REVIEW ARTICLE

Integrating Family Empowerment Into Thalassemia Care for Adolescents in Indonesia: A Synthesis of Recent Evidence

Ai MARDHIYAH[1], Santhna Letcmi PANDURAGAN[2], Henny Suzana MEDIANI[3], Ramesh Prasath RAI[2]

[1] Faculty of Health Science, Lincoln University College, Jalan Stadium SS 7/15, Ss 7, 47301 Petaling Jaya, Selangor, Malaysia
[2] Faculty of Nursing, Lincoln University College, Jalan Stadium SS 7/15, Ss 7, 47301 Petaling Jaya, Selangor, Malaysia
[3] Department of Pediatric Nursing Universitas Padjadjaran, Jl. Raya Bandung Sumedang KM.21, Hegarmanah, Jatinangor, Kabupaten Sumedang, Jawa Barat 45363

ABSTRACT

Thalassemia is a significant public health issues not only for patients and their families but also caused an economic burden of the country. Adjusting condition of children with chronic disease to family is considered as a crisis with different response of family. There are limited data on family empowerment in the thalassemia and few countries have a structured guideline how to empower family into the thalassemia care. The purpose of this study was to give health care providers caring for adolescents with thalassemia in Indonesia an overview that takes family empowerment issues into account. The review were focus on effectiveness of family empowerment on patients with thalassemia, special consideration to empower family in adolescence with thalassemia, feasibility of Applying family empowerment into the Indonesian thalassemic care system for Adolescent. Studies have showed that family empowerment program with various model significantly improve self-efficacy and quality of life of the patients, also improve knowledge and attitude of family caregivers. However, some critical issues need to be considered prior to implementing family empowerment into thalassemia primary care, including disease burden, adolescent growth and development, institutional factors, social and culture beliefs. Integrating family empowerment into thalassemia care system is essential, especially in Indonesia. Developing recommendations for adopting family empowerment in thalassemia primary care is still a major priority. In order to produce comprehensive family empowerment guidelines for thalassemia care, further study into thalassemia and empowerment is required..

Keywords: Adolescent, Family empowerment, Family centered-care, Review, Thalassemia

Corresponding Author:
Ai Mardhiyah, S.Kp., M. Kes
Email: amardiyah182@gmail.com
Tel: +60 3-7886 7315

INTRODUCTION

Current estimates suggest that 60% of 229 countries were originally endemic of inherited hemoglobin disorders potentially affecting 75% of births, the majority of being affected by Thalassemia (1–3). Approximately, 80% of thalassemia cases occur in the low- or middle- income countries including South and Southeast Asia ((3)). The World Health Organization (WHO) (2018) reported that at least 20% of the world's population carries α+ thalassemia, and 2% of children are born with transfusion- dependent β thalassemia (3). About 56,000 have a major thalassemia, including at least 30,000 who need regular transfusions to survive. The mortality rate due to thalassemia was higher, about 5,500 die perinatally due to α thalassemia major and at least 3000 die annually in their teens or early 20s from uncontrolled iron overload (3). Indonesia is a country with in which 3.8% carry thalassemia and about 2,500 children born with thalassemia annually (4). The prevalence of thalassemia continues to increase from 4,896 in 2012 to 9,028 in (4).

In addition to affecting patients and their families, thalassemia has a substantial economic impact on the country. The cost of treatment is high involving routine transfusion, frequent hospitalization, and medical checkups (5,6). Furthermore, many elements of the patient and family's lives are affected by the long-term nature of thalassemia, including their well-being and mental health. (7). Previous studies reported that adolescents with thalassemia have

more depressive symptoms, aggression, anxiety that impaired their quality of life (8,9). These findings highlighted the need for a comprehensive program to support mental health, Adolescent motivation and quality of life are improved in thalassemic adolescents. Accordingly, improving the quality of life among thalassemic adolescents is one of the main goals of treatment. Therefore, there must be an effective method for healthcare workers (especially nurses), who have the closest ties to patients and their families, in order to better their patients' lives. (10).

