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Tel : (+603) 6126 6633
Fax : (+603) 6126 6103
E-mail : annapurny@uitm.edu.my
Website : <https://mapd.my>

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Alia Aqilah Binti Mohd Suhaimi
Klinik Pakar Pergigian Pediatrik
Hospital Tunku Azizah, Kuala Lumpur

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TABLE OF CONTENT

73

73-77

NARRATIVE REVIEW

Paediatric Dentistry In Malaysia: From Evolution To The Horizon
Bahadun J

78

78-89

ORIGINAL RESEARCH

Evaluating The Knowledge Of Medical Practitioners Towards Traumatic Dental Injuries In The Emergency Department Of Hospital Serdang

Nair V, Subramaniam SD

90-100

Post-operative Pain Control in Children After Dental Extractions Under General Anesthesia (GA): A Service Evaluation Utilizing the Modified Post Anesthetic Discharge Scoring System (PADSS) Form

Jamil A, Vijayakumar T, Norazman Shah NN

101-112

Prevalence Of Unerupted Permanent Maxillary Central Incisors Among Primary School Children In Perak, Malaysia

Saharudin S, Perumal S, Ibrahim R, Chan YP, Mahussain WA, Vijayakumar M

113-124

Knowledge and Perception of Dental Trauma Among Students At A Faculty of Education

Al-Naser MG, Venkiteswaran A, Md Sabri BA, Abdul Halim R, Zulkifli AF

125-132

Oral Health Related Quality of Life (OHRQoL) In Children After Comprehensive Dental Treatment Under General Anaesthesia

Mohd Kasim F, Olikh SK, Bahadun J

133

133-138

CASE REPORTS

Oral Intravascular Papillary Endothelial Hyperplasia in a child: A Case Report

Ong YY, Sockalingam G

Paediatric Dentistry in Malaysia: From Evolution to the Horizon

Juanna Bahadun¹

(1) Department of Paediatric Dentistry, Hospital Shah Alam.

Corresponding Author

Juanna Bahadun

Address

Department of Paediatric Dentistry,
Hospital Shah Alam
Selangor, Malaysia

Email

juannabahadun@yahoo.com

DOI

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Paediatric dentistry in Malaysia has come a long way since its first inception in 1987; when it was approved and recognized as a specialty by the Ministry of Health Malaysia (MOH). The first paediatric dental specialist in the MOH, Dr. Noraini Nun Nahar Yunus, came back from postgraduate study in New Zealand in 1986 and went on to head the first Paediatric Dental Unit in the country within the newly established Institute of Paediatrics, Kuala Lumpur Hospital in 1991. By eight years later, 3 more paediatric dental specialists had come back from their postgraduate study in the United Kingdom (UK). The new specialists were entrusted to establish the specialty on a regional basis; namely south in Johor Bharu, north in Ipoh and east coast in Kuala Terengganu.

From then on, more young dental officers were sent abroad on scholarships to specialize in the field. Upon returning to the country, these specialists were posted to state hospitals to develop the service in the state. Currently, for MOH, paediatric dental service is available in all states in the country.

In 2007, began a new chapter in the paediatric dentistry fraternity in the country. The Malaysian Association of Paediatric Dentistry (MAPD) was founded on 27th May 2007 by several bold and visionary pioneer members. The paediatric dental specialists scattered in various institutions, were brought together under one umbrella. An association of our own allowed us to conduct scientific programmes, hands-on courses and put forward our ideas as one voice. Indeed, just a year after its formation, MAPD took the mammoth challenge of hosting the 6th Paediatric Dentistry Association of Asia (PDAA) and came out triumphant and became a model for others.

With regards to capacity-building of the specialty, year 2000 became a game-changer. The University of Malaya embarked on the first local paediatric dentistry postgraduate programme. National University of Malaysia accepted the first batch of paediatric dentistry postgraduate students in 2010, and soon after Universiti Sains Malaysia came on board. Henceforth, with local and overseas

training in paediatric dentistry made available, the number of paediatric dental specialists thrived. To date, from local postgraduate Paediatric dentistry programmes alone, 57 specialists have graduated and 16 more are currently in training. As of 2019, there were 80 paediatric dental specialists in the country with the majority in the government service, while the remaining numbers are in the teaching institutions, private practice and army dental service.

Over the years, paediatric dental service continued to escalate and became an integral part of comprehensive care of the paediatric patient. Certainly, there was a marked increase in total number of patients attending the Paediatric Dental specialist clinics in the Ministry of Health from 2015 through to 2019.¹ A similar trend was seen in new patients presenting to the Paediatric Dental facilities. In line with the increasing number of patients, workload in treatment rendered exhibited a similar increasing pattern.

Table 1 *Number of New Patient and Total Attendance for Paediatric Dental Specialty in MOH, 2015 to 2019 (Oral Health Programme Annual Report 2019)¹*

	2015	2016	2017	2018	2019
New Patient	32,130	34,637	37,009	42,600	45,548
Total Attendance	94,669	103,754	113,164	128,418	136,559

Table 2 *Treatment rendered by Paediatric Dental Specialists MOH, 2015 to 2019 (Oral Health Programme Annual Report 2019)¹*

	2015	2016	2017	2018	2019
Trauma	4,026	5,089	5,667	7,367	5,660
Pulp Therapy	2,789	2,829	2,753	3,519	6,338
Oral surgery	3,013	3,279	3,334	3,839	3,388
Restorations	33,588	36,763	39,783	45,729	49,301

National Oral Health Survey for Pre-school (2015) demonstrated that caries prevalence for 5-year-olds was 71.5%.² Mean DMFT

Score for 12-year-olds for 2019 was 0.68, and 1.31 for 16-year-olds.¹

Table 3 Mean DMFT Score for 12 and 16 year-olds, 2015 to 2019 (Oral Health Programme Annual Report 2019)¹

	2015	2016	2017	2018	2019
12-year-olds	0.82	0.79	0.75	0.71	0.68
16-year-olds	1.35	1.34	1.4	1.35	1.31

Caries experience showed evidence of improvement among the 12-year-olds in Malaysia. Dental caries prevalence among 12-year-old schoolchildren demonstrated a decline from 41.5% in 2007 to 33.3% in 2017.³

Children with special needs are patients categorized as those with physical

abnormalities, mental or learning issues, those with medical problems and children who have multiple issues. Number of cases of children with special needs cases attended to by paediatric dental specialists in MOH showed a steady incline from 2015 to 2018 and a slight dip in 2019.

Table 4 Children with Special Needs seen by Paediatric Dental Specialists (Oral Health Programme Annual Report ,2019)¹

2015	2016	2017	2018	2019
31621	33801	36055	38958	36600

The prevalence of 12-year-olds with traumatised anterior teeth demonstrated and increasing trend from 2.5% in 1997 to 5.4% in 2007 to 7.2% in 2017.² The same survey revealed that unfortunately only 17.7% had treatment for their traumatic injuries, while majority of the children did not receive treatment.

The data from the Department of Statistics Malaysia showed that there were 8.3 million children (0-16 years old) in 2019. Hence the ratio of paediatric dentists per 100,000 children in the country in 2019 was 0.86 per 100,000. In comparison, ratio of paediatric dentists per 100,000 children in the UK was 1.88 per 100,000.³ It is projected that by 2030,

number of children in Malaysia will increase to 10.3 million.⁵

In view of the expanding paediatric age group in the country, it is evident that the number of paediatric dental specialists must grow in tandem to meet the expected needs and demand of the increasing children population. To achieve that, all sectors of the specialty must play their role. It would be ideal if universities will provide more placements for postgraduate training in the field. At the same time, more scholarships will be made available to the aspiring young dental officers who are keen to do postgraduate in paediatric dentistry. New pathways must be explored to provide more

avenues for the young, enthusiastic dentists to pursue postgraduate studies without being fully dependent on the limited number of scholarships.

The importance of development of paediatric dental service in district hospitals cannot be emphasized enough. These will allow the paediatric population in the districts to access specialized care that would otherwise be difficult.

On the other end of the spectrum, established paediatric dental centres should be identified to focus on niche areas and given support to develop more advanced service. Other than support in infrastructure, capacity building must also be looked into. This is in the form of sending specialists for structured training in the specific areas, locally and abroad. These centres will play a crucial role in providing clinical service in these niche areas, as well as becoming training centres for other specialists in the country. In addition to service provision and

training centres, they will also lead in high quality, multi-centre research in various aspects of paediatric dentistry. These proposals are by no means a detailed roadmap, but rather some ideas to be added and expanded to for future actions.

In conclusion, oral health is an integral component in the overall health of a child. As a dental specialty that is age-defined, paediatric dental specialists play a key role in ensuring access to high-quality oral health care for children from birth through adulthood. Paediatric dentistry in Malaysia has thrived in leaps and bounds. Uptake of service has been on an upward trend and yearly more and more young specialists graduate from the programme which augurs well for the specialty. Nonetheless, we must not underestimate the challenge of meeting the oral health needs of an expanding paediatric population. Opportunities to grow must be constantly sought and new avenues explored with the paramount aim of providing the best care to our patients.

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Evaluating the Knowledge of Medical Practitioners Towards Traumatic Dental Injuries in The Emergency Department of Hospital Serdang.

Vilashni Nair¹, Shanthini Devi Subramaniam¹

ABSTRACT

Aim: To assess the knowledge of the emergency room medical practitioners regarding traumatic dental injury as medical practitioners of the ER are stationed at the frontlines of the hospital.

Methods: A close ended questionnaires consisting of 28 questions adapted from previous studies was handed personally to a group of 60 medical practitioners in the Emergency department of Hospital Serdang.

Results: The response rate of the subjects were seen to be 100%. Forty percent were seen to have fairly good knowledge regarding the basic management of traumatic dental injuries. It was also observed that about 73.8% knew the importance of extra oral time of an avulsed tooth. The storage media for an avulsed tooth which was deemed to be important by the majority practitioners were found to be ice cubes with the result of 41.7%. Majority of the practitioners was interested to increase their knowledge on the management of traumatic dental injuries.

Conclusion: Most participants had reasonable amount of knowledge, however their dilemmas towards certain aspect of management of traumatic dental injuries can be solved with further education and awareness by the dental team in order to further improve the quality of dental treatment given to patients attending the Emergency Department.

Keywords: *traumatic dental injuries, knowledge, medical practitioners*

(1) Department of Paediatric Dentistry, Hospital Serdang
Ministry of Health, Malaysia

Corresponding Author

Vilashini Nair

Address

Department of Paediatric Dentistry
Hospital Serdang.

Telephone Number

-

Email

vilashninair@gmail.com

DOI

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INTRODUCTION

Traumatic dental injuries are considered the most serious oral health problem amongst children and adolescents. One variant of traumatic dental injury is tooth avulsion which is defined as total dislodgment of the tooth from its socket. It represents about 16% of all traumatic dentoalveolar injuries in the permanent dentition.¹ Epidemiological studies indicate the annual incidence of dental trauma globally was at about 4.5%.² Crown fractures form the most frequent type of injury, comprising about 26%–76% of injuries to the permanent dentition³ whereas luxation injuries comprise 15%–61% of all dental injuries and mainly involving maxillary central incisors.⁴ Luxation injuries are the most common type in primary dentition whereas crown fractures are common in permanent dentition.

Maxillary central incisors have been regarded as the most affected tooth due to their labial projection making them more susceptible to facial trauma. These teeth have minimal resistance to frontal impact which are commonly seen among children aged 7–9 years because of the lack of the periodontal ligament's resiliency and the low mineralization of the surrounding bone.⁵ Permanent anterior teeth are not only essential for aesthetics but also for phonetics, mastication and psychological health of young patients. Thus, an immediate and appropriate management of the avulsed tooth by replantation is critical for long term prognosis, especially in children. Failure to replant the tooth will lead to expensive, time-consuming, and complex multidisciplinary approaches.⁶ Incorrect handling and late referrals to the dentist will lead to tooth

resorption and tooth ankyloses, resulting in poor prognosis. Other injuries such as luxative injuries and complicated crown fractures should also require prompt referral in order to ensure increased possibility of saving the tooth.

The prognosis of tooth replantation is dependent on factors such as time of endodontic intervention, storage media, extra-alveolar period and type of retention employed. The most critical factor for an excellent prognosis is the preservation of viable periodontal ligament cells (PDL) covering the root and this can be established by storing the tooth in a suitable media such as milk, saliva, or saline.³

Tooth avulsion usually occurs because of road traffic accidents, falls and other physical impacts. It could happen in schools during sports or because of physical violence. In these situations, parents usually take their children to visit the emergency department of the hospital to seek treatment. Therefore, to ensure proper management of traumatic dental injuries, it is important for the medical practitioners in the emergency department to have sufficient knowledge about the emergency management of such cases and with that avoid late referrals to the dentist.⁷

Several studies have shown that medical physicians lack adequate knowledge regarding the emergency management of traumatic dental injuries and avulsed tooth.^{8, 9, 10, 11} Up to this point, no studies focusing on medical practitioners have been reported in Malaysia. Therefore, the purpose of this study is to evaluate the knowledge and awareness of the medical practitioners from various age groups, designation and years of experience towards the management of traumatic dental

injuries in the emergency rooms of one major hospital in Malaysia and to evaluate their attitude towards receiving further dental education

METHODS

This is a cross-sectional study among medical practitioners consisting of house officers, medical officers and specialists on duty in the emergency department of Hospital Serdang. This present study is a cross-sectional survey with a sample size of 60 medical professionals from the emergency department of Hospital Serdang. All participants who were willing to participate were included in the study.

This study targets included medical practitioners working in the emergency departments comprising of specialists, medical officers, and house officers assigned to the green, yellow and red zones. A sample of 60 practitioners were collected from the emergency rooms during the period of the study. A 28-item closed ended questionnaire was as an instrument of the study.¹⁰ Permission for the modification and adaptation of the questionnaire for this study has been obtained appropriately from the author, Dr.K. Subhash raj, University of Toronto, Canada. Confidentiality and privacy of subjects were maintained as the questionnaire did not require names or contact numbers of the participants. A consent form was attached to the questionnaire explaining the purpose of the study. It stated that the participation in this survey is voluntary, and anyone can choose not to participate in the research or exit the survey at any time without penalty.

This study was analysed using the Statistical Package for the Social Sciences (SPSS) and Microsoft excel using SPSS. Section A will be the tabulation of demographic data and years of working experience of the medical practitioners involved in the survey. In the section of knowledge assessment (Section B), each correct answer will be given a score of one (1) and for a wrong answer will be given a score of zero (0). The total score for this section will be from zero (0) to fifteen (15), zero being no knowledge and fifteen being highly knowledgeable. Thus, the level of knowledge will be assessed through the determination of total score, namely poor knowledge score being (0– 8.0), and a good knowledge score being (9.0 – 15.0). The guidelines of the International Association of Dental Traumatology will be used as a reference for correct answers (Andersson, Andreasen, et al., 2012; DiAngelis, Andreasen, et al., 2012; Malmgren, Andreasen, et al., 2012). In Section C (Assessment of Attitude of Medical Practitioners towards receiving knowledge in management of traumatic dental injuries), each of the four (4) questions will be analysed independently.

This survey has been approved by National Medical Research Register (NMRR), with implementation of the National Institute of Health (NIH) guidelines on the conduct of research in the Ministry of Health Malaysia (MOH). The registration number of this research paper is NMRR-19-4018-51489(IIR).

METHODS

The majority the subjects were female (60%). About 78% were from the age group of 25 to 30 years old, 11.7% was from the age group

of 30 to 35 years old and about 10 % were from the age group between 35 to 45 years old. Out of this group there were 6 specialists (11.7%) ,23 medical officers (38.3%) and about 31 house officers (51.7%). (Table1)

Table 1 Socio demographic characteristics of respondents (n= 60)

	n	%
<i>Gender</i>		
Male	24	40.0
Female	36	60.0
<i>Designation</i>		
Specialist	6	10.0
Medical Officer	23	38.3
House Officer	31	51.7
<i>Age</i>		
25 – 30 years old	47	78.3
30 – 35 years old	7	11.7
35 – 45 years old	6	10.0
<i>Number of years working in the Emergency Department</i>		
< 1 year	39	65.0
1-5 years	11	18.3
5-10 years	4	6.7
> 10 years	6	10.0
<i>Previous exposure to knocked out tooth cases</i>		
Yes	31	50.8
No	29	47.5
<i>Have received advice / knowledge</i>		
Yes	27	44.3
No	33	54.1

<i>If yes, source of knowledge?</i>		
Book/Literature	18	29.5
Seminar/Workshop	7	11.5
Part of the medical syllabus	2	3.3
Course in first aid	0	0.0

It was observed that a 40% of the respondents were males and 60% of the respondents were female. A majority of the participants was from the house officer category and about 50.8 % of these

participants were seen to have previous exposure to a case with avulsed tooth. More than half of the subjects said that they did not receive any advice or knowledge regarding traumatic dental injuries.

Table 2 Knowledge, experience with dental avulsion, crown fractures and luxation injuries among medical practitioners in the Emergency Department of Hospital Serdang (n= 60)

	n	%
<i>Type of dentition (primary)</i>		
Primary	24	40.0
Mixed	20	33.3
Permanent	16	26.7
<i>Type of dentition (mixed)</i>		
Primary	25	41.7
Mixed	26	43.3
Permanent	9	15.0
<i>Duration of Extra Oral Time of avulsed tooth important?</i>		
Yes	45	73.8
No	15	24.6
<i>Urgency for referral to the dentist</i>		
Within 30 minutes	12	20.0
Within few hours	36	60.0
The next day	12	20.0
<i>Would you refer the child/instruct parents to go to the dentist?</i>		
Yes	60	100
No	0	0

<i>If yes, when will you advice the parents to go the dentist?</i>		
Immediately	44	73.3
Next day	10	16.7
After a few days	3	5.0
When pain arises	3	5.0
<i>Do you think the knocked out adult tooth can be put back / reimplanted?</i>		
Yes	30	50.0
No	30	50.0
<i>What would you do if a knocked-out tooth was covered with dirt and you have decided to replant it?</i>		
Rinse tooth with tap water	22	36.7
Rinse with H2O2	27	45.0
Clean with soap or disinfecting solution	11	18.3
<i>How would you hold a knocked-out tooth?</i>		
From the crown	24	40.0
From the root	9	15.0
Not sure	26	43.3
<i>If you did not replant the tooth, how would you advice patient to transport it to the dentist?</i>		
Milk	15	25.0
Ice cubes	25	41.7
Alcohol	5	8.3
Disinfecting solution	15	25.0
<i>Refraining from replacement of a completely knocked out teeth relates to</i>		
Primary	25	41.7
Permanent	25	41.7
Both primary and permanent	10	16.7
<i>Do you think a fractured crown of an adult/baby tooth is an emergency?</i>		
Yes	34	56.7
No	26	43.3

<i>Would you refer this case to the dentist immediately?</i>		
Yes	44	73.3
No	16	26.7
<i>Do you think carrying a segment of the crown to the dentist is beneficial</i>		
Yes	40	66.7
No	20	33.3
<i>Which terminology best describes the injury below?</i>		
Subluxation	44	73.3
Concussion	12	20.0
Lateral luxation	3	5.0
Intrusive luxation	1	1.7

It can be noted that only 40% identified the primary dentition correctly and 43.3 % of the participants identified the mixed dentition correctly. Majority of the participants from the survey did mention

that they knew that the extra oral time of the teeth was important however about 80% did not feel the urgency for a patient to visit the dentist within a time frame of 30 minutes. (Table 2)

Table 3 *Attitude of the medical practitioners towards receiving further knowledge concerning traumatic dental injuries (n=60)*

	n	%
<i>What do you think is your level of information about traumatic dental injuries?</i>		
Adequate	0	0.0
Inadequate	60	100.0
<i>In your opinion, learning about traumatic dental injuries is:</i>		
Very Important	8	13.3
Important	44	73.3
Somewhat important	8	13.3
Not important	0	0.0

<i>Would you like to receive more information to properly manage TDI?</i>		
Yes	60	100.0
No	0	0.0
<i>Are you interested in knowing the emergency management of knocked out teeth?</i>		
Yes	60	100.0
No	0	0.0

As a whole, the medical practitioners had positive attitude towards receiving further knowledge regarding traumatic dental injuries as they felt that their level of knowledge was inadequate. Thus, their

attitude towards receiving further education was also positive especially in the area of management of traumatic dental injuries. (Table 3)

