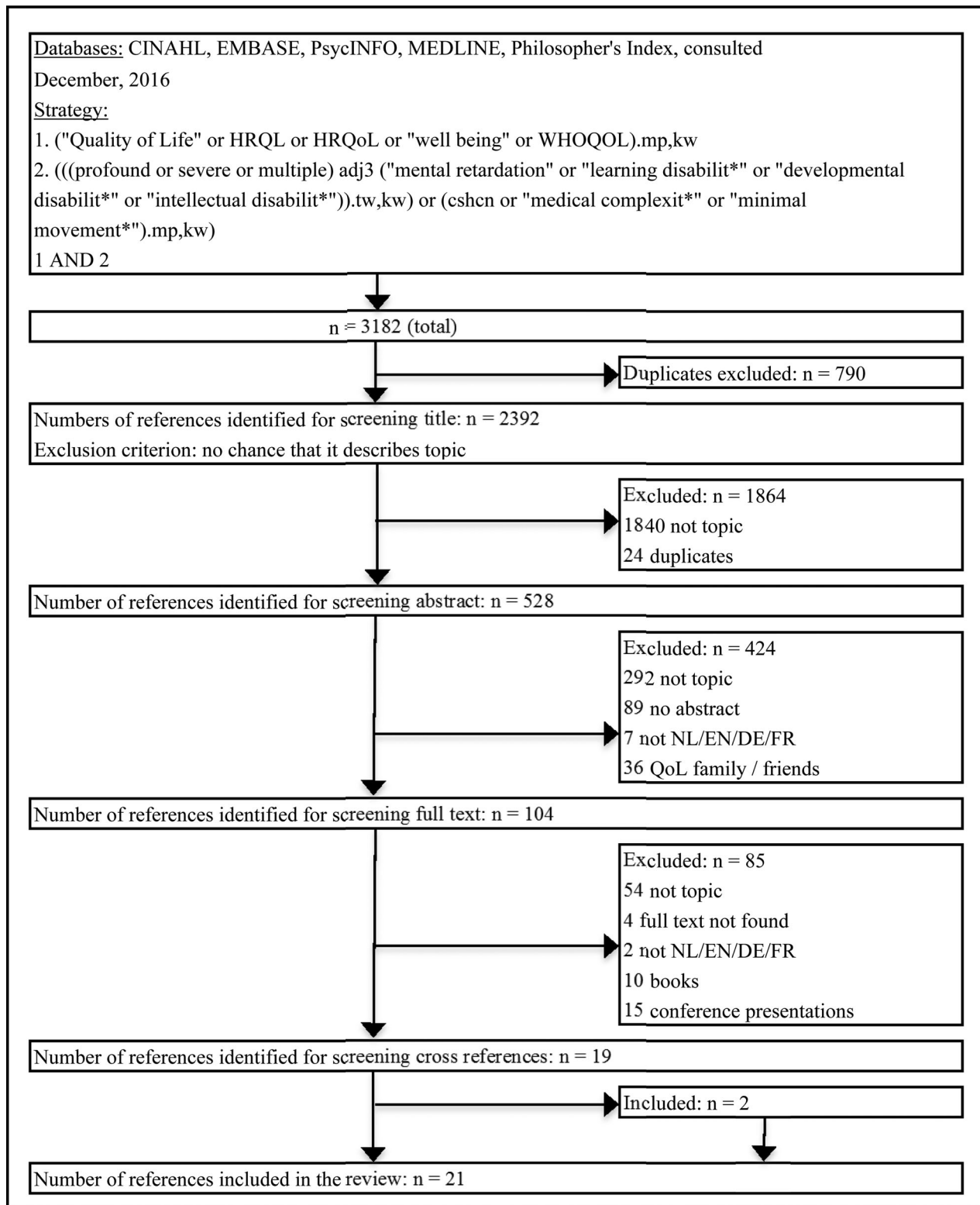


Figure 1. Flowchart.

an inventory was drawn up, both the instruments and methods of assessment described and an inventory of the assessors. After that, we searched for relationships between the concepts and the assessment methods

used. The results of these thematic analyses were discussed by the first and the last researcher, who went back and forth several times between the included articles and the thematic analysis in order to refine the

analyses. For example, many articles appeared to refer to both the subjective and the objective component (as one part of the concepts). Therefore, we searched in the articles for the definitions of subjective and objective components and the possible relationships between their definitions and the assessment methods. We found that subjective components were assessed by the use of different methods and objective components mostly by questionnaires. In order to sharpen and critically test the initial analyses, they were also discussed with the other authors of this paper.