Families respond differently to children with chronic diseases. A study reported that some families can successfully adapt themselves to having a child with a chronic disease. However, some people may struggle to adapt due to a lack of information, a lack of social support, a financial burden, and psychosocial issues (11). On the other hand, children's health, including disease prevention and treatment, relies heavily on their families (12). That condition become the reason why nowadays the attention of health care services has changed from child-centered care to family-centered care (13). As a result, the family, particularly the parents, should be involved in all aspects of the child's care, both in and out of the hospital (14). To enhance the role of family participation in child care, they are required to understand the disease, various aspect of treatment, care, and rehabilitation through empowerment (15).

Family empowerment is defined as a process to increase the autonomy and self-determination to enable them to represent their interest to build, develop, and increase the ability and effectiveness to set and reach goals for individual and social ends (16,17). There are various models of family empowerment applied in children with chronic or critical (12,18–24). Evidence has been suggested that family empowerment improves the quality of life and self-efficacy of children with thalassemia also improves the knowledge and attitude of mothers (12,18,22). According to the previous research, empowering sick children and their families is key to helping them cope with disease's symptoms and complications, as well as giving them greater influence over how they live their lives going forward (19).

There is a limited research available on family empowerment in the thalassemia population. The majority of studies only focus on disease management such as transfusion and prevention of complications. Moreover, few countries have a structured guideline on how to empower families in thalassemia care. Particularly in the Indonesian thalassemia care system, family empowerment receives less attention than transfusion management. The purpose of this paper is to give health care providers caring for a thalassemia-affected adolescent in Indonesia an overview of family empowerment concerns in thalassemia care management.

METHODS

Selection criteria

The purpose of this study was to conduct a systematic evaluation of the scientific literature using the PRISMA approach in order to identify evidence-based, which covered both theoretical and empirical literature on three topics: (a) effectiveness of family empowerment on patients with thalassemia; (b) special consideration to empower family in adolescence with thalassemia; (c) feasibility of Applying family empowerment into the Indonesian thalassemic care system for Adolescent. Inclusion criteria were defined as (1) type of participants: adolescence with thalassemia; (2) type of study: either randomized control trial, observational study, or review; (3) outcome: family empowerment, quality of life and other behavioral outcomes. Exclusion criteria were: a research conducted only in a specified target population such as just males or females.

Searching strategy

The source of the literature review was an online journal database as indexed in PubMed, Medline, Google Scholar, and Cochrane Library. We searched a combination of keywords including thalassemia mayor, thalassemia minor, adolescent, family empowerment, family-centered care, and quality of life. The search included the title, abstract, keywords, and text. The review was conducted in July 2019, and we focused our review on published papers (January 1998 - July 2019). Only publications in English and Indonesian languages were considered for inclusion.

1314 abstracts were found using the original keyword search. Of these, 1,297 were removed as not focused on family empowerment on thalassemic or chronic sickness. A total of 17 papers met all inclusion criteria; the majority were clinical trials, correlation studies, and review articles.

Data extraction

Data from the literature review was extracted by two reviewers independently using a structure data form developed by the investigator. We extracted data regarding the study information, including years of publication, study design, sample, intervention content/phase, clinical endpoints, features, usability, and feasibility. Any differences in data extraction between two reviewers were resolved by discussion and rechecked all entries paper.

Assessment quality of study

Studies were assessed by two reviewers using the standard form of the Joanna Briggs Institute Critical Evaluation Checklist for Observational Studies and Analysis (http:/www.joannabriggs.org/). The criteria

for appraisal of observational studies included subject, measurement, statistical test, and confounding. The criteria for reviews included review questions, inclusion and exclusion criteria, searching strategy, and appraisal. For the clinical trial, studies have assessed the risk of bias of included selection, reporting, performance, and attrition. The difference in the evaluation of studied quality mostly about which studies had a high risk of bias or low risk of bias. All disagreement was resolved by discussion and consulted with a third reviewer to help decide.

Family empowerment program in patients with thalassemia

Family empowerment included a concept of capacity building that highlighted the strength of the family as a unit not just focus on the sick child (25). Family empowerment has been implemented to patients with thalassemia, especially children and adolescents. Studies have shown that family empowerment programs with various models significantly improve self-efficacy and quality of life of the patients (12,18), also improve the knowledge and attitude of family caregivers (21,23) (Table I). Empowerment was also related to maintaining access to care, providing adequate data on wellbeing and engaging patients, families, and communities in care (26). It requires a multi-faceted approach, including an important approach of psychosocial educational support to explore family strengths, develop confidence and competence to take care of children with thalassemia.

