Oral health-related quality of life and orthodontic treatment need in thalassemia major patients

Adel Tabesh¹, Fatemeh Abbasi¹, Mojgan Shavakhi², Mahboobeh Mahmood³

¹Dental Research Center, Department of Oral Medicine, Dental Research Institute, Isfahan University of Medical Sciences, Isfahan, Iran, ²Dental Research Center, Department of Orthodontics, Dental Research Institute, Isfahan University of Medical Sciences, Isfahan, Iran, ³Department of Restorative Dentistry, School of Dentistry, Tehran University of Medical Sciences, Tehran, Iran

ABSTRACT

Background: Thalassemia major (TM) is a severe life-threatening hemoglobinopathy. It causes a typical chipmunk face due to increased hematopoiesis. Severe malocclusion often accompanies facial deformity, which may affect Oral Health-related Quality of Life (OHRQoL). The aim of this study was to assess the relationship between orthodontic treatment needs and OHRQoL in TM patients.

Materials and Methods: One hundred and five adult patients with TM participated in this cross-sectional study. Oral Health Impact Profile-14 (OHIP-14) questionnaire was used to measure OHRQoL and the patient’s need for orthodontic treatment was evaluated using the Index of Orthodontic Treatment Need (IOTN). The comparison of OHIP-14 scores between groups regarding orthodontic treatment need was carried out using t-test (SPSS software); \( P < 0.05 \) was considered statistically significant.

Results: The mean age of participants was 24.92 (±9.33) years, with 52% female versus 48% male. Orthodontic treatment need was 27.6%. The mean score of OHIP-14 was 12.95 (±7.02). A statistically significant relationship was found between OHIP-14 score and age, gender, and orthodontic treatment need \( (P < 0.05) \). All domains of OHIP-14 were significantly related to orthodontic treatment need \( (P < 0.05) \), except for “physical disability” \( (P = 0.282) \).

Conclusion: OHRQoL was lower in TM patients with orthodontic treatment needs. Planning to treat malocclusion seems necessary to improve the quality of life in these patients.

Key Words: Index of orthodontic treatment need, oral health, quality of life, thalassemia

INTRODUCTION

Beta thalassemia is a genetic disorder affecting hemoglobin synthesis.⁴ The major form expresses many characteristics, including the “chipmunk” face.⁵ It is the side effect of extreme maxillary protrusion, due to the increased volume of hematopoietic bone marrow.⁶ The observed maxillary enlargement can cause structural changes in the oral cavity such as teeth protrusion, spacing, occlusal deep bite, open bite, and different degrees of malocclusion that predispose patients to dental problems.⁶,⁷,⁸ Many thalassemia major (TM) patients suffer from severe Angle class II malocclusion that may affect the patient’s life quality.⁶,⁷,⁸

Index of Orthodontic Treatment Need-Dental Health Component (IOTN-DHC) has been developed to...
objectively assess the impact of malocclusion on oral health.\cite{9,10} It has been shown that the majority of TM patients have Class II skeletal and dental malocclusion with increased overjet and severe tooth displacements compared to controls, and has consequently been graded to have a high need for orthodontic treatment.\cite{11}

Oral Health-related Quality of Life (OHRQoL) is one’s satisfaction regarding his/her functional, emotional, and esthetic oral expectations.\cite{12} The OHRQoL questionnaires measure self-perceived oral need, which is an essential aid to plan suitable dental treatment for each individual.\cite{13} Several studies have measured OHRQoL in TM patients. Amirabadi et al. found lower OHRQoL in TM patients than controls;\cite{14} Phrai-In et al. reported a high prevalence of oral impacts on TM patients’ daily performances;\cite{15} and Ebeid et al. reported a negative impact of TM on emotional well-being.\cite{16} On the other hand, Fadel et al. reported an acceptable OHRQoL in TM.\cite{17}

Oral Health Impact Profile-14 (OHIP-14) is one of the most utilized questionnaires in assessing OHRQoL, with proven validity and reliability.\cite{18} Some studies have used this means to assess OHRQoL in systemic diseases, including TM. Mohamadi et al. found a negative impact of poor oral health status on OHIP-14 score in TM.\cite{19} and Motallebnejad et al. reported that oral health affects OHIP-14 in TM patients, especially regarding its psychological aspects.\cite{20}

Of note, Liu et al. found that orthodontic treatment need is associated with OHRQoL,\cite{21} and Dalaei et al. reported a negative impact of malocclusion on OHRQoL in the general population.\cite{22} However, since the occlusal problems in TM patients may significantly affect OHRQoL as well,\cite{21} the aim of the present study was to evaluate the relationship between orthodontic treatment need and OHRQoL in TM patients living in Isfahan, Iran.

