



# JOCPPD

Journal of Clinical Pediatric Dentistry

Review Articles  
Clinical Articles  
Clinical Research for Better Practice  
Oral Pathology  
Hospital Dentistry  
Growth and Development  
Dentofacial Orthopedics and Orthodontics

The Source for Modern Pediatric Dentistry

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# The Journal of Clinical Pediatric Dentistry

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# Function of the Family Unit, Oral Hygiene Rules and Attitudes to Dental Health in Children During First-Wave 2020 COVID-19 Lockdown

Sigalit Blumer\*/ Nurit Dagon \*\*/Benjamin Peretz\*\*\*/ Tal Ratson \*\*\*\*/ Johnny Kharouba \*\*\*\*\*

**Objective:** To examine whether general and dental health and habits of families were affected by the first-wave lockdown due to COVID-19, and whether these habits were related to family functioning, resilience and stress. **Study design:** A cross-sectional study using an online survey disseminated among families with kindergarten and primary school-aged children during the lockdown of March and April 2020. **Results:** A total of 361 respondents completed the survey. Most respondents adapted well to the changes imposed by lockdown and reported that they and their children had low anxiety levels and high mental resilience. Family functioning and behavior were positively correlated with nutrition habits and hygiene. General hygiene was positively correlated with oral hygiene. Respondents who reported requiring dental care had difficulties gaining access to it. Most respondents perceived that it is important to improve patients' digital access to pediatricians and dentists during crises. **Conclusion:** The study showed that better family functioning was associated with better family hygiene and nutrition, parental resilience and lower mental stress among children.

**Keywords:** oral health, corona virus epidemic, children, hygiene, family functioning

## INTRODUCTION

On 31 December 2019, an outbreak of a new corona virus in Wuhan (Hubei Province) in China was first reported. The World Health Organization (WHO) declared the outbreak a public health emergency of international concern on 30 January 2020. The disease caused by the virus was named COVID-19 on 11 February 2020, and was declared a pandemic by the WHO on 11 March, 2020.<sup>1</sup> The massive spread of the disease around the globe required citizen of many countries to change their behavior and normal daily functioning, practice social distancing and isolate to curb viral spread.

COVID-19 spreads mainly through droplets and fomites; therefore, dental professionals are at high risk for COVID-19 infection due to the nature of dental interventions, which include proximity of the dentist to the patient's oropharyngeal region and aerosol generation. Moreover, patients may be exposed to cross contamination if appropriate precautions are not taken.<sup>2</sup> In the wake of the pandemic, following guidance by professional dental associations, many countries saw the cancellation of routine dental care followed by urgent care, so that only emergency dental care was provided. The American Dental Association (ADA) suggested on March 16, 2020 that dentists defer all elective dental care for 3 weeks. The United States Centers for Disease Control and Prevention recommended in March 2020 that dental settings should prioritize urgent and emergency visits and delay elective visits and procedures to protect staff and preserve personal protective equipment and patient care supplies, as well as expand

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available hospital capacity.<sup>3</sup> In New Zealand, all non-essential and elective dental treatment was stopped on March 23 and on the same date all routine face-to-face dentistry was halted in Scotland, Wales and Northern Ireland.<sup>4,5</sup> During the same month, the Israeli Ministry of Health instructed the public to postpone non-essential dental treatments and dental offices were instructed to treat only emergency cases while maintaining strict preventive measures.<sup>6</sup> Guidelines were also published by various other dental organizations.<sup>7-10</sup>

Maintaining oral health in children is based on periodical check-ups and on provision of information on oral hygiene to the parents. Following the cessation of routine regular dental care during the pandemic, it was essential to focus on prevention and maintenance of oral health in the home. A diet rich in fruit and vegetables helps to protect against the onset of gum diseases and caries. Moreover, it is important to limit sugar sweetened beverage consumption<sup>11</sup>. Stressful life events, such as separation, unemployment, chronic illness or depression, may affect the functioning of families. Self-isolation measures and the lockdowns imposed by many countries meant that many families had to adjust and adapt to living together without going outside. Such changes could cause mental pressure, loneliness, and existential anxiety regarding family, friends and the changing world due to the many unknowns related to the new disease, including mortality and infection rates, symptoms, infectiousness, the economic situation and not knowing how long this situation would last. Studies on previous public health emergencies showed that they triggered stress emotional responses comprising higher anxiety levels and other negative emotions.<sup>12</sup> A study that evaluated the impacts of the declaration of a public health emergency in China due to COVID-19 on people's mental health, reported an increase in negative emotions (anxiety, depression, and indignation) and sensitivity to social risks, as well as a decrease in positive emotions and life satisfaction. The study also showed that people were more concerned about their health and families, and less concerned about friends and leisure.<sup>13</sup>

The unique situation imposed on families during lockdown has led us to investigate if dental health and habits were affected during lockdown and if these habits are related to family functioning, resilience and stress.

## MATERIALS AND METHOD

### Setting and participants

This was a cross-sectional study conducted during March and April 2020 among families with kindergarten and school-aged children in Israel. The study was approved by Tel Aviv University's ethics committee (research number 0001233-2). All participants provided their consent for study participation.

### Questionnaire and data collection

The study questionnaire (Figure 1) was constructed by the researchers. The questionnaire was validated for clarity and reliability on a sample of 10 respondents. Due to the lockdown, the questionnaire was disseminated to the study population electronically via the authors' email list and Facebook contacts. To access the questionnaire, the participants clicked a link and completed and submitted the questionnaire online through Google Forms. As such, the data were collected anonymously and automatically transferred to an Excel sheet on the researchers' server.

The questionnaire comprised 8 core variables, each consisting of several questions: 1) Family unit functioning and behavior (16 questions); 2) Nutrition habits (6 questions); 3) Family oral and hand hygiene (5 questions); 4) Access to medical treatments (12 questions); 5) Children's mental stress; 6) Parental functioning and mental resilience, 7) Coronavirus self-isolation and 8) Participant demographics.

### Statistical analysis

The data were analyzed using SPSS software version 25. Categorical variables were summarized as number and percent and continuous variables were summarized as mean and standard deviation. Then, the components of the core variables were summarized by mean and standard deviation, and associations were assessed using Spearman tests. The associations between the core variables, and the demographic characteristics were assessed using Mann-Whitney tests. To examine the relationships between socio-demographic variables, parental resilience, mental stress of children and nutrition habits, a hierarchical multiple linear regression analysis was conducted.

To assess the correlation between the general and oral hygiene, Spearman correlation tests were conducted between the general hygiene questions to the oral hygiene questions.

## RESULTS

### Participant demographics

A total of 361 individuals completed the questionnaires. Respondent demographics are shown in Table 1. The respondent's average age was 42.0±6.2 (SD) years. Most were female (87.0%), born in Israel (92.2%), were married (87.3%) and had an academic education (97.0%).

None of the respondents were diagnosed with COVID-19. Four respondents (1.1%) reported that a member of their family was diagnosed with the disease. Twenty-six respondents (7.2%) reported being instructed to self-isolate and 63 respondents (17.5%) reported that a member of their family had to self-isolate.

### Family behavior and functioning during lockdown

Over half of the respondents (54.0%) perceived that their lives were moderately at risk due to COVID-19, while 39.6% did not perceive any risk. Conversely, 54.0% did not perceive any risk to their children's lives, while 40.2% perceived a moderate risk to their children's lives.

Almost half of the respondents (48.2%) reported that their children were moderately concerned due to the COVID-19 outbreak while 44.9% reported that their children were not concerned at all. According to the respondents, most children (82.3%) were only moderately concerned or not at all concerned about their parents' health, but 83.9% were moderately to very much concerned about their grandparents' health.

Over two-thirds of respondents replied that their children were adapting well or extremely well to the changes in schedule brought about by lockdown and to this situation (68.1 and 65.1%, respectively).

Most respondents reported managing to create a schooling schedule and perceived that are managing with their children's online learning (82.0% and 92%, respectively); 44.9% of

Figure 1: Study questionnaire

**Family and children's behavior**

For each statement below describing the behavior of the family and children during the corona crisis, please mark the number that describes your conduct since the beginning of the crisis (lockdown): (1) Not at all (2) Somewhat (3) To a great extent (4) To a very great extent.

- To what extent do you feel your life is at risk due to the corona virus?
- To what extent are you afraid that your children are at risk of dying?
- How much are your children afraid of the situation?
- How much do your children fear for their parents' health?
- How much do your children fear for their grandparents' health?
- To what extent do you buy food products for fear of a shortage?
- To what extent have your children managed to adapt to the changes in the daily routine?
- To what extent have you succeeded in getting your children to adapt an online studying routine?
- Do you feel that the children have adapted to the situation?
- To what extent does your child cope with online learning?
- To what extent do you, as a parent, cope with operating the computer and online learning?
- To what extent do you feel that learning in this way would help your child complete the required study material?
- To what extent do you feel that under the current situation you are able to focus and think clearly in order to convey your messages to your children?
- To what extent do you feel that the relationship among the siblings is good in the current situation?
- To what extent do you feel that your children can deal with unpleasant feelings?
- To what extent do you currently feel under pressure to focus and think clearly in order to convey your messages to your child?

**Nutrition habits**

For each statement below describing nutrition during this period, please mark the number the represents the most appropriate answer: (1) Not true at all (2) Sometimes true (3) Often true (4) True almost all the time.

- I manage to get my kids to have an eating schedule.
- I feel I control the quality of the food the children eat.
- I feel I control the amount of food the children eat.
- We talk more about eating and the types of food being eaten.
- Consumption of salty and sweet snacks has increased considerably.
- I forbid my children to consume large amounts of snacks because of dental health fears.

**Hygiene habits**

For each statement below describing hygiene during this period, please mark the number the represents the most appropriate answer: (1) Not at all (2) Somewhat (3) To a great extent.

- Are the rules of hygiene observed at home according to the guidelines?
- Tooth brushing habits are maintained in a similar way to those before the crisis.
- I put more emphasis on brushing teeth with the children.
- The children are compliant and obey the new rules of hygiene (hand washing).
- The children are compliant and obey the rules of hygiene relating to brushing teeth more than usual.

**Access to medical treatments**

For each question below about accessibility to medical treatments please indicate (1) Yes (2) No.

- During this period of staying at home, did your child need medical care (not teeth)?

If the answer is "Yes",

- Was treatment immediately accessible?
- Were you please with the accessibility of the pediatrician?
- Did your child have a toothache or discomfort during this period?

If the answer is "Yes",

- Have you had a problem accessing dental care in the medical setting to which you belong?
- Do you think it is necessary to intentionally improve medical accessibility to pediatricians in times of crisis?
- Would provision of access to dentists in order to ask questions help you in times of crisis?
- Are you afraid to take your child to the dental clinic for emergency care because you fear getting infected by the corona virus?
- Are you refraining from going to the dental clinic these days due to the financial situation?
- Did your child receive dental care during this period?

If the answer is "Yes",

- Was treatment provided by the public healthcare services?
- Was treatment provided by private services?

**The children's feelings**

Below is a list of feelings and problems that children may have encountered during this period. Please read each section and mark the appropriate answer: (1) Not at all (2) Slightly (3) Somewhat (4) To a large extent.

- Nervousness.
- Loneliness.
- Upset.
- Disinterest.
- Anger.
- Fear.
- Anxiety of going to public places.

**Parent's emotional state**

Please indicate your feeling mood as a parent during the Corona crisis (lockdown) in the following situations: (1) Extremely bad (2) Not good (3) Quite good (4) Very good.

- Work.
- Health.
- Free time.
- Social connections.
- Family relationships.
- Daily functioning.
- Giving confidence to your child.

**Demographics**

Age

Marital status: 1. Married 2. Divorced. 3. Widow. 4. In a relationship. 5. Single parent. 6. Other.

Education: 1. Elementary school. 2. High school. 3. Academic.

Were you born in Israel? 1. Yes. 2. No.

Were you instructed to self-isolate? 1. Yes. 2. No.

Has anyone in your family been instructed to self-isolate? 1. Yes. 2. No.

Were you instructed to self-isolate due to proximity to a patient with verified COVID-19 infection? 1. Yes. 2. No.

Has anyone in your family been instructed to self-isolate due to proximity to a patient with verified COVID-19 infection? 1. Yes. 2. No.

Have you been diagnosed with COVID-19? 1. Yes. 2. No.

Has anyone in your family been diagnosed with COVID-19? 1. Yes. 2. No.

respondents reported that their children adapted well to extremely well to online school lessons and 38.5% reported that their children adapted moderately to online lessons. About two-thirds of respondents (67.3%) felt that online school lessons would help their children to catch up with school material that was not taught due to closure of schools.

Most respondents (75.6%) reported that they remain focused and think clearly despite the situation. Most (74.2%) reported good relationships among siblings in the household and 61.2% perceived that their children could cope with bad feelings.

### Nutrition habits

Most respondents (69.0%) reported that there is a regular eating schedule at home almost all the time or very often, 76.2% and 64.3% perceived that they are in control—almost all the time or very often—of the quality and quantity of food, respectively, that their children eat. Over half of the respondents (55.4%) felt that food and eating are being discussed more often at this time, and 60.9% reported that consumption of snacks and sweets has increased in their household. Over half of respondents (52.5%) reported that their children are usually or very often not allowed to consume large quantities of snacks in order to maintain dental/oral health.

### Hygiene habits

Most respondents (78.9%) reported maintaining high levels of hygiene according to the guidelines in the household and 75.6% reported that their children comply with the new guidelines of washing their hands more often. A similar percentage (78.1%) reported that tooth brushing habits are the same as before lockdown, 33.2% reported that they attribute greater importance to tooth brushing than before, but only 22.7% reported that their children comply with brushing their teeth more often.

**Table 1. Participant demographics**

Parameter	Study population N=361
	Mean
<b>Age, mean ± SD</b>	42.0 ± 6.2
<b>Gender, n (%)</b>	
Male	47 (13.0)
Female	314 (87.0)
<b>Marital status, n (%)</b>	
Married	315 (87.3)
Divorced	22 (6.1)
Widow	4 (1.1)
In relationship	11 (3.0)
Single parent	7 (1.9)
Other	2 (0.6)
<b>Education, n (%)</b>	
Academic (>12 years)	350 (97.0)
High school (≤12 years)	11 (3.0)
<b>Country of birth, n (%)</b>	
Israel	333 (92.2)
Abroad	28 (7.8)

n= number, SD=standard deviation

### Access to urgent medical and dental care

Most respondents (83.4%) reported that their children did not require medical treatments during lockdown. The vast majority of those who did require medical treatments (59 of 60 respondents, 99.3%) did not have any problems in receiving it. Regarding dental treatments, 12.5% of respondents reported that their children had a toothache or some other related oral discomfort. Of these respondents, 60% (27 of 45) reported receiving dental care as well as difficulties in accessing it.

Most respondents responded that in times of crisis it is important to improve digital access to pediatricians and dentists (87.3% and 85.2%, respectively).

Two third of respondents reported being apprehensive in seeking out dental treatment for their children because of fear of COVID-19 infection.

### Children’s mental stress during lockdown

When asked about their children’s feelings during this period, most respondents reported that their children were not at all or only slightly anxious (74.5%), lonely (73.1%), moody (80.3%), disinterested (80.6%), or afraid (88.9%). Over two-thirds of respondents (67.9%) reported that their children were not fearful at all or only slightly fearful of going to public places.

### Parental functioning and mental resilience during lockdown

When asked about their own feelings during the COVID-19 crisis, the majority of respondents perceived their health, daily functioning and family relations (96.1, 93.1 and 88.9%, respectively) as quite good to very good, and over two-thirds (67.6%) reported that they felt quite good to very good about their work. About half of the respondents (52.1%) felt the quantity of their free time was quite good to very good and 56.8% reported their social relationships were quite good to very good.

### Association between family functioning and behavior, hygiene, nutrition, parental mental resilience, children’s mental stress and studying routine

Table 2 shows the means and standard deviations of core variables in the study and Table 3 shows the means and standard deviations of the items relating to nutrition and hygiene habits.

Analysis of the association between the core variables in the study, using Spearman correlation (Table 4), showed that family functioning and behavior are positively correlated with nutrition habits ( $r=0.20, p<0.01$ ), hygiene habits ( $r=0.21, p<0.01$ ) and parental mental resilience ( $r=0.39, p<0.01$ ), and negatively correlated with their children’s mental stress ( $r=-0.15, p<0.01$ ).

Nutrition habits are positively correlated with hygiene habits ( $r=0.23, p<0.01$ ), and with parental mental resilience ( $r=0.17, p<0.01$ ), and with lower mental stress of children ( $r=-0.18, p<0.01$ ). Parental mental resilience was positively correlated with nutrition ( $r=0.17, p<0.01$ ) and hygiene habits ( $r=0.14, p<0.01$ ). Married parents reported marginally better nutrition habits in comparison with non-married parents ( $p=0.07$  by Mann-Whitney test) but no association was found between marital status and hygiene habits.

A negative correlation was found between children’s mental stress and nutrition habits ( $r=-0.18, p<0.01$ ), but no correlation was found between the children’s mental stress and oral hygiene ( $r=-0.06, p=0.27$ ).

**Table 2: Means, standard deviations and ranges for the core variables**

	Mean	Standard Deviation	Range
Family behavior	2.34	0.32	1 (not at all)–4 (to a very great extent)
Nutrition habits	2.85	0.53	1 (not true at all)–4 (true almost all the time)
Hygiene habits	2.43	0.39	1 (not at all) – 3 (to a great extent)
Access to urgent medical care	3.59	1.66	0 (none of the answers were positive) – 10 (all answers were positive)*
Mental stress of children	1.91	0.59	1 (not at all) – 4 (to a great extent)
Parental mental functioning and resilience	3.02	0.45	1 (extremely bad) – 4 (very good)
Coronavirus social isolation	0.38	0.84	1 (none of the answers were positive) – 4 (all answers were positive)*

\*Excluding two duplicate questions

**Table 3: Means, standard deviations, and ranges for the nutrition and hygiene questions**

	Mean	Standard deviation	Range*
<b>Nutrition</b>			
I manage to get my kids to have an eating schedule.	2.92	0.89	1-4
I feel I control the quality of the food the children eat.	3.07	0.83	1-4
I feel I control the amount of food the children eat.	2.78	0.88	1-4
We talk more about eating and the types of food being eaten.	2.58	0.93	1-4
Consumption of salty and sweet snacks has increased considerably.	3.15	0.82	1-4
I forbid my children to consume large amounts of snacks due to risk for oral health.	2.61	0.94	1-4
<b>Hygiene</b>			
Are the rules of hygiene observed at home according to the guidelines?	2.78	0.43	1-3
Tooth brushing habits are maintained in a similar way to those before the crisis.	2.76	0.48	1-3
I put more emphasis on brushing teeth with the children.	2.02	0.80	1-3
The children are compliant and obey the new rules of hygiene (hand washing).	2.74	0.47	1-3
The children are compliant and obey the rules of hygiene relating to brushing teeth more than usual.	1.86	0.76	1-3

\*Range for questions on nutrition habits: (1) Not true at all (2) Sometimes true (3) Often true (4) True almost all the time; Range for questions on hygiene habits: (1) Not at all (2) Somewhat (3) To a great extent.

**Table 4: Associations between the core variables.**

	1	2	3	4	5	6
Family behavior						
Nutrition habits	0.20**					
Hygiene habits	0.21**	0.23**				
Accessibility to medical treatments	0.03	-0.07	0.03			
Mental stress of children	-0.15**	-0.18**	-0.11*	0.20**		
Parental mental resilience	0.39**	0.17**	0.14**	-0.14*	-0.31**	
Coronavirus social isolation	0.01	0.04	-0.05	0.02	-0.03	-0.02

\*p <0.05, \*\*p <0.01

Oral hygiene was assessed using the questions “*Tooth brushing habits are maintained in a similar way to those before the crisis*”, “*I put greater emphasis on brushing teeth with the children*” and “*The children are compliant and obey the rules of hygiene relating to brushing teeth more than usual*”. The average score was 2.21 (SD=0.52, range=1-3). General hygiene was assessed using the questions “*Are the rules of hygiene observed at home according to the guidelines?*” and “*The children are compliant and obey the new rules of hygiene (hand washing)*”. The average score was 2.76

(SD=0.38, range=1-3). General hygiene was positively correlated with oral hygiene (r=0.35, p<0.01 by Mann Whitney test).

Greater access to urgent medical care was positively correlated with higher parental mental resilience (r=0.20, p<0.01), and negatively correlated with children’s mental stress (r=-0.14, p<0.01). Married parents reported less access to urgent medical care (Mean=3.53, SD=1.64) than non-married parents (Mean=4.00, SD=1.70), but this difference was only marginally significant (p=0.06).

No correlation was found between Coronavirus self-isolation and the other variables. Difference between participants who were in self-isolation and participants who were not in self-isolation, were assessed using Mann-Whitney tests. The results showed a marginal difference between self-isolation and hygiene habits ( $p=0.053$ ). That is, parents who had been in self-isolation (Mean=2.58, SD=0.33) reported better hygiene habits in comparison parents who had not been in self-isolation (Mean=2.42, SD=0.39). No difference in hygiene or nutrition habits were found between those who had a family member in self-isolation and those who did not. There were no differences in nutrition and oral habits between respondents who had a family member with COVID-19 and those who did not.

To assess the correlation between the children’s learning routine and nutrition and hygiene habits at home, the questions “*To what extent have your children managed to adapt to the changes in the daily routine*”, “*To what extent have you succeeded in getting your children to adapt an online studying routine*”, “*Do you feel that the children have adapted to the situation?*”, “*To what extent does your child cope with online learning?*”, “*To what extent do you, as a parent, cope with operating the computer and online learning?*” and “*To what extent do you currently feel under pressure to focus and think clearly in order to convey your messages to your child?*” were averaged for one variable. The average score was 2.55 (SD=0.53, range=1.14-4.00). The children’s studying routine was positively correlated with nutrition ( $r=0.27$ ,  $p<0.01$ ) and hygiene habits ( $r=0.21$ ,  $p<0.01$ ).

**Multivariate analysis for predicting nutrition habits during lockdown**

To examine the relationships between socio-demographic variables, parental resilience, children’s mental stress and nutrition habits, a hierarchical multiple linear regression analysis was conducted. The variables in the model accounted for approximately 16.20% of the total variance in nutrition habits ( $F_{(10, 348)}=6.73$ ,  $p<0.01$ ).

Results showed a negative correlation between respondents’ age and nutrition habits ( $\beta=-0.20$ ,  $p<0.01$ ), meaning that younger parents maintain better nutrition habits in comparison with older parents. Family functioning and behavior ( $\beta=0.18$ ,  $p<0.01$ ) and hygiene habits ( $\beta=0.14$ ,  $p<0.01$ ), positively predicted nutrition habits, while mental stress of children ( $\beta=-0.14$ ,  $p=0.01$ ) negatively predicted nutrition habits. (Table 5).

Multivariate analysis for predicting hygiene habits during lockdown

To examine the relationships between socio-demographic variables, parental attitudes, children’s feelings and nutrition habits, a hierarchical multiple linear regression analysis was conducted. The variables in the model accounted for approximately 11.70% of the total variance in hygiene habits ( $F_{(10, 348)}=4.60$ ,  $p<0.01$ ).

Results showed a negative correlation between age and hygiene habits ( $\beta=-0.13$ ,  $p<0.05$ ), meaning younger parents keep better hygiene habits in comparison with older parents. Family behavior ( $\beta=0.22$ ,  $p<0.01$ ) and nutrition habits ( $\beta=0.15$ ,  $p<0.01$ ), positively predicted hygiene habits (Table 6).

**Table 5: Standardized coefficients (β) to predict nutrition habits during lockdown**

Nutrition habits			
1	Age	-0.15**	-0.20**
	Gender (male)	0.00	0.02
	Marital status (married)	0.05	0.01
	Country of birth (Israel)	-0.05	-0.05
2	Family behavior		0.18**
	Hygiene habits		0.14**
	Accessibility to medical treatments		-0.03
	Mental stress of children		-0.14*
	Parental resilience		0.09
	Coronavirus insulation and adhesion		0.06
	R <sup>2</sup>	0.03*	0.16**
	R <sup>2</sup> change	0.03*	0.13**

\* $p<0.05$ , \*\* $p<0.01$

**Table 6: Standardized coefficients (β) to predict hygiene habits during Coronavirus social isolation**

Hygiene habits			
1	Age	-0.08	-0.13*
	Gender (male)	-0.03	-0.02
	Marital status (married)	-0.02	-0.06
	Country of birth (Israel)	-0.06	-0.05
2	Family behavior		0.22**
	Nutrition habits		0.15**
	Accessibility to medical treatments		0.04
	Mental stress of children		-0.03
	Parental resilience		0.07
	Coronavirus insulation and adhesion		-0.02
	R <sup>2</sup>	0.01	0.12**
	R <sup>2</sup> change	0.01	0.11**

\* $p < .05$ , \*\* $p < .01$

**DISCUSSION**

Our findings show that lockdown due to the COVID-19 pandemic did not greatly affect most of our study population: According to the respondents they adapted well to the changes imposed by lockdown and reported low anxiety levels and high mental resilience – both for themselves and their children. Our findings also show that better family functioning and behavior and better nutrition habits are correlated with improved hygiene, and that better general hygiene is correlated with better dental hygiene.

Parents’ oral health-related attitudes and their dental behaviors, such as locus of control and self-efficacy impact their children’s dental health,<sup>14-18</sup> In addition, parental psychosocial factors, such as low sense of coherence, maternal depression, parental stress and indulgent parenting, have shown to negatively affect their children’s oral health.<sup>19-21</sup>

Renzaho and de Silva-Sanigorski investigated the relationships between child dental health family structure, parental distress, and family functioning. They reported that poor family functioning,

parental mental illness and child mental health or conduct problems were all associated with poor oral health in children. These findings were independent of parental education, household income, language spoken at home, country of birth, child gender and parent's age.<sup>22</sup> Similarly, Duijster *et al* reported that lower family functioning was associated with an increased likelihood of engaging in less favorable oral hygiene behaviors.<sup>23</sup> Family functioning did not significantly differ between single parents and those who raised their child with a partner. In the current study married parents reported marginally better nutrition habits in comparison with non-married parents but no association was found between marital status and hygiene habits.

The vast majority of those who required medical care did not have any problems receiving it. In contrast, those who reported requiring dental care had difficulties gaining access to such treatments because dentists feared being infected with corona virus, or due to travel restrictions. Importantly, most respondents perceived that it is important to improve digital access to pediatricians and dentists during times of crises. Indeed, the pandemic may have hastened the process of using telehealth in dentistry to screen patients for dental problems.

Our study is limited by the cross-sectional nature of the data, which limit our ability to infer casual relationships. In addition, the data are self-reported, which introduces risks of recall biases and social desirability. Moreover, some of the measures should be considered as the parental perception of their children's mental state rather than the child's mental state. Sociodemographic variables, including the number of children in the family and their ages were also not assessed; however, questionnaires were sent to families with children who attend kindergartens and primary schools. Family structure was previously found to be associated with children's dental health: children from larger households, single-parent families, and those with higher birth order showed higher levels of caries.<sup>24</sup> The study participants were obtained through a convenience sample because, due to the lockdown, the only possibility to reach the target population was electronically by email or social media (Facebook); therefore, the sampled population behavior may not be generalizable to the general population in Israel. The vast majority of respondents were women with an academic education, indicating a higher socioeconomic position.<sup>25</sup> Data on family functioning relied on the point of view of one individual family member, which may have resulted in a biased view of the family functioning.

## CONCLUSIONS

The current study showed that better family functioning during lockdown due to the COVID-19 epidemic was associated with better family hygiene and nutrition, parental resilience and lower mental stress among children, as reported by their parents.

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### Conflicts of Interest:

The authors declare no conflict of interest

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## Prefabricated Zirconia Crowns – A Solution to Treat Hypomineralized Permanent Molars: Report of a Case

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*The presence of carious lesions in children associated with developmental defects of enamel is frequently observed. Restoring these affected teeth can be a challenge for the clinician. Teeth with enamel defects may have poor or limited resin adhesion and some may require repeated restoration. Prefabricated zirconia permanent molar crowns were recently introduced as an option for restoring severely decayed and broken down young permanent molars. These new restorations offer an efficient, esthetic, and economic option to restore severely broken down carious permanent molars that may be associated with enamel defects in partially or fully erupted molars.*

*A clinical case of a 13-year-old female patient is presented. She had a mandibular second permanent molar that demonstrated significant caries and loss of much of the clinical crown, which was treated with a vital pulpotomy and restored with a prefabricated zirconia crown.*

**Keywords:** Molar Incisor Hypomineralization, Enamel defects, prefabricated zirconia crown, Mineral Trioxide Aggregate.

### INTRODUCTION

**M**olar Incisor Hypomineralization (MIH) is the term used to describe a clinical condition that is reported as increasing in frequency and whose prevalence ranges between 2.8 and 40.2% of the population<sup>1,2</sup>. It is estimated that this condition affects, on average, one in six children worldwide<sup>1</sup>. The term Molar Incisor Hypomineralization was first introduced by Weerheijm in 2001<sup>3,4</sup>. These authors defined this entity as hypomineralization of systemic origin, presenting with defects in tooth enamel, in at least one and in up to four permanent first molars and is frequently associated with similarly affected incisors<sup>4,5</sup>.

Clinically, MIH is considered an opacity in the enamel, whose color could range from shades of white to cream to brown<sup>4,6</sup>. It is usually found in at least one of the first permanent molars, although MIH can also be detected in both primary and permanent second molars<sup>4,5,7</sup>. It is noteworthy that the presence of hypomineralized second primary molars increases the possibility of MIH in the permanent dentition; however, the absence of hypomineralized second primary molars does not eliminate the possibility of presenting MIH<sup>7,8</sup>. In MIH, the permanent molars may show rapid caries progression starting shortly after the eruption of the tooth.

