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Validation of a Quality of Life Assessment Questionnaire for Patients with Infantile Hemangioma and Their Families – the IH-QOL-RO

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ABSTRACT

Introduction: *Despite the fact that infantile haemangioma is one of the most common benign tumour of the child, the impact of this pathology on the quality of life of patients and their families has not aroused much interest in health services research. Several tools have been proposed to date to assess this, but there is no Romanian standardised instrument yet. Taking as a model the questionnaire developed and validated in English by Chamlin* et al *(2015), we translated, culturally adapted and piloted this instrument into Romanian. The questionnaire was administered during two years, between August 2019 and August 2021, to the parents of children with a diagnosis of infantile hemangioma who attended "M. S. Curie" Children's Emergency Hospital, Bucharest, Romania. Inclusion criteria were the diagnosis of infantile hemangioma and children under the age of 24 months. Other comorbidities which may have caused other health impairments were considered as exclusion criteria. Response rate was 100% for all items in the questionnaire. A total of 112 family respondents were included for analysis. Classic psychometric tests were used.*

Results: *Based on the 29 standardized original items, the four scales have Cronbach-alpha values ranging from 0.489 (CSI), 0.609 (PSF), 0.689 (PEF) to 0.719 (CPS). The proposed final Romanian version includes 26 standardised items. The Cronbach-alpha values improve marginally: 0.63 (PSF), 0.67 (CSI), 0.72 (PEF) and 0.733 (0.78) (CPS).*

Conclusion: *We propose the 21-item scale of the IH-QOL-RO as the Romanian version of the IH-QOL®. The instrument has been culturally adapted and is ready to use in paediatric clinics. We recommend the use of IH-QOL-RO in a longitudinal study design as a measure of health-related quality of life and to complete the classical set of psychometric tests with the 48-hour test-retest reliability.*

Keywords: infantile hemangioma, quality of life, questionnaire, validation.

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INTRODUCTION

Infantile hemangioma (IH) is the most common paediatric vascular tumour with an estimated incidence of 4%, though the reported incidence ranges from 2-10% (1-4). This difference in incidence can be explained by the fact that IH can occur after a child's discharge from a health unit and the registry data may lag registrations or remain missing (5). Further challenges in determining the true incidence of IH include non-attendance for medical review and referral bias. We must also take into account the difference in diagnosis and reporting, especially since until 1982 there was no clear classification of these vascular anomalies (6).

In recent years there has been an increase in the incidence of childhood hemangiomas, which was partly explained by an increase in the number of premature babies and low birth weight babies (two of the factors mentioned in the literature as risk factors) (1).

Most infantile hemangiomas are not present at birth, but a proliferative phase commonly begins during the first month of life, and at the age of five months, 80% of patients reach complete growth. In some cases, this growth can continue until the age of one year and in exceptional cases up to two years. The third and final phase (involution phase) follows, with a slow and gradual decrease in size, which is usually completed within 10 to 12 years of life (7).

Most of the hemangiomas are benign; however, there are also cases in which the proliferation phase is accentuated and complications may occur, including ulceration, bleeding, visual impairment or airways obstruction if periorbital region or upper airways are involved respectively, scars and disfigurement. The occurrence of complications depends on the size and location of the haemangiomas (8). As a result of all leading evidence-base, it is essential to consider how this vascular anomaly affects the lives of children and their families.

So far, the published literature showed that location, size and appearance of hemangiomas are three important features that influence the attitudes towards this condition, making them somehow comparable to other dermatological conditions.

Dermatological conditions often remain a hidden affliction and the impact of skin diseases on the quality of life (QoL) of patients and their families is often underestimated. For most of the patients with dermatological diseases, these health conditions are not not very painful and rarely life-threatening, which is one of the reasons why they may have received less attention in terms of impact assessment.

Unlike many other serious illnesses which can be entirely 'hidden' or 'out of sight', haemangiomas are visible, 'in plain sight', for patients on a daily basis. Such self-awareness can have a very profound psychological and social impact and patients often learn how to hide these, so that they could escape unnoticed.

A comprehensive assessment of the QoL of these patients and their families is an essential step in the clinical and general management of the condition at the individual, family and societal levels. Although many studies have explored the QoL of patients with dermatological problems, only few of them evaluated the family impact of those dermatological conditions and even fewer investigated their impact on families of children with haemangiomas. It is therefore important to extensively assess how these conditions affect the lives of children and their families.

