Vol. 4. No.2. 2026 E-ISSN 2987- 2081

Holistic Nursing PLUS
Pillars, Longevity, Ubiquitary, Solicitude

SAKU
SAHABAT PUBLIKASI KUU

Child, Parental, and Family-Related Characteristics Associated with Caregiver Burden Among Parents of Children With β-Thalassemia: A Cross-Sectional Study

Nanang Saprudin[1,2] and Devi Nallappan[1]

[1] School of Health & Applied Science, Lincoln University College, Petaling Jaya, Selangor, Malaysia
[2] Faculty of Nursing, Universitas Bhakti Husada Indonesia, Kuningan, West Java, Indonesia

Academic Editor: Rika Monika
Received: 15 August 2026
Accepted: 21 September 2026
Published: 31 October 2026

Publisher's Note: HNP stays neutral with regard to jurisdictional claims in published maps and institutional affiliations.

Copyright: © 2026 by the authors. Submitted for possible open access publication under the terms and conditions of the Creative Commons Attribution (CC BY) license (https://creativecommons.org/licenses/by/4.0/).

Abstract

Background: Transfusion-dependent thalassemia is a chronic blood disorder that can affect psychological well-being and quality of life among patients and their families.**Purpose:** This systematic review examined the psychological challenges and quality-of-life concerns experienced by patients with transfusion-dependent thalassemia and their families.

Methods: A systematic review was conducted using the Preferred Reporting Items for Systematic Reviews and Meta-Analyses framework. Scopus, ProQuest, ScienceDirect, and Taylor & Francis were searched for relevant studies published from 2022 through December 2024. Eligible studies used standardized, validated instruments to assess psychological health or quality of life in patients with transfusion-dependent thalassemia and their families and employed a cross-sectional design.

Results: Eleven studies met the inclusion criteria. Patients experienced psychological difficulties, including anxiety, depression, panic symptoms, social phobia, and problems with self-concept. Children also exhibited behavioral difficulties, attention-related problems, and challenges in peer relationships. Quality-of-life concerns included impaired school functioning, emotional well-being, social participation, physical health, pain, fatigue, and work limitations. Families and caregivers experienced emotional distress, financial strain, and challenges associated with regular blood transfusions. Caregivers also reported feelings of discouragement and powerlessness, underscoring the demands of long-term care.

Conclusions: Transfusion-dependent thalassemia is associated with psychological challenges and reduced quality of life among patients and their families. Comprehensive, family-centered support may help address these multidimensional needs. Further research should evaluate interventions that support psychological well-being and quality of life in both patients and caregivers.

Keywords: Thalassemia; Quality of Life; Mental Health; Caregivers; Family

Introduction

β-Thalassemia is an inherited hemoglobin disorder that can require lifelong treatment and remains an important health concern in regions where the disease is prevalent, including Southeast Asia. Children with transfusion-dependent β-thalassemia require regular blood transfusions, iron-chelation therapy, and continuing monitoring of hematologic and iron status. Consequently, the

effects of the disease extend beyond the child and create sustained responsibilities for parents and other family members (Aydinok et al., 2024; Musallam et al., 2023).

Parents may need to accompany children to transfusion and follow-up appointments, administer or supervise medications and iron-chelation therapy, monitor symptoms, and provide ongoing emotional and practical support. These responsibilities may interfere with employment, household routines, social participation, and family relationships. Previous research involving caregivers of children with thalassemia has documented psychological distress, reduced quality of life, emotional strain, and substantial practical challenges associated with long-term disease management (Dewi et al., 2024; Othman et al., 2022; Sahu et al., 2023). Caregiver burden is therefore multidimensional and may encompass emotional, physical, financial, and social demands rather than reflecting only the severity or frequency of children's clinical needs.

The caregiving experience is also embedded within the resources and circumstances of the family. Parental age, education, employment, health status, household income, and family or spousal support may influence the time, financial resources, and organizational capacity available for caregiving. Studies among parents of children with chronic conditions have linked caregiver burden with social support, perceived stress, family resources, and socioeconomic circumstances (Alay & Kaçan, 2024; Almulla et al., 2024; Shattnawi et al., 2023). Employment may have particularly complex implications. It may provide financial and social resources while also creating competing demands and limiting flexibility for repeated healthcare appointments (Stevens et al., 2024).

Child-related characteristics may also contribute to caregiving demands. Transfusion requirements, iron-chelation therapy, duration of illness, and other clinical characteristics represent continuing treatment responsibilities. Nevertheless, clinical characteristics alone may not explain differences in parental burden. Evidence from β-thalassemia populations indicates that quality of life and psychosocial outcomes may be shaped by interactions between disease-related demands and family circumstances (Hood et al., 2024; Mardhiyah & Panduragan, 2024; Zhang et al., 2024).

Despite increasing recognition of the psychosocial consequences of thalassemia, characteristics associated with caregiver burden among parents of children with β-thalassemia remain incompletely characterized. Previous studies have frequently examined caregiver burden, psychological well-being, quality of life, or selected clinical characteristics separately rather than

considering child-, parental-, and family-related domains simultaneously. Research among adults with β-thalassemia has also demonstrated that caregiver burden is multidimensional and associated with sociodemographic and clinical circumstances (Yasmeen et al., 2024; Zhang et al., 2024). An integrated examination of these domains may therefore provide a more useful basis for understanding parental needs within family-centered nursing care.

This issue has direct relevance to nursing practice. Pediatric nurses have repeated opportunities to interact with children and parents during transfusion and follow-up encounters. These encounters provide opportunities to identify parents experiencing substantial caregiving demands and to initiate education, psychosocial support, or referral to family and community resources. Family-centered nursing emphasizes partnership with parents and recognition of parental needs alongside the child's clinical care (Zia et al., 2025). From this perspective, identifying characteristics associated with caregiver burden may help nurses broaden assessment beyond disease-related indicators and consider the circumstances in which caregiving occurs.

