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Infantile Haemangioma – Elaboration and Piloting a Specific Quality of Life Questionnaire

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ABSTRACT

Background: *Infantile haemangiomas are the most common benign tumours of the child with clinical manifestations in the first two years of life, which is an additional cause of parents' concerns.*

Objective: *This study describes the first stage in elaborating a specific instrument to evaluate the quality of life of both patients with infantile haemangioma under two years of age and their parents, adapted to the reality of the Romanian context.*

Methods: *Items were generated from a literature review – from both the current generic pediatricians' instruments and specific tools in dermatology for assessing quality of life and the existing consensus among experts – as well as from a qualitative analysis of parents' concerns. The instrument was piloted on a group of patients' relatives.*

Results: *We have developed a 28-item specific infantile haemangioma quality of life questionnaire with four sub-scales to assess physical health, the social function of the child, parents' emotional health and the social function of parents. Demographic data and clinical features (meanings of symptoms and outcomes) that have an impact on the quality of life were obtained.*

Conclusion: *It is important to be able to measure and compare the quality of life of both patients with infantile haemangioma and their parents for adapting the treatment to the specific needs of patients and their family. The effectiveness of new therapeutic options which are especially useful for infants with haemangiomas can be checked by using the questionnaire as a measure of patient-reported outcome. The questionnaire developed by us was well accepted by the patients' parents.*

Keywords: quality of life, infantile haemangioma, health, questionnaire.

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INTRODUCTION

The term "quality of life" (QoL) is relatively new, being firstly used only in the second half of the twentieth century. However, the interest towards the new term is constantly growing since its first use until today. This fact is proved by the large number of publications that refer to it. The QoL means that the various aspects related to the physical and mental impact must be primarily seen in terms of their general resonance in a person's life (1, 2).

The World Health Organization (WHO) defines the QoL as "the perception of individuals about their position in life, in the context of the cultural and value system in which they live, in relation to their goals, expectations, values and concerns. The QoL incorporates physical health, psychological state, level of independence, social relationships, one's own beliefs and the relationship with environmental factors. The QoL refers to a subjective assessment that is included in the cultural, social and environmental context." (3 - 5).

It is generally agreed that QoL assessment tools should include both objective and subjective criteria.

The questionnaires that assess the QoL of patients with dermatological conditions, although schematic and easy to apply, do not include all the dimensions of childhood haemangioma, especially as it is a visible condition in the first weeks of life, when the emotional status of the family, especially of the mother, is influenced (6-14). Unlike many other serious diseases that can be completely "hidden", these conditions are sometimes carried "in plain sight" by patients every day. This can have a profound psychological and social impact that patients often learn to hide, so it goes unnoticed. That is why a correct and extensive assessment of the QoL of these patients and their families is an essential step in order to be able to formulate specific methods for addressing these conditions at both the social and individual level. Numerous studies on the QoL of dermatological patients have already been performed, but only few of them regarded these patients' families too and even fewer evaluated the impact of pediatric haemangiomas.

Therefore, it is important to make an extensive assessment of the ways in which these conditions affect the lives of patients and those close to them. Given the particularity of dermatological conditions in terms of the psychological and social impact of the change in aesthetic appearance, it is necessary to adapt the assessment tools in such a way as to take into account such criteria. General evaluation questionnaires that overlook social and psychological aspects may omit the major influences on the QoL caused by skin damage.

The difficulty of their elaboration results not only from the great diversity of the pathology of this population group, but especially from the age subgroups' different reactions to disease, the evaluation of these reactions representing a great challenge for researchers.

In 2015, Chamlin and colleagues proposed the validation of IH-QoL as the first scale in English designed to measure the impact on patients with infantile haemangioma (IH) and their parents (11). This research tool includes four categories of symptoms and signs (physical symptoms of the patient, social function of the patient, social and psychological function as well as the emotional state of the caregiver) and comprises 29 questions. It is a scale for measuring the QoL based on hypotheses developed and refined through questions addressed directly to parents by reviewing published papers and feedback from clinicians. The questionnaire was evaluated, piloted and validated. IH-QoL was designed to measure the predominant impact on parents, with more items being included in subscales for the mother (20) compared to only nine items for the child, as follows: child physical symptoms (CPS), child social interactions (CSI), parent psychosocial functioning (PSF), and parent emotional functioning (PEF) (11). There is no such tool in Romania, hence the concern for developing one adapted to the Romanian socio-cultural context.