Results

Twenty-one articles were included. For a summary of the included articles and results, see [Table 1](#). The included studies were performed in Europe, the US, Canada and Australia. Also included were studies aiming to develop a procedure for evaluating QoL of people with PIMD, conducted in Belgium, the Netherlands and Germany (Petry, Maes, & Vlaskamp, 2001).

Which concepts of QoL are used?

The articles included did not directly answer the question “which concepts are used for QoL?” However, 12 of the included articles referred to Schalock’s principles relating to QoL (Bigby, Knox, Beadle-Brown, & Bould, 2014; Hartnett et al., 2008; Lyons, 2005; Petry, Kuppens, Vos, & Maes, 2010; Petry & Maes, 2006; Petry et al., 2001; Petry, Maes, & Vlaskamp, 2007b, 2009a; Vehmas & Watson, 2014; Vos, De Cock, Petry, Van Den Noortgate, & Maes, 2010a, 2010b, 2013). Two articles referred to the SIRG QoL (Hartnett et al., 2008; Petry et al., 2007b) in which QoL was conceptualised by the following descriptions: (a) QoL is multidimensional and influenced by personal and environmental factors and their interactions; (b) it has both subjective and objective components; (c) it has the same components for all people and (d) it is enhanced by self-determination, resources, purpose in life and a sense of belonging. The last element was not examined in the studies included in this review.

With respect to multidimensionality, several combinations of domains were found. For an overview of domains, see [Table 2](#). These several combinations of domains were operationalised in items (Bigby et al., 2014; Gómez, Arias, Verdugo, Tassé, & Brown, 2015; Petry, Maes, & Vlaskamp, 2005, 2007a), and twice a Delphi round was used (Gómez et al., 2015; Petry, Maes, & Vlaskamp, 2007a). In addition, in the article of Petry et al. (2005), the basic domains of Felce and Perry (1995) were described as valid and relevant for people with PIMD, which may refer to the third SIRG QoL

statement that QoL has the same components for all people.

References to objective and subjective components of QoL were found in 10 studies (Lyons, 2005; Ouellette-Kuntz, McCreary, Minnes, & Stanton, 1994; Petry et al., 2010; Petry & Maes, 2006; Petry et al., 2007b, 2009a, 2009b; Vos et al., 2010a, 2010b; Vos, De Cock, Petry, Van Den Noortgate, & Maes, 2013). Vos et al. (2013, p. 441), following Cummins (2005), Landesman (1986) and Perry and Felce (2005), described the objective component of QoL as the “information on the life of the person that has properties that can be measured in the public domain and verified by different parties (e.g., how many visitors one has during the week).” On the other hand, Vos et al. (2013, p. 441) defined the subjective component as the information “about the private world of experience of the person, which can be viewed as an estimate of the true subjective experiences and cannot be verified directly through any evaluation of the variable in question beyond the level of the individual.” The following subjective components were examined: happiness (Favell, Realon, & Sutton, 1996; Green & Reid, 1996), pleasure (Petry & Maes, 2006), emotions (Vos et al., 2013), mood (Petry et al., 2010; Ross & Oliver, 2003; Vos, De Cock, Petry, Van Den Noortgate, & Maes, 2010b, 2013), life satisfaction (Lyons, 2005) and interest (Petry et al., 2010; Ross & Oliver, 2003). However, the studies made different distinctions. For example, one author includes emotions in the subjective components of QoL (Vos et al., 2010a, 2013), whereas the QoL-PMD includes emotional wellbeing in the objective components of QoL (Petry et al., 2005, 2007a, 2009a).

Some of the included articles referred to the (strategic) use of the concept of QoL (Petry et al., 2010, 2001, 2009a; Vos et al., 2010b, 2013). They stated that QoL was used as a conceptual framework for assessing quality outcomes, that is, as a social construct that guides quality enhancement strategies and as a criterion for assessing these strategies (Verdugo, Schalock, Keith, & Stancliffe, 2005). Petry, for example, mentioned that persons with PIMD need special care, and consequently that attention should be given to the optimisation and evaluation of the quality of care because its most important result is an enhanced QoL (Petry et al., 2001).

Which instruments/methods were used and who determined QoL?

