

Measuring quality of life in dysphonic patients: a systematic review of content development in patient-reported outcomes measures

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The purpose of the presented paper was to review the patient reported outcome measure protocols of treatment results used in dysphonic population to understand whether the used procedures contemplate the development standards of contents and psychometric evaluation. A systematic review was performed in papers indexed in Medline, Cumulative Index to Nursing & Allied Health and, Health and Psychosocial Instruments, using as keywords “voice”, “dysphonia”, “quality of life”, “instrument”, “scale”, “score”, “research instrument”, “inventory” and, “patient reported outcome measures”.

Voice disorders occur in 3%-9% of the population and have profound effects in Quality of Life (QoL). To quantify this impact, as well as evaluate patient outcomes and guide informed therapeutic decisions, numerous patients reported outcome measures has been developed. These questionnaires have gained popularity in both the clinical and research milieus. However, the aspects related to the development of a self-evaluation questionnaire receive more attention only in the past 15 years, resulting in SAC (Scientific Advisory Committee) report, of Medical Outcome Trust (2002).

The instrument development involves three stages, as the first stage the development of conceptual model and questionnaires items, the second stage includes field-testing in a large group of patients and, the third stage is the final one which the psychometric evaluation must be performed in the definitive form of the instrument. In the first stage qualitative methods are used including patients' interviews, focus groups and literature review as primary source of information. The main aim of this stage is to comprehensively evaluate any pre-existent evaluation in contents and psychometrics tests, in order to verify the quality of measures. Whenever a developed scientific instrument and clinical significant does not exist, it is judge to have a potential area to develop a new instrument based on existing measures. Then a list of items is done and administered in a small group of patients to reduce the ambiguities in the wording, confirm appropriateness and, determine the acceptability and time required for instrument completion. In the second stage

involving field-testing in a large sample of patients, the results must allow revision and reducing items, as observing redundancies for instance. In this step, factor analysis helps to refine the items and to perceive the way they reflect attitudes and specific abilities. In third stage, the questionnaire is again applied in a large population to determine the acceptance of data quality, analyzing whether the score distribution, the internal consistency reliability, items correlation, test-retest, validity within scale, validity within comparing with others questionnaires and, the ability to detect clinically significant change after treatment. To clinicians and researchers whom search for self-evaluation protocols to dysphonic patients is important to perceive the weak and strong points of each one of the evaluated questionnaires. Although it is considered the development of the two pioneers self-evaluation protocols in voice field, VHI and V-RQOL, a new and progressive approach to evaluate patients with voice disorders, these instruments were created as an answer to an age devout to objective analysis and without major control of the adopted criteria that, in fact, were organized afterwards.

It was identified and analyzed as the self-evaluation protocols as the instruments directed to parents of patient, to measure quality of life associated to voice issues, evaluated according to the guide to development and evaluation of SAC of Medical Outcomes Trust. Nine instruments fulfilled the inclusion criteria and the quality of these questionnaires was variable, related to instrument development, as none of them fulfilled all the current recommended criteria. Of the nine questionnaires, the VoiSS was the one submitted to the most rigorous process of development. Besides, many instruments were derived to allow its application in a proxy respondent (father, spouse or caregiver) which frequently represent a failure in tempt to approach the quality of life aspects specific to target population. The development of instrument to dysphonic patient analysis is frequently flawed and the performed review suggests that the deficits in psychometrical proprieties of instruments are at least partial result of deficiencies in the development process. Suggests yet that the development of a new protocol respecting all the international standards is going to assure clinicians are using the most relevant variables to patients with voice disorders.