Patients presenting with visible enamel defects may report dental sensitivity to heat, cold, certain foods and, sometimes also to toothpastes and tooth brushing. The breakdown and roughness of the hypomineralized enamel can favor the accumulation of biofilm and the consequent development of tooth decay.<sup>1,2,9</sup>

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Presented here is a report of a 13-year-old female patient with enamel defects in the maxillary central incisors and significant defects and breakdown of a mandibular second permanent molar. She reported frequent episodes of dental sensitivity. The permanent molar was restored with a prefabricated zirconia crown. Prior to placement of the crown, a vital pulpotomy was performed by the placement of Mineral Trioxide Aggregate (MTA), a bioactive material. After 40 months the tooth demonstrated very good clinical and radiographic results.

**Case Report**

A 13-year-old female patient presented to the office of the principal author(JC) with the chief complaint of an extremely sensitive mandibular right second permanent molar (Figure 1). The patient reported pain caused by temperature changes, particularly cold, which prevented her from chewing any food on the right side of her mouth. Toothbrushing with toothpaste also caused some discomfort; thus, the patient avoided toothbrushing.

The tooth had previously been restored using composite resin restorations, but those restorations had failed and been lost. Clinically, the affected molar exhibited a large, extensive carious lesion and had lost much of the clinical crown.

Due to the extensive carious lesion associated with the MIH and the lack of sound enamel capable of being etched and bonded, a resin restoration was contraindicated. It was decided that full coverage restoration was needed. Historically, preformed metal crowns have been used to restore young, badly broken down permanent molars, but in this instance there was a desire for a more esthetic restoration. It was decided to restore the tooth with a prefabricated zirconia crown (NuSmile ZR Permanent Molar Crown; NuSmile, Ltd., Houston, TX, USA).

The child was anesthetized with the administration of a single 1.8-ml cartridge of Articaine 4% with epinephrine 1/200,000 (Medicaine 1/200,000; Septodont, Saint-Maur-des-Fossés Cedex, France). A rubber dam was placed, isolating the affected tooth. With

a low-speed #6 carbide round bur, the carious tooth structure was carefully excavated. This, however, lead to a carious exposure of the pulp, which was found to be vital. As this was a young, permanent molar it was decided to treat the pulp with an MTA vital pulpotomy. (Figure 2).

The coronal pulp was removed and a wet cotton pellet moistened with a 1.25% solution of sodium hypochlorite was placed on the pulp stumps for approximately 1 min. The pulp chamber was then rinsed with sterile saline solution. A portion of Mineral Trioxide Aggregate cement (NeoMTA; NuSmile, Ltd, Houston, TX. USA) was mixed following the manufacturer’s instructions to a putty-like consistency. The MTA was placed into the pulp chamber with an amalgam carrier and was gently condensed with a wet cotton pellet. The MTA was then covered with a 2-3mm layer of resin-modified glass ionomer cement (BioCem; NuSmile, Ltd, Houston,TX. USA) to protect the MTA during the subsequent process of tooth preparation and crown placement.

Preparation for the zirconia crown was done as conservatively as possible. This consisted of occlusal reduction of approximately 1.5 mm; and circumferential reduction of 1-1.5mm, removing all surface convexity, with a gingival feather-edge margin extending approximately 1.5 mm subgingivally. (Figure 3) As this was a molar in an active process of eruption it was desired to place the gingival margin at a depth that would remain subgingival at the completion of eruption.

**Figure 1. Initial image of a hypomineralized and carious right mandibular second permanent molar with a very extensive occlusal lesion associated with developmental defects of the enamel.**



**Figure 2. Occlusal view of the pulpotomy.**



**Figure 3. Preparation performed on the mandibular second molar.**



After fitting the crown to a passive fit, the occlusion was checked, and the crown was deemed ready to cement. Adhesion of resin modified glass ionomer cements to zirconia contaminated by saliva has been shown to be poorer than to non-contaminated zirconia, so prior to cementation the crown was thoroughly rinsed and its inner surface was cleaned with a cleaning solution containing zirconium oxide particles (Ivoclean; Ivoclar Vivadent, Schaan, Liechtenstein) to remove traces of phosphates contained in the saliva that contaminated the crown during try-in. (It should be noted that the NuSmile Permanent Crown kits now come equipped with pink “try-in” crowns which can be used to trial fit the prefabricated zirconia crowns, avoiding the contamination of the zirconia crowns to be cemented.) The crown was then cemented with resin-modified glass ionomer cement (BioCem; NuSmile, Ltd, Houston, TX, USA) following the manufacturer’s instructions. A post-operative radiograph was taken to evaluate the final fit and cementation. Excess retained cement was noted on the radiograph and removed. The fit of the crown and evaluation of the pulpal treatment was found to be very satisfactory. (Figure 4).

One week later the tooth was reassessed. Tooth sensitivity had completely disappeared, the gum around the crown was healthy, and the patient was very pleased with the treatment, reporting that she was able to eat, chew, and brush her teeth in a normal manner (Figure 5).

**Figure 4. X-ray of the cemented crown. Some excess of cement can be noted on the mesial of the crown.**



Fourteen months later, the patient initiated orthodontic treatment in the same dental office, allowing for continued follow-up of the tooth. At 40 months post-operatively the crown had retained its esthetics and function and the tooth had remained vital and asymptomatic. (Figures 6 and 7)

**Figure 5. Photograph of the crown 1 week after cementation. The crown appears clean, the gingival tissue is healthy, and tooth sensitivity has disappeared.**



**Figure 6. Follow-up at 40 months after crown placement. The tooth is asymptomatic and the gingival tissue appears healthy.**



**Figure 7. Periapical control x-ray 40 months post-op.**



## DISCUSSION

MIH has been classified into three severity levels. Mild MIH shows demarcated opacities located at non-stress bearing area, with no caries associated with the affected enamel. Moderate MIH demonstrates demarcated opacities present on molars and incisors, with post-eruptive enamel breakdown limited to one or 2 surfaces without cuspal involvement. Severe MIH (as seen in the present case) demonstrates post-eruptive enamel breakdown, crown destruction, caries associated with affected enamel, and a history of dental sensitivity<sup>10</sup>.

Hypomineralized molars often present a restorative challenge for the dentist, particularly due to the difficulty of restoring them with adhesive systems such as composite resins<sup>11</sup>. This is due to the poor or inadequate etching of the defective enamel obtained after the application of phosphoric acid<sup>11-14</sup>. This inadequate etching a result of higher than normal organic/protein content of the affected enamel and significantly lower mineralization than is found in sound, unaffected enamel. Another common problem lies in the difficulty in determining where affected enamel ends, and normal, strong enamel begins. This is important for creating an adequate finish line for the restoration, especially in partially erupted molars. Occasionally glass ionomers, or resin modified glass ionomers may be used to restore MIH teeth, but these are usually considered intermediate restorations. Prefabricated metal crowns have been used to restore severely damaged MIH teeth with high long-term survival rates<sup>2,14</sup>. While durable and economical, these crowns lack esthetic appeal. Full coronal custom coverage with laboratory fabricated crowns can also be done, particularly in older children, but is time-consuming, technique-sensitive and expensive.

In the present case, there had been previous attempts to restore the tooth with resin, but failure of those restorations left the patient's parents seeking another restorative option; one that would solve dental sensitivity and that was esthetically pleasing, as well as durable.

The pulp exposure of an immature permanent molar during caries excavation required the decision of either performing a full root canal treatment or a pulpotomy. While the root apices were nearly closed, it was decided that a vital pulpotomy would be the best treatment to maintain vitality and allow for completion of root end closure<sup>15-17</sup>.

Yet another treatment option to pulpal therapy and placement of a crown was the removal of the affected molar and allowing the permanent third molar to erupt as a replacement a few years later. After discussing the pros and cons of both treatment alternatives with the patient's parents, it was decided to restore the affected second molar.

Due to the age of the patient, the second molar was still in active eruption. The crown margin was placed approximately 1.5 mm subgingivally in order to compensate for the subsequent eruption of the tooth, yet still maintain a subgingival margin

Forty months after restoration the molar was asymptomatic; and clinically and radiographically sound with an esthetic appearance. Both clinical and radiographic monitoring is ongoing to evaluate the continued vitality and clinical function of the crown. Zirconia is extremely durable and biocompatible, and the post-operative radiographs show good marginal adaptation of the crown. Replacement of this pre-fabricated crown with a custom crown may be necessary in the future, but that decision will be dictated only if failure of the present treatment occurs.

## CONCLUSION

Use of a prefabricated zirconia crown to restore a severely destroyed permanent second molar was shown to have excellent results in this case. Advantages of this treatment include the ability to provide an esthetic, durable restoration at a low cost that can be placed in a single appointment with no impressions or laboratory costs. In partially erupted molars, it is possible to place this type of crown since a precise finish line is not required as is necessary with a laboratory processed crown. Additionally, It is not necessary to refer the patient to another specialist because the crowns can be placed by a pediatric dentist.

Prefabricated zirconia crowns hold promise as a clinically and economically viable alternative for restoring permanent molars affected by enamel defects, tooth decay, and molar incisor hypomineralization.

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## Postoperative Bleeding Complications after Stainless Steel Crown Placement: A Case Series

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*Stainless steel crown (SSC) placement is a common pediatric restorative treatment, generally completed with minimal complications. Discussed in this case series are two patients who presented to the emergency department (ED) with moderate oral bleeding persisting more than 12 hours after oral rehabilitation under general anesthesia. Bleeding incidence after pediatric oral rehabilitation has been found to range from 20% to 40%, with most being considered mild and significantly associated with extractions. There is limited documentation regarding moderate, persistent postoperative bleeding associated with placement of preformed metal crowns. The objective of this paper is to raise awareness of postoperative bleeding following SSC placement, discuss probable causes to minimize complications in the future, and discuss the local measures that were used to obtain hemostasis.*

**Keywords:** *delayed postoperative bleeding, postoperative complications, stainless steel crowns, preformed metal crowns, pediatric dentistry.*

### INTRODUCTION

For almost 70 years, stainless steel crowns (SSC) have been used in the treatment of dental decay because of their durability, inexpensiveness, efficiency, and efficacy.<sup>1</sup> It is estimated that 20% of children two to five years of age have dental caries,<sup>2</sup> with many likely receiving restorative therapies such as SSCs. Indications for SSC placement include extensive caries, cervical decalcification, and developmental defects. The durability and predictability of SSCs make them particularly useful in management of high-risk children undergoing oral rehabilitation under general anesthesia.<sup>3</sup> The technique involved usually generates a minimal amount of self-limiting bleeding. Continuous oral bleeding is an unusual event in a patient without an underlying medical disorder.<sup>4</sup> More unusual is persistent moderate postoperative bleeding following SSC placement.<sup>5,6</sup> This case series is the first of our knowledge to formally discuss persistent postoperative bleeding associated with SSC placement.

### Case 1

A three-year-old female with a history of asthma, obstructive sleep apnea, and four-month post-surgical history of uncomplicated tonsillectomy and adenoidectomy (T&A) presented to the Nemours/Alfred I. duPont Hospital for Children's (AIDHC) emergency department (ED) 12 hours after oral rehabilitation because of persistent gingival bleeding around a lower right SSC. The treatment was completed in an outpatient ambulatory surgical center (ASC), and the patient presented with bilateral first mandibular molar SSCs, two anterior crowns, and composite restorations. The history as reported by the patient's mother included oral bleeding

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that was present upon discharge and continuation after application of gauze pressure at home. There were no known familial bleeding disorders; however, the father's medical history was unknown.

The patient presented with pallor, nausea, and some body bruising, but vital signs and capillary refill remained within normal limits. Upon examination, moderate bleeding was associated with the mandibular right first molar prepared with an SSC. The remaining oral cavity exhibited no bleeding. A liver clot was removed three separate times by the ED team prior to the dentist's ED arrival. The initial treatment was compression with gauze using digital pressure in five-minute increments, but bleeding continued, causing gauze saturation. It did, however, allow for visualization of subgingival cement in the distal buccal sulcus of the tooth. The cement was debrided, revealing a deep periodontal pocket. The area was then irrigated, and digital compression was repeated, with no resolution. A dental radiograph (Fig. 1) was taken, showing no abnormal findings. Absorbable sterile mesh was compressed into the distal buccal sulcus, followed by syringing of 15.5% ferric sulfate around the remaining circumference of the SSC. Hemostasis was achieved, and the patient was monitored for 90 minutes to confirm resolution before discharge. Coagulation abnormalities, malnutrition, and pulpal involvement were unlikely but unable to be fully ruled out. Post-discharge, the patient's mother consulted with hematology; no clotting abnormalities were identified.

**Figure 1. Periapical radiograph associated with Case 1. The presence of SSC and normal trabecular pattern of bone is observed.**



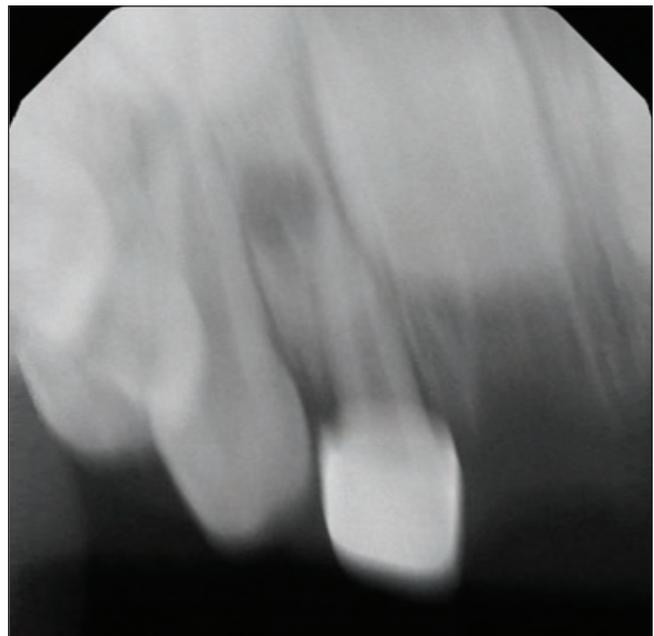
### Case 2

A three-year-old female with a seven-month post-surgical history of uncomplicated T&A presented to the Nemours/AIDHC ED because of persistent gingival bleeding following a maxillary right lateral incisor crown procedure during oral rehabilitation under general anesthesia. The treatment had been completed at an ASC and included extractions, a white-veneered SSC, and restorations. Her dental history was significant for trauma three weeks previously, which the mother reported involved her maxillary anterior teeth, leading to the central incisors requiring extractions and the right lateral incisor a crown. Her family history was not significant for inherited bleeding disorders.

The patient presented hemodynamically stable and well appearing. Intraoral examination revealed bleeding to be localized to the maxillary anterior ridge. Gauze pressure was used to achieve temporary hemostasis to determine the precise area of bleeding. Upper right and left maxillary central incisor extraction sites contained what appeared to be absorbable gelatin sponge. The upper right maxillary lateral incisor, which had been restored with a white-veneered SSC, had persistent bleeding from the sulcus and from a 3-mm laceration on palatal tissue, adjacent to the crown. The tooth had class I mobility, and there were no signs of swelling or infection. Gauze pressure alone was not adequate to achieve hemostasis. A 15.5% ferric sulfate solution was then applied to the area using sterile gauze, along with digital pressure. This medicament only provided for 20-minute increments of hemostasis, after which very small amounts of breakthrough bleeding occurred from the gingival sulcus.

A radiograph was taken (Fig. 2) revealing a periapical radiolucency and a widened periodontal ligament, suggestive of necrosis secondary to the reported previous trauma. There was no sign of non-vital pulp therapy. The clinical impression was one of iatrogenic trauma involvement during tooth preparation, and, with the radiographic findings, this was likely exacerbated by a luxation injury. It was determined that extraction and placement of an absorbable gelatin sponge would be the best course of action. The upper right maxillary lateral incisor was extracted under moderate sedation and local anesthesia. Absorbable sponge was placed in the extraction socket, and hemostasis was achieved almost immediately. No direct pressure or suture was needed. The patient was monitored for an additional 50 minutes postoperatively with continued hemostasis. The dental follow-up the next day revealed a clotted extraction site with continued hemostasis. Mother reported that there was no further bleeding after ED discharge.

**Figure 2. Periapical radiograph associated with Case 2. The presence of periapical radiolucency and widened periodontal ligament space associated with the right lateral incisor and recent extractions of the central incisors are observed.**



## DISCUSSION

In restorative dental procedures, some level of iatrogenic damage is inherently associated even with careful surgical technique.<sup>7</sup> An SSC preparation is likely to result in soft tissue trauma to the immediately adjacent gingivae, even with utmost care. This is generally minimal, resulting in negligible amounts of self-limited bleeding. If, however, bleeding persists, systemic hematological bleeding disorders should be considered. In these cases, bleeding disorders were unlikely because of the lack of bleeding complications after T&A for both patients, the lack of known familial clotting disorders, and the fact that bleeding was limited to one area of the mouth, even though similar procedures occurred throughout the mouth.

In the first case, it appears iatrogenic trauma to the gingivae and alveolus likely occurred but was exacerbated by the presence of dental cement. Dental cement, like other foreign body matter, can cause delayed and continual reorganization of blood coagulum, resulting in liver clots.<sup>8</sup> Liver clots are typically removed easily with suction, curette, or gauze,<sup>8</sup> as was the case here. Because of bruising presentation, the oral bleeding, and lack of paternal history, outpatient coagulation studies were completed; however, no hematological abnormalities were found. The probable diagnosis is delayed post-surgical bleeding due to residual subgingival cement and iatrogenic soft tissue trauma during preparation.

In the second case, the etiology of bleeding was also likely multifactorial. It appeared to be caused by the palatal laceration of the periodontium; however, the tooth was also necrotic, likely secondary to previous luxation. It is plausible that treatment caused an acute exacerbation of a preexisting inflammatory process within the periodontal space of the tooth. Worth noting also was that no pulpal therapy was appreciated. This suggests that preoperative radiographs were likely not taken the day of surgery, or, if taken, the apical region was not captured or evaluated. An important consideration for traumatized teeth is that it can take up to three weeks for pulp necrosis and inflammatory root resorption to become radiographically evident.<sup>9</sup> Preoperative films on the day of treatment should be considered when planning restorative therapy for recently injured teeth. In this case, non-vital pulp therapy or extraction would have been indicated.

Hemostasis was achieved locally in the first case by way of debridement, absorbable mesh, and ferric sulfate. Removal of debris decreases inflammation and promotes clot formation. The absorbable mesh, a sterile regenerated cellulose sheet, can be applied directly to the area of bleeding,<sup>10</sup> in this case, the sulcus. Once in place, it swells into a brownish/black gelatinous mass promoting clot formation.<sup>10</sup> The low pH of the mesh lends to wide-range bactericidal properties<sup>10</sup> but may also cause delayed healing.<sup>4</sup> It is normally absorbed within two weeks.<sup>4</sup> Lastly, ferric sulfate reacts with blood causing agglutination of blood proteins, therefore occluding capillary orifices.<sup>11</sup>

In the second case, hemostasis was achieved by tooth extraction and placement of a hydrocolloid absorbable gelatin sponge. This gelatin sponge is composed of porcine-derived collagen that is whipped into foam and then dried.<sup>10</sup> Absorption of blood causes it to expand,<sup>10</sup> lending to its hemostatic properties. Although it was not needed in our case, it is commonly used in conjunction with a figure-of-eight suture and application of pressure for several minutes.<sup>4</sup> The gelatin sponge has a neutral pH, which allows it to be used as an adjunct with topical thrombin,<sup>10</sup> and, lastly, it is absorbed in four to six weeks.<sup>4,10</sup>

There are additional topical hemostasis agents that can be considered in these cases, including topical thrombin, epinephrine, and tranexamic acid. Although these cases were treated satisfactorily with local measures, differential diagnosis should include systemic considerations. These clinical scenarios are a reminder of the importance of careful diagnosis, procedure precision, and postoperative management to minimize postoperative bleeding complications.

## Financial support and conflicts of interest:

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# Efficacy of a Modified Audio-Tactile Performance Technique with Braille (ATPb) on the Oral Hygiene Status of Visually-Impaired Children

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**Objective:** We assessed the effectiveness of a modified audio-tactile performance (ATP) technique with braille (ATPb) on the oral health statuses of visually-impaired children. **Study design:** Ninety visually-impaired institutionalized children received oral hygiene instructions using audio (AM), ATP or ATPb techniques. Plaque scores were assessed at baseline and after reinforcement and non-reinforcement periods. **Results:** In the totally visually-impaired, mean reductions in plaque scores in the ATPb, ATP and AM groups during the reinforcement period were  $1.119 \pm 0.260$ ,  $0.654 \pm 0.239$  and  $0.237 \pm 0.255$ , respectively ( $p < 0.001$ ), worsening to  $0.107 \pm 0.160$ ,  $0.083 \pm 0.193$  and  $-0.208 \pm 0.267$  during the non-reinforcement period ( $p < 0.001$ ), before culminating at 6 months at  $1.227 \pm 0.261$ ,  $0.737 \pm 0.317$  and  $0.029 \pm 0.108$  ( $p < 0.001$ ). In partially visually-impaired children, reductions during the reinforcement period were  $0.934 \pm 0.279$ ,  $0.762 \pm 0.270$  and  $0.118 \pm 0.237$ , respectively, dropping to  $0.176 \pm 0.166$ ,  $0.083 \pm 0.169$  and  $-0.128 \pm 0.114$  without reinforcement and culminating at  $1.109 \pm 0.258$ ,  $0.845 \pm 0.292$  and  $-0.010 \pm 0.226$  ( $p < 0.001$ ). There were significant inter-group differences during the three periods ( $p < 0.001$ ), except in the ATP and ATPb groups during the non-reinforcement period for totally impaired ( $p = 0.157$ ) and during reinforcement ( $p = 0.155$ ) and non-reinforcement ( $p = 0.051$ ) periods for partially impaired children. **Conclusions:** All three techniques were successful when reinforced periodically. However, only ATP and ATPb were successful during periods without reinforcement. The modified audio-tactile performance technique with braille (ATPb) was most effective, allowing visually-impaired children to retain oral hygiene information without intervention.

**Keywords:** Audio-tactile technique, Braille, Oral health education, Visually impaired children.

## INTRODUCTION

People with disabilities constitute a large percentage of the population and the number of individuals with disabilities is increasing due to significant medical developments in recent years which have enhanced survival.<sup>1</sup> During the early years of life, a child learns to understand and construe their environment with the help of their primary senses, of which vision is the most important. Thus, when sight is impaired or absent in childhood, it can have detrimental effects on their physical, neurological, cognitive and emotional development.

In 2012, the World Health Organization (WHO) estimated that approximately 285 million people were visually impaired, of which 39 million were blind and 246 million had low vision.<sup>2</sup> In India, about 280,000–320,000 children are visually-impaired.<sup>3</sup> As visually-impaired individuals may be able to differentiate images, light and colours or even read large print, the term does not necessarily indicate a complete lack of sight.<sup>4</sup> Nevertheless, the visually-impaired tend to depend on their other senses to adapt themselves to their surroundings, such as sound, speech, and touch.<sup>5</sup>

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Oral health is fundamental to the overall health of an individual. However, visually-impaired children often have poorer oral hygiene compared to their sighted counterparts and usually experience difficulty in maintaining their oral health as they cannot detect and recognise early signs of dental disease.<sup>4,6,7</sup> The absence of visual-motor coordination, insufficient parental supervision and the child's lack of concern for their appearance are other contributory factors.<sup>8</sup> Visually-impaired children deserve the same opportunities for treatment as those who are otherwise healthy. Unfortunately, factors associated with unmet dental care needs among children with special healthcare needs include living in low-income households or rural areas, a failure to receive adequate medical care, increased special needs severity and lower degree of functioning, poor psychological adjustment and caregiver-related factors such as depression and low levels of functioning.<sup>9</sup>

Oral health education is key in the prevention of oral diseases. However, conventional methods of health education may not be effective to motivate visually-impaired children.<sup>10</sup> Various studies have investigated the use of verbal and tactile aids, braille and music-based education programs to educate and train such children on oral hygiene maintenance. Verbal health talks are the most convenient means of delivering such oral health education.

Braille is a tactile writing system introduced by Louis Braille and is traditionally written on embossed paper. It consists of small rectangular blocks called cells that contain tiny raised dots corresponding to letters of the alphabet. Audio-tactile performance (ATP) is an oral health education technique for the blind that was introduced by Hebbal *et al* in 2011. In this method, children are taught techniques for brushing their teeth using both audio and tactile aids.<sup>11</sup> The aim of the current study was to modify the ATP technique by incorporating braille and to compare its effectiveness with conventional audio methods (AM) and ATP techniques.

### MATERIAL AND METHOD

Ninety institutionalized children between 6–15 years old with either total or partial visual impairment were selected from three schools in two adjoining southern states in India. Ethical clearance from our institution's ethics board and permission from the school authorities was obtained prior to the study. As these children were institutionalised, informed consent for their inclusion in the study was obtained from their parents by the school authorities. The ability to read braille was deemed an essential criterion for inclusion. Subjects were excluded from the study if they had any underlying systemic illnesses or other handicapping conditions, were on any long-term medications or had rampant caries. They were also not included if the child was uncooperative or if they had attended camps or received oral health education in the preceding six months prior to the study.

A random selection of 30 children from each school were allocated to one of three groups, with each group receiving a different form of dental health education. The first group received AM, the second received ATP and the third received a modified ATP technique with braille (ATPb). The study comprised four phases, consisting of interaction and collection of baseline data (phase I), oral health education (phase II), a reinforcement period (phase III) and a non-reinforcement period (phase IV). The efficacy of the education techniques was assessed separately among children with total and partial impairment.

### Phase I: Interaction and Collection of Baseline Data

An interactive session was carried out with the visually-impaired children to build a good rapport and to assess their level of cooperation. These sessions also aimed to assess the children's baseline knowledge and attitudes towards maintaining adequate oral hygiene and their capacity for self-maintenance. As this was their first time interacting with the dentist for most children, a relaxed and friendly environment was maintained.

The personal details (name, age and gender), degree of visual impairment (total or partial) and existing oral hygiene maintenance habits (material, frequency and method of tooth brushing) of each child were recorded in a specially-designed data sheet. All questions were asked individually to each child and confidentiality was maintained. Subsequently, a clinical examination was performed within school premises in natural daylight with a sterile mouth mirror and an explorer while the child sat on a simple chair. Mouth masks and gloves were not utilized as the children were noted to be generally apprehensive towards these accoutrements during the preliminary sessions. In order to ensure familiarity and cooperation, each instrument and its function was explained to the child and they were allowed to handle the instruments prior to the start of the examination. Baseline plaque scores were subsequently recorded using the simplified Silness-Löe Plaque Index.<sup>12</sup>

### Phase II: Oral Health Education

Three different techniques for oral health education were utilized for children in each school.

#### *Group I: Audio Method (AM)*

Thirty children from a school in Kerala State were included in the first group and received oral health education and instructions on brushing techniques via audio means alone. A health talk was delivered explaining the importance of teeth and the need to maintain adequate oral hygiene. Various methods of oral hygiene maintenance such as brushing and flossing of the teeth were explained to the children. The modified Bass technique was described and the children were made to repeat it verbally until they were deemed to have comprehended the method well.

#### *Group II: Audio-Tactile Performance (ATP) Technique*

Thirty children from another school in the same state were included in the second group and received oral health education via a conventional ATP technique. The children were first verbally taught the importance of oral hygiene and brushing techniques, before later being made to feel large models of a tooth set using their fingers. Brushing techniques were demonstrated using these models. A zinc oxide eugenol cement was used to simulate calculi in the cervical areas of the posterior teeth and the lingual areas of the anterior teeth. This was done with the intention of helping the children identify such areas in their own mouths. The modified Bass technique was taught by guiding the child's hands over the models and performing the necessary strokes with accompanying verbal explanations. The children were then requested to try the technique themselves. They then repeated the brushing technique several times without a time frame until they had mastered it.

### Group III: Modified Audio-Tactile Performance Technique with Braille (ATPb)

Thirty children from a school in the adjoining state of Karnataka were included in the third group and received oral health education using a modified ATP technique which incorporated instructions in braille. The conventional ATP technique as taught in Group II was followed; however, the children also received a braille pamphlet detailing a custom-designed story explaining the need for oral hygiene maintenance along with instructions for the modified Bass brushing technique. The story starts with an entry into “Tooth city” where they meet familiar cartoon characters. They then take them around showing the plaque accumulations, cavities, gingival status and explain in common terms about oral hygiene. The conversation between these characters dwells further into the bad and good tooth brushing techniques. The story was first explained to the children verbally and was then read aloud, from braille pamphlet, first by each child individually and then later in a group setting. The children were also instructed to read the pamphlet at least once a week during class.

### Phase III: Reinforcement Period

Following the initial education sessions, the children underwent a three-month reinforcement period, during which monthly visits were made to each school and the brushing technique was reinforced using the respective oral health education methods described above. This was done with the intention of further acquainting the children with the brushing technique so that they could put it into practice in daily life. The teachers were also educated and trained during this time, so that they could supervise the children during the brushing process. Plaque scores using the simplified Silness-Löe Plaque Index were re-assessed at the end of this period.

### Phase IV: Non-Reinforcement Period

The children then underwent a non-reinforcement period over the final three months of the study period. During this time, no oral health education was given. This period was intended to assess which of the three oral health education methods was most effective for good oral hygiene maintenance and was most likely to be retained without intervention. Plaque scores using the simplified Silness-Löe Plaque Index were once again re-assessed after three months of non-reinforcement.

### Statistical Analysis

Collected data were summarised using descriptive statistics such as frequencies, percentages, means and standard deviations. In order to compare outcome measures between the totally and partially visually-impaired children, an independent sample t-test was used. In addition, pairwise comparisons of consecutive measurements within groups were analysed using a paired t-test. Kruskal-Wallis and Man-Whitney U tests were performed for data which did not follow a normal distribution. Data management and analysis was performed using Microsoft Excel and SPSS Version 20.0 (IBM Inc., Chicago, IL, USA).