Most of the family empowerment programs are guided by the concepts of illness belief model and social cognitive theory (12,18,22). (18) developed a program to empower families, based on four pillars: education, self-efficacy, involvement in training, and assessment. The stages included: (1) perceived threat: two sessions of group discussion and problem solving and an individual counselling session (topics : nutrition & blood transfusion); (2) self-efficacy: discussion and problem-solving in a group setting; (3) training perception: transfer information from the training sessions and pamphlets to the active member of the family in order to enhance self-efficacy; (4) evaluation program: the first stage included asking 2 oral questions at the beginning of each training session and the second phase was the final evaluation. (22) implemented family empowerment program through four strategies: (1) searching realities and exploring hope and illness beliefs about thalassemia; (2) sharing experiences and feelings and encouraging social support among the group members; (3) developing ability to manage care by sharing information and experiences and learning from other families; (4) enhancing self-assurance in order to succeed in family settings, & reflecting and providing feedback.

Family empowerment programs included health education programs for the family and encouraged them to participate in the patient's care to ensure that they had gained the skills and confidence to perform the task effectively at home (12,18). The current model is limited because it doesn't keep up with new theory and research-based evidence. Instead, it only studies attachment theory. On the other hand, a review of the family-centered care model found that the physical and emotional influences of parents and family early experiences were associated with lifelong health outcomes. There were four common model of family-centered care, namely bioecological model of human development, the family and community resource framework, allostatic load and biological embedding, nurse–family partnership (27).

Special consideration for applying family empowerment in thalassemia care

Disease burden

Thalassemia affects the life and health of both the affected child and family due to disease severity, long-term treatments and care, and psychological consequence as a result of caring for children with a serious chronic illness (28). Although the treatment could increase the life expectancy of the children, children with thalassemia still suffer from complex problems, disruptions to school and social activities are also included, and lack of skill of the parent to take care their children (29,30). There are a number of factors that influence health outcomes and disease prevention in the family. The quality of life of thalassemic patients has seen substantial improvement in Western countries, but even greater strides will be required to aid the needs of children in underdeveloped countries. Thalassemic children receive frequently blood transfusion and take deferoxamine to detoxify iron, that also induces iron-related issues and causes life threatening illnesses such as cardiovascular disease (31). As a result, providing excellent parental care for children with chronic illnesses necessitates a high degree of family competence, which involves an awareness of the facts of a child's chronic disease as well as the skill to handle child care over time.

Adolescent' growth and development

The adolescent patient must develop social independence while he/she has a dependency on family, doctors and nurses and a need to rigidly adhere to a demanding schedule. Non-adherence will result in problems and possible fatality. Family and caretakers may become overprotective. An adolescent with thalassemic may suffer from short stature, bone deformities and poor sexual development that could influence self-image and self-confidence (32). There was a lack of pubescent changes among adolescents with thalassemia both males and females, less than

Table I : Family empowerment program in patients with thalassemia

Authors, Years	Design	Sample	Intervention	Results
Borimnejad et al, 2017	• Quasi-experimental • Pre- and post-test	Adolescent with thalassemia major (n=35)	Empowering the patients in this program was based on four constructs including knowledge, self-efficacy, training participation and evaluation.4 stages: 1. Perceived threat: two sessions of group discussion and problem solving and an individual counselling session (topics : nutrition & blood transfusion). 2. Self-efficacy: training sessions with group discussion and problem solving. 3. Training perception: increase self-efficacy by transfer information from the training sessions and pamphlets to the active member of the family. 4. Evaluation program: the first stage included asking 2 oral questions at the beginning of each training session and the second phase was the final evaluation	The use of empowerment program and the participation of a family member helped to increase the self- efficacy of adolescents.
Wacharasin et al, 2015	Qualitative analysis of pre-intervention interviews and focus groups with family caregivers	25 Thai parents and family members who served as the primary caregiver for a child with thalassemia participated	1. Phase I: Creating a context for the family empowerment program (FEP). 2. Phase II: Implementation of FEP (Searching realities and exploring hope and illness beliefs about thalassemia, sharing experiences and feelings and encouraging social support among the group members, developing ability to manage care by sharing information and experiences and learning from other families, Strengthening confidence for mastery in family situations, & Reflecting and providing feedback.	FEP provided more knowledge, confidence, and competence in caring for their child with thalassemia
Borhani et al, 2011	• Clinical trial • Pre-poste test • 1.5 months follow up	Children aged 6-12 years with thalassemia major (n=86)	3 stages: 1. 3-5 sessions of group discussion for the children with their parents to improve perception, increasing knowledge and. awareness in association with the disease. 2. Increase participation in order to increase self-esteem and self- efficacy of the individual in coping with problems. 3. Presentation in the previous sessions were given to the child and his/her parents as CD.	After the intervention, there was a significant difference in level of the quality of life in all dimension
Najafi et al, 2011	Semi-experimental stud	86 children (6-12 y.o) with thalassemia	4 stages: 1. In the group sessions and discussed the use of pamphlets and slides about the disease. 2. Decoding phase problem 3. Educational pamphlets and other handouts and CD audio tracks were given to family members. 4. Evaluation was performed one and half month after intervention	Knowledge and attitude of the parents about their children disease were significantly increased after intervention

