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Original Article

Oral health-related quality of life in patients with osteogenesis imperfecta in Taiwan

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KEYWORDS
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Abstract *Background/purpose:* Osteogenesis imperfecta (OI) is a rare genetic disorder that affects bone and dental structures, often reducing oral health-related quality of life (OHRQoL). Maintaining OHRQoL in individuals with OI depends greatly on their dental conditions. Thus, this study identified key dental factors associated with OHRQoL in this population.
Materials and methods: Thirty-seven patients with OI participated in this cross-sectional study. Data were collected using structured questionnaires on sociodemographic characteristics, oral habits, self-perceived oral health, and OHRQoL. Clinical examinations were performed to evaluate dental status. Associations between dental variables and OHRQoL were analyzed using the Mann–Whitney U test and Kruskal–Wallis test. Multiple logistic regression analysis was used to identify the significant predictors of poor OHRQoL.
Results: Multiple logistic regression analysis revealed that the number of posterior functional tooth units (P-FTUs) was the only significant dental predictor of OHRQoL. A higher P-FTU count was significantly associated with better OHRQoL scores.
Conclusion: An adequate number of P-FTUs is essential for maintaining OHRQoL in patients with OI. In addition to retaining natural teeth or fixed prostheses, ensuring proper distribution and functional occlusion is critical. Clinicians should prioritize treatment strategies that

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preserve posterior occlusion and provide prosthetic rehabilitation when required to support optimal oral function and quality of life.

Introduction

Osteogenesis imperfecta (OI), also known as brittle bone disease, is a rare hereditary connective tissue disorder primarily characterized by increased bone fragility, which results in frequent fractures after minimal trauma. OI is most often inherited in an autosomal dominant pattern and caused by mutations in genes responsible for producing type I collagen, a critical structural protein present in the bone, dentin, cornea, dermis, and tendons.[1] Most patients with OI have mutations in the COL1A1 and COL1A2 genes. These mutations impair the quality or quantity of type I collagen and result in skeletal abnormalities, such as short stature, bone deformities, and decreased mobility. In addition to skeletal fragility, patients may present with blue sclera, joint and skin hyperlaxity, wormian bones, and hearing loss.[2] These systemic manifestations can substantially affect patients' overall quality of life (QoL).[3,4] Therefore, when evaluating the health status and treatment outcomes of individuals with OI, clinicians should consider not only the physical manifestations of the disease but also its broader impact on QoL.[5,6]

Oral diseases can disrupt essential daily activities such as eating, sleeping, working, and social interaction, making oral health a critical component of overall health-related QoL (HRQoL). Traditional clinical indicators, such as dental caries and periodontal disease, do not fully capture the psychological and social dimensions of oral health, as defined by the World Health Organization (WHO).[7,8] Oral health-related quality of life (OHRQoL) provides a multidimensional, patient-centered perspective that includes the physical, psychological, and social effects of oral conditions. Therefore, assessing OHRQoL provides a comprehensive understanding of oral health and its effect on daily life.[9–11]

A notable dental manifestation of OI is dentinogenesis imperfecta, a condition caused by defective dentin structure.[12,13] This defect often results in severe tooth wear and early tooth loss, creating challenges for endodontic treatment and long-term oral health maintenance.[14,15] Effective chewing promotes better digestion and nutrient absorption and enhances overall well-being and daily convenience. By contrast, impaired mastication can restrict an individual's diet; reduce their eating efficiency; and adversely affect their psychological, emotional, and social well-being. In the first phase of our study[16] conducted with patients with OI in Taiwan, we examined factors affecting masticatory ability by using questionnaires and clinical dental assessments. In statistical analyses, the number of posterior functional tooth units (P-FTUs) was indicated to be a significant predictor of self-reported masticatory difficulty. These findings highlight the importance of maintaining an adequate number of teeth or prostheses and ensuring their proper alignment and functional occlusion.