Given the particularity of dermatological conditions in terms of psychological and social impact of changes in appearance, it is necessary to adapt or design assessment tools which take into account QoL criteria. Generic measures or profiles may overlook sensitive social and psychological aspects. Generic measures include the ITQOL and ITQOL-SF47, instruments which measure items which generate a health profile (10). ITQOL was validated in other countries such as China (11), whereas part of the measure has also been included in a respiratory disease specific QoL measure which was validated in the Netherlands (12).

This article is a follow-up of an already published paper (9) and describes the Romanian validation of a infant haemangioma disease-specific questionnaire (IH-QOL) developed by Chamlin *et al* in 2015, (8) with the aim to evaluate the quality of life of paediatric haemangioma patients and their families.

MATERIAL AND METHODS

Study objective: to validate the translated adaptation of IH QoL and test its properties with clas-

sic psychometric testing criteria, including reliability, validity and responsiveness, in the Romanian population.

Study design: observational, cross-sectional study, carried out between August 2019 and August 2021, on 112 children and their families who attended "M. S. Curie" Emergency Hospital for Children, Bucharest, Romania.

The questionnaire was administered to all children's legal guardians. Children with IH were at their first hospital attendance. The reason of presentation was either to confirm the diagnosis made by another (family) doctor or to confirm the diagnosis as well as to enrol the child in treatment. The following inclusion criteria were used: legal guardians of IH children less than 24 months of age who had no other significant comorbidities which could have led to measurement errors in either of the four-scale self-assessed QoL measurements. We used the translated and Romanian adapted IH-QOL. The pilot project was published in a previous paper (9).

The piloted questionnaire used 28 items out of the original 29 items, which were grouped under the same four scales: physical health (CPS five items), the social function of the child (CSI four items), parents' emotional health (PEF 10 items) and the social function of parents (PSF 10 items) (8). We dropped one item (item 30), which was culturally too sensitive to be included, as identified by the research team. This was flagged up during the pilot stage (9).

Responders chose answers for items along a five-point Likert scale ranging from 1 to 5, where 1 = strongly disagree, 2 = disagree, 3 = neither agree nor disagree, 4 = agree, and 5 = strongly

TABLE 1. Psychometric testing and criteria used at field test evaluation of IH-QOL-RO 28-item questionnaire (13-15)

Test	Definition; concepts	Criteria tested
Acceptability	Quality of data assessed by completeness of data and floor and ceiling effects	Proportion of missing data for scales (<10%)
Test of scaling assumptions	Evidence that an item belongs in its own scale and not another scale (item convergent and discriminant validity for each of the four scales)	*Scaling success/failure* (item does/ does not correlate significantly higher with own scale than other scale) *and probable scaling success/failure* (item does/does not correlate more highly, *but not significantly*, with own scale than other scales)
Reliability		
Internal consistency	Extent to which items in a scale measure the same construct (such as homogeneity of the scale); assessed by Cronbach's alpha, item-total correlations, and value of alpha if an item is deleted from a scale	Cronbach's alpha for scales >0.70 • Item-total correlations >0.30 • Value of alpha if an item is deleted from scale should not 'substantially increase'
Test-retest	Repeatability – the degree to which scores are consistent over time (when no change is expected) (48 hours)	Intraclass correlation coefficient >0.70 (future field test)
Construct validity		
Within scale analyses	Evidence that each scale measures a single construct and items can be combined to form scales; assessed on the basis of evidence of good internal consistency, factor analysis, and correlations between scale scores	Internal consistency (Cronbach's alpha >0.70) • Principle Axis Factor Analysis (factor loadings ≥0.30); • Moderate intercorrelations between scale scores and evidence of unique reliable variance (reliability coefficients with values greater than the intercorrelations between scales)
Analyses against external criteria: convergent and discriminant validity	Evidence that scales are correlated with other measures of the same or similar construct, and not correlated with other measures of different constructs; assessed on the basis of correlations between IH-QOL-RO items and independent variables (sex)	Magnitude and direction of correlations expected to vary according to the similarity of constructs being measured: • Very low correlations (<.30) expected for independent variables, *e.g.*, sex
Analyses against external criteria	Evidence that scales differentiate between known groups; assessed by comparing IH-QOL-RO scores between groups hypothesised to differ	IH-QOL-RO scores should be significantly different for groups expected to differ, *e.g.*, different localisation of the haemangioma ($p < .05$) (third manuscript)

agree. Extreme values express the stronger attitudes. Collected data were analysed with IBM SPSS 23.0.

Taking into account that sensitive personal information was included in the instrument, the study was carried out in compliance with the ethical rules as well as the present legislation regarding the protection of personal data. Each respondent actively consented to data collection, analysis and publication of results in an aggregated form prior to questionnaire completion. The hospital board and the ethics committee were consulted and their approvals were obtained in this respect.