20% of females had menarche that resulted in sexual immaturity (33). Adolescents with thalassemia also suffer from different social disadvantages such as school dropout, family conflict, and declining social connections. Furthermore, personal physical manifestations induced by excessive deposition of iron discourages patients from active daily participation and lead to social exclusion. Family support perception of thalassemia is heavily influenced by their religious and educational backgrounds, thus their assistance may be little.. These include ethical themes relating to notions of sin, evil, destiny, and female sexuality (34). In such situations, the experiences of families not only hinder the normal development of adolescents with thalassemia but also serve as barriers to their psychosocial growth at times.

Social and culture beliefs

Caring for children with thalassemia requires families to keep up-to-date information and diagnosis for children with thalassemia (35,36) However, studies in Malaysia indicate that most families were unaware of the treatment and/or prognosis of the disease (37). In some countries, probably including families were focused on finding a way to cure thalassemia rather than learning to live with their child's chronic disease (28). Changing family beliefs about thalassemia and focusing on overcoming the suffering of the disease could be an important intervention to increase family empowerment.

An obstacle that is often encountered in respect of culturally competent treatment is the disparity between cultural values, behaviors and language or style of interaction between patients and health professionals (38). The study also highlighted the lack of social justice as a major deficit in implementing family empowerment. Social justice is an important requirement for the achievement of health services quality and fairness. Therefore, social justice must be considered based on the following principles: (a) equitable and equitable access to all health services for thalassemia patients; (b) not giving different treatment based on power; (c) focusing on social determinants of health; (d) making policies and procedures that promote the principle of justice for all users of health services; (e) protecting human rights; and (f) facilitating human development and self-actualization (39).

Institutional factors

Several studies have highlighted the essential factors of successful implementation of family empowerment as organizational support. This support includes policies, practices and staffing models that promote family-friendly care; build physical facilities to meet the needs of patients and families. Therefore, the capacity of health care staff must be improved to improve family autonomy in patient care, communication skills, family philosophy and methods of learning for patients and families (28).

Challenges for integrating family empowerment into thalassemia care

The application of family empowerment in the thalassemia population may have challenges. Firstly, children with thalassemia are diagnosed, typically, parents are unaware of the one in four chances they face in each new pregnancy. Individuals affected majorly with beta-thalassemia are not well integrated into society (40). Unfortunately, many of these children die before they turn ten years old because their parents cannot afford to take them out of their homes for medical treatment (40).

Second, while blood transfusions for those with health insurance are given free of charge at the hospital, parents are still billed for the required supportive medications. The Red Cross also offers blood transfusions, but even in this case, even when officially declared indigent, the parents have to pay a small amount of money. In such situations, they are asked to pay IDR 80,000 (around US$ 8), with the Indonesian Red Cross covering the remaining costs (40) with on average their monthly income was around US$ 50-100. Moreover, a blood transfusion usually necessitates a two- or three-day hospital stay., it means a loss of income during which one or both parents may not be able to work.

