

## ORIGINAL RESEARCH

# Oral health knowledge, attitude and practice among parents of children with craniofacial syndromes

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**Abstract**

Parents play an important role in caring for their children's oral health, especially for those with craniofacial deformities. In this study, we analyzed the oral health knowledge, attitude and practice (KAP) among parents of children of 1 to 16 years-of-age with craniofacial syndromes (CS) at Universiti Malaya Medical Center (UMMC), Malaysia. This was a case-controlled study conducted between March and December 2021 involving 30 parents of children with CS and 30 parents of normal children as controls. A modified validated KAP questionnaire was distributed to all parents. Statistical analysis was carried out using SPSS 26.0 and descriptive analysis was performed, with data expressed as mean, standard deviation, frequency and percentage (%). Most respondents from both groups were mothers (73.3%) between 31 to 40 years-of-age. Both groups of parents had similar levels of oral health knowledge; there was no significant difference between the two groups for 10 of the KAP questions ( $p > 0.05$ ). However, there was a significant difference between the two groups with regards to two questions relating to the definition of plaque and its relationship to dental caries ( $p = 0.035$  and  $p = 0.032$ , respectively). Some parents of CS children believed that primary teeth were not important (23.33%) and were not concerned if their children showed changes in tooth color (26.67%). Despite parental acknowledgement of ideal practice, both groups of children showed irregular dental attendance and reduced toothbrushing frequency. Parents of children with CS had a similar depth of oral health knowledge and a slightly reduced positive attitude when compared to parents in the control group. However, both groups of parents had poor knowledge relating to the transmission and causes of dental caries. Healthcare providers should increase their awareness strategies for parents to be more aware of the ways to improve their children's oral health.

**Keywords**

Attitude; Craniosynostosis; Craniofacial syndrome; Knowledge; Oral health; Practice; Parents

## 1. Introduction

Craniofacial deformities are structural malformations involving the head and facial regions that stem from birth. Among others, this condition may coalesce with clinically documented syndromes such as Apert, Pfeiffer and Crouzon. Although the prevalence of these conditions is reported to be as low as 0.0003% [1], the oral health of children with craniofacial syndromes should not be neglected. In Malaysia, the Combined Oro-Craniomaxillofacial Clinic at the Universiti Malaya Medical Center (UMMC) is the only established multidisciplinary clinical and research center that enables clinicians to provide comprehensive care for these patients.

Previous reports found that the craniofacial deformities of some children were linked to alterations in the homeobox protein *MSX1* and paired box gene 9 (*PAX9*) genes resulting in various dental anomalies, including enamel hypoplasia and

ectopic teeth. Most children with craniofacial syndromes had to undergo surgeries whilst growing up; this would have led to possible complications and changes to their oral cavity, including hypoplastic maxillae and residual scar tissues [2]. Therefore, caring for the oral health of these children is vital as they present with limitations that can impede them from achieving even the bare minimum of oral health practices. However, some parents have been reported to be rather oblivious to their children's dental condition, especially the primary teeth, as they are under the false misinterpretation that the primary teeth are not essential as they are superseded by the permanent teeth [3].

In retrospect, research has proven that patients with craniofacial deformities, such as cleft lip and palate (CLP), are much more predisposed to developing caries, periodontal problems and have significantly poorer oral hygiene than those without CLP [4]. This insinuates that there may be special dental

care requirements amongst this group of children that could have been overlooked. No previous study has investigated the specific knowledge of parents with respect to the oral health of children with craniofacial syndromes.

Thus, the objectives of this study were to: (1) analyze and compare the oral health knowledge, attitude and practice (KAP) between parents of children with craniofacial syndromes (CS) and parents with normal children, and (2) relate the knowledge of parents with respect to oral health knowledge and the oral health practice of their children.