The Oral Health Impact Profile (OHIP) is among the most widely used instruments for measuring OHRQoL. The OHIP evaluates seven dimensions of oral health burden, namely functional limitation, physical pain, psychological discomfort, physical disability, psychological disability, social disability, and handicap. This validated instrument captures the social and psychological effects of oral health conditions at both the individual and population levels.[10,11,17] Tooth loss, often resulting from untreated dental caries or periodontal disease, is a well-documented factor that adversely affects OHRQoL.[8,18] Gerritsen et al.[6] observed a consistent association between tooth loss and poor OHRQoL, regardless of the assessment tool used, and determined that the distribution of missing teeth affected the extent of impairment. Mack et al.[19] reported that individuals with fewer than nine teeth experienced a greater decline in OHRQoL than individuals living with chronic conditions such as cancer, hypertension, or allergies.

Although previous studies[17–19] have provided valuable insights, only a limited number of studies[20,21] have examined the optimal dental status, including the number and types of teeth, that individuals with OI should maintain to preserve their OHRQoL. This study represents the second phase of our research on patients with OI in Taiwan and builds on the findings from the first phase.[16] In this phase, we identified dental factors most strongly associated with OHRQoL in this population. The findings of this study can inform clinical strategies that improve patient-centered outcomes.

Materials and methods

Study design and participants

This cross-sectional study included Taiwanese patients with OI. Data were collected using structured questionnaires and clinical oral examinations. The participants were the same individuals enrolled in the first phase of our study,[16] and recruitment was conducted in collaboration with the Taiwan OI Association during organized outreach activities. Participants were enrolled between May 2022 and November 2022. Before participation, all individuals received a full explanation of the study's objectives and significance, and their written informed consent was obtained. The inclusion criteria were having (1) a confirmed diagnosis of OI, (2) the ability to understand and complete the questionnaire, and (3) the ability to undergo an oral

examination. Participants were excluded if they were unable to complete the questionnaire or declined the oral examination. The study protocol was approved by the Institutional Review Board of Kaohsiung Medical University Chung-Ho Memorial Hospital, Taiwan (approval number: KMUHIRB-SV(II)-20220011).

Data collection

A structured questionnaire was used to collect information on sociodemographic characteristics (age, sex, education level, marital status, and employment status), self-perceived oral health, oral habits (smoking and alcohol consumption), and OHRQoL. OHRQoL was examined using the Oral Health Impact Profile-18 for Taiwanese patients with OI (OHIP-18TOI), a modified version of the OHIP-49.[17] The OHIP-18TOI consists of 18 items selected by a panel of five experts due to their relevance to specific oral health challenges to OI. The questionnaire covers seven dimensions of oral health burden, namely functional limitation, physical pain, psychological discomfort, physical disability, psychological disability, social disability, and handicap. Responses are recorded on a 5-point Likert scale: 0 (never), 1 (rarely), 2 (occasionally), 3 (often), and 4 (always). The total OHIP-18TOI score ranges from 0 to 72, with higher scores indicating a greater oral health burden and poorer OHRQoL. The internal consistency of the OHIP-18TOI was evaluated using Cronbach's alpha. The Cronbach's α with each item was 0.804, and values ranged from 0.788 to 0.806 without each item, indicating good internal reliability.

Dental examination

All clinical oral examinations were performed by two calibrated dentists following the WHO oral health survey methodology.[22] Before the study, both examiners underwent standardized training and calibrated their assessments by evaluating five pilot cases each. The interexaminer agreement was high, with a kappa coefficient (κ) of 0.850, and any discrepancies in assessments were resolved through discussion. During examinations, the following dental parameters were recorded: the number, type, and location of natural and artificial teeth. Dental classifications were defined as follows: functional natural teeth (FNT) were defined as natural teeth excluding third molars (range: 0—28), with teeth having grade III mobility, retained roots, or more than 75 % crown destruction excluded; functional teeth (FT) were defined as FNT and fixed prosthetic teeth supported by natural teeth or implants (range: 0—28, excluding third molars); functional tooth units (FTUs) were defined as pairs of opposing FT in occlusion (range: 0—14); P-FTUs were defined as occluding pairs in the premolar and molar regions, with premolars counted as 1 unit and molars as 2 units (range: 0—12). The Eichner index was used to evaluate occlusal status on the basis of FT, with patients given classifications of Class A (contact in all four posterior occlusal zones), Class B (contact in one to three occlusal zones or only in the anterior region), or Class C (no occlusal contact in any zone).