We used standard psychometric tests and criteria to evaluate the acceptability, reliability, validity (internal consistency), and responsiveness of the IH-QOL-RO. These are summarised in Table 1 in accordance with the literature on field test evaluation of psychometric criteria for QoL instruments, including acceptability (low missing data, good response rates), scaling assumptions (good item convergent and discriminant validity), reliability (good internal consistency) and responsiveness, for the adapted 28-item IH-QOL-RO questionnaire (13-15). A classical statistical analysis was carried out by performing a principal component analysis (PCA) with Promax with Kaizer Normalization and by performing all reliability analyses listed in Table 1, with the exception of the test-retest reliability. This is further addressed in the discussion section. Factors with variance under 5% were dropped. Factor loading of 0.4 or higher are considered significantly loaded to that factor and were retained in the scale. Discriminant analyses (external) was carried out with reporting on the Spearman RHO correlation coefficient (sex) (13, 15).

Scoring the subscales

Likert-type scale responses were used in all questionnaires: 0 = never a problem, 1 = almost never a problem, 2 = sometimes a problem, 3 = often a problem and 4 = almost always a problem. Scores of each item were next linearly transformed into a 0 to 100 scale (0 = 100, 1 = 75, 2 = 50, 3 = 25, 4 = 0) (8, 14).

The IH-QoL 29-item questionnaire can be reported as a total score (range, 0-116, where 0 represents no effect and 116 significant effects on the QoL), but subscale reporting is suggested. The subscale scores include *child physical symptoms* (range 0-16), *child social interactions* (range 0-20), *parent psychosocial functioning* (range 0-40), and *parent emotional functioning* (range 0-40).

The IH-QOL-RO 28-item questionnaire can also be reported as a total score (range, 0-84, where 0 represents no effect and 84 significant effects on QOL). We also recommend that the following subscale scores are used: CPS range 0-16, CSI range 0-12, PSF range 0-24 and PEF range 0-32. We used the same coding for a linearly transformed score (0 to 100 scale). ❑

RESULTS

Acceptability

The condition on proportion of missing data for scales to be less than 10% was observed. In fact, this condition was met on all domains, as there were no missing data for any of the items in the four scales (100% responses).

Tests of scaling assumptions

Scaling success/failure (when item does/does not correlate significantly higher with own scale than other scale) and *probable scaling success/failure* (when item does/does not correlate more highly, *but not significantly*, with own scale than other scales).

Factor loading of 0.4 or higher are considered significantly loaded to that factor and were retained in the scale. The original instrument was developed with factor loadings as follows: CPS with five items, CSI with four items, PEF with 10 items and PSF with 10 items. The IH-QOL-RO obtained loadings as follows: CPS with four items, CSI with three items, PSF with six items and PEF with eight items.

Factor analysis – Principal component analysis

Eigenvalues and variance explained

Six out of eight components were identified with variance explained from 22.47 to 5.43% at first extraction with the exploratory factor analysis. The screeplot identifies the eight components of the factor analysis (Figure 1).

Reliability

Internal consistency

Four scales/components were identified by the developers of the measure, including CPS (child physical symptoms), CSI (child social interac-

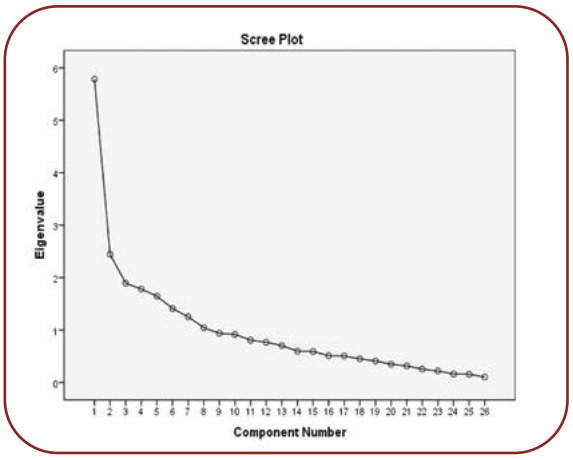

FIGURE 1. Screenplot identifying eight components of the factor analysis

tions), PSF (parental social functioning) and PEF (parental emotional functioning).

Construct validity (within scale analyses)

Evidence that each scale measured a single construct and items can be combined to form scales was assessed on the basis of evidence of good internal consistency, factor analysis, and correlations between scale scores.

Statistics are reported in Table 2 for the 21-item version of the four scales.

The following values were taken by item-total correlations from: 0.360 to 0.771 (items 1, 2, 4 and 5) for CPS scale; 0.130 to 0.496 (items 8, 14 and 24) for CSI scale; 0.150 to 0.496 (items 11, 18, 22, 23, 25 and 26) for PSF scale, and 0.203 to 0.645 (items 9,10, 12, 16, 17, 19, 20 and 21) for PEF scale.

