Assessment of Dental Development in Children with Hypodontia and Hyperdontia: A Case Control Study

Yunita Dewi Ardini[1], Noraini Abu Bakar[2], Nabila Farhana Kamalul'lail[3], Noor Syazwani Mohd Rasdi[3].
[1] Paediatric Dentistry Unit, Kulliyyah of Dentistry, International Islamic University Malaysia Kuantan
[2] Unit of Orthodontics, Kulliyyah of Dentistry, International Islamic University Malaysia Kuantan
[3] Kulliyyah of Dentistry, International Islamic University Malaysia Kuantan

ABSTRACT

The aim is to investigate the dental development (DD) in children with hypodontia and hyperdontia compared to age, gender, and race matched controls.A match-pair case control study was conducted among children aged 5 to 14-year-old, attending the Dental Polyclinic, IIUM, from December 2011 until September 2014. Willem's method was used to analyze the dental age (DA) using panoramic radiograph. The difference between dental age of hypodontia/hyperdontia children and the controlswas analyzed using analysis of covariance (ANCOVA). The differences between dental and chronological ages (CA) and the differences in DA and CA with the numbers of missing teeth were analyzed using t-test. Intra-class correlation coefficient between examiners ranged from 0.98-0.99. The result is that27 hypodontia and 12 hyperdontia samples were recruited and compared to a total of 78 matched controls. Both male (n=12) and female (n=15) hypodontia children had statistically significant delay in DD (p=0.000). There was no significant difference in the DD of male (n=7, p=0.811) and female (n=5, p=0.235) in hyperdontia children compared to the matched controls. No differences were observed between DA and CA with the number of missing teeth. Unlike hyperdontia children, hypodontia children showed delayed DD in comparison to age, gender, and race matched controls. There was no association between magnitudes of dental development delay in hypodontia children with the number of missing teeth. This study provides valuable baseline information for provision of better treatment planning for those hypodontia and hyperdontia children that may involve inter-disciplinary management.

KEYWORDS: dental development, hypodontia, hyperdontia

INTRODUCTION

Dental development is a process of tooth formation, beginning during the fifth week of embryonic life until the roots of the third permanent molars are completed, at about the age of 20 years.[1] Many growth and transcription factors are involved in order to ensure that all the teeth develop in an ordered and controlled process. It is crucial to know the normal sequence of dental development, in order to provide good treatment plan for patients and for age estimation in forensic odontology. Mitchell L.[2] clarified that calcification times in the permanent dentition should be known in assessing the dental age, which can be compared to the chronological age of a patient, to determine whether a missing developing tooth on a radiograph can be considered as absence. Karp J.M.[3] stated that chronological age ranges are mostly used to present the possible time of dental development. Besides that, dental development also shows less variability with chronological age compared to any other developmental features. Small variation in tooth formation and eruption among persons has made dental estimation of chronological age the primary method of age determination for younger persons.[4] Thus, dental age assessment by various methods has been developed to estimate the age based on different dental development phases.[5] Willem's method was chosen in this research as it has been proven to be more realiable in estimating dental age for Malaysian children.[6]

Both hypodontia and hyperdontia are examples of anomalies of tooth formation and eruption. Hypodontia is the term most often applied to a situation where a patient has missing teeth as a result of failure of development.[1] Other terms often used are congenitally missing teeth, agenesis of teeth and also lack of teeth. On the other hand, hyperdontia is defined as presence of any tooth or tooth substance in excess of normal dental formula.[7] Morphologically, it is classified into supplemental which resembles a tooth and occur at the end of tooth series, conical or peg-shaped which often occur between upper central incisors, tuberculate which does not fall into conical or supplemental categories and lastly odontome which is rare. By position, it is divided into mesiodens, distomolar, paramolar and parapremolar.