Six instruments and assessors. Five questionnaires determining QoL in persons with PIMD were found, as well as one instrument using observations and interviews, which are presented in [Table 1](#). According to the authors (Ouellette-Kuntz et al., 1994), the QUOLIS measured

Table 1. Characteristics of the included articles.

1st author	Year	Objective	Persons with PIMD	Study design	Components of QoL	instruments or methods to assess QoL PIMD	Assessors	Psychometric properties
Ouelette-Kuntz	1994	To develop the Quality of life Interview Schedule (QUOLIS) and the intra- and inter-reliability	$n = 10$, age >18	Qualitative (interviews)	Twelve domains of Ouelette	QUOLIS: questionnaire (filled in by interviewer)	Trained interviewer, interview with two people who know the person well, family member or friend and staff member	Intra- and inter-rater reliability
Green	1996	To operationalise, measure and increase happiness among people with profound disabilities	$n = 5$, age 18–41	Qualitative (observations)	Happiness	Two experiments using behavioural observations & video tapes	Familiar and unfamiliar observers	NA
Favell	1996	To develop and demonstrate methodologies for measuring happiness in people with profound mental retardation and other severe handicaps	Study 1: $n = 7$, age not mentioned, study 2: $n = 1$, age 30, study 3 $n = 20$, age not mentioned, study 4: $n = 1$, age 30	Qualitative (observations of video tapes and other observations)	Happiness	Four studies using observations of video tapes and other observations	Supervisory staff (familiar), trained observer (unfamiliar)	NA
Petry	2001	To develop and implement a valid and reliable procedure for evaluating the QoL of people with profound and multiple learning disabilities	NA	NA (Study protocol)	NA	NA	NA	NA
Ross	2003	To develop an informant questionnaire on affect for use to adults with PIMD and examine its psychometric properties	$n = 53$, age >18	Quantitative (questionnaires, psychometric analysis)	Mood, interest and pleasure	MIPQ: questionnaire	Familiar informants	Inter-rater reliability; internal consistency; construct validity
Lyons	2005	To underpin the assumptions and face validity of the Life Satisfaction Matrix	$n = 22$, age <18	Qualitative (participant observations, semi-structured interviews)	Satisfaction	Life Satisfaction Matrix: observations and interviews	Most familiar communication partner, next communication partner and researcher (unfamiliar)	Face validity
Petry	2005	To consider the general validity of the basic domains of QoL in persons with PIMD	$n = 42$, age <18; $n = 34$, age >18	Qualitative (interviews)	Five domains of Felce and Perry	NA	NA	NA
Petry	2006	To explore a procedure for drafting individualised profiles of how people with profound multiple disabilities express pleasure and displeasure	$n = 6$, age 9–29	Mixed method (video tapes, questionnaires) ^a	Pleasure/ displeasure	Behavioural observations, questionnaire, videotape	Parent, direct support staff, supervisory staff and independent researcher (unfamiliar)	NA
André	2007	To describe QoL in children with severe disability and to look for factors that influence this QoL, in particular gastrostomy	$n = 28$, age 1–18	Qualitative (interviews)	Five domains of André	QUALIN: questionnaire (filled in by interviewer)	Parents	NA
Petry	2007a	To assess items on QoL and related aspects of support in people with PIMD	NA	Qualitative (Delphi study)	Five domains of Felce and Perry	NA	NA	NA
Petry	2007b	To examine which support characteristics are associated with QoL of people with PIMD	$n = 42$, age <18; $n = 34$, age >18	Qualitative (semi-structured interviews)	NA	NA	NA	NA
Hartnett	2008	To explore the contributions of day programmes to QoL	$n = 8$, age 21–32	Mixed method (interviews, questionnaires) ^a	Eight domains of Schalock	Interviews and questionnaire	Parents and staff	NA

(Continued)

Table 1. Continued.