Accordingly, this study examined the associations of child-, parental-, and family-related characteristics with parent-reported caregiver burden among parents of children with β-thalassemia receiving routine transfusion therapy. The examined characteristics included child sex, child age, duration of illness, iron-chelation therapy, pretransfusion hemoglobin concentration, annual transfusion frequency, recent transfusion frequency, responding-parent age, parental education, parental employment, parental chronic illness, family structure, and monthly household income.

Materials and Methods

Study Design and Setting

A quantitative observational study with a cross-sectional design was conducted from August 2025 to February 2026 at the Thalassemia Day Care Unit of a tertiary referral hospital in Indonesia. The unit had 30 beds and provided routine transfusion services for approximately 25–30 children with β-thalassemia per day. The study was reported in accordance with the Strengthening the Reporting of Observational Studies in Epidemiology (STROBE) recommendations (Elm et al., 2007).

The primary outcome was parent-reported caregiver burden, defined as the emotional, physical, financial, and social demands experienced by parents while caring for a child with β-thalassemia. Child-, parental-, and family-related characteristics were examined as candidate variables because of their potential relevance to caregiving demands and family functioning.

Participants and Sampling

The target population comprised parents of children with confirmed β-thalassemia who received regular transfusion therapy at the study site. Approximately 140 potentially eligible parents were identified during the predefined recruitment period. Parents were eligible if they were the biological or adoptive parent of a child with β-thalassemia receiving routine transfusion therapy, could understand the study information, and were willing to provide informed consent. Parents were excluded if their child had a documented neurological comorbidity or major congenital abnormality that could substantially influence caregiving demands or family functioning.

Of the approximately 140 potentially eligible parents, 22 were excluded during screening because they did not meet the eligibility criteria or declined participation. An additional 14 eligible parents were not recruited because they did not attend the study setting during the recruitment period. Consequently, 104 parents were consecutively recruited (Figure 1). Only one parent per child was included to avoid non-independent observations. The responding parent was defined as the parent who completed the study questionnaires and provided the parent-reported caregiver-burden assessment.

No separate a priori sample-size calculation was performed specifically for the regression analysis. The final sample represented the accessible eligible population recruited consecutively during the predefined study period. Accordingly, the precision and generalizability of the regression estimates were interpreted cautiously, particularly for characteristics with small numbers of observations.

Variables and Measures

The primary outcome was parent-reported caregiver burden. Candidate variables were classified into child-, parental-, and family-related characteristics based on their potential relevance to caregiving demands and family functioning.

Caregiver Burden

Caregiver burden was assessed using an 8-item instrument adapted from the conceptual domains of the Caregiver Strain Index (Robinson, 1983). Because the item structure and construct validity of the adapted instrument differed from those of the original scale, it was referred to throughout the study as an adapted caregiver-burden instrument rather than as the Caregiver Strain Index. The instrument was adapted for parents caring for children with β-thalassemia and addressed emotional, physical, financial, and social caregiving demands. The initial instrument contained 10 items rated on a 5-point Likert scale ranging from 1 (never) to 5 (always), with higher scores indicating greater caregiver burden.

Psychometric evaluation of the instrument was conducted in an independent sample and completed before the present study. Content validity was evaluated by six experts with expertise in pediatric nursing, family caregiving, chronic illness management, and thalassemia care. Item-level content validity indices ranged from .87 to 1.00, and the scale-level content validity index/average was .94. Construct validity was subsequently examined in an independent sample of 150 caregivers using principal axis factoring with Promax rotation. Sampling adequacy was supported by a Kaiser–Meyer–Olkin value of .819 and a significant Bartlett's test of sphericity, χ^2 (28) = 503.80, $p < .001$.

Parallel analysis supported a two-factor structure representing subjective and objective caregiver burden, accounting for 66.12% of the variance. Items 3 and 4 were removed according to the prespecified retention criteria, resulting in an 8-item instrument. Internal consistency was satisfactory, with Cronbach's α = .845 and McDonald's ω = .854. The total score of the final 8-item instrument was used as the primary outcome in the present study.

Child-Related Characteristics

Child-related variables included sex, age, duration of illness, iron-chelation therapy, pretransfusion hemoglobin concentration, annual transfusion frequency, and the number of transfusions received during the preceding 3 months. Pretransfusion hemoglobin concentration was categorized as <7, 7–9, and >9 g/dL. Annual transfusion frequency was categorized as <12, 12–16, and >16 transfusions per year. Clinical information was obtained from medical records and clarified with parents when necessary.

Parental Characteristics

Parental characteristics included responding-parent age, maternal and paternal educational levels, maternal and paternal employment status, and maternal and paternal chronic illness. Maternal employment status was categorized as private-sector employee, self-employed, civil servant, or homemaker/unemployed. Chronic illness was based on parental reports of documented ongoing health conditions.

Family Characteristics

Family characteristics included family structure and monthly household income. Family structure was categorized as two-parent or single-parent household. Monthly household income was categorized according to the predefined questionnaire categories.

Data Collection and Quality Control

Following written informed consent, participants completed the sociodemographic, parental, and family questionnaires and the caregiver-burden instrument during their child's scheduled visit to the Thalassemia Day Care Unit. The questionnaires were self-administered. Research team members clarified questionnaire items when necessary without influencing participants' responses.

Clinical information, including pretransfusion hemoglobin concentration, transfusion frequency, and treatment characteristics, was extracted from medical records. Medical-record information was checked for completeness and clarified with parents when necessary. Completed questionnaires were reviewed before data entry. All 104 participants had complete data for variables included in the final regression model; therefore, no missing-data imputation was required. Each participant was assigned a study code, and identifying information was stored separately from the analytic dataset.