The aetiology of hypodontia and hyperdontia can be

Corresponding author:
Yunita Dewi Ardini
Paediatric Dentistry Unit,
Faculty of Dentistry
International Islamic University Malaysia
Jalan Sultan Ahmad Shah, 25200 Kuantan, Pahang
Email: dryunita@iium.edu.my

multifactorial including environmental and genetic factors. In the case of hypodontia, it can be due to changes of dental lamina formation, failure of a tooth germ to develop at optimal time, space limitation, systemic conditions and genetic factors. However, in hyperdontia, there is a familial tendency. Systemic conditions such as cleidocranial dysplasia, Gardner's syndrome and cleft lip and palate has also been found to be associated with hyperdontia.[8] Thus, both hypodontia and hyperdontia can also be classified as syndromic or non-syndromic cases where non-syndromic cases present without any systemic disorders or syndromes.[9]

Morphologic, radiographic, histologic and biochemical methods have been developed to estimate age based on dental tissue and tooth morphology. Demirjian's method, revisited by Willems G. *et al.*[10] for age estimation using panoramic radiographs involved observation of the morphologically different stages of mineralization. It simplified dental age estimation and restricted the number of stages of tooth development to 8 (Figure 1) giving them a score from A to H (Table2 and 3). This method confines the analysis to the first seven teeth of the left lower quadrant.

The main purpose of this study is to observe the dental development in patients with hypodontia and hyperdontia in terms of dental age and chronological age of permanent teeth, whether it is delayed, normal or premature compared to their respective control groups.

To date, to the best of our knowledge, there are no studies that look at the dental development rate in both hypodontia and hyperdontia, and investigating it's relation to the amount of missing teeth in Malaysian population.

MATERIALS AND METHOD

Study Design
A match-pair case control study.

Study Population
Total paediatric patients attending Dental Polyclinic, Dental Faculty, International Islamic University Malaysia (IIUM) Kuantan.

Sample Population
Paediatric patients, age between 5-14-year-old attending Dental Polyclinic of Dental Faculty, IIUM from December 2011 toSeptember 2014, and requiringdental panoramic radiograph as part of the treatment planning.

Sample Unit
Inclusion criteria:

- Malay ethnicity.
- Medically fit.
- Non-syndromic.
- Patient whom require panoramic radiograph for treatment planning.

Exclusion criteria:

- Patients on long-term medication (with chronic diseases).
- Patients with history of tooth loss due to trauma.
- Patients with missing teeth in both lower right and left quadrant.
- Hypodontia children with missing third molar only.
- Low quality panoramic radiographs.

Data Collection and Data Analysis
Data collection began on March 2014 upon receiving the ethical approval from IIUM Research Ethics Committee (IREC). Children with either hypodontia or hyperdontia were identified through screening of existing panoramic radiographs (via Planmeca Romexis Software Database) of patients who have had treatment in IIUM Polyclinicfrom December 2011 to September 2014.These radiographs were taken with Orthopantomogram (OPG) machine, Planmeca ProMax 3D, which provide a resolution of 3 lp/mm image. The exposure time was 12 seconds with a magnification factor of 1.45. Total of 1371 dental panoramic radiographs were screened. Radiographs with low quality images were rejected.

In this study, a tooth was diagnosed as congenitally missing (hypodontia) when no mineralization of its crown could be identified on the panoramic radiographs, with no evidence of it had been extracted, excluding the third molar. Hyperdontia children were identified when there was extra number of tooth/teeth seen from the radiographs, exceeding the normal dental formula. Next, the archived case sheets of these children were collected to analyze the medical history and dental history in order to match the inclusion and exclusion criteria. 27 hypodontia and 12 hyperdontia samples were recruited. This sample was matched for race, age, and sex with 78 healthy, normal control subjects. Two controls were used for each hypodontia/hyperdontia sample.

The chronological age for both groups was calculated by subtracting the date of birth from the date of the radiograph taken which specified the actual age of these children when the radiograph was taken. The age was then converted into years with one decimal point. The chronological age for matched control group was carefully selected to the accuracy of ±1month as compared to their respective hypodontia/hyperdontia children.