## RESULTS

A total of ninety visually-impaired children received oral hygiene instructions using three different oral health education techniques. Each group included 30 children from one of three schools. The mean age was  $10.23 \pm 2.41$  years and there were 48% males and 52% females. Overall, 40 children (44.4%) had total visual impairment, while 50 (55.6%) had partial visual impairment. Thirteen (43.3%) of the children in Group I receiving AM oral health education, 13 (43.3%) in Group II receiving ATP oral health education and 14 (46.7%) in Group III receiving modified ATPb oral health education had total blindness (Table 1).

The oral hygiene status of the children was poor, with a mean baseline plaque score of  $2.031 \pm 0.411$ . Although the mean plaque scores of the totally visually-impaired children ( $2.104 \pm 0.390$ ) was greater than that of the partially visually-impaired children ( $1.972 \pm 0.422$ ), this difference was not statistically significant ( $p = 0.13$ ). The demographic characteristics and baseline plaque scores of the different groups are shown in Table 1. In Groups I and III, the partially visually-impaired children had better oral hygiene statuses; in contrast, in Group II, the totally visually-impaired fared better. However, there were no statistically significant differences in mean plaque scores based on degree of blindness within any of the three groups (Figure 1).

The effectiveness of each oral health education technique was compared separately among the totally and partially visually-impaired children to assess the most effective means of delivering oral hygiene instructions to such children.

### Children with Total Visual Impairment

Among the totally visually-impaired children, the maximum reduction in plaque scores during the reinforcement period was seen in Group III receiving ATPb education, with a mean difference of  $1.119 \pm 0.260$ , followed by Groups II and I receiving ATP and AM education, with mean differences of  $0.654 \pm 0.239$  and  $0.237 \pm 0.255$ , respectively (Fig. 2). There were significant differences in effectiveness during this period within each group ( $p < 0.001$ ) (Table 2). When effectiveness between groups was compared, we found significant differences between Groups I and II, Groups I and III and Groups II and III ( $p < 0.001$ ) (Table 3).

During the three-month period where there were no attempts at reinforcement, the maximum reduction in scores was seen in Group III, followed by Group II, with mean differences of  $0.107 \pm 0.160$  and  $0.083 \pm 0.193$ , respectively. However, in Group I, scores worsened over time, with a mean difference of  $-0.208 \pm 0.267$  (Figure 2). There was significant difference in effectiveness within groups during this period ( $p < 0.001$ ) (Table 2). In terms of effectiveness between individual groups, significant differences were noted only between Groups I and II and Groups I and III ( $p < 0.001$ ), with no difference in effectiveness between Groups II and III ( $p = 0.157$ ) (Table 3).

At the end of the six-month study period (i.e. after both periods of reinforcement and non-reinforcement), the maximum reduction in scores was seen in Group III, followed by Groups II and I. Mean differences during this time were  $1.227 \pm 0.261$ ,  $0.737 \pm 0.317$  and  $0.029 \pm 0.108$ , respectively (Figure 2). There were significant differences in effectiveness within the groups ( $p < 0.001$ ) (Table 2). We also noted significant differences in effectiveness between Groups I and II, Groups I and III and Groups II and III (Table 3).

Table 1. Table showing the demographics and characteristics of the study group

Number of children		30	30	30	
Blindness	Total n (%)	13 (43.3)	13 (43.3)	14 (46.7)	
	Partial n (%)	17 (56.7)	17 (56.7)	16 (53.3)	
Gender	Male n (%)	Total	7 (53.8)	3 (23.1)	10 (71.45)
		Partial	6 (35.3)	8 (47.1)	9 (56.3)
	Female n (%)	Total	6 (46.2)	10 (76.9)	4 (28.6)
		Partial	11 (64.7)	9 (52.9)	7(43.7)
Age	Total Mean ± SD	9.69±2.63	12.08±2.50	9.57±2.31	
	Partial Mean ± SD	10.53±2.21	9.47±2.10	10.25±2.27	
Plaque score	Total Mean ± SD	1.839±0.226	2.092±0.272	2.362±0.195	
	Partial Mean ± SD	1.541±0.348	2.120±0.259	2.260±0.225	
		<b>Group I AM</b>	<b>Group II ATP</b>	<b>Group III ATPb</b>	

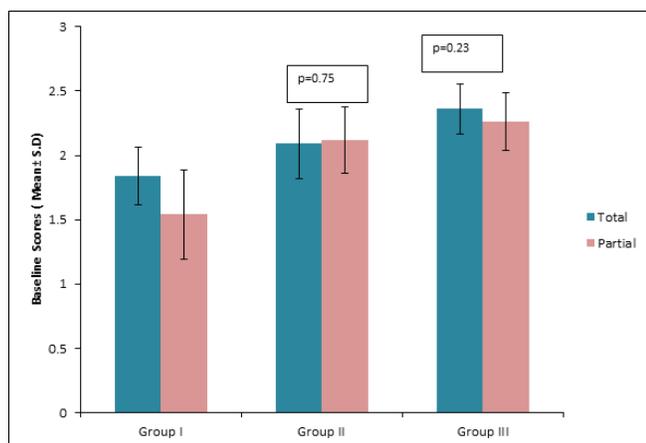
Table 2. Table showing the effectiveness of the three techniques on the oral hygiene status of the totally and partially visually impaired children during the study period.

Blindness	Groups	Baseline–Reinforced 0-3 months		Reinforced – Non-reinforced 3-6 months		Baseline- Non- reinforced 0-6 months	
		Mean difference	p value	Mean difference	p value	Mean difference	p value
Total	Group I (AM)	0.237±0.255	<0.001	-0.208±0.267	<0.001	0.029±0.108	<0.001
	Group II (ATP)	0.654±0.239		0.083±0.193		0.737±0.317	
	Group III (ATPb)	1.119±0.260		0.107±0.160		1.227±0.261	
Partial	Group I (AM)	0.118±0.237	<0.001	-0.128±0.114	<0.001	-0.010±0.226	<0.001
	Group II (ATP)	0.762±0.270		0.083±0.169		0.845±0.292	
	Group III (ATPb)	0.934±0.279		0.176±0.166		1.109±0.258	

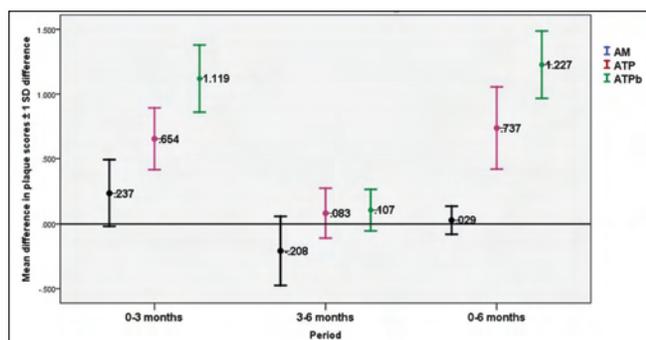
Table 3: Table showing the group wise comparison of effectiveness of the three techniques based on the degree of blindness

Blindness	Groups	Baseline–Reinforced 0-3 months		Reinforced – Non-reinforced 3-6 months		Baseline- Non- reinforced 0-6 months	
		Mean difference	p value	Mean difference	p value	Mean difference	p value
Total	Group I and II (AM and ATP)	0.417	<0.001	0.291	0.000	0.708	<0.001
	Group I and III (AM and ATPb)	0.882	<0.001	0.315	0.001	1.198	<0.001
	Group II and III (ATP and ATPb)	0.466	<0.001	0.023	0.157	0.489	<0.001
Partial	Group I and II (AM and ATP)	0.644	<0.001	0.21	<0.001	0.855	<0.001
	Group I and III (AM and ATPb)	0.816	<0.001	0.303	<0.001	1.119	<0.001
	Group II and III (ATP and ATPb)	0.172	0.155	0.092	0.051	0.264	0.015

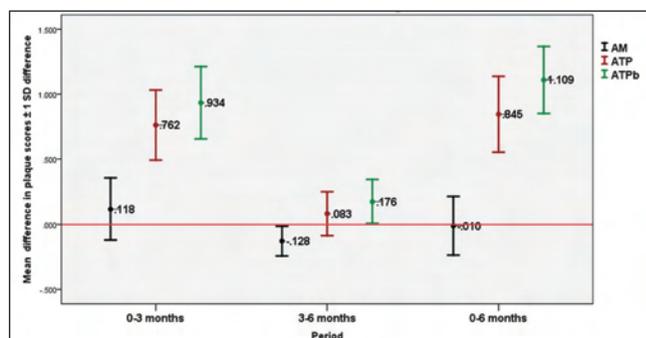
**Figure 1. Comparison of the mean baseline plaque scores of the totally and partially visually impaired children within the groups.**



**Figure 2. Comparison of the effectiveness of the three oral health education techniques on the oral health status of totally visually impaired children.**



**Figure 3. Comparison of the effectiveness of the three oral health education techniques on the oral hygiene status of the partially visually impaired children.**



### Children with Partial Visual Impairment

During the reinforcement period, the partially visually-impaired children in Group III showed the greatest reduction in scores, with a mean difference of  $0.934 \pm 0.279$ . This was followed by Groups II and I, with mean differences of  $0.762 \pm 0.270$  and  $0.118 \pm 0.237$ , respectively (Figure 3). There were significant differences in effectiveness within groups during this period ( $p < 0.001$ ) (Table 2). When effectiveness between groups was compared, there were significant differences between Groups I and II and Groups I and III ( $p < 0.001$ ), but not between Groups II and III ( $p = 0.155$ ) (Table 3).

During the subsequent three-month non-reinforcement period during which no intervention was performed, the greatest reduction in scores was seen in Group III, followed by Group II, with mean differences of  $0.176 \pm 0.166$  and  $0.083 \pm 0.169$ , respectively. In Group I, an increase in plaque scores was seen during this period, with a mean difference of  $-0.128 \pm 0.114$  (Fig. 3). There were significant differences in effectiveness within groups during this period ( $p < 0.001$ ) (Table 2). In terms of between-group differences, significant differences were noted only between Groups I and II and Groups I and III ( $p < 0.001$ ), with no difference in effectiveness between Groups II and III ( $p = 0.051$ ) (Table 3).

After six months, the greatest reduction in scores was seen in Group III, with a mean difference of  $1.109 \pm 0.258$ , followed by Group II, with  $0.845 \pm 0.292$ . However, partially visually-impaired children in Group I showed an overall worsening of scores, with a mean difference of  $-0.010 \pm 0.226$  (Fig. 3). There was significant difference in effectiveness within groups ( $p < 0.001$ ) (Table 2). Overall, there were significant differences in effectiveness between Groups I and II, Groups I and III and Groups II and III by the end of the six-month study period (Table 3).

### DISCUSSION

The lack of one of the primary senses—in this case, vision—which orients an otherwise healthy individual to their surroundings may be the main reason for poor oral hygiene in the visually impaired. An effective preventive program that focuses on educating and training the child to perform adequate oral hygiene maintenance technique is necessary as such individuals cannot visualise signs of dental disease.<sup>13</sup> We found that adding braille instructions to conventional audio-tactile methods not only resulted in a meaningful reduction in plaque scores, but also continued to improve the scores significantly, even without any additional intervention.

Institutionalised children were chosen in order to avoid any bias in outcome, as all children had similar dietary and oral hygiene practices. Only children aged 6–15 years old were included, as such children would have sufficient manual dexterity to brush their teeth independently and understand instructions clearly.<sup>14,15</sup> Moreover, schools are considered the best environment to teach preventive oral health practices as schoolchildren are already in a learning environment and are easily accessible.<sup>16</sup> The modified Bass technique is superior to the horizontal scrub technique as the former cleanses the proximal areas as well as the gingival third and sulcular regions of the oral cavity.<sup>17,18</sup> However, several studies have reported no significant difference in efficacy between these methods.<sup>19,20</sup> The modified Bass technique, even though time-consuming, was chosen for the current study as it has a wider cleansing zone.

Overall, the oral health status of the visually-impaired children was poor, with a mean baseline plaque score of  $2.031 \pm 0.411$ .

Previous research has confirmed that visually-impaired often have poor oral hygiene.<sup>8,21-25</sup> However, Prashanth et al. reported that only 8% of the blind children in their study population had poor oral hygiene.<sup>26</sup> Reasons for poor oral hygiene among visually-impaired children could be the lack of development of necessary self-maintenance skills, inability to see and remove plaque, lack of supervision while brushing, limited intellectual capacity in some children, lack of knowledge regarding oral hygiene and effective tooth brushing and the reduced amount of time spent brushing their teeth.<sup>27-29</sup> Local factors like malocclusion and dental crowding may also lead to the accumulation of plaque, contributing to reduced oral health. However, Mitsea et al. observed better oral hygiene among visually-impaired children in comparison to children with cerebral palsy and mental retardation.<sup>30</sup> Institutionalised blind children may have poorer oral hygiene compared to those who are non-institutionalised, possibly due to a lack of caregiver assistance while brushing and during other oral hygiene practices.<sup>31</sup> Lack of supervision and help from parents may affect factors like brushing technique and motor skills.<sup>25</sup> However, this may not always be true and depends to a large extent on the infrastructure of the institution in question and the enforcement of routine hygiene practices.<sup>32</sup>

There was no significant difference in oral hygiene status of the totally and partially visually-impaired children as a whole and within groups at baseline. In contrast, Bennadi et al. reported an increase in plaque levels among those who were totally blind.<sup>33</sup> This difference in findings in the present study is probably due to the fact that many were classified medically with partial impairment because of an enhanced sensitivity to light rather than an actual improvement in vision.

Conventional methods of oral health education are directed towards visualising and identifying early signs of dental disease. Naturally, this is near impossible for visually-impaired individuals.<sup>26</sup> Hence, customised methods of education and motivation to correct oral hygiene practices are essential to maintain good oral hygiene. The visually-impaired depend on various other sensations such as sound, speech and touch along with kinaesthetic and olfactory sensations to adapt themselves to their immediate surroundings.<sup>34</sup>

In the present study, three different approaches of providing oral hygiene instructions were assessed. In each group, the respective method was reinforced for a period of three months, following which there was no intervention for another three months (the non-reinforcement period). Among the children who received oral health education via a verbal health talk, a reduction in plaque scores was seen during the reinforcement period. However, by the end of the study period, the final scores had worsened in comparison to baseline scores. This suggests that even though this technique was capable of marginally improving oral hygiene status, the children could neither improve nor maintain their oral hygiene without intervention.

In contrast, children who took part in the ATP intervention had significantly reduced plaque scores compared to those receiving AM. Although there was only a marginal improvement seen during the non-reinforcement period, there was a significant improvement in mean plaque scores from baseline by the end of the study. This clearly shows that this technique was very effective if reinforced. In the absence of the intervention, the children could maintain their oral hygiene, but could not improve further. Earlier studies have similarly shown significant improvement in oral hygiene using ATP techniques.<sup>4,11</sup> However, reinforcement in these studies was

primarily given by teachers or caregivers and there was no well-demarcated period of reinforcement and non-reinforcement.

Finally, the modified ATP technique with braille resulted in a significant reduction in plaque scores by the end of the reinforcement period. Furthermore, a meaningful improvement in plaque scores was observed even without intervention during the non-reinforcement period. This improvement was greater than that observed in the ATP group during this time, although the difference was not statistically significant. By the end of the study, a significant drop in plaque scores was seen in comparison to baseline scores; moreover, this improvement was greater than that seen during the reinforcement period. Thus, the newly developed technique was very effective in improving and maintaining plaque scores, both with and without reinforcement. The children were able to retain the instructions and incorporate them into their daily practice, thereby considerably improving their oral hygiene.

When the effectiveness of the three techniques was compared, it was observed that the ATPb technique resulted in the greatest reduction in plaque scores during both the reinforcement and non-reinforcement periods. This was probably due to the fact that it was the most customised technique of the three. In addition, the inclusion of braille may have helped to convert the tactile sensations into learned information. Moreover, as the braille pamphlet included a story, it may have been more appealing to the children and therefore better remembered. Possibly, the efficacy of the next best technique, ATP, can be further improved with repeated instructions over longer periods of reinforcement.

Chowdary *et al* showed that combining verbal, braille and tactile oral hygiene instructions most effectively reduced plaque scores compared to verbal and braille or verbal and tactile means.<sup>35</sup> Surprisingly, a study from Hyderabad of four different oral health education techniques found the highest improvement in scores in the audio group, with the least reduction in those who received instructions via braille alone. In this study, the braille group did not receive individualised attention, which may explain their poor performance.<sup>36</sup> Another study found that tactile and auditory means were equally effective in educating and motivating visually-impaired children and that these methods can be made interesting by utilising braille, plastic models, an audio story or the computer software Job Access With Speech (JAWS).<sup>37</sup>

In the current study, the improvement in plaque scores with the modified ATPb intervention was similar in both totally and partially visually-impaired children. However, in a study done by Joybell et al., it was observed that partially blind children mastered brushing techniques better than totally impaired children following oral health education using the ATP technique.<sup>4</sup> Another study found a greater decrease in plaque control record values among partially visually-impaired children following oral health education, with a definite advantage over completely blind children.<sup>34</sup>

In general, the present study clearly indicates that the oral health practices of visually-impaired children can be improved by deviating from traditional approaches of oral health education. We developed a modified ATP technique including braille in order to ensure that visually-impaired children can be taught to perform basic oral hygiene practices independently. Nevertheless, as the study population comprised children from three institutions in three different cities, their dietary and oral hygiene practices could only be standardised to a certain extent; this could be considered a

limitation of our study. We recommend that techniques to educate and train visually-impaired children be customised on an individual basis in accordance with their needs. Though most of the techniques follow the principles of 'tell-feel-do', no standard technique has been adopted so far. ATPb is a step in that direction to formulate a technique that can be used by everyone with ease to create positive impacts on behavioural outcomes. More studies exploring this approach will definitely reveal more interventions, simple and feasible. Newer technologies should be combined with conventional methods for maximum benefit.

### CONCLUSION

The present study aimed to develop a new oral health education program for visually-impaired children to ensure adequate oral hygiene maintenance. While all three methods of oral health education—AM, conventional ATP and modified ATPb techniques—were successful in improving oral hygiene status with reinforcement, only the last two improved oral hygiene even during periods of non-reinforcement. However, the newly developed modified technique using braille, ATPb, was the most effective method with the advantage of encouraging retention of the learned information even without intervention.

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# Success of Biodentine and Ferric Sulfate as Pulpotomy Materials in Primary Molars: A Retrospective Study

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**Objective:** To determine the clinical and radiographic success of Biodentine® (BD) and Ferric Sulfate (FS) as primary molar pulpotomy materials and to compare their outcomes. **Study design:** Retrospective data was obtained from the electronic health records (EHR) of a university-based pediatric dental clinic. Participants were enrolled according to specified inclusion and exclusion criteria. Two trained and calibrated examiners evaluated the EHR using validated criteria for clinical and radiographic outcomes. Study data was numerically coded and analyzed. Cohen's Kappa and Chi-square tests were used ( $p < 0.05$ ). **Results:** Eighty-three participants (62.7% females, age range two to eight years, average age of 4.5 years) with 102 pulpotomies were enrolled. FS was used in 78% ( $n=79$ ) and BD in 22% ( $n=23$ ) of the cases. Follow-up periods ranged from six to 36 months (mean of 17 months). BD showed 100% clinical and radiographic success, while FS demonstrated 84% clinical and 70% radiographic success. The two groups were compared at one year with no statistically significant differences. At 18 months, BD outperformed FS clinically ( $p=.012$ ) and radiographically ( $p=.001$ ). Intra-rater and inter-rater agreement were  $\kappa > 0.88$ . **Conclusions:** Both materials can be recommended for clinical practice, however BD may be the preferred choice for its better outcomes at 18 months.

**Keywords:** Biodentine, Ferric Sulfate, Pulpotomy, Primary Molar, Pediatric Dentistry

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## INTRODUCTION

Primary molar pulpotomy is a type of vital pulp therapy indicated for treatment of teeth diagnosed with reversible pulpitis.<sup>1</sup> The procedure entails amputation of the inflamed coronal pulp and application of a suitable therapeutic material over the remaining radicular pulp stump, which can promote disease-free survival of the deciduous tooth.<sup>1,2</sup> While the immediate outcome of successful pulpotomy is an asymptomatic tooth with preserved radicular pulp, its ultimate objective is to prevent premature tooth loss and to maintain integrity of the dental arch.<sup>2,3</sup> The clinical technique and indications for the pulpotomy procedure have remained unchanged for decades, however the utilized therapeutic materials have been widely diverse and are continuously evolving over the years.<sup>4</sup> The existing pulpotomy materials can be classified into three main categories based on their effect on the remaining radicular pulp tissue.<sup>5</sup> These include agents producing devitalization (e.g., formocresol, gluteraldehyde), preservation in a healthy state (e.g., ferric sulfate, bioactive cements, calcium hydroxide, sodium hypochlorite, lasers, etc.) or promoting regeneration (e.g., bone morphogenetic proteins).<sup>5,6</sup>

For many years since the 1930s, the Buckley's solution (19% formaldehyde and 35% cresol diluted with glycerin and distilled water) has been the benchmark pulpotomy medicament.<sup>2</sup> Formocresol (FC) has protein-binding properties and works by fixation of

the radicular pulp tissue, making it inert.<sup>2,6</sup> Its overall success has been estimated around 87%.<sup>2</sup> However, in 2004 the International Agency for Research on Cancer classified formaldehyde as carcinogenic to humans.<sup>7</sup> Such toxicity and mutagenicity concerns have led to the discouragement of FC wider use and prompted new research looking for viable replacement options.<sup>2</sup> Ferric Sulfate (FS) in a 15.5% solution is a hemostatic agent and has been proposed as one such alternative.<sup>2,4</sup> Animal and human studies have reported that its success as pulpotomy medicament (79.3%–93%) is similar to that of FC.<sup>6,8,9</sup> FS forms a ferric ion-protein complex when it comes to contact with blood which upon agglutination produces plugs that mechanically occlude the cut blood vessels to achieve coagulation and hemostasis.<sup>10</sup> This reaction causes minimal damage to the radicular pulp tissue and helps preserve its vitality.<sup>10</sup> While FS has gained popularity over the past decade as the pulpotomy agent of choice in pediatric dentistry, more recently bioceramic materials such as Mineral Trioxide Aggregate (MTA) and Biodentine® (BD) have come to the scientific spotlight as successful endodontic therapy materials.<sup>2,11</sup>

BD is a novel calcium-silicate based cement, first introduced on the market in 2010 by Septodont (St Maur des Faussés, France).<sup>11</sup> It has gained attention as a contemporary dentine replacement and repair material and has been proposed for use in therapeutic pulpotomies in primary teeth due to its excellent salability and biocompatibility.<sup>11,12</sup> BD is a commercialized tricalcium silicate and thus undergoes an extensive manufacturing process utilizing active biosilicate technology, which eliminates aluminates and other metal impurities.<sup>11</sup> Therefore, the manufacturer claims that BD has improved mechanical properties when compared to MTA products including faster setting time, increased compressive strength, increased density, decreased porosity, induction of reparative dentine synthesis, and more manageable handling characteristics.<sup>11,12</sup> When used as a pulpotomy agent, BD creates a layer of superficial necrosis on contact with vital pulp tissue.<sup>12</sup> This stimulates a healthy inflammatory response of the radicular pulp and the formation of reparative dentin layer that serves as a hard tissue barrier sealing off and protecting the vital radicular pulp.<sup>11,12</sup> As a primary molar pulpotomy agent, BD has shown promising initial clinical results with reported 92% to 100% success rates.<sup>12-14</sup> However, the research investigating the outcomes of BD remains limited and further trials are needed for clinical practice recommendations.

The purpose of this study was to determine the clinical and radiographic success of primary molar pulpotomies completed with BD and FS at a university-based pediatric dental clinic and to compare the outcomes of the two materials.

## MATERIALS AND METHOD

The Institutional Review Board of the University of Illinois at Chicago (UIC), Chicago, Ill., USA granted permission to conduct this retrospective cohort study. To estimate the required sample size, a prospective power calculation, based on reported clinical success rates for BD and FS from previous studies, showed that 60 teeth would achieve a power of 80%. To account for inconsistent patient attendance at recall, our study included for initial review the first consecutive 400 pulpotomies completed since April 1, 2016. The list was generated from the dental electronic health record (EHR) system of the College of Dentistry, UIC and was based on

completed insurance codes for therapeutic pulpotomy. The specific date was chosen because at that time both materials were used in the university-based pediatric dental clinics. FS was available as Astringent™ (Ultradent Products, Inc., 3935, 505 W 10200 S, South Jordan, UT 84095, USA) and BD is a product of Septodont (94100 St Maur des Faussés, France). The principal investigator (PI), who was a post-graduate student in Pediatric Dentistry (PD), accessed all patient records and evaluated the information against the inclusion and exclusion criteria of the study (Table 1). Patients, who had teeth with completed pulpotomies that met the inclusion criteria, were enrolled as participants. Each of these teeth received a specific study number. All pulpotomies were completed by post-graduate PD students. The procedures were supervised by faculty members, specialists in PD. The EHR notes and radiographs corresponding to the dates of completion of the pulpotomy procedures of the study teeth, as well as all consecutive notes and radiographs from the periodic oral examinations were reviewed by the PI. Typically, patients with completed comprehensive exam and/or dental care are recalled at the clinics every six months for regular periodic dental examinations. Two sets of clinical and radiographic criteria validated by previous research<sup>14</sup> were used in this study to evaluate the information about the study teeth gathered from the EHR (Tables 2 and 3).

The participants' demographic information was collected, such as age at the time of pulpotomy, sex, ethnicity and medical background. Race was not included due to lack of consistent documentation. For each study tooth, the PI searched the EHR for information on the primary molar diagnosis (i.e., caries, trauma, developmental defect) that provided justification for carrying out the pulpotomy procedure. The type of behavior management modality (i.e., nitrous oxide sedation, oral sedation or general anesthesia) used at the time of pulpotomy procedure as well as the type of dental visit (i.e., routine or urgent care) were also recorded. A second examiner, faculty member and specialist in PD, evaluated the radiographs. This was done first independently and then the cases were discussed between the two examiners in order to resolve any disagreements. The PI and the second examiner were calibrated using a questionnaire with 20 randomly selected radiographs of teeth with pulpotomies (for inter-rater reliability). The questionnaire was created by an independent to the study person. The PI completed the questionnaire on two separate occasions with two weeks apart (for intra-rater reliability). The examiners were trained for the study purposes by reviewing pertinent literature discussing therapeutic materials and primary molar pulpotomies. Research data was numerically coded. The clinical and radiographic scores were further converted into dichotomous outcomes combining scores of 1 and 2 into category "Success" and scores 3 and 4 into category "Failure". The inter- and intra-examiner reliability were assessed using Cohen's Kappa, and statistical analysis was completed using Statistical Product and Service Solutions (SPSS) software (Version 25.0, IBM SPSS Statistics, Armonk, N.Y., USA). A chi-square was utilized to assess for difference in outcomes between FS and BD groups with a level of significance of  $p < 0.05$ .

**Table 1. Inclusion and Exclusion Criteria of the Study**

Inclusion Criteria	Exclusion Criteria
Deciduous molar	Tooth other than a deciduous molar
Radiograph with clear visualization of the furcation pre-operatively	No pre-operative radiograph or a pre-operative radiograph without clear visualization of the furcation
Radiograph with clear visualization of the furcation at least 6-months post-operatively	No post-operative radiograph or a post-operative radiograph without clear visualization of the furcation
Clinical evaluation at least 6-months post-operatively	No clinical evaluation or clinical evaluation only less than 6-months post-operatively
Adequate documentation in the patient record on the date of the procedure and at all follow-up examinations	Poor/Improper documentation on the date of the procedure or at any recall examinations
Pulpotomy completed with Ferric Sulfate (FS) or Biodentine (BD)	Pulpotomy completed with a medicament other than FS or BD or medicament not documented in the chart

**Table 2. Criteria for Clinical Success (adapted from Rajasekharan et al.<sup>14</sup>)**

Score	Clinical Criteria	Description
1	Asymptomatic	<input type="checkbox"/> Pathology: Absent <input type="checkbox"/> Normal functioning <input type="checkbox"/> Naturally exfoliated <input type="checkbox"/> Exfoliation prematurely due to ectopic eruption <input type="checkbox"/> Mobility (physiological) ≤1 mm
2	Slight discomfort	<input type="checkbox"/> Pathology: Questionable <input type="checkbox"/> Percussion sensitivity <input type="checkbox"/> Chewing sensitivity, short-lasting <input type="checkbox"/> Gingival inflammation (due to poor oral hygiene) <input type="checkbox"/> Mobility (physiological) >1mm but <2mm
3	Minor discomfort	<input type="checkbox"/> Pathology: initial changes present <input type="checkbox"/> Chewing sensitivity, long lasting <input type="checkbox"/> Gingival swelling (not due to poor oral hygiene) <input type="checkbox"/> Periodontal pocket formation (no exudate) <input type="checkbox"/> Mobility >2mm but <3mm
4	Major discomfort	<input type="checkbox"/> Pathology: Late changes present <input type="checkbox"/> Spontaneous pain <input type="checkbox"/> Gingival swelling (not due to poor oral hygiene) <input type="checkbox"/> Periodontal pocket formation (exudate) <input type="checkbox"/> Sinus tract present <input type="checkbox"/> Mobility ≥ 3mm <input type="checkbox"/> Premature tooth loss, due to pathology

**Table 3. Criteria for Radiographic Success (adapted from Rajasekharan et al.<sup>14</sup>)**

Score	Radiographic Criteria	Description
1	No changes present	<input type="checkbox"/> Internal root canal form tapering from chamber to the apex <input type="checkbox"/> Periodontal ligament (PDL)/periapical regions: normal width and trabeculation
2	Pathological changes of questionable clinical significance	<input type="checkbox"/> External changes are not allowed (widened PDL) <input type="checkbox"/> Abnormal inter-radicular trabeculation or variation on radiodensity <input type="checkbox"/> Internal resorption acceptable (nonperforated) <input type="checkbox"/> Calcific metamorphosis is acceptable and defined as: uniformly thin root canal; shape (non-tapering); variation in radiodensity from canal to canal (one cloudier than the other) <input type="checkbox"/> Dentin bridge formation (one or more canals)
3	Pathological changes present	<input type="checkbox"/> External changes are present, but not large <input type="checkbox"/> Mildly widened PDL <input type="checkbox"/> Minor inter-radicular radiolucency with trabeculation still present <input type="checkbox"/> Minor external root resorption <input type="checkbox"/> Internal resorption changes are acceptable, but not if external change is also present (perforated form)
4	Pathological changes present requiring an immediate extraction of the tooth	<input type="checkbox"/> Frank osseous radiolucency present, endangering permanent successor

**RESULTS**

After reviewing the 400 EHR, 102 deciduous molars in 83 pediatric patients satisfied the selection criteria and were included in the study sample. The flow diagram of the study process is illustrated in Figure 1.

