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Double primary teeth and their relationship with the permanent successors: a radiographic study of 376 cases

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Abstract

Double primary teeth were observed in 367 children and their relationship with the permanent successors was studied with the aid of radiographs. Double primary teeth involving 2 adjacent teeth and those involving the same tooth have different patterns of association with the permanent successors. Double primary teeth involving 2 adjacent teeth are associated more frequently with congenitally missing permanent successors. A combination of factors such as decreased hereditary dependent mitotic potential of the tooth germ and presence of an inhibitory zone around the tooth germ might have been responsible for the formation of these anomalies. It is suggested that double primary teeth probably result from unseparated rather than fused tooth masses.

Double tooth is the term frequently used to describe the anomaly of conjoined teeth.[1] Other terms, such as fusion (Livitas 1965; Mader 1979) gemination (Levitas 1965; Mader 1981) connation (Hutchin and Morris 1966) linking tooth (Sprinz 1953) synodontia and schizodontia (DeJonge 1955) also have been suggested. In this article, the term double tooth is used because in many instances, it is clinically difficult to decide whether fusion or gemination has occurred.[2]

The anomaly occurs more frequently in the primary than in the permanent dentition and has been reported to be more prevalent in Mongoloid (3%: Saito 1959; Niswander and Sujaku 1963) than Caucasian (1.6%)[3] populations. No preference of sex has been apparent.

Although double teeth have been reported in premolar and permanent molar areas, the condition is seen predominantly in the incisor and canine regions. Double primary molars only have been reported as anomalies in the otodental syndrome (Winter 1983). Double teeth can occur unilaterally or bilaterally in either the maxillary or mandibular arch. The simultaneous bilateral occurrence in both arches has not been reported.

The association of double primary teeth with variation in tooth number, both in the primary and permanent dentitions, has been documented.[4] In general, congenital absence of a permanent successor often is associated with a double primary tooth if the normal number of teeth exists when the double tooth is counted as 2 teeth (Grahnen and Granath 1961; Gellin 1984). On the other hand, a supernumerary tooth is sometimes present if hyperdontia exists when the double tooth is considered as 2 teeth (Brook and Winter 1970; Ravn 1971).

Although esthetic and functional problems created by double primary teeth are relatively transient in the primary dentition, proper monitoring of occlusal development should not be overlooked to prevent deviation of the midline and abnormal delay in eruption of the permanent successors. In cases where permanent successors are congenitally absent or in double tooth formation, esthetic and functional problems in the permanent dentition should be anticipated. Extensive carious involvement on the labial and lingual vertical grooves of the double primary tooth often requires early treatment (Lowell and Solomon 1964).

The purpose of this study was to compare the distribution of the different types of double primary teeth and their relationship to the permanent suc-

[1] Moody and Montgomery 1934; Miles 1954; Grahnen and Granath 1961; Brook and Winter 1970; Itkin and Barr 1975; Weiss 1980; Gellin 1984.
[2] Miles 1954; Brook and Winter 1970; Itkin and Barr 1975.
[3] Grahnen and Granath 1961; Clayton 1956; Ravn 1971.
[4] Grahnen and Granath 1961; Brook and Winter 1970; Gellin 1984; Ravn 1971.

TABLE 1. Distribution of Maxillary Double Primary Teeth as Related to the Permanent Successors

	|		2M|		|2M		peg2|		|peg2		2M|2M		2M|peg2		peg2|2M		|S		Total			
	♂	♀	♂	♀	♂	♀	♂	♀	♂	♀	♂	♀	♂	♀	♂	♂	♂	♀	♂	♀	♂/♀	%
aa|	1																		1		1	(2.6)
ab|	1	1	6	5			4	1											11	7	18	(47.4)
|ab					3					1			1		1				5	1	6	(15.8)
bb|	1	1																	1	1	2	(5.3)
|bb	2								1								1		4		4	(10.5)
cc|	1																		1		1	(2.6)
|cc		1																		1	1	(2.6)
ab|ab											3								3		3	(7.9)
ab|bM															1				1		1	(2.6)
cc|cc		1																		1	1	(2.6)
Total	6	4	6	5	3		4	1	1	1	3		1		2		1		27	11	38	(100.0)
(%)	(15.8)	(10.5)	(15.8)	(13.2)	(7.9)		(10.5)	(2.6)	(2.6)	(2.6)	(7.9)		(2.6)		(5.3)		(2.6)		(71.1)	(28.9)		

cessors in a sample of Hong Kong school children. The possible etiology and development of the anomaly are discussed.

