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Awareness On Phace Syndrome Among Dental Students.

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Abstract

INTRODUCTION:
PHACE syndrome comprises posterior fossa malformations, haemangioma, arterial anomalies, cardiac defects, eye anomalies and less commonly, sternal cleft or supraumbilical raphe. It is a neurocutaneous condition in which extensive or segmental Infantile haemangioma, usually on the face, scalp, or cervical region, can be associated with posterior fossa malformations, arterial abnormalities. PHACE syndrome can rarely cause oral manifestations also.

AIM:
To assess the knowledge and awareness on PHACE syndrome among the dental students.

MATERIALS AND METHOD:

This is an online study setting with a sample size of 100 dental practitioners. The sampling method used is non-probability convenience sampling with minimal sampling bias. The questionnaire comprised 20 questions and the method of representing each output variable was pie chart and bar graph. The results were analyzed using SPSS statistical software. Chi square test and pearson correlation was done to check the association, and a *p-value* of less than 0.05 was said to be statistically significant.

RESULTS:

From the present study, third year students comparatively have more awareness and knowledge about PHACE syndrome, 36% were aware that PHACE syndrome can cause dental root abnormalities($p= 0.059$, $p>0.05$), 34% were aware that PHACE syndrome can cause enamel hypoplasia($p=0.098$, $p>0.05$), 24%

were aware about the teeth that are most likely to get affected by PHACE syndrome($p=0.018$, $p<0.05$), 36% were aware that PHACE syndrome can cause localized upper lip infantile hemangioma($p=0.026$, $p<0.05$).

CONCLUSION:

Within the limits of the study it can be concluded that third year students had a high level of awareness and knowledge about the syndrome when compared to the other study year students.

Key Words: Hemangioma; Hypoplasia: Infants; PHACE; Syndrome.

INTRODUCTION:

PHACE syndrome is a neurocutaneous syndrome that refers to the association of large, plaque-like, segmental hemangiomas of the face, with one or more of the following features or anomalies: Posterior fossa brain malformations, Arterial cerebrovascular anomalies, Cardiovascular anomalies, Eye anomalies, and Ventral developmental defects, especially sternal defects and supraumbilical raphe(1). Among these the most remarkable feature of PHACE syndrome is Infantile hemangioma, and abnormalities involving brain, aortic, thoracic, and cervical arteries, are not obvious under clinical examination, however they have considerable potential for morbidity(2).

PHACE syndrome is observed in 2% to 3% of Infantile hemangioma cases. It is considered as one of the most frequent neurocutaneous vascular disorders occurring in childhood. According to recent studies, when the Infantile hemangioma develops on the face irrespective of whether it is segmental or large, the probability of it being associated with PHACE syndrome is about 20% to 31%. PHACE most commonly affects females than males (9:1)(3). The pathogenesis of PHACE syndrome is still unknown, however it has been postulated that it might result from defective embryogenesis occurring between 3rd and 12th week of gestation, before or during the vasculogenesis, because certain malformations and Infantile hemangioma can affect the same side of the body. There is no evidence of genetic abnormalities that could possibly contribute to the development of PHACE syndrome(4). In 2009, Various medical specialties established criteria for the diagnosis of PHACE syndrome which are stratified into 2 categories: The first one being, PHACE syndrome, defined by the presence of segmental Infantile hemangioma larger than 5cm on the face, scalp, or cervical region, associated with 1 major criterion or 2 minor criteria. Secondly, possible PHACE syndrome, defined by the presence of Infantile hemangioma and 1 minor criterion. Major and minor criteria of PHACE syndrome have been determined, based on the vascular and structural involvement of the brain, cardiovascular system, eye, and midline(5). Certain patients with PHACE syndrome may have dysphagia, feeding difficulties, and delay in speech. These features are usually seen in patients with posterior fossa brain malformations; lip, oropharyngeal, and airway infantile hemangiomas; and history of cardiac surgery. Dysphagia can be secondary to the location of the Infantile hemangioma like lips, oral cavity, and pharynx or to even motor coordination(6,7).

