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Quality of life assessment in people with intellectual disability
An ethical or methodological issue?

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Abstract

Health-related quality of life is one of the outcomes proposed today for assessing the effectiveness of therapeutic interventions, especially in conditions with no medical cure and in which it is expected that the healthcare interventions will have an impact on the way people live. In the case of people with intellectual disability, there is controversy not only about the assessment of quality of life as an outcome, but also over the ethical and methodological considerations involved in its use. This paper addresses the ethical and methodological issues of including health-related quality of life as a clinical outcome in people with intellectual disability. **(Acta Med Colomb 2022; 47. DOI: https://doi.org/10.36104/amc.2022.2019).**

Key words: *intellectual disability, health-related quality of life, effectiveness, patient-centered outcomes*.

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Introduction

The assessment of health-related quality of life (HRQOL) has received considerable attention since the 80s. Some controversial points have been raised in the discussion including, first, the importance of HRQOL as an outcome of therapeutic interventions in people with intellectual disability (ID), and second, whether the HRQOL assessment should be done from the perspective of people with this condition or from the caregivers' perspective (1). This essay supports the HRQOL evaluation as an outcome of therapeutic health interventions from the perspective of people with ID. To defend this position, both controversies are addressed from ethical and methodological perspectives.

Undoubtedly, HRQOL is one of the main goals of current medicine and public health policies. In addition, there is growing interest in the topic among the scientific community, given that more than 2,000 HRQOL-related articles are published every year in medical journals (1, 2). But where is the term "quality of life" (QOL), as applied to the health sciences, derived from?

The QOL construct has commonly been associated with a clinical term, but is really a philosophical and ethical concept related to goodness, charity, brotherhood and happiness, among others. Goodness is an Aristotelian principle which should be triggered by all human actions. When we call ourselves healthcare professionals, we seek the good of those who use our services, and we abide by the ethical principle of beneficence in all our actions. Thus, according to Aristotelian ethics, it would be the set of actions which seek the human good and happiness in clinical practice. Therefore, clinical and research interventions in the ID population constitute a set of actions -in many cases institutional- which are necessary for safeguarding a good life for these people. The QOL concept is closely linked to the ultimate goal of healthcare interventions (3).

The good we healthcare professionals seek for our patients should include, first, the intention to do good, and second, that something good should be sought by the person receiving the action. In this case, the patient with ID should understand the good the healthcare professional desires to provide and, in addition, agree to accept the good that comes from the professional's interventions. Otherwise, the good would be incomprehensible. There is no other way of knowing if the user understands and wants to receive the interventions other than asking him/her.

Thus, and if the intention is to implement healthcare actions to improve the HRQOL of patients with ID, they must be aware of and in agreement with receiving these interventions. In addition, if we as professionals seek the good of the patients, and the good is linked to good living and, therefore, to quality of life, the initial conditions on which our actions intend to impact must be evaluated, especially when our decisions are motivated by objectivity. All actions

which seek the good should cause a change in this initial condition, which is why the direct evaluation of HRQOL in patients with ID is a measure of the expected effect of our healthcare interventions (3).

Positivist healthcare researchers, as staunch representatives of the biomedical model, assert that, in fact, medicine has impacted people's QOL through the objective results of rigorous scientific research. As proof, they refer to vaccines and complex pharmacological treatments which may save patients' lives. This would be precisely the contribution of the researchers and medicine to people's QOL. But the evaluation of HRQOL as a healthcare outcome deals with subtleties and complexities in people's lives which cannot be addressed with the objectivity pursued by the positivist paradigm (4).

We must understand that the at times indiscriminate application of new healthcare technologies, with the capacity of prolonging life at any cost, the complicated decision between quantity versus quality of life, and the ethical dilemma of distributing economic resources among populations with different health problems, have historically ignored the patients' opinions. This is something which the HRQOL incorporates, and therefore it has been classified as a "patient reported" outcome measure (PROM) (5). As an outcome measure, HRQOL not only seeks to determine the patients' preference regarding their health conditions, but also regarding the impact which treatment has on other aspects of their lives.

It is increasingly recognized that decision making in the healthcare sector must take into account the users' perception, without losing sight of objectivity. This perception should be supported on profound scientific empirical evidence which takes into consideration not only the classical quantitative indicators of morbidity, mortality and costs, but also qualitative indicators which express the impact on the patients' quality of life and satisfaction. The traditional biomedical model excludes the fact that, in most diseases, the health status is profoundly influenced by the state of mind, the ability to carry out activities of daily living independently, and social support, among others. Clearly, these aspects of highest importance in people's lives will have the greatest influence on their health status, and therefore should be considered in the patients' clinical evaluation (1).

Intellectual disability and the diseases that cause it are generally incurable; they present as chronic conditions in which medical treatments and rehabilitation, while considered effective, will not "cure" ID. Therefore, the measurement of the effectiveness of interventions in these patients should be mainly focused on the effect on HRQOL, as perceived by the patients and caregivers (6). The therapeutic interventions applied in this population should seek the greatest good, which is considered by Aristotelian ethics to be happiness, or the final goal, which is conceived as a life condition, not as a temporary state. Thus, actions to improve the HRQOL of a group should be evaluated within the permanence of an experience for life and not just a temporary experience in life, as are several of the outcomes we consider to be health intervention goals.