1st author	Year	Objective	Persons with PIMD	Study design	Components of QoL	instruments or methods to assess QoL PIMD	Assessors	Psychometric properties
Petry	2009a	To evaluate different dimensions of QoL of people with PIMD and the association between their QoL and personal and setting characteristics	<i>n</i> = 49, mean age 23.7 year	Quantitative (questionnaires)	Six subscales, based on five domains of Felce and Perry	QoL-PMD: questionnaire	Parent, direct staff, indirect staff	NA
Petry	2009b	To examine the possibilities for item reduction and the psychometric properties of the QoL-PMD	<i>n</i> = 49, mean age 23.7 year	Quantitative (further development of questionnaire)	NA	NA	NA	Internal consistency, construct validity
Petry	2010	To evaluate the psychometric properties of the Mood, Interest and Pleasure Questionnaire	<i>n</i> = 360, age >18	Quantitative (questionnaires, behaviour checklist)	Mood, interest and pleasure	MIPQ: questionnaire	Direct support staff	Test-retest, inter-rater reliability
Vos	2010a	To measure a non-interpretative measure of subjective wellbeing, that is, how the respiratory, cardiovascular and electro dermal response system associate with the valence dimension of emotion	<i>n</i> = 3, age 23, 23, 52	Mixed method (video observations, physiological measurements) ^a	Emotions (physiological)	Physiological measure and behavioural observations, video tape	Direct support staff and researcher (unfamiliar)	NA
Vos	2010b	To explore the client and service characteristics contributing the subjective wellbeing of persons with severe and profound ID measured by the Mood, Interest and Pleasure Questionnaire	<i>n</i> = 360, age >18	Quantitative (questionnaires)	Mood, interest and pleasure	MIPQ: questionnaire	Direct support staff	NA
Vos	2013	To examine the relationship between mood and emotions in people with severe and profound intellectual disabilities	<i>n</i> = 27, mean age 34.5	Mixed method (video observations, questionnaires)	Mood and emotion	Experiment using behavioural observations, video tapes and MIPQ: questionnaire	Two direct support staff members and independent researcher (unfamiliar)	NA
Bigby	2014	To explore the conceptualisation of good outcomes and support in terms of QoL and staff practices	<i>n</i> = 37, age >18	Qualitative (participant observations)	Eight domains of Schalock	Qualitative indicators extracted from participant observation	Author and research fellow	NA
Verdugo	2014	To provide reliability and validity evidence of the San Martin Scale	<i>n</i> = 1770 ^b , age >16	Quantitative (questionnaires)	Eight domains of Schalock	San Martin Scale: questionnaire	Parent, family or direct support staff	Reliability and construct validity
Gomez	2015	To contribute to the knowledge and understanding of QoL by developing a set of QoL indicators	NA	Qualitative (Delphi study)	Eight domains of Schalock	NA	NA	NA

Note: NA: not applicable; PIMD: Profound Intellectual and Multiple Disabilities; QoL: Quality of Life.

^aMixed method design but very small sample

^bMost participants had PIMD

Table 2. QoL domains used by different authors.

Author	Domains
Schallock	Emotional wellbeing, interpersonal relationship, material wellbeing, personal development, physical wellbeing, self-determination, social inclusion, rights
Felce and Perry	Physical wellbeing, material wellbeing, social wellbeing, development and activity, emotional wellbeing
André	Psychological conditions, physical conditions, familial context, abilities to communicate, abilities to stay alone
Ouellette	Health services, family guardianship, income maintenance, education training and employment, housing and safety, transportation, social and recreational services, religious/cultural supports, case management, aesthetics, advocacy, counselling

both objective and subjective components. The questionnaire, filled in by an interviewer, was based on questions posed to a family member, friend or staff member. The QoL-PMD is used to exclusively measure objective components of QoL (Petry et al., 2009a). This questionnaire is completed by a parent, direct staff and indirect staff. As the authors noted (Petry et al., 2010; Ross & Oliver, 2003; Vos et al., 2010a, 2013), the MIPQ only measures subjective components and is filled in by direct support staff. However, the other questionnaires, the QUALIN, based on interviews with parents (André, Hodgkinson, Bérard, & des Portes, 2007) and the San Martin Scale, filled in by parents, family or direct support staff (Verdugo, Gómez, Arias, Navas, & Schallock, 2014), did not distinguish between subjective and objective components. The development of the QoL-PMD was based on interviews with parents and professionals (Petry et al., 2005) and on a Delphi study (Petry et al., 2007a), in which experts selected items based on the domains of Felce and Perry (1995). Most questionnaires were based on a combination of several domains (see Table 2).

The Life-Satisfaction-Matrix (LSM, Lyons, 2005) combined participant observations and semi-structured interviews, which assessed the subjective component “satisfaction with life.” The assessors were the most familiar communication partner, the next most familiar communication partner and one unfamiliar partner. The LSM is based on four assumptions for persons with PMD (Lyons, 2005, p. 767):

- (a) for persons with PMD, life satisfaction is improved when more time is spent on preferred activities and less on non-preferred activities;
- (b) individuals with PMD express their inner states through consistent behavioural repertoires;
- (c) these behavioural repertoires can be discerned by familiar others, and validated by an independent other;
- (d) an individual’s routine daily activity preferences can be determined from their affective behavioural repertoires.