Third, culture has a significant impact on disability. Disability experiences are absent from major cultural images of a society's life. Furthermore, they stereotype and stigmatize physical and other differences, assign a variety of meanings to various types of disability and disease, and exclude handicapped individuals from activities that they cannot or are not supposed to participate in (34). Adolescents with thalassemia reported being significantly stressed, according to a study conducted in India, and patients suffer a multitude of physical, psychological, and social issues. They may be officially classified as a handicap in India, necessitating a multi-theoretical and multi-pronged intervention strategy to appropriately address them (34)s.

Fourth, in collectivist cultures such as Indonesia, families are perceived as having a collective face. In this sense, an individual's actions will affect other people's perceptions of one's entire family. Individuals should, therefore, strive to give a good name to their family and honor their parents. Before any other connections, they are also expected to be loyal to their family. Generally, the father or the oldest male is the patriarch, while women are responsible for domestic duties.

Applying family empowerment into the Indonesia Thalassemia care system for Adolescent

In 2011, the Indonesian government issued a policy to provide free access for people diagnosed with thalassemia, including consultation, laboratory examination but only for routine blood exam, urea,

creatinine, SGOT, and SGPT, medical equipment, drugs (folic acid, Vitamin E & C, xylocaine Jelly), transfusion, iron-chelation drugs, and ferritin examination for every three months. However, we don't have a specific guideline on thalassemia care that incorporates the concept of family empowerment yet. The family empowerment could be elaborate in disease management and prevention. The study noted that children with thalassemia could have a decent quality of life by expanding the availability of a regime of blood transfusions, iron chelation therapy, proper management of problems, and strong supportive care (41). Empower family on increasing availability of blood transfusion could be one of the solutions to increase blood availability through family networking and relationship. Nowadays, the extensive development of technology helps people to communicate with others easily throughout social media that can be utilized for a family to get the blood donor. The study also highlighted that proper management of complications is a key aspect but the majority of families had a lack of knowledge on caring for their chronic ill' child. Health care professionals may provide comprehensive training routinely and integrated into core disease management to improve the competency of the family to take care of their children properly.

A study reported that prevention programs in some countries have decreased thalassemia birth prevalence and may have saved patient care resources. These programs require preparation and commitment to include public awareness, testing for carriers, genetic counseling to help couples make informed choices, and eventually solutions such as prenatal diagnosis (32). There are significant differences in people's attitudes to screening as well as prenatal diagnosis and pregnancy termination. Cultural, social, ethical and legal factors must be taken into account in each country, but also in this age of increasing population mix, specific attitudes must be taken into account in planning services within communities in any country (42). As a largely Muslim nation this must impact on reproductive choices for families even with genetic disease. Despite the cost-effectiveness of prevention, very few countries have adopted nationally planned programs (43). Better understanding and awareness of the inherited disease is needed to empower the family for prevention

CONCLUSION

Especially in Indonesia, the thalassemia care system must incorporate family empowerment. Developing recommendations for implementation of family empowerment in thalassemia primary care remain a priority. Empowerment studies in thalassemia, particularly in the adolescent population, are necessary to determine the effectiveness and applicability. To make sure that family empowerment is consistently used by an adolescent with thalassemia, a systematic implementation of recruiting and training collaborative clinical team members will be needed to make sure that it is used. Defining the role of the nurse in thalassemia care and working with primary care providers to make sure they work together can help patients get the best possible care and avoid risks because of a lack of coordination. Further research is needed to better understand thalassemia and family empowerment, as well as to write comprehensive family empowerment guidelines for thalassemia care.