Plato also contributed to this reflection when he considered which type of good is necessary for achieving happiness. This good, according to Plato, is derived from the ethics of governing, and in academic and professional settings could be understood as the application of research-derived knowledge in serving vulnerable groups. That is, clinical research could objectively contribute to the ultimate goal: patients' happiness. Healthcare professionals and researchers, using knowledge generation as the exercise of the power to govern, can implement actions for the social good, to achieve the consumers' happiness and improve their HRQOL (7).

Another ethical argument for considering HRQOL assessment as a healthcare outcome in people with ID is the theory of ethical minimums, based on human rights, which is supported by Jürgen Habermas's theory, especially by concepts such as «deliberative democracy,» «civic confidence,» «legitimacy» and «moral duties.» Ethical minimums refer to the minimal conditions and behaviors for coexistence common to all social environments, considering basic elements on which all can agree and which make coexistence and tolerance possible (8).

The concept of deliberative democracy in Habermas's theory helps explain how the participation of vulnerable people with ID in processes related to them, such as their health, through PROM instruments (9), is an exercise which helps process their health problems better, promotes society's contribution to protecting this participation and dignifies the rights of people with ID as active citizens (10). This theory agrees with what was proposed in Article 8 of UNESCO's Universal Declaration on Bioethics and Human Rights (9), which states that "*in applying and advancing scientific knowledge, medical practice and associated technologies, human vulnerability should be taken into account. Individuals and groups of special vulnerability should be protected and the personal integrity of such individuals respected.*" *Thus, assessing HRQOL as an outcome is a means of fostering this participation and the protection of their human rights and, at the same time, is an effort towards ensuring social coexistence.*

So far, we have proposed ethical and philosophical arguments for considering HRQOL in people with ID from their own perspective. However, these ethical arguments also pose dilemmas regarding the use of HRQOL in people with ID which must be considered.

From a political perspective, HRQOL is already considered as a health outcome in therapeutic intervention cost effectiveness studies. Therefore, it is a key aspect in the consideration of the use of financial resources, which are always limited in healthcare systems. The right to health is influenced by the scarcity of resources, which means that spending must be aimed at interventions with proven effectiveness, and a reasonable cost. In this case, HRQOL

may be considered in determining interventions' "proven effectiveness" (11).

This situation poses an ethical dilemma regarding resource utilization which impacts healthcare decision making at a microlevel -the person- and at a public policy and healthcare financial resource allocation level. It poses concerns such as whether HRQOL could be used as a means to justify limiting resources for people with ID in interventions which are not cost effective, or whether healthcare interventions which promote HRQOL in people with ID could be more expensive.

In this case, we must clarify that the measurement of HRQOL can decrease costs without sacrificing user benefits, as health problems are prevented and treatments and medications are followed up, allowing the best and least costly ones to be chosen.

Some researchers who consider HRQOL to be an important outcome in this population have measured it using proxy instruments for evaluating HRQOL from the caregivers' perspective rather than the ID patients' perspective. This allows an HRQOL approach to the patient without dealing with the difficulties of measuring it directly in the patients, since people may have varying degrees of disability, which would entail measuring their ability to understand how the test and scale work and their ability to assent, to know if they really want to participate in completing an HRQOL instrument.

This situation poses another ethical dilemma in assessing HRQOL in people with ID: the tension between autonomy and beneficent paternalism. Beneficence refers to doing the patient good rather than harm, while autonomy promotes the patient's right to make decisions regarding which interventions he/she will receive. The paternalistic aspect of beneficence has led healthcare professionals to make decisions for the patients' wellbeing even without their consent. The principle of autonomy incorporated in the United States Bill of Rights covers the patients' right to know, the patients' right to consent to treatment, the right to refuse treatment, the right to confidentiality and the right to privacy.

If we choose to measure HRQOL as an outcome in people with ID from their caregivers' perspective, the principle of beneficence will prevail in considering the voice and opinion of parents and caregivers over that of the patients. In addition, a difference has been found between the caregiver's and the patient's perspective in the assessment of the patient's HRQOL, because, among other things, it is very hard for the caregiver to distinguish between his/her own quality of life and the patient's (12).

Another controversy regarding HRQOL as an outcome in this population concerns the methodological aspects and complexity of its measurement. Those who disagree with measuring HRQOL as an outcome consider that it cannot be feasibly evaluated, since the data obtained from these evaluations are imprecise and the measurement methods are vague. Thus, it could be considered a "soft outcome," with the concern that what has been held to be "hard science" may be replaced by soft outcomes (13).

To begin with, it is important to identify what science knows and recognize what it does not know, based on the quantitative paradigm's scientific method. That is, what can be learned through the scientific method, when we manage to extract a process from its environment, control all variables but one (the independent variable), and quantify the given effect through the changes in the dependent variable or the outcome (14).