**Demographic Results**

The study sample consisted of 62.7% (n=52) female and 37.3% (n= 31) male participants. The mean age of the participants was 4.5 years and the median was four years, while the age range was two to eight years. The sample included 1.2% (n=1) two years old, 25.3% (n=21) three years old, 27.7% (n=23) four years old, 21.7% (n=18) five years old, 19.3% (n=16) six years old, 1.2% (n=1) seven years old, and three 3.6% (n=3) eight years old participants. Ninety four percent of the participants were within the age group of 3 to 6 years. There was a higher prevalence of Hispanics (53%, n=44), which mirrored the population seen in our PD clinics. Ninety-five percent (n=78) of the participants were healthy and the remaining 4.8% (n=4) had well-controlled asthma.

**Descriptive Results of the Study Teeth**

Almost 80% of the participants had a single pulpotomy (n=66), while 22.5% (n=17) had two. With respect to the diagnostic justification for the pulpotomy procedures, all of the study teeth were described as having deep caries approaching the pulp and with pulpal exposure occurring during caries excavation. Seventy-eight percent of the teeth (n=79) were treated with FS and 22% (n=23) with BD. Specific considerations for using FS or BD by the clinicians were not provided in the EHR notes. All study teeth were restored with prefabricated stainless-steel crowns immediately after the pulpotomy completion. Eighteen percent (n=18) of the pulpotomies were performed on maxillary first primary molars, 16% (n=17) on maxillary second primary molars, 27% (n=28) on mandibular first primary molars, and 39% (n=39) on mandibular second primary molars.

**Descriptive Results on Behavior Management Modality**

With regard to patient management modality at the time of pulpotomy completion, 41 % (n=34) of the participants (with 41.2%, n=42 study teeth) received comprehensive care under general anesthesia (GA), 12% (n=10) of the participants (with 13.7%, n=14 study teeth) were managed with oral conscious sedation and 47% (n=39) participants (with 45.1%, n=46 study teeth) had treatment carried out under nitrous oxide inhalation sedation. Out of the 23 BD pulpotomies, 30.4% (n=7) were completed under GA while 69.6% (n=16) were done with nitrous oxide inhalation sedation. In the FS group, 44.3% (n=35) of the pulpotomies were completed under GA, 17.7% (n=14) under oral conscious sedation and 38% (n=30) with nitrous oxide inhalation sedation. All pulpotomies were completed during routine dental care and none were done as urgent care procedures.

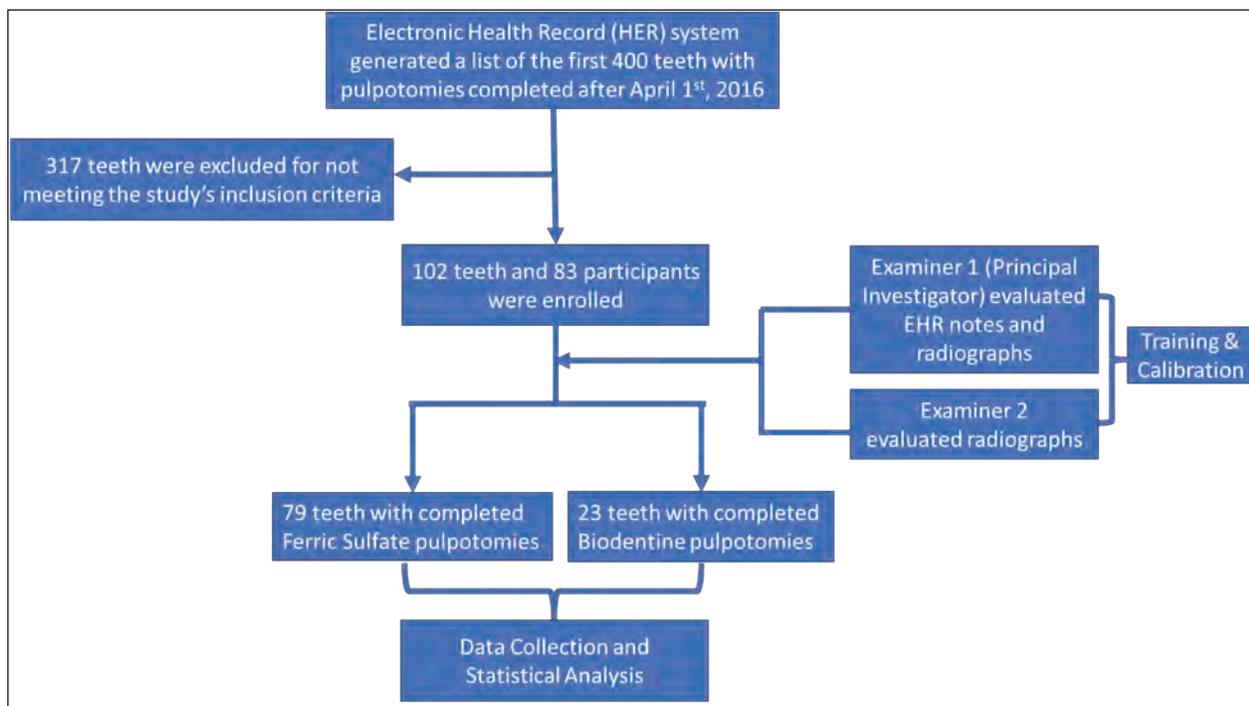
**Descriptive Results of Follow-up Periods**

The follow-up evaluation period for all study teeth ranged from six to 36 months (mean was 17 months). The follow-up time for the BD group ranged from 12 months to 20 months (mean was 15 months). Eleven BD pulpotomies were reviewed at around one year (12 ± 2 months) and 12 at around a year and a half (18 ± 2months). For the FS group, the follow-up periods ranged from six to 36 months (mean was 17 months). Five FS pulpotomies were reviewed at half a year (6 ± 2 months), 19 at a year (12 ± 2 months), 18 at a year and a half (18 ± 2 months), 18 at two years (24 ± 2 months), 16 at two and a half years (30 ± 2 months) and 7 at three years (36 ± 2months).

**Descriptive Results of Clinical and Radiographic Outcomes**

A total of 87.3% (n=89) of all pulpotomies had no clinical signs or symptoms of pathology (scores 1 and 2) and were categorized as successful. The remaining 12.7% (n=13) were categorized as

Figure 1. Flow Chart of Study Progress

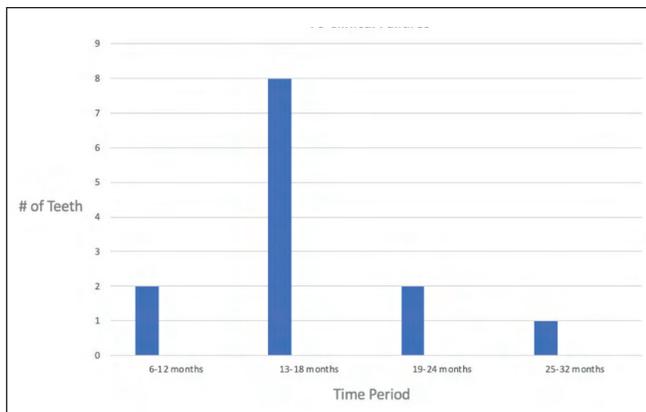


failures (combined scores 3 and 4). Minor discomfort (score 3) was reported for 5.9% (n=6) of the study teeth, while 6.9% (n=7) had major discomfort (score of 4). Radiographically, 23% (n=24) of the whole tooth sample failed (combined radiographic scores of 3 and 4), of which 5.9% (n=6) had required an immediate extraction (score 4).

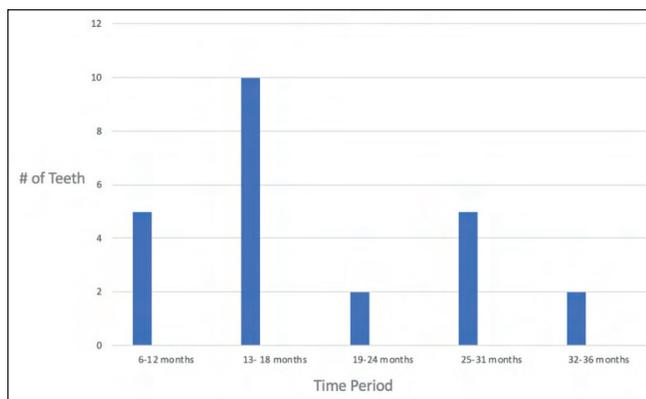
The sample of BD pulpotomies included teeth that were clinically asymptomatic (score 1) and radiographically successful (score 1 for n=19; score 2 for n=4). Four BD pulpotomies (17.4%) had a radiographic score of 2 at a year and a half (18 ± 2 months) as changes consistent with calcific metamorphosis of the intracanal space (pulp canal obliteration) were found. The overall success rate of BD was 100% both clinically and radiographically.

The FS group, however had both clinical and radiographic failures. Overall, 16.4% (n=13) of the teeth treated with FS displayed signs of clinical failure (n=6 with score 3; n=7 with score 4) and 30% (n=24) displayed signs of radiographic failure (n=18 with score 3; n=6 with score 4). Consistent with the descriptions from the scoring system we used, the most common cause for radiographic failure was internal root resorption (IRR) with external changes (41.7%, n=10 out of 24). The clinical and radiographic failures for the teeth treated with FS occurred at different time intervals over 36 months (Figures 2 and 3). The highest number of FS failures occurred between 13 to 18 months post-operatively with 61.5% (n=8 out of the 13) clinical and 41.7% (n=10 out of the 24) radiographic failures.

**Figure 2. Time Intervals of FS pulpotomies Clinical Failures**



**Figure 3. Time Intervals of FS pulpotomies Radiographic Failures**



**Comparative Analysis and Differences between FS and BD Groups**

Clinical and radiographic data was available for both BD and FS at one year (12 ± 2 months) and at a year and a half (18 months ± 2 months). Eleven BD pulpotomies showed 100% clinical and radiographic success at one year, while at a year and a half 12 BD pulpotomies were found 100% successful. From the FS group, at one year 89.5% (n=17/19) of the pulpotomies were clinically successful, while 73.7% (n=14/19) were successful radiographically. At a year and a half, 55.6% (n=10/18) of the FS pulpotomies showed clinical success and 44.4% (n=8/18) had radiographic success. Statistical analysis was run using Chi-square test and it was determined that there was no statistical difference between the two groups at one-year clinical (p=0.220) as well as radiographic (p=0.289) follow-up. At a year and a half follow up, statistically significant differences were found with BD outperforming FS both clinically (p=0.012) and radiographically (p=0.001).

Intra-rater and inter-rater agreement were assessed with Cohen’s Kappa Statistic with both generating a score of κ>0.88 (good reliability).

**DISCUSSION**

The ideal pulpotomy material should be bactericidal, harmless to the pulp and its surrounding structures, promote healing and continuous vitality of the radicular pulp, as well as allow for normal physiological process of root resorption.<sup>2,4</sup> All commercially available products have different advantages and disadvantages and the search for the ideal one is ongoing.<sup>2,15</sup> Furthermore, there is a lack of general consensus regarding the best therapeutic pulpotomy agent.<sup>15</sup> While FS has been widely used in clinical practice, BD is a relatively novel material that has similar composition and indications as MTA products.<sup>12,15</sup> Calcium-silicate-based cements are increasingly popular in regenerative endodontics as they are biocompatible (nontoxic, noncarcinogenic), bactericidal (highly alkaline), insoluble in tissue fluids, and dimensionally stable.<sup>16</sup> BD has been shown to be capable of inducing pulp healing and dentin formation.<sup>12,16</sup>

Our study contributes to the literature evaluating the performance of FS and BD as pulpotomy materials in primary molars in a short and medium term by reviewing retrospectively a sample treated at a university-based PD clinic. We found that the pulpotomies completed with BD were 100% successful a year and a half post-operatively While all teeth remained clinically asymptomatic, 17.4% (n=4) of the BD group developed radiographic changes consistent with pulp canal obliteration (PCO). We considered these teeth successful radiographically (score 2). However, there is controversy in the literature regarding the categorization of PCO. Some authors deem PCO as a radiographic failure acknowledging that it presents an aberration from normal pulp.<sup>17</sup> Others, argue that PCO is an evidence of preserved pulp vitality because is a result of hyperactivity of odontoblast-like cells, hence it should not be classified as a pathologic change or failure.<sup>18</sup> Our results are consistent with recent studies.<sup>14, 19, 20</sup> In a randomized controlled trial conducted by Rajasekharan *et al.*<sup>14</sup>, BD was compared to ProRoot® White MTA and Tempophore™ over a period of 18 months in a total sample of 69 teeth. The authors reported 95.7% clinical and 94.4% radiographic success for BD with statistically significantly more PCO compared to the other two materials.<sup>14</sup>

El Meligy *et al.*<sup>19</sup> compared the outcomes for BD and FC in a sample of 108 primary teeth. The authors determined 100% clinical and radiographic success in the BD group, while FC had also similar outcomes (100% clinical and 98.1% radiographic success).<sup>19</sup> Sirohi *et al.*<sup>20</sup> conducted a randomized controlled trial and evaluated the performance of BD versus FS in a sample of 50 primary molars (25 per group) over a period of 9 months. BD outperformed FS with 100% clinical and 92% radiographic success, versus 96% clinical and 84% radiographic success for the FS group.<sup>20</sup>

In our study, the FS pulpotomies had an overall clinical success rate of 84% and a radiographic success of 70%, which fell within the range reported in the literature.<sup>2,20-22</sup> Havale *et al.*<sup>21</sup> demonstrated that in 12 months in a sample of 30 participants FS had 96.7% clinical and 63.3% radiographic success. Odabas *et al.*<sup>22</sup> examined a sample of pulpotomies completed by dental students and reported 84.7% clinical and 78.2% radiographic success for the FS group after one year. Our cohort of FS pulpotomies showed signs of radiographic and clinical failures as early as six months post-operatively and as late as 36 months. However, the highest number of FS failures occurred between 13 to 18 months follow-up. We also determined that the most common cause for radiographic failure was IRR with external changes. Such findings are consistent with other studies.<sup>2,15,22,23</sup> IRR is believed to be caused by a chronic inflammation in a tooth with a vital pulp.<sup>23</sup> Osteoclasts break down dentin and cementum, which leads to progressive loss of tooth structure.<sup>23</sup> The process of IRR stops once the pulp becomes necrotic, and thus no more viable osteoclasts and nutrients are available for the destructive process to continue.<sup>23</sup> FS is a known irritant, especially at higher concentrations, which can lead to the cascade ultimately causing IRR.<sup>15,23</sup>

Our study had limitations, most of which are inherent to the retrospective design. Our sample size was relatively small, but it is consistent with most published studies, particularly in the BD group, and a loss to follow up is a common issue.<sup>18-22</sup> Another limitation was the reliance on EHR past notes of multiple providers and existing radiographs, which were not standardized. There was a lack of information describing specific considerations regarding the choice of pulpotomy material, hence we can only hypothesize that this decision was made based primarily on clinician's preference. The inconsistent follow-up periods of the pulpotomies was a challenge but expected in the population seen in a public health setting.

Our study also had strengths. Two trained and calibrated examiners evaluated the radiographs. Prospective power analysis was utilized to estimate the initial number of EHR for assessment, as well as only consecutively completed procedures were reviewed to minimize tooth selection bias. All pulpotomy procedures were completed by post-graduate PD students with various clinical experience. However, all critical steps of their work were overseen by attending faculty members, specialists in PD. The pediatric endodontics training of the post-graduate students included the regular pulp therapy course provided by the PD department and the standard pulpotomy technique recommended by the American Academy of Pediatric Dentistry was followed in the clinic.<sup>1</sup> Given that FS was traditionally the standard choice of pulpotomy agent, it is understandable that more teeth of the study sample were treated with this product. Furthermore, providers have had longer experience and better expertise using FS, while utilization of BD was a novelty.

While our study showed promising results for using BD in primary molar pulpotomies, perhaps of even higher practical value to pediatric dental providers is learning more about our experience with the failed teeth treated with FS. We found that FS pulpotomies failed most frequently within 13-18 months post-operatively. Although BD is more costly than FS, its price is not prohibitive for wide clinical use, given its higher success rate. Since pediatric dentists strive to provide the most optimal therapeutic solutions for their patients, FS could be a reasonable choice for teeth with more limited life span remaining (up to two years until exfoliation), while BD can be the preferred material for younger children or those treated with advanced behavior management modalities (e.g. oral sedation, general anesthesia).

Future high-quality clinical trials with longer follow-up are needed to enhance our knowledge of the different therapeutic pulpotomy materials and for more definitive clinical practice recommendations.

## CONCLUSIONS

Based on the results of this study, we can conclude that:

1. FS showed good outcomes, both clinically and radiographically, and its performance may be considered appropriate for clinical practice.
2. The highest number of FS failures occurred between 13 to 18 months post-operatively and the most common cause for FS radiographic failures was advanced internal root resorption.
3. BD outperformed FS and should be the preferred choice of primary molar pulpotomy material.
4. Pulp canal obliteration was the most common radiographic finding in the BD group.

## Data Accessibility:

Data is available on request from the corresponding author.

## Funding:

This was an unfunded study.

## Conflict of Interest:

The authors have no conflicts of interest to declare

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## Bond strength of Ion-releasing Restorative Materials to Sound and Caries-affected Dentin

Gül Keskin\*/Zübeyde Uçar Gündoğar\*\*/ Merve Yaman\*\*\* / Gün Burak Tek\*\*\*\*

**Objective:** This study evaluated the microtensile bond strength ( $\mu$ TBS) of ion-releasing restorative materials to sound and caries-affected dentin (CAD). **Study design:** 60 teeth were randomly divided into 2 groups (sound dentin, CAD) and 5 subgroups of 6 samples each: conventional glass ionomer cement (GIC), resin-modified GIC (RMGIC), glass hybrid reinforced GIC (EQ), giomer (BII), and bioactive restorative material (ACT).  $\mu$ TBS analyses were performed and data were analyzed statistically. **Results:** The ACT group bonded to sound dentin and the BII group bonded to CAD showed the highest  $\mu$ TBS ( $p < 0.05$ ). The GIC, RMGIC, and ACT groups, showed significantly lower  $\mu$ TBS when bonded to CAD compared with sound dentin ( $p < 0.05$ ). However, in the BII group, there were no statistically significant differences between the samples bonded to sound and CAD ( $p > 0.05$ ). All groups except EQ that bonded to sound dentin showed predominantly adhesive failure. **Conclusion:** The use of the giomer can be recommended due to its more stable bond durability.

**Keywords:** Bond strength, giomer, bioactive material, caries-affected dentin, therapeutic ions.

### INTRODUCTION

Dental caries is defined as a localized, multifactor pathological process that softens hard tooth tissues and causes cavitation, and it is a common disease worldwide.<sup>1</sup> In recent years, a partial caries removal technique, which is a minimally invasive approach to protect sound and potentially remineralized tooth tissue, has been recommended, instead of removing the carious tissue completely.<sup>2</sup> Complete removal of caries increases pulp exposure risk and postoperative pulpal symptoms, especially in acute and deep caries lesions. Furthermore, the partial caries removal method is a less invasive alternative, making this technique more advantageous.<sup>3,4</sup> In this technique, the contaminated dentin (caries-infected layer), which indicates the degradation of collagen fibril significantly, is removed. Caries-affected dentin (CAD), consisting of a collagen matrix with less bacterial infection than the contaminated dentin and a regular crossband infrastructure, can be remineralized.<sup>5</sup> It is crucial for a restorative material to have strong adhesion to the tooth to create a suitable microenvironment for dentin remineralization.<sup>6</sup> The formation of a compact and integrated structure between collagen fibrils and restorative material components, preventing permeability against oral and dentin fluids, provides a strong adhesion between adhesive materials and the dental substrate.<sup>7</sup> However, studies have reported that the bonding strength of restorative materials to CAD is generally 20–50% lower than to sound dentin.<sup>8,9</sup> Lower mineral content and cross-link, increased porosity of intertubular dentin, and the lower final tensile strength of carious dentin have been shown to cause lower bond strength for CAD.<sup>10,11</sup> The CAD layer consists of approximately 14–53% water. It has been

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argued that, replacing the water with minerals will increase mechanical properties and bond strength.<sup>12</sup> Previous studies have focused on the use of remineralizing agents to achieve this outcome.<sup>13-15</sup> However, only a limited number of studies evaluating the effect of ion-releasing restorative materials have examined the remineralization potential to increase the bond strength to dental tissues.<sup>7,16,17</sup> Moreover these studies were focused only on the effectiveness of glass ionomer cement (GIC).

Restorative materials with the ability to release “therapeutic” ions (e.g., fluoride, phosphates, calcium, and other minerals) include GIC and resin composites. These materials might reduce the risk of secondary caries by reducing biofilm penetration into the marginal gap of dental restorations and by promoting remineralization throughout the tooth-restoration interface.<sup>18</sup> These materials could reduce the activity of metalloproteinase and proteases such as cathepsins involved in collagen degradation which is considered one of the leading causes of reduced bonding longevity.<sup>19</sup>

This study aimed to evaluate the microtensile bond strength ( $\mu$ TBS) of ion-releasing restorative materials including conventional GIC, its reinforced modifications, surface pre-reacted glass-ionomer (S-PRG) fillers, and bioactive materials to sound and CAD tissue. The hypothesis is that no difference will be found between sound and CAD groups.

## MATERIALS AND METHOD

### Sample preparation

Sixty freshly extracted human third molar teeth were used for this study. Teeth were collected with patients’ informed consent, as approved by the Gaziantep University Clinical Research Ethics Committee (process no:2020/79). The teeth were stored in 0.5% chloramine solution at +4 °C for no longer than 1 month until used. The roots of teeth were embedded in acrylic resin (Imicryl SC; Imicryl Dental Materials, Inc, Konya, Turkey), 1.0 mm below the cemento-enamel junction, using a Teflon mold. The occlusal enamels were abraded perpendicularly to the long axis of the teeth to obtain flat midcoronal dentin surfaces under water cooling and constant pressure, using 600-grit abrasive discs with a polishing machine (Ecomet 3, Bueller, IL, USA). Thus, uniform and standardized smear layers were obtained at the dentin surface. Specimens were examined for any signs of pulp exposure and absence of enamel islets under a stereomicroscope (Leica M125, Leica Microsystems; Heerbrugg, Switzerland) at 30 $\times$  magnification, and then randomly divided into 2 groups: sound dentin and CAD ( $n = 30$ ). Specimens in the sound dentin group were stored in distilled water until the restorative procedure was performed. The pH cycle was performed for 14 days to form artificial caries lesions for the specimens in the CAD group.

### Artificial caries induction

All samples of the CAD group were immersed in demineralization solution (2.2 mM NaH<sub>2</sub>PO<sub>4</sub>, 2.2 mM CaCl<sub>2</sub>, 0.05 M acetic acid, pH=4.5) for 8 hours and in remineralization solution (0.9 mM NaH<sub>2</sub>PO<sub>4</sub>, 1.5 mM CaCl<sub>2</sub>, 0.15 mM KCL, pH=7.0) for 16 hours.<sup>20</sup> The solutions were refreshed daily, and the pH was periodically checked using a pH meter. When the samples were removed from one solution, they were washed with distilled water and dried before immersion in the other.

Image analysis was performed using a scanning electron microscope (SEM; Zeiss Gemini 300 FEG-SEM, Carl Zeiss, Oberkochen, Germany) to evaluate the superficial differences that could affect the bonding to sound and CAD samples.

### Restorative procedure

The specimens were divided into 5 subgroups of 6 samples each: conventional GIC (GIC; Fuji IX extra, GC, Tokyo, Japan), resin-modified GIC (RMGIC; Fuji II LC, GC, Tokyo, Japan), glass hybrid reinforced high-viscosity GIC (EQ; Equia Forte, GC, Tokyo, Japan), giomer (BII; Beautifill II LS Shofu Inc., Kyoto, Japan) and bioactive restorative material (ACT; ACTIVA BioACTIVE Restorative, Pulpdent, Watertown, MA, USA). One calibrated operator performed all restorative protocols according to the manufacturer’s instructions (Table 1). The materials were built up to 4–5 mm in height using a Teflon mold. Chemically cured restorative materials were protected for 2.5 min to avoid moisture contamination or drying out, and the light-cured materials were polymerized using an LED light source with 1000 mW/cm<sup>2</sup> standard power (Valo Cordless, Ultradent, South Jordan, UT, USA). All samples were stored in distilled water at 37 °C for 24 hours.

### Thermocycling procedure and $\mu$ TBS

After storage, to obtain beams of approximately 1 $\times$ 1 mm<sup>2</sup>, each bonded sample was sectioned longitudinally in 2 directions perpendicular to each other across the bound interface for the  $\mu$ TBS test using a diamond disc and a low-speed cutting machine (Isomet, Buehler Ltd, Lake Bluff, IL, USA) under water cooling. The beams’ cross-sectional areas were measured with a digital caliper (Insize 1108–200, Jiangsu Province, China). Thermocycling (THE-1100, SD Mechatronik GmbH, Germany) was applied for 10,000 cycles<sup>21</sup> at 5 °C and 55 °C in distilled water baths with a waiting time of 60 sec and a transfer time of 5 sec. The specimens were inspected under a stereomicroscope at 400 $\times$  magnification to check for cracks or gaps at the tooth-restoration interface after aging. Samples with gaps were excluded from the study. Only 2 beams per tooth were used for the test. Thus, 12 beams in each group were evaluated. All specimens were fixed with cyanoacrylate adhesive system (Pattex, Turk Henkel AŞ, Turkey) to 2 surfaces on a microtensile testing device (MicroTensile Tester, BISCO, Schaumburg, IL, USA). The beams were stressed to failure. The  $\mu$ TBS was expressed in MPa, as determined by dividing the imposed force (N) at the time of fracture by the bonding area (mm<sup>2</sup>). Data were statistically analyzed.

### Failure mode

All debonded specimens were evaluated under a stereomicroscope at 400 $\times$  magnification to determine failure mode. Failure was classified as adhesive, cohesive (in material or dentin), and mixed.

### Statistical analysis

Data were analyzed using SPSS v22.0. The normality of numerical data was tested by the Shapiro- Wilk test. One-way ANOVA and post-hoc Tukey comparison tests were used to compare the groups in normally distributed numerical data. The descriptive statistics are given as mean  $\pm$  std. A  $p < 0.05$  was considered significant.

**Table 1: Composition of the restorative materials used in the study, and their application procedures**

Material	Manufacturers	Composition	Application
Fuji IX extra (Shade A2)	GC, Tokyo, Japan	Polycarboxylic acid, water, polybasic carboxylic acid. Fluoroaluminosilicate glass, particle size of 0.3–200 $\mu\text{m}$ (8% m/m)	-Apply cavity conditioner -Rinse and dry by gently blowing with an air syringe -Apply the restorative material to the dentin surfaces
Fuji II LC (Shade A2)	GC, Tokyo, Japan	2-hydroxyethyl methacrylate, Polyacrylic acid and water. 58 wt% Fluoro-aluminum-silicate glass	-Apply cavity conditioner -Rinse and dry by gently blowing with an air syringe -Apply the restorative material to the dentin surfaces -Light-cure for 20 sec. at 1000 mW/cm <sup>2</sup> standart power.
Equia Forte (Shade A2)	GC, Tokyo, Japan	Carboxylic acid, polyacrylic acid, water. Fluoro-aluminumsilicate glass surface treated glass (wt% not applicable)	-Apply cavity conditioner -Rinse and dry by gently blowing with an air syringe -Apply the restorative material to the dentin surfaces
Beautifill II (Shade A2)	LS Shofu Inc., Kyoto, Japan	Bis-GMA, UDMA, Bis-MPEPP, TEGDMA. 83.3 wt% Fluoro-silicate glass	-Etch for 10-15 sec., rinse for 5 sec. and dry. -Apply G-Premio Bond, light-cure for 10 sec. at 1000 mW/cm <sup>2</sup> standart power. -Apply the restorative material to the dentin surfaces. -Light-cure for 20 sec. at 1000 mW/cm <sup>2</sup> standart power.
ACTIVA BioACTIVE Restorative, (Shade A2)	Pulpdent, Watertown, MA, USA	Blend of diurethane and other methacrylates with modified polyacrylic acid. 55.4 wt% Bioactive glass and sodium fluoride	-Etch for 10-15 sec., rinse for 5 sec. and dry. -Apply G-Premio Bond, light-cure for 10 sec. at 1000 mW/cm <sup>2</sup> standart power. -Apply the restorative material to the dentin surfaces. -Light-cure for 20 sec. at 1000 mW/cm <sup>2</sup> standart power.
Total etch	Ivoclar, Vivadent AG, Schaan, Liechtenstein	%37 phosphoric acid gel	-Apply the cavity for 10-15 sec. -Rinse and dry
Cavity Conditioner	GC, Tokyo, Japan	20% polyacrylic acid solution	-Apply the cavity for 10-15 sec. -Rinse and dry
G-Premio Bond	GC, Tokyo, Japan	4-MET, 10-MDP, MDTP, phosphoric acid ester monomer,	-Apply the adhesive on the dentin surface for 10 sec. -Dry with air gently for 5 sec. -Light-cure for 10 sec. at 1000 mW/cm <sup>2</sup> standart power.