With respect to dental abnormalities, enamel hypoplasia may be a feature of PHACE syndrome due to an intraoral hemangioma.. Enamel hypoplasia also increases the incidence and risk of dental caries. In recent studies, children with PHACE syndrome exhibited root abnormalities especially in permanent first molars. The crown portion of the tooth looked normal but roots of the permanent first molars could barely be seen and were very short with tapered stumps(8).

Our team has extensive knowledge and research experience that has translated into high quality publications.(9–17),(18–23),(24–30). .

The aim of the present study is to assess the knowledge and awareness on PHACE syndrome among the dental students.

MATERIALS AND METHOD:

This is a cross sectional study conducted with a sample size of 100 dental students in chennai. Ethical approval for the study was obtained

from the International review board prior to the start of the study. A self- structured questionnaire consisting of 15 questions was framed based on knowledge and awareness about PHACE syndrome using google forms, an online survey application. The questions were checked for validity by three internal experts and three external experts.

The questionnaire was distributed to the participants through various social media platforms. The sampling method used in this study is non-probability convenience sampling. In order to minimise the sampling bias, simple language and concepts were used. The Inclusion criteria was the study year and the exclusion criteria was age and gender. These questions were carefully studied and corresponding answers were marked by 100 dental students.

The responses from 100 dental students were received and results were analysed using SPSS software version 23 and represented as bar graphs. Chi square test and pearson correlation was done to check the association, and a *p-value* of less than 0.05 was said to be statistically significant.

RESULTS:

Out of 100 participants, 87% were about PHACE syndrome (Figure 2), among which 50% were aware about the abbreviation of PHACE (Figure 3). 65% of the population were aware that Infants are most likely to get affected by PHACE syndrome and 66% were aware that this syndrome is not fatal (Table 1). 75% were aware about the type anomaly that occurs at a higher rate in PHACE syndrome and 83% were aware that PHACE syndrome can cause oral manifestations (Table 1). 78% of the population were aware that clinicians should refer children with PHACE syndrome to a pediatric dentist by 1 year of age and only 25% were aware that propranolol is the beta blocker preferred for managing PHACE syndrome (Table 1)

The results reveal that third year students comparatively have more awareness and knowledge about PHACE syndrome, 36% were aware that PHACE syndrome can cause dental root abnormalities (Figure 4), 34% were aware that PHACE syndrome can cause enamel hypoplasia (Figure 5), 24% were aware about the teeth that are most likely to get affected by PHACE syndrome (Figure 6), 36% were aware that PHACE syndrome can cause localized upper lip infantile hemangioma (Figure 7).

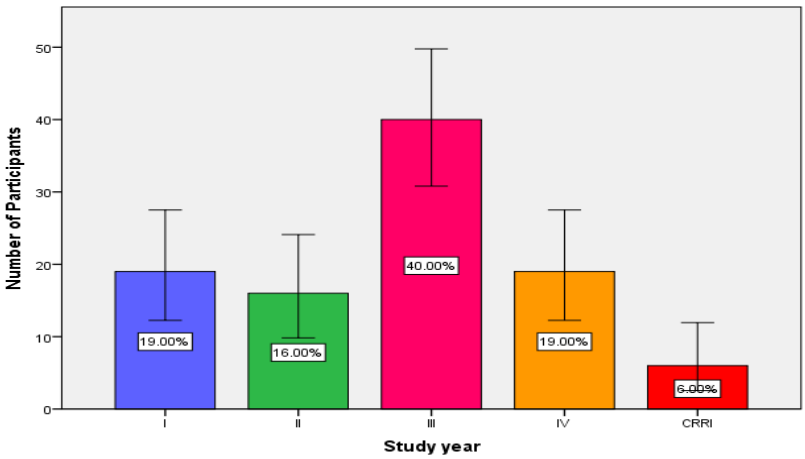

Figure 1: Bar chart showing percentage distribution on year of study. X-axis represents the study year and the Y-axis represents the number of participants. Blue colour represents Ist year students (19%), green colour represents IInd year students (16%), pink colour represents IIIrd year students (40%), yellow colour represents IVth year students (19%) and the red colour represents CRRI students(6%).

