STUDY PROTOCOL

Study Protocol: A Mixed-methods Study to Develop and Validate a Questionnaire of Self-management and Its Relationship With Quality of Life Among Thalassemia Adolescents

Siti Munirah Abdul Wahab[2], Siti Khuzaimah Ahmad Sharoni[1], Wan Ismahanisa Ismail[2], Noor Shafina Mohd Nor[3]

[1] Centre for Nursing Studies, Faculty of Health Sciences, Universiti Teknologi MARA Selangor, Kampus Puncak Alam, 42300 Puncak Alam, Selangor, Malaysia
[2] Nursing Department, Faculty of Health Sciences, Universiti Teknologi MARA, Cawangan Pulau Pinang, Kampus Bertam, 13200 Kepala Batas, Pulau Pinang, Malaysia
[3] Paediatric Department, Faculty of Medicine, Kampus Sungai Buloh, 47000 Sungai Buloh, Selangor, Malaysia

ABSTRACT

Introduction: Few studies have been published on self-management among Malaysian adolescents with thalassaemia due to the paucity of specific theoretical and culturally appropriate measures, which has led to psychological effects and non-compliance with conventional treatment. This study aims to develop and validate a questionnaire to evaluate self-management among Malaysian adolescents with thalassaemia. **Methods:** An exploratory mixed-methods design will be used in this study. In Phase I, we will recruit adolescents with thalassaemia aged 10 to 19 and their caregivers from one of the thalassaemia societies for a semi-structured interview. Participant recruitment will cease upon achieving thematic saturation. Coding and theme analyses will be used to identify self-management domains and items. In Phase II, we will develop self-management instruments based on the domains and items generated in Phase I. Content validity will be reviewed by ten expert panels, and a pilot study will be tested with 42 conveniently selected adolescents aged 10 to 19. Four hundred twenty-two participants from eleven thalassaemia NGOs will participate in the psychometric evaluation of the items in the self-management instruments. **Expected Outcome:** With these instruments, healthcare providers will be able to gather crucial information regarding thalassaemia adolescents' self-management, which may improve adolescents' adherence to treatment and lower their levels of serum ferritin, enhancing their health-related quality of life. **Trial registrations:** Ethical approval was obtained from the UiTM Ethics Research Committee (REC/10/2023-PG/MR/401) and the Malaysia Thalassaemia Society. *Malaysian Journal of Medicine and Health Sciences* (2024) 20(SUPP9): 268-275. doi:10.47836/mjmhs20.s9.42

Keywords: Thalassaemia, Adolescents, Self-management, Questionnaire, Health-related quality of life

Corresponding Author:
Siti Khuzaimah Ahmad Sharoni, PhD
Email: sitik123@uitm.edu.my
Tel : +603-3258 4349

INTRODUCTION

Living with thalassaemia is very challenging due to the many demands of the changes in lifestyle that occur from an early age. According to a study published in 2020, the annual expected number of newborns with beta-thalassaemia is 20,420 in Southeast Asia, 9,914 in the Eastern Mediterranean region, and 1,019 in South Asia (1). Until early 2012, there were 4,715 patients with thalassaemia in Malaysia, many of whom were between 10 and 15 years old (2). Adolescence is a crucial developmental period for establishing lifelong health-related behaviours (3). Patients with intricate chronic illnesses and developmental changes during adolescence encounter difficulties with their health-related quality of life (HRQoL) (4).

Like other chronic disorders, thalassaemia has substantial physical, socio-mental, congenital, and economic repercussions on individuals and their families (5). Physical anomalies, growth retardation, hepatosplenomegaly, yellow skin, and disease-like facial lines are seen (6). This bodily appearance lowers self-esteem and makes people feel different. Such individuals have anaemia, tiredness, and a lack of tolerance to physical activity, as well as needing lifetime care, blood transfusions, and iron chelation (7). As with other chronic disorders, thalassaemia requires ongoing follow-up and medical care monitoring, which affects physical, psychological, social, and school functioning in children and adolescents. Blood transfusion and iron chelation therapy make thalassaemia a chronic illness that requires self-management (8). For personal

well-being, HRQoL, and cost reduction, lifelong self-management is needed (9, 10).