The systematic review was performed only in English literature and revealed nine instruments, which characteristics are pointed out as follow:

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- Voice Handicap Index – VHI: an initial pool of items was developed from case history interviews with patients with voice disorders over a 7-year period. The items were grouped into three content domains: functional (25 items), emotional (31 items), and physical (29 items). Sixty-five dysphonic patients completed the preliminary version of the VHI. They were asked to circle one of five responses on an equal-appearing five-point scale. This initial pool of items was then reduced via assessment of internal consistency reliability, gender-related responses, and redundancy of group content. In addition, if 50% of the cohort did not respond to a particular item, it was omitted from the final version. The final version was then presented to 63 adult patients as a final component of the development process.
- Voice Handicap Index 10 – VHI-10: in 2004, a different group subjected the 30-item VHI to item reduction and factor analysis, resulting in VHI-10. Questions with the largest mean differences between 159 nondysphonic subjects and 100 dysphonic patients were identified. In addition, a cohort of clinicians was asked to select the 10 most “clinically relevant” VHI items for both the assessment of voice-related handicap and responsiveness to treatment. The resulting ten items were then evaluated using pre- and post-treatment item analysis.
- Voice-Related Quality of Life – V-RQOL: the authors of the V-RQOL report that both clinician input and informal patient interviews were used in the development of the V-RQOL. No further specific information regarding the development process was identified. An original version of the questionnaire was piloted on 20 patients to evaluate content and phrasing. The revised instrument was then administered to 109 new voice-disordered patients, and 21 patients without a voice disorder. Three questions were found to have low item-to-total correlation; two were retained in the instrument due to high face validity. One additional question dealing specifically with employment-related telephone use difficulties was omitted from the final version.
- Voice Outcome Survey – VOS: the VOS was constructed by an expert panel (physicians and voice therapists) in addition to patients with vocal cord paralyses. The initial pool of items was selected based on symptom frequency and “importance.” After initial testing, a pilot questionnaire was created. Additional items were inserted by the authors to evaluate patient limitations in certain daily, social, or work-related activities that require vocalization. The piloting process involved the addition of two questions identified to be particularly relevant to patients with unilateral vocal fold paralysis: straining while speaking and aspiration. Following psychometric analysis, two items were removed from the pilot instrument for inadequate test-retest reliability.
- Voice Activity and Participation Profile – VAPP: interviews of 45 dysphonic subjects as well as the input from 10 speech pathologists were utilized to develop the Voice Activity and Participation Profile. Subjects were asked to list on-going life situations that they felt were adversely affected by their dysphonia. To assess content validity, three groups (speech language pathologists, speech language pathology students, and voice patients) commented on how well the questions described dysphonia-related issues, as well as how clearly the items were presented. Forty subjects with dysphonia and 40 control subjects piloted the initial questionnaire. The instrument was then divided into five domains: self-perceived voice problems, social, emotional, work, and communication issues.
- Voice Symptom Scale – VoiSS: the development of the VoiSS involved three steps. First, 133 patients prospectively reported 467 difficulties associated with their voice disorder. A preliminary instrument was developed based on these issues. The pilot questionnaire was then administered to 168 patients with dysphonia. Item reduction was then performed using principal component analysis to detect latent items. In the final step, 180 new patients with dysphonia completed the refined VoiSS questionnaire. Content validity was determined using 13 items from the VHI. This process resulted in a 43-item instrument, further refined the psychometric structure of the VoiSS resulting in the final 30-item instrument.
- Pediatric Voice Outcome Survey – pVOS: this is a four-item instrument derived from the VOS to allow for parent proxy administration. Principal component and factor analysis was performed to evaluate the internal structure and the design of the instrument with reference to its application to the pediatric population. One hundred eight caregivers of children or adolescents requiring tracheotomies participated in the study and completed the instrument. Reliability testing and factor analysis supported the overall structure with one swallowing related omitted. To broaden the applicability of the PVOS to children with other vocal disorders, the instrument was included in another study of 385 parents of children aged 2–18 years with a range of conditions, including velopharyngeal insufficiency; subglottic stenosis; vocal cord nodules; reflux laryngitis; vocal cord paralysis; obstructive sleep apnea; adenotonsillar hypertrophy; otitis media; and sinus disorder.
- Pediatric Voice-Related Quality of Life – pV-RQOL: the pV-RQOL was adapted from the V-RQOL instrument to allow parent proxy administration. The ten-item pV-RQOL was jointly administered along with the PVOS to 104 caregivers of children aged 2-18 years with a variety of otolaryngological problems including velopharyngeal insufficiency, dysphonia, adenotonsillar hypertrophy, otitis media, and sinus disorders. This preliminary study suggested that the pV-RQOL was a valid instrument that correlated highly to the PVOS.
- Pediatric Voice Handicap Index – pVHI: the pVHI was adapted from the VHI. Primarily, the language was altered to reflect parental responses about the child’s voice disorder. Any questions deemed irrelevant to the pediatric population by the authors were eliminated, resulting in a 23-item instrument. The pVHI was administered in conjunction with another questionnaire consisting of 10 open-ended questions regarding the impact of the child’s voice quality on overall communication, development, education, social life, and family circumstances. Psychometric