## 2. Materials and methods

This was a case-controlled study conducted on parents of children with CS and other disorders. A priori power analysis was conducted using G\*Power [5] to determine the minimum sample size required to test the difference between two independent proportions. Thirty participants per group were required to achieve 84% power for a given difference between a proportion of 0.35 at a significance criterion of 0.05.

Considering the possibilities of no response and incomplete survey responses, a total of 44 participants were recruited from the Combined Oro-Cranio-maxillofacial Clinic, UMMC, Kuala Lumpur, Malaysia and denoted as the test group. The inclusion criteria for the test group were parents or guardians who had children aged 16 years and below who had been diagnosed with craniofacial disorders. For comparison purposes, the parents of children who were free from craniofacial syndromes were matched for age and gender for the test group (parents of children with CS) and labelled as the control group. These samples were randomly selected from the faculty's pediatric clinic. All parents or guardians must have acquired a good comprehension of either English or Malay language. Written informed consent was obtained from the questionnaire. The exclusion criteria for both groups included children that had not been diagnosed with craniofacial disorders or parents who refused to participate in this study.

The closed-ended questionnaire was adapted from an existing and published KAP questionnaire established by Jain *et al.* [6] and Salama *et al.* [7] to cater for the added test group for comparison purposes. In these former papers, experts were asked to analyze the content of the questionnaire with regards to the relevance and provide ratings on a four-point scale as per the guideline below:

- 4—Relevant (simple and clear)
- 3—Relevant but requires minor alteration
- 2—Requires major alteration
- 1—Not so relevant (could be removed)

The percentage of total items rated by experts as either 3 or 4 was regarded as a content validity index. A score of greater than or equal to 80% was deemed to have good validity. The content validity index of the questionnaire was 95% after calculation. The reliability of the questionnaire was calculated by Statistical Package for the Social Sciences Version 20 (SPSS ver. 20.0. IBM Corporation, USA) using Cronbach's  $\alpha$ ; the reliability was  $r = 0.8$ ; thus, the questionnaire was reliable.

The content of the questionnaire was then proofread by two specialists from the Department of Pediatric Dentistry and Oral Maxillofacial Surgery. Prior to distribution, the questionnaire

was pre-tested on five parents to ensure that the questionnaire was clear and lucid. Positive feedback was obtained, and the average time taken to answer the whole questionnaire was 15 minutes. These five subjects were excluded from the final results.

Due to the COVID-19 pandemic and lockdown restrictions in Malaysia, the participants were contacted and asked to fill in an online questionnaire *via* Google Form. The data were collected between March and November 2021. The first section of the questionnaire assessed the socio-demographic background of the parents which consisted of their age, sex and income. The second section of the questionnaire was related to their knowledge, attitude and practice towards their children's oral health.