Lifelong treatment and care to reduce problems in children with thalassaemia can influence HRQoL (11). One study conducted in Cairo stated the need for thalassaemia self-management and treatment plans to enhance HRQoL in children with chronic diseases such as thalassaemia (12). Some studies have identified that self-management is linked to HRQoL, distress, and depression (9). Enhanced self-management is associated with enhanced illness management, patient well-being, quality of life, and individual growth, potentially decreasing depression and anxiety, particularly in individuals with asthma and type 1 diabetes (13). Most studies have connected self-management and quality of life in paediatrics to chronic disease but not specifically to thalassaemia. Ultimately, the relationship between self-management and adolescents' quality of life is complex and elusive.

Self-management (SM) is the "day-to-day management of chronic conditions by individuals throughout an illness" (14). Self-management improves the health outcomes of chronic illness by enhancing treatment adherence and problem-solving skills, while individuals can better navigate challenges (15). Adolescents with chronic diseases need to acquire self-management abilities to handle their condition as they go into adulthood (16). A successful transition to adult healthcare requires self-management skills such as appointment-making/-keeping, medicine refills, and insurance (17), the lack of which raises the risk of poor long-term health (18). For adolescents with chronic disorders like thalassaemia, self-management measures can improve their health and well-being (8,19). Primary care physicians, nurse practitioners, and paediatric subspecialists should assess self-management skills, self-efficacy, and relevant contextual factors to support self-management (15). Therefore, establishing the nurse's role in thalassaemia care and in collaborating with other healthcare providers can improve patient care and reduce the risk of a lack of coordination (20).

Different scales measure quality of life, resilience, coping methods, and self-efficacy. No single scale measures all the aspects of thalassaemia adolescents' self-management. Quality measures are needed to evaluate teenage thalassaemia self-management. Researchers should concentrate on measuring self-management practices in children with chronic illnesses such as thalassaemia (15). These assessment tools should be adapted for shared management, meaning that both the healthcare provider and the patient should be involved in managing the condition. Of the existing tools, Self-Management of Type 1 Diabetes in Adolescence (SMOD-A) (21) and Adolescent Self-Management and Independence Scale II (AMIS II) Original (22) were created for adolescents with chronic conditions. Additionally, the Paediatric Epilepsy Medication Self-Management Questionnaire (23) and the Family Asthma Management System Scale (FAMSS) Original (24) were created for parents to evaluate family and child self-management. The features and psychometric aspects of the self-management questionnaire are shown in Table I.

Table I: Self-management related questionnaires

Questionnaires	Author and Year	Indications	Number of Items	Aims	Reliability	Validity	Face and Content Validity	Developed Upon Theoretical Framework
Family Asthma Management System Scale (FAMSS) Original	Klinnert et al. (24)	Parents of children with asthma	11 sub-scales	The FAMSS evaluates the effectiveness of the family asthma management system.	Cronbach's alpha for the FAMSS Summary Score was 0.91.	Not reported	Not reported	Family asthma management system
Self-Management of Type 1 Diabetes in Adolescence (the SMOD-A) Original	Schilling et al. (21)	Patients with type 1 diabetes	33	The SMOD-A evaluates self-management in youths with type 1 diabetes.	Cronbach's alphas for the five subscales ranged from 0.71 to 0.85	EFA revealed five sub-scales	Assessed by multiple groups of experts	Not reported
Pediatric Epilepsy Medication Self-Management Questionnaire (PEMSQ) Original	Modi et al. (23)	Parents of children with epilepsy	15 of 27	The PEMSQ evaluates medication self-management in children with epilepsy.	Cronbach's alphas ranged from 0.63 to 0.93	Not reported	Not reported	Not reported

CONTINUE

Table I: Self-management related questionnaires. (CONT.)

