#### Association of consanguineous marriages with severe early childhood caries: a pilot study

Ahmet Aras<sup>1</sup>, Mehmet Sinan Dogan<sup>2</sup>, Şemsettin Yıldız<sup>3</sup>, Muhammed Demir<sup>4\*</sup>

1. Department of Pediatric Dentistry, Private Oral and Dental Health Clinic, Şanlıurfa, Turkey.

2. Department of Pediatric Dentistry, Faculty of Dentistry, Harran University, Sanlıurfa, Turkey.

3. Department of Pediatric Dentistry, Faculty of Dentistry, Firat University, Elazig, Turkey.

4. Department of Pediatric Dentistry, Private Oral and Dental Health Clinic, Ankara, Turkey.

\*Corresponding author: Denir M. MSc. PhD. Asst. Prof. Department of Pediatric Demistry, Private Oral and Denial Health Clinic, Ankara Turkey. E-mail : mini. demir@icloud.com

#### Abstract

**Objective:** Consanguineous marriages have been reported to cause various genetic problems and lead to a number of syndromes, including dental syndromes that can result in dental caries. The aim of this pilot study was to determine whether consanguineous marriage is a predisposing factor for severe early childhood caries (S-ECC).

**Methods:** Ninety S-ECC patients aged 36–71 months were included in the study. Patients were divided into two groups; first group included children of first-cousin consanguineous marriages (n1 = 45), and the second group comprised children whose parents did not have consanguineous marriages (n2 = 45). Dental caries were recorded by two experienced paediatric dentists using the dmft index based on the ICCMS<sup>TM</sup> criteria. Between-group comparisons of dmft data were carried out using the Mann–Whitney U-test.

**Results:** The 90 paediatric patients in this study included 42 (46.7%) females and 48 (53.3%) males with a mean age of  $4.39 \pm 1.07$  years. The median dmft value for Group 1 was 9 (5-14) and 8 (5-15) for Group 2. However, this difference was not statistically significant (p>0.05).

**Conclusion:** Although the median dmft score was found to be higher in children with consanguineous parents, this difference was not statistically significant.

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## 1. Introduction

Early childhood caries (ECC) is a persistent and possibly growing public health problem (1). ECC is defined as the presence of one or more decayed (cavitated or non-cavitated), missing, or filled tooth surfaces in any primary tooth in a child 71 months of age or younger (2). In addition, early childhood caries is considered severe if it occurs in children younger than 3 years old or in children 4–6 years of age with elevated caries scores (3). Severe early childhood caries (S-ECC) is an aggressive form of dental caries that is characterised by a decayed, missing (due to caries), or filled tooth (dmft) index score  $\geq 4$  (age 3),  $\geq 5$  (age 4), or  $\geq 6$  (age 5) (4). S-ECC is destructive, and when it progresses, it can cause acute pain, sepsis, and potential tooth loss (5).

ECC is recognised as a serious public health problem due to its high prevalence, impact on quality of life, increased risk of caries in the permanent dentition, and its role in general health (6). One to twelve percent of children younger than 6 years in the developed world experience ECC (7). Because of the young age of these children, S-ECC is difficult to treat successfully in the dental chair, and these children frequently require treatment under general anaesthesia, which increases treatment costs (5,8).

Numerous risk factors have been associated with ECC. Epidemiological studies have found correlations between ECC and low socioeconomic status, minority status, low birth weight, and transfer of microbes from mother to child. In addition, child oral healthcare, feeding and cleaning behaviours, night-time bottle feeding, consumption of cariogenic frequent food. late commencement of child tooth brushing, and irregular brushing habits are also associated with ECC (7,9). predisposition, environmental Genetic and socioeconomic factors, parental education and awareness, ethnicity, marital status, and family size also affect caries formation (6). However, little is known about the host genetic factors that influence susceptibility (10).

The role of genetic factors continues to be an active area of research, and recent studies have described a potential genetic contribution to the risk for dental caries (1,11). A study conducted in Najran found that, in a region where consanguineous marriages are widespread, genetic factors may underlie the high prevalence of dental caries (10).

In addition, considerable genetic research has examined consanguineous marriage and its detrimental effects on offspring. Although consanguineous marriages are common in the world, their relationship with oral and periodontal diseases has not been thoroughly investigated, and research on the association between consanguinity and the various parameters of oral and periodontal health is needed (12,13). Therefore, the aim of this pilot study was to investigate whether consanguineous marriages leading to genetic diseases are a risk factor for S-ECC.

## 2. Materials and methods

Prior to the study, the patients' parents signed an informed consent form, and approval for the study was obtained from the Clinical Research Ethics Committee of Harran University (Reference no: 19.07.04). Inclusion criteria were as follows: a) 36–71 months old, b) no chronic illnesses, c) previously diagnosed with S-ECC, and e) registered at the dental clinics of Harran University Faculty of Dentistry. Families who did not complete the questionnaire and children who came to the clinic with an adult who was not a parent were excluded from the analysis.

Participation involved a thorough dental examination followed by a comprehensive interview with the parent(s). Clinical oral examinations were performed by two paediatric dentists with more than 5 years of experience. Prior to the study, both dentists conducted an examiner training and calibration session to compare their scoring of ten patients. Inter-examiner and intra-examiner reliability were assessed using Cohen's Kappa statistic ( $\kappa = 0.90$  for inter-examiner agreement and  $\kappa = 0.96$  for intra-examiner agreement).