Statistical analysis

Categorical variables are presented as frequencies and percentages, and continuous variables are expressed as means $\pm$ standard deviations. The Mann—Whitney U test and Kruskal—Wallis test were used to examine associations between OHIP-18TOI scores and various sociodemographic, behavioral, and dental factors.

The participants were stratified into two age groups ($\leq$35 and $>$ 35 years) on the basis of their median age. OHIP-18TOI scores were also categorized using the median score of 11, with scores $\leq$11 indicating good OHRQoL and scores $>$11 indicating poor OHRQoL. Multiple logistic regression analysis was performed to identify significant predictors of poor OHRQoL. The model was adjusted for potential confounders, including age, sex, education level, and other variables significantly associated with OHIP-18TOI scores. All statistical analyses were conducted using SPSS software (version 25.0; IBM Corp., Chicago, IL, USA). A P value of $<$0.05 was considered statistically significant.

Results

This study included 37 participants in the cohort of the first phase of the research, comprising 21 women (56.8 %) and 16 men (43.2 %).[16] The mean age was 38.6 $\pm$ 16.3 years (range: 14—75 years). Nineteen participants (51.4 %) were younger than 35 years, and 18 participants (48.6 %) were aged 35 years or older. Regarding education level, 17 (45.9 %) participants had completed high school or less, and 20 (54.1 %) had attained a university-level education or higher. Most participants were unmarried (n = 25 [67.6 %]), whereas 12 (32.4 %) were married. Seventeen (45.9 %) participants were employed, and 20 (54.1 %) were unemployed. Self-perceived oral health was rated as good or fair by 23 (62.2 %) participants. Most reported that they had never smoked (n = 35 [94.6 %]) or consumed alcohol (n = 32 [86.5 %]). In terms of dental parameters, the mean numbers of FNT, FT, FTUs, and P-FTUs were 23.5 $\pm$ 8.3, 23.7 $\pm$ 8.3, 9.3 $\pm$ 5.1, and 9.3 $\pm$ 5.1, respectively. According to the Eichner index, 23 (62.2 %), 11 (29.7 %), and 3 (8.1 %) participants received classifications of Class A, Class B, and Class C, respectively.

The mean OHIP-18TOI score was 10.8 $\pm$ 7.2 (Table 1). The most frequently reported concern was "feeling self-conscious about teeth" (mean score: 1.2 $\pm$ 1.3), followed by "bad breath" (1.1 $\pm$ 1.0), "dissatisfaction with brushing" (0.9 $\pm$ 1.1), "worsening general health" (0.8 $\pm$ 1.1), and "painful gums" (0.8 $\pm$ 0.8). OHIP-18TOI scores, including all seven dimensions, had no statistically significant associations with sociodemographic variables, or oral habits. Similarly, OHIP-18TOI scores were not significantly associated with occlusal status, as determined by the Eichner index. However, patients who rated their self-perceived oral health as poor had significantly higher OHIP-18TOI scores, indicating poorer OHRQoL (Table 2).

In terms of dental status, the numbers of FT and FTUs were not significantly associated with OHIP-18TOI scores. However, lower numbers of FNT and P-FTUs were significantly associated with higher OHIP-18TOI scores, indicating that patients with fewer FNT or P-FTUs tended to have

Table 1 Mean score of oral health-related quality of life (N = 37).

Variables	Mean ± SD
Functional limitations	1.9 ± 1.6
Difficulty chewing	0.8 ± 1.0
Breathstale	1.1 ± 1.0
Physical pain	2.8 ± 2.5
Sore jaw	0.5 ± 0.7
Toothache	0.7 ± 0.7
Painful gums	0.8 ± 0.8
Sensitive teeth	0.7 ± 0.9
Uncomfortable dentures	0.3 ± 0.6
Psychological discomfort	1.2 ± 1.3
Self-conscious about teeth	1.2 ± 1.3
Physical disability	2.4 ± 2.3
Less favor in food	0.2 ± 0.6
Avoid smiling	0.6 ± 1.0
Brush unsatisfactory	0.9 ± 1.1
Speech unclear	0.7 ± 0.9
Psychological disability	0.2 ± 0.7
Difficult to relax	0.2 ± 0.7
Social disability	0.9 ± 1.8
Avoid going out	0.4 ± 0.9
Trouble getting on with others	0.2 ± 0.6
Difficulty doing your usual jobs	0.4 ± 0.9
Handicap	1.3 ± 1.8
Life was less satisfying	0.5 ± 0.9
General health has worsened	0.8 ± 1.1
OHIP-18TOI scores	10.8 ± 7.2