Questionnaires	Author and Year	Indications	Number of Items	Aims	Reliability	Validity	Face and Content Validity	Developed Upon Theoretical Framework
Adolescent-Self-Management and Independence Scale II (AMIS II) Original	Sawin et al. (22)	Patients with spina bifida and their parents	5 of 17	The AMIS II evaluates the adolescent's increasing responsibility for and implementation of self-management behaviours.	Cronbach's alpha for the scale and subscales were 0.78–0.82. Cronbach's alphas for parent and adolescent were 0.87–0.89 for the total scale.	EFA, using principal axis analysis with an oblique rotation, conducted on parent data, supported two related self-management factors.	Content validity was conducted using healthcare professionals, parents, and AYA.	Not reported

Of the existing tools, Self-Management of Type 1 Diabetes in Adolescence (SMOD-A) (21) and Adolescent Self-Management and Independence Scale II (AMIS II) Original (22) were created for adolescents with chronic conditions. Additionally, the Paediatric Epilepsy Medication Self-Management Questionnaire (23) and the Family Asthma Management System Scale (FAMSS) Original (24) were created for parents to evaluate family and child self-management. The features and psychometric aspects of the self-management questionnaire are shown in Table I.

No measure exists to assess adolescent thalassaemia self-management and its components in Malaysia, while no inductively constructed and psychometrically verified instrument has measured teenage thalassaemia self-management. An efficient and reliable tool for measuring adolescents thalassaemia self-management could assist healthcare administrators in identifying adolescents' requirements and assessing support initiatives in regions with high frequency. The current study aims to identify self-management themes in thalassaemia adolescent patients (Phase I); develop and validate a Self-Management Questionnaire (SMQ) (Phase II); and determine the relationship between self-management and HRQoL. This exploratory mixed-methods project will run from May 2023 through December 2025.

MATERIALS AND METHODS

Phase I

In Phase I, we will utilise a qualitative method to explore the reality of adolescent thalassaemia SM from the viewpoints of adolescents and caregivers in identifying items of SM.

Study Participants

Due to the high prevalence of thalassaemia in Alor Setar, Kedah, northern Malaysia, "Kelab Thalassaemia Kedah" (KTK) will gather data and recruit participants for Phase I (2). Thalassaemia adolescents and carers registered with "Kelab Thalassaemia Kedah" in Kedah will be recruited to participate through purposeful sampling. Thalassaemia-afflicted adolescents will be interviewed separately to learn how they achieve SM. Carers will be included because of their vital role in helping thalassaemia adolescents in their SM. Potential participants will be contacted by phone based on the inclusion and exclusion criteria provided. Twenty thalassaemia adolescent participants and 20 of their parents will be interviewed, meeting the sample size recommendations for this type of research (25).

Participant Recruitment

Thalassaemia adolescent participants can be recruited for this study if they meet the following inclusion criteria: (a) are aged 10 to 19 years old, have been diagnosed with (any type of) thalassaemia, and have undergone conventional treatment for at least six months; and (b) can communicate in English or Bahasa Malaysia. The caregiver can be one parent or guardian of each adolescent who has lived with the adolescent for at least one year. The adolescents who agree to participate will be given letters of ethical approval, patient information sheets describing the study, and written permission forms for their parents.

For exclusion criteria, individuals who have trouble communicating because of thalassemia or other reasons will not be considered for this study.

Data Collection

To ensure the validity and dependability of the data, a semi-structured interview guide will be created in multiple steps. Based on the purpose of the research, open-ended questions will be provided in the interview guide (e.g., How do you take your iron chelation drugs?) in Step 1. In Step 2, two supervisory committee members will independently review the interview guide and provide input. Presenting the questions in English will help the committee comprehend them. After that, the approved interview guide will be translated into Malay

by an independent translator.

"Kelab Thalassaemia Kedah" has a quiet discussion room for the interview. They will be held in English or Malay since this study will include urban and rural participants who speak English and/or Malay. The interview goals will be discussed at the start of each session. To develop rapport, the participants will introduce themselves. Their viewpoints will be encouraged to be expressed freely. We will elicit self-management domains from the participants using all the interview questions. The interviewer will reflect after each 40-minute session. Audio and verbatim transcription will be done for all the sessions. Until thematic saturation is achieved, participant recruitment will be ongoing. There will be no disclosure of any personally identifiable information.