Construct validity (analyses against external criteria): convergent and discriminant validity results for sex

This is evidence that the scale is correlated with other measures of the same or similar construct, but not also with other measures of different constructs – here, this was assessed on the basis of correlations between IH-QOL and sex (n=112). Discriminant analysis has shown that all item scores and the scale scores were correlated with sex with coefficients <0.30 as hypothesized. Results are shown in Table 3. ❑

TABLE 3. Construct validity (discriminant analysis results – sex)

Scale	Item	Spearman's RHO correlation coefficient
CPS	**Q1**	**-0.074**
CPS	**Q2**	**0.053**
	Q3	0.071
CPS	**Q4**	**-0.070**
CPS	**Q5**	**-0.010**
	Q6	-0.046
	Q7	0.100
CSI	**Q8**	**0.032**
PEF	**Q9**	**0.004**
PEF	**Q10**	**-0.045**
PSF	**Q11**	**-0.099**
PEF	**Q12**	**-0.134**
	Q13	-0.041
CSI	**Q14**	**-0.003**
	Q15	-0.015
PEF	**Q16**	**-0.052**
PEF	**Q17**	**-0.133**
PSF	**Q18**	**-0.073**
PEF	**Q19**	**-0.154**
PEF	**Q20**	**-0.002**
PEF	**Q21**	**-0.052**
PSF	**Q22**	**-0.025**
PSF	**Q23**	**-0.138**
CSI	**Q24**	**-0.018**
PSF	**Q25**	**0.010**
PSF	**Q26**	**-0.078**
	CPS (four items) (0-16)	**-0.010**
	CSI (three items) (0-12)	**-0.072**
	PSF (six items) (0-24)	**-0.190***
	PEF (eight items) (0-32)	**-0.053**
	Total score (21 items) (0-84)	**-0.089**

*The result was statistically significant at p=0.05 (two-tailed)

DISCUSSION

Values for the Romanian scales were lower than those registered with the original 29-item scales, especially with the CSI scale (0.489 compared with 0.79 in the IH-QOL). Of the four scales, CSI was the weakest but could slightly improve its coefficient after one of the three items was dropped, which would leave the

	Standardised Cronbach-alpha (improved if one item dropped)			
Scale	**CPS**	**CSI**	**PSF**	**PEF**
IH-QOL-RO	0.719 (0.732)	0.489 (0.668)	0.609 (0.629)	0.689 (0.718)
IH-QOL ® (8)	0.78	0.79	0.86	0.84
IH-QOL (14)	0.75	0.75	0.65	0.68

TABLE 2. Construct validity of the four subscales (Romanian and original version)

scale with two highly correlated items (within scale analysis).

The IH-QOL was designed to measure the predominant impact on parents with more items in the parent subscales, 20 compared to 9 for the child. The Romanian version retains 14 items from the parent subscales (6 and 8 items respectively) and seven items from the child subscales (four and three items, respectively). Although the instrument is shorter, the remaining items confer appropriate reliability, despite the fact that items may need further reviews for better definitions.

The discriminant analysis (external) showed reliability with correlation coefficients <.30 as hypothesized, including for one subscale (PSF) where the correlation coefficient showed a two-tailed p=0.05 statistical significance. This information could not be checked against the information obtained from the use of the original scale.

Test-retest reliability (48 hours) needs to be included in a future study. A few months within the period of the admnistration of the questionnaire, the World Health Organisation declared a Public Health Emergency of International Concern (PHEIC) soon after the COVID-19 pandemic, so data collection was interrupted during the lockdown period (15 March-15 May 2020).

Item total correlations among factor scores ranged from 0.130 (CSI) to 0.63 (PEF). This compares with 0.17 (child physical symptoms and child social interactions) to 0.63 (parent psychosocial functioning & parent emotional functioning) from the original study (8).

Correlations of subscales with the total score were 0.41–0.93.

The developers of the current instrument highlighted the fact that worry about location and specific complications were deliberately not included in the IH-QOL as this disease specific measure was designed to be used for all hemangioma locations (8). However, location could be evaluated if anonymously and confidentially location data were analysed in line with the culturally adapted 21-item questionnaire. For this purpose, a third manuscript is in preparation.