poorer OHRQoL (Table 3). To identify the predictors of poor OHRQoL, a multiple logistic regression analysis was performed with adjustment for age, sex, education level, self-perceived oral health, and dental variables that were determined to be significantly associated with OHIP-18TOI scores (Table 4). Self-perceived oral health and P-FTUs were identified as the significant predictors of poor OHRQoL. Participants who rated their oral health as poor were 9.06 times more likely to have poor OHRQoL than did those who rated their oral health as good or fair (odds ratio [OR]: 9.06; 95 % confidence interval [CI]: 1.56–81.24). Participants with fewer than eight P-FTUs were 11.60 times more likely to have poor OHRQoL than did those with eight or more P-FTUs (OR: 11.60; 95 % CI: 1.94–111.97). Among all dental parameters evaluated (FNT, FT, FTUs, and P-FTUs), only P-FTUs remained a statistically significant predictor of poor OHRQoL in patients with OI.

Discussion

OI is a rare genetic disorder that has increasingly attracted the attention of oral health researchers. Nguyen et al.[23] reported that the risk of dental caries in individuals with OI is comparable to or even higher than that in the general population. This finding indicates that patients with OI are equally susceptible to tooth decay as individuals without OI.[24,25] Oral manifestations of OI, including changes in tooth number, morphology, and alignment, can cause pain, hypersensitivity, speech difficulties, masticatory impairment, esthetic concerns, and functional limitations. Each of these complications can negatively affect overall QoL.[3] A comprehensive assessment of QoL is essential for evaluating the broader impact of OI because clinical parameters alone do not fully reflect a patient's overall health status or the outcomes of medical and dental interventions.[11] Recently, the management of OI has shifted beyond conventional medical, orthopedic, and rehabilitative treatments that address bone fragility and mobility limitations. Current approaches also focus on community participation and the improvement in overall QoL.[26]

In the present study, the number of P-FTUs was identified as a significant predictor of OHRQoL in patients with OI. A higher P-FTU count was associated with lower OHIP-18TOI scores, indicating better OHRQoL. This finding is consistent with the results of Tsakos et al.[27] who reported that a decreased number of posterior occluding pairs was associated with poor OHRQoL. In addition, Naito et al.[28] observed that a greater number of FNT was correlated with improved OHRQoL, as measured using the Geriatric Oral Health Assessment Index. Although the findings of the current study revealed that FNT was negatively correlated with OHRQoL scores, it was not identified as a statistically significant predictor in the multivariate analysis. This difference may be explained by the multifactorial nature of OHRQoL, which includes four key domains, namely oral function, orofacial pain, orofacial appearance, and psychosocial impact.[29] The number of natural teeth plays a role in mastication but also affects esthetics and thus psychological and social well-being. In patients with OI, the effect of FNT on OHRQoL may therefore depend not only on functional performance but also on the anatomical location of the teeth and their esthetic contribution.

In the present study, P-FTUs comprised both posterior FNT and FT. Maintaining a sufficient number of FNT is crucial for achieving adequate P-FTUs, which in turn support better masticatory efficiency and improved OHRQoL. Ueno et al.[30] reported that individuals with 20 or more natural teeth typically had FTUs consisting of natural-to-natural occlusion (equivalent to P-FTUs in our study), whereas those with fewer than 20 natural teeth were more reliant on removable prostheses for occlusion. In addition, Gerritsen et al.[6] indicated that tooth loss negatively affects OHRQoL, with the distribution and location of missing teeth exerting a greater effect than the total number lost. Although FNT did not directly predict OHRQoL in our model, its indirect effect through its contribution to P-FTUs and occlusal function should not be overlooked.