Radiographic assessment of dental development wasthen performed for both hypodontia/ hyperdontia groups and the control group. Willem's method was used to analyze the dental age of the first seven permanent teeth on the left side of the mandible as shown on the panoramic radiograph. Scoring was done on each of the developing tooth referring to the written criteria described by Demirjian *et.al.*[11] for each stage and by comparing the tooth with the diagram and radiographic image. The written criteria is shown in Table1 and the

sample of the radiographic images in every stage was shown in Figure 1. The developmental stage was then converted into score by referring to Willems' Table (Demirjian Revisited) according to the genders (refer Table 2 for males and Table 3 for females). The total scores for the seven teeth will represent the dental age of the child.

The chronological age and dental age of the hypodontia/hyperdontia children and their controls were recorded in Microsoft Excel. Data obtained was analyzed using SPSS software version 16.0. Since the accuracy of the chronological age of matched control was ± 1 month, Ancova was used to analyze the difference between dental age of hypodontia/hyperdontia children and the control group, adjusting for the differences in chronological age of the hypodontia/hyperdontia children. Independent Sample t-test was also used in order to determine the mean difference between dental age and chronological age (mean DA-CA). The positive value of mean DA-CA shows no delay in dental development whereas a negative value will be interpreted as there is a delay dental development in the group.

Calibration of the first and second examiners was assessed twice. A sample of 20 panoramic radiographs, which were not part of this study were examined one-week apart. The reliability between examiners were checked using Intraclass Correlation Coefficient where the Intraclass value between the first examiner and second examiner was 0.984, whereas the Intraclass value between the first examiner and the third examiner was 0.990 and the Intraclass value between the second examiner and the third examiner was 0.992. These showed an excellent agreement and consistencies between the three examiners.

Table 1: Criteria of each dental developmental stage

TDS	Single Rooted Teeth and Multi-Rooted Teeth (Descriptions)
A	In both uniradicular and multiradicular teeth, a beginning of calcification is seen at the superior level of the crypt in the form of an inverted cone or cones. There is no fusion of these calcified points.
B	Fusion of the calcified points forms one or several cusps, which unite to give a regularly outlined occlusal surface.
C	Enamel formation is complete at the occlusal surface. Its extension and convergence toward the cervical region is seen. The beginning of the dentine deposit is seen The outline of the pulp shape has a curved shape at the occlusal border.
D	Crown formation is complete down to the cemento-enamel junction. The superior border of the pulp chamber in uniradicular teeth has a defined curved form, being concave towards the cervical region. The projection of the pulp horns, if present, gives an outline like an umbrella top. In molars, the pulp chamber has a trapezoid form. Beginning of root formation is seen in the form of a radiopaque spicule.
E	UNIRADICULAR TEETH The walls of the pulp chamber now form straight lines, whose continuity is broken by the presence of the pulp horn, which is larger than in the previous stage. The root development is still less than the crown. MULTIRADICULAR TEETH Initial formation of the radicular bifurcation is seen in the form of either a calcified point or a semilunar shape. The root length is still less than the crown height.
F	UNIRADICULAR TEETH The walls of the pulp chamber now form a more or less isosceles triangle. The apex ends in a funnel shape. Root development is equal to or greater than the crown height. MULTIRADICULAR TEETH The calcified region of the bifurcation has developed further down from its semilunar stage to give the roots a more definite and distinct outline, with funnel shaped endings The root length is equal to or greater than the crown height.
G	The walls of the root canals are now parallel(distal root of molars) The apical ends of the root canals are still partially open.
H	The apical end of the root canal is completely closed (distal root in molars) The periodontal membrane has a uniform width around the root and apex.

