

Bilateral Double Primary Teeth Associated with Multiple Odontogenic Anomalies in Permanent Dentition: A Case Report

SVSG Nirmala^{1*}, Sandeep Chilamakuri², Sunny Priyatham Tirupathi³

¹Department of paedodontics & Preventive Dentistry, Narayana Dental College & Hospital, India

²Former Post Graduate student, India

³Post graduate student, India

***Corresponding Author:** SVSG Nirmala, Department of Pedodontics & Preventive Dentistry, Professor, Narayana Dental College & Hospital, India.

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Abstract

Bilateral gemination in primary dentition is very rare. Bilateral gemination is seen in 0.01% to 0.04% in primary dentition and in 0.02% to 0.05% in permanent dentition. Here we report a unique case of multiple developmental abnormalities in a 7 year old boy which includes bilateral gemination in the maxilla. Orthopantomograph showed talon cusp in the left maxillary permanent lateral incisor; congenitally missing left second premolar in the mandible along with taurodontism of first permanent molars in the maxilla and mandible. Preventive measures were done and carious mandibular right second primary molar was restored with type IX GIC. Root stumps were present in the left mandibular primary molar which was extracted and lingual arch was given to maintain the space bilaterally. Geminated teeth were exfoliated normally. The possible etiological factors for all the three conditions along with treatment options are discussed.

Keywords: Bilateral gemination; bifid crown; congenitally missing premolar; first permanent molars; primary dentition; taurodontism; talon cusp

Geminated teeth are anomalies which arise from an attempt at division of a single tooth germ by an invagination, with resultant incomplete formation of two teeth. The structure is usually one with two completely or incompletely separated crowns that have a single root and root canal [1]. Gemination is often confused with fusion and distinction between both is difficult so the term "double teeth" is often used to describe both anomalies [2]. Gemination is a single enlarged tooth or joined tooth in which the tooth count is normal when the anomalous tooth is counted as one differentiation of fusion and gemination can be done by applying "Maders two tooth rule" If fused teeth are counted as one and the number of teeth in the dental arch is less than the term fusion is considered [3]. Etiology of gemination remains unknown. Evolution, trauma, heredity, and environmental factors are thought to play a role in germination [4]. This anomaly may also be associated with syndromes such as chondroectodermal dysplasia and achondrodysplasia [5].

Gemination occurs in both primary and permanent dentitions but more frequently it occurs in primary dentition and more frequent in maxilla than in mandible and more frequent in anterior region [6]. Unilateral gemination has a prevalence rate of 0.5% and 0.1% in deciduous and permanent dentition, respectively.

Bilateral gemination in primary dentition is very rare. Bilateral gemination is seen in 0.01% to 0.04% in primary dentition and in 0.02% to 0.05% in permanent dentition [7]. Turkestan., *et al* and Sener., *et al* reported cases of bilateral gemination [8,9].

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A survey of the literature showed that the prevalence estimates for bilateral double teeth range from 0.01% to 0.04% in the primary dentition, and 0.05% in the permanent dentition [10].

The purpose of this article is to present an unusual case of bilateral gemination in primary dentition with implications in the permanent dentition with underlying left lateral incisor with talons cusp, congenitally missing left lower second premolar as well as taurodontism of all four permanent molars in the maxilla and mandible.

Case Report

A 7 year old boy visited the department of Pediatric dentistry, for routine dental check-up. Medical history was non contributory and there were no apparent systemic manifestations. The clinical extra oral examination did not show any different alteration. Intra oral examination showed that he was in the early mixed dentition period with good oral hygiene. In the anterior region of the maxilla, double tooth due to gemination of primary lateral incisor was observed on both right and left sides (Figure 1&2).



Figure 1: Intra oral picture showing gemination of maxillary primary central and lateral incisors on right and left side of Maxilla.



Figure 2: Intra oral picture showing bilateral gemination from the occlusal aspect.