Adults with OI often experience substantial impairments in HRQoL, and the extent of these impairments varies in accordance with OI subtype.[31] The severity and clinical presentation of OI affect different QoL domains to varying degrees. Wehrli et al.[4] reported that individuals with OI frequently experience mobility limitations and difficulties in employment and sports participation, all of which have a substantial impact on mental health. Many patients also report higher levels of anxiety and depression and have markedly lower mental health scores compared with the general population. QoL assessments using instruments such as the Pediatric Quality of Life Inventory (PedsQL) for children aged 8–12 years and the Short Form-36 (SF-36) for

Table 2 Oral health-related quality of life (OHIP-18TOI) scores by participants' characteristics and Eichner Index (N = 37).

Variables	OHIP-18TOI Score	FL	PP	PsyD	PD	PsD	SocD	Handicap
	mean ± SD	mean ± SD	mean ± SD	mean ± SD	mean ± SD	mean ± SD	mean ± SD	mean ± SD
Gender								
Male	10.2 ± 7.2	1.7 ± 1.5	2.1 ± 1.6	1.3 ± 1.4	2.4 ± 2.7	0.4 ± 1.0	1.0 ± 2.3	1.5 ± 2.0
Female	11.2 ± 7.4	2.1 ± 1.7	3.4 ± 2.9	1.1 ± 1.2	2.4 ± 2.0	0.1 ± 0.4	0.8 ± 1.5	1.2 ± 1.6
Age, years								
< 35	8.7 ± 7.6	1.6 ± 1.3	2.1 ± 2.0	1.0 ± 1.1	2.6 ± 2.8	0.1 ± 0.2	0.6 ± 1.2	0.9 ± 1.5
≥ 35	12.9 ± 6.3	2.2 ± 1.8	3.7 ± 2.8	1.5 ± 1.5	2.2 ± 1.7	0.4 ± 1.0	1.1 ± 2.4	1.8 ± 2.0
Education level								
High school or below	10.0 ± 7.0	2.1 ± 1.9	2.8 ± 2.4	1.2 ± 1.5	1.4 ± 1.5	0.1 ± 0.3	0.9 ± 1.5	1.5 ± 1.7
College/University	11.4 ± 7.5	1.7 ± 1.2	2.9 ± 2.7	1.3 ± 1.1	3.3 ± 2.6	0.4 ± 0.9	0.8 ± 2.1	1.2 ± 1.8
Marital status								
Unmarried	10.6 ± 7.8	1.7 ± 1.3	2.2 ± 1.9	1.2 ± 1.3	2.4 ± 2.6	0.3 ± 0.8	1.2 ± 2.2	1.6 ± 2.0
Married	11.2 ± 6.1	2.3 ± 2.0	4.2 ± 3.1	1.2 ± 1.3	2.3 ± 1.8	0.2 ± 0.4	0.3 ± 0.6	0.8 ± 0.8
Occupation								
Employed	12.9 ± 7.5	2.1 ± 1.6	3.2 ± 3.1	1.3 ± 1.2	3.4 ± 2.7	0.5 ± 1.0	1.1 ± 2.3	1.4 ± 1.9
Unemployed	9.0 ± 6.6	1.8 ± 1.6	2.5 ± 1.9	1.2 ± 1.4	1.5 ± 1.5	0.1 ± 0.2	0.7 ± 1.4	1.3 ± 1.7
Self-perceived oral health								
Good/Fair	7.8 ± 5.5*	1.6 ± 1.3	2.2 ± 1.7	0.9 ± 1.1	1.9 ± 1.9	0.0 ± 0.2	0.4 ± 0.9	0.8 ± 1.4*
Poor	15.6 ± 7.3	2.4 ± 1.9	3.9 ± 3.2	1.7 ± 1.5	3.1 ± 2.8	0.6 ± 1.1	1.7 ± 2.6	2.1 ± 2.0
Smoking								
Never	11.0 ± 7.4	1.9 ± 1.6	2.8 ± 2.6	1.2 ± 1.3	2.5 ± 2.4	0.3 ± 0.7	0.9 ± 1.9	1.4 ± 1.8
Current smokers/Ex-smokers	7.0 ± 1.4	1.0 ± 0.0	3.0 ± 1.4	2.0 ± 0.0	1.0 ± 0.0	0.0 ± 0.0	0.0 ± 0.0	0.0 ± 0.0
Alcohol								
Never	10.3 ± 6.9	1.9 ± 1.5	2.9 ± 2.6	1.2 ± 1.3	2.2 ± 1.9	0.2 ± 0.4	0.7 ± 1.3	1.3 ± 1.7
Current drinkers/Ex-drinkers	13.6 ± 9.4	2.0 ± 1.9	2.2 ± 1.9	1.6 ± 1.7	3.6 ± 4.3	0.8 ± 1.8	1.8 ± 4.0	1.6 ± 2.6
Eichner index								
A	10.7 ± 7.5	1.7 ± 1.4	2.7 ± 2.4	1.1 ± 1.2	2.4 ± 2.6	0.3 ± 0.9	1.0 ± 2.1	1.3 ± 1.7
B	10.6 ± 6.6	2.0 ± 1.9	3.1 ± 3.0	1.4 ± 1.4	2.2 ± 1.8	0.2 ± 0.4	0.5 ± 0.9	1.3 ± 1.7
C	12.3 ± 10.1	2.7 ± 2.1	2.7 ± 2.1	1.3 ± 2.3	2.7 ± 2.3	0.0 ± 0.0	1.3 ± 2.3	1.7 ± 2.9