Data Analysis

Following each interview, the audio recordings will be stored on a laptop protected by a password. The audio will be transcribed verbatim using NVIVO software. The transcripts will be cross-referenced with the audio of the interviews to guarantee precision. Coded names will replace participant names for confidentiality. Interview recordings in Malay will be translated into English. Constant comparative analysis will be used. The researchers will read the transcripts before extracting and coding meaning units. Next, codes will be categorised. Credibility, transferability, dependability, and confirmability assessments will assure the quality and rigour of this qualitative inquiry.

Triangulation and member checks will be used to make this study credible. Triangulation will involve comparing two different researchers' conclusions on the same dataset. Restoring the analysed data to the thalassaemia patients and carers will verify membership. Researcher reflexivity and peer review will ensure reliability. The raw data will be reviewed and determine whether the research conclusions are credible based on the interview data by the qualitative researcher. The interviewers will also be candid about any implications of their own personal beliefs, assumptions, theoretical leanings, and connections to the study for the outcome of the inquiry. Transferability will be sought by thoroughly describing the study environment and participants' characteristics (e.g., the adolescents' age, gender, and ethnicity, as well as the caregivers' level of education). Providing verbatim transcripts will ensure confirmation.

No participant-related information, such as names or addresses, will be revealed in the interview or on any of the recorded audio. Only the audio will be used for transcription purposes; no copies or usage will be made of the recordings. The audio recordings will be properly discarded after transcription. All audio recordings in MP3 format will be kept until the transcription process is finished. A password-protected PC will store each transcription in a Microsoft Word file. Data will be restricted to the principal and coordinating investigators.

Phase II

A quantitative study will be conducted to, firstly, develop and validate the Self-Management Questionnaire (SMQ) among a sample of thalassaemia adolescent patients and, secondly, determine the relationship between self-management and Health-related Quality of Life (HRQoL) among Malaysian adolescents with thalassaemia. This study is designed as a cross-sectional quantitative analysis.

Item Generation

The preliminary SMQ will include topics from Phase I, in which self-management domains among thalassaemia adolescent patients were identified. Using these themes, we will develop preliminary SMQ items. The content validity index (CVI) will be calculated using empirical methods. The CVI is a commonly used measure for assessing content validity in instrument development, can be calculated based on the Item-CVI (I-CVI) and the Scale-level-CVI (S-CVI) (26).

According to Yusoff (27), the content validation specialists used in a study should number at least six but no more than 10. Eight expert panelists from various fields with expertise and knowledge in thalassaemia management, such as two nursing lecturers, one psychologist, two paediatric haematology specialists and two nurses who manage thalassemia patients, including one president of the thalassaemia society will assess each SMQ item independently. Two adolescents with thalassemia as representatives of thalassaemia patients will be invited to be part of the expert panelists who ensure accurate and sensitive SMQ items. Using a four-point Likert scale, they will evaluate and score the items based on appropriateness, representativeness, and explicitness. The latter will range from 1 (irrelevant and should be deleted) to 4 (relevant, clear, and precise).

The I-CVI is calculated by dividing the number of experts who rated an item as three or four on the scale by the total number of experts. Statistics suggest each I-CVI should be over 0.78 if six or more experts are involved. Lower-rated items should be removed or updated. The process should persist until each item/statement achieves an I-CVI of 0.78 or above. Through S-CVI/Ave, an expert committee will decide whether to delete or keep each item. The percentage of content-valid items is the S-CVI. Since this measures the average item quality, it is calculated by averaging the I-CVI. The recommended S-CVI/Ave is 0.90. S-CVI/Ave and I-CVI are reported individually. In addition to the relevancy of the items, the experts will grade their clarity from 1=very confusing to 4=very clear. Any item with a clarity score

below 0.8 will be eliminated. This will produce the preliminary version of the SMQ. English will be used to create the SMQ.