Methods and Materials

The sample consisted of 376 children (186 boys and 190 girls) with ages ranging from 5 to 9 with a mean age of 6.8 $\pm$ 0.7 years. They were all normal and healthy Hong Kong school children participating in the school dental care service. Double primary teeth were found during routine clinical examination. All children were of Chinese ethnic origin and most had not been examined previously by a dentist.

Upper and lower anterior occlusal radiographs were obtained from each child. Occasionally periapical and panoramic radiographs also were taken to verify the condition of the permanent successors.

Children with a history of trauma to the primary dentition or premature loss of the primary teeth were excluded.

The "two-tooth rule" similar to that frequently applied to differentiate between fusion and gemination (Mader 1979) was employed to designate the types of double primary teeth according to their position in the dental arch. If the double tooth was counted as 2 teeth and if the normal number of teeth was present, the condition was designated as double primary tooth involving 2 adjacent teeth.

If the double tooth was counted as 2 teeth and if an extra tooth was present, the condition then was designated as double primary tooth involving the same tooth.

The clinical findings and corresponding radiographic interpretations were coded into the computer database for analysis. Other dental anomalies such as congenitally missing primary incisors and supernumerary teeth also were recorded.

In order to simplify the lengthy descriptions of the types of double primary teeth and their permanent successors, symbols were used to represent their conditions in the clinical observations and radiographic interpretations.

In the clinical observations, 'ab', 'bc', and 'de' represented the double primary teeth involving 2 adjacent teeth, while 'aa', 'bb', and 'cc' represented the double primary teeth involving the same tooth. a, b, c, d and e were the primary central incisor, lateral incisor, canine, first and second molars respectively. bM indicated that the primary lateral incisor was congenitally missing.

For the radiographic interpretations, the conditions of the permanent successors were represented as:

- 1M = permanent central incisor was congenitally missing
- 2M = permanent lateral incisor was congenitally missing
- peg2 = peg-shaped permanent lateral incisor was present
- $\widehat{23}$ = double tooth involving the permanent lateral incisor and canine was present
- S = the presence of a supernumerary tooth

The symbols ⊥ and ⊤ were used to indicate the corresponding right and left quadrants of the maxilla and mandible respectively.

Statistical analysis was performed using the single binomial probability test (Pollard 1977).

Fig 1. ab-type double tooth with 2M in the right maxillary arch.

Results

Double primary teeth were present in the maxillary arch in 38 children (27 boys, 11 girls) and in the mandibular arch in 334 children (158 boys, 176 girls). Four children (1 boy, 3 girls) had double primary teeth present in both the maxillary and mandibular arches.

The distribution of the 38 cases of maxillary double primary teeth with respect to sex, position in the dental arch and condition of the corresponding permanent successors is shown in Table 1.

A significant sex preference of double primary teeth in the maxillary arch in favor of boys was observed ($P < 0.05$) but with no preference for types. The majority of the maxillary double primary teeth were present as the ab-type (63.2%) and their association with 2M (Fig 1), peg2, 2M/peg2 (Fig 2) and normal permanent successors was in the ratio of 7:3:1:1. While all of the 3 bilateral ab-types were associated with bilateral missing permanent lateral incisors (Fig 3), the bb-types had normal, peg2 and supernumerary permanent successors in the ratio of 4:1:1. The aa- and cc-types had no influence on the shape and number of the permanent successors.

The distribution of the 334 cases of mandibular double primary teeth with respect to sex, position in the dental arch and condition of the corresponding permanent successors is shown in Table 2.

Statistically, there were significant preferences of mandibular ab-types occurring in boys ($P < 0.05$) and bc/bM combination types in girls ($P < 0.05$). Of the 31 ab-types, 30 were associated with normal numbers of permanent successors (Fig 4), with only 1 associated with a missing permanent lateral incisor. However, the bc-types which were the majority type of the anomaly (65.5%) were associated with 2M (Fig 5), normal, 2M/2M, and $\widehat{23}$-type permanent successors in the ratio of approximately 50:15:7:1. Of the 50 bc/bM combination types, 40 presented with bilateral missing mandibular permanent lateral incisors (Fig

Fig 2. ab/bM combination type double tooth with the corresponding peg2/2M combination in the maxillary arch.