UDMA, urethane dimethacrylate; BisGMA, bisphenol-A glycol dimethacrylate; Bis-MPEPP, Bisphenol A polyethoxy; TEGDMA, triethylene glycol dimethacrylate; 4-MET, 4-[2-(methacryloyloxy)ethoxycarbonyl] phthalic acid; MDTP, 1-methyl-4-phenyl-1,2,3,6-tetrahydropyridine; 10-MDP, 10-methacryloyloxydecyl dihydrogen phosphate.

## RESULTS

### SEM image analysis

SEM images obtained from sound and CAD surfaces are shown in Figure 1. Dentinal tubules are clearly visible on the sound dentin surface. Conversely, most of the dentinal tubules on the CAD surface are occluded by mineral crystals. This occlusion is probably due to continuous mineral deposition that occurs within the tubule lumen and thicker smear layer with enriched organic components.

### $\mu\text{TBS}$ results

Means of  $\mu\text{TBS}$  (MPa) in sound and CAD are presented in Table 2. ANOVA results showed statistically significant differences between the dentin substrates and the restorative materials ( $p < 0.05$ ). The ACT group bonded to sound dentin showed the highest  $\mu\text{TBS}$  values, and this was statistically significant ( $p < 0.05$ ). The next-highest values were obtained from the BII and RMGIC groups bonded to sound dentin. The difference between these 2 groups was not statistically significant ( $p > 0.05$ ). The GIC and EQ groups bonded to sound dentin showed the lowest  $\mu\text{TBS}$  values, and there was no statistically significant difference between these 2 groups ( $p > 0.05$ ).

The BII group bonded to CAD showed the highest  $\mu\text{TBS}$  values, and this was statistically significant ( $p < 0.05$ ). After the BII

group, the highest values were obtained from the RMGIC and ACT groups bonded to CAD. The difference between these 2 groups was not statistically significant ( $p > 0.05$ ). Furthermore, there was no statistically significant difference between the ACT and EQ groups ( $p > 0.05$ ). The EQ and GIC groups bonded to CAD showed the lowest  $\mu\text{TBS}$  values, and there were no statistically significant differences between the groups ( $p > 0.05$ ).

The GIC, RMGIC, and ACT groups bonded to CAD showed significantly lower  $\mu\text{TBS}$  values compared with the same materials bonded to sound dentin ( $p < 0.05$ ). However, in the EQ and BII groups, there were no statistically significant differences between the bonds to sound and CAD samples ( $p > 0.05$ ).

### Failure mode analysis

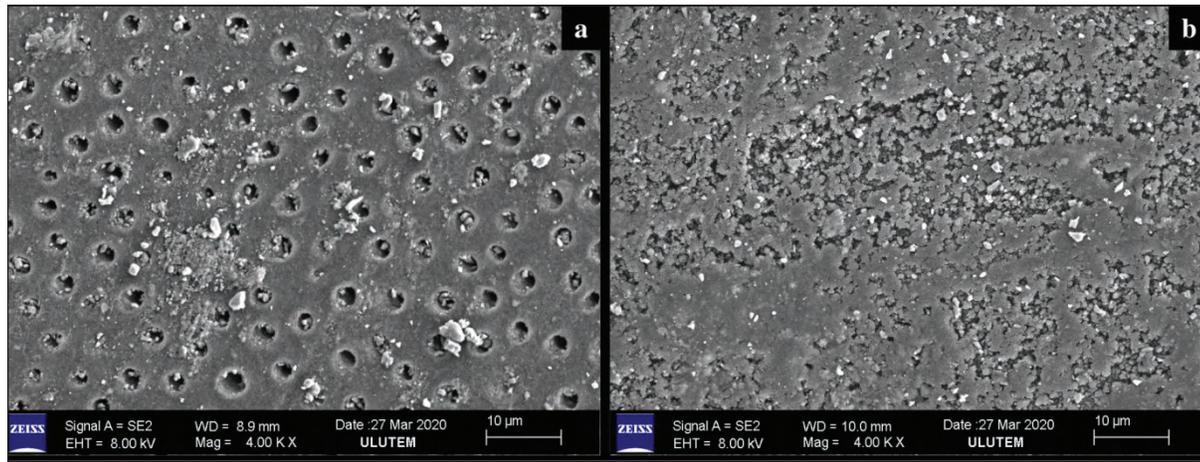
Evaluating the failure modes, Figure 2 shows the percentages of the fracture patterns. All groups showed predominantly “adhesive” failure, except EQ bonded to sound dentin. This group predominantly showed cohesive failure in the material. The fracture pattern of “cohesive failure in dentin” was seen only in the sample of ACT bonded to sound dentin. Mixed fracture patterns were not observed in any group.

**Table 2: Microtensile bond strength values (MPa) in sound and caries-affected dentin**

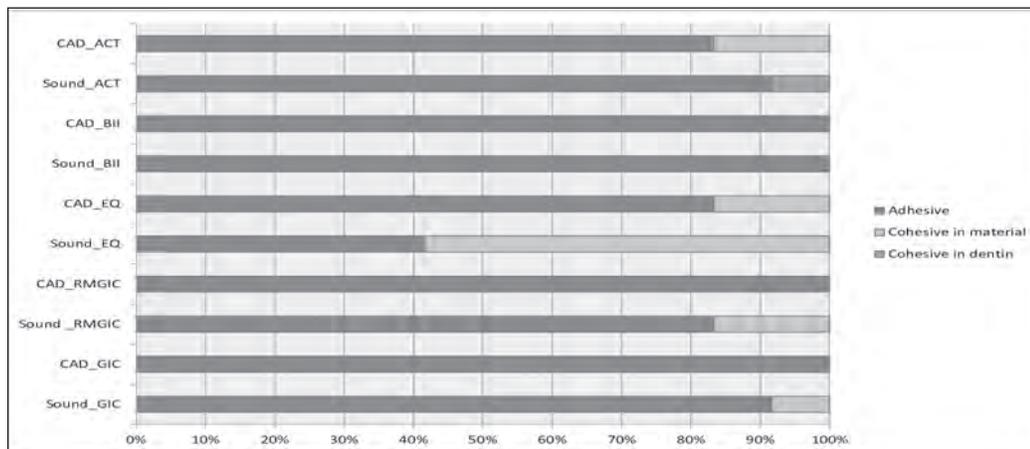
Groups	Sound dentin Mean (std.)	Min / Max	Caries-affected dentin Mean (std.)	Min / Max
GIC	14,72 (4,135) <sup>c*</sup>	10,51 / 22,60	7,06 (2,25362) <sup>c</sup>	2.80 / 10.10
RMGIC	26,17 (7,050) <sup>b*</sup>	14.20 / 40.40	14,68 (3,85200) <sup>b</sup>	9.70 / 21.40
EQ	15,15 (4,559) <sup>c</sup>	6.90 / 23.00	9,78 (3,86590) <sup>c</sup>	4.00 / 17.90
BII	26,75 (5,592) <sup>b</sup>	16.80 / 34.40	26,11 (7,56473) <sup>a</sup>	10.10 / 40.40
ACT	39,06 (8,282) <sup>a*</sup>	25.10 / 49.30	11,84 (2,66679) <sup>bc</sup>	8.10 / 19.00
All groups	24,37 (10,811) <sup>*</sup>	6.90 / 49.30	13,89 (7,92342)	2.80 / 40.40

Superscript different letters in the same column mean statistically significant differences (p<0.05), \* means statistically significant difference when compared to the sound and caries-affected dentin (p<0.05).

**Figure 1. SEM image analysis. a; Sound dentin, b; Caries-affected dentin (4.00 kx magnification).**



**Figure 2. The percentages of fracture patterns according to the groups.**



**DISCUSSION**

In the present study, all groups except BII and EQ showed lower bonding values to CAD when compared with sound dentin. Thus, the hypothesis that there would be no differences between materials' bond strength values to sound and CAD samples was rejected. These findings are compatible with those of previous studies in which reduced  $\mu$ TBS values were observed in the CAD group compared with sound dentin, regardless of adhesive systems or strategies.<sup>22,23</sup> This finding has been associated with the formation of a deeper demineralized zone with acid etching in CAD.<sup>24</sup> The high amount of water in this zone competes with the penetration of adhesive resin monomers. Therefore, it becomes difficult for the resin monomer

to penetrate the base of the exposed collagen matrix. Furthermore, caries-affected surfaces might contain substances that interfere with the formation or spread of free radicals and lead to poor polymerization of adhesive monomers.<sup>25</sup> Thus, weaker and unstable bonding is obtained in resin restorations that are bound to the tooth through an adhesive system.

The situation is slightly different in GIC materials. These materials are bound to the tooth by chemical adhesion, provided by ionic and polar interactions between polycarboxylate radicals and hydroxyapatite. This interaction is also thought to be beneficial in reducing hydrolytic degradation, thereby increasing the restoration longevity.<sup>7</sup> However, CAD contains more residuals and b-tricalcium

phosphate<sup>18</sup> minerals (whitlockite) in the dentinal tubules, which are less soluble than hydroxyapatite compared with sound dentin and could negatively affect this ionic interaction.<sup>22</sup> Potentially unstable adhesive interfaces can degrade slowly and continuously through water absorption. In this case, dentin biomodification is crucial to strengthen the bonding stability.<sup>26</sup> Ion-releasing restorative materials were used to provide the dentin biomodification in this study.

In the present study, the hypothesized results were obtained only from the BII group. The similar binding to sound and CAD samples could mean this material eliminates the negative factors that affect bonding to CAD. Gioners (glass ionomer+polymer) are resin-based, fluoride-releasing, PRG (pre-reacted glass-ionomer) fillers containing restorative materials. In the presence of water, PRG fillers are prepared through an acid-base reaction between fluoroaluminosilicate glass and polyalkenoic acid.<sup>27</sup> Unlike GICs, the acid-base reaction in the giomer occurs in S-PRG fillers during the production phase. This reaction forms a modified layer on the material's surface, which protects against the harmful effects of moisture.<sup>28</sup> The S-PRG fillers can release the ligand in the pre-reacted hydrogel, increasing the rapid release of fluoride, and the fillers can also release Al, Na, B, Si, and Sr ions. In this way, fluoride and silicate encourage remineralization of the dentin matrix, and hydroxyapatite crystals are converted to fluorapatite and strontium apatite by fluoride and strontium, thereby increasing the tooth resistance to acid.<sup>29</sup> In this study, giomer might have caused the substrate to be more hydrophilic and a suitable substrate for bonding, with the formation of feasible and regular reconstruction in demineralized dentin. The restructured mineralized surface, which forms through an organized crystal formation guided by a collagen matrix scaffold, could experience a high level of wettability and high surface energy by resin monomers.<sup>30</sup> In addition, there is a functional 10-methacryloxidesyl dihydrogenphosphate (10-MDP) monomer in the content of the universal bond (G-Premio Bond) used in our study. Pinna *et al.*<sup>31</sup> argued that there might be a possible chemical interaction between 10-MDP and CaF<sub>2</sub>, which occurs on the demineralized surface after fluoride application. Similar bonding values to sound and CAD dentin were associated with this situation in their study. The protective effect of the calcium salts of the formed 10-MDP and the resin-coated collagen, as well as the formation of more homogeneous hybrid layers, could explain the superior bond stability, as in the present study.<sup>32</sup>

Another striking result of this study was that the best bonding stability to sound dentin was found in the ACT group. This material is a new concept that combines the ion-release capacity of GICs with the optimal mechanical and aesthetic properties of resin materials.<sup>33</sup> ACT has a fluoroaluminosilicate glass structure similar to GIC. This structure can dissolve in acidic conditions, and the material gains the ion-releasing ability.<sup>34</sup> This restorative material includes a triple setting mechanism, according to the manufacturer: The acid-base neutralization reaction of GICs, light-cure, and self-cure of the matrix. Furthermore, it is recommended that ACT be applied as a self-adhesive or with a universal adhesive. Latta *et al.*<sup>35</sup> reported higher bonding strength values to enamel and dentin when using a universal adhesive with this material compared with a self-adhesive application. The universal adhesive could be the reason for the superior bond stability to sound dentin in the ACT group of the present study. However, it is interesting that the material had a lower bond strength to CAD than to the sound dentin. The presence of denatured collagen fibrils, lack of crossbanding, and inadequate resin

infiltration might occur in the interfiber collagen spaces, which could compromise bond strength.<sup>5</sup> In addition, irregular deposition and precipitation of mineral on dentin could mechanically destroy tubules and reduce the material's bonding performance.<sup>36</sup>

In our study, the lowest  $\mu$ TBS values were obtained from the GIC and EQ groups bonded to both sound and CAD samples. In these 2 groups, adhesion to dentin is occurs through both chemical bonding and micro-mechanical locking. RMGIC contains a resin monomer 2-hydroxyethyl methacrylate (HEMA) to provide better adhesion than conventional GIC.<sup>37</sup> The RMGIC group in this study also showed bonding stability superior to that of the GIC and EQ groups. However, its bonding to CAD was lower compared with its bonding to sound dentin. These results are compatible with the literature.<sup>16,17</sup> In a study evaluating conditioning effects, the root dentin bond strength of RMGIC was found to be lower when bonded to CAD than to sound dentin, regardless of conditioner application.<sup>16</sup> Contrary to these results, only one previous study showed that the bond strength of RMGIC in primary teeth was similar to that of sound and CAD samples. Researchers have claimed that this finding might be due to the use of cured primers on the dentin surface before RMGIC application.<sup>7</sup>

Adhesive failure affected most of the specimens in this study. The percentage of cohesive failure was predominant only in the EQ group, bonded to sound dentin. Aligning the specimen along the long axis of the test device, micro-cracks of the sample produced by slicing and the fragility of the material were reported as causes of cohesive failure. It is recommended to discard cohesive failure samples and to select only adhesive failure or a small portion of mixed failure (<10%) specimens for more reliable bond strength estimation. However, none of the studies excluded cohesive failure samples from the bond strength analyses.<sup>37</sup> In the present study, cohesive failure was not excluded because it involved a small percentage of the samples.

The current concept suggests a less invasive approach to the treatment of carious lesions. The main principle is to remove only the contaminated dentin and create a biological seal for the remaining tissue. In this case, the restorative materials' bonding ability to the CAD is crucial. In this study, restorative materials with different contents and bonding ability were tested. Although the obtained data offer an idea for clinical practice, they do not exactly mimic in vivo conditions. In addition, it is recommended to compare the bonding according to different adhesive systems and strategies as those differences could cause differences in bonding stability.

## CONCLUSION

Within the limitations of this study, the use of the giomer can be recommended, especially for the partial caries removal technique, because it showed more stable bond strength than other materials did in both sound and CAD samples. Moreover, bioactive restorative materials showed superior bond stability to sound dentin. Ion-releasing restorative materials have remineralization potential on the carious dentin. In addition, the hypothesis of increasing bond strength to CAD is essential for biomimetic and preventive dentistry that supports the minimal loss of dental tissues and aims to reconstruct the remaining structure. Further clinical and in vitro studies are needed to realize this idea.

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## Evaluation of Serum 25(OH)D Levels in Obese and Normal-Weight Children with Carious and Hypomineralized Teeth

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**Aim:** The aim of this study was to assess the association between dental caries, molar incisor hypomineralisation (MIH) and obesity in relationship with different vitamin D levels in children. **Study design:** This retrospective case-controlled study enrolled 455 children aged 6-18 years, who attended to both pediatric endocrinology and pediatric dentistry clinics at the Aydın Adnan Menderes University Hospital, Turkey. Vitamin D status was measured with serum (25(OH)D) concentrations. Body mass index (BMI) were used to determine adiposity. Caries status was assessed using the decayed-missing-filled teeth (dft) and (DMFT) index for primary and permanent dentitions using WHO standard methodology. MIH were diagnosed according to the EAPD criteria. **Results:** DMFT did not show any significant difference between obese and normal weight children in both age groups. However, in 6-11 age group, obese children had lower dft and the difference was statistically significant ( $p < 0.001$ ). Median caries index values and MIH prevalence among the obese and normal weight children found similar with deficient, insufficient and sufficient levels of serum 25(OH)D in both age groups. **Conclusion:** Our analyses provide no evidence to suggest that obese children are at increased risk for dental caries. Serum 25(OH)D concentrations would not seem to have a significant effect on dental caries and MIH in children.

**Keywords:** obesity, children, vitamin D, dental caries, MIH

### INTRODUCTION

Dental caries is the most common childhood chronic disease, with a prevalence of more than 50% in many countries.<sup>1</sup> Untreated dental caries can lead to serious problems, such as caries-related pulpitis, pain, tooth loss, and other co-morbidities, which can affect nutritional status, growth, development, and quality of life in children.<sup>2</sup>

There is currently an increasing interest in the association between oral health and adiposity status of children as well as adults. Dietary habits, poor oral health behaviors, genetic, and environmental risk factors were assumed to be common contributors to both obesity and dental caries.<sup>3</sup> and 282 of them completed all 3 phases of data collection. Body mass index, waist circumference, waist-to-hip (WHR) Systematic reviews have examined the cross-sectional relationship between anthropometric values and dental caries, and a vast of majority of these studies have inconclusive associations.<sup>4-6</sup> A longitudinal study of the association between obesity and dental caries showed that there was no relationship between the two chronic diseases in children and adolescents.<sup>7</sup>

Another important topic addressed by researchers is vitamin D, which has been identified as one of the key nutrients that contributes to the development and maintenance of optimum bone mass, tooth formation, and metabolic reactions.<sup>8</sup> Theodoratou *et al*<sup>8</sup> in their umbrella assessment using a systematic review and meta-analyses showed that dental caries in children is one of the health outcomes associated with vitamin D deficiency. Due to vitamin D's key role

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in hard tissue formation, vitamin D deficiency was also suspected to be one of the causative factors of molar-incisor hypomineralization (MIH)<sup>9</sup>, of which the precise etiology is uncertain. MIH is a type of enamel defect affecting, as the name suggests, the first molars and incisors in the permanent dentition.<sup>10</sup> Teeth affected with MIH are at an increased risk of acquiring dental caries and breakdown.<sup>11</sup>

It is well known that even in sunny countries, vitamin D deficiency is observed in different regions and have become a common problem in the world not only for adults, but also for the pediatric population.<sup>12</sup> Measurement of total 25(OH)D levels is the best test to assess body stores of vitamin D; however, there are still different opinions about the threshold value of 25(OH)D for deficiency.<sup>13</sup> Earlier studies from Turkey have noted an extended diversity of vitamin D deficiency in children depending on the population size and threshold used.<sup>13,14</sup>

Since the studies regarding serum 25(OH)D levels in children with dental caries and its connection to body mass index (BMI) are questionable, we designed a retrospective case-controlled study aiming to 1) examine serum 25(OH)D levels in children living in the Aegean region of Turkey, 2) determine the association between obesity and caries status, and 3) investigate the relationship between 25(OH)D, MIH, and caries status.

## MATERIALS AND METHOD

Data were collected from patients who attended to both the pediatric endocrinology and pediatric dentistry clinics at the University of Adnan Menderes between December 2016 and June 2017. The medical files of all children were reviewed. Non-syndromic obese children were compared with an age- and sex-matched control group of normal-weight counterparts. The subjects comprised 455 children aged 6–18 years old living in the city of Aydın (Aegean region, Turkey) who participated in blood tests and oral examinations. The Ethics Committee of the University of Adnan Menderes, Faculty of Medicine (Protocol Number: 2016/934) approved the study, which was performed according to the Helsinki Declaration.

Our sample was purposely divided into two main groups and two subgroups depending on BMI and dentition status, respectively. For BMI, the study population was classed into two groups based on whether the children were normal weight or obese using the international BMI cutoff points described by Cole et al<sup>15</sup> Moreover, the BMI percentile specific to age and gender of Neyzi *et al*,<sup>16</sup> a national standard of growth and body composition, was used to classify children as obese (BMI percentile  $\geq 95^{\text{th}}$ ), overweight ( $\geq 85^{\text{th}}$  to  $< 95^{\text{th}}$ ) and normal weight ( $> 16^{\text{th}}$  to  $< 85^{\text{th}}$ ).

All children in the study had the criteria of their first permanent molars and incisors being fully erupted. The caries assessment was determined as a tooth- and surface- related DMF index for the primary dentition (dft) and permanent dentition (DMFT) using the WHO standard methodology.<sup>17</sup> The caries status stated in the study was caries prevalence (at least one decayed tooth in any of the dentitions) and caries experience (the total count of decayed and filled teeth in both dentitions). Caries-free children are defined as children with no prior caries experience.

The MIH assessment was determined as follows: demarcated opacities, post-eruptive breakdown (PEB) of the hypomineralized enamel and atypical restorations (ARs) were diagnosed as enamel hypomineralization (EH), according to the EAPD criteria,<sup>11</sup> on

all permanent teeth and surfaces. Hypomineralized lesions with a diameter  $< 1$  mm were not recorded. EH-associated defects were not scored in the DMF index.

Laboratory results were extracted from the patient records. Children with chronic conditions and those using medication or supplements were excluded from the study. To classify serum 25(OH)D concentrations in our population, the cutoffs reported in the consensus created by Saggese *et al*<sup>18</sup> were used. Vitamin D-deficient concentrations were considered as those  $\leq 12$  ng/ml; a level of 12–20 ng/ml was considered to indicate vitamin D insufficiency, and a level of 20 ng/ml or greater was considered sufficient.

Statistical analyses were performed using IBM SPSS Statistics 17.0 (IBM Corporation, Armonk, NY, USA) Mean and standard deviation or median (minimum–maximum) values are given for continuous variables. Categorical variables are expressed as frequencies and related percentage values. They were compared using the chi-square test or Fisher’s exact test. Nonparametric tests (Mann–Whitney U, Kruskal–Wallis) were used for data without normal distribution. The DMFT/dft of both groups was divided into caries experience (total number of DMFT and dft separately) and caries prevalence (DMFT $>0$ ; dft $>0$ ) subgroups. Correlation analysis was carried out to determine the effects of demographic factors on these subgroups of obese and normal-weight children. The effects of oral index scores greater than 0 and all possible factors thought to be effective in predicting the presence of MIH were investigated using a multivariate logistic regression analysis. The adjusted risk ratios were calculated with 95% confidence interval. A p value  $< .05$  was considered to be statistically significant.

## RESULTS

A total of 455 children were included into the study. The mean age of the children was 12.3 years (standard deviation: 3.0). The distribution of boys and girls between normal weight and obese children was homogeneous in both 6-11 and 12-18 age group (p=0.442 and p=0.725).

The corresponding median values and standard deviation for the recorded caries index parameters are documented in Table 1 and 2, according to the BMI and 25(OH)D levels. The caries prevalence was found as (DMFT  $> 0$ ) %67; (dft  $> 0$ ) %54.1. Caries indexes according to the age groups in the study population found as follows: 6-11 age: DMFT: 1.77 $\pm$ 1.99 , dft: 2.71 $\pm$ 3.24. 12-18 age: DMFT: 3.54 $\pm$ 2.91.

DMFT did not show any significant difference between the obese and normal weight children in both age groups (p $>0.05$ ). On the other hand, in 6-11 age group, obese children had lower dft and the difference between obese and normal weight children was statistically significant (p $<0.001$ ). Caries prevalence and experience between the obese and normal weight children were similar in 12-18 age group (p $>0.05$ ); but the same result was not valid for the 6-11 age group.

Given the whole of the study population, MIH prevalence was %7.3 and ratios of having MIH were statistically similar in both obese and normal weight children with no gender predilection. (p=0.995)

Table-2 shows the multivariate analysis of the association between 25(OH)D levels, caries and MIH prevalence or experience. There were no statistically significant differences in the median caries

indexes among the groups with deficient, insufficient and sufficient levels of 25(OH)D in the both 6-11 and 12-18 age groups ( $p>0.05$ ).

Table-3 shows the median 25-(OH)D levels comparing the caries and MIH prevalence in age groups. As an interesting result, in 6-11 age group, median 25(OH)D level was significantly higher in children had dft than whom with caries free ( $p=0.013$ ). When controlling the MIH and 25(OH)D levels, there was no statistically significant association between the two variable in both 6-11 and 12-18 age groups ( $p>0.05$ ).

Among the whole study population, the proportion of children who had sufficient serum 25(OH)D levels in the 6-11 age group was statistically higher than the 12-18 age group; while the ratio of children who had deficient serum 25(OH)D levels were statistically lower when comparing the 12-18 age group ( $p=0.021$ ). Boys had significantly higher 25(OH)D levels than in girls ( $p <0.001$ ). Furthermore, there was a statistically significant and inverse correlation between age and serum 25(OH)D levels ( $r=-0.154$  and  $p<0.001$ ).

**Table-1: Caries prevalence and experience and MIH status of the children according to body mass index in age groups.**

Demographics	6-11 age			12-18 age				
	Girls	128 (%57.7)		132 (%56.7)				
	Boys	94 (%42.3)		101 (%43.3)				
	Normal weight(n=74)	Obese (n=148)	p-value	Total (n=222)	Normal weight(n=71)	Obese (n=162)	p-value	Total (n=233)
DMFT	1.85±1.87	1.72±2.05	0.398†	1.77±1.99	3.51±3.12	3.56±2.83	0.793†	3.54±2.91
DMFT > 0	46 (%62.2)	77 (%52.0)	0.152‡	123 (%55.4)	52 (%73.2)	130 (%80.2)	0.308¶	182 (%78.1)
dft	4.22±3.79	1.95±2.64	<0.001†	2.71±3.24	-	-	-	-
dft > 0	51 (%68.9)	69 (%46.6)	0.002‡	120(%54.1)	-	-	-	-
MIH								
Teeth (n)	0.34±1.14	0.25±0.99	0.595†	0.28±1.04	0.13±0.61	0.17±0.67	0.392†	0.16±0.65
Children (n)	7 (%9.5)	11 (%7.4)	0.794¶	18 (%8.1)	3 (%4.2)	12 (%7.4)	0.563\$	15 (%6.4)

† Mann Whitney U test, ‡ Pearson's chi square test, ¶ Continuity-corrected chi-square test, \$ Fisher's exact test.

**Table-2: Caries prevalence and experience and MIH status of the children according to 25-(OH) D levels in age groups.**

Serum 25-(OH)D	6-11 age				12-18 age			
	Deficiency (≤12 ng/ml) (n=41)	Insufficiency (12-20 ng/ml) (n=118)	Sufficiency (20 ng/ml) (n=63)	p-value	Deficiency (≤12 ng/ml) (n=68)	Insufficiency (12-20 ng/ml) (n=114)	Sufficiency (20 ng/ml) (n=51)	p-value
DMFT	2.24±2.46	1.74±1.92	1.51±1.74	0.383†	3.99±2.94	3.36±2.92	3.37±2.86	0.258†
DMFT >0	23 (%56.1)	68 (%57.6)	32 (%50.8)	0.675‡	56 (%82.4)	86 (%75.4)	40 (%78.4)	0.550‡
dft	2.12±3.26	2.87±3.41	2.78±2.90	0.213†	-	-	-	-
dft > 0	16 (%39.0)	65 (%55.1)	39 (%61.9)	0.069‡	-	-	-	-
MIH								
Teeth (n)	0.07±0.47	0.25±1.03	0.46±1.28	0.161†	0.01±0.12	0.23±0.82	0.20±0.63	0.123†
Children (n)	1 (%2.4)	9 (%7.6)	8 (%12.7)	0.166‡	1 (%1.5)	9 (%7.9)	5 (%9.8)	0.075¶

† Kruskal Wallis test, ‡ Pearson's chi square test, ¶ Likelihood-Ratio test.

**Table-3: Median 25-(OH)D levels comparing the caries and MIH prevalence in age groups.**

	6-11 age			12-18 age		
	n	Median 25(OH)D (ng/ml)	p-value	n	Median 25(OH)D (ng/ml)	p-value †
DMFT			0.672			0.562
DMFT = 0	99	17.62±6.82		51	17.24±10.40	
DMFT>0	123	17.39±7.09		182	15.53±6.87	
dft			0.013			-
dft=0	102	16.52±6.76		-	-	
dft>	120	18.31±7.05		-	-	
MIH			0.061			0.069
-	204	17.26±6.95		218	15.73±7.81	
+	18	20.11±6.66		15	18.50±7.22	

† Mann Whitney U test.

## DISCUSSION

The purpose of our study was to examine a possible association between dental caries and obesity and investigate the potential role of vitamin D on this association in children 6-18 years of age using data collected from the records. Our analyses provide no evidence to suggest that obese children are at increased risk for dental caries. Indeed, while not entirely consistent, the data suggest that adiposity might be associated with a slightly reduced risk for caries in 6-11 age in primary dentition. No significant association was found in caries status and BMI in permanent dentition both in 6-11 and 12-18 age groups.

The previous studies that examined the relationship between BMI and caries have been sparse and inconclusive. Studies linking obesity and caries in children have including positive associations<sup>19,20</sup> elementary school children (648 boys, 642 girls, no associations<sup>20</sup>, and inverse associations.<sup>21,22</sup> Chen et al<sup>23</sup>, in their recent systematic review and meta-analysis, declared that no consistent association between dental caries and any weight group in children both primary and permanent dentition.

Similarly, to the above, there have been contradictory findings from studies in Turkey. The cross-sectional study by Köksal *et al*<sup>24</sup> showed that overweight children had lower prevalence (66.1%) of dental caries than the underweight ones (89.7% ) whose age ranged from 5 to 9 years. Bulut *et al*<sup>25</sup>, in their case-control study, suggested that obesity seems to be not a potential risk factor for dental caries in primary dentition, but there was a statistically significant association in permanent dentition.

In our study, 6-11 age group, caries prevalence and experience was lower in obese children than in normal weight children. Similar observation was made in several studies on primary dentition.<sup>26-29</sup> A recent review<sup>6</sup>, which evaluated the correlation between obesity and dental caries in children, found that dental caries were more common in obese children than in normal weight children in permanent dentition; but the same study did not find any correlation in primary dentition. As an interesting result, we found not only a lack of association but a reverse correlation between dental caries and body weight for the primary dentition; confirming the results of previous studies.<sup>26,29,30</sup> We might explain this finding possibly that carious primary teeth may have exfoliated and caries in the permanent dentition may not have had sufficient time to develop. Accelerated linear growth and sexual maturation is associated with adiposity and primary teeth exfoliate earlier in overweight and obese children.<sup>31</sup> This could lead to false positive assessment of caries status.