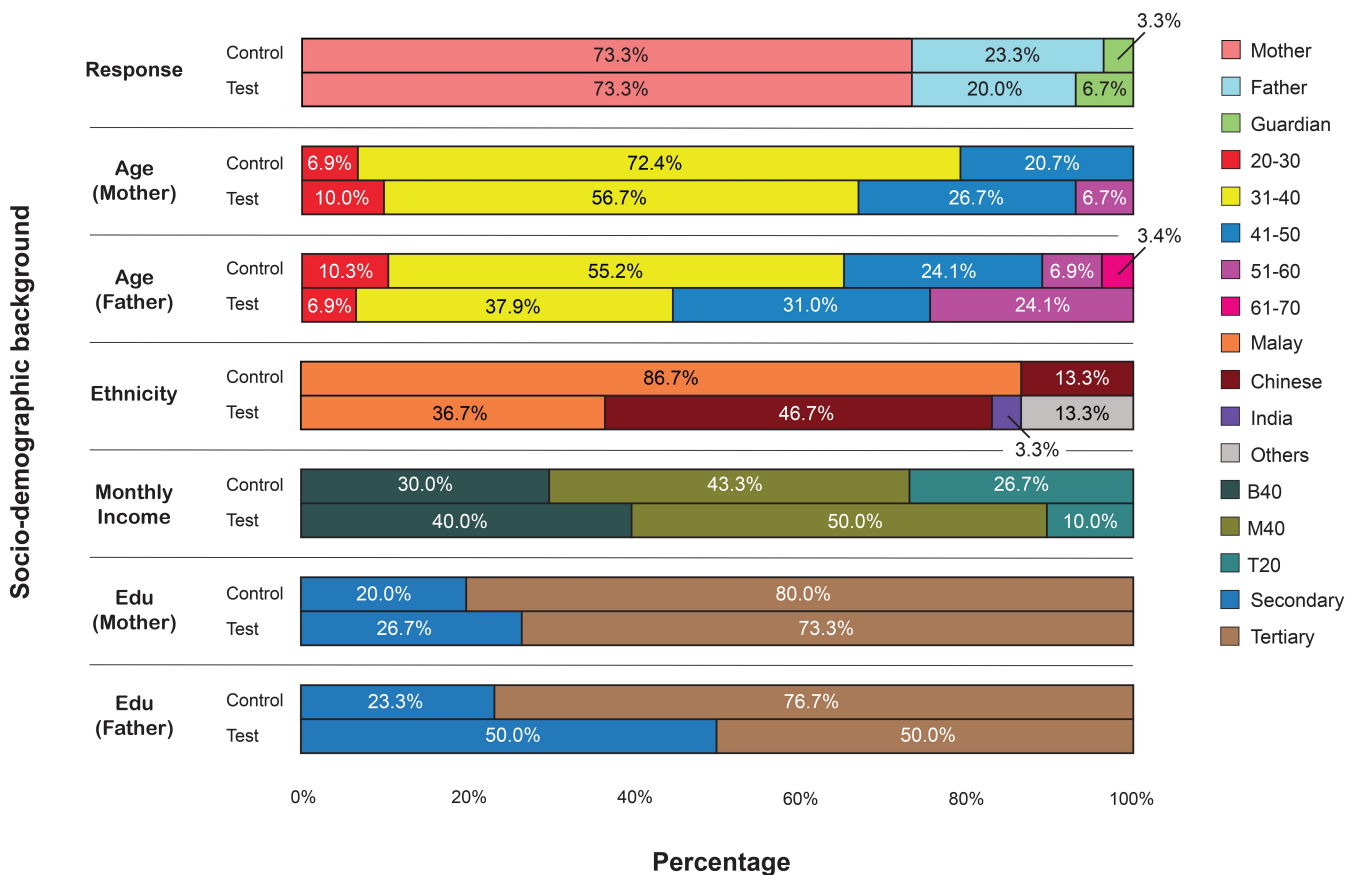
For the knowledge section, 12 multiple-choice questions were given with only one correct answer. For the attitude and practice sections, a 5-point Likert Scale was used (1 = Strongly agree, 2 = Agree, 3 = Neutral, 4 = Disagree, 5 = Strongly disagree) [1]. An answer of "strongly agree/agree" was considered as a positive statement of attitude. "Neutral" and "disagree/strongly disagree" were defined as a negative attitude.

All data were summarized in a Microsoft Excel spreadsheet before further statistical analysis was performed using Statistical Package for the Social Sciences Version 26 (SPSS ver. 26.0. IBM Corporation, USA). Both descriptive and inferential statistics were carried out. Results were mainly reported in the form of percentages. The socio-demographic background of parents and the CS children were presented as a clustered stacked bar chart. The number of correctly answered questions was recorded in the knowledge section and the chi-squared test was then used to determine the statistical difference or association between the knowledge of parents of children with CS and parents in the control group. Questions that resulted in a  $p$  value of  $< 0.05$  were considered significant.