In the quantitative scientific method, which is perhaps the most frequently used method in the health sciences, the interpretation of the results depends on the success in implementing the process, the selection of the dependent variables, the instruments and operative characteristics, the validity and reliability of the results obtained, and the characteristics of the population. The more satisfactorily these criteria are met, the narrower the confidence intervals of the results obtained for these variables, to indicate if an intervention is or is not effective. These are the benefits of experiments and what they can tell us about the patient but, at the same time, these limited contributions of the scientific method to the understanding of a health problem are heavily criticized when the results are extrapolated to a real, less controlled context (15).

In the clinical setting, a treatment will not be effective if at least two criteria are not met. First, the treatment must be able to be administered to patients in the real world, with all its limitations. Second, there must be outcome measurements to quantify the treatment's effect. These outcome measurements must be understandable and relevant for the patients, whose perspective is emotional and personal, as opposed to the outcome measurements of interest to the professionals from a positivist paradigm. Thus, the measurement of HRQOL as an outcome would lack rigor.

Favoring the patient's perspective in the healthcare research setting, many countries have actively committed to the public participation of patients; for example, in our Colombian context, patients' participation in the creation of clinical practice guidelines and evidence-based recommendations (16). In these participation scenarios, patients can determine their priorities and healthcare intervention needs both during their development as well as their evaluation, and their participation improves the way in which research is prioritized, communicated and used (17).

The English National Health Service (NHS) asserts that all countries should contribute to encouraging the public participation of patients, especially those who face the greatest health disadvantages and poorest health outcomes. People with ID are known to face disadvantages and have worse health outcomes and greater mortality compared with the general population (18).

This fostering of the public participation of people with ID has been termed "inclusive research." To be considered inclusive, it must have five characteristics: the research

problem should be a priority for people with ID; the research should foster the interests of people with disabilities; the research should be carried out in collaboration with people who have the condition; people with ID must be able to participate in all phases of the research; and the research question, process and results should be accessible to people with ID (19).

Although inclusive research is more expensive and takes more time to carry out, its contributions to knowledge have been reported, especially in settings in which people with ID can make a different contribution than their caregivers and when their participation contributes towards improving their own HRQOL (19).

The participation of people with ID presents some methodological challenges which, in turn, become arguments against, that must be evaluated. Albeit, this poses the need to develop exact and precise instruments for measuring HRQOL. To achieve this goal, the investigator would have to accept that, due to the characteristics of the outcome and population, precision would indeed be sacrificed. However, the benefit would lie in using an outcome measure which includes the patient's perspective, with less influence from the biomedical model we have been using, and which would bring us a little closer to the human aspect, to the complicated task of seeing others as a "whole," from a more constructivist paradigm, fostering the transformation of the healthcare systems' capacity to deal with the issues of greatest concern for the patients (20).

There is also concern in the scientific community regarding how changeable and personal the HRQOL construct can be, even in the same person. That is, the important dimensions of the HRQOL for a given person may change over time, according to their health condition or the presence of comorbidities. This is a concern when considering whether it is really possible to create instruments from which such a complex and individual construct may be generalized to the population, which once again raises ethical and moral doubts about whether we can defend an HRQOL model for the ID population (21).

In light of these concerns, apparently simplistic arguments could be proposed for these deep philosophical uncertainties raised by the definition of the QOL construct, which is undeniably subjective. The first argument is that when proposing a definition of this construct, the standpoint of the groups or categories of people should be considered, more than the truths which individual people may contribute. In these cases, the HRQOL evaluation tools should approximate the experiences of groups or populations, which may be defined as those who share a particular health condition (for example, the HRQOL of people living with cancer); or groups who share given services or healthcare areas, such as HRQOL in intensive care units; or those who have the shared characteristic of a therapeutic intervention, such as HRQOL evaluation in patients treated with chemotherapy (1, 22).

This would be the same principle that is applied in clinical studies, which seek to have a representative sample from which to infer the result of a variable in a given population, without attempting to explain what occurs to a particular person.

The second argument is that HRQOL evaluations in health care should not be concerned with absolute judgements about what is a good or bad HRQOL, but rather with aspects which help to distinguish mainly between one or another specific circumstance, or between one intervention or another. The HRQOL evaluation of a group of people is not intended for making value judgements or comparing the group's HRQOL to a pattern or model of a good HRQOL. On the contrary, it seeks to explore whether a group of people reports more favorable experiences in response to a given circumstance, which may be a therapeutic intervention. That is, if in the dimensions which are important for this group, the patients note a difference between one intervention and another.

In conclusion, if the goal of health care is to increase the wellbeing of people with ID and cause impacts that cut across all their lives, not just the pathophysiological aspects, HRQOL measurements must undoubtedly be integrated with the measurement of hard quantitative outcomes so that, together, they can guide the diagnosis, treatment and care. The inclusion of HRQOL places us within a constructivist paradigm in which the patients' reality may be interpreted from their valuable perspective, their autonomy is promoted, and the therapeutic accomplishments garner a more human meaning.

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