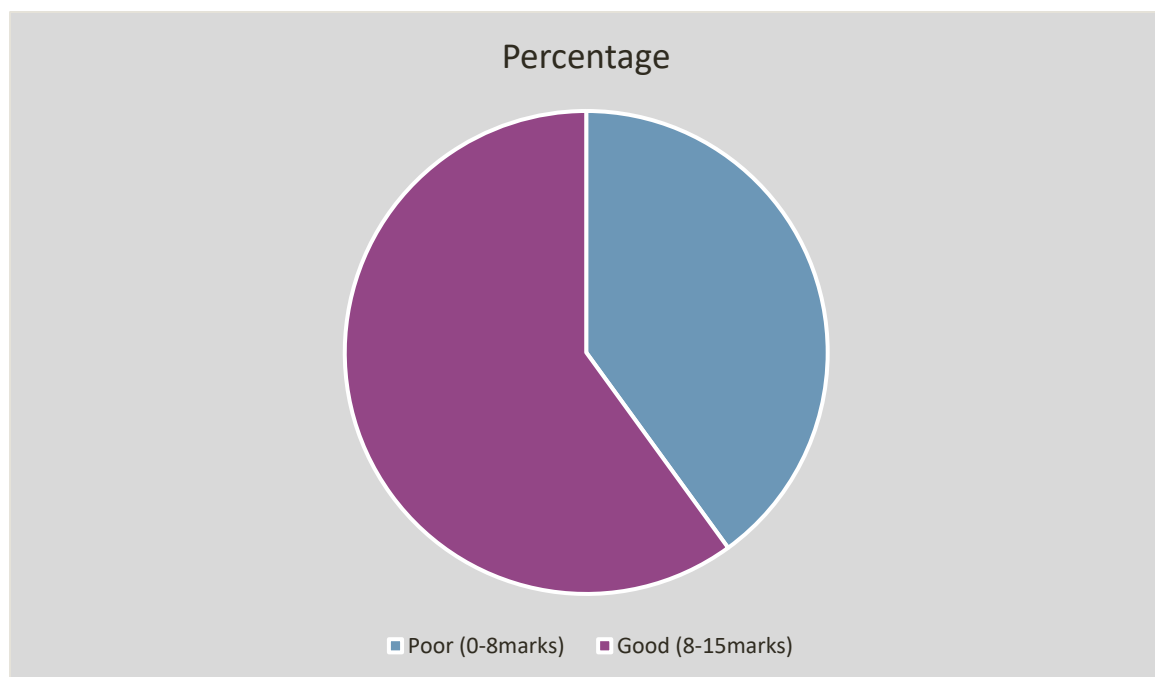


Figure 1 Level of knowledge among medical practitioners based on correct answers in section B (Knowledge score)

In this section of knowledge assessment, each questions will be given a score of one point, and wrong answer will be given a score of zero .The total score will be from zero (0) to fifteen (15) , zero being no knowledge and fifteen being full knowledgeable. A correct answer in this section was awarded with one point. Thus,

the level of knowledge was assessed through the determination of the total score, namely poor knowledge score being (0– 8.0), and a good knowledge score being (9.0 – 15.0). As a whole, it was seen that about 60% of the participants had poor knowledge score whereas 40% had good knowledge score. (Figure 1)

Table 4 *Group comparisons on knowledge level and designation categories using the Mann Whitney U – Test*

DESIGNATION		
	SPECIALIST	MEDICAL OFFICER / HOUSE OFFICER
STATISTICS		
MEDIAN SCORE	10.50	8.00
INTERQUARTILE		
RANGE	2.75	3.25

In order to compare the differences between two independent groups in which the dependent variable is not normally distributed, the Mann Whitney U-Test is used. In this case, the mean knowledge score of the group of specialists were made in comparison with a group of medical officers/house officers. (Table 4)

The statistics were seen to be statistically significant with $p=0.017$. Thus, from the

statistics it was observed that the knowledge score for the group of specialists were generally higher compared to that of the medical/house officers. It can be observed that the number of years of experience of these practitioner correlated better knowledge exposure on traumatic dental injuries.

DISCUSSION

This study provides a baseline information about the current level of knowledge of emergency department medical practitioners regarding tooth avulsion and traumatic dental injuries. Dental trauma can

present as an isolated injury or as multiple extended injuries.¹⁰ In either case they are attended by medical practitioners of the emergency department. As these medical practitioners are at the fore front of the

hospital, an immediate and prompt actions during dental referrals are necessary in giving good dental service. However, from this study, it was observed that even though some had a personal experience in coming across a child with traumatic dental injuries, only a few had the urge to actually seek the dental team for advice for future references.

Among these practitioners, 60% were unable to identify the type of dentition in children thus by understanding this basic foundation, further management of traumatic dental injuries such as early dental referrals can be done.

A positive finding is that 66.7 % believed that the dentist can do something with a broken fragment of tooth. With the advancement in restorative dentistry and bonding techniques, reattachment of the fractured crown section should always be considered today.

Another form of confusion centred on the significance of transport media in which an avulsed tooth can be place whereby the purpose of the transport media is to preserve the vitality of the periodontal ligament.¹¹ It was also observed that 41.7 % considered ice cubes as the best storage media. Only 26 % were aware that milk is the best storage medium.

In one study by Holan and Shmueli (2003), it was observed that 55% of the physicians had never received information regarding dental trauma.¹² Thus the highlight of our

study is that almost all participants accepted that their knowledge on the management of dental trauma is definitely inadequate. From this study, it was observed that these participants had interest to attend an additional educational program on dental trauma, and with a good attitude towards expanding their knowledge. With that, it reflected that these practitioners did not receive an opportunity to attend any dental health program.

To this end, it is seen that a simple procedure of reimplantation of a tooth could make a huge difference not only in the prognosis of the tooth but also influence the facial growth, functions, aesthetics, and psychological impact of the patient.¹³

An author also suggested that since there are guidelines in the form of flow charts which are seen to be available in the emergency departments such as that pertaining to myocardial infarctions, poisoning and animal bites, he believes that the guidelines produced by the International Association of Dental Traumatology (IADT) should also be placed in the emergency departments in the form of flow charts or posters.¹⁰ This will be very informative to the medical practitioners and it will help them provide appropriate primary care to the child with dental trauma, indirectly avoiding late referrals to the dental team.

CONCLUSION

In conclusion, though there is lack of proper knowledge among these medical practitioners in regard to management of traumatic dental injuries, there is certainly motivation and willingness among these

practitioners to gain further knowledge. Thus, by providing better dental programs and spreading awareness in the emergency department, this will indirectly help provide better service to the public.

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This research presents no conflict of interest.

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Post-operative Pain Control in Children After Dental Extractions Under General Anesthesia (GA): A Service Evaluation Utilizing the Modified Post Anesthetic Discharge Scoring System (PADSS) Form.

Adibah Jamil¹, Thaarani Vijayakumar¹, Nur Najmina Norazman Shah¹

ABSTRACT

Introduction: Pain in children's post-extractions under GA has raised concerns in parents as well as clinicians. The Post-Anaesthetic Discharge Scoring System (PADSS) provide objective evaluation of pain amongst its other criteria, and it is utilized in hospitals across Malaysia prior to discharging patients from daycare services.

Objective: To report on post-operative pain in children's post-extractions under GA using the PADSS form.

Method: A retrospective service evaluation utilizing patient's case notes as well as PADSS forms for cases done in 2020.

Results: A total of 35 children were included in this study. The average age of a child was 5.2 years old with a mean of 11 extractions. All children reported a pain score of 0 on discharge with 22.8% reporting pain score of 1 24 hours post-procedure. Perioperatively, intravenous (IV) Fentanyl was used in all children in combination with IV Acetaminophen (PCM) (82.8%) or PCM Suppository (17.1%). IV Morphine was given in addition to IV PCM in 20% of cases. All children had local anaesthesia administered peri-operatively and prescribed an oral analgesic (PCM) upon discharge. The children that reported a pain score of 1 post-procedure had an average of 11 teeth extracted. Conclusion: None of the children reported pain prior to discharge. Pain score of 1 was reported 24 hours later in children who had 10 or more teeth extracted. Non-compliance to oral analgesics was reported in children with pain. Following this study, the areas requiring improvement have been identified and measures have been established to improve quality of care.

Keywords: Child, Analgesia, General Anaesthesia, Dental, Extractions, Pain Control, Acetaminophen

(1) Department of Paediatric Dentistry, Slim River Hospital, Perak, Malaysia

Corresponding Author

Adibah binti Jamil

Address

Jabatan Pergigian Pediatrik
Hospital Slim River
35800 Perak.

Telephone Number

05-450 8000 (2057)

Email

dr.adibah@moh.gov.my

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INTRODUCTION

Dental caries remains as the highest primary diagnosis for hospitalization of children aged 5-9 years old. In the year 2018-2019, the number of Finished Consultant Episodes for dental caries in the United Kingdom stands at 25,702. This figure supersedes the next highest diagnosis for admitted patient care for the same age group by two-fold which is acute tonsillitis admission figuring at 11,811 episodes¹. This high figure for hospital admission is mainly contributed by the need for dentistry under general anaesthesia as very young children are usually either uncooperative or unable to tolerate multiple teeth extraction or complex dental procedure for very broken-down teeth or infection.

On the other hand, beds shortage in the hospital and long waiting times for elective operation has pushed the day-care services to become an immensely popular treatment of choice for both the surgeons and the parents². The concept of Day care Services entails patients coming into the hospital to undergo diagnostic or therapeutic procedure and be able to go home on the same day. In Malaysia, the Ministry of Health has built 9 dedicated "Ambulatory Care Centres" (ACC) consisting of multidisciplinary units and over 130 public hospitals providing Day care services for this purpose³.

With increasing number of Day care Surgical Services being done, the Modified Post-Anaesthetic Discharge Scoring System (PADSS) was developed by the Malaysia Medical Development Division in 2012 to provide 6 key objectives evaluation before discharging a patient home. The form evaluates pain, nausea and vomiting, vital

signs, activity level, and surgical bleeding. To be discharged from the Day-care unit, the patient has to have no or minimal pain as well as demonstrate that the pain that is controllable by local analgesics. The same form is also used as a post-operative assessment tool at home. Twenty-four hours post-discharge; a member of the day-care team would contact the patient and conduct a survey based on the Modified PADSS form to assess the post-operative pain.

Despite recent advances in the development of both paediatric pharmacological and non-pharmacological measures, children undergoing extractions under GA have reported significant physiologic morbidity reporting pain and discomfort post-procedure⁴. As high as 74% of children aged 6 to 16 years reported postoperative pain following dental treatment under GA⁵. Routine management of post-operative pain after dental extractions include Paracetamol and/or non-steroidal anti-inflammatories (NSAIDs) such as Ibuprofen⁶. However, medication adherence in children remains an on-going challenge due to difficulty in recognizing pain and underrating of pain in children by parents and healthcare professionals⁷⁻¹⁰. In a study by Atan et. al in 2004, 74% of children aged 6 to 16 years reported postoperative pain following dental treatment under GA⁵.

Against this background, this research aims to appraise the efficacy of current postoperative pain control practice of children undergoing dental extractions under general anaesthesia in day care service utilising a readily available Modified PADSS form.

The primary purpose of this study is to report on the prevalence of pain for children that has underwent tooth extractions under GA.

Secondary objectives include to report the number of teeth extracted and intra-operative analgesic administered; medication adherence post dental extractions and to study the use of self-reported pain scale

METHODS

This is a retrospective study using secondary data (Modified PADSS) utilizing a universal sampling method. We included all children under the age of 16 years old that underwent dental extractions under GA in Slim River Hospital between 1st Jan 2020 to 31st Dec 2020 (Figure 1).

All children under the age of 16 years, classified as American Society of Anaesthesiologists (ASA) physical status 1-2, that underwent a minimum of 1 primary/permanent tooth extracted under Day Care GA Services in Slim River Hospital between 1st Jan 2020 to 31st Dec 2020.

Our exclusion criteria included ASA III and higher patients, patients having procedures other than dental extractions under GA (e.g., surgical removal of impacted tooth) and cases that were done as in-patients.

This study approved by the Medical Research & Ethics Committee (KKM/NIHSEC/ P20-2586 (4)) and is registered with the NMRR (National Medical Research Register) with NMRR ID: NMRR-20-2811-57613.

The Anaesthesia Protocol conducted in this study were done as per individual Anaesthetist's preference and were not pre-emptively planned with the surgeon. 6-8% sevoflurane and oxygen FiO₂ 100% was administered under mask for anaesthetic induction. Oro-tracheal intubation was then performed under intra-venous (IV) administration of Fentanyl (0.5-2 mcg/kg). All sample received acetaminophen (15mg/kg) differing either by IV or suppository administration. Selective pain control intra-operatively were done by the anaesthesiologists on duty where 42.8% of the sample received an additional (IV) administration of Propofol (2 mg/kg) and only 20% of the sample received an addition of IV Morphine.

All dental extractions in this study were carried out by the same resident Paediatric Dentist Specialist in less than 30 minutes of operation time. All samples received local anaesthetic 2% Mepivacaine Hydrochloride Adrenaline 1:100.000 appropriated by weight prior to dental extraction.

Post-operatively, a joint assessment using the Modified PADSS form would be done by the surgeon and the anaesthesiologist teams. Only patients scoring more than 9 on the Modified PADSS form would be allowed to be discharged home with take-home analgesics, standardised to Syrup Acetaminophen 15mg/kg QID/PRN. Twenty-four hours post-discharge, the patients will be contacted by the Day care team to assess compliance to medication as well as presence of pain as per the modified PADSS form (Figure 2)

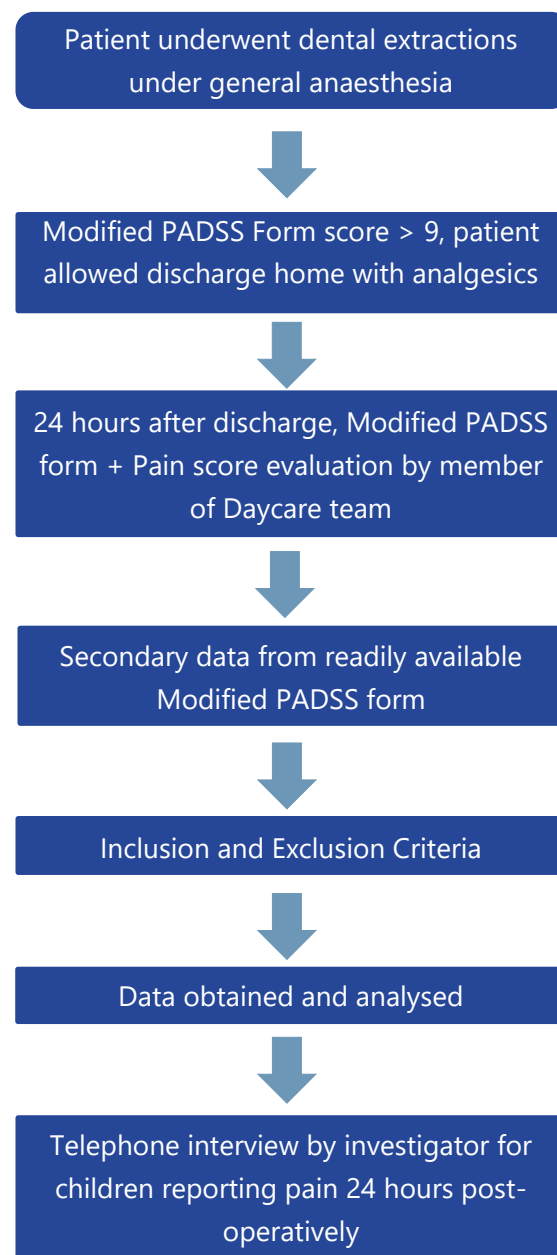


Figure 1 Flow of study

Appendix 4

DISCHARGE FORM

Modified Post Anaesthetic Discharge Scoring System (PADSS)
(Marshall and Chung).
(To accompany the Anaesthetic Record Form for each patient)

	Score
Vital signs	<i>Vital signs must be stable and consistent with age and preoperative baseline.</i>
	BP and pulse within 20% of preoperative baseline
	BP and pulse within 20-40% of preoperative baseline
	BP and pulse >40% from preoperative baseline
Activity Level	<i>Patient must be able to ambulate at preoperative level.</i>
	Steady gait, no dizziness (or meets preoperative level)
	Requires assistance
	Unable to ambulate
Nausea & Vomiting	<i>The patient should have minimal nausea and vomiting prior to discharge</i>
	Minimal: successfully treated with oral medication
	Moderate: successfully treated with IM/IV medication
	Severe: continues after repeated treatment
Pain	<i>The patient should have minimal or no pain prior to discharge.</i>
	<i>The level of pain that the patient has should be acceptable to the patient.</i>
	<i>Pain should be controllable by oral analgesics.</i>
	<i>The location, type and intensity of pain should be consistent with the anticipated postoperative discomfort.</i>
	Acceptability: Yes
	No
Surgical Bleeding	<i>Postoperative bleeding should be consistent with expected blood loss for the procedure</i>
	Minimal: does not require dressing change
	Moderate: up to two dressing changes required
	Severe: more than three dressing changes required
TOTAL: _____	
SN name: _____ Signature: _____	

Figure 2 Scoring system prior to discharge

Appendix 6

POST OPERATIVE FOLLOW UP FORM

(To be filled by a nurse or doctor on the day of surgery, before the patient is discharged)

Anaesthetist: _____

Surgeon: _____

Operation Date: _____

Procedure: _____

Patient contact phone number: _____

Section 1 (Before discharge on day of surgery)

Any post-operative problems? e.g. nausea / pain

Discharge analgesia:

Paracetamol ☐ Tramadol ☐NSAIDs ☐ Other ☐ _____

Section 2 (24 hours after discharge)

(Nurse/Doctor who calls the patient at home to ask the following questions)

1. Have you had any problems since you were discharged? Yes
- ☐
- No
- ☐

If yes what was the problem?

Pain ☐ Nausea/vomiting ☐

Others (specify)

2. Have you sought outside assistance since discharge? Yes
- ☐
- No
- ☐

If yes who did you see/contact and why?

GP ☐ ED ☐ Given contact no. ☐ Other ☐ _____

What advice was given?

Figure 3 Checklist prior to discharge and 24 hours post-surgery

RESULTS

Fifty-six patients had dental treatment under GA in the Day care Unit of Slim River Hospital from 1st Jan 2020 to 31st Dec 2020 and from this number, 35 patients fit the inclusion criteria. The ages of the children ranged from 2.5 years old to 9.8 years old with a mean of 5.2 years old. Each patient underwent tooth extraction ranging from 1 to 20 teeth with a mean of 11 teeth.

Prior to discharge, all children reported a pain score of 0. At 24 hours later, 22.8% of patient reported a pain score of 1 while the remaining 77.2% reported a pain score of 0 (Table 1). Of those reporting pain score of 1 at 24 hours post-operatively, 2.5% had 0-5 teeth extracted, 25% had 6-9 teeth extracted and 62.5% had more than 10 teeth extracted (Table 2).

Table 1 Baseline Pain Score Upon Discharge Vs 24 Hours Post-Discharge

Pain Score	Pain Score Post-op = 0	Pain Score Post-op = 1
Upon Discharge	100%	-
24 hours after discharge	77.2%	22.8%

Table 2 Pain Score Upon Discharge Vs 24 Hours Post-Operatively Comparing No Of Tooth Extracted

No. of Tooth Extracted	Pain Score at discharge = 1	Pain Score 24 Post-op = 1
0-5	29.6%	12.5%
6-9	14.8%	25%
More than 10	55.5%	62.5%

From the group that had received an addition of IV Morphine peri-operatively, 85.7% reported no pain and 14.3% reported a pain score of 1 at 24 hours post-surgery. A telephone-based interview by the investigator was conducted with parents of

children reporting a pain score of 1 at 24 hours post-surgery to assess the compliance to analgesics administered at home. A total of 87.5% of parents reported of not serving the prescribed analgesic (Table 3).

Table 3 Pain Score Upon Discharge Vs 24 Hours Post-Operatively Comparing Intra-Operative Analgesia

Intra-Op Analgesic	Pain score at discharge = 1	Pain score 24 post-op = 1
IV Morphine	85.7%	14.3%
IV Fentanyl	100%	100 %
IV/Suppository Acetaminophen	100%	100%
2% Mepivacaine Hydrochloride/ Adrenaline 1:100.000	100%	100%

DISCUSSION

In this study the children ages ranged from 2.5 years old to 9.8 years old with a mean of 5.2 years old. This finding is similar to other studies done on the demographic data of children receiving dental treatment under GA as children in this age group tend to not cope very well with extensive dental treatment (e.g., multiple extractions) without general anaesthesia^{11,12}.