The Pilot Version

Pilot testing will ensure that the Malaysian thalassaemia adolescents find the items significant. We will pilot the SMQ and additional measurements, including the PedsQL™ 4.0 Generic Core Scale (GCS). Pilot research is needed to reduce misunderstandings, measurement mistakes, and poorly phrased items. Pilot testing will be done with 42 thalassaemia adolescents, representing 10% of the anticipated 422 participants from the actual study sample, from "Kelab Thalassaemia Kedah", the same society involved in Phase I. All the adolescents who have attended the society and meet the study inclusion criteria will be asked to complete the questionnaire. Participating adolescents will complete an assent form and their carers' consent will be obtained. The researcher will help the teenagers complete the questionnaire and answer the questions. The researcher will organise the questionnaire, time it, and assist participants. Post-questionnaire, the participants will be briefly asked about their understanding and any confusion. They will also be able to add to, explain, and edit each preliminary SMQ question to make it more exact, culturally acceptable, and time-efficient. This will create the SMQ pilot.

Participant Recruitment

In Phase II, data collection and participant recruitment will involve 11 of the 12 non-government organisations (NGOs) of the Thalassaemia Society in Malaysia. Since "Kelab Thalassaemia Kedah" will be used for data collection for Phase I and the pilot testing, this club will be excluded to avoid confounding bias. Eligible thalassaemia adolescent patients will be randomly selected. The participants will be selected based on the inclusion and exclusion criteria used in Phase I.

A minimum of 200 participants is required for factor analysis sample size calculation (28). Hence, 422 participants will be appointed for the final sample through proportionate stratified random sampling to determine the total number of participants from each society and ensure that representative examples will be obtained.

Measures

Participants will fill out a study questionnaire that includes sociodemographic information such as age, gender, ethnicity, carers' level of education, carers' level of income, and serum ferritin status. The questionnaire will be available in either printed or online format and will also include the pilot version of the SMQ. Our goal is to create a questionnaire including less than 100 items to avoid respondent fatigue and enhance data quality (29). The overall score will be calculated by adding together the scores from all items. Higher total scores indicate weaker self-management. This SMQ questionnaire will use a Likert scale ranging from 1 to 5 (where 1 = strongly disagree and 5 = strongly agree). For the lowest possible score, sum up the lowest scores for all items; for example, if each item has a minimum score of 1, the lowest possible total score would be the sum of the lowest scores across all items). Meanwhile, the highest possible score sums up the highest scores for all items, for example, if each item has a maximum score of 5, the highest possible total score would be the sum of the highest scores across all items. This SMQ questionnaire will include subscales that represent different aspects of self-management. For example, the SMQ might have subscales related to emotional regulation, adherence to treatment, or lifestyle choices. Each subscale will have its own set of items and scores for each subscale separately.

The Final Version

We will analyse the factor structure of the revised SMQ using the validation sample. Exploratory factor analysis (EFA) and Cronbach's alpha will be applied to assess the adequacy of the measurement model of the SMQ. Missing values in EFA will be handled using 'pairwise' deletion, which assumes missing values are random and retains most of the data. EFA will be performed using principal component analysis (PCA). The data's appropriateness for factor analysis was evaluated, indicating correlation coefficients of 0.3 or higher in the correlation matrix. The Kaiser-Mayer Olkin (KMO) result of 0.794 surpassed the acceptable threshold. The Bartlett's Test of Sphericity yielded a statistically significant result, suggesting structured relationships among the items and confirming the correlation matrix's factorability (30).

Test for Validity

The predictive validity of the SMQ will be evaluated against the PedsQL™ 4.0 Generic Core Scale (GCS) using the Pearson correlation (31) to determine the relationship between self-management and HRQoL among Malaysian adolescents with thalassaemia. The 23-item instrument assesses various dimensions, including physical (eight items), emotional (five items), social (five items), and school (five items). It inquires how problematic a specific item from a dimension has been for a child over the past month. Each item has a five-point Likert scale that corresponds to five levels: "never a problem", "rarely a problem", "sometimes a problem", "often a problem", and "almost always a problem". The items are reversed and transformed into a 0 to 100 scale, where higher scores represent better HRQoL (31). The outcome is the average value calculated by considering the weight of each item in the Total Summary Score (TSS). The Physical Health Summary Score (HSS) and the Psychosocial Health Summary Score (PCHS) are subscales of the Total Summary Score (TSS). The PCHS comprises three components: emotional, social, and school functioning scores. We propose that there is a negative correlation between the total score of the SMQ

and the PedsQL™ 4.0 Generic Core Scale (GCS); for instance, poorer self-management is associated with lower HRQoL.