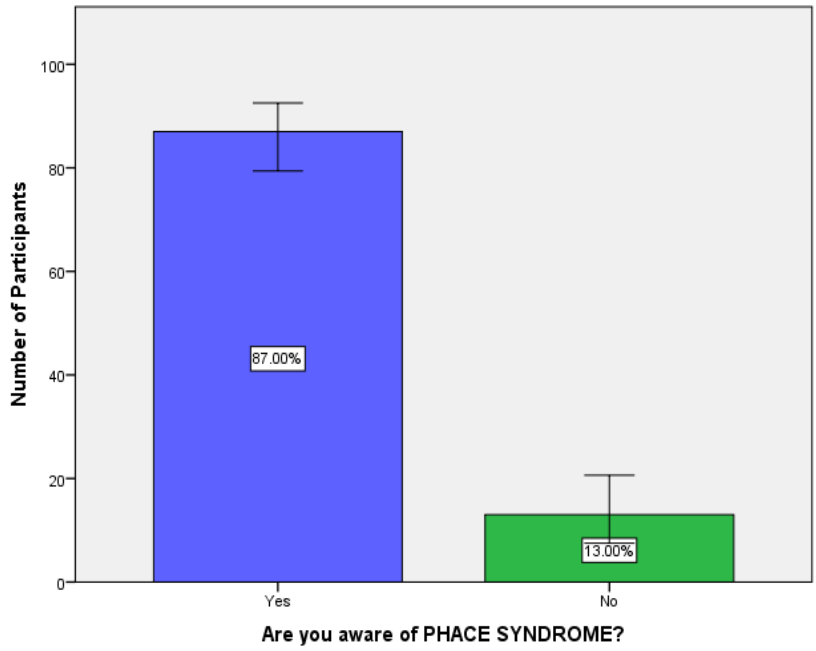

Figure 2: Bar chart showing percentage distribution on awareness of PHACE syndrome. The X-axis represents awareness and the Y-axis represents the number of participants. Blue colour represents Yes (87%) and the green colour represents No (13%).