All children in this study reported no pain prior to discharge but 22.8% reported pain score of 1 twenty-four hours later. The absence of pain upon discharge (4-6 hours post operatively) could be explained by the judicious decision of the Slim River Hospital anaesthetists to administer both IV Fentanyl (short-acting opioid) and IV Paracetamol

intra-operatively to all the children as well as the utilization of a local anaesthetic peri-operatively. A recent study by Alohal et al (2019) demonstrated better pain control for children having tooth extractions under general anaesthesia when IV Fentanyl and IV Paracetamol were administered intra-operatively¹³. These patients also demonstrated lower pain score and subsequently lower need for analgesics post-operatively in ward and at home¹⁴.

The addition of LA during dental treatment under GA possesses the benefits of decreased postoperative pain as well as improved haemorrhage control. In regard to postoperative pain, Atan et al (2004), in their study also found a significant decrease in pain at the operation site in patients who received supplemental local anaesthesia⁵.

Sixty-two percent of children with more than 10 teeth extracted reported a pain score of 1 twenty-four hours post-operatively and from this category, 87% of them were not compliant to the prescribed analgesic. In study done on 67 children aged 4 to 7 years old having acute pain, it was concluded that parents rated the pain considerably lower than the children themselves¹⁰. Another study by Rony et al (2010) concluded that fear of adverse effects from oral analgesics cause reluctance in parents to medicate their children¹⁵. These factors are in line with the present study where parents reported incompliance in administering prescribed oral analgesics for their children.

As analgesics are more effective in preventing pain than in the relief of established pain; it is important that they are given regularly in the early postoperative period and then 'as required' according to assessed pain¹⁶. An important facet of this study is the difficulty to adequately assess pain in children. This is because children only develop the ability to adequately assess pain around the age of five¹⁷. Infants and children also easily mistake new environments and absence of parents and/or security objects as pain¹⁸.

Children experience pain differently than that of adult and their inability to evidently express their pain level and their need for analgesics may result in zero pain behaviour especially in the mild pain score of 0-1¹⁸. As such, parents/caregiver may presume that pain is not present and thus put off

administering prescribed analgesics as has been demonstrated in this study. This highlights a need for a take-home pain assessment tool for parents/caretaker that is easy to use, objective, and is validated against children's self-report of pain and age.

The main limitation of this study was the limited beds in day care unit, thus lowering the sample size attainable. The varying assessors: trained nurses fill the form upon discharge and the parents fill it up 24 hours later could also lead to discrepancies. The Modified PADSS form assess the patient's pain score but not analgesics compliance at home. Children experiencing pain score of 1 at 24 hours post operatively could be attributed as a true experience of pain or could be attributed to the failure of parents/caretaker to serve the medications.

CONCLUSION

According to the results of this study, children do experience pain after dental extractions under general anaesthesia. Patient with more than 10 teeth extracted receiving none of the analgesics prescribed are more likely to report a pain score of 1. A study utilizing a child-self reporting measure such as a visual

analogue scale (VAS) would enable the investigator to obtain a more accurate picture of pain as quantifying pain especially in children. This remains the biggest challenge in conducting any high-quality pain related research in children.

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Prevalence of Unerupted Permanent Maxillary Central Incisors Among Primary School Children in Perak, Malaysia

Suhailiza Saharudin¹, Sumathy Perumal¹, Rashima Ibrahim², Chan Yen Pei¹, Wan Asmat Mahussain², Magesh Vijayakumar³

ABSTRACT

Introduction: The eruption of human dentition occurs in a chronological sequence. Delayed eruption of permanent teeth especially maxillary central incisors may cause cosmetic and pathological complications.

Objective: To determine the prevalence and management of unerupted permanent maxillary central incisors in the developing dentition among primary school children.

Methods: This was a cross sectional study conducted in three districts of Perak by examining the dental records of standard 3 primary school children who aged 8 - 9 years old in 2015. Multistage sampling method was performed to select the schools and the dental records of study subjects. The records were screened to obtain the demographic data, the presence of maxillary central incisors and the management performed which included the referral records.

Results: A total of 2,267 dental records were screened and it revealed 48 children (2.1%) had unerupted permanent maxillary central incisors by the age of 9. Males were commonly affected than females with a ratio of 1.2: 1 and mainly occurred in Malay population. Equally distribution of children (50%) were missing either one or both permanent maxillary central incisor with no propensity of occurrence between right or left central incisors. All 48 children (100%) with unerupted permanent maxillary central incisors were not referred for further management.

Conclusion: The prevalence of unerupted permanent maxillary central incisors was 2.1%. There was a high prevalence of delay in diagnosing and referring cases of unerupted permanent maxillary central incisors by the primary dental care providers. Awareness in diagnosing and management of delayed tooth eruption is necessary to ensure the proper referral is made for early intervention to avoid complicated treatment in the future.

Keywords: *Unerupted tooth · Delayed eruption · Permanent maxillary central incisors*

- (1) Department of Paediatric Dentistry, Hospital Raja Permaisuri Bainun, Ipoh, Perak.
- (2) Department of Paediatric Dentistry, Hospital Taiping, Perak.
- (3) Department of Paediatric Dentistry, Hospital Teluk Intan, Perak.

Corresponding Author

Suhailiza Saharudin

Address

Department of Paediatric Dentistry,
Hospital Raja Permaisuri Bainun,
30450 Ipoh, Perak.

Telephone Number

05-2087482

05-2087481 (Fax)

Email

dr.suhailiza@gmail.com

DOI

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INTRODUCTION

The maxillary incisors and canines are the most prominent teeth in an individual's smile and are referred to as the "social six".¹ Missing or delayed eruption of those teeth, particularly the maxillary central incisors may cause major impact on phonetics and dentofacial aesthetics, which later compromised individual's self-esteem and social interaction.¹⁻³

The permanent maxillary central incisors erupt approximately between the ages of 7 to 8 years.⁴ Early study by Davis (1987) stated maxillary central incisors are the third most common impacted teeth, following the third molars and maxillary canines.⁵ Those permanent maxillary central incisors are vulnerable to eruption failure secondary to space loss, obstruction or trauma.^{2,3}

The permanent maxillary central incisors are considered delayed in eruption when:⁶

- a) *there is no history of previous extraction,*
- b) *eruption of contralateral incisor has occurred 6 months earlier,^{7,8} or if both central incisors are unerupted and the lower incisors have erupted more than one year previously,² or*
- c) *there is a deviation from the normal sequence of eruption (e.g., lateral incisors erupt before the central incisors).^{2,8}*

Failure of the maxillary incisors eruption usually presents in the mixed dentition stage and is often noticed between the ages of 7 – 9 years.^{2,3} Eruption disturbances of the maxillary incisors can be a harbinger of a local or systemic condition,¹ but almost half (47%) of all unerupted maxillary incisors are due to

supernumerary teeth.⁹ Thorough and careful examination including history, clinical and radiographic investigations should be carried out to diagnose delayed eruption of permanent maxillary central incisors.¹ This study was aimed to determine the prevalence of unerupted permanent maxillary central incisors among primary school children in Perak, Malaysia.

MATERIALS AND METHODS

The study was approved by the Medical Research and Ethics Committee of Ministry of Health Malaysia [KKM/NIHSEC/P20-600(6) and KKM/NIHSEC/P20-600] with registered ID NMRR-19-3990-52332 (IIR). The study was conducted in compliance with the ethical principle outlined in the Declaration of Helsinki and Malaysian Good Clinical Practice Guideline.

This was a cross sectional study conducted in three districts of Perak, Malaysia. This study was performed by examining the stored dental records of standard 3 primary school children who aged 8 - 9 years old in year 2015 of the Kinta District Dental Health Office, Larut, Matang, Selama (LMS) Dental Health Office and Hilir Perak Dental Health Office. There was a total of 17,367 primary school children and by using the Sample Size Calculator for Estimations software, 1,526 were determined to be the sample size. In view of study limitation, 5% dropout rate was calculated to achieve a total minimum sample size of 1,609.

This study employed a multistage convenient sampling method. The first stage was a systematic selection of every 4th school from a list of 325 primary schools in the districts that were visited by the Mobile School Dental

Teams. 136 schools were selected using this method. A total of 10,160 dental records were collected from the total number of schools selected. Then, every 5th dental record was systematically selected to make up the final total sample size of 2,498 dental records which complied the minimum total sample size of 1,609.

The exclusion criteria included incomplete dental records (demographic data/ dental charting etc.) and history of extraction of permanent maxillary central incisors. After implying the exclusion criteria, a total of 231 dental records were excluded. Thus, the final total sample size was 2,267 dental records.

Dental records were screened to obtain the socio-demographic data and the presence of maxillary central incisors in the dental charting. Then, the dental records with missing or unerupted permanent maxillary central incisors will be examined further for

any referral for treatment. Delayed diagnosis will be determined if no referral has been made. Data were collected and processed using Microsoft® Excel for Mac (Version 16.16.7).

RESULTS

This study comprised a total of 2,267 primary school children with a mean age of 8.99 ± 0.52 years. There was an equal distribution of 50.8% male and 49.2% female subjects respectively. Majority of the children were Malays (57.1%), followed by Chinese (28.2%), Indians (13.9%) and other races (0.8%).

There were 48 children (2.1%) presented with a total of 72 unerupted permanent maxillary central incisors at the age of 9 years. The involvement of maxillary right central incisors was relatively higher compared to the maxillary left central incisors. (Table 1)

Table 1 Distribution of unerupted permanent maxillary central incisors (N = 72)

TOOTH	TOTAL	PERCENTAGE %
Maxillary right central incisor (FDI* notation: 11)	37	51.4
Maxillary left central incisor (FDI* notation: 21)	35	48.6

*FDI (World Dental Federation)

Further distribution of 48 children with unerupted permanent maxillary central incisors was displayed in Table 2. There were 26 boys (54.2%) and 22 girls (45.8%) had unerupted permanent maxillary incisors with the gender ratio of 1.2: 1. The predilection for occurrence is mainly in Malays [36 (75.0%)], followed by Chinese [9 (18.8%)], Indians [2 (4.2%)] and others [1 (2.1%)]. Study revealed

that 50% of children had at least one unerupted permanent maxillary central incisors, and other 50% of children were missing both of permanent maxillary central incisors. Study also showed that all the 48 children (100%) with delayed eruption of permanent maxillary central incisors were not referred to the dental clinic for further management.

Table 2 Distribution of children with unerupted permanent maxillary central incisors

Variable	Unerupted 11	Unerupted 21	Unerupted 11 & 21	Total (%)
Gender				
Male	6	7	13	26 (54.2)
Female	7	4	11	22 (45.8)
Race				
Malay	9	7	20	36 (75.0)
Chinese	3	3	3	9 (18.8)
Indian	1	-	1	2 (4.2)
Others	-	1	-	1 (2.1)
Referral				
Yes	-	-	-	-
No	13	11	24	48 (100)

*Teeth denoted by the FDI two-digit system

DISCUSSION

Human dentition consists of 20 teeth in the primary dentition and 32 teeth in the permanent dentition. Teeth begin to develop around the sixth week in utero and will reach completion during early adult life.¹⁰ Then it will move axially from its non-functional position in the bone to functional occlusion

to be in contact with its antagonist.¹¹ This tooth eruption is an orderly sequential and age specific event, but it may vary individually.^{12,13} Table 3 shows the tooth eruption and exfoliation times which are usually presented as ranges of chronological age.⁴

Table 3 Tooth eruption and exfoliation

	PRIMARY DENTITION			
	Mandible		Maxilla	
	Eruption (months)	Exfoliation (years)	Eruption (months)	Exfoliation (years)
Central incisors	5 - 8	6 - 7	6 - 10	7 - 8
Lateral incisors	7 - 10	7 - 8	8 - 12	8 - 9
Canines	16 - 20	9 - 11	16 - 20	11 - 12
First molars	11 - 18	10 - 12	11 - 18	9 - 11
Second molars	20 - 30	11 - 13	20 - 30	9 - 12

PERMANENT DENTITION				
	Mandible		Maxilla	
	Eruption (years)	Exfoliation (years)	Eruption (years)	Exfoliation (years)
Central incisors	6 - 7	9 - 10	7 - 8	9 - 10
Lateral incisors	7 - 8	10	8 - 9	11
Canines	9 - 11	12 - 15	11 - 12	12 - 15
First premolars	10 - 12	12 - 13	10 - 11	12 - 13
Second premolars	11 - 13	12 - 14	10 - 12	12 - 14
First molars	5.5 - 7	9 - 10	5.5 - 7	9 - 10
Second molars	12 to 14	14 - 16	12 - 14	14 - 16
Third molars	17 - 30	18+	17 - 30	18+

Adapted with permission from Logan WHG, Kronfeld R. Development of the human jaws and surrounding structures from birth to the age of fifteen years [4].

Delayed tooth eruption is commonly diagnosed by using two fundamental parameters as below.¹¹ First parameter is an "expected tooth eruption" which is based on average chronological age tooth eruption that derived from population studies. Nichifor et al (2011) suggested the normal variations of the eruption age are within the limit of one year (6 months sooner or later) in relation to the average age of eruption.¹⁴ The significant deviations from accepted norms of eruption time is considered as delayed tooth eruption.¹³

Second parameter is a "biologic tooth eruption" which is indicated by progression of root development. Becker (1998) suggested that root development alone should be the basis for defining the expected time of eruption for different teeth.¹⁵ Thus, if the tooth has developed more than the root length expected for eruption and remains unerupted, it should be defined as having delayed eruption.¹¹

This study used the expected chronological age tooth eruption whereby the permanent maxillary central incisor should be erupted at age of 7 to 8 years old.⁴ The permanent maxillary central incisors are considered delayed when they are not erupted within the limit of one year.^{2,3} The mean age of study subjects was 8.99 ± 0.52 years and theoretically those children should have both permanent maxillary central incisors at this age. This study identified 48 children (2.1%) who had unerupted permanent maxillary central incisors by the age of 9. This finding is close to the figure stated by Tan et al. (2018) and Di Biase (1969) which were 2.0% and 2.6% respectively.^{16,17} However, this finding is higher than those reported in several western countries before, which were 0.03% - 1.0%.¹⁸⁻

²⁰ Other research done in Nigeria showed 1.7% children from total of 2,240 patients presented with unerupted maxillary incisors.²¹

Studies on teeth emergence generally shows that permanent teeth erupt earlier in females than in males.²² Earlier eruption of permanent teeth in females is attributed to earlier onset of maturation.²³ Particularly in the permanent maxillary incisors, Sonu et al. (2002) observed females were more frequently affected than males.²¹ The increase ratio of the female to male patients is well recognised by most orthodontist and may reflects a difference in perception of malocclusion between the sexes. It may be since females rate dental appearance to be more important and tend to seek for aesthetics improvement more than the males.²¹ In the other hands, Bartolo et al. (2010) and Lygidakis et al. (2015) stated that incidence of unerupted permanent maxillary central incisors was more common in males than females with a minimum ratio of 2.7: 1.^{24,25} In agreement with the current study, it showed that males were more commonly affected than females with a small ratio of 1.2: 1. The contributing factors for unerupted incisors among male patients could be explained by a greater prevalence of supernumerary teeth development, or tooth dilacerations and ectopic position of tooth bud following trauma events in males. There is also possibility involvement of sexual chromosomes in the etiology of tooth eruption disturbances.²⁶

The majority occurrence mainly in Malays as Malays constitute the majority population of the study sample. Racial, ethnic, sexual, and individual factors can influence eruption and are usually considered in determining the standards of normal eruption.²⁷⁻²⁹

This study revealed that 50% of children had at least one unerupted permanent maxillary central incisors, meanwhile the rest of 50% of children found to be missing both of permanent maxillary central incisors. Contradicted with our findings, Betts et al. (1999) and Sonu et al. (2003) found the majority of patients (70 - 82%) had only one unerupted maxillary central incisor.^{9,21} Bilateral involvement of permanent maxillary central incisors were noted to be very rare.^{9,21} From the total of 72 permanent maxillary central incisors involved, there was no propensity of occurrence between right or left central incisors and this finding was similar with a study by Betts and colleagues previously.⁹ However, Sonu et al. (2003) found that right-sided delayed eruption was more common in both central and lateral incisors but no conclusive reasons behind it.²¹

Those delayed eruptions of permanent maxillary incisors can be influenced by a number of factors including local problems, systemic conditions and genetic factors.¹³ Physical obstruction is a commonly identified local cause of delayed eruption of maxillary central incisors. These obstructions can result from many different causes, such as supernumerary teeth, mucosal barrier, scar tissue, and tumors.¹¹ Endocrine disorders such as hypothyroidism, hypopituitarism, hypoparathyroidism, and pseudo-hypoparathyroidism are the most common systemic conditions associated with delayed tooth eruption.³⁰ Occasionally, some syndromes or genetic disorders which associated with multiple supernumerary teeth (e.g., cleidocranial dysplasia), formation of tumors or cysts in the jaws (e.g., Gorlin syndrome) or disturbances of periodontal tissues development (e.g., Gardner

syndrome) might lead to generalized delayed tooth eruption too.^{11,13}

Delayed eruption of permanent maxillary incisors is common in childhood and adolescence, yet it is often overlooked or dismissed in paediatric primary care.²⁷ It is crucial for the clinician to determine the etiology and formulate an appropriate treatment plan.¹ Literature suggests clinicians should recognize the teeth that fail to emerge within 12 months of the normal range are considered delayed eruptions. In these cases, referral to a dentist is warranted for further clinical and radiographic assessment.^{1,27,31} However, in this study, all 48 children (100%) with unerupted permanent maxillary central incisors were failed to be diagnosed, and therefore not been referred for further treatment. There was no documentation of the need for monitoring eruption in the dental records too. Early diagnosis is very important and can directly affect overall treatment planning and timing of interceptive orthodontic or surgical treatment. Interceptive treatment at the correct timing may improve skeletal malrelationship, eliminate functional interferences, and correct disturbances during the eruption.^{11,31}

Clinical Practice Guideline for Management of Unerupted Maxillary Incisor (2015) by Ministry of Health Malaysia stated that a well-planned multidisciplinary approach will ensure a successful treatment outcome. Recommendations of management of unerupted maxillary incisors are as follows,⁶

- *Early removal of obstructions and creation of sufficient arch space would facilitate spontaneous eruption of unerupted permanent maxillary incisors.*
- *The unerupted permanent maxillary incisors should be observed for about 12-18 months after interceptive treatment for spontaneous eruption.*
- *Failure of eruption of permanent maxillary incisors with no obvious obstruction, then surgical intervention needs to be considered.*

Currently in our local settings, treatment planning of such cases is discussed in a multidisciplinary combine clinic. The decisions are made between dental specialists in the field to achieve a favorable outcome. The potential risks of treatment, such as failure of eruption, ankylosis, external root resorption and poor gingival aesthetics, may still occur despite efforts made by the multidisciplinary team

CONCLUSION

The prevalence of unerupted permanent maxillary central incisors in developing dentition among primary school children was 2.1%. There was a high prevalence (100%) of delay in diagnosing and referring cases of unerupted permanent maxillary central incisors by the primary dental care providers which needs to be taken seriously. It is essential that the clinicians have a thorough knowledge of these eruption disturbances in order to make an appropriate, as well as timely intervention for adequate treatment. Therefore, awareness in diagnosing and management of delayed tooth eruption is necessary. Improvement measures such as continuous dental education and inter-operator calibration practice should be scheduled regularly. This is to ensure the proper referral is made for early intervention to avoid complicated treatment in the future.

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CONFLICT OF INTEREST

No conflicts of interest were reported by the authors.

ETHICAL APPROVAL

The study was approved by the Medical Research and Ethics Committee of Ministry of Health Malaysia [KKM/NIHSEC/P20-600(6) and KKM/NIHSEC/P20-600] with registered ID NMRR-19-3990-52332 (IIR). The study was conducted in compliance with the ethical principle outlined in the Declaration of Helsinki and Malaysian Good Clinical Practice Guideline.