analysis revealed that the pVHI was a valid and reliable instrument.

As main comment is worth to point out that VHI was the first questionnaire introduced, in 1997, to specifically evaluate the impact of dysphonia in quality of life during a period in which the tendency was to use heavy instrumentation in vocal analysis. Since then, many others instruments were described in literature fulfilling a higher or lower number of essential prerequisites. The mainly deficiencies are related to questionnaire development, especially in the pool of initial items and in the reducing of items process. These deficits, even in the considered popular instruments, may be the source of limitations in psychometrical validity and potentially confuse the observed clinical data.

By the time the instruments psychometrical validity was already investigated, the focus of this present article was to analyze the development of its contents. The development of contents of a test requires the pool of items to be generated by patients' interview, literature analysis and, experts opinion. From the nine analyzed questionnaires, only five used interviews with patients; two of them were developed by retrospectively analyzed anamnesis and, three of them used as primary and only source the experts' opinion.

The data about the numbers of interviewed patients in the beginning of the process was generally omitted from publications, as the VoiSS the only instrument with this data. The most part of instruments used few interviews and did not discuss properly the heterogeneity of dysphonic population. Regarding the items reduction, the VHI, V-RQOL, VOS and, VoiSS appear to fulfill the criteria. From the rest, five used the experts opinion as a criteria to reduce the items. These deficits probably are the reason for questionable psychometric measures. An additional aspect, it may have a tendency to modify the instruments that already exists to administer in other patient population. The manipulation of instruments to other populations and even to be used to proxy respondents, violate the fundamental criteria to develop itself. This com-

promise all the pediatric questionnaires, developed from the adult version. Pediatric quality of life questionnaires must necessarily include the important domains to children and the questionnaires developed to adults may flaw in this evaluation of relevant aspects to pediatric population. Besides, it is not clear how these instruments capture the quality of life aspects to this population.

Another aspect is the language modification has been used to adapt the instrument to a specific subgroup of dysphonic population, as singers and spouses. To complete, a good number of questionnaires had been translated to other languages without properly language validation. Finally, none of these protocols used contemporary psychometrical methods, such as RASCH method and Item Response Theory, which is a statistics modeling used in psychometrical measures, mainly in ability and knowledge evaluation field. These approaches to develop scales and reducing items increases the clinical utility, because offers an individual attention to patient and evaluate important aspects in quality of life lived by dysphonic people. Although the use of RASCH analysis and Item Response Theory has been suggested to dysphonic population, it seems to have little progress in this direction.

To sum up, although clinicians and researchers had improved their capacities to detect an illness progression and the treatment efficacy with the use of instruments that may allow guided therapeutic decision, unfortunately the result of this review revealed none of the instruments of quality of life current used in voice field contemplates the described criteria as essential to its development. This article offers a potential impetus to create a new metric that rigorous respect the recommendations to develop instruments with the incorporation of new psychometric methods to improve clinical utility. Must be clarified this do not suggest the elimination of vocal self-evaluation measures, since it offer a unique dimension in patient diagnosis and in determining clinical approach, but to understand the limitations inherent to deficiencies in the development process of these protocols.