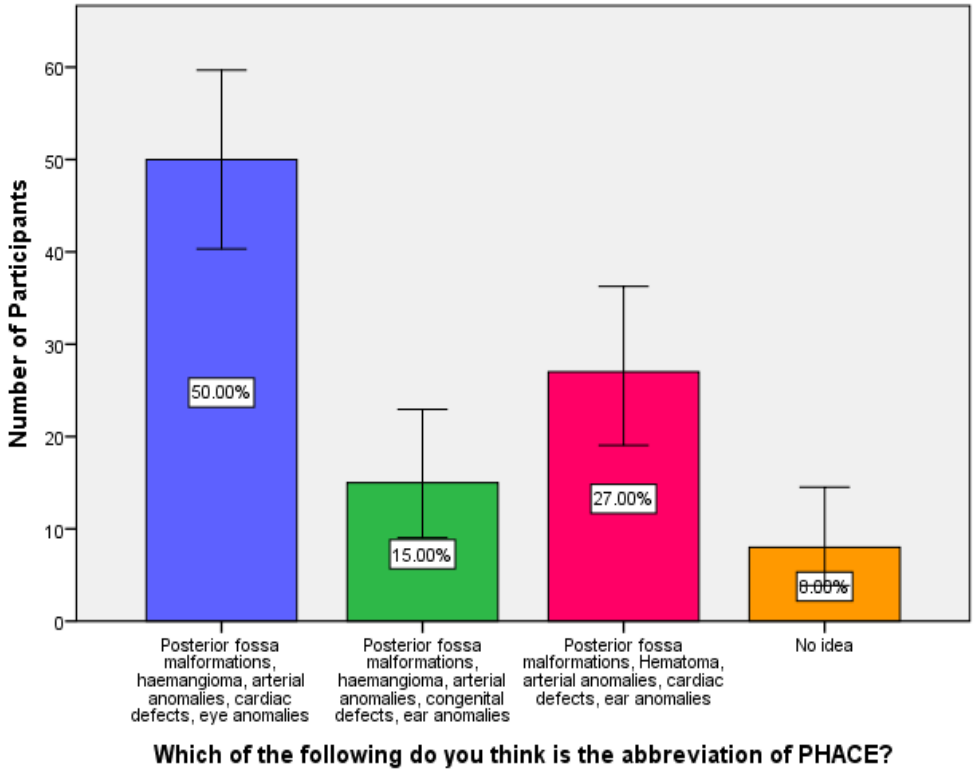

Figure 3: Bar chart showing percentage distribution about knowledge on abbreviation of. X-axis represents the study year and the Y-axis represents the number of participants. Blue

colour represents Posterior fossa malformations, haemangioma, arterial anomalies, cardiac defects, eye anomalies (50%), green colour represents Posterior fossa malformations, haemangioma, arterial anomalies, congenital defects, ear anomalies (15%), pink colour represents Posterior fossa malformations, hematoma, arterial anomalies, cardiac defects, ear anomalies (27%), and yellow colour represents no idea (6%)

Table 1: Table showing the percentage distribution of the responses to the questionairre.

Question	Response	Percentage of responses
Who is most likely to get affected by PHACE syndrome?	Infants	65%
	Children	21%
	Adults	8%
	No idea	6%
Which type of anomaly occurs at a much higher frequency in PHACE syndrome?	Arterial anomalies	75%
	Venous anomalies	6%
	None	10%
	No idea	9%
Is PHACE syndrome fatal?	Yes	26%
	No	66%
	Not aware	8%
Are you aware that PHACE syndrome can cause oral manifestations too?	Yes	83%
	No	17%
Are you aware that PHACE syndrome can cause dental root abnormalities?	Yes	79%
	No	21%
Are you aware that PHACE syndrome can cause Enamel hypoplasia?	Yes	80%
	No	20%
Which teeth are most likely to get affected by PHACE syndrome?	Permanent first molars	54%
	Primary first molars	10%
	Primary second molars	24%
	No idea	12%
Are you aware that PHACE syndrome can cause localized upper lip infantile hemangioma?	Yes	86%
	No	14%
Do you know that clinicians should refer children with PHACE syndrome to a pediatric dentist by 1 year of age?	Yes	78%
	No	22%
Which beta blocker is preferred in case of PHACE syndrome?	Propranolol	25%
	Nadolol	54%
	Both	12%
	No idea	9%