REFERENCES

1. Christianson A, Howson CP, Modell B, others. March of Dimes: global report on birth defects, the hidden toll of dying and disabled children. March Dimes Glob Rep birth defects, hidden toll dying Disabl Child. 2005;
2. Weatheral DJ. Keynote Address: The Challenge of Thalassemia for the Developing Countries. Ann N Y Acad Sci [Internet]. 2005 Nov 1;1054(1):11–7. Available from: https://doi.org/10.1196/annals.1345.002
3. WHO. Thalassemia data and statistic. 2018.
4. Ministry of Health. Thalassemia data and statistic. 2018.
5. Wong LP, George E, Tan J-AMA. Public perceptions and attitudes toward thalassaemia: Influencing factors in a multi-racial population. BMC Public Health [Internet]. 2011;11(1):193. Available from: https://doi.org/10.1186/1471-2458-11-193
6. Loukopoulos D. Haemoglobinopathies in Greece: prevention programme over the past 35 years. Indian J Med Res [Internet]. 2011 Oct;134(4):572–6. Available from: https://pubmed.ncbi.nlm.nih.gov/22089622
7. Gholizade L. Determine and compare the psychosocial problems of adolescents with thalassemia referred to medical centres with healthy adolescents selected from secondary schools and Kohkiluyeh and Boyer Ahmad province. [Thesis] Tehran: Shahid Beheshti University.; 2002.
8. Khani H. . Quality of life in Iranian Beta-thalassemia major patients of southern coastwise of the Caspian Sea. Int J Behav Sci [Internet]. 2009;2(4):325–32. Available from: http://www.behavsci.ir/article_67609.html
9. Pakbaz Z, Treadwell M, Yamashita R, Quirolo K, Foote D, Quill L, et al. Quality of life in patients with thalassemia intermedia compared to thalassemia major. Ann N Y Acad Sci. 2005;1054:457–61.
10. Mazzone L, Battaglia L, Andreozzi F, Romeo MA, Mazzone D. Emotional impact in β-thalassaemia major children following cognitive-behavioural

family therapy and quality of life of caregiving mothers. Clin Pract Epidemiol Ment Health. 2009 Mar 1;5:5.
11. Yazdi K, Sanagou A, Jouybari L. Psychosocial, social and financial problems with coping strategies among families with thalassemic patients. J Gorgan Univ Med Sci. 2007;9:71–5.
12. Borhani F, Najafi MK, Rabori ED, Sabzevari S. The effect of family-centered empowerment model on quality of life of school-aged children with thalassemia major. Iran J Nurs Midwifery Res. 2011;16(4):292–8.
13. Faramarzi S, Malekpour M. The effect of educational and psychological family-based early intervention on the motor development of children with down syndrome. 2009;
14. Knight L. Child health. Negotiating care roles. Nurs Times. 1995;91(27):31–3.
15. Boudreaux ED, Francis JL, Loyacano T. Family presence during invasive procedures and resuscitations in the emergency department: a critical review and suggestions for future research. Ann Emerg Med. 2002 Aug;40(2):193–205.
16. Hawks JH. Empowerment in nursing education: concept analysis and application to philosophy, learning and instruction. J Adv Nurs. 1992 May;17(5):609–18.
17. Harris LM. Empowerment and involvement. J Vasc Surg. 2018;67(4):997–1004.
18. Borimnejad PhD L, Parvizy PhD S, Haghaani PhD H, Sheibani MS B. The Effect of Family-Centered Empowerment Program on Self-Efficacy of Adolescents with Thalassemia Major: A Randomized Controlled Clinical Trial. Int J community based Nurs midwifery [Internet]. 2018 Jan;6(1):29–38. Available from: https://pubmed.ncbi.nlm.nih.gov/29344533
19. Heidari M, Alhani F, Kazemnejad A, Moezzi F. The effect of empowerment model on quality of life of Diabetic adolescents. Iran J Pediatr (ISSN 1018-4406) Vol 17 Num s1. 2007 Mar 1;17.
20. Nurhaeni N, Rustina Y, Agustini N, Enis Rosuliana N. Impact of family empowerment model on satisfaction and children's length of stay in hospital. Enferm Clin. 2018 Feb;28 Suppl 1:36–40.
21. Najafi KM, Borhani F, E DR, Sabzevari S. The Effect of Family-Centered Empowerment Model on the Mothers ' Knowledge and Attitudes about Thalassemia Disorder. Iran J Pediatr Hematol. 2011;1(3):98–103.
22. Wacharasin C, Phaktoop M, Sananreangsak S. Examining the usefulness of a Family Empowerment Program guided by the Illness Beliefs Model for families caring for a child with thalassemia. J Fam Nurs. 2015 May;21(2):295–321.
23. Wacharasin C, Phaktoop M, Sananreangsak S. A family empowerment program for families having children with thalassemia, Thailand. Nurs Health Sci. 2015 Sep;17(3):387–94.
24. Yeh H-Y, Ma W-F, Huang J-L, Hsueh K-C, Chiang L-C. Evaluating the effectiveness of a family empowerment program on family function and pulmonary function of children with asthma: A randomized control trial. Int J Nurs Stud. 2016 Aug;60:133–44.
25. Dunst C, Trivette C. Capacity-Building Family-Systems Intervention Practices. J Fam Soc Work. 2009 May 19;12:119–43.
26. Coyne I. Families and health-care professionals' perspectives and expectations of family-centred care: hidden expectations and unclear roles. Heal Expect an Int J public Particip Heal care Heal policy. 2015 Oct;18(5):796–808.
27. Tallon MM, Kendall GE, Snider PD. Rethinking family-centred care for the child and family in hospital. J Clin Nurs. 2015 May;24(9–10):1426–35.
28. Wacharasin C, Phaktoop M, Sananreangsak S, Teerarungsikul N, Tunsawad T. Evidence, problems, impacts, and empowerment in family caregivers of chronically ill children. J Public Heal Nurse, Fac Public Heal Mahidol Univ. 2008;22(23):25–39.
29. Greenberg P, Gordeuk V, Issaragrisil S, Siritanaratkul N, Fucharoen S, Ribeiro R. Major hematologic diseases in the developing world-new aspects of diagnosis and management of thalassemia, malarial anemia, and acute leukemia. Hematology Am Soc Hematol Educ Program. 2001 Feb 1;479–98.
30. Thavorncharoensap M, Torcharus K, Nuchprayoon I, Riewpaiboon A, Indaratna K, Ubol B-O. Factors affecting health-related quality of life in Thai children with thalassemia. BMC Blood Disord. 2010 Jan;10:1.
31. Caro JJ, Ward A, Green TC, Huybrechts K, Arana A, Wait S, et al. Impact of Thalassemia major on Patients and Their Families. Acta Haematol [Internet]. 2002;107(3):150–7. Available from: https://www.karger.com/DOI/10.1159/000057633
32. Angastiniotis M. The adolescent thalassemic. The complicant rebel. Minerva Pediatr. 2002 Dec;54(6):511–5.
33. Borgna-Pignatti C, De Stefano P, Zonta L, Vullo C, De Sanctis V, Melevendi C, et al. Growth and sexual maturation in thalassemia major. J Pediatr. 1985 Jan;106(1):150–5.
34. Roy T, Chatterjee SC. The experiences of adolescents with thalassemia in West Bengal, India. Qual Health Res. 2007 Jan;17(1):85–93.
35. Atkin K, Ahmad WIU. Family care-giving and chronic illness: how parents cope with a child with a sickle cell disorder or thalassaemia. Heal \& Soc care community. 2000;8(1):57–69.
36. Prasomsuk S, Jetsrisuparp A, Ratanasiri T, Ratanasiri A. Lived experiences of mothers caring