Purpose

The present study describes the first stage in elaborating a specific instrument to evaluate the QoL of both patients with IH aged under two years and their parents, adapted to the reality of the Romanian context. The questionnaire aims to evaluate the impact of the disease on the level of social integration, fulfilment on intra-family relationships, emotional insecurity and globally

assessed happiness under the name of quality of life. ❑

MATERIALS AND METHODS

Description of the concept

Because "skin health and appearance have the ability to influence self-esteem, mental status, social interactions and overall quality of life" worldwide as well as in the Romanian society, we made a literature review to identify tools used to measure health related QoL for pediatric patients in general and particularly those with IH (12, 13).

Based on clinical experience, expert opinion, and literature review, we hypothesized that the effects of haemangiomas on the QoL of both affected infants aged under two years and their families could be grouped into four dimensions: 1) the child's physical health (CPH) (*e.g.*, pain, sleep problems); 2) the social function of the child (SFC) (*e.g.*, other children avoid him); 3) the emotional health of parents (EHP) (*e.g.*, sadness, disappointment, worry); and 4) the social function of parents (SFP) (*e.g.*, affecting social life).

Review of instruments for quality of life assessment

We used as a model for developing our specific questionnaire the generic questionnaires for assessing the QoL of paediatric patients aged under two years and their families, including PedsQL™, PedsQL™ (FIF), pedsQL™ 4.0 FIM, PedsQL™ 4.0 GCIS, for patients aged one month to 12 months, and PedsQL™ 4.0 Core Infant for those aged 13 to 24 months) (14-17). Especially the above-described IHQoL developed by Chamlin *et al* (11) has been also taken into account, and items from this instrument were selected to be included in the final questionnaire.

The Romanian language versions of the above-mentioned questionnaires were translated according to the standardized procedures and were performed in several steps: Romanian translation of items from all questionnaires, reverse translation (English language), defining the concept, selection of items, creating the questionnaire, preliminary test, field test. The ISPOR Principles of Good Practice for the Translation and Cultural Adaptation Process for Patient-Reported Outcomes (PRO) Measures were applied during the process (18).

Participants and procedures

The population of the study comprised the legal relatives of patients with IH under two years of age (mother/father/grandmother/grandfather/legal guardian).

The following inclusion criteria were used: 1) the age of the child up to 24 months; 2) the presence of an infantile haemangioma diagnosed by a specialist; 3) the ability of parents to read and understand the Romanian language; 4) to sign the informed consent form and to answer the questionnaire. Patients older than two years at the time of the interview and those with comorbidities unrelated to haemangioma or other major developmental disorders were excluded.

In the present study we took into account the Romanian context, considering the limited financial possibilities of the family members in terms of internet access and possession of electronic devices that would allow them to complete the online questionnaire. Thus, we decided that the information should be obtained by physically completing the questionnaire during the first outpatient consultation. Patients were recruited from those who went to a pediatric surgery department at a pediatric clinical hospital between October 2019 and March 2020. ❑

RESULTS

First stage of developing the questionnaire

When reviewing the literature we searched for the elaboration of a research questionnaire with 60 items, considering the questionnaires used to measure the QoL in pediatric patients (14-17) and the specific questionnaire developed by Chamlin (11).

The research instrument is divided into three sections: one dedicated to collect demographic characteristics of responders, another one dedicated to collect data specific to IH characteristics (clinical data, including the location of the haemangioma, its size and stage of growth, and the prescribed treatment), and a third section consisting of 28 items specifically designed to measure the QoL containing self-assessment data on physical condition, emotional status and social function, through which we set out to evaluate the QoL of both IH patients and their families.