Data Analysis

Data were analyzed using IBM SPSS Statistics version 24. Categorical variables were summarized using frequencies and percentages. Continuous variables were summarized using means and standard deviations or medians and interquartile ranges, as appropriate. Caregiver burden was treated as a continuous outcome.

Bivariate associations were examined using the Mann–Whitney U test for categorical variables with two groups and the Kruskal–Wallis H test for variables with three or more groups. Spearman's

rank-order correlation was used for ordinal or nonnormally distributed continuous variables, whereas Pearson's correlation was used for child age and the number of transfusions during the preceding 3 months.

Candidate variables for multivariable analysis were selected on the basis of both conceptual relevance to caregiver burden and a prespecified bivariate screening criterion of $p < .25$. This threshold was used solely for candidate-variable screening and was not interpreted as a criterion for statistical significance. Six candidate variables met the screening criterion: child sex, child age, maternal employment status, paternal chronic illness, maternal chronic illness, and family structure.

These variables were entered simultaneously into a multiple linear regression model using the enter method. Maternal employment status was represented by three dummy-coded terms, resulting in eight regression parameters in the original model. Regression coefficients were reported as unstandardized B values with bootstrap standard errors, 95% confidence intervals, and two-sided p values. Two thousand bootstrap samples were generated for interval estimation. Bias-corrected and accelerated confidence intervals were used when available; percentile intervals were used when BCa intervals could not be calculated.

Model performance was summarized using R, R^2, adjusted R^2, and the F statistic. Cohen's f^2 was calculated as $R^2/ (1 - R^2)$. Multicollinearity was evaluated using tolerance, variance inflation factor (VIF), and condition-index statistics. Residual diagnostics included the Shapiro–Wilk test, standardized residuals, Cook's distance, and centered leverage. Visual inspection of residual plots was used to assess linearity and homoscedasticity.

A leave-one-observation sensitivity analysis was subsequently performed by excluding the observation with the highest centered leverage (.501) and re-estimating the model. Because the self-employed and civil-servant subgroups each contained only two mothers, exclusion of the high-leverage observation most likely reduced one of these subgroups to a single observation; the corresponding regression parameter could then no longer be reliably estimated and was therefore omitted from the sensitivity model.

Consequently, the sensitivity model contained seven regression parameters rather than eight, and its results were interpreted as a sensitivity analysis of the fitted model rather than as a strictly identical leave-one-observation comparison. The sensitivity model was evaluated by comparing

coefficient estimates, significance levels, and overall model fit with the original model. Given the cross-sectional design, regression coefficients were interpreted as adjusted associations rather than causal effects. Statistical significance was defined as a two-sided $p < .05$.

Ethical Considerations

The study protocol was reviewed and approved by the institutional Research Ethics Subcommittee (Ref. No. 074/KEP/6/2025). The study procedures followed applicable ethical standards for research involving human participants and the principles of the Declaration of Helsinki. Participants received information about the study purpose, procedures, potential benefits and risks, confidentiality, and voluntary participation. Written informed consent was obtained from all participants before data collection.

Results

Participants and Caregiver Burden

A total of 104 parent–child dyads were included in the analysis, and complete data were available for all variables included in the statistical analyses (**Figure 1**). The mean parent-reported caregiver burden score was 20.10 (SD = 4.94), with a median of 20.50 (IQR = 8.00; range = 9–33). Among the 104 children, 60 (57.7%) were boys and 44 (42.3%) were girls. Mean child age was 10.37 years (SD = 4.28; range = 2–18 years). Most children had lived with β-thalassemia for more than 5 years (70.2%) and received deferiprone for iron chelation (81.7%). The mean number of transfusions during the preceding 3 months was 4.86 (SD = 1.56; range = 1–8), and most children had pretransfusion hemoglobin concentrations between 7 and 9 g/dL (73.1%).

Responding parents had a mean age of 38.40 years (SD = 7.30; range = 24–55 years). Primary education was the most common educational level among fathers (54.8%) and mothers (60.5%). Most fathers were unemployed or engaged in casual labor (42.3%), whereas 85.6% of mothers were homemakers or unemployed. Chronic illness was reported by 9.6% of fathers and 12.5% of mothers. Most families were two-parent households (94.2%). Monthly household income was below IDR 1,500,000 for 41.4% of families, IDR 1,500,000–2,500,000 for 26.9%, and >IDR 2,500,000–3,500,000 for 31.7% (**Table 1**).

Bivariate Screening

None of the child-related characteristics reached the conventional $p < .05$ threshold. However, child sex ($p = .101$) and child age ($p = .136$) met the prespecified $p < .25$ screening criterion. Iron-chelation therapy, duration of illness, pretransfusion hemoglobin level, annual transfusion frequency, and the number of transfusions during the preceding 3 months did not meet the screening criterion.

Among parental characteristics, maternal employment status ($p = .010$), paternal chronic illness ($p = .014$), and maternal chronic illness ($p = .056$) met the screening criterion. Father's education, mother's education, father's employment status, and parent age did not meet the criterion. Family structure met the screening criterion ($p = .037$), whereas monthly household income did not ($p = .892$), and therefore was not entered into the multivariable model. Thus, six variables were retained for multivariable analysis (**Table 2**).

Multiple Linear Regression

The original multiple linear regression model was statistically significant, $F (8, 95) = 3.569$, $p = .001$, with $R = .481$, $R^2 = .231$, and adjusted $R^2 = .166$. The regression parameters collectively explained 23.1% of the variance in parent-reported caregiver burden, corresponding to a Cohen's f^2 of .300.