Orthopantomograph showed Talon cusp on the Maxillary left permanent lateral incisor and right side showed normally erupting permanent lateral incisor without any talons cusp, congenitally missing lower left second premolar as well as taurodontism of the first permanent molars both in the maxilla and mandible on either side (Figure 3). None of the family members were known to have the same trait. Preventive measures were performed. Oral and dental structures had a normal pattern obeying the chronology of eruption, therefore no immediate treatment was proposed and the patient's parents were only instructed to attend the clinic on a regular basis for follow-ups. Case was followed up for one year and fused primary incisor /primary canine tooth was exfoliated and succedaneous permanent maxillary left lateral incisor erupted with Talon cusp on the palatal aspect.



Figure 3: Orthopantomograph showing gemination of maxillary primary central and lateral incisors on right and left side along with Talon cusp on maxillary permanent left lateral incisor, agenesis of mandibular left second premolar as well as taurodontism of first permanent molars of maxilla and mandible.



Figure 4: Intra oral picture showing cemented Lingual Arch space maintainer in the mandible.

Discussion

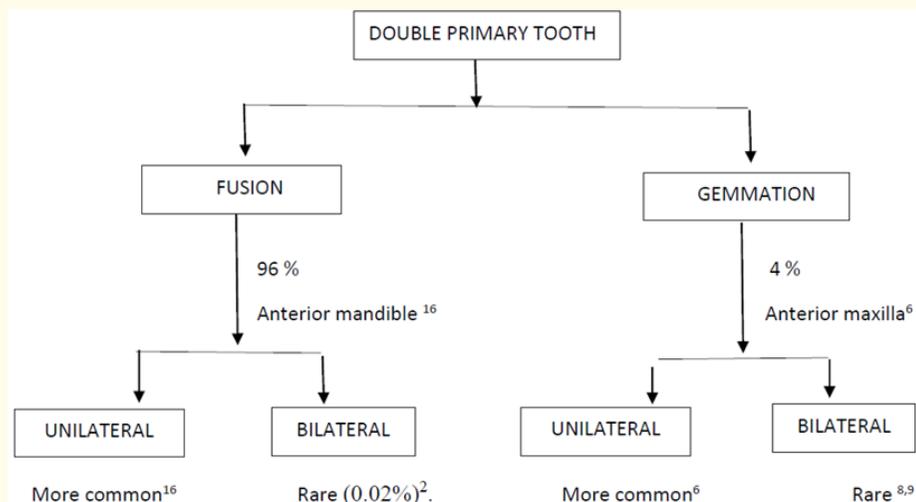
Prevalence Chart for Double Primary Teeth

The prevalence of double teeth varies between 0.1 and 3%. No gender differences were observed [11]. Double teeth occur more commonly in primary dentition than in permanent dentition. Double primary teeth due to fusion is more prevalent than double primary teeth due to gemination (In the primary dentition, with 94% of fusion and only 6% of germination) [12]. (Table 1) In our case, bilateral germination in the maxilla was present which is relatively rare.

Fusion commonly occurs unilaterally in mandible usually involving the lateral incisor and canine [13]. And gemination usually occurs unilateral in maxillary anteriors [14].

The clinical problems associated with double teeth includes, caries, delayed exfoliation and anomalies in the permanent dentition [15]. Caries was not observed in our case, only mild gingival inflammation was noted with respect to the geminated tooth.

Flow Chart



S No	Author	Age/Gender	Double Teeth		Associated anomaly with double teeth		Clinical follow up period	Radiographic follow up period	Management
			Maxilla Tooth no.	Mandible Tooth no.	Primary Tooth	Perma- nent Tooth			
1	Archana [29]	12 Male	22			Talon cusp	NS	NS	Extraction
2	Prabhakar [30]	9 years Male		41 & 42		Talon cusp	NS	NS	Treatment Not required
3	Nirmala [21]	8 years Female	52		Talon cusp		1 year	1 year	Extraction
4	Neeraja [31]	10 years Male	22			Talon cusp	NS	NS	Restored with Flowable composite
5	Shalini Gupta [32]	12 years Male	11			Talon cusp	NS	NS	Hemi section
6	Present Case	7 years Male	52 & 53 62 & 63			Talon cusp	1year	1 year	Exfoliated

NS: Not Stated

The presence of double primary teeth can also cause delayed resorption of root due to greater root mass and increased area of root surface relative to the size of permanent successor or crown [16] which may lead to delayed or ectopic resorption of permanent successor [17]. This finding is contradicting to our case as Double teeth were exfoliated within normal time.

