DENTAL FINDINGS OF PERSONS WITH OSTEOGENESIS IMPERFECTA IN VIETNAM

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ABSTRACT

Background Osteogenesis imperfecta (OI) is the collective term for a heterogeneous group of connective tissue syndromes. The aim of the current study is to describe dental characteristics, including dentinogenesis imperfecta (DGI), dental wear, occlusal features, and dental caries experience of Vietnamese persons with OI.
Methods The sample consisted of 74 individuals with OI classified into type I (n=25), type III (n=24), and type IV (n=25). All participants were examined for DGI through the evaluation of intrinsic color variation, dental wear using Hooper's index, occlusal features (including Angle's classification, overjet, overbite, crossbite) and dental caries experience by using the dmft/DMFT index.
Results DGI was found in 62.2% of the sample and was significantly related to OI type III and type IV (p=0.019). Dental wear occurred in 36.5% and was equivalent among OI types. Angle Class III malocclusion was more prevalent in type III (66.7%) and type IV (54.5%) than in type I (37.5%). High prevalence of reverse overjet (60.3%), posterior crossbite (32.2%), and missing teeth (23.3%) were found in the OI sample. The mean dmft/DMFT score was 3.0/2.2. The dental findings related to dental wear, occlusal features, and dental caries did not show significant differences among type I, III, and IV.
Conclusion There was a high prevalence of DGI and dental wear in the Vietnamese OI sample. Occlusal features were related to a high prevalence of class III malocclusion, overjet, open bite, posterior crossbite, and missing teeth. Dental caries experience of persons with OI was at a moderate level.

KEYWORDS

Dental Care for Disabled; Dentinogenesis Imperfecta; Dental Occlusion; Osteogenesis Imperfecta.

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1. INTRODUCTION

Osteogenesis imperfecta (OI) is the collective term for a heterogeneous group of connective tissue syndromes. Seventeen mutated genes have been found related to OI syndromes [1,2]. The mutation incidence varies in different populations from 1/20,000 to 1/10,000 OI cases [3–5].

The clinical classification of OI includes five types (type I–V) [2,6]. Type I is a mild phenotype with dominantly inherited OI and blue sclerae; type II is related to perinatal lethality; type III results in progressive deformity throughout the lifespan; type IV is similar to type I, but sclerae are normal; and type V has mesh-like bone appearance due to calcification in inter-osseous membranes. In clinical studies, types I, III and IV are often mentioned to describe the clinical features of living individuals with OI syndrome. Collagen mutation may influence the dental-facial structures of individuals with OI. Previous studies indicated that more than 50% of individuals with OI had class III malocclusions [7-9]. The OI syndromes cause not only maxillary deficiencies but also hypodontia [8,10]. Dentinogenesis imperfecta (DGI) and dental abnormalities are also observed in intra-oral and radiologic examination of persons with OI, such alterations may result in premature wear of dental structure [10-13]. This suggests that OI has an impact on many oral conditions. Because it is a rare disease, many OI studies focus on finding genetic mutations and on the treatment of bone fractures. There are still gaps in the scientific literature about the dental health of individuals with OI. In Vietnam although OI has been assessed medically [14], to the authors' knowledge, the dental aspects of OI types have not been reported. Therefore, the aim of the current study is to investigate the dental characteristics of persons with OI, including dentinogenesis imperfecta, dental wear, dental caries experience, and malocclusion.

2. MATERIALS AND METHODS

2.1. Study sample

This is a cross-sectional study and the total sample consisted of 74 OI persons aged 2–37 years (mean age = 10.6 ± 7.1, median age = 9.0) from 34 healthcare centers across Vietnam. The OI diagnosis was based on Sillence's classification [5] and was confirmed by two orthopedic experts. OI participants or their legal representatives signed informed consent forms. The Danang University of Medical Technology and Pharmacy (No. 523/CN-DHKTYDDN) approved this study. All procedures were performed according to the World Medical Association Declaration of Helsinki.

2.2. Examination of dentinogenesis imperfecta

DGI was clinically diagnosed according to the Shields' classification [13]. The clinical examination of DGI was based on evaluating color variation, including lightness level, saturation, and hue of teeth, using the Vita System 3D-Master.

The system consists of six lightness level groups from 0 to 5 (0=lightest, 5=darkest), five grades of color saturation (chroma) with a given score of 1, 1.5, 2, 2.5 and 3, and three levels of hue (L=yellowish, M=intermediate hue, R=reddish). In the current study, the lightness level was divided into grades of lightness (score 0–2) and darkness (score 3–5); chroma was grouped into low saturation (score 1–2) and high saturation (score 2.5–3).