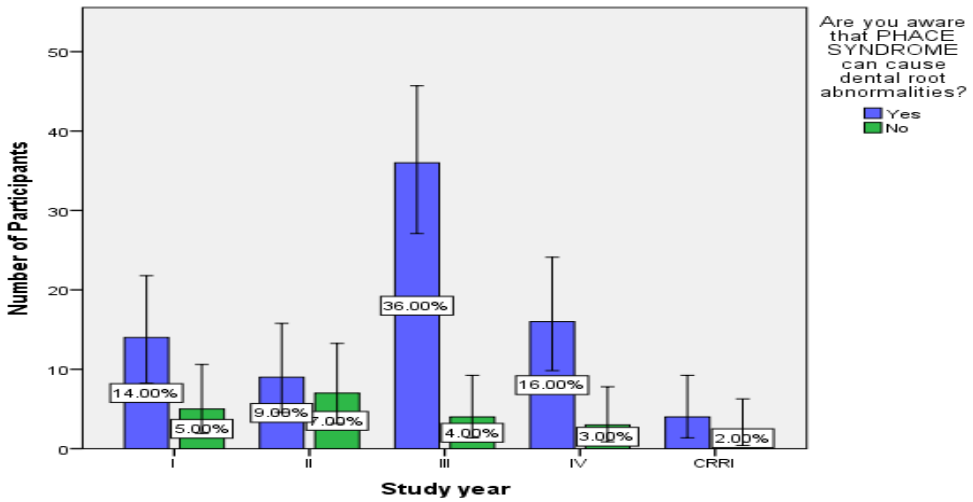

Figure 4: Bar graph showing association between the study year and knowledge on PHACE syndrome causing dental root abnormalities. Blue colour denotes Yes and green colour denotes No. X-axis represents the study year and the Y-axis represents the number of participants. Among Ist year students, 14% answered yes and 5% answered no. Among IInd year students, 9% answered yes and 7% answered no. Among IIIrd year students, 36% answered yes and 4% answered no. Among IVth year students, 16% answered yes and 3% answered no. Among CRRI students, 4% answered yes and 2% answered no. Chi square test was done and association was found to be statistically insignificant. Pearson's chi square value:9.094, df: 4, p value: 0.059 (p>0.05) hence statistically insignificant.

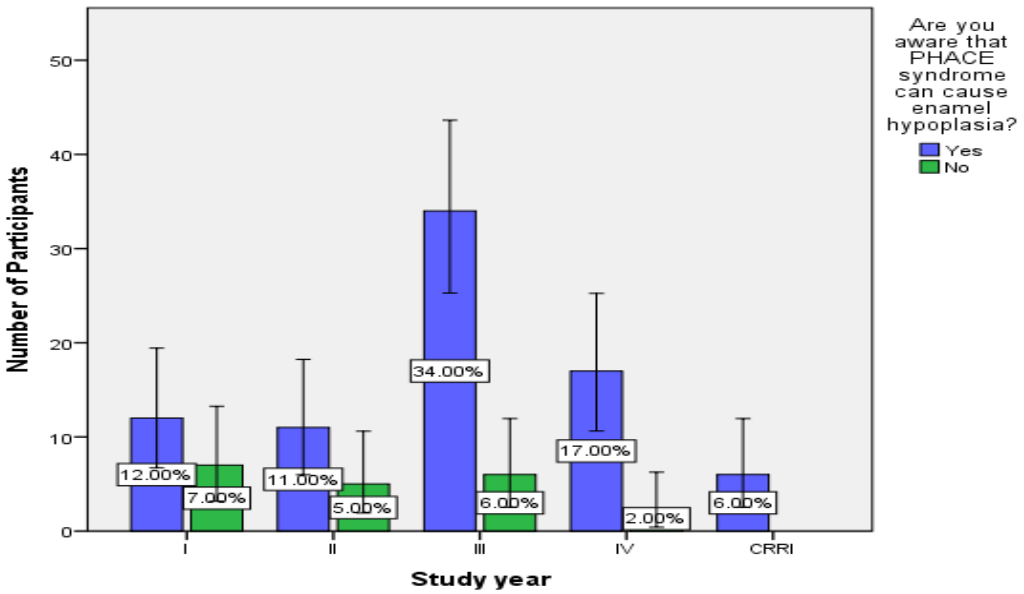

Figure 5: Bar graph showing association between the study year and knowledge on PHACE syndrome causing enamel hypoplasia. Blue colour denotes Yes and green colour denotes No. X-axis represents the study year and the Y-axis represents the number of participants. Among Ist year students, 12% answered yes and 7% answered no. Among IInd year students,11% answered yes and 5% answered no. Among IIIrd year students, 34% answered yes and 6%

answered no. Among IV[th] year students, 17% answered yes and 2% answered no. Among CRRI, all the students answered yes. Chi square test was done and association was found to be statistically insignificant. Pearson's chi square value:9.094, df: 4, p value: 0.098 (p>0.05) hence statistically insignificant.