TABLE 1. Dimensions and items of the research questionnaire

Dimensions	Items
1) the child's physical health (CPH)	3. My child has pain because of this haemangioma. * 4. My child seems sickly or prone to illness because of the haemangioma.* 5. My child is not growing or developing normally because of the haemangioma.* 6. My child has trouble sleeping because of the haemangioma.* 7.Because of the haemangioma my child has problems being soothed or comforted when crying.*
2) the social function of the child (SFC)	8. My child's haemangioma prevents him/her from participating in social events such as parties and play groups.** 9. Friend or relatives avoid touching and holding my child because of his/her haemangioma.** 10.Children seem to avoid touching or playing with my child because of his/her haemangioma.** 16. I am bothered when strangers stare at my child.* 26. I am bothered when strangers offer opinions or ask questions about my child's haemangioma.*
3) the emotional health of the parents (EHP)	11. My child's haemangioma makes me feel sad or depressed. * 12. I am disappointed that my child has this haemangioma.* 14. My child's haemangioma makes me feel anxious or nervous.* 15. I am bothered by how much time is needed to care for my child because of the haemangioma. * 17. I worry about the amount of money I have to spend because of the haemangioma. * 18. I am embarrassed by the way my child looks because of his/her haemangioma. ** 19. I am worried that in the future, my child will not make friends as easily because of the haemangioma. * 21. The haemangioma has affected how confident I feel about my child's medical care.* 22. I get worried when I see changes in my child's haemangioma. * 23. I have been frustrated with my child's medical care for the haemangioma.*
4) the social function of the parent (SFP)	1. Has your child's condition interfered with your daily routine at work in the last month? *** 2. Has your child's condition interfered with your ability to concentrate at work in the last month? *** 13. I experience more headaches than usual as a result of my child's haemangioma.* 20. I blame myself or my child's other parent that my child has this haemangioma. * 24. I am bothered that my child needs to be watched more closely at home because of the haemangioma.* 25. I feel physically weak as a result of my child's haemangioma. 27. My child's haemangioma affects our social life.* 28. My child's haemangioma has strained my relationship with my spouse or partner.*

*Items included from the initial version of the questionnaire developed by Chamlin *et al*

**Items excluded by Chamlin at all from the initial questionnaire and included in the research questionnaire

The creation of our questionnaire took into account the cultural and social characteristics of Romanian people, so that the four dimensions of the above-described concept (the child's physical health, the social function of the child, the emotional health of parents and the social function of parents) should be best covered.

After translation of items from the selected questionnaires, colleagues (paediatric surgeons, paediatricians and dermatologists), otherwise

not involved in this study, were asked to give their opinion on the content and form of the questions in terms of scientific relevance according with the developed concept.

We have not included all items from the initial version of Chamlin et al's questionnaire (prototype IH-QoL35-item). From the original questionnaire (35 items in the initial form) used by Chamlin et al, only 26 items have been chosen, including five items that were eliminated by Chamlin *et al* in the final version because of low consistency. Two different items were added by us, so the final version of our questionnaire had 28 items. Both additional items were included in the dimension regarding the social function of the parent (SFP).

The items of the research questionnaire split by the four dimensions of the QoL concept are described in Table 1.

Because the difference between the questionnaire elaborated by Chamlin *et al* and ours consists of five questions that were eliminated by us and two items that were added, we may consider that this version is a Romanian adaptation of IH-QoL.

Nine items were excluded from the initial version of Chamlin's questionnaire because we took into consideration the specific Romanian socio-cultural context:

– two items from the CSI sub-scale: item 27 ("I have been accused of child abuse because of my child's hemangioma") and item 31 ("I am bothered that children touch or comment on my child's hemangioma");
– four items from the PSF sub-scale: item 28 ("Our family is less likely to go to public places – e.g., grocery store – because of the hemangioma"), item 29 ("My child's hemangioma affects my or my spouse/partner's work due to missed time"), item 34 ("I feel too tired to do the things I like to do because of my child's hemangioma") and item 35 ("I have felt sick to my stomach as a result of my child's hemangioma");
– two items from the sub scale PEF 9: item 32 ("I worry about my child based on information I read on the internet") and item 33 ("I worry about side effects of the medication(s) used to treat my child's hemangioma");
– one item excluded by Chamlin *et al* from the final version of the questionnaire: item 30 ("I am considering not having more children because of my child's hemangioma").

For example, item 27 ("I have been accused of child abuse because of my child's hemangioma") has been excluded because the authors considered it non-specific for the Romanian society, characterized by a higher prevalence of using violence as a form of children's discipline or instruction and a general tolerance of a violent behavior (22).