Tooth shades were determined in daylight in 5–7 seconds. In our study, DGI was confirmed by intrinsic color variation with teeth that were darker, had high color saturation and displayed a reddish hue.

2.3. Measurement of dental wear

A dental impression was taken for pouring a dental cast to measure tooth wear using a millimeter probe. Tooth wear was evaluated based on Hopper's index [15]. A six-point scale (0=no wear, 5=the most severe) was used to identify a tooth as having incisal/cuspal wear. The level of tooth wear was grouped into no/mild and moderate/severe levels.

2.4. Examination of dental caries

The dental caries experience of persons with OI was recorded using the Decayed, Missing and Filled Teeth index for permanent teeth (DMFT) and for primary teeth (dmft). A decayed tooth was registered as presenting primary caries or secondary caries next to a filling. A missing tooth was a tooth that was extracted due to caries. A filled tooth was a tooth with restoration but without additional caries. The dmft/DMFT score was the sum of decayed, missing and filled teeth.

2.5. Analysis of occlusion

Occlusal variables of OI dentition consisted of 10 variables including overbite, overjet, open bite, posterior crossbite, contact point displacement, midline diastema, molar Angle classification, incisal segment crowding, and incisal segment spacing.

2.6. Calibration procedure

The dental status of an OI person was examined twice on the same day by the first author (MSN) at local healthcare centers to ensuring reliability between the interval examination and inter-examiner. The Kappa value of 0.92 and 0.87 indicated high reliability of the clinical examination. The first author also conducted an analysis of occlusion and dental wear on dental casts. The calculated Kappa values were above 0.85, indicating a high degree of intra-examiner and inter-analysis reliability.

2.7. Statistical analysis

Data entry and analyses were performed with the Statistical Package for the Social Sciences software

Table 1. Prevalence of dentinogenesis imperfecta and tooth discoloration in persons with osteogenesis imperfecta.

Variable		Total N = 74	OI classification			p-value[a]
			Type I n = 25	Type III n = 24	Type IV n = 25	
Dentinogenesis imperfecta						
	No	37.8	60.0	25.0	28.0	0.019*
	Yes	62.2	40.0	75.0	72.0	
Tooth coloration						
Lightness level						
	0-2	12.2	28.0	0	8.0	0.008*b
	3-5	87.8	72.0	100	92.0	
Saturation (Chroma)						
	Low	75.7	72.0	79.2	76.0	0.842
	High	24.3	28.0	20.8	24.0	
Hue						
	Yellowish (L)	10.8	4.0	8.3	20.0	0.418
	Intermediate hue (M)	79.7	88.0	79.2	72.0	
	Reddish (R)	9.5	8.0	12.5	8.0	

[a]Chi-square test; b: Fisher's test
*Significant.

Table 2. Prevalence of dental wear in persons with osteogenesis imperfecta.

Dental wear	Total N = 74	OI classification			p-value
		Type I n = 25	Type III n = 24	Type IV n = 25	
Prevalence of OI persons with dental wear					
No/mild	63.5	55.6	69.6	63.6	0.652[a]
Moderate/severe	36.5	44.4	30.4	36.4	
Mean percentage of teeth showing wear in dentition					
No/mild	90.4	89.7	93.5	87.9	0.647[b]
Moderate/severe	9.6	10.3	6.5	12.1	

[a]Chi-square test; [b]ANOVA test.

version 17.0 (SPSS Inc., Chicago, IL, USA).
The Chi-square test and ANOVA test were used to identify the differences related to DGI, dental wear, occlusal features, and dmft/DMFT score among OI types I, III, and IV. An interval confidence level of 95% and a two-sided p-value of .05 were set for significant difference.