After simultaneous adjustment, child sex was not independently associated with caregiver burden ($B = 1.948$, 95% CI [−0.004, 4.208], $p = .051$). Child age was also not associated with caregiver burden ($B = 0.007$, 95% CI [−0.191, 0.205], $p = .941$). Paternal chronic illness showed a positive but nonstatistically significant association ($B = 3.484$, 95% CI [−0.584, 7.089], $p = .058$), as did maternal chronic illness ($B = 2.446$, 95% CI [−0.414, 5.240], $p = .094$). Family structure was not independently associated with caregiver burden ($B = 2.887$, 95% CI [−0.597, 6.995], $p = .112$).

Maternal employment status showed statistically significant associations in the original model. Compared with the homemaker/unemployed category, caregiver burden was lower when the mother was self-employed ($B = −4.498$, 95% CI [−7.782, −1.276], $p = .009$) or a civil servant ($B = −8.020$, 95% CI [−12.796, −3.627], $p = .001$). The association for private-sector employment was not statistically significant ($B = 1.451$, 95% CI [−0.920, 3.728], $p = .287$). Because only two mothers were classified as self-employed and two as civil servants, these estimates should be regarded as exploratory rather than definitive, and were interpreted cautiously. Detailed regression coefficients, confidence intervals, and sensitivity estimates are presented in **Table 3**.

Sensitivity Analysis

The maximum centered leverage was .501. After exclusion of the corresponding observation and re-estimation of the model, the sensitivity model remained statistically significant, $F(7, 94) = 3.111$, $p = .005$, with $R^2 = .188$ and adjusted $R^2 = .128$. However, several individual associations changed.

The coefficient for child sex changed from B = 1.948 (p = .051) in the original model to B = 2.099 (p = .038) in the sensitivity model. Paternal chronic illness changed from B = 3.484 (p = .058) to B = 3.508 (p = .028). In contrast, the association for maternal self-employment was attenuated, changing from B = −4.498 (p = .009) to B = −4.461 (p = .187), and was no longer statistically significant. Child age remained nonstatistically significant (B = 0.002, p = .983), as did maternal chronic illness (B = 2.477, p = .075), family structure (B = 2.913, p = .150), and private-sector maternal employment (B = 1.448, p = .340).

Because the sensitivity model retained an estimate for the self-employed category but not for the civil-servant category, the excluded high-leverage observation most likely belonged to the civil-servant subgroup. With only one civil-servant observation remaining, the corresponding regression parameter could not be reliably estimated and was therefore omitted from the sensitivity model (denoted NE, not estimable, in **Table 3**).

Because the sensitivity analysis involved both exclusion of the high-leverage observation and a reduction in the number of regression parameters, the changes in individual coefficients should be interpreted cautiously and should not be regarded as a pure influence analysis. No individual association demonstrated consistent statistical significance across the original and sensitivity models. The clearest and most defensible finding was that the overall multivariable model remained statistically significant after sensitivity analysis, although the associations for individual characteristics were sensitive to the influential observation and to the small employment subgroups.

Regression Diagnostics

There was no evidence of substantial multicollinearity. Tolerance values ranged from .857 to .992, VIF values ranged from 1.008 to 1.167, and the maximum condition index was 6.872. The Shapiro–Wilk test did not indicate a statistically significant departure from residual normality (p = .251), and no substantial departure from residual normality was otherwise detected. Standardized residuals ranged from −2.009 to 2.983, and the maximum Cook's distance was .143. The maximum centered

leverage was .501; although leverage is not equivalent to an outlier, this value indicated a high-leverage observation that warranted the sensitivity analysis described above. Visual inspection of the residual plots showed no apparent systematic curvature or pronounced funnel-shaped pattern, supporting the assumptions of linearity and homoscedasticity. Nevertheless, the relatively high leverage of one observation and the small employment subgroups warranted cautious interpretation of individual regression coefficients.

Discussion

This study examined the associations of child-, parental-, and family-related characteristics with parent-reported caregiver burden among parents of children with transfusion-dependent β-thalassemia. The overall regression model was statistically significant and explained 23.1% of the variance in caregiver burden. Maternal employment status showed statistically significant associations in the original adjusted model, whereas child characteristics, parental chronic illness, and family structure did not reach statistical significance in that model. However, the employment associations should be interpreted cautiously because the relevant subgroups were very small, and the maternal self-employment association was not stable in sensitivity analysis. These findings suggest that caregiver burden may be better understood within the broader parental and family context than through children's clinical characteristics alone (Almulla et al., 2024; Heller & Melnikov, 2024; Liu et al., 2023).

The observed burden is consistent with the continuing demands associated with caring for a child with thalassemia. Parents must accommodate repeated transfusion visits, iron-chelation therapy, symptom-related care, and the emotional and practical demands associated with long-term caregiving (Dewi et al., 2024; Klaassen et al., 2026). Previous studies have documented psychological distress, reduced quality of life, emotional strain, and practical difficulties among parents and caregivers of individuals with (Othman et al., 2022; Sahu et al., 2023; Yasmeen et al., 2024). Qualitative evidence from Indonesia similarly identified substantial emotional and practical challenges among mothers caring for children with thalassemia (Dewi et al., 2024). The present findings extend this perspective by suggesting that variation in caregiver burden may not

correspond closely with routine clinical characteristics among children receiving ongoing transfusion therapy.

Maternal Employment

Maternal employment showed the strongest adjusted associations in the original model. Compared with homemaker/unemployed mothers, caregiver burden was lower among mothers who were self-employed or civil servants. Employment, however, should not be interpreted as a protective factor. It may represent several interconnected socioeconomic resources, including income, social engagement, autonomy, employment benefits, and greater control over household resources (Cohn et al., 2020). Conversely, employment may increase competing demands, reduce flexibility, and make repeated treatment attendance more difficult (Stevens et al., 2024).