## 3. Results

### 3.1 The socio-demographic background of parents

A total of 44 parents of children with CS registered at UMMC were approached for this study. However, eight parents gave no response, one withdrew and five parents of children that were not diagnosed with craniofacial disorders were omitted. Hence, only 30 parents of children with CS were able to participate in this study. An additional 30 parents with normal children that matched the age and gender of the study group were used as controls. Fig. 1 depicts the summary of the socio-demographic background of the parents involved. The participants of our study were mainly mothers (73.3%). Parents of children with CS were mostly Chinese (46.7%) whereas parents of other children in the control group were mostly Malay (86.7%). The mean age (and standard deviation) of the mothers and fathers of children with CS were 39.3 ( $\pm 6.8$ ) years and 43.21 ( $\pm 8.4$ ) years, respectively. The mean age (and standard deviation) of mothers and fathers in the control group were 36.9 (6.02) years and 39.1 (8.3) years, respectively. The mean age and standard deviation of mothers of children



**FIGURE 1. Socio-demographic background of parents of children with craniofacial syndromes and children in the control group.**

with CS are comparable. On the other hand, the mean age of fathers of children with CS was slightly higher than that of fathers in the control group with relatively equal standard deviations. Most of the parents had tertiary education and came from the middle 40% group (M40) of the Malaysian household class; their monthly household income ranged from RM4851 to RM10,970. Some of the subsections were short of one sample due to irretrievable data (such as death). No statistical significance was detected between the control and test groups ( $p > 0.05$ ) with this respect.

### 3.2 Socio-demographic background of children with CS

Fig. 2 presents a summary of the children with craniofacial disorders. We found that more than half of the children in our study were girls (56.7%) and were aged 5 years and below (53.3%). The mean age of the children was 6.0 years with a standard deviation of 3.6; these children were mostly affected by Apert syndrome (26.67%) followed by Crouzon (23.3%) and Pfeiffer syndrome (13.3%).

### 3.3 Knowledge of oral health

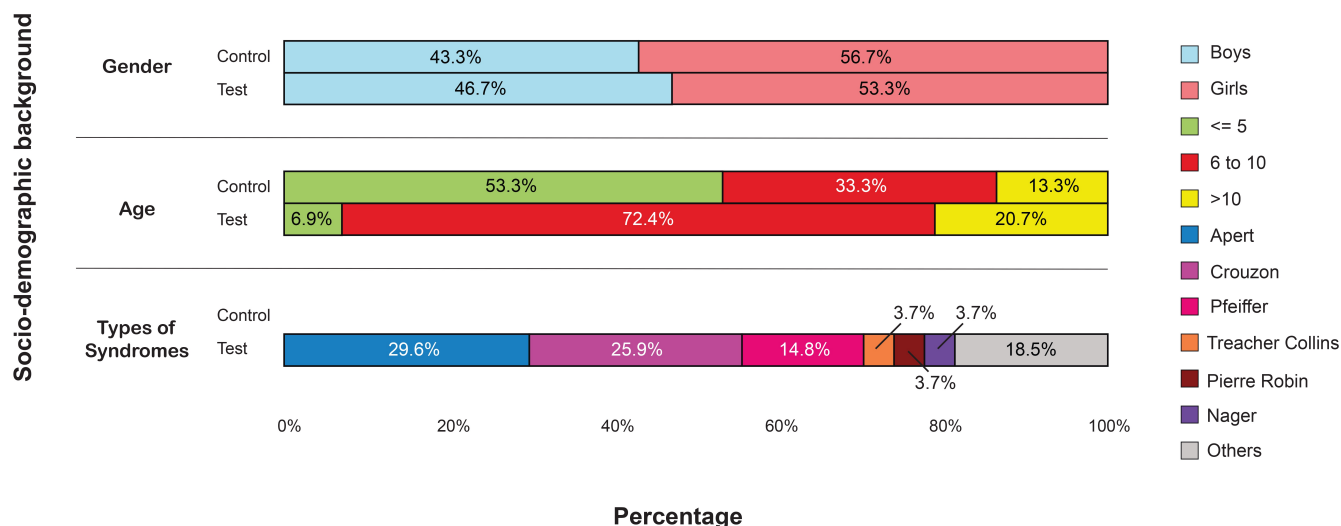
Both groups of parents had a relatively similar depth of knowledge on oral health; answers to only two of the 12 questions assessed were significant when compared between the two