3. RESULTS

The study included 74 persons with OI aged 2–37 years (50% females and 50% males). The distribution of clinical diagnoses was 33.8% type I (n=25), 32.4% type III (n=24), and 33.8% type IV (n=25). Primary dentition was accounted for in 31.1% of the sample, mixed dentition was in 37.8%, and permanent dentition was 31.1%. Dentinogenesis imperfecta was found in 62.2% of the total sample. Prevalence of DGI was statistically more frequent in type III (75%) and color variation, 72% dentition of type I, 92% of type IV, and 100% of type III were graded in darkness levels. Prevalence of high saturation was fairly equivalent among type I (28.0%), type III (20.8%), and type IV (24.0%, p=0.842). The hue component of the total dentitions was 10.8% of yellowish, 79.7% of intermediate hue, and 9.5% of reddish.
There were no differences in hue components among OI types (p = 0.418) (Table 1). Dental wear occurred in 36.5% of OI individuals, of which 44.4% presented in type I, 30.4% in type III, and 36.4% in type IV. The mean percentage of the amount of tooth wear in dentition was found in 12.1% of type IV, 10.5% of type I, and 6.5% of type III. However, there were no differences in the distribution of tooth wear among three types of OI (p>0.05, Table 2). Table 3 shows the characteristics of dentition associated with each type of OI. According to Angle's classification, class III was more prevalent in type III (66.7%) and type

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Table 3. Prevalence of occlusal features in persons with osteogenesis imperfecta.

Variable	Total N = 74	OI classification			p-value
		Type I n = 25	Type III n = 24	Type IV n = 25	
Angle's classification					
Class I	27.1	43.8	23.8	18.2	0.231
Class II	18.6	18.8	9.5	27.3	
Class III	54.2	37.5	66.7	54.5	
Overbite >3.5mm					
No	80.7	64.3	90.5	81.8	0.155
Yes	19.3	35.7	9.5	18.2	
Increased overjet > 3.5 mm					
No	100	100	100	100	-
Yes	0	0	0	0	
Reverse overjet					
No	39.7	60.0	33.3	31.8	0.173
Yes	60.3	40.0	66.7	62.8	
Posterior crossbite					
No	67.8	75.0	57.1	72.7	0.424
Yes	32.2	25.0	42.9	27.3	
Open bite					
No	82.5	100	75.0	77.3	0.133
Yes	17.5	0	25.0	22.7	
Diastema					
No	86.4	87.5	90.5	81.8	0.702
Yes	13.6	12.5	9.5	18.2	
Displacement > 2mm					
No	66.1	75.0	52.4	72.7	0.251
Yes	33.9	25.0	47.6	27.3	
Incisal segment crowding					
No	53.4	75.0	42.9	47.6	0.121
Yes	46.7	25.0	57.1	52.4	
Incisal segment spacing					
No	66.1	62.5	76.2	59.1	0.466
Yes	33.9	37.5	23.8	40.9	
Missing teeth					
No	76.7	76.5	76.2	77.3	0.996
Yes	23.3	23.5	23.8	22.7	

Chi-square test; *Significant.

IV (54.5%) than in type I (37.5%). No OI persons had an increased overjet (>3.5mm), but 60.3% of individuals had reverse overjet. Posterior crossbite occurred in 32.2% of the total OI sample, of which 42.9% presented in type III, 27.3% in type IV, and 25.0% in type I. High prevalence of incisal segment crowding (46.7%), incisal segment spacing (33.9%), displacement of tooth>2mm (33.9%), missing teeth (23.3%), open bite (19.3%) and diastema (13.6%) were also found in the total sample, but no significant differences were observed among type I, III and IV (p>0.05). Table 4 indicates dental caries experience within the OI sample; the mean of the dmft and DMFT score was 3.0 ± 4.1 and 2.2 ± 4.6 respectively. The dmft and DMFT scores were equivalent among types of OI (p>0.05). None of the persons with OI received restorative treatment for decayed teeth, and none of the permanent teeth in type III and IV were extracted due to caries.

4. DISCUSSION

4.1. General information about Vietnamese persons with OI. This is a preliminary study conducted in Vietnam to collect the dental characteristics of persons who suffer from OI. The total sample of our

Table 4. Mean score of dental caries components of primary and permanent teeth in osteogenesis imperfecta patients.

Dental caries experience	Total N = 74	OI classification			p-value
		Type I n = 25	Type III n = 24	Type IV n = 25	
Primary teeth					
dt	1.8 ± 3.2	0.9 ± 2.0	3.2 ± 4.3	1.7 ± 3.1	0.111
mt	1.3 ± 2.4	1.1 ± 2.8	1.6 ± 2.3	1.2 ±2.0	0.771
ft	0	0	0	0	-
dmft	3.0 ± 4.1	1.9 ± 3.4	4.9 ± 4.4	2.8 ± 4.4	0.119
Permanent teeth					
DT	1.7 ± 2.4	1.0 ± 1.6	2.3 ±2.9	1.6 ± 2.3	0.295
MT	0.3 ± 1.8	0.9 ± 3.5	0	0	0.272
FT	0	0	0	0	-
DMFT	2.2 ± 4.6	2.9 ±7 .9	2.3 ± 2.8	1.6 ± 2.3	0.753