INFORMED CONSENT

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Knowledge and Perception of Dental Trauma Among Students at A Faculty of Education

Mohammed Gh Al-Naser¹, Annapurny Venkiteswaran², Budi Aslinie Md Sabri³, Rohaida Abdul Halim², Ahmad Fahim Zulkifli⁴

ABSTRACT

Introduction: Knowledge of emergency dental trauma management is regarded essential for teachers as schools are a common place for such injuries to occur. Unfortunately, the level of dental knowledge remains poor among teachers worldwide. . The purpose of this study is to assess dental trauma knowledge of university students training to become teachers.

Methodology: A validated questionnaire was used in this study which contained demographic details and two hypothetical scenarios to assess dental trauma knowledge. A total of 83 subjects responded to the online questionnaire.

Results: Majority of the respondents were female (74,7%) and the mean age was 23.4 years. With regards to prior training or education on dental trauma, only 1.2% of the subjects had experience with first-aid training, Only 15.7% were able to identify the correct tooth in first scenario. Most of the subjects recognized that tap water was the best choice of rinsing solution but the response for suitable medium of transport for an avulsed tooth was wrong in more than 85% of the subjects. Although the level of knowledge was rather low, it was good to note that more than 90% of the subjects were keen to have more exposure and knowledge in dental trauma.

Conclusion: The knowledge on dental trauma among students from a faculty of education was poor. Intervention is recommended to improve the level of knowledge.

Keyword: *Dental trauma knowledge, Traumatic dental injuries, students, avulsion, perception*

- (1) Primary Care Clinic, Faculty of Dentistry, UiTM
- (2) Centre of Paediatric Dentistry & Orthodontics Studies, Faculty of Dentistry UiTM
- (3) Centre of Population Oral Health & Clinical Prevention Studies, Faculty of Dentistry UiTM
- (4) Physical & Health Education Department, Faculty of Education UiTM

Corresponding Author
Dr.Mohammed Gh Al-Naser

Address
Faculty of Dentistry UiTM
Sungai Buloh, Selangor
Malaysia

Telephone Number
-

Email
dr_moh_87@yahoo.com

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INTRODUCTION

Children spend most of their day time in school and daycare institutions, hence it is

not surprising that schools are a common place for dental injuries to occur^{1,2}. Inadequate treatment provided or failure of providing treatment in time may cause aesthetic defects and ultimately loss of the

tooth itself, particularly in children who still have immature permanent teeth³. The traumatic dental injuries and its subsequent sequelae are expected to exceed the load of dental caries and periodontal disease in younger patients^{4,5} not only physically and economically but also with unquantifiable psychological effect on the victim. Traumatic dental injuries may lead to serious aesthetic, social and psychological impairments as well as the imposed cost of traumatic dental injuries to the victims and their families⁶.

In most instances, the first person who sees a child with dental trauma is the schoolteacher. However, poor knowledge and lack of first aid training render the teachers inadequate to handle such injuries and this has an impact on the prognosis of the tooth⁷⁻¹⁰.

Multiple studies were done to assess schoolteachers knowledge of traumatic dental injuries and dental trauma first aid care and found that the schoolteachers knowledge was limited¹¹⁻¹⁴. The common trend seen was absence of dental first-aid training and lack of information on dental trauma¹¹⁻²⁰. Although prior experience of witnessing a dental trauma was thought to correlate with better knowledge¹¹, findings from large scale studies contradict this notion^{14,21}.

In Malaysia, future school teachers are either trained at Teacher's Training Colleges or Faculty of Education in a university. The Dental Health Division, Ministry of Health (MOH) is tasked with the provision of essential dental knowledge for future teachers attending teacher's training colleges

with the help of a guideline which outlines the method of delivery and content²². Currently there are no studies which have evaluated the effectiveness of the dental knowledge module carried out by MOH. The programme by MOH does not include future teachers enrolled at universities in Malaysia. Therefore, this aims to evaluate the knowledge of dental trauma among future teachers attending a local university.

METHODS

The study was a cross-sectional study that surveyed students from Faculty of Education – UiTM. Ethical approval (REC/394/18) was obtained from the Institute of Research and Management – UiTM prior to data collection commenced and participation was voluntary. There were no exclusion criteria except for inability to read and understand English and Malay languages. Participants for this study were from the Education of Faculty, Universiti Teknologi MARA, Puncak Alam. The subjects were randomly selected using random number generator and the number was matched against a master student list.

A three-part questionnaire was used to extract information from the students. The bilingual questionnaire was obtained from Hassan et al (2018)¹⁰ which derived the questions from multiple studies^{12,22-24} from previous studies questionnaire which was done by^{12,23-24}.

The questionnaire was divided into 3 sections. The first part of the questionnaire contains questions regarding the

demographic data of the participants, their teaching field, their teaching experience and first aid and dental trauma first aid training. The second part has questions regarding dental trauma knowledge and its management. It was divided into two scenarios. Each scenario contained a simulated traumatic dental injury case and was followed by a number of questions regarding each. The scenario of the first simulated case was regards broken permanent teeth of 8 years old boy associated with bleeding from gingiva and lips while the scenario of the second simulated case was about an avulsion avulsed upper incisor in a 12 year old girl. The third part of the questionnaire consisted of questions about students' satisfaction with the current level of knowledge regarding emergency management of dental injuries,

the ability to handle a dental emergency in school. The respondents were also asked what the best method is to enhance their knowledge of dental trauma.

RESULTS

Out of the 300 questionnaires sent, there were 83 subjects who responded to the questionnaire. Demographic part showed majority of the participants were female (74.7%) and mean age was 23.39 years old. The majority of participant were arts and design (59%). The participants who joined the study hailed from all over Malaysia with highest percentage of Kelantanese 19.3% and least number from Penang and Perlis (Table 1).

Table 1 Distribution of subjects according to gender, state of origin, field of study and current academic year

		Frequency	Percent
Gender	Male	21	25.3
	Female	62	74.7
State of Origin	Johor	9	10.8
	Kedah	8	9.6
	Kelantan	16	19.3
	Malacca	3	3.6
	Negeri Sembilan	2	2.4
	Pahang	4	4.8
	Penang	1	1.2
	Perak	7	8.4
	Perlis	1	1.2
	Sabah	2	2.4
	Sarawak	2	2.4
	Selangor	11	13.3
	Terengganu	10	12.0
	Kuala Lumpur	7	8.4

Study	Languages	6	7.2	
	Arts and Design	49	59.0	
	Biology	10	12.0	
	Physical Health	18	21.7	
Year of Study	2nd year	33	39.8	
	3rd year	17	20.5	
	4th year	33	39.8	
Age	Min	Max	Mean	Std. Deviation
	21	29	23.39	1.505

Although more than half the participants claimed they had some sort of first aid training during their course, only 1.2% of them noted having any dental related

emergency training. There was 18.1% of the participants who encountered dental trauma incident. (Table 2).

Table 2 <i>Past experience with dental trauma teaching</i>		Frequency	Percent
Have you had first aid training during your teaching training?	No	40	48.2
	Yes	43	51.8
If you did, did it include dental trauma first aid?	No	82	98.8
	Yes	1	1.2
Have you ever dealt with a dental injury prior to this?	No	68	81.9
	Yes	15	18.1

In the first scenario, the first question was about tooth identification in which the lowest percentage of participants (15.7%) were able to get the correct answer "permanent tooth". The second question was immediate management after the trauma. Most of the participants (49.4%) chose the answer "Contact his parents and advise them to send the child to a dentist immediately" while the only 1 participant (1.2%) answered "Reassure the child and send him back to class". (Table 3)

The second scenario contains three questions about avulsion. The first question inquired

about the immediate management after tooth avulsion. The answers showed that 10.8% would "put the tooth back in the socket and send the child to the dentist immediately". While majority of the answers (49.4%) were "put the tooth in solution and send the child to a dentist immediately". The second question was about the suitable method to rinse the tooth in which "Tap Water" was the most picked at 43.4%. The last question was about the choice of medium to transport the tooth. There were only 10.8% who answered "Milk" and 3.6% "Saliva". (Table3)

Table 3 *Knowledge regarding dental trauma*

		Frequency	Percent
Are the broken teeth likely to be primary (baby) or permanent teeth?	Primary Teeth	52	62.7
	Permanent Teeth	13	15.7
	Do not know	18	21.7
Which of the following actions would you consider most appropriate for this situation?	After school, contact his parents to explain what happened	2	2.4
	Contact his parents and advise them to send the child to a dentist immediately	41	49.4
	Reassure the child and send him back to class	1	1.2
	Send the child to the school nurse if available	39	47.0
The immediate action you would take is:	Put the tooth back in its place in its socket and send to the dentist immediately	9	10.8
	Stop the bleeding and send the child home	10	12.0
	Put the tooth in a solution and send the child to a dentist immediately	41	49.4
	Send the child to the nearest hospital	23	27.7
In your opinion, which of the following is most suitable to rinse the tooth?	Tap water	36	43.4
	Antiseptic solution	22	26.5
	Alcohol	5	6.0
	Mouthrinse solution	20	24.1
What is the best medium to transport the tooth to the clinic?	Milk	9	10.8
	Clean Tissue Paper	24	28.9
	Clean Handkerchief	15	18.1
	Ice	12	14.5
	Salted Water	20	24.1
	Saliva	3	3.6

Majority of the participants (80.7%) were not satisfied with their knowledge of dental trauma management as well as most of them (73.5%) though they are unable to handle dental emergency if they encountered.

However, 91.6% of the participants felt they need more traumatic dental injuries knowledge and 96.4% were interested to get more skills and exposure. (Table 4)

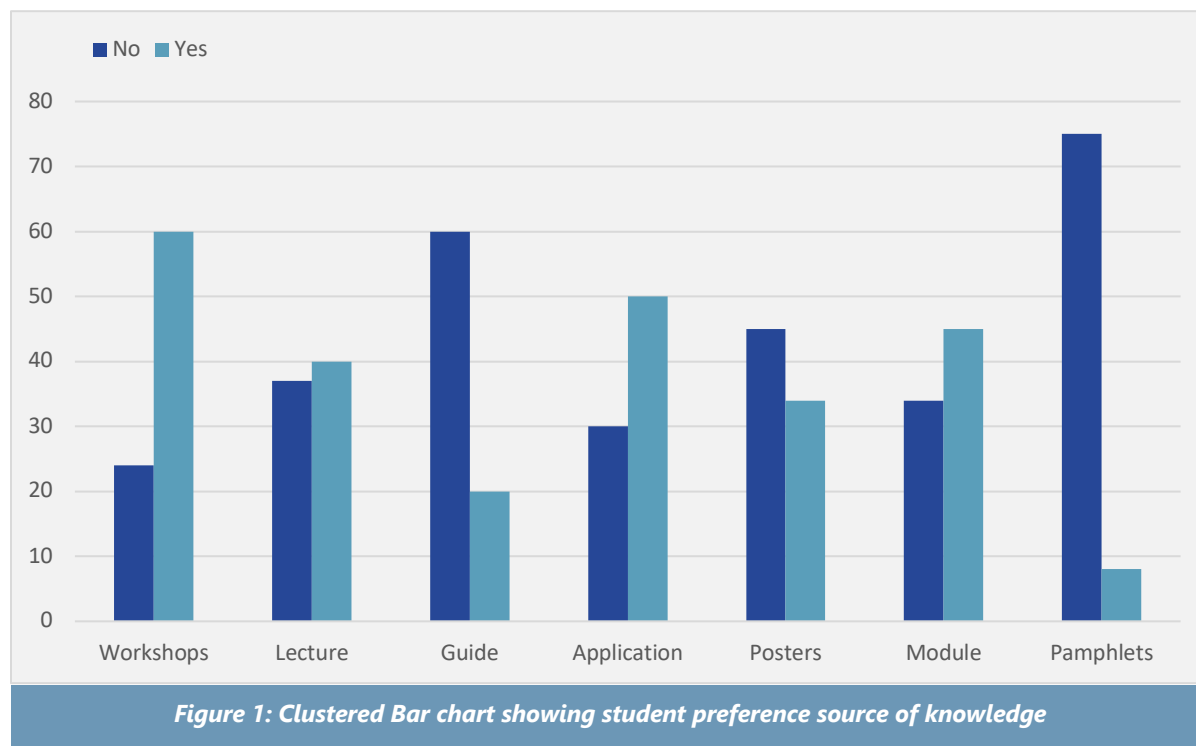
Table 4 *Perception of knowledge on management of dental injuries*

		Frequency	Percent
1. Are you satisfied with your level of knowledge regarding emergency management of dental injuries	No	67	80.7
	Yes	16	19.3
2. Do you think you will be able to handle a dental	No	61	73.5

emergency in school adequately?	Yes	22	26.5
3. Do you think you need more knowledge and exposure regarding dental emergencies?	No	7	8.4
	Yes	76	91.6
4. Would you be interested in having more exposure and skills in dealing with dental emergencies in school?	No	3	3.6
	Yes	80	96.4

When asked about their preferred tool of learning with regards to dental trauma, the participants were mostly inclined towards workshops and web applications.

Figure 1 showed the participants reflection of their choice of method to learn about traumatic dental injuries. Workshop was the choicest method to increased traumatic dental injuries knowledge and skill.



DISCUSSION

Teachers play an important role in the prevention and immediate management of dental injuries sustained on the school grounds. Hence, it is crucial to identify their knowledge level of traumatic dental injuries and find the best ways to improve their

knowledge and skills in managing dental injuries.

Majority of the participants in this study were females (75%) and the mean age of participants was 23.39 years.

In this study, 51.8% of the participants had first aid training. It is lower in comparison to

studies from India²⁵ and Iran²⁶ but 87.6% by (Pujita et al, 2013) similar to a Malaysian study⁹ which showed 42% with exposure to first-aid training.

The percentage of subjects who had prior exposure to first aid in dental trauma was a mere 1.2% which fared poorly in comparison to 5.3% (Marcano et al, 2018), 8% (Pujita et al, 2013), 7.6% (Arikan et al, 2013) 19.6% (Niviethithia et al, 2018) and 25% (Al Sari et al, 2019)^{27,28,30,32,33}. This could be attributed to the low weightage given to dental trauma in the training module. The module recommended by MOH has 11 topics of which only one slot is dedicated to dental trauma.

The identification of tooth type showed that only 15.7% answered correctly "permanent tooth". It is a low percentage in comparison to other studies in which participants able to identify the correct type of tooth was 67% (Tzimpoulas et al, 2020), 56.7% (Kaul et al, 2017), 60.4% (De Lima et al, 2012) and 44.1% (Hashim et al, 2011)^{11,20,34,35}. It is in contrary to (Mohandas and Chandan, 2009), and (Nirwan et al, 2016) in which tooth identification by participants was 24.4% and 33.2% respectively^{36,37}. The identification of the type of tooth is of utmost importance as the treatment approach is different for primary and permanent teeth. Since the majority of the respondents thought it was a primary tooth, they assumed that the tooth should not be replanted. If this were to occur in real life, the child would have lost a permanent tooth and be subjected to a long process of wearing prosthesis.

With regards to the immediate action taken

in the case of permanent tooth avulsion, more than 10% of the respondents would put the tooth back in the socket and send to the dentist while another 50% of them would put the tooth in a solution and send to the dentist. Both these options are favourable and would ensure better prognosis for the avulsed tooth. This study showed better results in comparison to another Malaysian study, Hassan et al (2018) who studied the response of primary school teachers in which only 4% would attempt replantation of an avulsed permanent tooth¹⁰. The majority of respondents in that study chose to send the child to the hospital without any management of preservation of the avulsed tooth¹⁰. The result of this study concurs with other global studies of reimplantation of avulsed tooth; immediate placement of tooth in socket was reported in 5.8% (Marcano-Caldera et al., 2018)¹³, 12% (McIntyre et al., 2008)¹⁵, 13% (Raoof et al, 2012)²¹, 14.4% (De Lima et al, 2012), 18.8% (Mori et al, 2007) and 19.3% (Hashim, 2011)^{21,32,34,35,38,39}.

The choice of rinsing solution was concerning as almost 50% of the respondents chose either antiseptic solution or mouthrinse. Rinsing with either of these solutions would gravely reduce the survival of the avulsed tooth. Only 43.4% the participants answered correctly, which was tap water and this results are similar to 38% (S. Hassan et al, 2018) and 51.8% (Marcano-Caldera et al., 2018). It was notably higher than 15.17% (Kaul et al, 2017) but much less impressive than reported by Hassan H et al (2018)⁹ which was 71%.

The preferred medium of tooth storage answers was tissue paper followed by a clean

handkerchief. Only 10.8% chose milk and 3.6% chose saliva. This is a similar result to 4.7% milk and 2.7% saliva (S. Hassan et al., 2018)¹⁰ (H. Hassan et al., 2018)⁹, and (Kaul et al, 2017)²⁰.

Although most of the participants (80.7%) expressed their dissatisfaction with their knowledge about emergency management of dental injuries, there was a great interest (96.4%) in improving their knowledge. This is a similar feeling of participants (64.7%, 59.44% in other studies done^{28 20}. However, 96.4% of the participants were enthusiastic to increase their knowledge.

CONCLUSION

There is low dental trauma knowledge among students training to become teachers at a higher learning centre. However, they showed a good attitude and interest to obtain dental trauma information and training. Therefore, this study suggests conducting educational intervention to provide traumatic dental injuries. The efficacy of various educational tools needs to be further studied to determine which would be the most suitable approach³⁸⁻⁴².

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CONFLICT OF INTEREST

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Oral Health Related Quality of Life (OHRQoL) in Children after Comprehensive Dental Treatment under General Anesthesia.

Fariza Md Kasim¹, Juanna Bahadun¹, Simran Olikh¹

ABSTRACT

Introduction: Early childhood caries (ECC) stands as a prevailing dental disease common among pre-schoolers globally and may lead to negative impact on their quality of life. Most cases often require treatment under general anaesthesia (GA) due to the extensive treatment required. The aim of this present study is to analyze the impact of comprehensive dental treatment under general anaesthesia on the oral health related quality of life (OHRQoL) of children and their families using the Early Childhood Oral Health Impact Scale (ECOHIS).

Methods: A prospective study was carried out on 70 parents of child patients (ASA I and II) aged 2 to 6 years old, who underwent comprehensive dental treatment under GA in Hospital Shah Alam. The parents were asked to complete the Early Childhood Oral Health Impact Scale (ECOHIS) questionnaire which was designed to assess 13 items related to quality of life of the patient. A pre-treatment form was filled and repeated again at six months after completion of treatment. The Wilcoxon signed-rank test served statistical analyses with the significance level at $p < 0.05$.

Results: The Wilcoxon Signed-Rank test indicated that the ECOHIS score after dental treatment were statistically lower than the score before dental treatment under GA, $Z = -17.61$, $p < 0.001$ revealing a large effect size of 1.49. Child symptom domain manifested the largest effect size ($r = 0.57$) compared to effect size of other domains. All domains of ECOHIS showed significance levels of $p < 0.001$.

Discussion: In this study, significant improvements were noted in both the child and family impact component. Parents reported that their children experienced less pain, improved oral intake, able to sleep better and less absence from school after dental treatment under general anesthesia. Parents also stated to have less distress and overall improvement in family function.

Conclusion: Comprehensive dental treatment under general anesthesia significantly improved the quality of life of children and their respective families.

Keywords: OHRQOL, Oral Health Related Quality of Life, Early childhood caries, ECOHIS, General anesthesia, Comprehensive dental treatment

(1) Department Of Paediatric Dentistry, Hospital Shah Alam, Selangor, Malaysia,

Corresponding Author

Fariza Md Kasim

Address

Department of Pediatric Dentistry, Hospital Shah Alam
Persiaran Kayangan, Seksyen 7,
40000, Shah Alam

Telephone Number

None

Email

fareezakasim@gmail.com
juannabahadun@yahoo.com
simran.olikh@gmail.com

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INTRODUCTION

Early Childhood Caries (ECC) is one of the most common health problems among toddlers and preschool age children^{1,7}. Early childhood caries (ECC) is characterized by the presence of one or more teeth decayed (cavitated or noncavitated lesions), missing (due to caries) or filled tooth surfaces in primary tooth in child 71 months of age or younger⁸. ECC is mostly found in young children under the age of 6 years⁷. Consequences of ECC includes a higher risk of pain or discomfort, abscesses, carious lesions in both the primary and permanent dentition, risk for delayed physical growth and development, increased days with restricted activity and diminished Oral Health Related Quality of Life (OHRQoL)^{2,9}.