Patients were examined for caries status and dmft score in a dental chair under good illumination using a sterilised mouth mirror, explorer, cotton rolls, and gloves, as recommended by the World Health Organization (WHO) (14). Radiographs were not obtained. Dmft scoring was based on the International Caries Classification and Management System (ICCMS<sup>TM</sup>) criteria (15). After the dental examination, the parents were asked whether they had a consanguineous marriage, and if so, the degree of relationship.

Ninety patients with S-ECC were divided into two groups; the first group included children of first-cousin consanguineous marriages (n1 = 45), and the second group included children of parents who did not have consanguineous marriages (n2 = 45). Between-group comparisons of dmft data were carried out using the Mann–Whitney U-test.

## 2.1. Statistical Analyses

Data were analysed using SPSS software (IBM Corp., Armonk, NY, USA). Mean, standard deviation (SD), median, minimum–maximum, and percent values were calculated for descriptive statistics. Normality of the data distribution was tested using histograms and the Kolmogorov–Smirnov test. As the data were not normally distributed, the Mann–Whitney U-test was used for between-group comparisons. Statistical significance was defined as p-values <0.05.

# 3. Results

Of the 90 children included in this study, 42 (46.7%) were female, and 48 (53.3%) were male. The mean age was  $4.39 \pm 1.07$  years (Table 1). The median dmft score for Group 1 was 9 (range: 5–14), and that for Group 2 was 8 (range: 5–15); this difference was not significant (p > 0.05) (Table 2).

Table 1. Group demographi	cs.
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	Ν	Gender		Age (years)	
		Female	Male	Mean ± SD	
Group 1	45	23	22	$4.40\pm1.05$	
Group 2	45	19	26	$4.38 \pm 1.09$	
Total	90	42	48 (53.3%)	$4.39 \pm 1.07$	
		(46.7%			
		)			

**Table 2.** Between-group comparisons of dmftscores.

	Group	Mean	Std.	Median	Min.	Max.	Р-
			Deviation				value
	Group	9.13	2.58	9	5	14	0.159
dmft	1						
score	Group 2	8.38	2.36	8	5	15	

# 4. Discussion

The aim of this pilot study was to determine whether consanguineous marriage is a risk factor for S-ECC. When the intraoral examination and information obtained from the families were examined statistically, consanguineous marriage was found to increase the mean and median dmft scores in patients with S-ECC. However, this increase was not statistically significant.

Previous studies have indicated that consanguineous marriage brings about various genetic problems, including dental syndromes that can increase the risk of tooth decay (10,12,13,16). However, to our knowledge no previous study has investigated the relationship between S-ECC and consanguineous marriage.

Early childhood caries is a result of a multifaceted interaction of biological, genetic, and biochemical factors with an overlaying complex of social determinants of oral health (17). It is well established that environmental factors such as diet, oral hygiene, other oral habits, and socioeconomic factors are risk or protective factors for caries. However, environmental factors can overcome the genetic component of this complex disease (11). Recent studies continue to demonstrate that genetic variation is associated with caries and that these variations can play a role in caries aetiology as risk factors or as protective factors (18–20). Some studies have shown significant differences in genetic disorders between children born to consanguineous marriage partners and those born to nonconsanguineous parents, while others have found no significant differences (16).

Consanguineous marriage is defined as a marriage between people who are related as second cousins or closer (21). Consanguinity rates differ in communities depending on religion, culture, and geography, and the prevalence is high among Middle Eastern and Arab citizens (22). The highest rates of consanguineous marriages in the world are seen in many Arab countries, where 20–50% of all marriages are consanguineous, especially marriages between first cousins (12). According to data from the Turkish Family Structure Survey, the consanguineous marriage rate in Turkey in 2011 was 23.3%, with a rate of 21.1% in urban areas and 28.2% in rural areas in urban areas, with the highest rate found in the Southern Anatolian Region (44.8%) (21).

Although consanguineous marriages are common in the world, their effects on oral diseases have not been thoroughly investigated, and our understanding of their effects on oral and periodontal diseases is currently inadequate (13). One of the few studies on this subject showed that amelogenesis imperfecta (AI) can develop as a result of consanguineous marriages and that similar enamel defects are found in families of patients with AI (23,24). A study conducted in Saudi Arabia, where consanguineous marriages are common, suggested that consanguinity could be the source of the genetic component behind the high prevalence of dental caries (10). Supporting that study's findings, our results showed that consanguineous marriage was associated with increased dmft scores in children.

# 5. Conclusion

The findings of this pilot study suggest that consanguineous marriage may be one of the risk factors for S-ECC. Due to the limitations of the present study, future investigations are necessary to validate these results.

Descriptions

Author Contribution

Study conception and design: AA, MSD, ŞY

Analysis and interpretation of the data: AA, MSD, SY, MD

Draft manuscript preparation: AA, MSD, MD

Critical revision of the work: AA, MSD

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**Ethical approval:** All procedures were approved by the Clinical Research Ethics Committee of Harran University (Reference no: 19.07.04).

**Conflict of interest:** The authors declare that they have no conflict of interest.

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