### MATERIALS AND METHODS

#### Patients and sampling

This was an analytic cross-sectional study approved in research and ethics committee of Isfahan (NO:399551). The participants were recruited from patients with TM referred to Oral Medicine Department, Faculty of Dentistry, Isfahan University of Medical Sciences, from June 2019 to June 2020.

The patients with a confirmed diagnosis of TM between 18 and 35 years of age were included in this study. The exclusion criteria were suffering from other systemic diseases, current drug consumption, need for hospitalization or blood transfusion, wearing complete or partial dentures, history of or current orthodontic treatment, and not willing to participate in this study. The study design was explained to all present patients, and a total of 105 volunteers took part.

#### Oral health-related quality of life

Data were collected using self-administered OHIP-14 questionnaire [Figure 1]. Reliability and validity of the Persian version of the questionnaire have been previously confirmed.\cite{18} Age and gender of patients were asked as demographic data and attached to their questionnaire. The OHIP-14 questionnaire consists of 14 questions which are rated on a 5-point scale, ranging from “never” (score 0) to “always” (score 4). The total score is therefore going to be from 0 to 56. A lower score indicates a better OHRQoL and vice versa. The score for each domain of the questionnaire was calculated as well, a score from 0 to 8 for each of the 7 domains.

#### Orthodontic treatment need

A thorough dental examination was performed by
a trained examiner, using a dental mirror, explorer, and periodontal probe. IOTN-DHC was used to define malocclusion severity. This index categorizes orthodontic treatment needs into five grades, from no need (grade 1) to very great need (grade 5), by means of examining over-jet, over-bite, open-bite, cross-bite, crowding, and some other clinical orthodontic parameters. The index output was dichotomized and recorded as 1: no/borderline need (grades 1-3) and 2: definitive need (grades 4 and 5) for orthodontic treatment.[13]

Ethical considerations
The local ethics committee passed the study design (ethical code IR.MUI.RESEARCH.REC.1399.651) and the researcher had to ethically refer the patients to be treated accordingly. All participants signed in an informed consent form too. Not being willing to participate in the study did not affect the proposed treatment plan for the patient.

Statistical analysis
The statistical analysis was done via SPSS version 22 (SPSS Inc., IL, USA) T-test was used to compare OHIP-14 scores between groups. Pearson correlation coefficient was used to analyze the relationship between OHIP-14 and age and the Chi-square test was used to compare IOTN regarding the gender. P < 0.05 was considered statistically significant.

RESULTS
One hundred and five TM patients participated in the present study. Fifty-five (52%) were women, and 50 (48%) were men. The mean (±standard deviation [SD]) age was 24.92 (±9.33). Definitive orthodontic treatment need frequency was 27.6%. The mean scores of OHIP-14 and orthodontic treatment need frequency for both genders are shown in Table 1. T-test showed that women had higher OHIP-14 scores (poorer OHRQoL) than men (P = 0.015). Chi-square test revealed no statistically significant difference in orthodontic treatment need frequency between men and women (P = 0.15).

Pearson correlation test showed that OHRQoL deteriorates as patients age (P < 0.001, r = 0.70). Also, t-test revealed that older patients experience more orthodontic treatment needs (P < 0.001). Table 2 shows the mean score of OHIP-14 in relation to orthodontic treatment needs. T-test showed better OHRQoL in patients with no need (P < 0.001). The mean (± SD) scores of OHIP 14 domains are shown

| Gender | OHIP-14, mean (±SD) | Orthodontic treatment need, frequency (%) |
|--------|---------------------|------------------------------------------|
| Male   | 11.22 (±6.37)       | 22                                       |
| Female | 14.56 (±7.42)       | 33                                       |
| P      | 0.015*              | 0.15                                     |

*Statistically significant. SD: Standard deviation, OHIP: Oral Health Impact Profile

| Orthodontic treatment need | Mean OHIP-14 score | P |
|----------------------------|--------------------|---|
| No                         | 11.67 (±6.52)      | <0.001* |
| Yes                        | 17.88 (±6.59)      |    |

*Statistically significant. OHIP: Oral Health Impact Profile

in Table 3, as well as the relationship between each domain and gender, age, and orthodontic treatment need.