Oral health plays a significant role in overall health and well-being of young children⁸. Poor oral health in children results in pain, refusal to eat, disturbance in their sleep and absence from school session indirectly affecting their social life⁹. Majority of early childhood caries are left untreated, and it presents as public health problem¹⁰. Findings from National Oral Health Survey of Preschool Children, 2005 showed caries prevalence of 87.19% in 1995, 76.2% in 2005 which was further reduced to 71.3% in 2015³. Although a wide number of children are able to receive dental treatment in a conventional setting, some patients fail to respond to the standard behavior management techniques and therefore require treatment under General Anesthesia (GA)^{10,11}.

Early Childhood Oral Health Impact Scale (ECOHIS) has been developed to evaluate the impact of OHRQoL on preschool children and their families^{4, 21, 22}. The original English version of ECOHIS has been translated into other languages including Malay language and is also found to be sensitive and responsive to Comprehensive Dental Treatment (CDT) under GA treatment effects⁴.

The purpose of this study was to analyze the impact of CDT under GA on OHRQoL of children and their families using ECOHIS. Three objectives were outlined in this study: to assess OHRQoL of children and their families before and after comprehensive dental treatment under GA as well as to determine the domains of OHRQoL most affected by severe ECC and having the most impact from CDT.

METHODS

Ethical approval was granted by the Medical Research and Ethics Committee (MREC), Ministry of Health (MOH), Malaysia. A prospective study with convenience sampling was carried out on a sample size of 70 parents of children with severe ECC aged 2 to 6 years old categorized as ASA 1 under the American Society of Anesthesiologist (ASA) classification, who underwent CDT under GA in Hospital Shah Alam, Selangor, Malaysia from year March 2017 to March 2019.

The study was voluntary as the explanation of the research was given to the parents / guardian and they provided their written consent prior to CDT under GA. The ECOHIS questionnaire was distributed and returned

on the same day which was collected as a pre-operative response.

The study consisted of clinical dental examinations and comprehensive dental treatment under GA. After 6 months post-operative CDT under GA recall visit, the ECOHIS questionnaire was distributed once again to all the parents / guardian and the feedback were collected as post-operative responses.

The survey tool used for assessing the OHRQoL was the Malay-ECOHIS questionnaire which has been validated. The Malay-ECOHIS comprises 13 criteria which has been divided into 2 main parts. It comprises the child impact section and family impact section. The child impact section encompasses 4 domains: child's symptom (1 item), child's function (4 items), child's psychology (2 items) and child's self-image with social interaction (2 items). The family impact section covers 2 domains: parental distress (2 items) and family function (2 items). The questionnaire has 5 response options; 0= never, 1= hardly ever, 2= occasionally, 3= often, 4= very often, and 5= don't know 4. The total score was calculated by summing up all the 13 items whereby the "don't know" will be scored as missing values

10, 16, 17.

DATA ANALYSIS

The Statistical Package of Social Science (SPSS) version 20.0 was used for data analysis and the level of statistical significance was set at $p < 0.05$. OHRQoL was measured by adding the criteria scores to create a total ECOHIS score which was tabulated from individual domains and sections. A higher score

displays greater oral health problems and poor OHRQoL. The magnitude of changes after CDT under GA was determined by subtracting the ECOHIS scores of pre-operative responses and post-operative responses.

The Wilcoxon signed-rank test was used to compare baseline and follow-up scores. Effect size statistics were calculated by dividing the mean of change score by the standard deviation of the baseline score as suggested by Cohen ⁵. An effect size of < 0.2 indicates a small but clinically meaningful magnitude of change whereas an effect size of $0.2 - 0.7$ shows a moderate magnitude of change while an effect size of > 0.7 shows a large change ⁵.

RESULTS

Ninety-five participants were recruited during the study period. Seventy participants (73.7%) completed the CDT under GA, of which 46 were boys with a mean age of 4.42, Standard Deviation (SD) of 0.96 and 24 were girls with a mean age of 4.22 (SD of 1.31). Twenty-five participants were excluded for reasons such as incomplete questionnaires and failed to attend follow up appointments. The parents / guardians who completed both pre-operative and post-operative ECOHIS questionnaires were 24 fathers (34.3%) and 46 mothers (65.7%).

The mean overall score of the ECOHIS pre-operatively is 7.61 (SD of 3.94) while for the post-operative total ECOHIS scores and domain scores declined remarkably to 1.79 (SD of 2.42) after CDT under GA. The child impact section pre-operatively showed a mean value of 5.26 (SD of 2.74) whereas the

post-operative showed a substantial reduction with a mean value of 1.14 (SD of 1.76). The most frequently reported item in the child impact section prior to intervention was child's symptom (pain) with a mean value of 2.11 (SD of 0.83) which showed an improvement post-intervention with a mean value of 0.39 (SD of 0.77). All domains in the child impact section showed a large effect size with a value of >0.7 except for child self-image and social interaction which exhibited a moderate effect size with a value of 0.62.

As for the family impact section, a mean value of 2.36 (SD of 1.70) was documented pre-

operatively whereas the post-operative showed a reduction with a mean value of 0.64 (SD of 1.02). Parental distress carrying a mean value of 1.40 (SD of 1.12) was the predominant reported criteria in the family impact section pre-operatively which displayed a significant improvement post-operatively with a mean value of 0.43 (SD of 0.68). Similarly, all the domains in the family impact section exhibited moderate effect size with values within 0.2 – 0.7. The results of the study were significant at $p < 0.001$. Therefore, this study indicates that dental treatments carried out under GA are likely to improve the quality of life in children and their families.

Table 1 The main ECOHIS domain scores before and after Comprehensive Dental Treatment under General Anesthesia (n=70).

ECOHIS domains (Number of items)	Pre-operative		Post-operative		p-value	Effect size
	Mean (SD)	Range	Mean (SD)	Range		
Total ECOHIS Score (13)	7.61 (3.94)	19.83	1.79 (2.42)	11.00	<0.001	0.85
Child Impact Section (4)	5.26 (2.74)	13.83	1.14 (1.76)	7.50	<0.001	0.82
Child symptom (1)	2.11 (0.83)	4.00	0.39 (0.77)	3.00	<0.001	0.80
Child function (4)	1.12 (0.77)	3.50	0.39 (0.25)	2.50	<0.001	0.73
Child psychology (2)	1.18 (0.92)	4.00	0.19 (0.52)	2.50	<0.001	0.72
Child self-image and social interaction (2)	0.84 (0.91)	3.50	0.18 (0.50)	3.00	<0.001	0.62
Family Impact Section (2)	2.36 (1.70)	7.00	0.64 (1.02)	4.50	<0.001	0.74
Parental distress (2)	1.40 (1.12)	4.00	0.43 (0.68)	3.00	<0.001	0.67
Family function (2)	0.96 (0.88)	4.00	0.21 (0.49)	2.50	<0.001	0.65

DISCUSSION

QHRQoL was formally conceptualized in 1978 and is described as the part of a person's quality of life that is affected by oral health²³. This research is aimed to analyze the impact of CDT under GA on the OHRQoL of children with severe ECC and the impact on their families using the Malay-ECOHIS questionnaire. This study found that substantial reduction was observed in the ECOHIS scores both in the children and family impact section. The greatest improvement was observed in the child symptom domain followed by child function which comprised of difficulty in eating and drinking, difficulty in pronouncing words and missing school. Interestingly, improvement in quality of life was also noted in the family impact section as this indicated that CDT under GA also benefited the family. This implies that caries in severe form has a significant influence on a child's daily function. It was also observed that the children were more social, smiled often and had better attendance in school⁶. Dental caries that is left untreated may associate with dental pain which leads to inability to eat and sleep, which can undesirably impact on child's daily activities such as absence from school due to symptoms^{6,18}. Additionally, some parents may need to take off or leave from work to care for the child^{6,23}. Evidence shows that untreated early childhood caries have consequences on the psychosocial, physical and functional of the child. In certain, the disease has also impact on the children's nutrition, growth and development²⁴.

The most worrying if children with severe ECC fail to cooperate with dental treatment in conventional setting^{23, 10, 11}. Thus, dental

treatment carried out in GA settings, can offer a better operative environment in which excellent quality of dental treatment can be completed in a single visit^{12, 13, 14}. Even though this form of treatment is deemed effective, it poses risks to a patient's overall health¹¹. It is also a costly as well as a resource intensive method which requires clear evidence of its benefits for children and their families¹. However, in uncooperative patients with severe ECC, CDT under GA is considered most suitable method of performing a complete oral rehabilitation in single visit²³. Despite the fact that many parents had expressed some levels of anxiety before and during GA due to the concern on serious morbidity or even in rare cases, mortality during GA²³. On the contrary, this research has shown significant improvement in the child's impact and family impact section where it has positive effects on the quality of life of the children and their families.

The strength of this study is a prospective study and the first study in Malaysia assessing the OHRQoL in children after CDT under GA. There are several limitations to this study which are worth mentioning for further study improvements. Firstly, selection bias could have risen from two sources where some parents did not return the survey questionnaire or either they did not answer all the questions sections completely and some manual questionnaire forms were also found to be missing throughout the study¹⁵. However, the incomplete survey questionnaire has been eliminated to avoid bias for this study. The second limitation of this study were patients who failed to attend the follow up visit after 1 month CDT under GA. Poor compliance with follow up indicates poor compliance with oral health habits that

is important for preventing recurrent caries^{12, 15}. Another limitation of this study is that it does not take into account of parents' socioeconomic status or educational background measure in relation to prevalence of Early Childhood Caries^{19, 20}.

Several recommendations in future studies may include the use of Google Form for the ease of respondents answering and responses can be analyzed automatically. Sending a written or a telephone call reminder 2 weeks prior to the follow-up visit can also be included. An additional recommendation is demographic data such as of parents' socioeconomic status, educational background could also be captured in future studies.

CONCLUSION

The OHRQoL of children with severe ECC and the impact on their families showed significant improvement after CDT under GA. Therefore, it appears that dental treatments are likely to improve the quality of life in children and their families. However, CDT under GA is not without risk and should be conducted only when necessary. In a nutshell, dental health professionals and public health programs should target their efforts toward prevention to reduce or decrease dental caries which may improve OHRQoL among children.

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CONFLICT OF INTEREST

The investigators declare they have no conflict of interest or competing interest.

ETHICAL APPROVAL

An ethical approval has been obtained from the Ministry of Health Medical Research Ethical Committee (MREC) (Approval Number: NMRR-17-1935-37027 (IIR)). We ensure written consent from all the respondents before beginning data collection. Participants in the survey are entirely voluntary.

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Oral Intravascular Papillary Endothelial Hyperplasia in a Child: A Case Report

Yan Yu Ong¹, Sockalingam G²

ABSTRACT

Intravascular papillary endothelial hyperplasia (IPEH) is a rare benign lesion that has a non-specific clinical presentation. It comprises about 2% of all the benign vascular tumours of the skin and subcutaneous tissue. Intraorally, it can mimic several pathological lesions such as pyogenic granuloma, mucocele, venous malformation, haemangioma, and to some extent, an angiosarcoma. Thus, diagnosis of oral exophytic lesion can be quite challenging for clinicians, especially in children. In such circumstances, a histopathological examination is crucial to achieve a correct diagnosis and prevent unnecessary aggressive treatment. Here, we discuss a case of recurrent intraoral swelling in a 10-year-old girl, which was first diagnosed as pyogenic granuloma, but subsequently revealed to be IPEH.

Keywords: Vascular tumour, Masson tumour, benign, hyperplasia

- (1) Department of Paediatric Dentistry, Tunku Azizah Hospital, Kementerian Kesihatan Malaysia.
- (2) Professor in Clinical Paediatric Dentistry Mahsa University.

Corresponding Author

Yan Yu Ong

Address

Department of Paediatric Dentistry
Tunku Azizah Hospital, Kementerian
Kesihatan Malaysia.

Telephone Number

+6012 3908 804

Email

ongyanyu28@gmail.com

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INTRODUCTION

IPEH, also known as Masson tumour, is an unusual benign vascular tumour composed of an excessive proliferation of endothelial cells in blood vessels.¹ It is commonly seen in the head, neck and the extremities.² Based on available literature, it is rarely reported in the oral cavity. Less than 120 cases of oral IPEH cases were reported in the English-language literature [3]. Although it is rarely described in the oral cavity, literature reviews showed that they were more commonly seen in the lower lip, followed by the upper lip, tongue, buccal mucosa and mandibular vestibule.^{1,3,4} In the oral cavity, IPEH normally present clinically as firm, and sometimes tender swelling, ranging from 0.5 to 1.8cm in diameter. It may appear as a reddish blue discoloration and most of the time is slow growing.⁵ The lesion has a

slight female predilection and is more common in adult with a mean average age of 42.6 years.⁴

Hashimoto et al (1983) described three distinct types of IPEH which are:

- i. Pure - Arising in a dilated vascular space
- ii. Mixed - Occurs within a pre-existing vascular lesion
- iii. Extravascular - Arising from haematoma

The significance that it may be mistaken for angiosarcoma is emphasized in multiple literatures.^{1,3} In this report, we discuss a case of IPEH that was diagnosed in a 10-year old child with a history of recurrent pyogenic granuloma.

CASE REPORT

A healthy 10-year-old girl presented to our department with a firm, painless, non-pulsatile, reddish blue swelling at the lower anterior gingiva. She had history of recurrent intraoral swelling over the lower anterior gingiva in the past 3 years (Figure 1), where excisional biopsies under a different hospital setting revealed a diagnosis of pyogenic granuloma.

At the time of presentation, the swelling was noticed by patient one year back. Clinical examination showed multiple reddish nodules on the left corner of her mouth (Figure 2) which was diagnosed as cherry angioma. Intraorally, there was a 1 x 1 cm swelling at the lower anterior gingiva in relation to lower right incisors (Figure 3). Oral hygiene was fair however there was noticeable plaque accumulation and calculus near the area of swelling.

Radiographic investigation (lower occlusal and orthopantomogram) showed no bony involvement. Based on her history and clinical presentation, a provisional diagnosis of pyogenic granuloma was made. To further confirm the diagnosis, excisional biopsy was carried out and a diagnosis of intravascular papillary endothelial hyperplasia was obtained. Histopathological examination revealed that beneath the surface covering of parakeratinised stratified squamous epithelium, numerous proliferations of plump endothelial cells predominantly arranged in lobular pattern and interspaced by dilated vascular channels having papillary like projections lined by monolayer endothelial cells. The intervening stroma has mild lymphocytic infiltration (Figure 4).

Up to date, no recurrence has been reported 18 months post-surgery



Figure 1 Clinical appearance of previous intraoral swelling at lower anterior gingiva diagnosed as pyogenic granuloma



Figure 2 Skin growth diagnosed as Cherry Angioma on the left side of lower lip area



Figure 3 Current intraoral swelling at lower right incisor

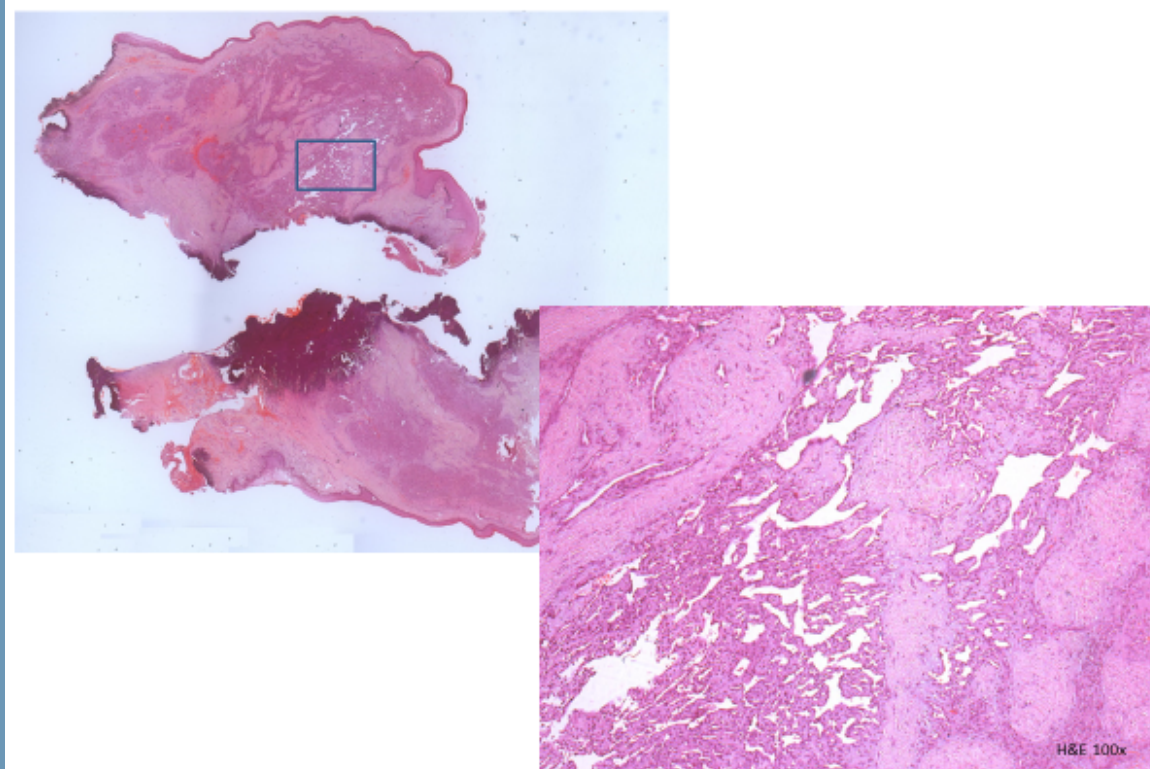


Figure 4 Histopathological findings of IPEH showing parakeratinised stratified squamous epithelium, with numerous proliferations of plump endothelial cells

DISCUSSION

Most intraoral lesions that occur in children present clinically as an exophytic growth.⁷ About 90-98% of soft tissue biopsies in

children are diagnosed as benign.⁷ This case report is about a child with a history of recurrent pyogenic granuloma who was later diagnosed with IPEH. It is the second case reported in children to date. The first case of

intraoral IPEH was reported in a 9-month-old boy by Devi et al (2004).⁸ Clinically, IPEH usually presents as a slow growing, firm, reddish-blue lesion with a slight elevation.¹ It is most seen in the head and neck region, but rarely occurs in the oral cavity.⁹ Based on clinical presentation alone, it is very difficult to differentiate a pyogenic granuloma from IPEH. Both lesions have been classified as benign vascular tumours under the ISSVA classification for vascular anomalies.¹⁰

Pyogenic granuloma on the other hand is a benign soft tissue lesion that is thought to result from chronic irritation, trauma, and hormonal factors. Oral pyogenic granulomas present as sessile or pedunculated nodules, ranging in size from a few millimetres to 2 cm, with a bright red colour and a smooth or ulcerated surface.⁷ In this case, the child has undergone two excisional biopsies where a diagnosis of pyogenic granuloma was made. When she presented to our department with another similar lesion, she was initially thought to have a recurrent episode of pyogenic granuloma based on history and clinical examination.

However, a third excisional biopsy has revealed a diagnosis of IPEH, which may be of pure form, arising in a dilated vascular space.⁶

CONCLUSION

In summary, IPEH is a benign lesion that can be managed by wide surgical excision with a low recurrence rate. Although benign, there is still a possibility that it may be mistaken clinically and histologically for angiosarcoma.

Even though the management for both lesions are the same, which is a total surgical excision, it is still important to diagnose patient accurately. Some immunohistochemical markers, such as the CD34, can also be useful in the identification and characterization of IPEH lesions.³ Another significance of this lesion that has been discussed in multiple literatures are the clinical and histopathological similarities it has with a malignant angiosarcoma. Inaloz et al. (2001) has described in his paper regarding the differential features of pyogenic granuloma, intravascular papillary endothelial hyperplasia (IPEH) and low-grade angiosarcoma.¹¹ The main features distinguishing IPEH from the other lesions are that they're commonly found in intravascular location and the presence of thrombotic zone. Incidental finding of extraoral cherry angioma in this patient is of no significance in relation to IPEH, however, it is also not commonly seen in children.

Management for IPEH is normally a wide surgical excision. Recurrence is rare and is most likely associated with a lesion that develops in a pre-existing vascular tumour (mixed form), such as the pyogenic granuloma.¹¹

A wider knowledge on the differential diagnosis of such lesion can help in achieving a correct diagnosis and subsequently appropriate management for the patient. Oral health practitioners should be aware of the clinical characteristics of these findings and the need for further workup or referral in selected cases.

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CONFLICT OF INTEREST

The authors declare no conflict of interest.

ETHICAL APPROVAL

The patient's anonymity is carefully protected, and the guardian of this patient consented to publication of this clinical data.