Other methods and assessors. To determine subjective components of QoL, most studies used behavioural observations (Favell et al., 1996; Green & Reid, 1996; Petry & Maes, 2006; Vos et al., 2013). Green and Reid (1996) and Favell et al. (1996), for example, selected observable responses (including facial responses such as smiling) to define happiness. This behaviour was observed by supervisory staff and unfamiliar observers. In the study of Petry and Maes (2006), staff members identified expressions of pleasure and displeasure and found out that persons with PIMD especially used sounds and facial expressions to express pleasure and displeasure and positive and negative moods. In three qualitative studies (Bigby et al., 2014; Hartnett et al., 2008; Petry et al., 2007b), in-depth interviews with parents and staff and participant observation were used. Through in-depth interviews, Petry et al. (2007b) examined which support characteristics were associated with QoL in persons with PIMD. Characteristics with regard to the structure of the setting, such as group size and staff turnover, and with regard to the organisation of the support, such as interdisciplinary, played a central role.

Methodological pluralism and triangulation of assessors. Answering the question which instruments/methods were used and who determined QoL, we found that Petry and Maes (2006) used methodological pluralism and assessor triangulation. They used several methods to determine QoL in persons with PIMD, such as behavioural observations and questionnaires (methodological pluralism). Triangulation of assessors means determining QoL in persons with PIMD by two familiar observers and a third unfamiliar observer. Although parents and proxies mostly were the assessors of QoL, in five articles their role was mentioned as a limitation (Green & Reid, 1996; Hartnett et al., 2008; Petry et al., 2007b, 2009a; Verdugo et al., 2014). These studies referred to research on other groups, in which proxy outcomes differed from self-assessments (Cummins, 2002; Heal & Sigelman, 1996; Stancliffe, 1995).

In their study, Petry and Maes (2006) used triangulation in the following way: first, staff members identified expressions of pleasure and displeasure, and then parents and direct support staff filled in a questionnaire about how the person expressed these. An independent researcher then coded behavioural signals. Vos et al. (2013) used two members of the direct support staff to identify behaviour expressing both positive and negative emotions (affective profile), followed by an independent researcher coding the emotions. For an overview of persons determining QoL in persons with PIMD, see Table 1.

Discussion

In line with the findings of Petry and Maes (2009) in their overview chapter, we found no direct answer to our first research question on which concepts were used. We did find that most authors referred to the theory of Schalock and the conceptualisation of the SIRG QoL, a result of a two-year consensus process with a panel of experts (Schalock et al., 2002). We found several combinations of domains as well as references to objective and subjective components. However, of the four elements of the SIRG conceptualisation described in the articles, one was not explored in the included studies, namely the one stating that QoL is enhanced by self-determination, resources, purpose in life and a sense of belonging.

However, in persons with PIMD, self-determination and purpose in life are concepts that can raise questions due to their developmental level ($IQ < 20$). If someone's IQ is low, does this mean that their QoL is low? The answer to this question is of moral importance because it implies a devaluing of the lives of persons with PIMD (cf. Vehmas, 1999; cf. Vehmas & Watson, 2016). On the other hand, it may be worth examining in clinical practice and in research what self-determination, purpose in life and being a person (could) mean in everyday life of persons with PIMD. Reinders (2008) did this from a theological and anthropological point of view. In this vein, Petry and Maes (2009) described self-determination in relation to having choices and control over activities and environment. Furthermore, Vlaskamp (1993) developed an educational program for children with PIMD in which the assumptions were that persons with PIMD had an influence on their environment and were able to develop meaningful relationships with others.

Another issue in the included articles and in the overview of Petry and Maes (2009) was the distinction between objective and subjective components of QoL. However, the included studies used different distinctions, which is confusing. Vos et al. (2010a, 2013) included emotions in the subjective components of QoL, whereas the QoL-PMD includes emotional well-being in the objective components (Petry et al., 2005, 2007a, 2009a).