P-value is calculated using Mann–Whitney U tests.
P-value of Eichner Index is calculated using Kruskal–Wallis Test.
FL: Functional limitations; PP: Physical pain; PsyD: Psychological discomfort; PD: Physical disability; PsD: Psychological disability; SocD: Social disability.
**P*-value <0.05.

Table 3 Pearson correlation coefficient between OHIP-18TOI scores and FNT, FT, FTUs and P-FTUs (N = 37).

	OHIP-18TOI Score	FL	PP	PsyD	PD	PsD	SocD	Handicap
	Coefficient	Coefficient	Coefficient	Coefficient	Coefficient	Coefficient	Coefficient	Coefficient
FNT	−0.3639*	−0.1955	−0.1767	−0.0093	−0.222	−0.052	−0.159	−0.262
FT	−0.3217	−0.1583	−0.1179	−0.0053	−0.254	0.036	−0.121	−0.265
FTUs	−0.2374	−0.1604	−0.1095	−0.0205	−0.027	0.247	−0.035	0.044
P-FTUs	−0.3781*	−0.2704	−0.2213	−0.1576	−0.11	0.169	−0.034	0.008

P-value is calculated using Spearman Rank Correlation, **P*-value <0.05.
FNT: Functional natural teeth; FT: Functional teeth; FTUs: Functional tooth units; P-FTUs: Posterior-Functional tooth units.
FL: Functional limitations; PP: Physical pain; PsyD: Psychological discomfort; PD: Physical disability; PsD: Psychological disability; SocD: Social disability.

individuals aged 14 years and older confirm that patients with OI have considerably poorer QoL than healthy peers.[4] McDonald et al.[31] observed a progressive decline in HRQoL with increasing age in patients with OI, indicating the importance of regular evaluation throughout adulthood. As OI-related complications accumulate over time, ongoing monitoring becomes necessary. HRQoL also differs by OI subtype; individuals with type III OI generally have

Table 4 Logistic regression analysis for the predictors of poor OHRQoL (N = 37).

Variables	Crude OR[a] (95%CI)	Model 1	Model 2
		Adjusted OR[b] (95%CI)	Adjusted OR[b] (95%CI)
Age, years			
< 35	1	1	1
≥ 35	3.40 (0.91–14.00)	2.03 (0.40–10.38)	1.73 (0.29–10.13)
Gender			
Male	1.33 (0.36–5.03)	1.68 (0.34–9.01)	1.46 (0.25–9.16)
Female	1	1	1
Education level			
Less than college	1.09 (0.29–4.03)	1.07 (0.21–5.45)	0.62 (0.08–3.95)
College or above	1	1	1
Self-perceived oral health			
Good/Fair	1	1	1
Poor	5.71 (1.41–27.31)*	4.76 (1.00–26.54)	9.06 (1.56–81.24)*
FNT			
0-19	7.92 (1.10–161.67)*	4.09 (0.45–93.63)	
20-28	1	1	
P-FTUs			
0-7	7.33 (1.79–35.97)*		11.60 (1.94–111.97)*
8-12	1		1