Test for Reliability

The Cronbach's alpha coefficient will assess the internal consistency of the final version of the SMQ. Items with a reliability coefficient of 0.70 or higher will be included in the SMQ questionnaire. An alpha coefficient of 0.70 is acceptable, whereas a value exceeding 0.80 indicates high internal consistency (32).

Ethics Approval

The UiTM Research Ethics Committee granted ethical approval for this research at UiTM (reference no: REC/10/2023 (PG/MR/401) on 16 October 2023. Before conducting the study, support from all presidents of the Thalassaemia Society will also be required.

DISCUSSION

Thalassaemia adolescents may face individual and environmental difficulties that hinder self-management and health outcomes (8). Inadequate research exists on self-management among adolescents with blood disorders like thalassaemia. Multiple self-management tools have been developed and utilised in other countries, but these have not been assessed for their cultural, contextual, and developmental appropriateness to Malaysia.

To address these demands, this study has two goals. First, through quantitative inquiry, we aim to identify self-management domains by exploring the realisation of adolescent thalassaemia self-management from the perspectives of adolescents and caregivers. Secondly, through a quantitative approach, we intend to develop and validate a self-management questionnaire called the Self-Management Questionnaire (SMQ). This newly developed questionnaire can be used to measure components of self-management behaviours. It will also precisely pinpoint the underlying HRQoL among thalassaemia adolescents.

The study will utilise the PedsQL™ 4.0 Generic Core Scale (GCS) to investigate the predictive validity of the SMQ in analysing the association between self-management and health-related quality of life (HRQoL). The Malay version of the PedsQL™ 4.0 Generic Core Scale (GCS) has been validated locally, and it shows moderate-to-good reliability and validity (33). Second, we will print and distribute questionnaires online and offline to boost the response rates. Eligible participants will be called to have the survey explained and provide their consent. We anticipate these sample procedures will improve the response rate and efficiency, as well as reduce the cost.

CONCLUSION

This study will offer comprehensive and detailed insights into the fundamental aspects of self-management behaviours in thalassemia teenagers. Based on these themes, we will create a local questionnaire (SMQ) to measure self-management levels in an adolescents thalassaemia patients accurately. This questionnaire aims to assist healthcare providers in comprehending the varying significance of various self-management elements and their interplay. This information will help the Ministry of Health make policies and help public health professionals create effective health promotion programmes and evidence-based interventions to decrease problems from not following traditional treatment. Better-controlled serum ferritin could reduce multiple organ complications, relieving the public healthcare burden. Improving adherence may also result in more cost-effective thalassaemia treatment in Malaysia.

ACKNOWLEDGEMENT

This paper is part of a research project funded by the Ministry of Higher Education Malaysia under the Fundamental Research Grant Scheme (FRGS) (FRGS/1/2023/SS10/UITM/02/6). The authors would like to thank the directors of Malaysia's Thalassaemia Societies for granting permission to perform the study within their societies.

REFERENCES

1. Kattamis A, Forni GL, Aydinok Y, Viprakasit V. Changing patterns in the epidemiology of β-thalassemia. Vol. 105, European Journal of Haematology. Blackwell Publishing Ltd; 2020: p. 692–703. doi: 10.1111/ejh.13512.
2. Mohd Ibrahim H, Muda Z, Othman IS, Mohamed Unni MN, Teh KH, Thevarajah A, et al. Observational study on the current status of thalassaemia in Malaysia: A report from the Malaysian Thalassaemia Registry. BMJ Open. 2020 Jun 29;10(6). doi: 10.1136/bmjopen-2020-037974.
3. Schulenberg JE, Sameroff AJ, Cicchetti D. The transition to adulthood as a critical juncture in the course of psychopathology and mental health. 2004. doi: 10.10170S0954579404040015.
4. Shackleford JL, Kelley SJ, Spratling R. Applying the self-determination theory to health-related quality of life for adolescents with congenital heart disease. J Pediatr Nurs. 2019 May 1; 46:62–71.