Furthermore, in order to measure subjective well-being, such as pleasure or happiness, researchers often used assumptions. For example, Lyons (2005) assumed that preferred activities improve life satisfaction and therefore QoL. However, we doubt the adequacy of this assumption because persons with PIMD may behave in a stereotypical way and prefer a certain behaviour (Poppes, van der Putten, & Vlaskamp, 2010), like playing

with (their own) hands or compulsively needing a specific toy. Although they seem to prefer this behaviour, it may prevent them from gaining (new) experiences that could improve their QoL (Poppes et al., 2010). Moreover, stereotypical "preferred" behaviour (such as self-injury) could indicate a low QoL. Another example is the assumption in Green and Reid (1996) that smiling reflects happiness. We find that this assumption needs more reflection. In fact, smiling can be a side effect of an epileptic seizure with no relationship to happiness. Additionally, laughing and smiling can be syndrome related, as with Angelman Syndrome (Adams & Oliver, 2011). If even parents or direct staff are unsure how they should interpret the behaviour of the person with PIMD, this may be even more difficult for distanced observers.

Based upon the conclusions of Petry and Maes (2009), we agree that assessing QoL in persons with PIMD is a challenge for researchers as well as caregivers. We think that one solution to this challenge may be an approach that recognises the importance of interpretation. For example, Goode and Hogg (1994) saw gaining information on QoL in persons with PIMD as an art or craft using careful judgment combined with empathy. They articulated that information can neither be easily operationalised nor described in a written document. Such an approach recognises the value of QoL in persons with PIMD without trying to objectify it. The importance of interpretation is also recognised in narrative approaches (Meininger, 2005). Methods like these should be developed further in future studies.

Another issue is triangulation and the use of proxies. When determining QoL in persons with PIMD, researchers and proxies should be critical about their own assumptions and interpretations of behaviour. In this regard, it is worth referring to Petry and Maes (2006) who aimed at methodological pluralism and triangulation to confront these difficulties. We found that triangulation was used by other researchers as well. In most studies, we found that parents or direct support staff assessed or determined QoL in persons with PIMD, sometimes as one of the partners in triangulation. Some authors argued that the person who is closest to the person with PIMD should determine his or her QoL (André et al., 2007; Ouellette-Kuntz et al., 1994), whereas many studies consider using a proxy a limitation. These different opinions on the use of proxies (such as parents) require attention, especially because parents play a significant role in decision-making (Zaal-Schuller et al., 2016). More in-depth research on the role of parents and proxies in assessing QoL when making difficult, life-altering decisions is necessary.

In accordance with the SIRG QoL, the concepts found in the included articles were not only used to describe the QoL of persons with PIMD, but they were also used with the strategic aim to be a change agent, with the main aim to improve quality outcomes (Lyons, 2010). This is confirmed by Petry and Maes (2009) in their overview chapter of QoL in persons with PIMD (Petry & Maes, 2009). According to them, the main purpose of measuring QoL is to improve QoL by detecting circumstances that decrease it and make changes based upon those findings. Therefore, they consider the use of the concept of QoL in end-of-life decisions very undesirable. “By adopting this value as a central tenet in QoL measurement, the application of QoL in ethical decisions on life and death or in refusing to treat certain people is discarded as an abuse” (Petry & Maes, 2009, p. 26). However, as recent research shows, QoL is certainly related to the decision-making processes in persons with PIMD, including end-of-life decisions (Zaal-Schuller et al., 2016).

The strengths of this study are its systematic research and explicit findings of the different uses and views on concepts in research on QoL of persons with PIMD. A limitation of our study is that the included studies were performed mostly in Western countries, for example, in Belgium and the Netherlands, which may limit the generalisability of the findings and requires further research in other contexts.

Conclusion

This review provides insight into several components of QoL in persons with PIMD and in the tools used to assess them, questionnaires and behavioural observations in particular. Methodological pluralism and triangulation of assessors with varying closeness in relation to the person with PIMD are often considered good assessment elements. However, we questioned some elements of the conceptualisation including the following: the difference between objective and subjective QoL, as well as underlying assumptions of behavioural observations. Instead of trying to objectify QoL in persons with PIMD, we argue that the existence of interpretation is recognised.

Research on QoL in persons with PIMD remains a challenge because of their incapability of expressing themselves verbally. To confront this challenge, in their overview chapter Petry and Maes (2009) argued for more valid and reliable research methods to measure QoL in persons with PIMD. We think that a narrative approach, which recognises both the role of interpretation and the storied nature of who a person is, can be

helpful in describing QoL in persons with PIMD, especially when we extend QoL to medical decisions in persons with PIMD. In these decisions, the role of parents is essential.

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