The small size of the employment subgroups substantially limits interpretation. Only two mothers were self-employed and two were civil servants, making the corresponding estimates potentially sensitive to individual observations. This concern is supported by the sensitivity analysis, in which the association for maternal self-employment was no longer statistically significant after exclusion of the high-leverage observation, although the overall regression model remained significant. Thus, the findings are better viewed as an indication that work and socioeconomic circumstances may be relevant to caregiver burden than as evidence of an employment-related effect.

For nursing practice, employment status should therefore not be used alone to classify caregiver risk. Assessment should explore working hours, job flexibility, income adequacy, workplace support, leave arrangements, and the ability to attend transfusion (Hellmers et al., 2025). These factors may provide more meaningful information about the practical resources and demands experienced by families.

Child, Parental, and Family Characteristics

No child-related characteristic was independently associated with caregiver burden in the original model. This finding does not imply that treatment demands are unimportant. Transfusion-dependent β-thalassemia involves continuing treatment and disease-management requirements that may affect quality of life and family functioning (Aydinok et al., 2024; Drahos et al., 2024; Lee et al., 2024). Rather, routine clinical indicators may have limited value for distinguishing parents

with greater burden within a population exposed to similarly intensive treatment. Psychosocial resources, coping, family functioning, social support, financial circumstances, and caregiving responsibilities may contribute to differences in burden but were not comprehensively assessed in this study.

Parental chronic illness was also not independently associated with burden in the original model, although both maternal and paternal chronic illness showed positive coefficients. The small number of affected parents limits statistical precision. Future studies should examine not only the presence of chronic illness but also its severity, functional consequences, and effects on caregiving responsibilities.

Family structure was associated with burden at the bivariate level but not after adjustment. This suggests that household composition may be less informative than the actual availability and distribution of support. Most families were two-parent households, but this does not necessarily indicate equal sharing of healthcare responsibilities. Previous research has identified differences in parental responsibility for children's healthcare and highlighted the potential relevance of spousal support and parent–nurse partnership (Heffernan et al., 2024; Kim et al., 2024). Nursing assessment should therefore focus on who manages treatment, accompanies the child, contributes financially, provides emotional support, and has opportunities for respite.

Nursing Implications

These findings support family-centered caregiver assessment during routine thalassemia care. Repeated transfusion encounters provide opportunities for nurses to identify parents experiencing substantial caregiving demands. Assessment should include socioeconomic circumstances, parental health, caregiving-role distribution, social support, and financial strain in addition to the child's clinical status. Individualized education, parent–nurse partnerships, and connections with psychosocial and community resources may further support parental coping and quality of life (Mardhiyah et al., 2025; Yildirim et al., 2022). Caregiver-burden assessment may therefore serve as an entry point for individualized family-centered support rather than simply as a method of risk classification.

Strengths and Limitations

A strength of this study was the simultaneous consideration of child-, parental-, and family-related characteristics. The adapted caregiver-burden measure demonstrated satisfactory measurement properties in an independent sample, and bootstrap estimation provided additional information for evaluating coefficient uncertainty. Nevertheless, the cross-sectional design prevents conclusions regarding temporal or causal relationships, and recruitment from a single tertiary hospital may limit transferability. Several predictor categories contained very small subgroups, and no separate a priori sample-size calculation was performed specifically for the regression analysis. In addition, the model explained 23.1% of the variance, indicating that important psychosocial and family-level factors were not captured. Finally, one observation had relatively high leverage. Although the overall model remained statistically significant after sensitivity analysis, changes in individual coefficients indicate that some associations were not robust. Larger multicenter and longitudinal studies incorporating psychosocial, socioeconomic, family-functioning, and caregiving variables are warranted.

Conclusion

Caregiver burden among parents of children with transfusion-dependent β-thalassemia should be considered within a broader parental and family context. The overall multivariable model was statistically significant, but individual associations were not consistently robust across sensitivity analysis. Maternal employment showed statistically significant associations in the original adjusted model; however, the very small employment subgroups and the sensitivity analysis indicate that these estimates require cautious interpretation. Child characteristics, parental chronic illness, and family structure were not independently associated with burden in the original model. Family-centered nursing assessment should therefore consider socioeconomic circumstances, caregiving responsibilities, parental health, and available support alongside children's clinical characteristics. Further multicenter longitudinal research is needed to identify potentially modifiable factors that can guide targeted nursing support.

Declaration of Interest

The authors declare that they have no known financial or personal relationships that could have appeared to influence the work reported in this article.

Acknowledgments

The authors acknowledge the parents and children who participated in this study and the healthcare personnel who supported data collection at the Thalassemia Day Care Unit.

Funding

This research received no specific grant from any funding agency in the public, commercial, or not-for-profit sectors.

Data Availability

The data supporting the findings of this study are available from the corresponding author upon reasonable request. The data are not publicly available because of ethical restrictions and confidentiality requirements to protect participant privacy.

References

Alay, G. K., & Kaçan, H. (2024). Care burden and resilience in parents of children with special needs and chronic diseases. *Journal of Pediatric Nursing*, *76*, 61–67. https://doi.org/10.1016/j.pedn.2024.02.001

Almulla, H., Aljaloud, O., Almulla, H., & Nasser, S. (2024). Caregiver burden , perceived stress , and social support among parents of chronically Ill children in Saudi Arabia. *BMC Nursing*, *23*(1), 4–11. https://doi.org/10.1186/s12912-024-02494-8.