doi:10.1016/j.pedn.2019.02.037.
5. Tarım HŞ, Öz F. Thalassemia major and associated psychosocial problems: A narrative review [Internet]. Iran J Public Health. 2022 [cited 2022 June 28]; vol.15. Available from: http://doi.org/10.18502/ijph.v51i1.8287.
6. Hakeem GLA, Mousa SO, Moustafa AN, Mahgoob MH, Hassan EE. Health-related quality of life in pediatric and adolescent patients with transfusion-dependent Я-thalassemia in upper Egypt (single center study). Health Qual Life Outcomes. 2018 Apr 10;16(1). doi: 10.1186/s12955-018-0893-z.
7. Pouraboli B, Abedi HA, Abbaszadeh A, Kazemi M. Living in a misty marsh: A qualitative study on the experiences of self-care suffering of patients with thalassemia. Iranian Journal of Nursing and Midwifery Research | Special Issue on Health and Wellbeing. 2014 [cited 2022 July 22]; vol.19. Available from: https://www.ncbi.nlm.nih.gov/pmc/articles/PMC4402994/.
8. Razzazan N, Ravanipour M, Jahanpour F, Ahmad ;, Zarei R, Motamed ; Niloofar, et al. Investigating challenges facing self-management empowerment education in adolescents and youths with Major Thalassemia. Jundishapur J Chronic Dis Care. 2014;3(4):23366. doi: 10.5812/jjcdc.23366.
9. Bravo L, Killela MK, Reyes BL, Santos KMB, Torres V, Huang CC, et al. Self-management, self-efficacy, and Health-related quality of life in children with chronic illness and medical complexity. Journal of Pediatric Health Care. 2020 Jul 1;34(4):304–14. doi: 10.1016/j.pedhc.2019.11.009.
10. Jiang X, Walker K, Topps AK. A systematic review of self-care measures for adolescents with health needs. Quality of Life Research. Springer Science and Business Media Deutschland GmbH; 2021; vol(30):p. 967–81. doi: 10.1007/s11136-020-02685-1.
11. Sawicki GS, Ostrenga J, Petren K, Fink AK, D'Agostino E, Strassle C, et al. Risk factors for gaps in care during transfer from pediatric to adult cystic fibrosis programs in the United States. Ann Am Thorac Soc. 2018 Feb 1;15(2):234–40. doi: 10.1513/AnnalsATS.201705-357OC.
12. Ahmed Khalil A, Mohamed Mahmoud Sarhan prof, Rizk Mohammed N, Moheb Mohammed Gomah N. Assess quality of life of children with Beta Thalassemia Major. Port Said Scientific Journal of Nursing. 2019; vol.6. doi:10.21608/pssjn.2019.34698.
13. Biagioli V, Spitaletta G, Kania V, Mascolo R, Gawronski O, Liburdi A, et al. Instruments measuring self-care in children and young adults with chronic conditions: A systematic review. Frontiers in Pediatrics. Frontiers Media S.A. 2022; vol.10. doi: 10.3389/fped.2022.832453.
14. Grady PA, Gough LL. Self-management: A comprehensive approach to management of chronic conditions. Public Health. 2014;104:25–31. doi: 10.2105/AJPH.2014.302041.
15. Lozano P, Houtrow A, Washington KP. Supporting self-management in children and adolescents with complex chronic conditions [Internet]. PEDIATRICS. 2018 [cited 2023 June 28]; vol. 141. Available from: https://doi.org/10.1542/peds.2017-1284h.
16. Gauci J, Bloomfield J, Lawn S, Towns S, Steinbeck K. Effectiveness of self-management programmes for adolescents with a chronic illness: A systematic review. Journal of Advanced Nursing. John Wiley and Sons Inc. 2021; vol.77: p. 3585–99. doi: 10.1111/jan.14801.
17. Wiemann CM, Graham SC, Garland BH, Raphael JL, Jones MD, Sanchez-Fournier BE, et al. In-depth interviews to assess the relevancy and fit of a peer-mentored intervention for transition-age youth with chronic medical conditions. J Pediatr Nurs. 2020 Jan 1;50:121–7. doi: 10.1016/j.pedn.2019.04.028.
18. Hergenroeder AC, Moodie DS, Penny DJ, Wiemann CM, Sanchez-Fournier B, Moore LK, et al. Functional classification of heart failure before and after implementing a healthcare transition program for youth and young adults transferring from a pediatric to an adult congenital heart disease clinics. Congenit Heart Dis. 2018 Jul 1;13(4):548–53. doi: 10.1111/chd.12604.
19. Catarino M, Charepe Z, Festas C. Promotion of self-management of chronic disease in children and teenagers: Scoping review. Healthcare (Switzerland). MDPI; 2021; vol. 9. doi: 10.3390/healthcare9121642.
20. Mardhiyah A, Letcmi Panduragan S, Mediani HS, Rai RP, Bandung R, 21 SK, et al. Integrating family empowerment into thalassemia care for adolescents in Indonesia: A synthesis of recent evidence [Internet]. Malaysian Journal of Medicine and Health Sciences. 2022 [cited 2023 June 23]; vol. 18. Available from: https://medic.upm.edu.my/upload/dokumen/2023010417021856_2021_1245.pdf.
21. Schilling LS, Dixon JK, Knafl KA, Lynn MR, Murphy K, Dumser S, et al. A new self-report measure of self-management of type 1 diabetes for adolescents. Nurs Res. 2009 Jul;58(4):228–36. doi:10.1097/NNR.0b013e3181ac142a.
22. Sawin KJ, Heffelfinger A, Cashin SE, Brei TJ. The development of the adolescent/young adult self-management and independence scale II: Psychometric data. J Pediatr Rehabil Med. 2018;11(4):311–22. doi: 10.3233/PRM-170479.
23. Modi AC, Monahan S, Daniels D, Glauser TA. Development and validation of the Pediatric Epilepsy Medication Self-Management Questionnaire. Epilepsy and Behavior. 2010 May;18(1–2):94–9. doi: 10.1016/j.yebeh.2010.03.009.
24. Klinnert MD, Mequaid EL, Gavin LA. Assessing the family asthma management System. J Asthma. 1997 [cited 2022 March 15]; vol. 34. Available from: https://doi.org/10.3109/02770909709071206.