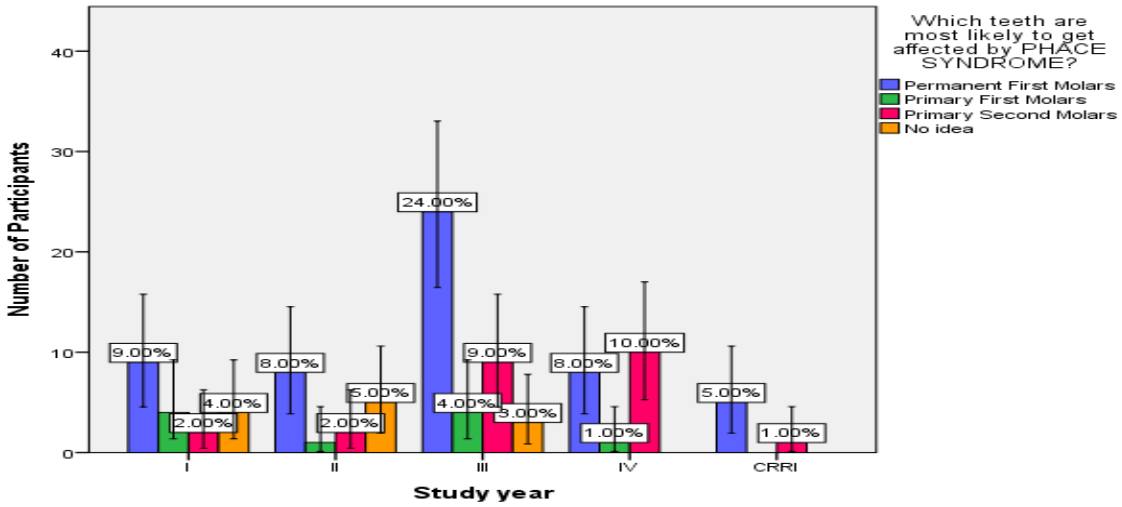

Figure 6: Bar graph showing association between the study year and knowledge on PHACE syndrome can most likely affect permanent first molars. Blue colour denotes permanent first molars, and green colour denotes primary first molars, yellow colour denotes primary second molars, and pink colour denotes no idea. X-axis represents the study year and the Y-axis represents the number of participants. Among I[st] year students, 9% answered permanent first molars. Among II[nd] year students,8% answered permanent first molars. Among III[rd] year students, 24% answered permanent first molars. Among IV[th] year students, 10% answered primary second molars. Among CRRI, 5% answered permanent first molars. Chi square test was done and association was found to be statistically significant. Pearson's chi square value:24.389, df: 12, p value: 0.018 (p<0.05) hence statistically significant.

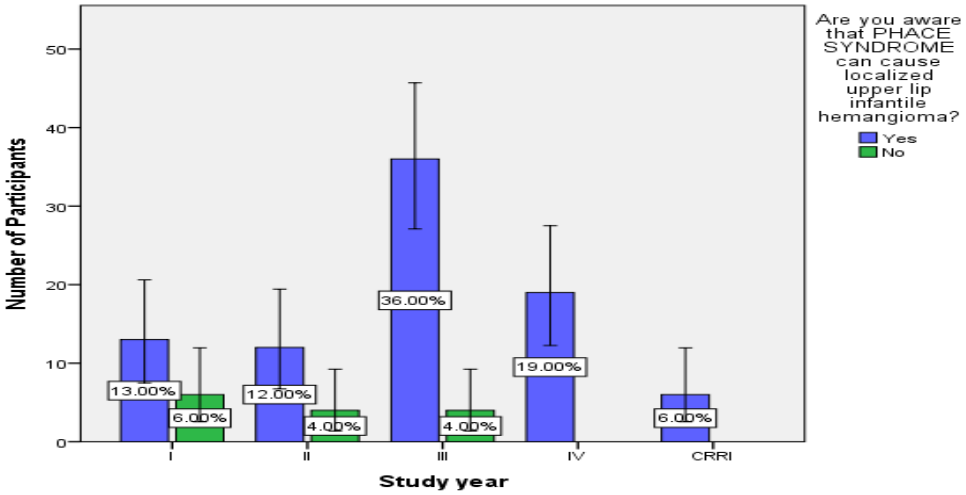

Figure 7: Bar graph showing association between the study year and knowledge on PHACE syndrome causing localized upper lip infantile hemangioma. Blue colour denotes Yes and green colour denotes No. X-axis represents the study year and the Y-axis represents the