CONCLUSION

The Romanian version of IH-QOL (IH-QOL-RO) shows a moderate reliability but has a high acceptability. The four scales have Cronbach-alpha values based on the 28 standardized items ranging from 0.489 (CSI), 0.609 (PSF), 0.689 (PEF) to 0.719 (CPS). Factor analysis extracted 21 standardised items. Cronbach-alpha values improve but two scales still give values under 0.70: 0.629 (PSF) and 0.668 (CSI); the two remaining scales slightly improve in their Cronbach coefficients: 0.718 (PEF) and 0.732 (CPS). We propose the 21-item IH-QOL-RO version for use to measure the QoL in both children up to two years of age with haemangioma and their legal tutors. The seven item reduction from 28 to 21 confers the optimum utility of the measure. The instrument has been culturally adapted and is ready to use not only in paediatric clinics but also in follow-up studies. Test-retest relaibility (48 hours) needs to be included in a future study. ❑

Conflicts of interest: none declared.
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References

1. **Anderson KR, Schoch JJ, Lohse CM, et al.** Increasing incidence of infantile hemangiomas (IH) over the past 35 years: Correlation with decreasing gestational age at birth and birth 444 weight. *J Amer Acad Dermatol* 2016;74:120-126.
2. **Seiffert A, Schneider M, Roessler J, et al.** Incidence, Treatment Patterns, and Health Care Costs of Infantile Hemangioma: Results of a Retrospective German Database Analysis. *Pediatr Dermatol* 2017;34:450-457.
3. **Kilcline C, Frieden IJ.** Infantile hemangiomas: how common are they? A systematic review of the medical literature. *Pediatr Dermatol* 2008;25:168-173.
4. **Dickison P, Christou E, Wargon O.** A prospective study of infantile hemangiomas with a focus on incidence and risk factors. *Pediatr Dermatol* 2011;28:663-669.
5. **Rodríguez Bandera AI, Sebaratnam DF, Wargon O, Wong LF.** Infantile hemangioma. Part 1: Epidemiology, pathogenesis, clinical presentation and assessment. *J Am Acad Dermatol* 2021;85:1379-1392. doi:10.1016/j.jaad.2021.08.019.
6. **Mulliken JB, Glowacki J.** Hemangiomas and vascular malformations in infants and children: a classification based on endothelial characteristics. *Plast Reconst Surg* 1982;69:412-422.

7. **Léauté-Labréze C, Harper JI, Hoeger PH.** Infantile Haemangioma. *Lancet* 2017;390:85-94. doi: 10.1016/S0140-6736(16)00645-0.
8. **Chamlin SL, Mancini AJ, Lai JS, et al.** Development and validation of a quality-of-life instrument for infantile hemangiomas. *J Invest Dermatol* 2015;135:1533-1539. doi: 10.1038/jid.2015.18.
9. **Raicu AM, Bratu EC, Buzatu M, et al.** Infantile Haemangioma-Elaboration and Piloting a Specific Quality of Life Questionnaire. *Maedica* (Bucur) 2022;17:576-582.
10. **Landgraf JM, Vogel I, Oostenbrink R, et al.** Parent-reported health outcomes in infants/toddlers: measurement properties and clinical validity of the ITQOL-SF47. *Qual Life Res* 2013;22:635-646. doi:10.1007/s11136-012-0177-8.
11. **Volger S, Landgraf JM, Mao M, et al.** Feasibility and Psychometric Properties of the Infant Toddler Quality of Life (ITQOL) Questionnaire in a Community-Based Sample of Healthy Infants in China. *Matern Child Health J* 2018;22:702-712. doi:10.1007/s10995-018-2439-8.
12. **Raat H, Landgraf JM, Oostenbrink R, et al.** Reliability and validity of the Infant and Toddler Quality of Life Questionnaire (ITQOL) in a general population and respiratory disease sample. *Qual Life Res* 2007;16:445-460. doi:10.1007/s11136-006-9134-8.
13. **Lamping DL, Schroter S, Marquis P, et al.** The community acquired pneumonia symptom questionnaire: a new patient-based outcome measure to evaluate symptoms in patients with community-acquired pneumonia. *Chest* 2002;122:920-929.
14. **Marquis P, Fayol C, Joire JE.** Clinical validation of a quality of life questionnaire in angina pectoris patients. *European Heart Journal* 1995;16:1554-1560.
15. **Steriu A, Bratu EC, Cioran NV, Brinduse LA.** Validation of the Romanian Communication Skills Ability Scale - CSAS-RO - A Tool for Medical Schools. *J Med Educ Curric Dev* 2022;9:23821205221096375. Published 2022 May 10. doi:10.1177/23821205221096375.
16. **Wang C, Li Y, Xiang B, et al.** Quality of life in children with infantile hemangioma: a case control study. *Health Qual Life Outcomes* 2017;15:221. doi:10.1186/s12955-017-0772-z.