25. Creswell JW, Poth CN. Qualitative inquiry and research design: Choosing among five approaches. Sage Publications, Incorporated; 2018.
26. Zamanzadeh V, Ghahramanian A, Rassouli M, Abbaszadeh A, Alavi-Majd H, Nikanfar AR. Design and Implementation Content Validity Study: Development of an instrument for measuring Patient-Centered Communication. J Caring Sci. 2015 Jun 1;4(2):165–78. doi: 10.15171/jcs.2015.017.
27. Yusoff MSB. ABC of content validation and content validity index calculation. education in medicine journal. 2019 Jun 1;11(2):49–54. doi: 10.21315/eimj2019.11.2.6.
28. Kyriazos TA. Applied psychometrics: sample size and sample power considerations in Factor Analysis (EFA, CFA) and SEM in general. Psychology. 2018;09(08):2207–30. doi:10.4236/psych.2018.98126.
29. Rolstad S, Adler J, Rydén A. Response burden and questionnaire length: Is shorter better? A review and meta-analysis. Value in Health. 2011 Dec;14(8):1101–8. doi: 10.1016/j.jval.2011.06.003.
30. Shrestha N. Factor analysis as a tool for survey analysis. Am J Appl Math Stat. 2021 Jan 20;9(1):4–11. doi:10.12691/ajams-9-1-2.
31. Varni JW, Seid M, Kurtin PS. PedsQLTM 4.0: Reliability and validity of the Pediatric Quality of Life InventoryTM Version 4.0 Generic Core Scales in healthy and patient populations [Internet]. 2001 [cited 2021 June 20]. Available from: http://www.pedsql.org.
32. McNeish D. Thanks coefficient alpha, We'll take it from here. Psychol Methods. 2018 Sep 1;23(3):412–33. doi: 10.1037/met0000144.
33. Hamidah A, Wong CY, Tamil AM, Zarina LA, Zulkifli ZS, Jamal R. Health-related quality of life (HRQOL) among pediatric leukemia patients in Malaysia. Pediatr Blood Cancer. 2011 Jul 15;57(1):105–9. doi: 10.1002/pbc.23125.