number of participants. Among Ist year students, 13% answered yes and 6% answered no. Among IInd year students,12% answered yes and 4% answered no. Among IIIrd year students, 36% answered yes and 4% answered no. Among IVth year students and CRRI, all the students answered yes. Chi square test was done and association was found to be statistically significant. Pearson's chi square value:11.086, df: 4, p value: 0.026 ($p<0.05$) hence statistically significant.

DISCUSSION:

In the present study, 36% of IIIrd year students, 16% of IVth year students, 14% of Ist year students, 9% of IInd year students and 4% of CRRI were aware that PHACE syndrome can cause dental root abnormalities(Figure 3). Youssef et al.,2019 presented four children with PHACE syndrome who have absence of or severe malformation of the roots of their permanent first molars. Root abnormalities in the children's molars were bilateral and not restricted to the segments affected by cutaneous hemangioma. The reason for root abnormalities is unknown, but given the rarity of these findings in healthy children, it is likely an additional dental manifestation of PHACE syndrome. The absence of functional roots in the permanent first molars can result in significant consequences(31). In the present study, 24% of IIIrd year students, 9% of Ist year students, 14% of Ist year students, 8% of IInd year students and IVth year students, and 5% of CRRI were aware that PHACE syndrome can most likely affect permanent first molars.

The etiology of the enamel hypoplasia seen in individuals with PHACE is unclear. The fact that the enamel hypoplasia was limited to patients with intraoral hemangioma suggests that a common primary event, process, or pathway is responsible for both or that the presence of abnormal vessels in the oral cavity has a direct secondary effect on enamel formation. Children with enamel hypoplasia are at higher risk of developing caries. Dietary factors of frequent or prolonged exposure of the teeth to food products sweetened with fermentable sugars may exacerbate this risk. Pediatric formulations of any medication can also promote dental caries through the presence of syrup or acidic pH. Because many children with PHACE syndrome are treated with propranolol or prednisolone, they may be at even greater risk of developing caries if enamel hypoplasia is present(8,32,33). In the present study, 34% of IIIrd year students, 17% of IVth year students, 12% of Ist year students, 11% of IInd year students and 6% of CRRI were aware that PHACE syndrome can cause enamel hypoplasia(Figure 4) and 19% of IIIrd year students, 11% of IVth year and Ist year students, 8% of IInd year students and 5% of CRRI were aware that propranolol is the beta blocker preferred in case of PHACE syndrome.

In a cohort study performed by Cawthorn et al., 2019 with infants having infantile lip hemangioma associated with PHACE syndrome. Approximately half (48.0%) of all lesions were of mixed depth (consisting of both a superficial and deep component); 31.4% were exclusively superficial, and 20.6% were exclusively deep. Hemangiomas were most commonly located on the upper lip (58.8%), as compared to the lower lip (35.3%); a minority was found at the commissure (5.8%). Violation of the vermillion border occurred in 39.2% of cases, and the oral mucosa was involved in 54.9% of cases(34). In the present study, 36% of IIIrd year students, 19% of IVth year students, 13% of Ist year students, 12% of IInd year students and 6% of CRRI were aware that PHACE syndrome can cause localised upper lip infantile hemangioma(Figure 6).

CONCLUSION:

Within the limits of the study it can be concluded that third year students had a high level of awareness and knowledge about the syndrome when compared to the other study year students. Long term documentations of such cases will enhance the understanding of PHACE syndrome and its manifestations thereby improving the management of PHACE syndrome in future and its related conditions.

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CONFLICT OF INTEREST:

No potential conflict of interest relevant to this article was reported.

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