Table 2: Scores according to dental developmental stages in males

TOOTH	A	B	C	D	E	F	G	H
CENTRAL INCISOR	-	-	1.68	1.49	1.5	1.86	2.07	2.19
LATERAL INCISOR	-	-	0.55	0.63	0.74	1.08	1.32	1.64
CANINE	-	-	-	0.04	0.31	0.47	1.09	1.9
FIRST PREMOLAR	0.15	0.56	0.75	1.11	1.48	2.03	2.43	2.83
SECOND PREMOLAR	0.08	0.05	0.12	0.27	0.33	0.45	0.4	1.15
FIRST MOLAR	-	-	-	0.69	1.14	1.6	1.95	2.15
SECOND MOLAR	0.18	0.48	0.71	0.8	1.31	2	2.48	4.17

Table 3: Scores according to dental developmental stages for females

TOOTH	A	B	C	D	E	F	G	H
CENTRAL INCISOR	-	-	1.83	2.19	2.34	2.82	3.19	3.14
LATERAL INCISOR	-	-	-	0.29	0.32	0.49	-0.79	0.7
CANINE	-	-	0.6	0.54	0.62	1.08	1.72	2
FIRST PREMOLAR	-0.95	-0.15	0.16	0.41	0.6	1.27	1.58	2.19
SECOND PREMOLAR	-0.19	0.01	0.27	0.17	0.35	0.35	0.55	1.51
FIRST MOLAR	-	-	-	0.62	0.9	1.56	1.82	2.21
SECOND MOLAR	0.14	0.11	0.21	0.32	0.66	1.28	2.09	4.04

RESULTS

A total of 1371 dental panoramic radiographs were screened from Planmeca Romexis Database where 27hypodontia children (12 males, 15 females) and 12hyperdontia children(7 males, 5 females) were found suitable with the inclusion and exclusion criteria of this study. A total of 78 age, gender, and race matched controls were recruited for this research where 2 controls were used to be compare to every hypodontia/ hyperdontia child. There were more samples recruited in hypodontia due to the fact that it was more common than hyperdontia.

Intra examiner consistency

Intra class correlation coefficient between three examiners ranged from 0.98-0.99. These showed an excellent agreement and consistencies between the three examiners.

Comparison of mean dental age for hypodontia and hyperdontia children compared to control group.

The Ancovastatistical results were shown in Table 4 for hypodontia cases and Table 5 for hyperdontia cases, comparing the mean dental age with their respective control groups. The mean dental age for hypodontia children were 10.3±2.2 years (females) and 8.9±2.4 years (males), both lower than their respective control group; 11.7±2.6for females and 9.9±2.7 years for males. These results were statistically significant with p=0.000 for both genders.

Contrarily, the mean dental age was lower in hyperdontia children which were 10.1±3 (females) and 10.4±2.9 (males) as compared to their controls; 10.9±2.7 years (females) and 10.5±2.3 years (males) but the results were not statistically significant with p=0.233 and p=0.815 respectively.

Table 4: Mean Dental Age of Hypodontia Children compared to control group using Ancova Statistical Analysis.

	Gender	n	Mean dental age(years)	Standard deviation (years)	Sig
Male	Samples	12	8.9	2.4	0.000
	Controls	24	9.9	2.7	
Female	Samples	15	10.3	2.2	0.000
	Controls	30	11.7	2.6	

Table 5: Mean Dental Age of Hyperdontia Children compared to control group using Ancova Statistical Analysis.

	Gender	n	Mean dental age (years)	Standard deviation (years)	Sig
Male	Samples	7	10.4	2.9	0.815
	Controls	14	10.5	2.3	
Female	Samples	5	10.1	3.0	0.233
	Controls	10	10.9	2.7	

The mean difference between dental age and chronological age (DA-CA) for hypodontia and hyperdontia groups compared to control group.

The mean difference between dental age and chronological age (DA-CA) for hypodontia and hyperdontia groups with their respective control groups were calculated using Independent Sample t-test and the results were shown in Table 6 and Table 7.