DISCUSSION
Occlusion morbidities are of great concern in TM patients. An excessive amount of hematopoietic bone marrow expands facial bones in these patients, due to extremely increased demand for hematopoiesis[2,3] and maxillary protrusion ensues, which may cause difficulty in oral function and esthetics, leading to OHRQoL impaction.[16]

The prevalence of malocclusion among TM patients was 27.6% in the present study. In a systematic review[24] by Eslamipour et al., it was reported that in the Iranian population, 23.8% of people had all types of malocclusion discovered by IOTN-DHC. Hedayati et al. reported a definitive need for orthodontic treatment in 18.39% of their Persian study population,[25] and the mean IOTN-DHC was 1.23 (±2.45) in the Persian participants of Naseri et al. study.[26] Therefore, the prevalence of malocclusion among TM patients was slightly higher than the general population based on the present study, which is in consensus with the results of previous studies.[11,27]

The impact of malocclusion on OHRQoL of the general population has been well shown in the previous studies.[21,22,28] Similar to our results, Dalaie et al. found a significant correlation between orthodontic treatment need and the total OHIP-14 score among the general population. No significant relationship
between physical disability and orthodontic treatment need was found in their study, in accordance with the results of the present study.}\textsuperscript{[22]} This may reflect the fact that, even in TM patients, esthetic and speech considerations, rather than food intake problems, have a substantial impact on OHRQoL.\textsuperscript{[29]} Furthermore, Sun \textit{et al}. reported that functional limitation and social well-being domains of OHRQoL are more easily affected by malocclusion than other domains,\textsuperscript{[30]} which is in agreement with our results.

Studies measuring OHRQoL in TM patients have bolded the impact of oral health status on psychological aspects of OHRQoL. Ebeid \textit{et al}. reported a negative impact of TM on the emotional well-being aspect of OHRQoL.\textsuperscript{[16]} In the study by Motallebnejad \textit{et al}., psychological aspects of OHIP-14 were more impacted by oral health condition, compared to functional aspects, in Persian TM patients.\textsuperscript{[20]} Therefore, psycho-social support seems mandatory to improve OHRQoL in TM patients.

This study detected lower OHRQoL scores in all domains of OHIP-14 among older people with TM than the younger ones. It sounds that as younger patients have more strictly been under care since their birth, they have developed less medical problems affecting their oral condition, resulting in a better OHRQoL. Furthermore, they might have paid more attention to their oral hygiene, which may have improved their OHRQoL. This fact urges the planning of prompt medical and dental support in TM patients, including primary medical intervention to alleviate facial bone marrow changes as early as possible, as well as orthopedic and functional appliances to prevent or at least reduce orthosurgical needs.\textsuperscript{[23]}

The present study showed a statistically significant relationship between orthodontic treatment need and OHIP-14 score. IOTN-DHC is an objective tool, helping the dentist to meter patient’s need for orthodontic treatment. On the other hand, OHIP-14 is quite a subjective means of assessing the impact of oral health on one’s daily life. As improving the OHRQoL is considered the final goal for every dental or orthodontic treatment,\textsuperscript{[31-35]} the results of the present study facilitate the use of subjective methods, such as IOTN-AC (esthetic component) or several OHRQoL questionnaires, in planning an appropriate orthodontic treatment for TM patients.

Of course, this study was done within the limitations of a cross-sectional research. Studies investigating the cause–effect relationship between variables could interpret the impact of malocclusion on OHRQoL domains more accurately. Besides, several oral health parameters might have influenced OHIP-14 score in the present study, including a number of present teeth or previous dental treatments. Future studies are recommended to further elucidate the exact role and interaction of such factors in determining OHRQoL in patients suffering from TM. Developing disease-specific questionnaires to executively measure OHRQoL in TM would help improving OHRQoL in these patients a great deal.