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Mandibular Osteomyelitis of A Young Adolescent Secondary to a Rare Cause: A Case Report

Girijaa Vathi Nachimuthu¹, Sumathy Perumal¹, Suhailiza Saharudin¹

ABSTRACT

Osteomyelitis of jaw is an inflammatory process of the entire bone including the cortex and the periosteum with multiple underlying aetiologies. Here we describe an interesting case report of an unusual cause of mandibular osteomyelitis. A healthy 15-year-old Indian girl presented to our centre with recurrent pain of the left mandible region for about 2 months. Clinical examination revealed an extra-oral left mandibular swelling with expansion of intraoral buccal bone. The clinical and the pre-operative radiological findings were suggestive of osteomyelitis of the left mandible and confirmed with histopathological examination. Initial resolution of the lesion was remarkable with the administration of antibiotic and surgical debridement. However shortly after the completion of antibiotics, she presented with recurrent pain on the left mandible, with generalized areas of radiolucency involving her entire mandible. Further investigations revealed unexpected haematological finding involving prominent vitamin deficiency. This case report emphasizes on identification of rare aetiologies, which should always be investigated and managed accordingly if the disease is not responding to conventional therapy. Identification and understanding the aetiology remains of paramount importance to aid in providing optimal treatment.

Keywords: *Osteomyelitis, mandible, vitamin deficiency*

(1) Department of Paediatric Dentistry, Hospital Raja Permaisuri Bainun, Ministry of Health Malaysia, Ipoh, Perak.

Corresponding Author

Dr.Girijaa Vathi Nachimuthu

Address

Department of Paediatric Dentistry
Hospital Raja Permaisuri Bainun
Ipoh, Perak.

Telephone Number

-

Email

girijaanachi@gmail.com

DOI

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INTRODUCTION

Osteomyelitis of mandible denotes the inflammation of bone, involving an inflammatory process in the medullary spaces or cortical surfaces of bone, that extends away from the initial site of involvement, which fails to heal after an appropriate

intervention. Commonly known as 'condensing osteitis', osteomyelitis of jaw is usually classified into acute (≤ 2 weeks), subacute (between 2 weeks to 3 months) and chronic (≥ 3 months) forms with various reported aetiologies¹. The primary cause of chronic osteomyelitis mainly could be from odontogenic infections, traumatic dental

procedures, inadequate antibiotic therapy, inappropriate selection of antibiotics, irradiation or diagnostic failures.² Osteomyelitis of mandible has also been associated with underlying systemic conditions such as diabetes, autoimmune diseases, malnutrition, acquired immunodeficiency syndromes and malignancies. Once the aetiology is identified, accurate diagnosis and the removal of source of infection are made feasible. This, in turn, improves the prognosis tremendously. While osteomyelitis of jaw in young adolescents has a good prognosis due to their better healing capacity, this condition should always be diagnosed early and treated promptly to avoid any serious complications. The following is an account of a rare and an unusual origin of mandibular osteomyelitis, highlighting on the rather, challenging diagnostic process.

CASE HISTORY

A healthy 15-year-old girl presented at our dental clinic with a chief complaint of recurrent pain at lower left jaw for the past 2 months. The described intermediate to severe pain, was radiating in nature involving the left body of mandible, extending towards the left temporal and left postauricular region. There were no apparent exacerbating factors contributing to the pain, neither was there a history of trauma. This worsening pain prompted her to seek treatment from various medical and dental practitioners. The patient's medical history was otherwise non-contributory.

Visual inspection revealed diffused extraoral left mandibular swelling measuring about 2 cm x 2 cm which was firm and non-tender upon palpation. The overlying skin appeared

to be normal in colour. The patient had no paraesthesia over left mandible region and her mouth opening was adequate. She also denied any history of trismus or previous temporomandibular joint pain.

Intraorally, the patient presented with prominent left mandibular buccal bone expansion. The region of lower left first premolar tooth to lower left first permanent molar tooth was very tender upon palpation and percussion. Then the teeth were further investigated to determine their pulpal integrity. The lower left second premolar was not responsive to pulp sensibility test including the electric pulp test and cold test, indicative of non-vitality. Meanwhile, the adjacent lower left first permanent molar, though presented with large occlusal tooth-coloured restoration, appeared to be vital. There was no positive finding of periodontal pocketing or pathological mobility. Otherwise, there was no signs of trauma recorded on any of the teeth at the site of complaint.

Her preliminary radiological work up comprised of an Orthopantomogram (OPG) and a Cone Beam Computed Tomography (CBCT), as showed in Figure 1(a) and 1(b), which revealed evidence of moth-eaten appearance on the left body of mandible extending from mesial of lower left first premolar to distal of lower left first permanent molar. There was significant bone loss noted at the buccal aspect and widening of the periodontal ligament space widening over distal aspect of the lower left first premolar root. The Computed Tomography (CT) (Figure 2) which was scheduled soon after, exhibited mixed lytic sclerotic lesion involving the left body up until the left angle of mandible. There was generalised thinning

of bone cortex with some areas of buccal cortical bone breached at the body of left mandible. Those lesions were associated with

periosteal reaction. Pre-operative blood investigations were within normal parameters.

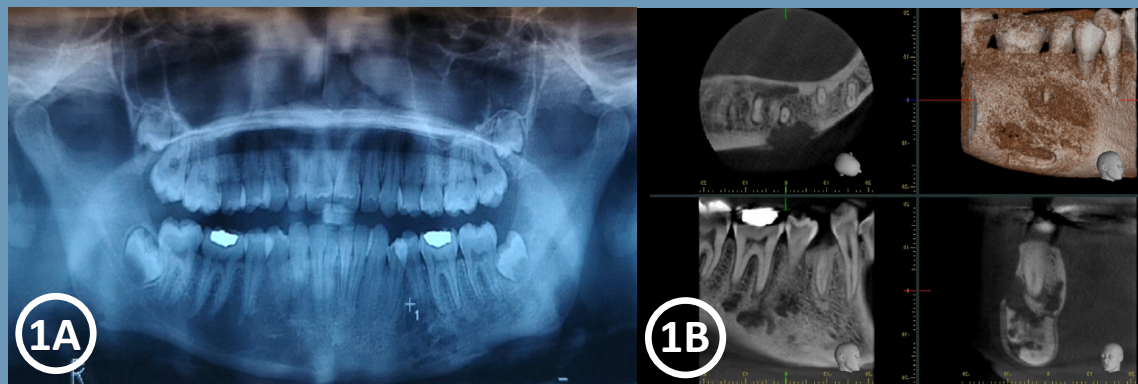


Figure 1 pre-operative OPG (1a) and CBCT (1b) depicting sites of lesion at left mandible.

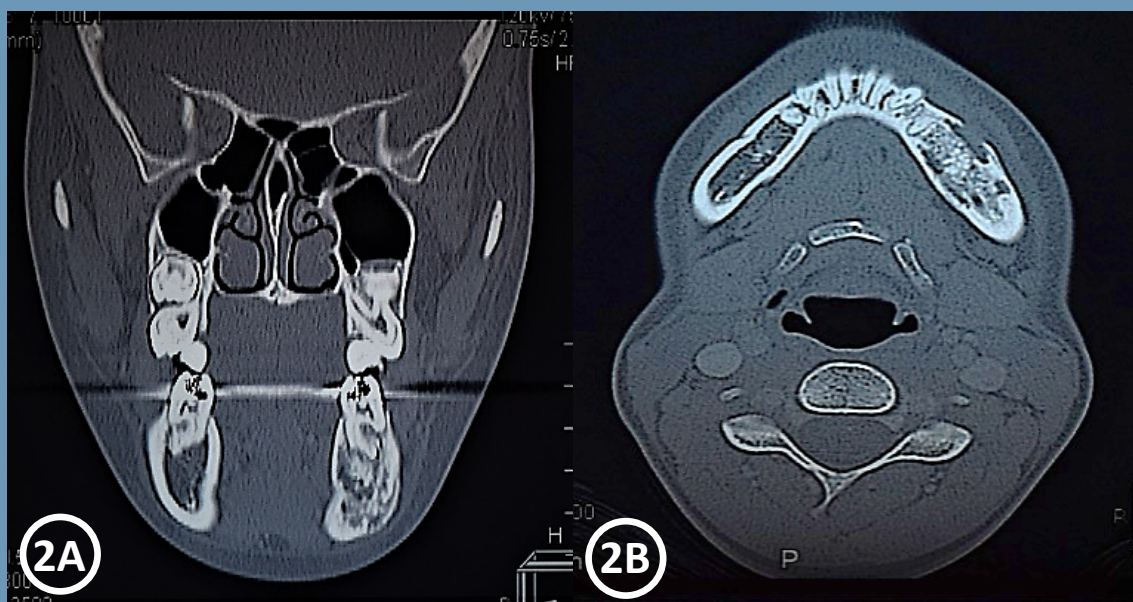


Figure 2 CT views; coronal (2a) and axial (2b); showing involved left mandible region.

Based on the history and clinical-radiological characteristics, a provisional diagnosis of osteomyelitis of left mandible was established. The other differential diagnosis were fibrous dysplasia, non-ossifying fibroma, florid cemento-osseous dysplasia, and osteosarcoma.

As part of initial pain management, pulp extirpation of lower left second premolar was performed since the tooth was symptomatic with signs of non-vitality. The patient was commenced on penicillin-based antibiotics for a period of eight weeks empirically along with adequate analgesics for infection and pain control. In the meantime, she was scheduled for exploration, debridement, and

incisional biopsy of left mandible under general anaesthesia (GA), during which, part of the affected bone was removed and sent for histopathological examination (HPE) for an advanced analysis. Complementary curettage and irrigation were performed supplemented with extraction of potential causative lower left second premolar tooth to eliminate possible residual infection.

HPE result confirmed the diagnosis of osteomyelitis. It demonstrated small, irregular fragments of bony trabeculae with empty lacunae and attached fibro-cellular tissue that displayed chronic inflammatory cell infiltration.

Post operatively, the patient completed the antibiotic therapy for a total of 8-weeks period. Initial phase of recovery was remarkable, whereby at 2 months follow-up visit, it showed a significant healing of lesion, and the patient was entirely asymptomatic. There was a complete resolution of facial swelling with notable reduction in left mandibular buccal bone expansion.

However, the patient complained of recurrent pain on left mandible at 3 months post-operative recall visit. She gave a history of generalized weakness, lethargy, loss of appetite and noticeable weight loss for the last one month. Apart from the tenderness at the left buccal mandible region, there was no other noticeable abnormality.

For further assessment, radiographic investigations were carried out, which revealed new lesions with evidence of

sequestrum formation over the left mandibular region. Presence of moth-eaten appearance at the contralateral side of the mandible was the new incidental finding (figure 3a), indicating generalized mandibular osteolytic lesion.

At that instant, it was vital importance to conduct the necessary haematological examination to rule out any undiagnosed underlying systemic bone disease, a point often overlooked. Our patient had normal parameters of full blood count, liver function test and renal profile. On the other hand, the test level of erythrocyte sedimentation rate (ESR) was elevated at 28mm/hour. The mineral metabolism tests comprising of serum levels of calcium, phosphate, potassium, parathyroid hormone, and alkaline phosphatase were within normal range but there was a notable deficiency of Vitamin D with 25-Hydroxy Vitamin D level of 7.5ng/ml.

Based on the history, clinical findings and investigations, the diagnosis was finally concluded as chronic osteomyelitis of mandible secondary to Vitamin D deficiency.

The patient was subsequently referred to an Endocrinologist for the management of Vitamin D deficiency, for which she was commenced on Vitamin D3 supplement (25'000 units/month). The patient had since been recovering well with significant mandibular bone healing consisting of sclerotic left mandible with no lytic lesion during a follow-up recall visit at 1 year as depicted in the OPG (figure 3b).

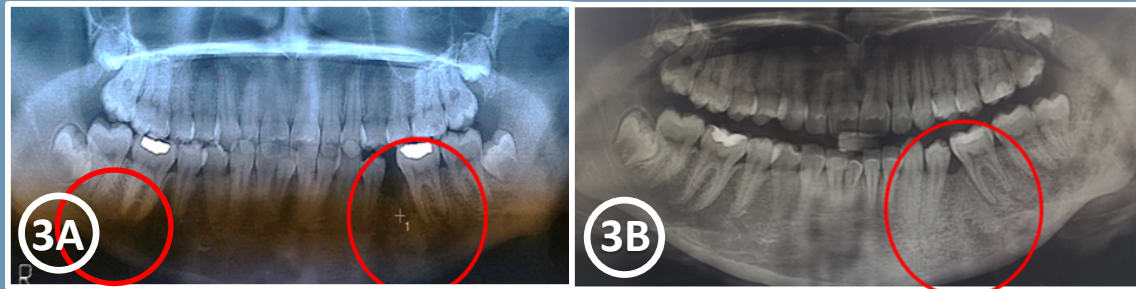


Figure 3 post-operative OPG taken at 3-months (3a) and 1-year (3b) depicting sites of lesion at left mandible.

DISCUSSION

Osteomyelitis is an inflammatory disease of the bone that usually begins as an infection of the medullary cavity, rapidly involves the haversian system, and quickly extends to the periosteum of the area². Chronic osteomyelitis of mandible is a bone disease that is characterized by inflammatory processes, mainly by bony destruction, and sequestrum formation³. It is a well-known entity in the historical literature where in the absence of antibiotics, infections and fractures of long bones frequently failed to heal⁴. Being a rare condition, osteomyelitis of jaw has been largely associated with odontogenic infections, and subtly associated with systemic diseases including diabetes, autoimmune states, malignancies, malnutrition, acquired immunodeficiency syndrome, and medications such as steroids and bisphosphonate. The disease is rare, accounting for 2% to 5% of all osteomyelitis cases. It primarily affects children, with a female to male ratio of 5:1, with no racial predilection⁵. In a five-year follow-up study of 23 patients published in 2002, the median age of onset was 10 years with a reported range of 4 to 14 years⁵. In childhood, the most common route of infection is via hematogenous spread of a microorganism. Less common, osteomyelitis may also occur

via contiguous spread such as after direct trauma. Most cases of acute hematogenous osteomyelitis in children are caused by Gram-positive bacteria, principally *S. aureus*. The bacterial contamination of bone tissue is best determined by bone biopsy under radiographic guide⁶. The most frequently bacteria associated with osteomyelitis are *Staphylococcus aureus*, Gram negatives (*Pseudomonas aeruginosa*), and anaerobe bacteria (*Bacteroides fragilis*)⁶. Chronic osteomyelitis is characterized by recurrent pain, swelling, limited mouth opening, absence of suppuration, periostitis, occasionally regional lymphadenopathy and reduced inferior alveolar sensation, neuropalsy in the involved area, pathologic fracture, and trismus.²

Chronic osteomyelitis seems to have a stage-like course. Initially it is often seen with a radiographically mixed sclerotic and osteolytic pattern, which later changes to sclerotic⁷. The sclerotic changes are characteristic of diffused sclerosing osteomyelitis. The term non-suppurative does not exclude the presence of pathogens. It seems unclear whether non-suppurative osteomyelitis of the jaw is aseptic or not, or if both possibilities exist. Chronic non-suppurative osteomyelitis can be caused by certain bacteria (*Actinomyces* and *Eikenella*

corrodens) presenting mild or no symptoms and so often diagnosed several years into the disease process⁷. Other authors report negative bacterial findings indicating a non-bacterial pathogenesis. Here, we have presented a case of non-infectious mandibular osteomyelitis, in which the unexplored aetiology posed a diagnostic dilemma at the initial stage.

Osteomyelitis is diagnosed based on the patient history, clinical examinations, and the surgical and radiographic findings. Non-suppurative osteomyelitis is characterized by chronic non-specific inflammation, increased bone resorption and deposition of varying degrees⁷. Histopathologic examinations can be consistent with the diagnosis and the microbiologic tests can be helpful. Histologically, an increased number of osteoblasts, thickened bony trabecula, and fibrous marrow replacement are found. Pathologic bone remodelling and the presence of chronic inflammatory cells are often cited as indicators of osteomyelitis². The general principal of management for an osteomyelitis is the antibiotic therapy along with removal of cause and surgical debridement⁸. Treatment varies from a range of simple non-invasive approaches to more invasive and radical treatment⁴. The non-surgical approach includes antibiotics, NSAIDS, hyperbaric oxygen therapy, bisphosphonate treatment, and muscle relaxants. Amoxicillin was chosen for non-surgical management because of its broad antibacterial activity against anaerobic organisms commonly present in chronic 'mixed' odontogenic infections and it has the potential to penetrate well and achieve high therapeutic concentrations in bone⁹. Following the failure of a non-surgical approach, a surgical intervention to consider

include decortications alone, decortication with bone grafting, partial (marginal) resection, and segmental resection⁴. In the event where the condition fails to heal following a routine therapy, one should explore other possible aetiological factors such as patho-physiological or underlying medical conditions⁸. Hence, further investigation is mandatory to clearly define the cause of disease to achieve a long-term successful outcome. This case highlights on how such omission influenced the long-term stability of healing, as shown by patient responding well at the early stage but developing recurrence with worsening of symptoms shortly after the initial therapy.

A systemic involvement of the host should not be overlooked when a condition does not respond to the conventional therapy. This clinical case was noteworthy to be reported as the oversight to identify and understand the actual rare systemic aetiology, caused recurrence of symptoms.. An advanced hematological analysis was performed in this patient which led to identification of the Vitamin D deficiency.

Vitamin D plays a pivotal role in keeping our body function at an optimal level; particularly in regulation of bone health, regulation of blood pressure, calcium haemostasis, neuro-development, regulation and differentiation of cell growth, and immunomodulation (prevention of autoimmune diseases and control of invading pathogens)¹⁰. In a healthy individual with sufficient host immunity mechanisms, these factors form a carefully balanced equilibrium¹¹. If this equilibrium is disturbed by altering one or more of these factors, deep bone infection establishes. Usually, there is an underlying predisposing factor like malnutrition, alcoholism, diabetes,

leukaemia, or anaemia⁸. Other predisposing factors are those that are characterized by the formation of avascular bone, e.g., therapeutically irradiated bone, osteopetrosis, Paget's disease, and florid osseous dysplasia¹¹. Osteomyelitis is more commonly observed in the mandible because of its poor blood supply as compared to the maxilla, and because the dense mandibular cortical bone is more prone to damage and, therefore, leading to infection. Deficiency of vitamin D in our patient was believed to have predisposed her to altered immune response, hence increasing the susceptibility of patient to infection, contributing to the osteomyelitis of left mandible and the generalized lysis of mandibular bone¹⁰. This was proven by a remarkable outcome which was observed with Vitamin D correction, in addition to the conventional regime of antibiotics and surgical therapy offered to this patient.

Furthermore, vitamin D has been increasingly recognized as an immune mediator that may play a role in the pathogenesis of infections¹². It showed that vitamin D deficiency (<50nmol/L) is associated with an increase in infection rate, sepsis, 30-day mortality, and in-hospital mortality in adult critically ill patients, worldwide. After a thorough literature search, only a few articles have been published regarding a possible connection between vitamin D deficiency with the development of bone infections. These studies showed that there was an increased prevalence in vitamin D deficiency among those patients who concurrently had a bone infection, but not in paediatric population specifically¹². An exact causal relationship could not be established, but it was suggested that vitamin D supplementation may be a possible way to lower the risk of periprosthetic joint infection.

Vitamin D associated osteomyelitis is relatively uncommon that documentation of such entity is very scarce. One study stated an almost comparable case history and observed a similar pattern of management and outcome of treatment for this rare condition⁵. Furthermore, we speculate whether the co-existing conditions may affect the patient's treatment, outcome, and prognosis, since vitamin D is extremely relevant in bone health.

If a patient is not recovering from osteomyelitis despite having treated for vitamin deficit, an alternative treatment needs to be considered⁵. Use of bisphosphonate showed remarkable recovery in bone abnormality in a 17-year-old patient with Vitamin D deficiency⁵. Bisphosphonates are pyrophosphate analogues capable of potentially inhibiting osteoclastic bone resorption/ remodelling⁷. Biphosphonate ranging from disodium clodronate, pamidronate, and alendronate are reported to be effective as pain relieving agents in diffuse sclerosing osteomyelitis of the mandible. The mechanism of pain relief is not known. Based on these evidence, should there be any future recurrence of symptoms in patient with refractory infection, bisphosphonate therapy could pose to be a viable option that is worthwhile to be considered⁵.