Poor OHRQoL = Poor oral health-related quality of life: OHIP-18TOI scores greater than 11 points.
OR[a] = Odds ratios[a]: were derived from univariate logistic regression model.
OR[b] = Odds ratios[b]: were derived from a multiple logistic regression model.
* *P*-value<0.05.

substantially poorer QoL than those with type I, likely due to more severe skeletal deformities, a higher frequency of fractures, and greater physical limitations. These findings highlight the importance of personalized clinical management tailored to each patient's needs.

Dahan-Oliel et al.[32] conducted a comprehensive review of QoL instruments used in OI research, including the SF-36, Life Satisfaction Questionnaire, PedsQL, Child Health Questionnaire, Health Utilities Index, and HO Quality of Life–Brief version. Their analysis consistently revealed that patients with OI have lower QoL than the general population, regardless of the assessment tool used. Congenital OI subtypes were associated with greater QoL impairment than acquired OI subtypes. Complications such as scoliosis, respiratory dysfunction, and thoracic or spinal deformities further exacerbate physical limitations. While individuals with OI often have lower physical QoL scores, their psychological well-being can be comparable to that of healthy individuals. Pain, mobility restrictions, and decreased social participation are key contributors to low QoL in patients with OI.

In the UK, Cachia et al.[20] assessed the OHRQoL in 106 children with OI and confirmed dental concerns in oral health, functional well-being, and socio-emotional well-being, which were associated with OI severity. In Brazil, Pantoja et al.[21] examined the connection between craniofacial and oral manifestations and OHRQoL in 30 children and adolescents aged 8–14 with OI. The study found no notable differences based on age group, disease severity, or the presence of dentinogenesis imperfecta. Oral manifestations did not have a direct impact on OHRQoL. The most affected areas were oral symptoms and emotional well-being. The findings of our study were consistent with those reported by Cachia et al.[20] and Pantoja et al.[21]

Given the complexity of OI, disease-specific HRQoL assessment tools are required to more accurately evaluate its impact and inform treatment decisions.[31] However, no HRQoL instrument specific to OI has been developed to date. Most studies[33–35] have used general tools, such as the SF-36[33] or SF-12,[34]or pediatric-focused instruments, such as the PedsQL.[35] Although these measures provide useful insights, they may not fully capture the distinctive oral and systemic challenges experienced by individuals with OI.[4] In the present study, we used the OHIP-18TOI, a modified version of the OHIP-49, comprising 18 items selected by five experts in special needs dentistry, oral surgery, family dentistry, and public health. This instrument demonstrated acceptable internal consistency and was deemed appropriate for evaluating OHRQoL in patients with OI.

This study has several limitations. First, participants were recruited through the Taiwan OI Association during organized activities, which likely resulted in a sample composed of functionally independent individuals who actively participate in community events. Their experiences may therefore not represent all patients with OI, particularly those with more severe impairments or limited access to social support. Second, although the OHIP-18TOI demonstrated acceptable internal consistency, it has not yet been fully validated in a larger, nationally representative Taiwanese OI population. Future studies should validate this tool to ensure its applicability to broader OI cohorts. Third, the small sample size, common in rare disease research, prevented the stratification of analyses by OI subtype, limiting our ability to examine how specific

dental factors affect OHRQoL in different clinical presentations. Nevertheless, this study is among the first to identify the dental predictors of OHRQoL in Taiwanese patients with OI. Its findings provide valuable insights into the unique oral health challenges in this population and highlight the importance of maintaining functional occlusion.

In summary, this study identified P-FTUs as a significant dental predictor of OHRQoL in patients with OI. Maintaining OHRQoL in this population requires retaining an adequate number of natural teeth or providing fixed prosthetic replacements, with particular attention to proper distribution and occlusal function. Health professionals should focus on treatment strategies that preserve functional posterior occlusion, thereby supporting effective mastication and improving overall well-being in individuals with OI.

Declaration of competing interest

All authors have no conflicts of interest relevant to this article.

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