6), 6 had only unilateral missing permanent lateral incisors and 4 had 2M/$\widehat{23}$ combination in the permanent successors. Bilateral bc-types were associated with 2M/2M, normal, 2M, and 2M/$\widehat{23}$ permanent successors in the ratio of approximately 10:3:2:1.

The bc-type double primary teeth showed significant preference to the right side of the mandibular arch ($P < 0.05$). Among the bc/bM combination types, the bM/bc type was significantly more than the bc/bM type ($P < 0.05$).

Two sisters presented with mirror images of bc-type double primary teeth with corresponding missing permanent lateral incisors, while 2 brothers exhibited identical forms of the bc-type with normal permanent successors.

Fig 3. Bilateral ab-type double teeth with bilateral 2M in the maxillary arch.

FIG 4. ab-type double tooth in the right mandibular arch with normal number of permanent successors.

FIG 5. bc-type double tooth with 2M in the right mandibular arch.

The positions and types of double primary teeth and conditions of their corresponding successors for the 4 cases having the anomaly present in both the maxillary and mandibular arches is shown in Table 3.

Bilateral occurrence of double primary teeth in both the maxillary and mandibular arches was not observed. The only case of double primary tooth involving the posterior teeth (Table 3, case #3) was found in the maxillary arch with concurrent bc/bM combination present on the mandibular arch.

In this sample, no bc-type double primary tooth was observed in the maxillary arch.

Discussion

The sex preference of maxillary and mandibular double primary teeth involving the central and lateral incisors in boys observed in this sample are consistent with findings reported for the Japanese population (Saito 1959). However, the frequency of double primary teeth involving the mandibular primary central and lateral incisors (9.3%) observed in this sample of mandibular double primary teeth is much lower. This probably reflects differences in sampling, as cases collected in this study were from a population with early mixed dentition where a number of mandibular double primary teeth involving the central and lateral incisors were likely to be lost due to exfoliation.

The frequency of double primary tooth formation involving 2 adjacent teeth, especially the primary central and lateral incisors in the maxillary arch or the primary lateral incisor and canine in the mandibular arch is much higher than any other type of double teeth. Only 10 of 372 cases presented with double formation involving the same tooth. Almost

TABLE 2. Distribution of Mandibular Double Primary Teeth as Related to the Permanent Successors

	|		2M|		|2M		|23		|1M		2M|2M		2M|23		23|2M		Total			
	♂	♀	♂	♀	♂	♀	♂	♀	♂	♀	♂	♀	♂	♀	♂	♀	♂	♀	♂/♀	%
ab|	15	4	1														16	4	20	(6.0)
|ab	8	3															8	3	11	(3.3)
bc|	6	11	46	52		1		1			4	6					56	71	127	(38.0)
|bc	13	15		1	26	25		2			7	3					46	46	92	(27.5)
bc|bc		6	1		1	2					13	8		2			15	18	33	(9.9)
bc|bM					1	2						9	1				2	11	13	(3.9)
bM|bc			2	1							10	21	1		1	1	14	23	37	(11.1)
bM|ab									1								1		1	(0.3)
Total (%)	42 (12.6)	39 (11.7)	50 (15.0)	54 (16.2)	28 (8.4)	30 (9.0)		3 (0.9)	1 (0.3)		34 (10.2)	47 (14.1)	2 (0.6)	2 (0.6)	1 (0.3)	1 (0.3)	158 (47.3)	176 (52.7)	334	(100.0)

FIG 6. bc/bM combination type double tooth with bilateral 2M in the mandibular arch.

TABLE 3. Positions and Types of Double Primary Teeth and Conditions of Corresponding Permanent Successors*

Case	Sex	Primary Double Teeth	Corresponding Permanent Successors
#1	♂	– / bb bc / –	– / – 2M / –
#2	♀	cc / cc – / bc	– / – – / –
#3	♀	de / – bc / bM	– / – 2M / 2M
#4	♀	ab / – aa / aa	2M / 2M – / –

* Cases having the anomaly present in both arches.

all cases occurred in the anterior region of the dental arches. Double primary posterior teeth are rare.

The high incidence of double teeth involving adjacent anterior teeth of the dentition has been attributed to the fact that such tooth germs are in the same developmental stage and are located close to each other (Yuasa 1944). The close proximity of the primary central and lateral incisors in the maxillary arch and the primary lateral incisors and canines in the mandibular arch have been shown by van der Linden et al. (1972) in both jaws before birth. Oöe (1972) argued that it is difficult to attribute the frequent occurrence of double teeth in the incisor region solely to the lack of space because double teeth can occur when there appeared to be ample space between the tooth germs. Furthermore, the tooth germs are surrounded by developing dental follicles which prevent the tooth germs from fusing with each other.