Both males and females hypodontia children had delayed dental development as denoted by a negative mean difference of DA-CA. The mean differences between DA and CA in male and female hypodontia children were -0.15 year and -0.71 year respectively, compared to 0.48 year and 0.74 year for the controls. These results were found to be statistically significant, where p=0.000 for both genders.

However, there was statistically no delay in dental development for hyperdontia children in males, (p=0.811), where the mean difference was almost similar which were 0.42 year for hyperdontia children and 0.49 year for their controls. There was delay dental development in female hyperdontia which were -0.15 year compared to control 0.25 year. However this result was not statistically significant with p=0.235.

Table 6: Mean Difference of Dental Age and Chronological Age of Hypodontia Children compared to control group using Independent Sample T-test.

	MEAN DIFFERENCE OF DENTAL AGE AND CHRONOLOGICAL AGE (DA-CA)			
	MALE (year)		FEMALE (year)	
SAMPLES	-0.15 (n=12)	p=0.000	-0.71 (n=15)	p=0.000
CONTROLS	0.48 (n=24)		0.74 (n=30)	

Table 7: Mean Difference of Dental Age and Chronological Age of Hyperdontia Children compared to control group using Independent Sample T-test.

	MEAN DIFFERENCE OF DENTAL AGE AND CHRONOLOGICAL AGE (DA-CA)			
	MALE (year)		FEMALE (year)	
SAMPLES	0.42 (n=7)	p=0.811	-0.15 (n=5)	p=0.235
CONTROLS	0.49 (n=14)		0.25 (n=10)	

Table 8: Mean Difference of Dental Age and Chronological Age (Da-Ca) in Children with one Missing Tooth versus Multiple Missing Teeth

		HYPODONTIA CHILDREN WITH ONEMISSING TOOTH	HYPODONTIA CHILDREN WITH MUL-TIPLE MISSING TEETH	P VALUE
SAMPLES	n DA-CA	18 -0.55	9 -0.76	p=0.000
CONTROLS	n DA-CA	36 0.70	18 0.47	p=0.000

Comparison of dental age and chronological age (DA-CA) in single missing tooth versus multiple missing teeth in hypodontia cases compared to control group.

Table 8 compared the means DA-CA between children with a single missing tooth and those with multiple missing teeth in relation to case controls. There were significant difference in both single and multiple missing teeth with p=0.00, where single missing tooth showed -0.55 years in hypodontia children and 0.7 year in controls with the delay of 1.25 years, whereas multiple missing teeth showed -0.76 year in hypodontia children and 0.47 year in controls with the delayof 1.23 years. This showed that both single and multiple missing teeth had equally significant delay in dental development.

DISCUSSION

This study showed that hypodontia children had statistically significant delay in dental development for both genders as compared to the control group. This result was consistent with the studies conducted by Uslenghi S. *et.al.*[12], Kan *et.al.*[13], Tunc E.S. *et.al.*[14] and Ruiz-Mealin E.V. et al.[15] However, studies conducted by Bailitetal.[16] and Odagami *et al.*[17] showed a different outcome wherei the hypodontia children had similar rate of dental development as in control group.

This research also revealedno significant delay in dental development of hyperdontia children as compared to their control group, suggesting that hyperdontia was not associated with general growth changes affecting dental development. The result was supported by the study previously conducted by Kan *et.al.*[13]

Another finding is that there was no association between magnitudes of dental development delay in hypodontia children withthe number of missing teeth. This result was similar to Rune B. &Sarnas K.[18] In contrast, Odagami *et.al.*,[17] and Uslenghi S. *et.al.*,[12] found that there was a relation between severity of missing teeth and developmental timing.