groups ( $p < 0.05$ ). These two questions were regarding the definition of dental plaque and its relationship to caries; we discovered that parents in the control group scored higher (73% and 77% respectively). In contrast, a lower percentage of parents of children with CS were able to answer these questions correctly (47% and 50% respectively) (Table 1).

Both groups of parents failed to acknowledge that dental caries were transmissible with each group scoring less than 30%. With regards to the causes of dental caries, both groups were unaware that crowding and irregular tooth morphology, such as pits and fissures, could increase the risk of caries. Most of these parents were well-equipped with basic knowledge relating to toothbrushing, the types of bristles to be used and the role of fluoride.

### 3.4 Attitude

Generally, most parents from both groups had a positive attitude towards oral health with a cumulative percentage of 70% and 80% respectively. The results obtained for each statement are illustrated in Fig. 3. A portion of the parents of children with CS (26.7%) were unconcerned if they saw changes in the tooth color of their child. In addition, 23.3% of these parents believed that primary teeth were not important as compared to permanent teeth. In contrast, the control group (93.33%) showed a more positive attitude towards the importance of primary teeth. However, these differences were



**FIGURE 2. Socio-demographic background of the children with craniofacial syndromes (the test group).**

**TABLE 1. Statistical analysis of the knowledge section of the questionnaire for both groups of parents.**

Questions related to parents' knowledge	Group	N answered correctly (%)	<i>p</i> value
Frequency of toothbrushing	Test	22 (73)	0.095
	Control	27 (90)	
The recommended type of bristles	Test	23 (77)	0.260
	Control	22 (73)	
Bacterial transmission between individuals	Test	7 (23)	0.766
	Control	8 (27)	
Role of fluoride: strengthens teeth	Test	12 (40)	0.791
	Control	11 (37)	
Role of fluoride: prevents tooth decay	Test	20 (67)	0.136
	Control	26 (87)	
Causes of tooth decay: Bacteria	Test	19 (63)	0.080
	Control	25 (83)	
Causes of tooth decay: the amount of sugary food intake	Test	19 (63)	0.584
	Control	21 (70)	
Causes of tooth decay: crowding	Test	6 (20)	0.739
	Control	5 (17)	
Causes of tooth decay: irregular tooth morphology	Test	4 (13)	0.068
	Control	3 (10)	
Causes of tooth decay: frequency of sugary food intake	Test	20 (67)	0.067
	Control	26 (87)	
Definition of dental plaque	Test	14 (47)	0.035*
	Control	22 (73)	
Dental plaque and its relation to caries	Test	15 (50)	0.032*
	Control	23 (77)	

\*Statistically different at the 0.05 significance level.

not statistically significant ( $p > 0.05$ ) when compared between the two groups.

### 3.5 Practice

With regards to practice, we assessed the relationship between parental oral health knowledge and the actual practice of their

children. This was to identify whether the parents had their children practice what they preached. Of the 67% of parents of children with CS who acknowledged that toothbrushing should be performed twice daily, only 75% of their children practiced the correct frequency. In contrast, 90% of parents in the control group knew the correct toothbrushing frequency; however, only 70% of their children practiced this strategy (Table 2).