Aydinok, Y., Purushotham, S., Yucel, A., Glassberg, M., & Deshpande, S. (2024). Systematic literature review of the indirect costs and humanistic burden of β -thalassemia. *Therapeutic Advances in Hematology*, *15*, 1–16. https://doi.org/10.1177/20406207241270872

Cohn, L. N., Pechlivanoglou, P., Lee, Y., Mahant, S., Orkin, J., Marson, A., & Cohen, E. (2020). Health Outcomes of Parents of Children with Chronic Illness: A Systematic Review and Meta-Analysis. *The Journal of Pediatrics*, *218*, 166–177. https://doi.org/10.1016/j.jpeds.2019.10.068

Dewi, N. H., Novieastari, E., Tutik, R., & Hariyati, S. (2024). Exploring experiences of mothers of children with thalassemia major in Indonesia : A descriptive phenomenological study. *Belitung Nursing Journa*, *10*(5), 585–592. https://doi.org/10.33546/bnj.3142

Drahos, J., Boateng, A., Melanie, K., Levine, L., & Dongha, N. (2024). Health - Related Quality - of - Life Impacts Associated with Transfusion - Dependent β - Thalassemia in the USA and UK : A Qualitative Assessment. *The Patient - Patient-Centered Outcomes Research*, *17*(4), 421–439. https://doi.org/10.1007/s40271-024-00678-7

Elm, E. von, Altman, D. G., Egger, M., Pocock, S. J., Gøtzsche, P. C., & Vandenbroucke, J. P. (2007). The Strengthening the Reporting of Observational Studies in Epidemiology (STROBE) statement: guidelines for reporting observational studies. *The Lancet*, *370*, 1453–1457. https://doi.org/10.1016/S0140-6736(07)61602-X

Heffernan, M. E., Alfieri, N. L., Keese, A., Bendelow, A. C., Casale, M., Smith, T. L., Menker, C. G., Parker, J. J., Garfield, C. F., Davis, M. M., & Macy, M. L. (2024). Differences in responsibility for child healthcare by parent gender: A cross-sectional study. *Social Science & Medicine*, *365*. https://doi.org/10.1016/j.socscimed.2024.117576

Heller, N., & Melnikov, S. (2024). Factors affecting caregiver burden among parents of children with cancer : A path analysis. *Journal of Clinical Nursing*, *34*(1), 128–138. https://doi.org/10.1111/jocn.17202

Hellmers, N., Stimpfel, A. W., Fletcher, J., & Dickson, V. V. (2025). Multi-contextual Factors that Influence Work-Life Balance of Family Caregivers of Adults with Parkinson's Disease. *Journal of Occupational and Environmental Medicine*, *67*(2), 1–25. https://doi.org/10.1097/JOM.0000000000003283.

Hood, A. M., Chaman, A., Chen, Y., & Mufti, S. (2024). Psychological challenges and quality of life in Pakistani parents of children living with thalassemia. *Journal of Pediatric Nursing*, *76*, 132–139. https://doi.org/10.1016/j.pedn.2024.02.015

Kim, J., Chae, H., & Kim, Y. (2024). Spousal support, parent–nurse partnership and caregiver burden among parents of children with chronic diseases: A cross-sectional study. *Journal of Clinical Nursing*, *33*(7), 2649–2661. https://doi.org/10.1111/jocn.16985

Klaassen, R. J., Khelifi, R. S., Barrowman, N., Merelles-Pulcini, M., Vichinsky, E. P., Kirby-Allen, M., Neufeld, E. J., Kwiatkowski, J. L., Wu, J., & Young, N. L. (2026). Taking Care of the Caregivers: A Quality-of-Life Measure for the Parents of Children With Transfusion-Dependent Thalassemia. *Pediatric Blood & Cancer*, *73*(2), e32163. https://doi.org/10.1002/pbc.32163

Lee, W. J., Ain, N., Tahir, M., Chun, G. Y., & Li, S. C. (2024). The impact of chelation compliance in health outcome and health related quality of life in thalassaemia patients : a systematic review. *Health and Quality of Life Outcomes*, 1–14. https://doi.org/10.1186/s12955-023-02221-y

Liu, F., Shen, Q., Huang, M., & Zhou, H. (2023). Factors associated with caregiver burden among family caregivers of children with cerebral palsy : a systematic review. *BMJ Open*, *13*(4), 1–12. https://doi.org/10.1136/bmjopen-2022-065215

Mardhiyah, A., & Panduragan, S. L. (2024). Factors Associated With Quality of Life Among Adolescent With Beta Thalassemia in Indonesia : A Cross-Sectional Study. *SAGE Open Nursing*, *10*, 7. https://doi.org/10.1177/23779608241255638

Mardhiyah, A., Yosep, I., Hendrawati, S., Rakhmawati, W., Khoirunnisa, K., & Eriyani, T. (2025). Nursing interventions for improving quality of life among parents with major Thalassemic children : a scoping review. *BMC Nursing*, *24*, 1–12. https://doi.org/10.1186/s12912-025-02994-1

Musallam, K. M., Lombard, L., Kistler, K. D., Arregui, M., Gilroy, K. S., Chamberlain, C., Zagadailov, E., Ruiz, K., & Taher, A. T. (2023). Epidemiology of clinically significant forms of alpha- and beta-thalassemia: A global map of evidence and gaps. *American Journal of Hematology*, *98*(9), 1436–1451. https://doi.org/10.1002/ajh.27006

Othman, A., Abdul Ghani, M. S. A., Taib, F., & Mohamad, N. (2022). Psychological distress and coping strategies among the caretakers of children with transfusion-dependent thalassemia.