Item 28 ("Our family is less likely to go to public places – e.g., grocery store – because of the hemangioma") has been excluded because culturally, young children were not usually exposed to crowded places.

Items 32 ("I worry about my child based on information I read on the internet") and 33 ("I worry about side effects of the medication(s) used to treat my child's hemangioma") have been also excluded due to the specific Romanian context – the use of internet to seek health information has one of the lowest levels in the EU, that is 31%, similarly to Bulgaria, ranked in last place (23).

In order to facilitate the analysis of information that would be obtained after completing the questionnaires, we decided to use a different scale from that used by Chamlin *et al* to evaluate the IH-QoL.

The new scale evaluated the items according to scores from 1 to 5 for each item: 1 = never a problem; 2 = almost never/rarely (ones/month) a problem; 3 = sometimes (2-3 times/month) a problem; 4 = often/frequently (4-5 times/month) a problem; 5 = almost always (more than six times/month)/always a problem.

Testing the first version of the questionnaire

Considering the young age of the patients (with special and individualized needs and programs) and the physical and financial effort of their relatives to come to the scheduled appointment with the child within a strictly set time interval, we made one focus group session with members of five of the IH children, to whom the questionnaires were also applied. Nine people (five mothers, two fathers and two grandparents) with various ages, educational levels and financial possibilities agreed to participate.

With the help of the focus group, we explored the range of issues related to the impact of IH on the QoL and recorded the key words and phra-

ses through which participants described their experiences and feelings, the extent to which they were found or not in the questionnaire, clarity of questions, etc. The meeting took place at a distance from the moment of initiating the treatment (three or four months), so that the already established relationship with the patients' relatives offered them the comfort and security of an open in-depth dialogue. We asked about the readability, intelligibility, and relevance of the questions. Some minor changes have been made to a few questions so that the content was not changed. There were also individual discussions with relatives who were unable to attend the focus group.

Several difficulties were encountered when applying the questionnaire, including reluctance to participate in the study/decision making without consulting the other parent (they asked for time to think); difficulties in understanding the content of the questions under the emotions of the first contact with the specialist doctor; difficulties in self-assessment of physical and emotional status; requesting help in completing the questionnaire that influenced the answers; extension of the completion time by requesting additional explanations.

In our opinion, these problems could be solved by providing a consent form and time to think during the initiation phase of the treatment, handing over the questionnaire to be completed at home and brought to the next visit, reformulating the questions by using a less abstract language, changing the content of questions with preserving their meaning in order to facilitate the answer, the most precise and complete explanation of all aspects of the study and the content of the questionnaire.

Applying the research questionnaire

The questionnaire was administered to the parents of 20 children with IH under 24 months of age. If parents had questions about the semantics or content of the items, we provided them answers as well as assistance for dealing with the questionnaire, which made it possible to follow the completion of the questionnaire in full and ensure there was no risk of missing data.

Telephone interviews were conducted when parents asked for time to think and had doubts about participating in the study. The completion time was three to five minutes if the completion was supervised, and eight to ten minutes if the respondent did not ask for help. The questionnaire once obtained was applied to 112 respondents; the results will be subject of another article. ❑

DISCUSSIONS

It is essential to achieve the most accurate measurement of the QoL of both IH patients and their families in order to succeed in taking steps to improve it by using measures specific to each affected area (for example, psychological counselling). Although there are several pediatric generic scales for measuring health, none of them is assessing the unique impact of the birth of a child with IH during the critical period of the first months of life. IH-QoL was designed to comprehensively measure the QoL in this single group of population, following the complexity of IH children's reactions. This scale is based on direct interaction with parents, review of the literature, feedback from doctors who use to treat this disease, so we believe that it can be successfully used in Romania too. ❑

CONCLUSIONS

This questionnaire is a good tool for assessing the quality of life of paediatric patients belonging to children with IH, with applicability in the Romanian population.

New therapeutic options, which are especially useful for infants with haemangiomas (oral and topical beta-blockers, laser therapy with high-performance devices, etc), can verify their effectiveness through the questionnaire developed by the authors of the present study as a measure of patient-reported outcome.

It is particularly important that this scale can also be used in practice to identify parents who need support either emotionally or financially. Patients' problems with limited access to specific health services and innovative therapies may also be identified. ❑

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