Embryologically, Oöe (1957) showed both the mandibular primary central and lateral incisors developed from a common primordium. This agreed with the clone theory (Osborn 1978) of tooth morphological development. In the clone model, Osborn proposed that the predifferentiated tooth-producing ectomesenchymal stem progenitors move to the presumptive jaws and then proliferate to produce teeth of certain shapes and sizes. The gradations of shapes and sizes in the series are expressions of intrinsic time-dependent alterations in the growing cell population which forms them (Lumsden 1979). Although development within the tooth-class may also occur interstitially,[5] the number of teeth within a tooth-class are limited by the mitotic potential of the elongating ends (the progress zones) of the tooth-class cell mass relative to the zone of inhibition created around it (Osborn and ten Cate 1983). An unimpeded continuation of the progress zone could have resulted in additional supernumerary teeth (Schwartz 1984). On the other hand, inhibition of the separation of neighboring tooth germs, perhaps by reduction of mitotic activity or increase of inhibition, could be the process for a missing tooth.

Obviously, by comparison with the field theory (Butler 1978) and the multifactorial model (Brook 1984), the clone theory alone is less able to account for the asymmetry common in tooth morphological development without considering the importance of environmental influence on the expression of the genome. ten Cate (1985) suggested that it is likely that a field initiates a clone.

Reports of inheritance of double teeth have been documented.[6] The presence of double primary incisors in mirror-image twins (Schneider 1985) and the occurrence of both double teeth and hypodontia in the same region in 3 of 8 sibs reported by Grahnen and Granath (1961) indicated a strong influence of genetic factors. If the condition is of genetic origin, it is probably either autosomal recessive or dominant with very little penetrance (Brook and Winter 1970). However, a clear Mendelian pattern cannot be established.

According to the concept of tooth development by means of its progress zone (Osborn and ten Cate 1983), the clone cells in the tooth-class cell mass 'I' (Fig 7A) would proliferate to become 2 separate cell masses 'I_a' and 'I_b' (Fig 7C) with the cell mass 'I_aI_b' occurring as an intermediate state (Fig 7B). And if the mitotic potential of the progress zone of the cell mass 'I_b' relative to the zone of inhibition created around it allows, it would continue to proliferate into another 2 separate cell masses 'I_b' and '$I_{b'}$' (Fig 7E) with the

[5] Berkowitz and Thomson 1971; Oöe 1971; Knapp and McMahon 1984.

[6] Moody and Montgomery 1934; Miles 1954; Brook and Winter 1970; Hutchin and Morris 1966.

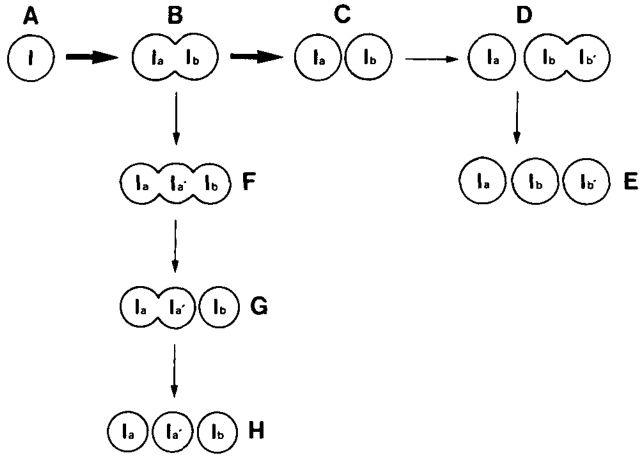

FIG 7. Schematic diagram of various types of double tooth formation according to the concept of tooth development by means of its progress zone. (A) incisor tooth-class cell mass 'I', (B) ab-type double primary tooth formed from intermediate cell mass 'I_aI_b', (C) primary central 'I_a' and lateral 'I_b' incisors, (D) bb-type double primary tooth formed from intermediate cell mass '$I_bI_{b'}$', (E) supernumerary primary lateral incisor '$I_{b'}$' in supplemental form, (F) triple primary tooth formed from intermediate cell mass '$I_aI_{a'}I_b$', (G) aa-type double primary tooth formed from cell mass '$I_aI_{a'}$', (H) supernumerary primary central incisor '$I_{a'}$' in supplemental form.