Limitation of this study

There are several limitations of this study. The sample size was small as it was restricted to patients attending dental polyclinic IIUM that has operated for less than 10 years. As hypodontia and hyperdontia are congenital in nature, both are considered rare and deviated from normal population. Hence, restriction of sample origin will reduce the sample size, which may affect the findings and generalizability of the study. Secondly, despite the widely used of Willem's method in dental age assessment, it had one crucialdisadvantage.Hypodontia children with bilateral missing teeth on lower arch must be excluded.This method had proposed that the dental age assessment was to be made on the first seven permanent teeth of lower left quadrant. As stated by Willems G.[10] only one side of mandible was used for this technique as there was a high degree of symmetry between right and left sides. If the missing tooth occurred on the left side, the right side could be used as reference. Therefore, cases of bilateral hypodontia of the mandible have to be excluded in this study, even though there were quite a number of such cases encountered during data collection.

Clinical significant of the finding of this study

Early detection of hypodontia and hyperdontia gives significant clinical implications in terms of dental management. Multidisciplinary consultation involving Paediatric, Orthodontic, Restorative, Prosthodontic and Oral Surgery departments during treatment planning with coordination and appropriate timing of subsequent dental care enables clinician to provide the optimum management for hypodontia and hyperdontia patients.

Hypodontia children

Orthodontic treatment and implants placement are amongst popular treatment option for patients with hypodontia. Appropriate timing is the key for long-term success and stability of the treatment outcome. Study has showed that implants placed before cessation of facial development tend to submerge relative to the adjacent erupting teeth.[13] Implants inserted during late puberty or early adulthood provide better long-term stability compared to implants placed before or during early puberty.[17] Using the finding in this research, the timing of such treatment in hypodontia children may need to be delayed by up to 1 year for boys and 1.4 years for girls compared to treatment for a normal child.

Hyperdontia children

According to this study, there was no significant delay in dental development as compared to normal child. Therefore, this can be taken into consideration during treatment planning and consultation to patients and parents.

Islamic perspective of this study

This study showed evidence of the greatness of Allah SWT)'s creation. The whole embryological and developmental process must be spot on for most of us to be normal. As the ayah in surah Az-Zumar"*He creates you in the wombs of your mothers in stages, one after another, in three veils of darkness*." (TMQ 39:06).The summary of Ibn Kathir's interpretation (may Allah have mercy on him) of this ayah is as follows: It means, Allah (SWT) preordained you in your mothers' wombs (in stages, one after another); each of you is first a *nutfa*(gamete: reproductive cell, whether sperm or ovum), then it develops into a '*alaqa* (leech-like clot), then into *amudgha* (the form of a chewed-up morsel) and then he is shaped

and formed with the development of bones, flesh, nerves and veins. Afterwards, the soul is blown into him and he becomes a different creation. (He is the Unique Creator.

Embryological process is complex and complicated. Million interactions and process that happen in the womb,in precise order , prove to us the glory of Allah (SWT) as mentioned in Surah *al-A'la* (87.1-87.3)*"Exalt the name of your Lord, the most high. Who create and proportioned and who destined and then guided."*

In dentistry, the tooth bud of the primary teeth developed in the first few weeks of intrauterine life, and for permanent teeth it developed much later. Lacking of this process will cause hypodontia and exaggeration of this process will cause hyperdontia. This display a small example ofhow precise human creation is and how we should be forever thankful to our Creator.

In term of management, having this knowledge of dental development in hypodontia and hyperdontia will give us, the clinician, a useful insight on how to better treat these cases. This will in return enhanced us in fulfilling our *amanah*towards the patients and promote professionalism. As in the case of all other concepts in Islam, professionalism inIslam is rooted and based upon the two basic and primary sources of reference in Islam, namely the Quran and the Hadith. The need to execute our duty in the whole submission and obedience to the almighty God.

CONCLUSION

These conclusions can be drawn based on this study's findings:

1. Hypodontia children have statistically significant delayed dental development in comparison to age, gender, and race matched controls.
2. Non-syndromic hyperdontia children exhibit similar dental development rate as compared to normal matched controls.
3. There was no association between magnitudes of dental development delay in hypodontia children with the number of missing teeth.

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