**CONCLUSION**

TM patients with the need for orthodontic treatment experienced worse OHRQoL compared to their counterparts without this need. It seems that more attention has to be paid to treat orthodontic problems in these patients to improve their oral health and quality of life.

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**Conflicts of interest**
The authors of this manuscript declare that they have no conflicts of interest, real or perceived, financial or nonfinancial in this article.
REFERENCES

1. Alqahtani H. Medically compromised patients in orthodontic practice: Review of evidence and recommendations. Int Orthod 2019;17:776-88.
2. Hattab FN. Thalassemia major and related dentomaxillofacial complications: Clinical and radiographic overview with reference to dental care. Int J Exp Dent Sci 2017;6:95-104.
3. Hattab FN. Patterns of physical growth and dental development in Jordanian children and adolescents with thalassemia major. J Oral Sci 2013;55:71-7.
4. Hattab FN. Mesiodistal crown diameters and tooth size discrepancy of permanent dentition in thalassemic patients. J Clin Exp Dent 2013;5:e239-44.
5. Mulimani P, Abas AB, Karanth L, Colombatti R, Kulkarni P. Treatment of dental and orthodontic complications in thalassemia. Cochran Database Syst Rev 2019;8:CD012969.
6. Movahhedian N, Akbarizadeh F, Kochjastepour L, Sardarani AR, Pakshir HR, Ahviri F. Assessment of mandibular characteristics in patients affected with β-thalassemia major: A retrospective case-control study. Int Orthod 2020;18:776-83.
7. Elangovan A, Mangana J, Joseph E, Guptha V. Prevalence of dentofacial abnormalities in children and adolescents with β-thalassemia major. Indian J Dent Res 2013;24:406-10.
8. Marghalani AH, Al-Mahmoud AS, Chaar DM, Rabie GM, Alzurqi NI, Al Zahra F. Prevalence of malocclusion in the southern region of Jeddah, KSA, and its impact on quality of life – A cross-sectional study. J Evol Med Dent Sci 2021;10:768-73.
9. Elyaskhil M, Shafai NA, Mokhtar N. Effect of malocclusion severity on oral health related quality of life in Malay adolescents. Health Qual Life Outcomes 2021;19:71.
10. Kaygusuz E, Hacıömeroğlu AB, Yüksel S, Mümün M, Tortop T. The influence of abnormalities in the profile and overjet on psychological well-being. Turk J Orthod 2021;34:54-60.
11. Jeelani W, Sher U, Ahmed M. Nature and severity of dental malocclusion in children suffering from transfusion-dependent β-thalassemia major. Dental Press J Orthod 2020;25:261-9.
12. Asgari I, Ahmady AE, Broder H, Eslamipour F, Wilson-Genderson M. Assessing the oral health-related quality of life in Iranian adolescents: Validity of the Persian version of the Child Oral Health Impact Profile (COHIP). Oral Health Prev Dent 2013;11:147-54.
13. Asgari I, Ahmady AE, Yadegarfar G, Eslamipour F. Evaluation of orthodontic treatment need by patient-based methods compared with normative method. Dent Res J (Isfahan) 2013;10:636-42.
14. Amirabadi F, Saravani S, Miri-Alibad G, Khorashadi-Zadeh M. The association between dental health status and oral health-related quality of life of children diagnosed with beta-thalassemia major in Zahedan city, Iran. Int J Pediatr Mashhad 2019;7:8985-91.
15. Phrai-In N, Noikeaw J, Sukprasert N, Taya T, Samnieng P. Oral health status and impact on oral health-related quality of life in children with thalassemia major. UI Proc Health Med 2017;1:144-6.
16. Ebeid FS, Khan NI. The adverse impact of thalassemia major on adolescents’ oral health-related quality of life. J Pediatr Hematol Oncol 2020;42:e345-51.
17. Fadel HT, Zolaly MA, Alharbi MO, Qarah LA, Alrehili MS, Alamri AD, et al. Oral health profiles and related quality of life in thalassemia children in relation to iron overload: A cross-sectional study. Int J Environ Res Public Health 2020;17:9444.