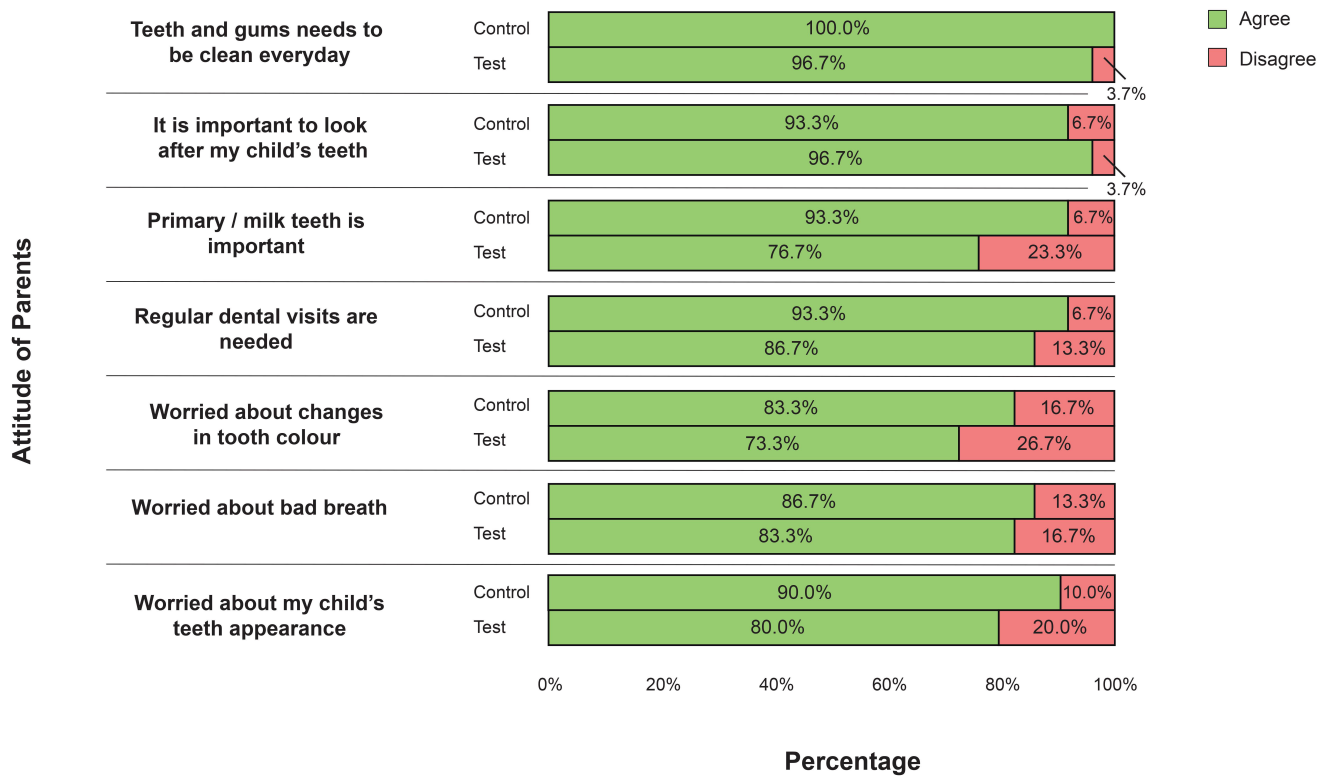


FIGURE 3. Attitude of parents of children with craniofacial syndromes and the control group towards their children.

TABLE 2. The relationship between parental oral health knowledge to the child's oral health practice in both groups of parents.

Questions in regard to parents' oral health knowledge	Percentage of parents that scored the correct answer, n (%)		Questions in regard to child's oral health practice	Percentage of parents that apply their knowledge onto their child, n (%)	
	Test group	Control group		Test group	Control group
How many times should you brush your teeth in a day?	20 (67%)	27 (90%)	How many times does your child brush his/her teeth in a day?	15 (75%)	19 (70%)
Can bacteria cause tooth decay and be transmitted from person to person?	7 (23%)	8 (27%)	Do you blow or share utensils with your child?	4 (57%)	5 (63%)
How often should you bring your child to visit the dentist for a regular checkup?	16 (53%)	23 (77%)	How often does your child visit the dentist?	10 (63%)	9 (39%)

\*The chi-square tests were performed on all pairs of test and control groups. Their p-values were all greater than 0.05.