Our findings that obesity is not linked to dental caries is in contrast with numerous clinical studies. According to many researchers, obese children have significantly more caries in their primary and permanent teeth than lean children. However, they stated that obesity is not the only etiological factor for the caries formation. These disparity may be caused by variations in study design, area and the age of the participants.<sup>32</sup> Furthermore, caries indexes and BMI cut-off values differed across the studies. Several authors also hypothesized that BMI may not be the most suitable tool for assessing a child's weight status when investigating the association between obesity and caries. Possibly we could say that the evidence is inadequate to explain the link between obesity and dental caries solely based on cariogenic food consumption.

To the best of our knowledge, there is no study in the literature that investigated the link between dental caries and obesity and the possible effects of vitamin D on these two chronic conditions. Although there are studies asserting a decrease in serum 25(OH)D levels in the presence of obesity; in our study, serum 25(OH)D levels did not show any difference between the obese and normal weight groups. In order to give an idea about this topic, children also need to be evaluated in more detailed which included serum glucose and lipid metabolism biomarkers.

Many studies in the literature have claimed that vitamin D can prevent inception and development of dental caries. Mellanby *et al*<sup>33</sup> showed an association between vitamin D supplementation and reduced caries risk in their results. Vitamin D in the first few years of life may be prophylactic for dental caries, as also studied by Hujoel *et al*<sup>34</sup> On the other hand, Gyll *et al*<sup>35</sup>, in their follow-up study, showed that vitamin D status at 6 years of age was unrelated to enamel defects; but associated with saliva LL37, an innate immunity peptide attacking cariogenic bacteria, for which expression has been linked to vitamin D status. In studies using national representative data from researchers, Schroth *et al*<sup>36</sup> suggested that there is an association between caries and lower serum 25(OH)D levels in Canadian children. Inversely, Herzog *et al*<sup>37</sup> concluded that they did not find a significant association between serum 25(OH)D levels and caries experience in US children.

Our findings showed that the prevalence of MIH was similar in the population; deficient, insufficient and sufficient 25(OH)D levels at any age group. MIH prevalence was %7.3 and the ratios of children with MIH were statistically similar in both obese and normal weight children. ( $p=0.995$ )

Kühnisch and colleagues<sup>9</sup>the precise etiology of molar-incisor hypomineralization (MIH, investigated the relationship between MIH and 25(OH)D for the first time in the literature. MIH was diagnosed in %13.6 of their study population and mean concentration of 25(OH)D was 75.8 nmol/l (standard deviation 22.0 nmol/l). Furthermore, they found that higher 25(OH)D values was significantly associated with a lower OR of having hypomineralized teeth, and a reduced incidence of caries. Ghanim et al<sup>38</sup>there has been increasing interest worldwide in investigation of the prevalence of demarcated opacities in tooth enamel substance, known as molar-incisor hypomineralisation (MIH, examined 810 children and (%20.2) presented MIH lesions at least one index teeth and they found interestingly that obesity was negatively correlated to children having MIH.

Median serum 25(OH)D level was measured as 16.67 (ng/ml) in our study. Unfortunately seen from this result, children with vitamin D deficiency (24.0%) and insufficiency (51.0%) were in the majority. Although our data being represented the different seasons to avoid the radiation effects in vitamin D, the high frequency of lower vitamin D levels in the population conferred a very narrow range of serum 25(OH)D concentrations, so making comparisons could be limited. Nevertheless, these results are in concordance with the studies performed in our country. Gultekin et al.<sup>39</sup> determined a vitamin D deficiency of 10.45% in children aged 6-17 years age. Another study in Turkey found subclinical vitamin D insufficiency in 40% of healthy female adolescents.<sup>40</sup> These data emphasize the problematic situation even in the sunny climate of Turkey.

Our findings must be evaluated within the confirmed limitations. First, it was a retrospective study; therefore, there is no inference

in terms of causes and effects. Long-term follow-up studies would give us more detailed information about these complex interactions. Second, we had no data about socioeconomic status of children; but we know that the university have a comprehensive hospital in the Aydın city where patients from many different social backgrounds apply. Third, dietary data was not available and we could not obtain data about oral hygiene habits due to the most of the children in the study did not know if they were using fluoridated paste. Moreover, those patients hadn't informed about correct tooth-brushing techniques or proper eating habits before the study. So, it could be difficult to speculate between the groups in terms of info and awareness background. For these reasons, although the sample size does not represent the whole population, it could be demonstrated that the number of patients are superior compared to other studies and represents a population with as homogeneous distribution as possible. Another issue to consider, caries indexes and obesity indices differed across the studies. Different methods complicate standardization and makes difficult to compare with studies in the literature.

Our research would allow considerations for the association of serum 25(OH)D concentrations during hard tissue mineralization and the clinical status after tooth eruption. Additionally, pubertal status of children has not been questioned in our research. Sexual maturation is associated with adiposity and growing age may alter vitamin D; biomarkers (eg: calcium, phosphate and PTH) having direct effects on final 25(OH)D concentrations. So, these important headings could lead another study subject.

## CONCLUSIONS

Our study provides no evidence to suggest that obese children are at increased risk for dental caries. Furthermore, serum 25(OH)D concentrations would not seem to have a significant effect on dental caries and MIH lesions. Given the lack of consensus regarding the association between vitamin D and dental caries, highlighting the issue by further studies is important to guide future prevention and treatment efforts. It is worthwhile to emphasize that pediatric dentists are reminded they still have an important role in influencing eating habits and food choices.

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# Oral Care in Kindler Syndrome: 7-Year Follow-up of 2 Brothers

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**Background:** Kindler poikiloderma is an inherited autosomal genodermatosis characterized by blistering of the epidermis and mucosae. Its prevalence is unknown. **Case report:** We monitored two brothers suffering from this pathology. Oral manifestations mainly take the form of periodontal lesions. In our patients we noted gingivitis progressing to periodontitis as follow-up care was not effective. We also diagnosed enamel hypoplasia, described more rarely in this pathology. **Conclusion:** Periodontitis in Kindler Syndrome responds to maintenance therapy, but the absence of surveillance is penalized by a deterioration in periodontal condition and complication of management. All restorative, endodontic, surgical, periodontal and orthodontic treatments should be performed with appropriate precautions.

**Keywords:** Kindler syndrome, epidermolysis bullosa,, enamel hypoplasia, periodontal disease.

## INTRODUCTION

Inherited epidermolysis bullosa (CIM-10 Q81.0, Q81.1, Q81.2, Q81.8, Q81.9; ORPHA:7936) is a group of rare genodermatoses. Four main subtypes are described: Epidermolysis Bullosa Simplex, Dystrophic Epidermolysis Bullosa, Junctional Epidermolysis Bullosa, and Kindler Syndrome (KS). In all these conditions, proteins involved in the junction between the epidermis and the dermis are abnormal, due to genetic mutation with consequences ranging from moderate discomfort to disorders that cause serious impairment according to the severity of the forms. Extreme fragility of the skin reduces resistance to shearing and frictional injury. A cleavage occurs between these two skin layers in the event of collision, friction or trauma and blisters can appear<sup>1,2</sup>.

Kindler poikiloderma is an autosomal recessive pathology, characterized by blisters arising in multiple levels of cleavage within the epidermis and mucosae, and by specific clinical phenotypic features, such as photosensitivity. Its prevalence is unknown. Persons of any race can be affected and there is no gender predilection. Symptoms begin at birth, or during infancy<sup>1</sup>. Management of these patients requires a multidisciplinary team, in which a dentist is essential, because the hard and soft tissues of the oral cavity, and particularly the periodontium are very often involved in the clinical manifestations (Table 1). We introduce two brothers E., born in 2006 and S., born in 2002, treated in the Competence Center for Rare Odontological Diseases at Timone University Hospital (Assistance Publique-Hôpitaux de Marseille) since 2009 which specialises in KS.

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Table 1: Case reports describing the main oral manifestations and management of patients with Kindler Syndrome: review of literature published between 1997 and 2019.

Authors of Articles	Number of cases	Gender/Age	Oral manifestations	Oral care described
24 ➤ Anwar et al. ➤ Barbosa et al. ➤ Burch et al. ➤ De Almeida et al. ➤ Fischer et al. ➤ Ghosh et al. ➤ Gkaitatzi et al. Krishna et al. ➤ Kustner et al. Mansur et al. Mendes et al. ➤ Nofal et al. ➤ Penagos et al. Ricketts et al. Sharma et al. ➤ Suga et al. ➤ Suman et al. Torkzaban et al. Wada et al. ➤ Wiebe et al. (1) Wiebe et al. (2) ➤ Yazdanfar et al. ➤ Yildirim et al. ➤ Zhou et al.	86	38 F / 2-54y 48 M/4-49y	<ul style="list-style-type: none"> <li>• Oral mucosa: Hyperkeratosis, Painful ulcers on soft palate, lips, floor of the mouth, labial commissure, hard palate</li> <li>• Gingivitis: Chronic, desquamative</li> <li>• Periodontitis: mild to severe with rapid progression</li> <li>• Tooth mobility, premature loss</li> <li>• Ankyloglossia</li> <li>• Difficulty in swallowing</li> <li>• Xerostomia</li> <li>• Microstomia</li> <li>• Synechiae between the lips and the gums</li> <li>• Poor hygiene</li> <li>• Carious lesions</li> </ul>	<ul style="list-style-type: none"> <li>• Instructions for tooth-brushing and use of chlorhexidine mouthwash</li> <li>• Dietary advice</li> <li>• Symptomatic treatments for oral dryness</li> <li>• Supragingival scaling</li> <li>• Gingivectomy</li> <li>• Tooth extraction</li> <li>• Implant treatment</li> <li>• Restorative dental treatment</li> </ul>

**Case report**

Two brothers (S. seven years old and E. three years old) diagnosed with a moderate form of KS, were treated in 2009 for dental care needs. The genetic analysis revealed a homozygous FERMT1 gene mutation g.80929\_89169del. Their mother presented with the same mutation but was heterozygous for this gene. The father was not affected. With regard to general health status, S and E had urethral stenosis. S. also suffered from a hydrocele of moderate size on the right and a tendency to develop ingrown toenails due partly to poor nail trimming. He suffered from hyperhidrosis. E. was undergoing treatment with an angiotensin-converting enzyme inhibitor (enapril maleate) because of his proteinuria. They had no other health problems, in particular no anemia, no bone abnormalities, no gastrointestinal tract lesions and no eye impairments except moderate photosensitivity. No allergies were present, and no growth failure was noted. Mental development was not impaired.

With regard to their oral habits, food was served three times a day, with a varied diet and little snacking. Tooth sensitivity was exacerbated by cold. S. brushed his teeth every morning and evening, E. brushed only in the morning. Difficulties were reported in the anterior part of the mouth, caused by severe pain and bleeding gums, complicated by lesions on the skin of the hands (figure 1A) where there were severe erosive lesions with the presence of blisters and scars left by blisters. Despite the advice given by the medical dermatologist, no photoprotection or emollients were used.

The perioral examinations of the two brothers showed erosive lesions, blisters, and scars were detected on the neck, around the mouth, on the bridge of the nose, and at the labial commissures.

There was noticeable pigmentation of the lips as well as erythematous zones around the corners of the mouth with angular cheilitis (Figure 1).

The key points of the dental, periodontal and mucosal approaches are summarized in Table 2.

*Case one:* Since the age of seven, S. has been seen regularly for dental management. Difficulties in maintaining good oral hygiene were observed in relation to the difficulty of using a toothbrush and oral pain due to blisters on the mucosae.

He presented with carious lesions on the primary molars. His lack of cooperation with dental care proved very difficult in the first appointment and we decided to avulse the primary teeth with local anesthesia in several sessions. We performed anesthesia with a topical xylocaine 2% gel followed by a prolonged, deep injection into the tissue to avoid blister formation using a periapical infiltration technique. Hemostasis was obtained by moderate compression with wet compresses. Post-operative advice emphasized the risk posed by biting the lips. No space-saving appliance was accepted.

At ten years old, S. started orthodontic treatment for Dento-Maxillary Disharmony. Clinical examinations showed a short, U-shaped arch, a small space for the two permanent maxillary canines (14mm), mesial positioning of teeth in the maxillary area, a lack of space for the development of the two left mandibular premolars and the first right mandibular premolar, and crowding of 14mm. The radiological examination (Figure 2) confirmed that the maxillary canines were impacted, that the eruption of three mandibular premolars was blocked and that four wisdom teeth germs were present.

Table 2: Key points in the dental, periodontal, and mucosal approaches for Kindler syndrome

Management	Dental Approach	Periodontal Approach	Mucosal Approach
<b>BEFORE COMMENCING ANESTHESIA</b>	<ul style="list-style-type: none"> <li>✓ Vaseline on lips and all instruments</li> </ul>		
	<ul style="list-style-type: none"> <li>✓ Topical anesthetic gel + local anesthesia</li> </ul>		<ul style="list-style-type: none"> <li>✓ Post-operative advice to reduce risk of self-inflicted lip trauma</li> </ul>
	<ul style="list-style-type: none"> <li>✓ Prolonged injection with deep placement</li> </ul>		
<b>RESTORATIVE SESSION</b>	<ul style="list-style-type: none"> <li>✓ All conventional restorative materials</li> <li>✓ Restoration perfectly adapted and polished</li> <li>✓ Avoid use of</li> <li>✓ air-water syringe</li> <li>✓ <b>▲</b> Photopolymerization: Specific protective glasses</li> <li>✓ Fluoride varnish</li> </ul>		
<b>PERIODONTAL MANAGEMENT</b>		<ul style="list-style-type: none"> <li>✓ Monthly monitoring</li> <li>✓ Scaling polishing ok with abundant irrigation</li> <li>✓ Prophylactic cleaning with polishing brushes and chlorhexidine mouthwash</li> </ul>	
<b>AVULSION</b>			<ul style="list-style-type: none"> <li>✓ Moderate compression with wet compresses for hemostasis</li> </ul>
<b>ORTHODONTIC TREATMENT</b>	<ul style="list-style-type: none"> <li>✓ As simple and short as possible</li> <li>✓ Orthodontic wax on brackets</li> <li>✓ Pansoral gel®</li> </ul>		
<b>END OF SESSION</b>	<ul style="list-style-type: none"> <li>✓ No material residues in the sublingual region and the vestibule</li> </ul>		<ul style="list-style-type: none"> <li>✓ Blisters drained with a sterile anesthesia needle</li> </ul>
<b>GENERAL ADVICE</b>	<ul style="list-style-type: none"> <li>✓ Cheek retractors rather than mirrors</li> <li>✓ Avoid lateral traction</li> <li>✓ No strong suction</li> <li>✓ Aspiration cannulas applied against a cotton roller</li> </ul>		
<b>INSTRUCTIONS REGARDING ORAL HEALTH</b>	<ul style="list-style-type: none"> <li>✓ Parents help with brushing</li> <li>✓ Toothbrush with small head/soft and short bristles</li> <li>✓ Soak toothbrush in hot water before brushing</li> </ul>		

Figure 1: localization of lesions: the skin of the hand (A) perioral (B)



The orthodontic treatment consisted in avulsing the first four premolars and applying braces to *straighten the teeth* and to preserve space for the canines. Pain management required orthodontic wax and the prescription of an oral gel (*cetalkonium chloride 0.01 %*, *choline salicylate 8.7 %*). It was reported that many brackets had come loose. This could be explained by enamel dysplasia.

Three years later, the clinical examination carried out at the end of orthodontic treatment showed enamel hypoplasia of all teeth with partial lack of occlusal enamel on the first permanent molars (Figure 3C). We also noted intra-oral blisters on the attached gingiva of the maxillary central incisors, and on the mucous membrane of the palate which also white lesions. The gums were erythematous and edematous. Periodontal sounding was associated with gingival bleeding. There was an extensive band of dental plaque around the cervical zone of the teeth, but there was no calculus (Score of 2 on the Silness-Löe plaque index). It was difficult to perform a radiological survey because mouth opening was limited to 37mm due to microstomia. We were unable to finish despite the use of small X-ray sensors, and Vaseline (petroleum jelly). The pain induced was too great. An orthopantomogram was performed to complete the X-ray examination (Figure 3A).

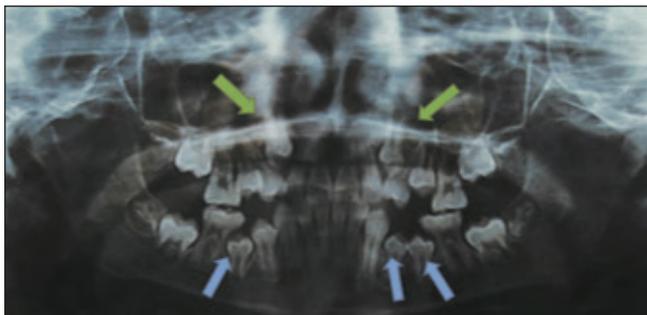
We advised S. of exercises to try to reduce his microstomia by gentle stretching of the mouth, repeated opening / closing movements, to be performed every day and half an hour before his appointment with the dentist.

At 13 years old, S. underwent an operation to dilate the urethra. A regular dental follow-up was medically indicated. Due to the moderate nature of the oral effects, no modification of the treatment plan was considered, only precautionary measures were implemented. Follow-up is provided by a pediatric dentist.

Before each session we took care to lubricate the lips and all instruments with Vaseline to reduce adhesion. The aspiration cannulas were pressed against a cotton roll to keep them away from soft tissue. No strong suction was used. We avoided lateral traction as much as possible, preferring light pressure by compressive force. When it was necessary to pull on the mouth, we used cheek retractors rather than a mirror. After each session, we checked for blisters.

This always involved cautious prophylactic cleaning using polishing brushes and alcohol-free 0.12% chlorhexidine gluconate mouthwashes and the application of a fluoride varnish to reduce sensitivity during meals. Immediate relief was reported by the patient. Indication for partial or total covering of the teeth concerned will be re-assessed when the patient is fully grown. At the moment, areas of exposed dentin could not be covered without creating a permanent vertical over-bite of the molars.

**Figure 2: Radiographic examination of S. at ten years**



During monthly checks, we noted a regression of the gingivitis. Plaque removal was performed efficiently by the patient. The gums retained a slightly edematous, red appearance. Fifteen small blisters were present on the palate. At the age of 14, we noticed a break of six months in follow-up. When the patient returned, his gums were very inflamed and edematous. We noted spontaneous bleeding during the clinical examination and brushing (score of 3 on the Löe-Silness Gingival Index). There was gingival recession (Miller's Class III Classification) at the lower right central incisor, with plaque accumulation (Figure 4A) and a significant presence of calculus around the mandibular incisors. No sensitivity was reported. We were also able to notice mild bone loss here, and an X-ray revealed subgingival calculus (Figure 4B). We performed scaling and polishing under abundant gentle rinsing, and applied fluoride varnish with great caution. Despite our precautions a blister developed on the lip, which we

**Figure 3: Clinical and radiographic (A) examination of S. at ten years: Enamel hypoplasia of the maxillary incisors (B) on the first permanent molar (C).**



drained with a sterile anesthesia needle. We repeated our advice concerning oral hygiene, in particular regarding the need for regular periodontal monitoring in order to achieve good periodontal health. We prescribed a plaque disclosing solution, a very flexible surgical toothbrush (7 / 100th), and chlorhexidine gel (Elugel®) for gentle application to the area of recession at the lower right central incisor, for seven days.

One month later, oral hygiene had improved, and the plaque level and the gingival index were normalized. We suggested that the gingival recession be treated. The patient did not follow up and did not show up for his check-up appointments.

*Case two:* E.'s dental health has also been monitored since the age of three. Like his brother, due to the moderate nature of the oral effects, no modification of the treatment plan was considered, only precautionary measures were implemented. Follow-up is provided by a pediatric dentist. Before each session we took care to lubricate the lips and all instruments with Vaseline. The aspiration cannulas were pressed against a cotton roll to keep them away from soft tissue, no strong suction was used. We extracted his first lower right primary molar because of an overly deep carious lesion.

At the age of nine, he presented with hypomineralization and punctiform hypoplasia of the central maxillary incisors (Figure 5) and blisters on the attached gingivae of the left central and

lateral maxillary incisors. We could see a blister on the lower lip. There was severe gingivitis (Score of 2 on the Löe-Silness Gingival Index) (Figure 5) around the anterior maxillary teeth, and moderate gingivitis (Score of 1 on the Löe-Silness gingival index) in other areas. Passage of the probe caused gingival bleeding. We noted a significant amount of dental plaque but no calculus.

Oral hygiene motivational sessions were repeated, but six months later we noticed the presence of cervical demineralization and an occlusal carious lesion of the enamel on the permanent maxillary molars. We treated this lesion by debridement after application of a topical xylocaine 2% anesthetic gel and periapical anesthesia (epinephrine 1:200,000 anesthetic carpule) and restoration with resin-modified glass-ionomer cement. The patient reported discomfort during photopolymerization of the restorative material, due to photosensitivity. We offered to reduce his discomfort by providing him with specific protective glasses. Particular attention was paid to polishing the restoration and we checked for the absence of material residues in the sublingual region and in the vestibule. There was profuse gingival bleeding. We performed prophylactic cleaning with an alcohol-free 0.12% chlorhexidine gluconate mouthwash and a toothbrush, descaling the maxillary and mandibular incisors and performing topical application of fluoride varnish. We repeated the encouragement to maintain oral hygiene.

At the age of ten, there was a break of 6 months in follow-up. On his return, E. had severe gingivitis related to dental plaque (Score of 2 on the Löe-Silness Gingival Index). His teeth were unbrushed, especially in the maxillary incisal block. We performed prophylactic cleaning with a brush and polishing paste, and a fluoride varnish (Fluor protector® 7100 ppm) was applied to all the teeth. We repeated the instructions regarding oral hygiene. Explanations were given to the patient regarding the need for regular periodontal monitoring in order to achieve good periodontal health. His mother must help him with toothbrushing.

**Figure 4: A. Gingival recession at tooth 41 B. Retroalveolar X-ray anterior region of the mandible**



**Figure 5: E., nine years old: Intraoral view of the maxillary incisors with enamel hypoplasia and stage two gingivitis (score of 2 on the Gingival Index)**



## DISCUSSION

The prevalence of Kindler syndrome is unknown. This rare skin disease affects all populations with no racial or sexual differences. The known features of this disease are grounded in case reports published in the literature. Since 1997, 24 case report articles have appeared in the PubMed database, reporting on a total of 86 cases. The oral manifestations found most frequently are gingivitis, periodontitis, and oral ulcerations, and xerostomia and microstomia have sometimes been described. Kindler Syndrome is characterized by weakness of the periodontium<sup>1</sup>. The two brothers affected by KS presented with all the oral disorders described in the literature: lesions on the perioral tissues, the oral mucosa, the periodontium and carious lesions. They also presented with enamel defects.

Perioral sites around the mouth, on the labial commissures, on the lips, have been described in the literature. It is likely that the microstomia was caused by a constant process of blister formation and scarring. The modification of oral epithelium architecture induces scarring at the labial commissure and loss of vestibular space. Access to the oral cavity is complicated and this contributes to the difficulty in maintaining good oral hygiene, increases the risk of carious lesions and affects comfort during dental treatment. No study has demonstrated an optimal solution while techniques have been proposed to improve mouth opening: the occasional use of resin pads, daily exercises with wooden spatulas, combined with gentle stretching of the mouth, by repeated opening-closing movements and the intermittent use of resin pads<sup>1</sup>.

The oral mucosa is one of the areas that is most frequently affected in children with KS<sup>3</sup>. All its surfaces can develop lesions such as blisters and erosions, ulcerations, erythema and atrophy<sup>1</sup>. All these forms were found in our patients. The palatal mucosa is the second most common site of lesion formation.

KS is particularly characterized by a greater predisposition to periodontal disease, which occurs early and progresses faster than in the general population<sup>2</sup>. The two brothers presented with the initial symptoms of periodontal disease: edematous and erythematous gums, with bleeding and dental plaque. Dental management needs to focus primarily on periodontal monitoring<sup>1</sup>. Initial periodontal therapy is essential, and it must be continued to be applied regularly. Indeed, we managed to stabilize a relatively good level of periodontal health during regular follow-up sessions. We observed a rapid, significant worsening of the periodontitis after a 6-month break in follow-up, with recession of the gingiva of the lower right central incisor for S. This deterioration complicates the management and maintenance of S's periodontal health. Gingival recession is caused by the lack of keratinized gingiva and is aggravated by the difficulties in maintaining oral hygiene. In these patients, complete and extensive destruction of the keratinized tissues can occur in a relatively short period of time, due to the constant cycle of mucosal lesion formation, with the development of blisters, then scars. Therefore, we think that it is essential to organize a monthly follow-up to maintain periodontal health in patients with Kindler Syndrome with the use of prophylactic cleaning and polishing techniques. Traditional non-surgical periodontal treatment has beneficial effects. Ultrasound should be used with care to avoid bleeding and blister formation. If blisters appear, they will need to be drained to avoid expansion of the lesion<sup>1</sup>. Extensive irrigation is necessary to avoid the heating of tissues, requiring the use of a suction cannula, which

can increase the risk of mucosal trauma. However, it is difficult for patients to maintain sufficient motivation to carry out the diligent care procedures which are essential for their periodontal health. This demotivation is immediately penalised by a deterioration in the periodontal condition and complication of management. No treatment of choice for gingival recession using a surgical approach is indicated in the literature. One case report describes periodontal surgery in a patient with inherited epidermolysis bullosa, which was not Kindler Syndrome<sup>4</sup>. Therefore, there is no consensus in the literature. We think, however, that periodontal surgery techniques pose a real risk of causing blister formation in these patients. The periodontal type limits choice of technique. The risk of blistering during preparation of the operating site seems high to us. This should be done using a pad rather than by rubbing with a compress. There is also a risk when using aspiration cannulas which can cause desquamation of the mucosae, during suturing and even throughout surgery when the tissues are held under lateral traction. The palate is also at high risk of blister formation and is the second most common site in this respect. Moreover, Budunelli indicates in his case that it is better to choose a single surgical site, to avoid causing blister formation in two areas<sup>4</sup>.

The prevalence of caries is significantly higher in patients with inherited epidermolysis bullosa than in the general population. The etiology is multifactorial, but the difficulty experienced by Kindler Syndrome patients in achieving good oral hygiene is particularly noted; it is difficult for their hands to hold the toothbrush properly, which leads to difficulty in brushing effectively, in the mechanical control of plaque, whilst blisters make brushing the teeth painful<sup>1</sup>. All conventional restorative materials can be used for restorative dental treatment. The choice is made according to the possibility of isolation, risk of caries, and socio-economic factors specific to each patient. Most of the recommended treatments use glass ionomer cement and preformed pedodontic caps, that are tried and tested methods of tooth restoration<sup>5</sup>. The restorations were perfectly adapted to the teeth and polished to prevent injuries, blisters and ulcerations<sup>1</sup>. Photosensitivity is a specific feature of Kindler Syndrome. Treatment was performed under local anesthesia, which is the best technique for moderate forms of inherited epidermolysis bullosa<sup>1</sup>. Topical anesthetic gel was used. To prevent blister formation, the anesthetic solution was injected slowly into the tissues with deep placement. Postoperative advice was given to prevent biting or trauma to the lips that may occur under local anesthesia<sup>1</sup>.

The goal of therapy is the prevention of caries and mucosal damage. Maintaining a functional dentition reduces the risk of injury by more effective chewing<sup>1</sup>. Despite the pain, toothbrushing must be performed, using a toothbrush with a small head and soft, short bristles<sup>3</sup>, softened by soaking in hot water<sup>1</sup>. Mouthwashes can be administered for one minute, once a day, for a month. Chlorhexidine 0.12% does little to prevent decay, when there is insufficient brushing, its antiseptic action could be helpful. It could be used as mouthwash, as well as in the form of gels or swabs<sup>1</sup>.

Both brothers presented with minor enamel defects, which are reported in all subtypes of inherited epidermolysis bullosa<sup>2</sup>, but E. presented with a rare form of enamel hypoplasia. These enamel defects take the form of a pit or groove on the crown and cause the retention of food fragments which leads to the development of caries. The prevalence of enamel defects in Kindler Syndrome

is similar to that in the general population (27%)<sup>3</sup>. The enamel hypoplasia present was therefore possibly independent of Kindler Syndrome. As recommended, topical fluoride was applied at each appointment in order to decrease tooth sensitivity.

S. received orthodontic treatment between the ages of 10 and 13 years. In these patients, the combined effects of malnutrition and mucocutaneous scarring leads to the inhibition of facial growth, and possibly dentoalveolar disharmony and dental malposition<sup>1,6</sup>. Because of possible gingival weaknesses, orthodontic treatment plans must be simplified and be of as short a duration as possible, with increased periodontal maintenance<sup>6</sup>. The difficulties encountered during orthodontic care are related to the use of orthodontic brackets and wires, which can cause ulceration of the mucous membranes. To avoid these injuries, orthodontic wax can be applied to the brackets<sup>1,6</sup>.

Selective extractions of the first premolars can be programmed. These are not contraindicated but a benefit-risk analysis must be carried out. These procedures should ideally be performed under local anesthesia. Blood tests for anemia should be performed if possible. An atraumatic technique should be used, and mucosal incisions should be clean to prevent the formation of blisters<sup>1</sup>. Hemostasis should be performed using moderate compression with wet compresses to avoid tissue adhesion<sup>1,5</sup>. Stitches may be inserted, but they must be placed so as not to irritate the mucous membranes<sup>20</sup>.

## CONCLUSION

Thus, the follow-up of these two children diagnosed with a moderate form of KS shows that, in their case, dental care is tricky and requires monthly monitoring. Periodontal maintenance therapy is essential, so the absence of surveillance leads quickly to the recurrence of gingivitis. This demotivation is immediately penalized by deterioration in the periodontal condition and complication of management. Therefore, it is essential for the dentist to be part of the care team. However, all restorative, endodontic, surgical, periodontal and orthodontic treatments should be performed with appropriate precautions for hemostasis and should aim to avoid as much mucosal damage as possible.