ANOVA test
dt/DT: decayed teeth; mt/MT = missing teeth; ft/FT = filled teeth.

study was 74 OI persons from thirty-four provinces that are home to approximately 60 million of the total population of Vietnam, meaning that the prevalence of OI in Vietnam is estimated at 1/480,000. Our prevalence might be lower compared to the prevalence of 1/25,000-1/10,000 reported in other countries [3-5]. Our study lacked information about OI in the newborn infant group, and OI's mild type might be undiagnosed in the general population; in addition, OI type II was excluded from the present study. Nonetheless, the distribution of OI types in our study approached a range distribution of 39-79% for type I, 9-24% for type III, and 13-40% for type IV, as reported in previous studies [10,11,16,17]. Bisphosphonate therapy has good results in increasing the bone mineral density; however, most of our participants could not follow bisphosphonate therapy due to inaccessibility of adequate medical care, sustainment or counseling from the medical professionals.

4.2. Dentinogenesis imperfecta and dental wear

A high prevalence of DGI was found in Vietnamese with OI. Our results were in accordance with a study by Majorana et al. [12] that indicated that 62.5% of an Italian OI sample had DGI. Conversely, Malgrem [11] and Saeves [10] found that the prevalence of DGI in OI samples in Sweden and Norway was 41.5% and 19.0%, respectively. Concerning DGI related to types of OI, our study is consistent with previous studies in showing DGI to be more prevalent in OI type III than in type I and IV [10,18], indicating that DGI is related to the severity of OI. DGI is a disorder of dentin formation causing deposition of dentine, obliteration of the pulp chamber and intrinsic discoloration. This could explain the finding that up to 80% of dentition in our OI sample were graded as having a dark lightness level and yellow-red intermediate to reddish color. Our study found that 36.5% of OI persons had dental wear. This is in line with previous investigations that found the prevalence of dental wear ranging from 37.5% to 66.5% in OI samples [11,12]. Dental wear occurring in OI person could be from DGI. Among DGI types of Shields' classification, DGI type I is associated with OI because of the inherited disorders of collagen metabolism; whereas, DGI type II and III are mutations affecting the dentin sialophosphoprotein gene [13]. The mutations in COL1A1 and COL1A2 genes would cause DGI type I that teeth easily expose the abnormal dentine and were typically worn.

Approximately 10% of teeth in both dentitions were showed a severely worn condition in the current study. Preventive and restorative care of DGI and dental wear are important for Vietnamese OI persons. The treatment considerations are preservation of occlusal height, maintenance of oral function and esthetic needs. Nonetheless, most of OI persons might not receive dental treatment because of the family economic hardship; thus, there is a need for more effective support for OI persons to approach treatment to restore the harmony of the oral functions.

4.3. Occlusal features

OI mutation not only has an impact on dental structure but also on dental occlusion. The findings of our study indicate that class III malocclusion presented in 54.2% of the sample, which was similar to 60-80% of class III malocclusion in investigations in Taiwan and Canada [7,9]. In the current study, class III malocclusion was more prevalent in OI type III compared to type IV and I. An individual with OI type III is described as having a triangular face, and this feature might be associated with class III malocclusion. High prevalence of class III in

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the Vietnamese OI sample corresponded with the prevalence of OI persons having reverse overjet. Reverse overjet is a manifestation of disharmony between the maxilla and the mandible. In the current study, OI persons were likely to have deficient growth of the maxilla. The evidence was that a posterior crossbite presented in 25.0-42.9% of OI types, with no OI persons having an overjet > 3.5mm, a parameter indicating a protrusive maxilla. Our study was strongly supported by previous studies. Chang et al. [7] reported a shorter upper facial length in an OI sample as compared to a healthy sample. By using the discrepancy index in orthodontic treatment, Rizkalla et al. [9] found OI to be related with anterior and posterior crossbite. Scalia et al. [19] concluded that malocclusion of OI was associated with a retrognathic maxilla. The impairment of maxilla growth could be from a lack of type I collagen due to mutation. In addition, the symptom of loose joints could be observed in severe cases of OI. When occurring in the temporomandibular joint, it can stretch more than normal and lead to abnormal jaw relations. The deficiency of maxillary length might also lead to teeth crowding. Our study found that both the prevalence of displacement of tooth > 2mm and incisal segment crowding gradually increased from OI type I to type IV and type III. According to Sillence's classification for living OI patients, type I is the mildest form, whereas type III is the most severe form. Such severe deformities of OI type III and IV influenced the defective growth of the maxilla and might also be associated with disharmony of growth between the two jaws.