for children with thalassemia major in Thailand. J Spec Pediatr Nurs. 2007 Jan;12(1):13–23.
37. Kukreja A, Khan A, Xian LT, Razley A, Rahim Z. Awareness Of Thalassaemia Among Rural Folks In Penang, Malaysia. Internet J Heal. 2009;12.
38. Ailinger RL, Martyn D, Lasus H, Lima Garcia N. The effect of a cultural intervention on adherence to latent tuberculosis infection therapy in Latino immigrants. Public Health Nurs. 2010;27(2):115–20.
39. Buettner-Schmidt K, Lobo ML. Social justice: A concept analysis. J Adv Nurs. 2012;68(4):948–58.
40. Widayanti CG, Ediati A, Tamam M, Faradz SMH, Sistermans EA, Plass AMC. Feasibility of preconception screening for thalassaemia in Indonesia: Exploring the opinion of Javanese mothers. Ethn Heal. 2011;16(4–5):483–99.
41. Dubey AP, Parakh A, Dublish S. Current trends in the management of beta thalassemia. Indian J Pediatr. 2008 Jul;75(7):739–43.
42. Cousens NE, Gaff CL, Metcalfe SA, Delatycki MB. Carrier screening for beta-thalassaemia: a review of international practice. Eur J Hum Genet. 2010 Oct;18(10):1077–83.
43. Koren, A., Profeta, L., Zalman, L., Palmor, H., Levin, C., Zamir, R.B., Shalev, S., Blondheim O. Prevention of -thalassemia in Northern Israel—A Cost-Benefit Analysis. Mediterr J Hematol Infect Dis. 2014;6: e201401