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## Teledentistry Platforms for Orthodontics

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*Technology has transformed almost every aspect of our lives. Smartphones enable patients to request, receive, and transmit information irrespective of the time and place. The global pandemic has forced healthcare providers to employ technology to aid in ‘flattening the curve. The Novel Coronavirus, which is responsible for COVID-19, is transmitted primarily through person-to-person contact but may also be spread through aerosol generating procedures, so many clinics have severely limited interpersonal interactions.<sup>1</sup> The purpose of this article is to provide helpful information for those orthodontists considering some form of remote practice. Various HIPAA-compliant telecommunication or teledentistry systems that can be used for orthodontic treatment are introduced and discussed. Detailed information about each platform that can potentially be used for orthodontics is provided in Figure 1. The authors do not endorse any of the products listed and the included software is not all inclusive but instead is a glimpse into the options available.*

**Keywords:** Telecommunication, Teleconsultation, Patient Management, Artificial Intelligence (AI) assisted

### INTRODUCTION

Teledentistry, a subset of telehealth, is the transmission of a patient’s dental health information via telecommunication platforms either asynchronously or synchronistically to diagnose and provide advice about treatment over a distance.<sup>2</sup> Asynchronistic software uses archived health information including radiographs, photographs, video and digital impressions which are analyzed by a doctor at some future time.<sup>3</sup> Synchronous telecommunication employs a live video chat in which the patient is directly examined.<sup>3</sup>

In orthodontics, advancement in technology and the need for social distancing due to COVID-19 has aided in transitioning in-person consultations and monitoring to virtual patient encounters where the patient is not physically in the office.<sup>4</sup> In addition, some patients prefer orthodontic treatment with less frequent in-office visits.<sup>4,5</sup> It has been postulated that the adoption of virtual check-ups to supplement chairside appointments could have a positive economic impact on orthodontic offices by decreasing chair time.<sup>2,5</sup> Teledentistry can be particularly useful when monitoring, leveling and aligning; evaluating maxillary expanders; checking patient cooperation with elastics; and guiding parents with minor emergencies that can be handled at home.<sup>5</sup>

Currently, three main forms of virtual communication technology are used in orthodontics: virtual consultation or live chats, treatment progress photo uploads by patients for orthodontic review at a later time, and artificial intelligence (AI) assisted treatment monitoring with photos or videos taken by patients. Various HIPAA-compliant telecommunication or teledentistry platforms are available for orthodontic use (Fig. 1).

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### Telecommunication Tools for Orthodontics

All communication about a patient’s medical information must be Health Insurance Portability and Accountability Act (HIPAA) compliant. For a telehealth service to be considered HIPAA compliant, it must have all the necessary controls to ensure that Personal Health Information (PHI) is sent securely, and must include access controls, audit controls, and full end-to-end encryption.<sup>6</sup> Some readily available web conferencing software such as Zoom, Google Meet, GoToMeeting, Skype, BlueJeans, Microsoft Teams, and ReadyTalk offer HIPAA compliant versions for healthcare professionals. Since most of these programs were not originally designed for medical or dental use, their functions are generally limited to live virtual consultation, a chat feature, and document transfer without additional features commonly found in systems built especially for medical or dental purposes. Other popular platforms such as Apple FaceTime, Facebook Messenger video chat, and WhatsApp lack HIPAA compliance.<sup>6</sup> All healthcare providers working with an outside vendor that handles PHI (this includes telecommunication platforms) are required to have a Business Associate Agreement (BAA) in place to ensure that these third-parties will only use PHI in a secure and established manner.<sup>7</sup>

While the standard version of Zoom is not HIPAA compliant, Zoom for Healthcare is HIPAA compliant, uses bank-grade 256-bit Advanced Encryption Standard (AES) encryption, and provides a signed BAA for a starting cost of \$200 per month with a 12-month commitment for 10 hosts.<sup>8,9</sup> Zoom for Healthcare also provides encrypted chats, dual screen-share, document transfer, live video recording, and screenshot functions.

Google Meet offers HIPAA compliant teleconsultation services through paid “G Suite” accounts starting at \$6 per month for each Gmail account the company uses.<sup>10</sup> The BAA (G Suite/Cloud Identity HIPAA Business Associate Amendment) can be reviewed and accepted through the Google Admin console page (Fig. 2).<sup>10</sup> This BAA also covers many other widely used Google services including Gmail, Google Calendar, Google Drive (including Docs, Sheets, Slides, and Forms), Google Hangouts (chat messaging feature only), Google Chat and many more.<sup>11</sup> For orthodontists and offices already familiar with Google services, the software can be utilized in numerous useful ways. Google uses 128-bit or stronger AES to protect data in transit and in its data centers.<sup>12</sup>

When using these universal telecommunication platforms for orthodontic purposes, care should be taken by the providers and their staff to adhere to the HIPAA Minimum Necessary Standard by properly configuring the settings and following compliant use

of the platform.<sup>13</sup> Many companies provide detailed guidelines for their customers to follow in order to ensure that the platform is used compliantly.

### Teleconsultation for New Patient Encounters

There has been an increase in the development of telecommunication tools designed specifically for dentistry.

Currently, a number of companies such as Dentulu and Toothpic provide software for limited dental conferencing. These programs allow a prospective new patient to seek out a provider virtually for a diagnosis or recommendation based on the photos they provide. The doctor sets his/her availability and allows patients to request an appointment similar to how UBER lets its employees set their own work schedules according to their availability. With this software, patients pay for the provider’s time and advice with the platform receiving a portion of the revenue generated.<sup>14,15</sup> The provider is not required to give definitive treatment but can invite patients to their office if desired.<sup>15</sup> While the American Association of Orthodontists believe that treatment should not begin prior to an in-person evaluation, these platforms allow orthodontists to initiate a discussion with patients about their treatment and to provide education regarding the orthodontic process.<sup>16</sup> While this type of teledentistry software is geared towards generating an extra revenue stream rather than as an alternative to in-office visits for regular patients, orthodontists can still use it to allow their patients to talk directly to them for a fee.

Figure. 1. Various teledentistry platforms for orthodontics.

Legend:	Telecommunication			Teleconsultation						Patient Management			AI
	Zoom Healthcare	Skype	Google Meet	Toothpic	Dentulu	Smile Virtual	Review Tool	Smile Snap	Rhinogram	Carestack	TeleDent	Teledentix	Dental Monitoring
<b>1. Privacy</b>													
HIPAA compliant/ Provides BAA	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green
<b>2. Online platform functions</b>													
Compatible with any phone brands/ computer processor	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green
Cloud back-up	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green
Screenshare	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green
Recording virtual consultation	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green
Live Chat (synchronous)	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green
Asynchronous Messaging	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green
Asynchronous photo-uploading/viewing	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green
Designated photo template	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green
Payment collection	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green
Self-scheduling	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green
Share documents and obtain signatures	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green
Customizable patient reminders and alerts	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green
<b>3. Practice management and marketing</b>													
Generates leads	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green
Generates reports	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green
Clinical data sharing with other providers	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green
Compatible with management software (e.g. Dolphin)	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green
Customizable: Dr.'s Name / logo	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green
<b>4. Advance Technology</b>													
Photo-taking device provided	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green
Compatible with intraoral camera	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green
Treatment outcome projection	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green
AI-assisted evaluation of pictures	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green	Green
<b>5. Cost</b>													
Cost	\$	Call Rep	\$	+\$	+\$	\$\$\$	\$	\$\$	Call Rep	\$	\$\$	\$\$\$	\$\$\$\$

Other platforms are available for virtual treatment consultations. Smile Virtual, Review Tool and Smile Snap are three of the teledentistry companies that allow dentists to asynchronously prescreen and recruit new patients virtually. These platforms generate leads with customizable landing pages that are integrated directly into the office website.<sup>17-19</sup> The patient uploads a few requested photos to the platform via the dentist’s website and shares concerns and desires regarding their teeth. The orthodontist reviews the information and records a personalized video consultation. After the patient views the virtual consultation, they can schedule a follow-up appointment.<sup>17-19</sup>

Smile Virtual is a tool for boosting new patient traffic and conversion rate for cosmetic dental procedures and orthodontic treatment, claiming an 87% treatment conversion. It has the additional feature of providing treatment outcomes of individuals with similar conditions.<sup>17</sup> Smile Virtual requests two patient-provided photos that are minimally diagnostic. The photos are best for brief treatment proposals but are not ideal for long-term patient monitoring. Review Tool requires five standard photos for initial orthodontic records<sup>17,18</sup> and provides synchronous consults through third-party platforms Zoom, FaceTime, Skype, iMessage and Gmail. With the built-in live video consult features, Smile Snap offers asynchronous and synchronous options for patients and is intended to be a communication tool both for new recruits and treatment monitoring. All three applications provide analytic tools to monitor conversion rate.<sup>17-19</sup>

Rhinogram is similar but comes with some very distinct features.<sup>20</sup> It allows for HIPAA compliant patient-to-practice texting for both new and existing patients. When a patient is interested in treatment, they send a standard text to the practice which responds in turn with a completely customizable reply through the app. It could be to schedule an appointment, request photos or offer a virtual appointment. No software download is required by the patient, as all communication including the virtual call and sending of secure legal documents is conducted via text or Facebook Messenger on the patient’s smartphone. With Rhinopay, it is possible to help collect balances.

## Teledentistry for Patient Management

A limiting factor of the previously mentioned teledentistry software is the inability to remotely manage the practice and patient data out of office.

TeleDent was created by MouthWatch and provides a secure means of synchronistic or asynchronistic remote consult with established patients and is currently used in many public health and dental school settings. It was designed for both solo and multi-practice use and is fairly user friendly.<sup>21,22</sup> The patient can easily send and receive pictures following the company-provided instructions that are individually sent out by the office. While it has the ability to be used on a mobile device, its best use is on desktop computers. It does not sync with any of the major orthodontic management software, so patients need to be manually entered into the software and pictures must be downloaded and reuploaded. There is no limit on the number of patients and each license covers ten providers.

Teledentix is a virtual practice management software built for teledentistry. Patients can use it to complete and sign forms, make payments, and receive health education. Patient restoration chartings, images, periodontal chartings, treatment plans, treatment progress and progress notes can be uploaded to the dashboard. In addition, there is a tab for medical records, prescriptions, patient education materials, and appointment history. With Teledentix, available hours can be set by the practice, thus enabling patients to book their own appointments. The provider can send, track and store referrals with a network of providers.<sup>23</sup> Teledentix can potentially be a one-stop shop for managing a practice remotely.

Carestack is primarily a virtual practice management software with some telecommunication capabilities. Carestack provides access to an odontogram for basic dental charting, note taking capability, individual treatment plans for patient review, and portals for signing legal documents.<sup>23,24</sup> Compared to other dental management software, providers can also conduct virtual sessions with patients in which patients can schedule a live consultation. Asynchronous options are limited on Carestack. Dentists can customize

Figure. 2. Step-by-step guide for accepting BAA with Google for paid G suite business account owners.

**Step 1.** Log into the Google Admin console and click on "Account Settings."

**Step 2.** In the Account Settings page, click on "Legal and Compliance."

**Step 3.** In the Legal and Compliance page, scroll to Security and Privacy Additional Terms. Click "G Suite/Cloud Identity HIPAA Business Associate Amendment" to view the amendment. After review, click "Not Accepted."

**Step 4.** Then click "Review and Accept."

**Step 5.** Answer all three questions, and if you are confirmed as a HIPAA covered entity, click "I accept" to accept the HIPAA BAA.

HIPAA Compliance with G Suite/Cloud Identity	
1. Are you a Covered Entity (or Business Associate of a Covered Entity) under HIPAA?	Yes
2. Will you be using G Suite/Cloud Identity in connection with Protected Health Information? (Note that a BAA is only available for services listed in the vHSA included functionality. Any other services must not be used in connection with PHI.)	Yes
3. Are you authorized to request and agree to a Business Associate Agreement with Google for your G Suite/Cloud Identity organization?	Yes

text messages to patients with a link that lets them view and pay their invoices. Additionally, Carestack plans to automate software to collect outstanding balances through pay-by-text and intelligent payment reminders.<sup>24</sup>

### AI-assisted Intelligent Oral Monitoring for Orthodontics

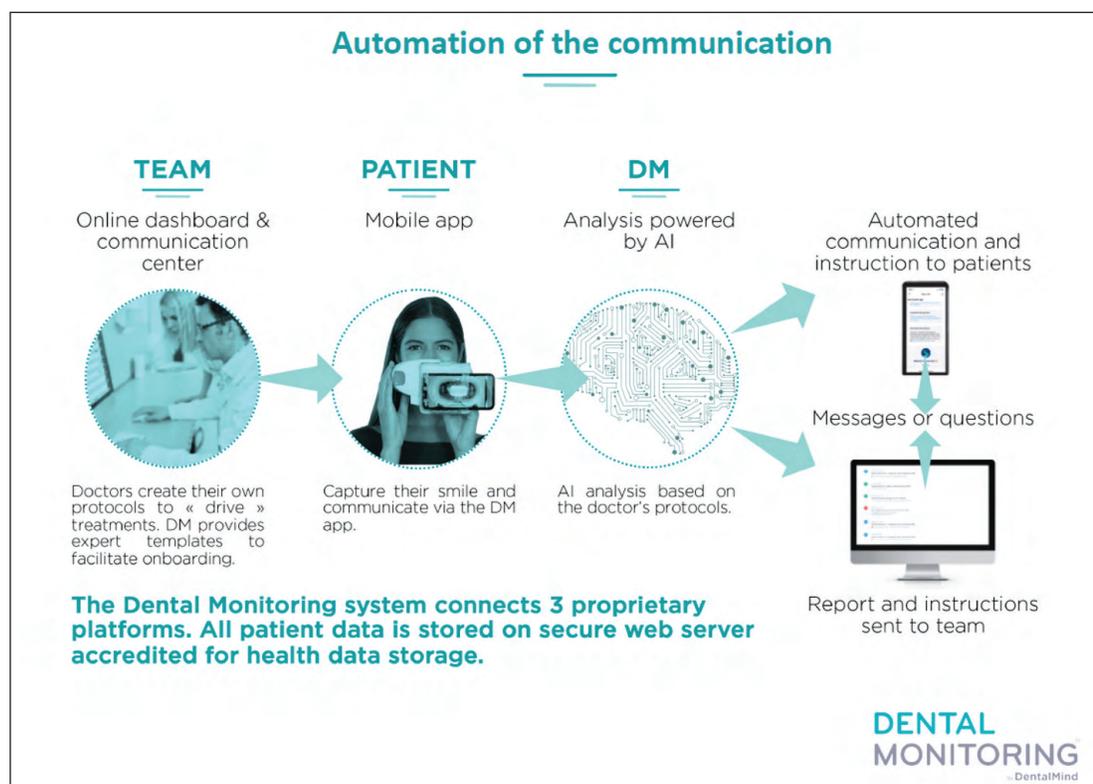
In an effort to reduce both the orthodontist's workload and the patient's chair time, Dental Minds has combined AI and teleorthodontics technology to provide a unique intelligent oral monitoring system called Dental Monitoring.<sup>25,26</sup> It uses AI to allow orthodontists to monitor orthodontic tooth movements and their patients' dental conditions remotely. The system works in three interconnected platforms: a smartphone application for patients, a patented tooth movement tracking algorithm, and an online doctor dashboard on which orthodontists can view patient treatment progress and posttreatment changes (Fig. 3). Dental Monitoring uses photographs or video scans taken and uploaded by patients with their smartphones with the assistance of its proprietary scan box, cheek retractor, and smartphone application. Dental Monitoring's patented AI system then remotely verifies and tracks the movement of each tooth with both fixed appliances and clear aligners. In addition, it can alert providers of common orthodontic conditions including distorted or broken brackets, worn-down or missing attachments, the condition and position of separators or bands, archwire engagement, and unseating of aligners. Dental Minds claims that it can monitor almost 180 dental conditions.<sup>26</sup> An automated communication system sends either an automated communication with instructions to the patient or a report to the orthodontic team for further analysis. Orthodontists can customize what information they see for each visit and under what conditions they are alerted.

In addition, its Smile Mate app uses self-uploaded pictures of patients' teeth, allowing the AI to detect and report notable dental and orthodontic conditions to the orthodontist. It does not have data storage functions but helps with synchronous new and existing patient consultations and integrates with a third-party app, Calendly, for scheduling activities. Dental Monitoring's Vision software uses AI for a more realistic orthodontic treatment projection to assist patients with treatment decisions. Its most novel technology enables reconstruction of three-dimensional (3D) digital models from these intraoral images by stitching them with the pretreatment CBCT and intraoral scan files for more accurate AI monitoring and 3D evaluation of tooth movements (including root movements). While the diagnostic accuracy of its AI-assisted dental monitoring and image reconstruction process still needs to be verified, Dental Monitoring is the only platform reviewed that has the capability of full AI-assisted monitoring.

### DISCUSSION

When selecting a teledentistry platform for an orthodontic practice, it is important to match the practice needs with the intended purpose and required functions of the software. Telecommunication platforms are great for general synchronous virtual communication but lack asynchronous communication options and are not designed specifically for dental practices. Teleconsultation platforms built for virtual consultations are not ideal for long-term patient care, but instead are designed to increase patient leads. Teledentistry software for patient management are well designed for long-term patient management and multipractice patient management. These platforms are comprehensive in that they provide some level of practice management and virtual patient interaction, but they do not sync with other orthodontic

Figure 3. Automation of communication with intelligent oral monitoring by Dental Monitoring.



practice management software. If an orthodontic office simply needs to incorporate virtual consultations with a photo-uploading feature, Smile Virtual, Review Tool, Smile Snap or Rhinogram are good options. Intelligent oral monitoring for orthodontics is completely unique in that artificial intelligence is used to help detect orthodontic treatment complications and tracking.

Ease of use for providers and patients is also a very important factor. If the platform is unfamiliar or difficult to use, there will naturally be less motivation to use it. Most telecommunication software is user-friendly since many of the applications are already widely used in various other applications. Teledentistry platforms built for virtual consultations are also very user-friendly on the patient end, but less so on the provider end due to the new functions that the staff must be introduced to. In general, teledentistry software for virtual dental management and intelligent oral monitoring require lengthy instructions for use and more troubleshooting should be expected due to the multiplicity of their functions.

Obtaining a BAA is an essential part of HIPAA compliance. Orthodontists should perform proper due diligence to identify technology vendors that offer “HIPAA compliant” telecommunication. In addition to privacy issues, orthodontic clinicians should consider that inaccurate or insufficient data will lead to misdiagnosis or under-diagnosis, resulting in compromised treatment. Another practical concern is how to efficiently manage the doctor’s and staff’s daily schedules between in-person patient visits and remote teledentistry sessions.

Due to the continual advancement of technology, it is likely that some aspects of this article will become outdated. Technology is always advancing, as are these platforms, so a thorough evaluation of each option should be considered prior to selection. Unfortunately, some information is vague or poorly described by the companies.

## CONCLUSIONS

- Advances in technology, increasing patient demands, and the current public health crisis have driven orthodontists to consider virtual appointments.
- Numerous platforms are available, but each offers different services. A thorough evaluation of intended use of the software should precede any commitment to a service.
  1. Zoom, Google, Skype, etc. have HIPAA compliant versions, but orthodontists must ensure all settings are properly configured.
  2. Dentulu and Toothpic are not suitable platforms for orthodontic patient monitoring or consultations as they are only intended for limited initial evaluations.
  3. Smile Virtual, Review Tools, Smile Snap and Rhinogram are great resources for increasing an orthodontist’s virtual presence while obtaining new leads. The software is not intended for managing patients but can offer a HIPAA compliant means of communication.
  4. Carestack, TeleDent Teledentix are intended for remote management of patients of record. They include many features that aid in comprehensive virtual monitoring.
  5. Dental Monitoring is a unique teledentistry technology developed for orthodontic use. Artificial intelligence is employed to track progress and alert providers of potential treatment complications.
- HIPAA compliance of the system and a BAA provision of the teledentistry platform must be checked to protect personal health information.

## Authors’ Contribution

Authors (Leah Rogowski, Janet H. Kim and Sumayah Al Shami) contributed to the outline, design and research for this manuscript. Authors (Jae Hyun Park and Scott E I Howell) contributed to the direction and editing of this manuscript.

## Conflict of Interest

Scott E I Howell is a clinical advisor for MouthWatch, LLC. The rest of authors do not have any conflict of interest.

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## Accuracy of Model Estimation versus Tanaka and Johnston Arch Length Analysis

Lauren M White\*/ Nathan E Kirk\*\*/ Jeffrey A Dean\*\*\*

**Objectives:** This study examines how accurate pediatric dentists are at estimating dental arch lengths by comparing their model estimations (guesstimating the arch length without measuring) to the Tanaka and Johnston mixed dentition arch length analysis. **Study Design:** This study consisted of two parts, a survey of practitioners and a model estimating and measuring component. The survey was designed and given to 44 pediatric dentists to determine how many were practicing orthodontics and using arch length analyses routinely. Then 18 pediatric dentists and 13 pediatric dental residents examined 20 sets of mixed dentition models and estimated how much space was available. These estimations were compared to the calculated gold standard, the Tanaka and Johnston arch length analysis of the same models. **Results and Conclusions:** More than half of the dentists surveyed that practice comprehensive orthodontics use arch length estimates. Pediatric dentists and pediatric dental residents are just as good as each other at estimating arch length. Pediatric dentists and pediatric dental residents underestimated arch length by  $-3.6$  and  $-3.1$  mm, respectively. More research needs to be done to determine if model estimation is a clinically acceptable way to judge arch length.

**Keywords:** arch length analysis, orthodontics, mixed dentition, pediatrics, Tanaka-Johnston, space maintenance

### INTRODUCTION

Many practitioners are practicing orthodontic treatment in the mixed dentition phase.<sup>1</sup> There are several advantages of early orthodontic treatment such as a decrease in overall cost and prevention of further orthodontic treatment.<sup>2</sup> However, with early orthodontic interception comes the importance of mixed dentition arch analysis. While somewhat time consuming, a mixed dentition arch analysis can help predict the mesial-distal width of the permanent canine and premolars which can reveal tooth-size arch length discrepancies.<sup>3</sup> Overprediction of the permanent dentition arch length may result in undesirable extraction therapy, which can significantly compromise the final orthodontic treatment and facial esthetics.

Many different mixed dentition analyses have been discussed in the literature. These analyses usually fall into one of three categories: measurements from radiographs, measurements from study models, or a combination of both.<sup>3</sup> While not as accurate for more diverse populations, some of the most popular methods are the Moyers and the Tanaka and Johnston which both use study models.<sup>3,4</sup> Studies have suggested that the various mixed dentition analyses have high validity and reliability.<sup>5</sup> The Tanaka and Johnston method is one of the most commonly used in the literature for comparison of new methods as it is universally accepted and relatively simple.<sup>3,5,6</sup> The Tanaka and Johnston method uses the mesio-distal widths of the mandibular permanent incisors from study models to predict the mesio-distal width of permanent canine and premolars accurate at the 75<sup>th</sup> percentile.<sup>7</sup>

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While there is an abundance of literature on the validity of various mixed dentition analyses, little research has been conducted on the number of practitioners that routinely utilize them. Furthermore, little has been published comparing the different accepted mixed dentition analyses versus model estimation via guesstimating without measurement. The aims of this paper are as follows:

- to determine the number of practitioners actively utilizing mixed dentition analyses
- to compare practitioner model estimations to the results from the Tanaka and Johnston mixed dentition analyses for accuracy.
- to determine if model estimations by more experienced pediatric dentists were more accurate than that of pediatric dental residents to evaluate the implications that this may have on treatment planning

## MATERIALS AND METHOD

A survey was given to forty-four pediatric dentists associated with the University of Tennessee Pediatric Alumni and the Indiana University Dental School Faculty to determine the percentage of pediatric dentists who perform arch length analysis and what method of analysis they use. The survey was given after an orthodontic seminar for pediatric dentists. The results of the survey were compiled and analyzed.

To compare the difference between practitioner model estimations and the Tanaka and Johnston method, twenty mixed dentition study models from the orthodontic dental clinic at the Indiana University Dental School were used for the analysis. Eighteen pediatric dentists and thirteen pediatric dental residents evaluated the same twenty study models. Each participant evaluated all twenty study models using a best guesstimate, or estimation method, without knowing the results of the Tanaka and Johnston analysis. Examiner A measured the mesial-distal width of the maxillary and mandibular incisors using a digital Boley gauge and performed the calculations described by Tanaka and Johnston<sup>7</sup>. Examiner A was blinded from the results of the participants. Examiner B performed the same measurements to show repeatability and accuracy. The Tanaka and Johnston analysis calculated by Examiner A were used as the control analysis.

### Statistical analysis

All analyses were conducted for the whole mouth, i.e., data for maxillary and mandible jaws combined, and for each jaw separately.

The repeatability of each examiners measurements was verified by having Examiner A repeat measurements for all 20 models and then Examiner B repeat calculations for 10 randomly selected models. The measurements were analyzed statistically via mean differences and paired t-tests to test the hypotheses of zero mean difference in measurements. The differences between the dentists' and residents' measurements and the gold standard (Examiner A's measurements) were then calculated for each model and categorized as being a clinically acceptable difference, absolute value of the difference was less than two mm. Differences were categorized as clinically acceptable if they were less than two mm as space excess or deficiencies above two mm typically require more extensive treatment planning and early intervention.<sup>8</sup> The frequency of a clinically

acceptable arch length was tabulated, and the mean difference was tested for statistical significance using a paired t-test.

The intra class correlation coefficient (icc) was also estimated using a mixed model with random terms for model and examiner. The icc is the amount of between model variability relative to the total variability. An icc value close to one shows that variability coming from repeated measurements on a model (within model variability) is small relative to the total variability. Total variability is comprised of variability from measurements of different models, from different examiners, and from repeated measurements on the same model.

The differences between the dentists' and residents' measurements versus the gold standard (Tanaka and Johnston measurements) were calculated for each model and categorized as being a clinically acceptable difference, absolute value of the difference was less than two mm. Using this dichotomized response, frequencies were tabulated for each professional type (dentist and resident) for each model. A Cochran Mantel Haenszel statistic (stratified by model) tested the hypothesis of no association between the clinically acceptable difference category and professional type (dentist versus resident).

## RESULTS

### Profile of pediatric dentists practicing orthodontics

Forty-four dentists were surveyed for the type of orthodontia they practice. Mean number of years practicing pediatric dentistry was 24 with a minimum of 5 years and maximum of 38 years. Of the 44 surveyed, 33 (75%) responded that they practice comprehensive orthodontics. The mean number of years of practicing pediatric dentistry for this subgroup was 24.3. Of these 33 pediatric dentists, 91% responded that they take study models during the mixed dentition and permanent dentition stages. Twenty-six of these 33 (78.79%) responded that they perform arch length analysis. Of these 26, the two most used methods of arch length analysis were model estimation and the Moyers method, used by 50% and 38.5% respectively. Many pediatric dentists indicated the use of more than one type of analysis (Table 1).

### Repeatability of each examiner's measurements:

The difference between measurements repeated by each examiner was analyzed for the whole mouth and by jaw. All mean differences by both examiner A and B were less than two mm. Paired t-tests were not significant indicating that repeated measurements by each individual examiner were on average not different from the original measurements thus verifying repeatability of individual measurements (Table 2).

The intra class correlation coefficients of just under one showed that the variability coming from within each model was negligible when compared with the total variability (Table 3).

Because the measurements taken by Examiner A and B were determined to be repeatable across examiners with low variability, measurements from one examiner, Examiner A, were used in the Tanaka and Johnston formula to calculate the predicted arch-length discrepancy. These calculations created the gold-standard for comparison with the estimations by pediatric dentist and residents. The differences in measurements from the gold standard were classified as being a clinically acceptable distance of less than or equal to two mm from the gold standard. For both pediatric dentists and residents, the majority of the estimations were over the clinically

acceptable distance, 78.19% and 73.85% respectively (Table 4). Additionally, the Cochran Mantel Haenszel test showed no association between professional type (dentist or resident) and the category of clinically acceptable difference (Table 5).

**Table 1: Profile of Pediatric Dentist that practice orthodontics**

<b>mean years practicing pediatric dentistry</b>	24.3 years
<b>Timing of study models</b>	
mixed dentition	9%
mixed dentition and permanent dentition	91%
<b>Type of Orthodontics practiced</b>	
comprehensive orthodontics	9.09%
early and comprehensive orthodontics	2.27%
space maintenance	6.82%
space maintenance and early orthodontics	2.27%
space maintenance, early orthodontics, comprehensive orthodontics	2.27%
space maintenance, space regaining	2.27%
space maintenance, space regaining, early orthodontics	13.64%
space maintenance, space regaining, early orthodontics, comprehensive orthodontics	61.36%
<b>Arch length analysis</b>	
use of arch length analysis	78.79%
no arch length analysis	21.21%
<b>Types of Arch Length analysis used</b>	
Model estimation	50.00%
Moyers	38.40%
Tanaka-Johnston	23.00%
Hixon-Oldfather	15.30%
Other	26.90%

**DISCUSSION**

The study revealed many of the pediatric dentists surveyed are practicing some sort of orthodontics. Of those that practice orthodontics, most use an arch length analysis with the most popular method being model estimation. Due to the wide use of estimation, it is important to see how reliable this method is because it can greatly influence how these dentists are planning orthodontic treatment.