The evidence was that over 20% of people with OI type III and IV had an open bite in contrast with 0% of type I. Waltimo-Siren et al. [17] indicated that the gonial angle of OI type I was 124.6 degrees, which was lower compared to the 126.3 degrees of OI type III/IV. Similarly, Chang et al. [7] found a clockwise rotation of the mandible of OI patients as compared to the control group. Such alterations in the mandible might be associated with an open-bite of occlusion in OI patients.

The mutations of COL1A1 and COL1A2 in persons with OI might prohibit tooth formation and development. In the current study, the prevalence of missing teeth was equivalent among OI types, and it ranged from 22.7-23.8%; that was much higher compared to 0.5-11.0% of missing teeth in the general population [20]. The findings related to missing teeth in our study are reinforced by previous OI studies. Tooth agenesis was found in 17% of the OI sample, including 11% hypodontia and 6% oligodontia [18].

Approximately 14% of individuals with OI type III had congenital missing teeth [21], and in our study, missing teeth accounted for 23.8% of OI type III. The odds of having missing teeth among persons with OI was more 2.0-4.7 times compared to the general population [8,10].

4.4. Dental caries experience

Dental caries is a major concern for persons with OI because of difficulties with physical activity for oral hygiene. The mean score dmft/DMFT of 3.0/2.2 indicated a moderate level of dental caries experience in OI sample. Our findings were in accordance with previous studies that highlighted oral problems among the OI population. Saeves et al. [10] described that although OI patients in Norway had regular dental visits and daily oral health habits, their oral status was not as good as compared to the general population. Differently from the findings of Saeves in Norway, none of the Vietnamese individuals with OI received any restorative treatment for decayed teeth. The physical disability of persons with OI might influence their ability to visit a dentist for dental treatment; in addition, most of them were from healthcare centers that only focused on rehabilitation of OI patients. The dental issues and lack of dental visits could accelerate caries development in OI persons; however, the number of decayed teeth of Vietnamese people with OI was lower compared to the general population in Vietnam [22]. This is possibly related to DGI presenting in persons with OI. In DGI dentition, the presence of obliterated dentinal tubules and pulp chamber can prevent penetration of harmful bacteria, although enamel has chipped away. This might explain that the missing teeth component in our study (mt/MT=1.3/0.3) resulted from hypodontia as aforementioned, not by the impact of caries. The shortcoming of our study is an absence the control group to compare with the OI sample in evaluation the risk of oral problems. In addition, radiographic examination was not carried out for OI participants to determine the reasons for missing teeth due to impacted teeth or hypodontia.

5. CONCLUSION

There was a high prevalence of dentinogenesis imperfecta and dental wear in the Vietnamese OI sample, especially in OI type III and IV. The occlusal features of OI persons were determined with a high prevalence of class III, malocclusion, overjet, open bite, posterior crossbite, and missing teeth. The dental caries experience of persons with OI was at a moderate level, and none of the study's subjects had received any dental restorations.

CONFLICT OF INTEREST

The authors declare no conflict of interest.

AUTHOR CONTRIBUTIONS

MSN: performed clinical studies, data acquisition, statistical analysis, and manuscripts writing, MS: analysis results, proofreading, BH: data acquisition, KM: data acquisition and interpretation of the results, SK: interpretation of the results, AM: data acquisition, TT: data acquisition and manuscript literature search, TJ: protocol, proofreading.

Original Articles

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Questions

1. Osteogenesis imperfecta is a genetic mutation affecting

❑a. Connective tissue;
❑b. Bone;
❑c. Eyes;
❑d. Dentition.

2. Which type of osteogenesis imperfecta cannot be observed in living persons?

❑a. Type I;
❑b. Type II;
❑c. Type III;
❑d. Type IV.

3. The high prevalence of malocclusion that can be observed in persons with OI is related to

❑a. Angle's class I;
❑b. Angle's class II, division I;
❑c. Angle's class II, division II;
❑d. Angle's class III.

4. Which type of dentinogenesis imperfecta is associated with osteogenesis imperfecta?

❑a. Only DGI type I;
❑b. DGI type I and type II;
❑c. DGI type I and type III;
❑d. DGI type II and type III.