When asked about the transmissibility of dental caries, 23% of parents of children with CS were able to answer the question correctly; of these parents, 57% of them blew on or shared their utensils with their children during feeding regardless. Similarly, 27% of parents in the control group were aware that dental caries was transmissible, yet 63% of them still blew on or shared their utensils with their children during feeding.

In total, 53% of parents of children with CS knew that they should take their child to their dentist every six months. However, only 63% of these parents practiced the correct frequency. In contrast, of the 77% of parents in the control group who knew they should take their child to the dentist every six months, only 39% did so.

## 4. Discussion

Owing to the COVID-19 pandemic and the frequent lockdown restrictions, the number of patients visiting the Combined Oro-Craniofacial Clinic at UMMC was reduced drastically from the usual 8 to 12 patients per month to half of that amount. However, the 30 participants involved in this study provide accurate representations of the views of parents with children who had been diagnosed with CS in Malaysia.

### 4.1 Knowledge

Our study revealed that parents from both groups were unaware that irregular tooth morphology can retain plaque and increase susceptibility to caries, particularly in molars where pits and fissures are present. This is especially worrying as the first molars are the earliest permanent dentition to erupt into the oral cavity [8]. Occlusal surfaces of the first permanent molars have been reported to be most likely to be diagnosed with caries within a short period of time after eruption due to high levels of plaque accumulation. This could be due to the lack of awareness amongst parents regarding the emergence of such teeth; hence their children's oral hygiene was overlooked [9]. Irregular tooth morphology can serve as a reservoir for bacteria to metabolize dietary sugars to produce organic acid; locally, this creates a low pH and subsequently causes demineralization of the tooth surface [10]. Thus, clinicians have been taking precautionary means over the years to abate this problem *via* the application of fissure sealants and fluoride varnish as soon as these teeth have erupted. Both groups of parents did not know that crowding is more susceptible to dental caries. In these cases, ineffective cleaning, especially in tight proximal contact areas, may lead to ineffective oral hygiene [11], thereby leading to the formation of proximal caries.

Dental caries has been reported to be able to transmit between individuals *via* saliva with *Streptococcus mutans* as the main etiological agent [12]. From our study, more than 70% of parents from both groups were unaware of this fact. This finding is similar to a study reported by Nassar *et al.* [13] in which a high percentage of parents in their study kissed or shared utensils with their child. To break the chain of caries transmission, parents and caregivers must avoid sharing utensils or blowing on food prior to feeding their children.

### 4.2 Attitude

Tooth discoloration has a broad array of causes. Some cases represent an indicator of tooth destruction, such as dental caries and microbial involvement; this leads to a black discoloration [14]. We found out that a quarter of parents of children with CS were not worried if there were any changes to their color of their children's teeth. This may be due to the fact that they are already preoccupied with multiple commitments for their child, namely medical follow-ups, speech therapy or psychological support; thus, oral health seems to be the least of their priorities [1]. A different study has shown that the children themselves, especially girls, are more disapproving of their tooth discoloration [15].

With regards to the importance of primary teeth as compared to permanent teeth, some respondents believed that the primary teeth are not as important as the latter; this was a similar discovery to a study reported previously by Jain *et al.* [6]. This finding is especially daunting because untreated carious primary teeth can cause harmful effects on a child's well-being. Their quality-of-life can be affected due to pain, along with acute and chronic infection, thus resulting in sleep disturbances [16], thereby affecting their performance in school. In contrast, parents in the control group performed well for this question (93.33% of those surveyed).

### 4.3 Practice

It is important to note that children with CS have different oral manifestations when compared to other children. The common presentations include retrognathic mandibles, limited mouth opening and hypodontia. Children with Apert syndrome may have severe crowding, delayed eruption and an anterior open bite [17] whereas Crouzon syndrome may manifest with impacted and malformed roots and crowns [18]. Thus, we need to be mindful of the challenges that these children may face when trying to maintain good oral hygiene.