18. Ravaghi V, Farrahi-Avval N, Locker D, Underwood M. Validation of the Persian short version of the Oral Health Impact Profile (OHIP-14). Oral Health Prev Dent 2010;8:229-35.
19. Mohamadi S, Moradveisi B, Rasouli MA. Assessment of oral health and quality of life in hemophilia and thalassemia major patients. J Dent Med 2021;33:157-67.
20. Motallebnejad M, Noghani A, Tamaddon A, Khafris S. Assessment of oral health status and oral health-related quality of life in thalassemia major patients. J Mazandaran Univ Med Sci 2014;24:83-94.
21. Liu Z, McGrath C, Hägg U. Associations between orthodontic treatment need and oral health-related quality of life among young adults: Does it depend on how you assess them? Community Dent Oral Epidemiol 2011;39:137-44.
22. Dalaie K, Behnaz M, Khodabakhshi Z, Hosseinpouir S. Impact of malocclusion severity on oral health-related quality of life in an Iranian young adult population. Eur J Dent 2018;12:129-35.
23. Einy S, Ben-Barak A, Kridin K, Aizenbud D. Craniofacial deformities in patients with beta-thalassemia: Orthodontic versus surgical correction – A systematic review. J Pediatr Hematol Oncol 2020;42:198-203.
24. Eslamipour F, Afshari Z, Najimi A. Prevalence of orthodontic treatment need in permanent dentition of Iranian population: A systematic review and meta-analysis of observational studies. Dent Res J (Isfahan) 2018;15:1-10.
25. Hedayati Z, Fattahi HR, Jahromi SB. The use of index of orthodontic treatment need in an Iranian population. J Indian Soc Pedod Prev Dent 2007;25:10-4.
26. Naseri N, Baherimoghadam T, Kavianirad F, Haem M, Nikmehr S. Associations between malocclusion and self-esteem among Persian adolescent population. J Orthod Sci 2020;9:6.
27. Gupta DK, Singh SP, Utreja A, Verma S. Prevalence of malocclusion and assessment of treatment needs in β-thalassemia major children. Prog Orthod 2016;17:7.
28. Ferrando-Magraner E, Garcia-Sanz V, Bellot-Arcis C, Montiel-Company JM, Almerich-Silla JM, Paredes-Gallardo V. Oral health-related quality of life of adolescents after orthodontic treatment. A systematic review. J Clin Exp Dent 2019;11:e194-202.
29. Farzanegan F, Jeravi F, Sooratgar A, Dastmalchi P. Evaluation of relationship between oral health-related quality of life and occlusion traits among female adolescents. Dent Res J (Isfahan) 2014;11:684-8.
30. Sun L, Wong HM, McGrath CP. Association between the severity of malocclusion, assessed by occlusal indices, and oral health related quality of life: A systematic review and meta-analysis. Oral Health Prev Dent 2018;16:211-23.
31. Oyapero A, Edomwonyi AI, Adeniyi AA, Olatosi OO. Can oral health-related quality of life be worsened by dental
appointments? Dent Res J (Isfahan) 2020;17:395-403.
32. Martins AM, Guimarães LS, Campos CH, Küchler EC, Pereira DM, Maia LC, et al. The effect of complete dentures on edentulous patients’ oral health-related quality of life in long-term: A systematic review and meta-analysis. Dent Res J (Isfahan) 2021;18:65.
33. Larsson P, Bondemark L, Häggman-Henrikson B. The impact of oro-facial appearance on oral health-related quality of life: A systematic review. J Oral Rehabil 2021;48:271-81.
34. Mandava P, Singaraju GS, Obili S, Nettam V, Vatturu S, Erugu S. Impact of self-esteem on the relationship between orthodontic treatment and the oral health-related quality of life in patients after orthodontic treatment – A systematic review. Med Pharm Rep 2021;94:158-69.
35. Alrashed M, Alqerban A. The relationship between malocclusion and oral health-related quality of life among adolescents: A systematic literature review and meta-analysis. Eur J Orthod 2021;43:173-83.