cell mass '$I_bI_{b'}$' occurring as another intermediate state (Fig 7D). In normal tooth development, such as in the primary maxillary incisors, the incisor tooth-class cell mass 'I' would proliferate to become the primary maxillary central and lateral incisors (i.e., cell masses 'I_a' and 'I_b' respectively). Occasionally, as the results of variations of local inhibitory influences, the cell masses 'I_aI_b', '$I_bI_{b'}$' and '$I_{b'}$' would be presented clinically as a double primary tooth involving 2 adjacent central and lateral incisors, a double primary tooth involving the same incisor tooth and a supernumerary lateral incisor in supplemental form respectively. Triple tooth (Ravn 1971; Knapp and McMahon 1984) would be formed if interstitial growth happened within the cell mass 'I_aI_b' to give the cell mass '$I_aI_{a'}I_b$' (Fig 7F). The timing and extent of inhibitory influences would determine not only the condition of the primary dentition but also the condition of the permanent successors. It would be obvious that double primary teeth involving the same tooth were developed either from tooth-class cell masses having lower mitotic potentials or developmental zones having more inhibitory potentials.

The results of this study showing a significant association of double primary teeth involving 2 adjacent teeth with anomalies in their permanent successors, as compared with those double primary teeth involving the same tooth, strongly suggest that the 2 patterns of double tooth formation may occur at different stages of the tooth germ development. The double primary teeth involving 2 adjacent teeth, having less tooth mass, would have occurred at a much earlier stage of tooth development than those involving the same tooth.

However, it is interesting to note that the patterns and types of double primary teeth formed in the maxillary and mandibular arches are different. In this sample, both the bc- and bc/bM combination types are observed only in the mandibular arch while bb- and cc-types are present only in the maxillary arch. Also, the ab-types in the mandibular arch differ from the maxillary counterparts by showing almost no association with anomalies in the permanent successors. This would suggest that either the environmental factors or the genetically determined tooth-class cell masses in the 2 arches are different.

The high frequency of congenital absence of primary incisor (i.e., bM) present concurrently with bc-type double primary tooth in the same arch as observed in this study would suggest that the bc-types, which we classified previously as double primary teeth involving 2 adjacent teeth, could have been classified as those involving the same tooth. In this case the tooth involved would probably be the primary canine with concurrent missing primary lateral incisor on the same side. The missing primary lateral incisor is the result of the incisor tooth-class cell mass failing to proliferate distally. This failure could in some cases allow the progress zone of the canine tooth-class cell mass to proliferate mesially. Similarly, the $\widehat{23}$-type double permanent teeth could better be classified as double teeth involving the permanent canine, with concurrent missing permanent lateral incisor. It is more acceptable than assuming 2 tooth germs at different developmental stages would fuse together to form the $\widehat{23}$-type double teeth. This concept could also help to explain why the 4 cases of $\widehat{23}$-type double permanent teeth were present as successors on the bM sides of the bc/bM combination types (as shown in Table 2). In other words, the bc-types presented in the mandibular arches are equivalent to the cc-types presented in the maxillary arches except that concurrent absence of primary lateral incisors is common in the mandibular arch. The common occurrence of missing primary lateral incisors would make the bb-type double tooth formation unlikely in the mandibular arch.

In terms of the ab-type double primary teeth, the significant difference in association with anomalies in permanent successors between the ab-types in the maxillary and mandibular arches cannot be due to differences in sampling. At this point, we can only suggest that the relationship between the double primary teeth and the types of anomalies in their permanent successors may be associated with the degree of separation of the double primary tooth mass. As the degree of separation of the tooth mass decreases, the permanent successors may appear peg shaped or may be missing altogether. If this is the case, the ab-type double primary teeth as observed in the mandibular arches in this study would have a higher degree of separation than those observed in the maxillary arches. Attempts have been made in this study to classify the degree of separation of the double primary teeth based on the radiographic interpretations in terms of the number of root canals or pulp chambers. However, it is difficult to derive a form of classification without additional information obtained from histological sections of the double teeth concerned.

Based on the results of this study, it seems that terms such as fusion or gemination commonly used to describe the anomaly of conjoined teeth may not be appropriate. Instead, a more appropriate description may be an unseparated tooth mass. Without establishing the etiology of the anomalies the term double tooth would seem to be more appropriate.

The close relationship of the double primary teeth with their permanent successors would justify the use of radiographs to confirm the number and condition of permanent successors in order to develop proper treatment plans for children with these anomalies.

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