CONCLUSION

In summary, rare, and unusual aetiologies should not be excluded in the diagnostic process of a disease. Vitamin D deficiency, though an uncommon cause of osteomyelitis, should always be investigated especially when the condition is not

responding to the conventional therapy. We present a rare case of mandibular osteomyelitis with mandibular localisations to emphasise that any disease that appears to be simple can lead to the discovery of an

unexpected and rare. Accurate identification of cause and the appropriate management would certainly improve the long-term prognosis.

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The authors declare no conflict of interest.

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Maxillary and Mandibular Osteomyelitis Associated with Osteopetrosis: A Case Report

Eunice Chong¹, Leong Kei Joe¹, Azalina Osman¹, Nurnabiha Mohd Jabid¹, Goh Dai Shan¹

ABSTRACT

Osteopetrosis is a rare hereditary bone disorder characterized by the malfunction of osteoclast resulting in bones hardening. Complications such as osteomyelitis are often difficult to treat and can be life-threatening.

A 3 year old boy, born of consanguineous marriage, was referred with concerns about his missing and abnormal-looking teeth. Apart from being diagnosed with learning disability and bilateral eye retinal dystopia, he presented with hypodontia and multiple grossly carious teeth. Total extraction was later performed under general anaesthesia.

Intraoperatively, his teeth were noted to be hypomineralised with abnormal morphology. Postoperative reviews showed poor wound healing of the sockets with exposed alveolar bone on both jaws. The child was later diagnosed with osteopetrosis after a chest x-ray was taken as part of the investigation due to severe anemia and pneumonia. His oral condition continued to deteriorate further despite multiple courses of antibiotics and repeated debridement of necrotic bone. He is currently on long term follow-up for oral wound care along with long term antibiotics.

Osteopetrosis with chronic suppurative osteomyelitis of the jaw is extremely rare. The clinical findings and course of treatment in this case serve as valuable information to understanding and managing the disease better.

(1) Sabah Women & Children's Hospital, Kota Kinabalu, Sabah

Corresponding Author

Eunice Chong

Address

Department of Pediatric Dentistry,
Women and Children Hospital,
Sabah.

Telephone Number

-

Email

eunice_le@hotmail.com

DOI

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INTRODUCTION

Osteopetrosis or also known as "marble bone disease" is a descriptive term that refers to a rare, heritable bone disorder characterized by increased bone density on radiographs

caused by failure of osteoclast development or gene mutations and function. The overall incidence of these conditions is 1 in 20,000 births for benign autosomal dominant osteopetrosis (ADO) and malignant autosomal recessive osteopetrosis (ARO) has an incidence of 1 in 250,000 births.¹

The benign autosomal dominant form, manifested in adults have complications that are confined to the skeleton, including fractures, scoliosis, hip osteoarthritis and osteomyelitis, particularly affecting the mandible in association with dental abscess or caries. The patients are often asymptomatic at birth but frequently exhibit fractures by the end of their first decade of life. Most patients survive into adulthood but with significant disability^{2,3,4}

The malignant autosomal recessive form has an onset in the first decade of life in association with a poor prognosis due to bone marrow compression and compressive neuropathies. The increase in bone density can paradoxically weaken the bone, resulting in a predisposition to fractures and osteomyelitis. Hematopoiesis is so severely decreased that anemia, thrombocytopenia, and hepatosplenomegaly are produced and some of these complications may even lead to death. Neuropathies related to cranial nerve entrapment occurs, leading to visual and hearing deterioration^{5,6}

Treatment of osteopetrotic conditions is largely symptomatic, although haematopoietic stem cell transplantation is

employed for the most severe forms associated with bone marrow failure and currently offers the best chance of longer-term survival in this group. The severe infantile forms of osteopetrosis are associated with shorten life expectancy. Life expectancy in the adult onset forms appears to be unaffected.¹

CASE REPORT

A 3 year old boy, born of consanguineous marriage was referred to us from the Department of Ophthalmology for missing and abnormal looking teeth. He came into our clinic always seated in a stroller with a cap on his head and a straw in his hand. The patient had short stature, frontal bossing, assisted ambulation with his head always looking down and his both eyes squinting [Figure 1]. Apart from being diagnosed with global developmental delay and having autistic features, he was under investigation for bilateral congenital retinal dystrophy, bilateral alternating esotropia and bilateral jerk nystagmus. He otherwise did not have any history of hospitalizations, past surgeries or medical allergies.



Figure 1 In a stroller, finding comfort under his cap and a straw in his hand. Frontal bossing with squinting eyes.

On our clinical examination, he presented with hypodontia and multiple grossly carious primary teeth. Due to a lack of cooperation, comprehensive dental treatment under general anaesthesia was performed. Intraoperative findings includes hypodontia, abnormal tooth morphology with short-

formed roots and hypomineralised crowns [Figure 2]. Sockets were sutured with resorbable sutures and packed with hemostatic agent. Post-operative recovery was uneventfully, patient was discharged home with analgesics and was given a date for post-op review.



During his 8 months' post op review, poor healing was noted over the extraction sockets, with presence of large necrotic bone [Figure 3]. We did further investigations to determine the cause of his poor wound healing. Peripheral blood film was performed

and the results came back as normochromic normocytic anemia. Multiple soft and hard tissue specimens were then sent for culture and sensitivity tests and the results are as seen in Table 1.

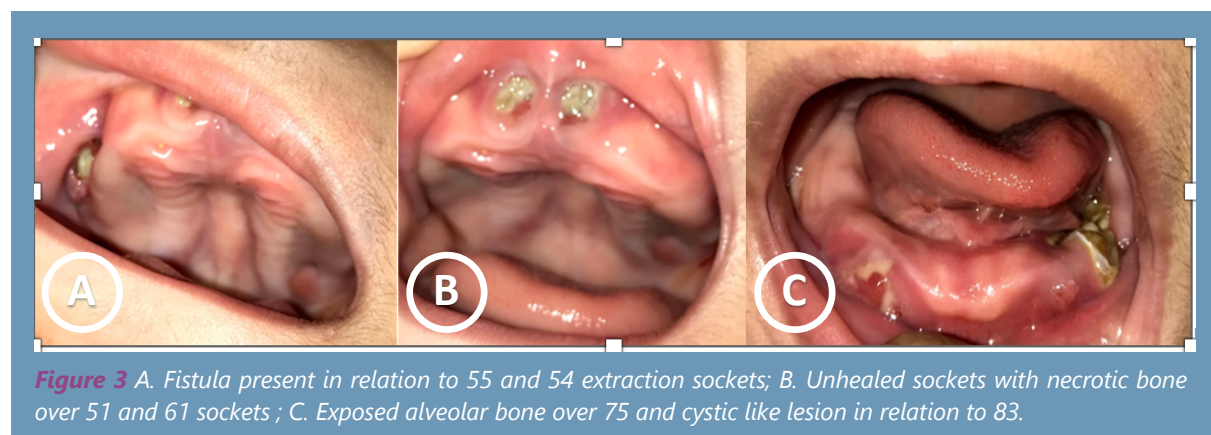


Table 1 *Types of specimen sent for culture and sensitivity tests and their results*

Anterior Maxillary Soft Tissue	Amp-C Producer Enterobacter Cloacae	Ampicillin Augmentin Cefoxitin
	Non-ESBL Serratia Marcescens	Ampicillin Augmentin Unasyn
Left Maxillary Bone	Klebsiella Pneumoniae	Ampicillin
	Escheriachia Coli	Ampicillin

He was prescribed multiple cycles of antibiotics as listed in Table 2 for a time period of 3 months but the lesions did not show any signs of healing, there was still pus

discharge and foul smell intraorally during his reviews. Workup by the Paediatric Medical team for possibilities of poor wound healing also did not show anything conclusive.

Table 2 *Type of antibiotics*

CYCLES	
Metronidazole 7.5mg/kg	8 hourly for 5 days
Amoxicillin 10mg/kg	8 hourly for 5 days
Metronidazole 7.5mg/kg	8 hourly for 5 days
Amoxicillin 10mg/kg	8 hourly for 5 days
Metronidazole 7.5mg/kg	8 hourly for 5 days
Cephalexin 25mg/kg	8 hourly for 5 days
Metronidazole 7.5mg/kg	8 hourly for 5 days
Cephalexin 25mg/kg	8 hourly for 5 days
Metronidazole 7.5mg/kg	8 hourly for 5 days
Cefuroxime 15mg/kg	12 hourly for 5 days

We finally came to a diagnosis when he was later admitted into the ward for severe anemia and pneumonia. A chest radiograph was taken as part of the investigation, and it showed an increase in opacity of the bones suggestive of sclerotic changes [Figure 4]. A skeletal survey was performed and confirmed

his diagnosis as osteopetrosis with suppurative osteomyelitis of the maxilla and mandible. Magnetic Resonance Imaging (MRI) was done under general anaesthesia and showed generalised thickening of skull vault likely systemic in nature.



Figure 4 Chest radiograph showing increased bone density and diffuse sclerosis suggestive of osteopetrosis

With a diagnosis in hand, our patient was referred to the Department of Oral and Maxillofacial Surgery for bone debridement and primary wound closure under general anaesthesia. Primary closure was attempted

during the first bone debridement, but unfortunately during his post op review, the wound had broken down [Figure 5, 6] and there was no positive progress to his condition after that.



Figure 5 sutures were loose and alveolar bone is visible



Figure 6 Wound breakdown on third and fourth quadrant

He subsequently underwent a total of 4 bone debridement procedures under general anaesthesia with similar outcomes. Intraoperative findings for the maxillary bone were sinus fistula over the first quadrant, exposed necrotic bone over the second quadrant,

quadrant with bulbous palatal and tuberosity bone. Over the mandible, there was also exposed necrotic bone over the third quadrant with thin alveolar bone and sinus opening over the fourth quadrant [Figure 7, 8].

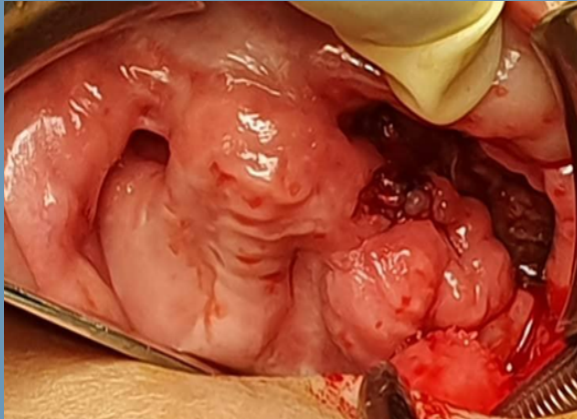


Figure 7 Maxillary- fistula over the first quadrant, exposed necrotic bone over the second quadrant and bulbous palatal bone



Figure 8 Mandible-exposed necrotic bone over the third quadrant with thin alveolar bone and sinus opening over the fourth quadrant

Over the past 3 years he has also been admitted multiple times for neutropenic fever and has undergone blood transfusion for anemia. He is currently on long term antibiotics (Amoxicillin and Clavulanic Acid) under the purview of the paediatric infectious disease team until they deem he is suitable to discontinue the antibiotics. His parents have been informed of his poor quality of life and guarded prognosis long term.

5 years following his first dental procedure, his current condition remained as quo. There

is necrotic palatal bone that bleeds easily with soft tissue bulging at the rugae area and exposed necrotic alveolar bone with irregular soft tissue margins over the right mandible [Figure 9,10]. He has also experienced submandibular swelling on a number of occasions that has required hospital admission for intravenous antibiotics.

This case is said to be the first case of osteopetrosis to be reported in Sabah. For now, a multidisciplinary approach is the best way forward.

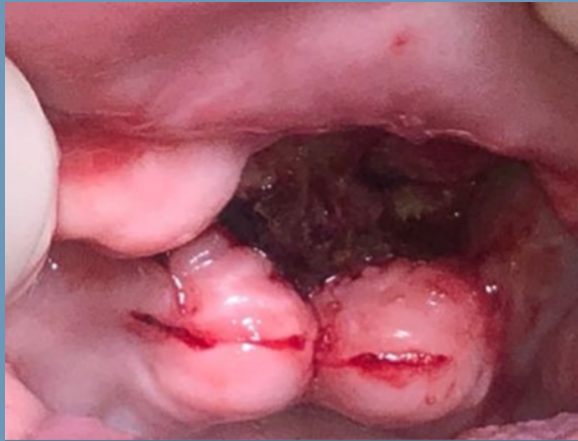


Figure 9 Necrotic palatal bone that bleeds easily with abnormal soft tissue bulge.



Figure 10 Irregular soft tissue margins with necrotic alveolar bone of the mandible.

DISCUSSION

Bone turnover and consequently, bone density is highly regulated by the balance processes of bone formation by osteoblasts and bone resorption by osteoclast. Osteopetrosis is a rare genetic bone disorder characterised by failure of osteoclastic cells. Marrow spaces decreased in the most severe and fatal form, which leads to extramedullary hematopoiesis and hypersplenism resulting in anemia, severe thrombocytopenia, and hepatosplenomegaly.⁷

Unlike the few reported cases elsewhere where the diagnosis was known before dental treatment, it was rather unfortunate for our patient to have endured such a painful journey of poor wound healing and bone necrosis before we could come to a definite diagnosis of Osteopetrosis. Radiographic images could not be taken due

to behavioural management difficulty during his post op reviews bearing in mind that this child also has autistic features. The rarity of the disorder and the insignificant level of difficulty during dental extraction did not prompt the paediatric dental and medical team to investigate further for other bone disorders.

Perhaps, looking in hindsight of the follow up care, a more aggressive approach such as CT scans (of the maxilla and mandible) under general anaesthesia together with long term antibiotics could have been prescribed earlier. Osteopetrosis with chronic suppurative osteomyelitis of the jaw is extremely rare. Despite establishing a diagnosis, once osteomyelitis has set in, bony debridement of the unhealthy jawbone and attempts for primary closure have proven to be extremely difficult despite multiple surgeries. Although the child is currently managed conservatively with long term antibiotics, we have yet to

know the long-term outcome which in our opinion is extremely guarded, especially when it is coupled with other medical issues due to bone marrow suppression. The clinical findings and course of treatment in this case

serve as valuable information to be added into the literature of rare diseases for the better understanding and management of this condition.

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Micro-Marsupialization in Ranulas: The Way Forward for Children?

Jamil A¹, Vijayakumar T¹

ABSTRACT

A ranula is a cystic lesion in the floor of the mouth arising from the sublingual gland as a result of extravasation of mucus in the surrounding tissue. It poses a challenge to manage surgically due to its location and proximity to vital structures. Current available treatment modalities include surgical

excision of affected sublingual gland, marsupialization, micro-marsupialization, laser, and sclerotherapy. Despite widely reported, there's currently no acceptable guideline for management of ranulas, especially in children. Current research advocates a less invasive approach to manage this lesion due to potential complications that may arise from removal of the salivary gland. This paper aims to highlight a case of a ranula in a 10-year-old that was successfully managed using micro-marsupialization and further analyse the success rates of micro-marsupialization in managing ranulas.

Keywords: *Ranula, Micro-marsupialization, Children, Case Report*

(1) Department of Paediatric Dentistry, Slim River Hospital, Perak

Corresponding Author

Dr. Adibah Jamil

Address

Jabatan Pergigian Pediatrik, Hospital Tengku Ampuan Rahimah, Klang

Telephone Number

None

Email

adibahjamil@gmail.com

DOI

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INTRODUCTION

Ranula is a pseudo-cystic lesion lined with granulation tissue instead of epithelium^{1,2}. It commonly arises from the floor of the mouth as a result of structural disturbances i.e trauma / obstruction / congenital malformations of the sublingual gland and /or its duct. This pathological disruption sets off mucinous extravasation of the gland onto surrounding tissue, giving rise to the distinctive fluctuant, frog-belly like appearance of ranula. They are generally asymptomatic and clinically may or may not present as a bluish fluctuant swelling at the floor of the mouth.^{3,4,5}

Depending upon herniation through mylohyoid muscle, ranulas can further be divided into superficial and plunging ranula⁶. The superficial intraoral ranula is limited to the floor of mouth and is commonly seen during the first and second decades of life^{1,7} however the plunging ranula presents itself as a submandibular mass that is herniated through or behind the mylohyoid muscle, potentially extending to the parapharyngeal and retropharyngeal spaces^{1,7,8} and rarely seen in children.^{7,9,10}

Regardless of its type, ranula poses a challenge to manage surgically due to its location and proximity to vital structures. Contemporary available treatment for ranula

includes surgical excision of the affected sublingual gland, marsupialization, micro-marsupialization, excision and drainage of the lesion, laser and sclerotherapy.¹¹ Despite widely reported, there's currently no acceptable guideline for management of ranulas, especially in children. Current research advocates a less invasive approach to manage this lesion in view of potential complications that may arise from removal of the salivary gland.^{11,12}

This paper aims to highlight a case of a superficial oral ranula in an 11-year-old that was successfully managed using micro-marsupialization and further analyse the success rates of micro-marsupialization in managing ranulas.

CASE REPORT

A fit and healthy 11 year-old boy came to us with the complaint of swelling over floor of mouth since a year ago. Swelling has been painless but recently is giving some discomfort. Patient claimed that swelling sometimes grew larger but always bursts spontaneously with yellowish, "salty" residue.

Upon examination, we noted a well-differentiated, solitary, semi-translucent swelling with smooth overlying mucosa over right floor of mouth measuring 8 x 4 x 2 mm that was fluctuant on palpation. No secondary changes such as ulceration, fistula formation, infection or discharge was noted and the tongue was not raised (Figure 1). The lymph nodes were not enlarged and there were no signs of cervical extension. A mandibular occlusal radiograph revealed no

evidence of sialoliths and on palpation it was noted there was no saliva draining from the sublingual area. Based on the clinical presentation, the lesion was diagnosed as a ranula.

Topical benzocaine 20% was given for 3 minutes before local anesthetic (Mepivacaine 2%) was administered surrounding the lesion. A slit was made on the roof of the lesion in anterior-posterior direction on its most fluctuant point. A blunt dissection with artery forcep was done to fully drain the mucinous content. The ensuing slit of the roof was sutured to the base of the lesion at its 4 corners using black silk 2.0 sutures (Vestisilk 2/0 Non-absorbable Black Silk, Reverse Cutting, 35mm 3/8c Worldwide Medivest) (Figure 2). Hemostasis was achieved with gauze pack compression for 2 minutes. A topical hyaluronic acid gel was applied onto surgical site to aid in healing and to increase patient's comfort.

The sutures were planned to be kept intact for 30 days but upon 1 weeks post operative review, 2 out of the 4 sutures were dislodged at home 5 days post-operatively. Upon 2 weeks post operative review, none of the sutures remained. The patient was reviewed for 1 week, 2 weeks, 1 month, 6 months and up till 1 year post-operatively. Clinically, the lesion was not visible after 2 weeks post-operatively except for a slight raise in the right floor of mouth measuring 2.5 mm x 1 mm that was only noted upon finger palpation. A period of active monitoring was employed and at the 6 months and 1 year review, the lesion showed a complete resolution with no sign of recurrence.