Frontiers in Pediatrics, *10*. https://doi.org/10.3389/fped.2022.941202

Robinson, B. C. (1983). Validation of a Caregiver Strain Index. *Journal of Gerontology*, *38*(3), 344–348. https://doi.org/10.1093/geronj/38.3.344

Sahu, S., Agrawal, A., Shrivastava, J., & Tonk, S. (2023). Psychiatric disorders and caregiver burden in children with transfusion dependent β-thalassaemia and their caregivers. *World Journal of Clinical Pediatrics*, *12*(3), 125–132. https://doi.org/10.5409/wjcp.v12.i3.125

Shattnawi, K. K., Ali, N. Al, Almanasreh, A. A., & Al-Motlaq, M. A. (2023). Caregiver burden among parents of children with chronic diseases: A cross-sectional study. *Journal of Clinical Nursing*, *32*(17–18), 6485–6493. https://doi.org/10.1111/jocn.16672

Stevens, E. K., Aziz, S., Wuensch, K. L., & Walcott, C. (2024). Caregivers of Children with Special Healthcare Needs : A Quantitative Examination of Work-Family Culture , Caregiver Burden , and Work- Life Balance. *Journal of Child and Family Studies*, *33*, 1365–1377. https://doi.org/10.1007/s10826-024-02822-1

Yasmeen, H., Khan, H. R., & Hasnain, S. (2024). Exploring caregiver burden of thalassemia major patients. *Proceedings of the Pakistan Academy of Sciences: B. Life and Environmental Sciences, 61*(2), 207–215. https://doi.org/10.53560/PPASB(61-2)941

Yildirim, M., Yayan, E. H., Dağ, Y. S., Sülün, A., & Akyay, A. (2022). The relationship between the care burden and quality of life of parents who have children with hematological problems and their perceived social support. *Journal of Pediatric Nursing*, *63*, e107–e112. https://doi.org/10.1016/j.pedn.2021.10.004

Zhang, R., Zhang, S., Ming, J., Xie, J., Liu, B., Jiang, W., Fu, Y., Zhen, X., & Sun, X. (2024). A cross-sectional study: caregiver burden and related determinants of adult patients with β-thalassemia major in mainland China. *BMC Nursing*, *23*(1), 1–17. https://doi.org/10.1186/s12912-024-01826-y

Zia, S. K., Aghamohammadi, M., Moshfeghi, S., & Vosoghi, N. (2025). Examining the relationship between nurses ' professional self-efficacy and parents ' perception of family-centered care. *BMC Nursing*, *24*. /https://doi.org/10.1186/s12912-025-03112-x

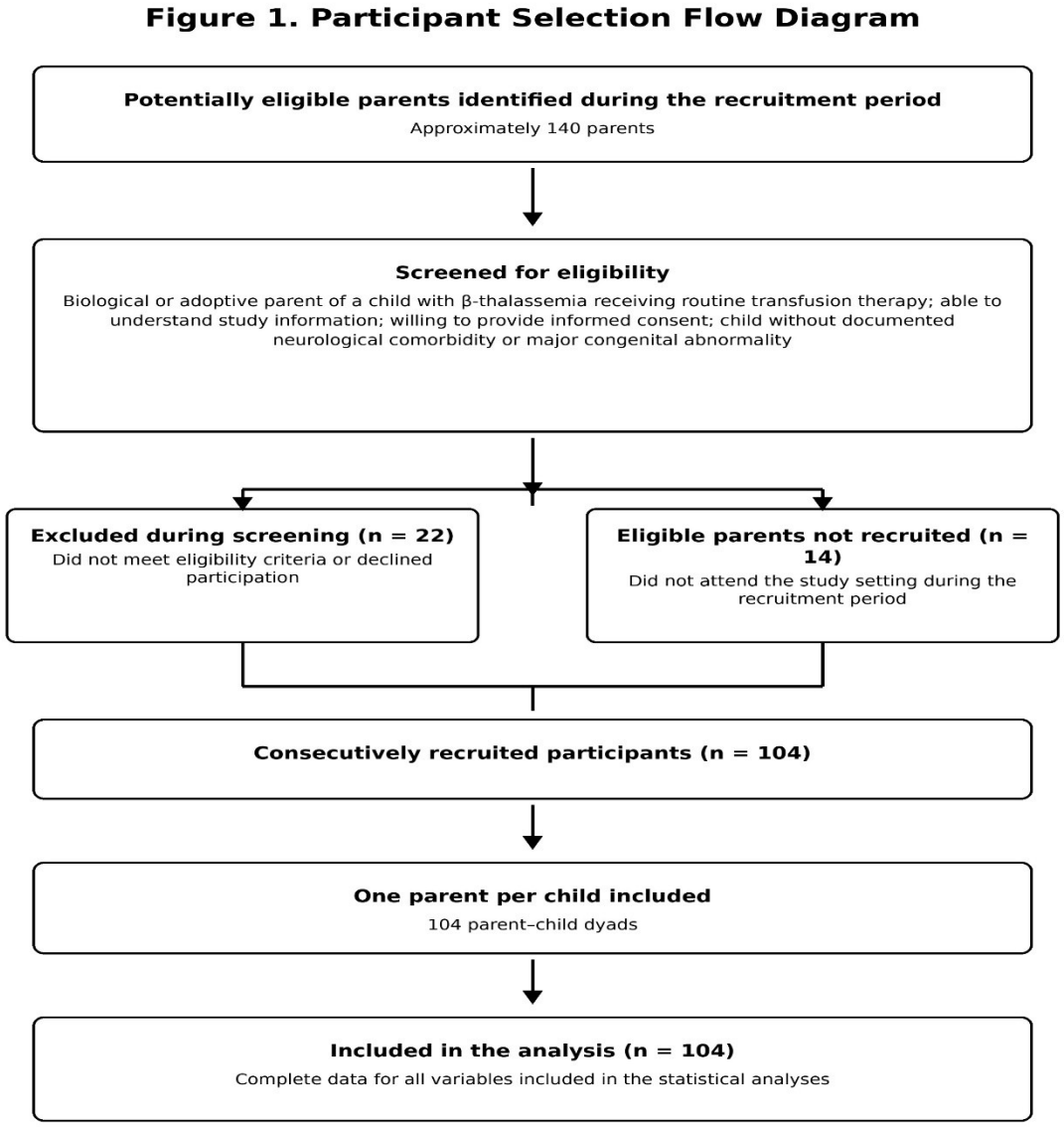


Figure 1. *Participant Selection Flow Diagram*

Table 1

Participant Characteristics and Parent-Reported Caregiver Burden (N = 104)