There is an international consensus that brushing twice daily is recommended by most practitioners as it helps in effective plaque control [19]. However, our study found that a minority of the parents of children with CS claimed that their children only brushed once daily, despite knowing the correct frequency. This may be because these children have small jaws the small jaws which limit their mouth opening, thus impeding them from brushing their teeth properly. In addition to this, children with craniofacial syndromes who have undergone surgeries may face postoperative complications such as hypoplastic maxillae, disfigured dental arches and oronasal fistula [2]. These features would require a separate approach for them to achieve a disease-free oral cavity as compared to normal children. For instance, the presence of an oral fistula may cause the regurgitation of water during toothbrushing, thus leading to inconvenience for both the child and parent when attempting to practice good oral hygiene.

Some children with CS that have issues with their psychomotor and intellectual skills can further aggravate the situation especially when parents are attempting to gain their cooperation and focus during tooth brushing. This dampens the ability and motivation of both parties to apply correct oral hygiene practices. Therefore, parents are responsible to work

hand-in-hand with clinicians *via* oral health education during dental visits.

Fluoride has been proven to bring a plethora of advantages to our teeth, especially in reducing the prevalence of caries by increasing remineralization and reducing demineralization of the enamel [20]. In the present study, we found that all parents had incorporated a fluoride source in their daily schedule.

Despite acknowledging that dental visits should be undertaken every 6 months, 37.5% of the parents of children with CS were irregular attendees. This correlates with a study conducted by Al-Oufi and Omar [21] which revealed that most mothers felt that regular dental visits are not essential. A potential cause to this finding can be explained by Chi Adam *et al.* [1], who discovered that parents of children with CS experience financial restraints from the numerous medical follow-ups they need to take their child to. In fact, these parents were mostly committed to dual occupations and do not have much time to engage their children with dental appointments. Furthermore, the emergence of the COVID-19 pandemic led the parents to refrain from going outdoors as a safety precaution.

The primary limitation of this study is the small sample size; this was due to its low incidence in Malaysia (0.003%) [1]. Therefore, when non-statistically significant outcomes were identified, it was not evident whether this was due to a true non-statistically significant outcome or whether there was inadequate power (from the small number of samples). In addition, these samples were obtained from a single center *via* Google Forms rather than physical forms; consequently, potential selection bias may have occurred. Furthermore, the data related to the practice section were based on only the respondents that had answered correctly in the knowledge section to match the objective of the study. Thus, future studies can improve these aspects for better data representation and research outcomes.

## 5. Conclusions

Parents of children with CS have a relatively similar depth of knowledge towards oral health as parents in the control group. However, the parents of children with CS have slightly less positive attitudes. Both groups of parents showed poor application of their knowledge to their children's oral hygiene practice. Dental professionals are responsible for educating and motivating the parents of children with CS. This can be achieved *via* oral health education and dietary counselling during appointments to apply their knowledge to their children to improve their oral hygiene, thereby enhancing their quality of life.

## ABBREVIATIONS

KAP, knowledge, attitude and practice; UMMC, Universiti Malaya Medical Center; CS, craniofacial syndrome.

## AVAILABILITY OF DATA AND MATERIALS

The data presented in this study are available on reasonable request from the corresponding author.

## AUTHOR CONTRIBUTIONS

NAR and NFAMF—designed and performed the research study. NZA and FH—provided help and advice. NAA—analyzed the data. All authors contributed to editorial changes in the manuscript. All authors read and approved the final manuscript.

## ETHICS APPROVAL AND CONSENT TO PARTICIPATE

The study was approved by the Medical Ethics Committee, Faculty of Dentistry, Universiti Malaya (DF CD2114/0043 (U)). Written informed consent was obtained from all parents prior to the study.

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## CONFLICT OF INTEREST

The authors declare no conflict of interest.

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