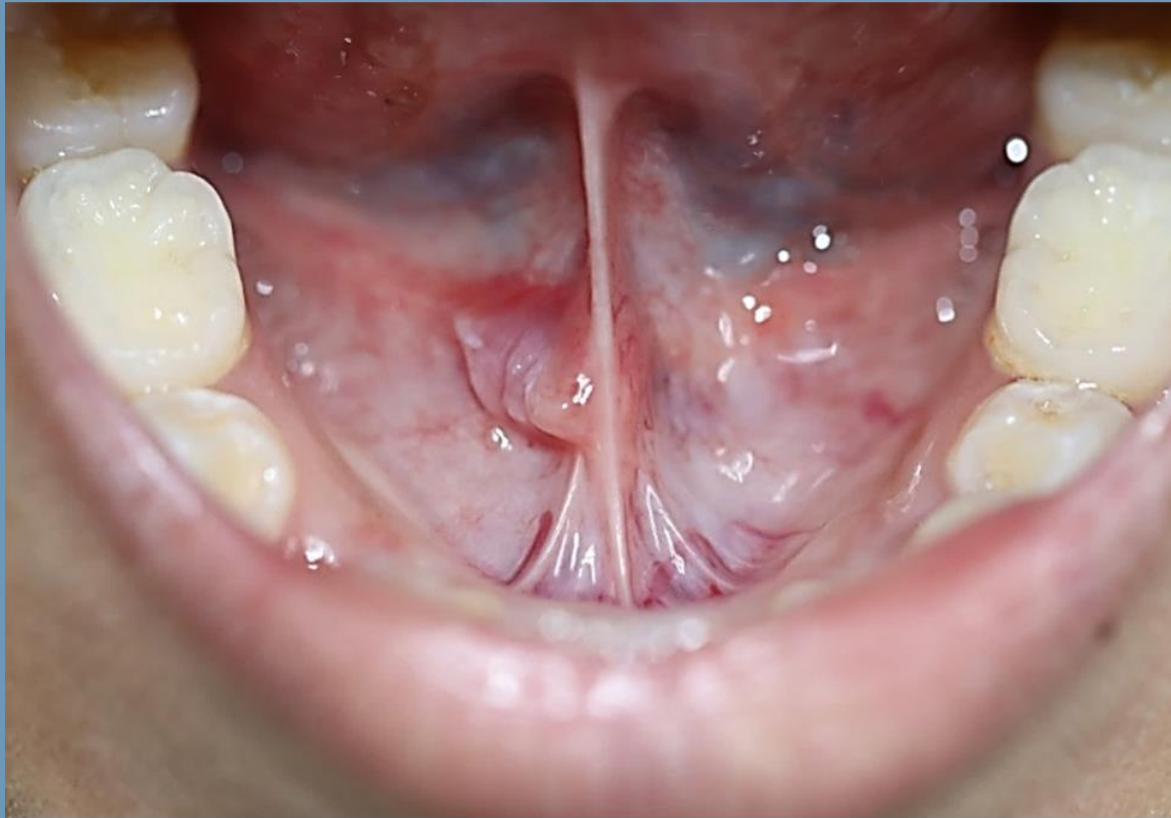


Figure 1: Initial clinical presentation



Figure 2: Intra-operative



Figure 3: 1 week post-operative



Figure 4: 6 months post-operative



Figure 2: 1 year post-operative

DISCUSSION

With incidence of 0.2 cases per 1000,¹³ ranulas account for approximately 6% of all oral sialocysts¹⁴ and arises from the sublingual salivary gland in 90% of the cases.¹³ Golden B, 2014 conducted an experimental study on the effect of salivary glands ducts ligation and finds out that ligation of sublingual salivary gland duct resulted in formation of ranula while ligation of submandibular salivary gland duct did not and ligation of parotid gland duct led to atrophy.¹⁵

The sublingual salivary gland lacks a true capsule and rather was enveloped by the mucosal fold of the floor of the mouth.¹⁶ Trauma to the sublingual salivary gland duct and/or its acini leads to ductal obstruction^{7,8,17} and consequently builds up increased hydrostatic pressure from the mucus extravasation¹⁸. Eventually, the acini may rupture, leading to the formation of pseudocyst from mucus escape reaction (MER)¹⁸. It may also develop due to chronic diseases of the sublingual gland¹⁷ or anatomical variation⁸.

Depending upon clinical extension of the lesion, ranulas are usually classified as either simple ranula (limited to the floor of mouth)

or plunging ranula (herniated through the mylohyoid muscle)¹⁹. The simple ranula are more commonly seen³ with a ratio of 10:1 to plunging ranula²⁰ and are also more commonly seen in paediatric population⁷ as has been corroborated in this case report. Regardless, any untreated ranula in paediatric patients under five years old can lead to obstructive sleep apnoea, and delay in seeking treatment can eventually lead to failure to thrive¹⁸.

Diagnosis of ranula is typically done via observation of its clinical characteristics. Imaging techniques such as ultrasonography (USG), Magnetic Resonance Imaging (MRI) or Computed Tomography (CT) are used mainly to assess the extension of swelling when the diagnosis is uncertain¹⁸. In a systematic review by Packiri et al done in 2017, it is recommended that MRI or CT scan is used when the lesion is more than 2 cm in diameter. In this case report, no USG, MRI or CT scan was done in view of limited logistics (closest MRI/CT Scan service was only available 1 hour away from current hospital) but a lower standard occlusal (LSO) radiograph was instead taken to ensure that no sialoliths were present in the lesion that might necessitate a different

management approach. As no evidence of sialoliths were present on the LSO and clinically there was no evidence that it could be a plunging ranula the decision was made to not send the patient for an USG.

Despite not routinely done (as was in this case), complementary cytochemical investigations like fine needle aspiration (FNAC) can be used to assist in definitive diagnosis. Chemical analysis of the viscous fluid content demonstrates increased amylase and protein content, numerous inflammatory cells and mucus; that is suggestive of salivary secretion¹⁸. FNAC is also beneficial in distinguishing ranulas from other oral or neck masses such as thyroglossal duct cyst, branchial cleft cyst, cystic hygroma, lymphatic malformation, intramuscular hemangioma, lymphangioma, abscess, or dermoid cyst²¹. In addition to FNAC, histopathological examination of ranula demonstrates presence of peripheral fibrosis capsule surrounding a central pool of mucin and mucinophages^{22,23}. Ranulas are thus considered as a pseudocyst and not a true cyst in view of the absence of an epithelial lining²⁴. In this case report, aspiration of the content was not done as consecutive loss of fluctuance may obviate the point for excision

Treatment of ranulas has long been a polarising debate between a conservative management with higher rate of recurrence or that of a more invasive, surgical methods but with a higher success rate. A middle ground treatment would be one that is fairly safe and easy to perform with a satisfactory success rate and is acceptable to patients. With this in mind, the primary aim in managing ranulas would be either to clear the mucinous blockage or to create an alternative pathway of drainage and maintain the drainage patency.

Total resection of the whole salivary gland has been advocated to prevent recurrence²⁵. In paediatric population however, total removal of sublingual salivary gland poses a risk to many critical, still developing

structures overlying the floor of the mouth such as the lingual nerve and the submandibular duct. There is also a risk of fistula formation². If the surgeon perforated the mylohyoid muscle, then the marginal mandibular and hypoglossal nerve is also at risk of injury. In addition, the long-term effect of early salivary gland removal in children has not been studied yet. In our view, total resection should be conserved for a more recalcitrant lesion and especially employed in that of plunging ranulas.

Creating an alternative drainage pathway through the pseudocyst wall can be done either by a physical approach i.e marsupialization, micro-marsupialization and the tunnelling method or through a chemical approach i.e., injection of a sclerotic substance such as OK-432^{10,17}. Although these methods do not have great predictability²⁰, good results have been documented for lesions smaller than 2 cm^{7,17,23}. Even with reported incidences of recurrences, in children however, the success rates of marsupialisation and micro-marsupialisation is significantly higher²⁶.

Morton and Bartley in 1995 proposed that oral ranula could be easily treated by the micro-marsupialization technique, which consists of placing a silk suture on the dome of the lesion along its widest diameter. This will drain accumulated saliva and creates a new epithelialized tract along the suture pathway. However, this method is reported to have high failure rates as lesions tend to reappear within 30 days²⁹. Therefore, a modified micro-marsupialization approach has been advocated consisting of an increase in the number of sutures that are closely placed and with longer period (minimum 30 days) of suture maintenance²⁹.

For this case report, we've utilised the modified micro marsupialization approach with a larger diameter needle (Black Silk 2.0). This creates a larger, alternative pathway of drainage and at the same time is well tolerated in children. The new drainage

patency is maintained by securing the sliced roof of lesion to the base at its 4 corners and left in situ for at least 2 weeks. This results in a total collapse of lesion while at the same time allows unhindered flow of trapped saliva onto the oral cavity. The sutures, however, were dislodged in 2 weeks. This is concurrent with the article by Piazzetta et al³⁰ where children had difficulty in maintaining the sutures due to discomfort and inadequate oral hygiene.

This modification resulted in a gradual size reduction of lesion within 2 weeks and a complete resolution in 1 months. We considered this case to be successful as there was no history of spontaneous pain or discomfort except during the first few days after treatment and there was no recurrence of the lesion after a year the procedure was done. Although this modification doesn't result in instantaneous resolution of lesion, it still achieves the treatment goal within an acceptable period of time. It also preserves many critical structures lying in the floor of the mouth by circumventing the need for a more aggressive and invasive surgical procedure.

The reduced invasiveness and simplicity of the method with minimal requirement for post-operative care enables this treatment option to be applied to the paediatric cohort. Limitations of this technique include its applicability to only small lesions (<2cm) and it does not enable a full biopsy examination.

CONCLUSION

This case was managed with the modified micro-marsupialization technique and no signs of recurrence were observed 12 months after surgery. This technique however does not enable a biopsy to be done thus making it an exclusively clinical diagnosis. However, the comparatively low invasiveness of the procedure and the minimal need for post-operative care makes this method a good treatment option for the management of ranulas, especially in children.

CONFLICT OF INTEREST

None

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Dentinogenic Ghost Cell Tumor: A Case Report In A Child

Geetha Siva Sankaran¹, Ganasalingam Sockalingam G¹

ABSTRACT

Dentinogenic Ghost Cell Tumor (DGCT) is defined by WHO as a locally invasive neoplasm. DGCT presents as intraosseous (central) and less commonly as extraosseous (peripheral) variants, with highest prevalence reported in the second and seventh decades. It occurs equally in the maxilla and mandible with no gender predilection. This is a case of a 10-year old female with clinical presentation of a painless, slow growing swelling at the left mandibular region of four months duration. Aspiration, incisional biopsy and marsupialization were performed in the district hospital and its histopathological examination revealed calcifying cystic odontogenic tumor. However, upon four months review, swelling was unresolved. The lesion was then enucleated, peripheral ostectomy and chemical curettage of lesion on the left mandible was performed under general anaesthesia. Histopathological examination revealed dentinogenic ghost cell tumor. The patient has remained disease free for 14 months postoperatively. The biologic behaviour of central DGCTs is considered to be more aggressive and requires radical treatment. However, in our case a more conservative approach was taken as patient is a growing child.

Keywords: *Dentonogenesis ghost cell tumour, children*

INTRODUCTION

Dentinogenic ghost cell tumor (DGCT), also known as odontogenic ghost cell tumor is considered a rare neoplastic variant of COC. Based on Buchner's research, it was found that COC represents 1-2% of all odontogenic tumors and of this only 2-14% were solid tumors, considered to be DGCTs¹.

DGCT are of two types, extra osseous (peripheral) and intraosseous (central) type. Intraosseous DGCT are more aggressive,

(1) Department of Pediatric Dentistry, Tunku Azizah Hospital, Kementerian Kesihatan Malaysia, Kuala Lumpur.

Corresponding Author

Dr. Geetha Siva Sankaran

Address

Department of Paediatric Dentistry, Tunku Azizah Hospital, Kuala Lumpur.

Telephone Number

010-4646462

Email

sgeetha88@live.com

DOI

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have an infiltrative growth pattern and a high recurrence rate after resection. Therefore, intraosseous DGCT should be treated by extensive surgical resection with an adequate safety margin as compared to extraosseous variant.

Different terminologies and classifications of Calcifying Odontogenic Cyst (COC) have been suggested and practiced in the literature since 1962 to date. Fejerskov and Krogh in 1972 were of the opinion that the term COC is not entirely appropriate, because there could be a possibility of cystic

degeneration taking place in the centre of the proliferating epithelial islands rather than epithelial changes developing in a pre-existing cyst wall². They have also stated that the presence of ghost cells which may subsequently show calcification and the proliferative potential of some lesions giving rise to lesions of considerable size.

In 2005, the WHO classified COC as an odontogenic, benign tumor rather than a cyst, re-naming this entity as calcifying cystic odontogenic tumor (CCOT). WHO in 2005 defined DGCT as 'A locally invasive neoplasm characterized by ameloblastoma-like islands of epithelial cells in a mature connective tissue stroma. Aberrant keratinization may be found in the form of ghost cells in association with varying amounts of dysplastic dentin'³.

CASE PRESENTATION

A 10 year old female, with history of childhood asthma, first presented to a district hospital with a painless, slow growing swelling at the left mandibular region of four months duration. The swelling was not associated with any pain, discomfort, functional disability or disturbance of her psychological well-being. Radiographically, the lesion appeared ill-defined, multilocular with non-corticated borders extending from the midline of the left mandible to the mesial of tooth 36 with severely displaced tooth 33 and 34 (Figure A).

Aspiration, incisional biopsy and marsupialization were performed and its histopathological examination revealed

calcifying cystic odontogenic tumor. Marsupialization was the initial treatment of choice because it relieves the pressure within the cavity, inducing bone formation and reducing the risk of mandibular fracture and inferior alveolar nerve damage. Marsupialization can be used as the only sole treatment for a cyst or as a preliminary step in management, with enucleation deferred until later.

Upon four months, patient referred to us complaining of unresolved swelling.

Clinical examination revealed facial asymmetry with a noticeable bulge at the left mandible. No cervical lymphadenopathy was noted. Intraorally, a swelling was noted buccally extending from distal of 31 till the mid portion of 36 and lingually at the lower left premolars region which does not interfere with occlusion (Figure B). The deciduous teeth 73,74 and 75 were retained with clinically missing 33, 34 and 35. Other permanent teeth were fully erupted. Upon palpation, the egg crackling features could be felt at the buccal expansion site close to the retained deciduous teeth. The overlying mucosa of the crepitus area appears reddish in color.

OPG and CT scan revealed a multilocular mixed radiopaque and radiolucent lesion, extending from the midline of the mandible to the mesial of 36, associated with severely displaced 33 and 34. Radiopaque masses were noted in close proximity to the roots of tooth 33 and 34. The lower border of mandible and lingual cortical plate were thin but intact.



Figure A OPG revealed a multilocular mixed radiopaque and radiolucent lesion.



Figure B Clinically shows a swelling buccally from distal of 31 till the mid portion of 36.



Figure C Intraoperative features; Cyst lining separated from the buccal bone at the left mandible.



Figure D Post-operative orthopantomogram view.

Enucleation, peripheral ostectomy and chemical curettage of lesion on the left mandible was performed under general anaesthesia (Figure C). The lesion measuring 4.0 x 3.5 x 1.8cm was removed. Histopathological examination revealed cystic lining lined by stratified squamous epithelium of varying thickness and palisaded basal cell layer. Numerous ghost cells keratinisations are present in superficial epithelium. Occasional dentinoid material and epithelial islands with ghost cells were seen in fibrous connective tissue wall. Dystrophic calcifications were observed in

areas. The lesion was diagnosed histologically as a Dentinogenic Ghost Cell Tumor of the left mandible based on the presence of dentinoid material and epithelial islands with ghost cells seen in the fibrous connective tissue. The lesion appeared to be completely removed. At 14 months postoperatively, there were no signs of local recurrence (Figure D).

DISCUSSION

DGCT is a solid variant of CCOT. It is an uncommon odontogenic neoplasm and can

be seen at any age with the highest prevalence at the second and seventh decades with no gender predilection and no preference for the maxilla and mandible. However, Shah et al found that this lesion occurs more commonly in males than in females⁴.

Intraosseous lesions have been reported to occur predominantly in canine to first molar region. De Arruda et al who reviewed 55 cases of DGCTs, observed that mandible is the most common site of involvement⁵. The present case was noted at the mandible between the lateral incisor and first molar in a 10-year-old female patient, which is a comparatively younger age than the average age as reported by Candido who found for DGCT with an average of 62years⁶.

The size of intra-osseous DGCT varies from 1 to more than 10cm in diameter and is usually asymptomatic. The clinical signs of intraosseous DGCT variants may include expansion of the jaw, clinically visible swelling and obliteration of the maxillary sinus and infiltration of the soft tissues. Swelling can be painful or painless and occasionally accompanied by pus discharge, tooth displacement or mobility. Buchner et al reported 45 cases and found that the lesion presents with dull, slight or mild pain in 52% of the cases⁷. The reported case presented with a diffuse extraoral bony hard swelling which was uniform throughout.

Radiographic examinations of this case revealed a multilocular, mixed radiopaque and radiolucent lesion, at left mandibular area displacing teeth 33, 34 and 35. This was in accordance with Konstankis et al in 2013, which states that DGCT may appear radiographically as radiolucent, radiopaque or mixed lesion, and lesions can be unilocular or multilocular with either well-defined or ill-defined margins⁸.

DGCTs can clinically and radiographically resemble other odontogenic tumors. A characteristic feature of DGCT is ghost cells. However, the presence of ghost cells alone is not pathognomic of DGCT (Figure E and F), since they can be identified in other neoplasms such as odontomas, ameloblastomas and ameloblastic fibroodontomas. Thus, it is important to understand the pathological and radiological features of DGCTs to differentiate from other lesions. Histopathologically, two characteristics features of DGCTs distinguish them from other odontogenic tumors are masses of keratinizing ghost cells and dysplastic dentin. In the current case, the microscopic features showed cystic lining lined by stratified squamous epithelium of varying thickness and palisaded basal cell layer. Numerous ghost cell keratinisations are noted. Occasional dentinoid material and epithelial islands with ghost cells were seen in the fibrous connective tissue. These histopathologic features are compatible with the diagnosis of a DGCT.

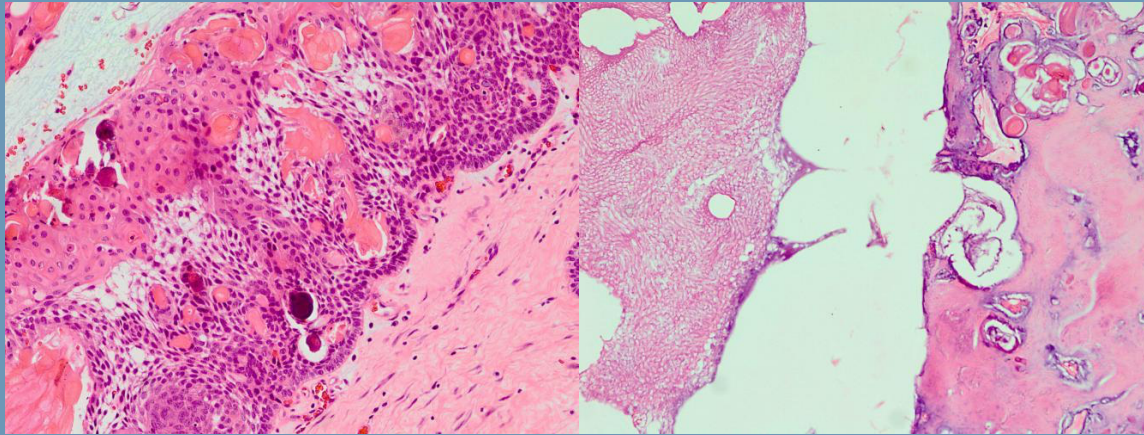


Figure E Odontogenic epithelial lining having palisaded basal cell layer with clusters of ghost cells and occasional calcification. (20x mag)

Figure F Enamel matrix, dentinoid and cementoid material together with clusters of ghost cells (Decalcified section, 10x)

Enucleation, peripheral ostectomy and chemical curettage was performed under general anaesthesia. Marsupialization may be an option, however, immediate enucleation should be considered due to its local invasive nature of the tumor. Enucleation is the surgical treatment that involves the complete removal of the entity. Physical or chemical curettage can be added to the enucleation procedure that require additional removal of surrounding bone to help ensure complete removal and decrease the persistence of the lesion. In this case, the addition of Carnoy's solution to curettage and peripheral ostectomy has been shown to be more effective in decreasing the recurrence rate than are the enucleation procedures alone. The patient is being followed up for a one year and has shown no signs of recurrence. Garcia et al and Sun et al central DGCTs have reported that DGCTs to have a high recurrence rate⁹. Hence, it is strongly recommended to keep patients under long term follow-up.

CONCLUSION

In this article, we reported a case of a 10-year-old female, who presented with a slow growing, painless swelling at the left mandibular region for four months prior that was initially treated with marsupialization. To arrive at a definitive diagnosis, routine histopathological examination was carried out and demonstrated the characteristics features of DGCT. When lesions shows large dimensions, as seen in the present case, a two stage approach allows important structures to be preserved. Marsupialization prior to enucleation proved to be favourable and effective treatment of choice in pediatric patients. However, owing to the aggressive nature and high rate of recurrence of DGCT, it should be treated with a more radical approach. Therefore, we believe that the choice of this surgical method is a more rational approach to reduce the recurrence. Long term patient follow up is mandatory as recurrences over 5-8 years following primary

treatment have been reported irrespective of the mode of surgical treatment.

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CONFLICT OF INTEREST

The authors have no conflict of interest to declare.

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The patient provided informed consent.

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