The statistical analysis of the data shows that the gold standard calculation, Tanaka and Johnston, is repeatable, reliable, and can be used to examine the clinical acceptability of the model estimations made by practitioners. The results indicated that model estimations by both pediatric dentists and pediatric dental residents were often outside the clinically acceptable range. When looking at the whole mouth (maxillary and mandibular) analysis for overall percent of clinically acceptable distance, dentists were within the clinically acceptable range 26.81 percent of the time and residents were in the clinically acceptable range 26.15 percent of the time. In the by jaw analysis, the maxillary arch percentage of clinically acceptable distance was 16.94 % for dentists and 18.46% for residents, which is much worse than the whole mouth percentages. On the other hand, the mandibular arch measurements were much better, with 63% of the pediatric dentists and 61% of the pediatric dental residents estimating the arch length within the clinically acceptable standard that was set at + or – two mm. That is to say, the dentists were much better at estimating the arch lengths in the mandibular arch than the maxillary arch.

Additionally, the Cochran-Mantel-Haenszel test stratified by model showed there was no association between professional type (dentist or resident) and the category of clinically acceptable difference. Pediatric dentists were no better than pediatric dental residents at predicting arch length which could mean that they do not improve upon their arch-length analysis skills past residency.

Because the present study indicates that most pediatric dentists are using model estimations that are often outside the clinically acceptable range to analyze space in the mixed dentition, it is imperative to determine what implications this has on treatment planning. The space requirements and conditions determined in the mixed dentition phase will often translate to the permanent dentition

**Table 2: Mean Difference for examiners A and B**

	N	Mean difference	Std dev	Std error	Min	Max	Difference	tValue	DF	pvalue
<b>Examiner A whole mouth</b>	40	-0.15	0.62	0.1	-1.52	1.52	y_A-y_A2	-1.58	39	0.12213
<b>Examiner B whole mouth</b>	20	-0.19	0.92	0.21	-1.87	1.65	y_b-y_b2	-0.93	19	0.36167
<b>Examiner A maxillary</b>	20	-0.18	0.67	0.15	-1.52	1.52	Mx_A-Mx_A2	-1.2	19	0.24669
<b>Examiner B maxillary</b>	10	-0.05	1.04	0.33	-1.87	1.65	Mx_B-Mx_B2	-0.16	9	0.87802
<b>Examiner A mandibular</b>	20	-0.13	0.57	0.13	-1.17	0.89	Md_A-Md_A2	-1	19	0.3278
<b>Examiner B mandibular</b>	10	-0.33	0.82	0.26	-1.54	0.88	Md_B-Md_B2	-1.29	9	0.22957

**Table 3: Mean difference between measurements of Examiner A and B**

	N	d <=2	Mean	Std dev	Std error	Min	Max	Difference	tValue	DF	pvalue	Between model var	Between examiner var	Within model var	icc
whole mouth	40	1	-0.04	0.75	0.12	-1.38	1.43	y_A-y_b	-0.3	39	0.7668	17.98	0	0.27	0.99
maxillary	20	1	0.03	0.73	0.16	-1.28	1.43	Mx_A-Mx_B	0.18	19	0.86076	14.68	0	0.25	0.98
mandibular	20	1	-0.1	0.78	0.17	-1.38	1.27	Md_A-Md_B	-0.57	19	0.57341	16.42	0	0.29	0.98

**Table 4: Mean difference and clinically acceptable distance in mm between Participant estimations and gold-standard calculations**

	N	Mean difference	Std dev	Std error	Min	Max	CAD  d ≤2	d >2
Dentist whole mouth	720	-3.6	4.1	0.2	-20.3	7.8	26.81%	73.19%
Resident whole mouth	520	-3.1	3.5	0.2	-12.7	8.9	26.15%	73.85%
Dentist maxillary	360	-4.7	3.7	0.2	-17.4	7.8	16.94%	83.06%
Resident maxillary	260	-4.2	3	0.2	-12.7	7.1	18.46%	81.54%
Dentist mandibular	360	-2.6	4.3	0.2	-20.3	7	36.67%	63.33%
Resident mandibular	260	-1.9	3.6	0.2	-11.3	8.9	33.85%	66.15%

**Table 5: Cochran-Mantel-Haenszel test to show association between profession**

	cmh	df	pvalue
WHOLE MOUTH	0.075	1	0.78388
MAXILLARY	0.314	1	0.57522
MANDIBULAR	0.632	1	0.42672

which can allow nonideal arch lengths to be detected early. Dental arch space excess of one-two mm is usually ideal which will often allow the permanent teeth to erupt with little crowding and without intervention. However, if there is too much space available (>3 mm), decisions have to be made regarding potential space closure, and more long-term planning. Space deficiencies, on the other hand, often require more immediate intervention. A space deficiency of less than -3 mm might necessitate a lower lingual holding arch, while a space deficiency of greater than -3 mm may require a space regaining, arch expansion or extraction treatment.<sup>8</sup> The present study indicates that pediatric dentists are often underestimating the space available by an average of -3.6 mm, which is roughly half the size of an average premolar.<sup>9</sup> Following the traditional space deficiency measurements, they may be inclined to undesirably create space with an appliance or extractions, when the dentition may have been acceptable for early observation with no intervention.

There are several theoretical advantages to early treatment such as improved control of growth, improved self-esteem of the patient, and decreased cost and extent of treatment in the permanent dentition.<sup>10</sup> Some studies suggest that early intervention may have two improved treatment outcomes of less relapse and treatment times.<sup>11</sup> However, overall there seems to be insufficient evidence to support the additional benefits of early treatment over traditional treatment options.<sup>12,13</sup> Early treatment may add additional cost as permanent teeth may erupt into undesirable locations even after intervention, which would require correction with stage II treatment.<sup>14</sup> The lack of evidence supporting early mixed dentition therapy combined with the results of the present study suggests that early treatment should be restricted to conservative treatment options especially if practitioners are not using accurate measurements of arch space.<sup>12</sup> More irreversible methods like extraction therapy should perhaps be reserved until the occlusion has further developed. Early intervention should only be done if it will be truly efficient and effective as suggested by Proffit.<sup>14</sup> If pediatric dentists are to begin treatment in the mixed dentition, the results of this study indicate that they should use an established mixed dentition analysis to aid in more accurate treatment planning.

A limitation to this study was that we were not able to determine how accurate the measurements and estimations were to the actual permanent dentition arch length. Future studies could follow that arch length development into the full permanent dentition to allow this consideration.

## CONCLUSIONS

1. More than half of the dentists surveyed that practice comprehensive orthodontics do model estimation arch length analysis.
2. Pediatric dentists and pediatric dental residents are just as good as each other at estimating arch length.
3. Pediatric dentists and pediatric dental residents underestimated whole mouth arch lengths by -3.6 and -3.1 mm, respectively.
4. Both pediatric dentists and pediatric dental residents are better at estimating mandibular arch lengths than maxillary arch lengths.

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# Prevalence of Premature Eruption and Agenesis of Premolars in Turkish Children: A Retrospective Study

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**Objective:** To determine the prevalence and distribution of premature eruption and agenesis of premolars in a sample of Turkish children. **Study design:** A sample of 1715 patients aged 5 to 11 years was selected. Panoramic radiographs were used to assess premature eruption and agenesis of premolars. Developmental stage of erupted premolars was assessed using Demirjian's method and selecting prematurely erupted premolars on the basis of clinical eruption with a root length less than half of their final expected root lengths. Statistical analysis was performed using chi-square test ( $p < .05$ ). **Results:** One hundred fifteen (6.7%) of 1715 patients presented at least one premolar agenesis with no significant sex difference (56 boys, 59 girls). Mandibular second premolars were the most absent teeth. Multiple agenesis of premolars (3.4%) was more common than single agenesis (3.3%). A total of 85 (5.0%) patients (51 boys, 34 girls; no significant sex difference) had at least one prematurely erupted premolar, and maxillary first premolars were most commonly affected. Early erupted premolars were in stage D or E based on Demirjian's dental formation scale. **Conclusions:** The prevalence of premature eruption and agenesis of premolars in Turkish children were 5.0% and 6.7%, respectively. Both conditions are not uncommon and may highlight the need for early diagnosis to prevent subsequent clinical problems.

**Keywords:** Dental Anomalies; Tooth Eruption; Panoramic Radiography; Tooth Agenesis; Premolars.

## INTRODUCTION

Anomalies of teeth are caused by genetic factors and their interactions with environmental influences involved in different stages of tooth development. Their effects may manifest as various deviations in tooth size, morphology, structure, and number; and such anomalies may occur together as an isolated entity or as part of a syndrome<sup>1</sup>.

Among dental anomalies, tooth agenesis is the most common developmental dental anomaly. It is estimated that prevalence of congenitally missing teeth in permanent dentition (except for the third molar) varies from 2.6% to 11.3%, depending on the population studied. The most common missing teeth are mandibular second premolars and maxillary lateral incisors<sup>2</sup>. Agenesis of permanent teeth may occur simultaneously with several clinical situations, such as abnormalities in tooth size or shape, delayed development, or ectopic eruption of permanent teeth<sup>3</sup>.

Tooth eruption is often defined as a physiologic process that consists of movements of a tooth during odontogenesis in its crypt through the alveolar process till its functional position in the oral cavity<sup>4</sup>. Although numerous theories have been postulated to explain the eruption of teeth, the underlying mechanism(s) still remains unclear. Because the eruptive movement begins with the initiation of root formation, root growth and elongation have been considered in the eruptive mechanism<sup>5</sup>. However, teeth can also

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erupt after completion of root formation or when their root is removed surgically<sup>6</sup>.

Eruption of teeth with little or no root development may rarely occur as a result of radiotherapy<sup>7</sup> and congenital kidney disease<sup>8</sup>. Tooth eruption with partial or complete arrest of root formation due to traumatic injury to overlying primary tooth has also been reported<sup>9,10</sup>. Even a rootless mandibular second premolar has been reported to erupt under a primary molar with no signs of decay or infection<sup>11</sup>.

Premature loss of deciduous teeth due to periapical infection or decay may cause its permanent successor to erupt prematurely in the very early stage of root development<sup>12</sup>. Abscess formation of the overlying primary tooth causing bone destruction may also result in very early emergence of premolars.

To date, there have been only a few reports to describe early eruption of premolars with inadequate root formation<sup>9,10</sup>. The aim of this study was, therefore, to determine the prevalence and distribution of two distinct entities in the premolar region: premature eruption and agenesis in a sample of Turkish pediatric patients.

**MATERIALS AND METHOD**

This retrospective study assessed 1715 digital panoramic radiographs from 1794 patient records taken at the Dental School of Hacettepe University, in Ankara, Turkey, between 2018 and 2019. All radiographs were obtained for routine dental examination of children aged 5 to 11 years using the same device (Veraview IC5 device, Morita Corporation, Japan) and parameters (67 kV, 5.0 mA, 13.9 s). The panoramic images were viewed simultaneously by an experienced radiologist and an experienced pediatric dentist. In case of disagreement between their assessments of any image, a senior reviewer made the final decision. The study protocol involving the use of panoramic radiographs was approved by the Local Ethics Committee.

Patients with disorders such as syndromes or clefts and those with previous jaw fractures were excluded. Agenesis and premature eruption of premolars were assessed on digital panoramic images. Only erupted premolars in full occlusion were selected and included. Based on the description of Gron<sup>13</sup>, a premolar was defined as prematurely erupted when the root length of the tooth at clinical eruption was less than half of its final expected root length. The developmental stage of premolars was recorded according to Demirjian’s dental maturity stages<sup>14</sup>, as defined below:

Stage D: Crown formation is completed up to the cementoamel junction.

Stage E: Root length is less than the crown height.

Stage F: Root length is greater than or equal to the crown height.

Stage G: Root development is completed, but apical foramen is open.

Stage H: Root apex is completely closed.

In cases involving more than one agenesis or premature eruption of premolar (PEP), the finding was recorded as multiple.

**Statistical analysis**

Statistical analysis was performed by using SPSS for Windows 21.0 (IBM Corp. Released 2012. Armonk, NY). The number, percentage, mean, standard deviation, median, 1st and 3rd quartiles, minimum and maximum values were estimated for descriptive statistics. Chi-square were used to assess the significance of the differences between categorical variables.

The level of significance was set at p-value < 0.05.

**RESULTS**

The study sample consisted of 926 boys and 789 girls with a mean age of 8.8±1.68 years and 8.9±1.56 years, respectively. One hundred fifteen patients (6.7%) had at least one premolar agenesis. The distribution of missing premolars is presented in Table 1. Mandibular left and right second premolars were the most absent teeth. Multiple agenesis of premolars (n=59, 3.4%) was more common than single agenesis (n=56, 3.3%). The prevalence of premolar agenesis was 7.5% (n=59) in girls and 6.0% (n=56) in boys, and the difference between them was not significant (Table 2, p> 0.05).

The frequency and distributions of PEP is presented in Table 3. Eighty-five (5.0%) patients had at least one PEP. The most common PEPs were maxillary left and right first premolars, respectively (Table 3). The prevalence of PEP was higher in boys (51, 5.5%) than in girls (34, 4.3%), in the absence of statistical significance (Table 4, p> 0.05). With regard to their development; stage D (Fig.1) and stage E (Fig. 2) were observed in 51 and 47 of the premolars respectively. In nine cases, PEP was accompanied by premolar agenesis (Fig. 3 and 4).

**Table 1. Prevalence and characteristics of premolar agenesis**

Total (1715)	n	%
Single	56	3.3
Multiple	59	3.4
<b>Total</b>	115	6.7
<b>Tooth number</b>		
35	75	4.3
45	69	4.0
25	30	1.7
15	24	1.3

**Table 2. Prevalence of premolar agenesis according to sex**

	Male		Female		Total		p*
	n	%	n	%	n	%	
<b>Absent</b>	870	94.0	730	92.5	1492	93.3	0.246
<b>Present</b>	56	6.0	59	7.5	115	6.3	
<b>Total</b>	926	100.0	789	100.0	1715	100.0	

\*Chi Square Test, p>0.05

**Table 3. Prevalence and characteristics of prematurely erupted premolars**

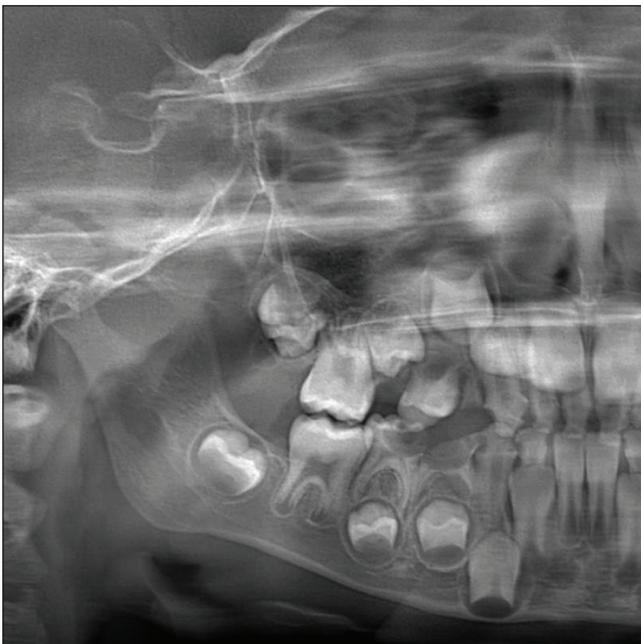
Total (1715)	n	%
Single	62	3.6
Multiple	23	1.3
Total	85	5.0
<b>Tooth number</b>		
24	29	1.7
14	28	1.6
44	24	1.4
15	13	0.8

**Table 4. Prevalence of premature eruption of premolars according to sex**

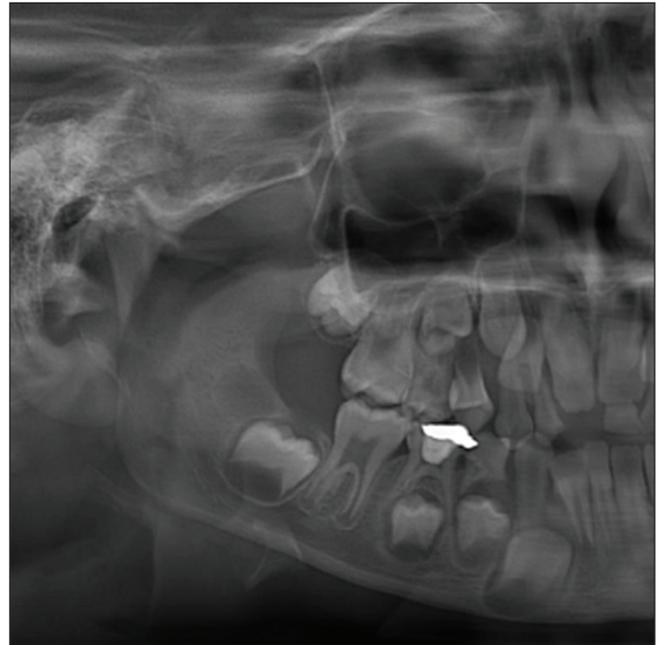
	Male		Female		Total		p*
	n	%	n	%	n	%	
<b>Absent</b>	875	94.5	755	95.7	1630	95.0	0.266
<b>Present</b>	51	5.5	34	4.3	85	5.0	
<b>Total</b>	926	100.0	789	100.0	1715	100.0	

\*Chi Square Test, p>0.05

**Figure 1. Panoramic radiograph showing a prematurely erupted maxillary right first premolar (stage D) in a 6-year-old boy. The tooth is in occlusion with almost initial root development.**



**Figure 2. Radiographic view of a 7-year-old girl with prematurely erupted maxillary right first premolar (stage E). The tooth is in occlusion with the root length less than that of the crown.**



**Figure 3. Radiographic view of a 10-year-old girl with prematurely erupted mandibular first premolar (stage E) and agenesis of mandibular second premolar.**



**Figure 4. Panoramic radiograph showing a prematurely erupted maxillary right second premolar (stage D) and congenital absence of the maxillary right first premolar in an 8-year-old boy.**



## DISCUSSION

The present study was designed to investigate the prevalence and characteristics of premature eruption and agenesis of premolars. This study supports evidence from previous observations that rootless teeth or teeth with little root formation can erupt into full occlusion, indicating that root formation is not required for tooth eruption<sup>7,8,10,11</sup>. Although root development does not seem to provide eruptive force or to be responsible for the active eruption of a tooth, it has correlations with eruption. According to Gron<sup>13</sup>, the gingival emergence of a premolar usually occurs when one-half to three-fourth of its final root length is formed. Based on the diagnostic criteria suggested by Becker<sup>15</sup>, if an erupted tooth has less root development than its expected root length at eruption, is defined as prematurely erupted. Gron's<sup>11</sup> method estimates the root length with reference to the adjacent roots and alveolar outlines of adjacent teeth, and the criteria of Demirjian's method<sup>12</sup> rely on the proportion of root length to crown height rather than its absolute tooth length. Thus, the well-known distortions of developing teeth in panoramic images were considered to have a minimal impact on measurement accuracy.

In the present study, the developmental stages of PEP were D and E according to Demirjian's classification<sup>14</sup>. This finding indicates that once the tooth prematurely erupts, it reaches the occlusal level in a very early stage of root development. This is also consistent with the findings of Brin and Koyoumdijsky-Kaye<sup>16</sup>, who demonstrated that permanent successors of prematurely extracted primary molars have a reduced root length than their usual root length at the time of normal eruption.

The mean ages of patients with prematurely erupted maxillary and mandibular first premolars were  $8.78 \pm 1.11$  years and  $9.06 \pm 1.20$  years, respectively; and the mean ages of the patients with prematurely erupted second premolars were  $9.66 \pm 1.15$  years. In a previous study<sup>17</sup>, the mean eruption times were  $10.25 \pm 1.44$  years

and  $10.22 \pm 1.39$  years for maxillary and mandibular first premolars; and  $11.00 \pm 1.10$  years and  $11.06 \pm 1.08$  years, for maxillary and mandibular second premolars, respectively. From a chronologic perspective, the present findings confirm the premature eruption times of premolars<sup>15</sup>.

The premature extraction of primary teeth may impact the eruption time of their permanent successors by either delaying or accelerating their eruption. According to Posen<sup>18</sup>, eruption is delayed if the extraction is done at ages four and five, while early eruption occurs if the primary tooth is lost at ages five to eight. Fanning<sup>12</sup> reported that eruption of a premolar is accelerated in the presence of an infected deciduous tooth and extraction of a deciduous molar before the premolar crown has formed. The premature loss of primary tooth resulting in accelerated vertical movement of the permanent tooth bud may cause bud to face occlusal stresses early, which negatively affects the root formation<sup>16,19</sup>. Maxillary deciduous molars, particularly the first molars, are one of the most frequently early extracted teeth due to extensive decay and periapical infection<sup>20,21</sup>. This may explain the higher prevalence of maxillary first premolars among the prematurely erupted teeth in our study. Further, the number of prematurely erupted premolars were higher in boys than in girls in the present study, probably because boys had more extracted deciduous molars compared to girls.

The prevalence and distribution of developmental dental anomalies have been widely studied among various populations, ethnic groups and races<sup>22-24</sup>. The premolar teeth, especially second premolars may present a wide variety of developmental anomalies, with tooth agenesis, supernumerary teeth, dens evaginatus, rotation, and root dilacerations being the most common<sup>24,25</sup>. The concurrent occurrence of these anomalies is not uncommon which may suggest a possible role of shared genetic mechanisms.

The reported prevalence rates for premolar agenesis vary from 0.1% to 11.3% in different populations<sup>2,26</sup>. In this study, the

overall prevalence of premolar agenesis (6.7%) was higher than those reported by Altug-Atac and Erdem<sup>22</sup> (0.46%), Uslu *et al*<sup>25</sup> (3.5%), and Kazanci *et al*<sup>27</sup> (1.58%). In line with previous reports<sup>20-22</sup>, mandibular second premolars were the most frequent missing premolar teeth in our study. Here, a minor, insignificant difference was observed between males and females as well as between single and bilateral agenesis. Agenesis of premolars is often associated with other dental anomalies such as distoangulation, delayed development of mandibular second premolars, and infraocclusion of primary molars<sup>3,28-30</sup>. However, the present study did not aim to investigate such concurrent conditions.

The premature loss of primary molars may cause succedaneous premolars to erupt early with a reduced root development than their expected root length at normal eruption time. The fate of PEP is unstudied, but it can be speculated that undeveloped root status makes them unstable to occlusal forces. Another complication of early loss of primary molars is the loss in arch length and subsequent malocclusion, which coincides with the effects of tooth agenesis on the permanent dentition. Long-term follow-up observations are necessary to manage complications in a timely fashion.

## CONCLUSIONS

The prevalence of premature eruption and agenesis of premolars in Turkish children were 5.0% and 6.7%, respectively, indicating that those conditions are not uncommon among Turkish children. Mandibular second premolars were the most frequently missing teeth, and the maxillary first premolars were most commonly affected by premature eruption. Neither the frequency of premature eruption nor that of the agenesis of premolars were significantly affected by sex. Both conditions may highlight the need for early diagnosis to prevent subsequent clinical problems.

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## Conflict of interest

The authors declare that they have no conflict of interest.

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## Delayed Development of Maxillary Second Premolar: Case Report and Literature Review

Chen Ying\*/ You Wen Zhe\*\*/ Xia bin \*\*\*

*Delayed tooth development (DTD) is the development progress of a tooth germ that takes place later due to local or general causes. This case report reviews a 16-year-old Asian adolescent whose bilateral upper second premolar germs were at Nolla's 6 stage as shown on a panoramic radiograph. It is unusual that tooth germs of the maxillary second premolar are developed after 11 years of age. To reduce the chance of misdiagnosis, clinicians should consider the possibility of DTD if a tooth germ does not present in radiographs.*

**Keywords:** Delayed tooth development, premolar, children

### INTRODUCTION

Delayed tooth development (DTD) is the development progress of a tooth germ that takes place later from local or general causes. Generally, tooth development occurs at particular age. For instance, the hard tissue of maxillary second premolars forms at 2-2.5 year old, and the enamel completed at 6-7 years old; Normally, They will erupt at 10-12 years old.<sup>1</sup> It is rare if the development of a tooth germ is delayed. This case exhibits delayed tooth development (DTD) in a male adolescent whose maxillary second premolars did not exist at the first visit when he was 11 years old, but the tooth germs could be seen on another panoramic radiograph when he was 16 years old. To reduce the chance of misdiagnosis, clinicians should consider the possibility of DTD if a tooth germ does not present in radiographs.

### Case report

A Han nationality male, born in Oct 2002, aged 11 years old was referred to the Peking University School and Hospital of Stomatology in November 2013 for a routine examination. The patient had no systemic disease or abnormal medication history. The intraoral examination revealed early permanent dentition with completely erupted lower premolars (34, 35, 44, 45), present and partially erupted upper premolars (14, 24), and clinically absent upper second premolars (15, 25). A panoramic radiograph revealed initial tooth bud formation of the upper right second premolar (25); however, there was no indication of the upper left second premolar (15), and the presence of all third molars was apparent (Figure 1). In July 2018, the patient underwent another panoramic radiograph; the upper left second premolar (Nolla's stage 6) was present, and the upper right second premolar was also at Nolla's stage 6 (Figure 2). The patient's father lost his upper central incisors (11, 21) because of trauma; his upper left first and second molars, upper left second premolar, and lower right second molar (25, 26, 27, 47) were missing because of periodontal disease; and his upper right second premolars were congenitally absent (Figure 3). His mother does not have missing teeth.

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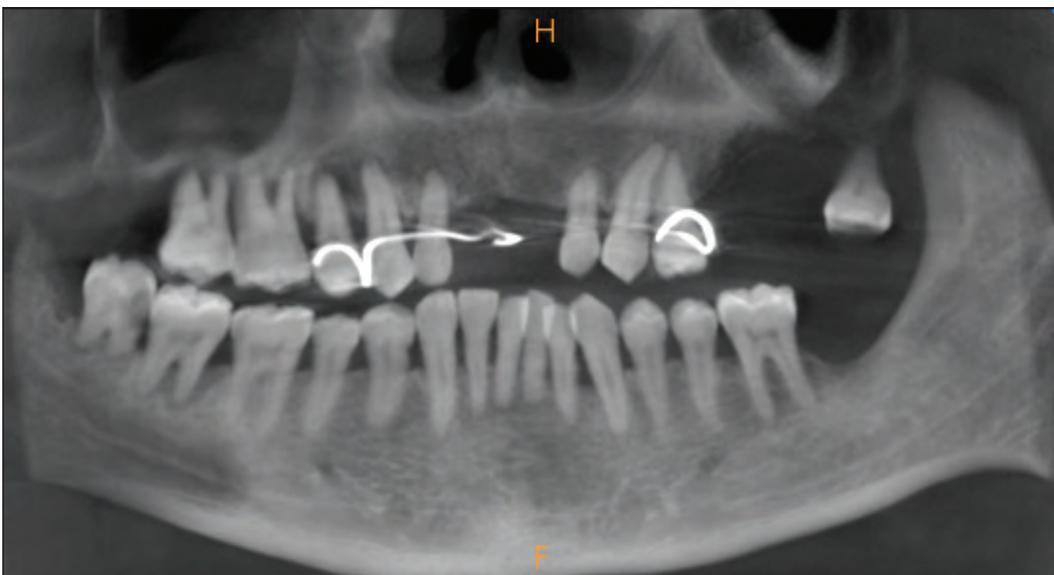
Figure 1. Panoramic radiograph showing missing upper right premolar bud (November 2013)



Figure 2. Panoramic radiograph showing upper second premolars were in Nolla's stage 6 (July 2018)



Figure 3. Cone-beam computed tomography section showing missing teeth in his father (March 2017)



## DISCUSSION

This case presents delayed development of bilateral maxillary second premolars in a 16-year-old male. The panoramic radiograph taken when he was 11 years old revealed initial tooth bud formation of the upper left second premolar. Another panoramic radiograph taken five years later presented the delayed development of the bilateral maxillary second premolars. The growth rate was not significantly different from the general interval of approximately 5 years from the onset of hard tissue formation until the crown completes for second bicuspid teeth.<sup>1,2</sup>

Tooth development involves a complex signaling network between endothelial mesenchymal tissue formed by the neural crest and dentinal epithelium. Delayed tooth development refers to the development of a tooth with Nolla's stage 2 or later than that of the same tooth in the same age group<sup>1,3</sup>. Park et al,<sup>4</sup> conducted a survey in 2017 amongst 4611 children aged 6-12.9 years in South Korea, and the prevalence rate of delayed tooth development was 3.4%, of which the second premolar of the upper jaw was the most commonly delayed tooth development, with 1.02%. The proportion of those with delayed development of the second maxillary second molar and second mandibular premolar was 0.88% and 0.74%, respectively.

Causes of this phenomenon may be related to premature birth, malnutrition, hormone secretion disorder and heredity.<sup>5</sup> Hitherto, more than 200 genes have been identified related to tooth development. Studies have shown that PAX9, MSX1 and WNT10A are associated with both congenital tooth loss and delayed tooth development.<sup>6,7</sup> Dharmo *et al*,<sup>8</sup> found a trend of lower stages of tooth development in patients with WNT10A variants. In this case, the presence of bilateral maxillary second premolar loss in the father of the child and delayed tooth development in the child suggested that delayed tooth development may be a genetic developmental abnormality.

In reviewing previous literature, Memmott *et al*,<sup>9</sup> reported a black female with delayed development of the upper left premolar and congenital loss of the upper right incisor. When she was 17 years and 1 month old, the tooth germ of the upper left premolar with Nolla's stage 6 was found. That is the latest age at which delayed tooth development has been reported. The male in this case is the latest age found in an Asian population. Because the incidence of delayed tooth development is low, compared with other tooth development obstacles, this phenomenon has not received sufficient attention. This case suggests that if tooth germs of maxillary premolars are absent in clinical or imaging examinations, it is necessary to read the film carefully to observe whether there is evidence of the early tooth germ. Treatment plans should be made after a comprehensive analysis. Clinicians should consider the possibility of delayed tooth development so that we can reduce the incidence of misdiagnosis of congenital missing teeth.

## CONCLUSION

The presence of delayed tooth development and the wrong diagnosis of congenital tooth loss due to the neglect of this condition will affect the choice of the orthodontic program and the correct time of intervention. To reduce the chance of misdiagnosis, clinicians should consider the possibility of DTD if a tooth germ does not present in radiographs in particular chronological age, and proper radiographs should be periodically examined.

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