Characteristic	n (%) or M ± SD	Range
Parent-reported caregiver burden, M ± SD	20.10 ± 4.94	9–33
Median (IQR)	20.50 (8.00)	—
Child sex		
Male	60 (57.7)	—
Female	44 (42.3)	—
Child age, years, M ± SD	10.37 ± 4.28	2–18
Iron-chelation therapy		
Deferasirox	19 (18.3)	—
Deferiprone	85 (81.7)	—
Duration of illness		
<1 year	3 (2.9)	—
1–5 years	28 (26.9)	—
>5 years	73 (70.2)	—
Pretransfusion hemoglobin level, g/dL		
<7	17 (16.3)	—
7–9	76 (73.1)	—
>9	11 (10.6)	—
Annual transfusion frequency		
<12 transfusions	23 (22.1)	—
12–16 transfusions	35 (33.7)	—
>16 transfusions	46 (44.2)	—
Transfusions during previous 3 months, M ± SD	4.86 ± 1.56	1–8
Father's educational level		
Higher education	14 (13.5)	—
Secondary education	33 (31.7)	—
Primary education	57 (54.8)	—
Mother's educational level		
Higher education	14 (13.5)	—
Secondary education	27 (26.0)	—
Primary education	63 (60.5)	—
Father's employment status		
Private-sector employee	14 (13.5)	—
Self-employed	38 (36.5)	—
Civil servant	8 (7.7)	—
Unemployed/casual laborer	44 (42.3)	—
Maternal employment status		
Private-sector employee	11 (10.6)	—
Self-employed	2 (1.9)	—
Civil servant	2 (1.9)	—
Homemaker/unemployed	89 (85.6)	—
Paternal chronic illness		
No	94 (90.4)	—
Yes	10 (9.6)	—
Maternal chronic illness		
No	91 (87.5)	—
Yes	13 (12.5)	—
Responding-parent age, years, M ± SD	38.40 ± 7.30	24–55
Family structure		
Two-parent household	98 (94.2)	—
Single-parent household	6 (5.8)	—
Monthly household income		
<IDR 1,500,000	43 (41.4)	—
IDR 1,500,000–2,500,000	28 (26.9)	—
>IDR 2,500,000–3,500,000	33 (31.7)	—

Note. IQR = interquartile range; M = mean; SD = standard deviation; IDR = Indonesian rupiah. Percentages are based on N = 104. Higher caregiver-burden scores indicate greater burden.

Table 2

Bivariate Screening of Candidate Variables for Parent-Reported Caregiver Burden

Candidate variable	Bivariate p	Screening decision
Child sex	.101	Retained
Child age, years	.136	Retained
Maternal employment status	.010	Retained
Paternal chronic illness	.014	Retained
Maternal chronic illness	.056	Retained
Family structure	.037	Retained
Iron-chelation therapy	.753	Not retained
Duration of illness	.967	Not retained
Pretransfusion hemoglobin level	.419	Not retained
Annual transfusion frequency	.303	Not retained
Transfusions during previous 3 months	.822	Not retained
Father's educational level	.353	Not retained
Mother's educational level	.804	Not retained
Father's employment status	.788	Not retained
Responding-parent age	.405	Not retained
Monthly household income	.892	Not retained

Note. Variables with $p < .25$ were retained as candidate variables for the multivariable model. The screening threshold was used only for candidate-variable selection and was not interpreted as a test of statistical significance; candidate variables were selected on the basis of both conceptual relevance to caregiver burden and this prespecified screening criterion. Bivariate p values were obtained using the Mann–Whitney U test, Kruskal–Wallis H test, Spearman's rank-order correlation, or Pearson's correlation, as appropriate to variable type.

Table 3

Multiple Linear Regression of Parent-Reported Caregiver Burden and Sensitivity Analysis

Predictor	Original B	Bootstrap SE	95% CI	p	Sensitivity B	Sensitivity p
Child sex	1.948	0.988	−0.004 to 4.208	.051	2.099	.038
Child age, years	0.007	0.100	−0.191 to 0.205	.941	0.002	.983
Paternal chronic illness	3.484	1.928	−0.584 to 7.089	.058	3.508	.028
Maternal chronic illness	2.446	1.481	−0.414 to 5.240	.094	2.477	.075
Family structure	2.887	2.096	−0.597 to 6.995	.112	2.913	.150
Maternal employment: private-sector employee	1.451	1.396	−0.920 to 3.728	.287	1.448	.340
Maternal employment: self-employed	−4.498	1.764	−7.782 to −1.276	.009	−4.461	.187
Maternal employment: civil servant	−8.020	2.811	−12.796 to −3.627	.001	NE	NE

Note. Dependent variable = parent-reported caregiver burden score. B = unstandardized regression coefficient after simultaneous adjustment for all variables; SE = standard error; CI = confidence interval; NE = not estimable. Reference category for maternal employment = homemaker/unemployed. Bootstrap estimates were based on 2,000 samples. The confidence intervals for the self-employed and civil-servant categories were estimated using the percentile method because BCa intervals could not be computed from the available jackknife samples. p values are two-tailed.

Original model: $R = .481$, $R^2 = .231$, adjusted $R^2 = .166$, $F(8, 95) = 3.569$, $p = .001$, Cohen's $f^2 = .300$. Sensitivity model after exclusion of the observation with centered leverage = .501: $R^2 = .188$, adjusted $R^2 = .128$, $F(7, 94) = 3.111$, $p = .005$. The civil-servant employment parameter was not estimable (NE) in the sensitivity model because only one civil-servant observation remained after exclusion of the high-leverage case; the sensitivity model therefore contained seven regression parameters and should not be interpreted as a strictly identical leave-one-observation analysis.