Studies have shown that double primary teeth have an influence on permanent successors, including hypodontia most common (missing teeth), supernumerary teeth, repeated double teeth and peg-shaped teeth [18,19]. But in this present case there was no hypodontia with respect to primary double teeth due to bilateral gemination and the underlying permanent successors are present

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It has been stated that double teeth associated with crowding [20] but in this case crowding is not present but on the left side the permanent lateral incisor in the OPG showed increased radio opacity which after eruption was found to be talon cusp on the palatal surface of Left maxillary permanent lateral incisor. Talon cusp is a developmental anomaly which projects lingually from the cingulum areas composed of normal enamel and dentin and contains a horn of pulp tissue. The problems it poses for the patient in terms of aesthetics, caries control, and occlusal accommodation it is recommended prophylactically restoring the groove to prevent caries. Talon cusp may be associated with other syndromes such as Rubinstein-Taybi syndrome. Nirmala et al reported a case of unilateral gemination with talon cusp [21].

According to Stewart agenesis is defined as absence of one or few teeth and in reference to second premolars, agenesis of single second premolar is most common form but associated with other developmental anomalies are very rare [22]. Literature showed that submerged mandibular left second primary molar associated with agenesis of mandibular left second premolar with supernumerary tooth [23].

Tooth agenesis of second premolar affecting 25% of population. The molecular basis of the defect is not completely understood, despite identification of several mutations in MSX1 and PAX9 genes that seem to be crucial for tooth agenesis.

The mechanism for the occurrence of congenitally missing teeth may be due to the expression or misexpression of certain genes at certain times in the development of a tooth germ. In some cases the developing tooth germ may be initiated normally, however, abnormal apoptosis leads to involution of the developing tooth. Certain genes promoting the progression of tooth morphogenesis are not expressed for the process to proceed. Alternatively, the genes that cause programmed cell death are inadvertently expressed causing the body to start resorbing the developing tooth germ [24].

Agenesis of mandibular second premolars leads to many problems. The clinicians must make the proper decision at the appropriate time, regarding management of the edentulous space. If the space is left open for an eventual restoration, the correct amount of space must be created and the alveolar ridge must be left in an ideal condition for a future restoration. In the past either conventional bridges or resin bonded bridges were used to fill edentulous space. However full coverage conventional bridges in young patients can result in devitalisation of the pulp and require root canal therapy. Resin bonded posterior bridges have questionable survival rates [14]. Today the first choice of restoration for a congenitally missing mandibular premolar should be a single tooth implant [25].

Case presented by us, mandibular left second primary molar was caries free; hence no treatment was carried out.

OPG of the present case showed all four permanent molars bilaterally exhibiting taurodontism. The term 'taurodontism' was proposed by Sir Arthur Keith in 1913 to describe a peculiar dental anomaly in which the body of the tooth is enlarged at the expense of the roots. The term means 'bull-like' teeth and its usage is derived from the similarity of these teeth to those of cud chewing animals. The prevalence of Taurodontism is reported to range from 2.5% to 11.3% of the human population [22-26].

A variety of possible causes of taurodontism have been enumerated by Mangion as follows: (1) a specialized or retrograde character, (2) a primitive pattern, (3) a mendelian recessive trait, (4) an atavistic feature, and (5) a mutation resulting from odontoblastic deficiency during dentinogenesis of the roots. Hammer and his associates believe that the taurodont is caused by failure of Hertwig's epithelial sheath to invaginate at the proper horizontal level. Goldstein and Gottlieb have stated that the condition appears to be genetically controlled and familial in nature. Taurodontism appears most frequently as an isolated anomaly, but it has also been associated with several developmental syndromes and anomalies including amelogenesis imperfecta, Down's syndrome, ectodermal dysplasia, Klinefelter syndrome, tricho-dento-osseous syndrome, Mohr syndrome, Wolf-Hirschhorn syndrome and Lowe syndrome and vander woude syndrome [22].

Taurodontism may affect either the primary or permanent dentition, although permanent tooth involvement is more common. The teeth involved are almost invariably molars, sometimes only a single tooth, at other times several molars in the same quadrant. The

condition may be unilateral or bilateral or may exhibit any combination of quadrant involvement. The teeth themselves have no remarkable or unusual morphologic clinical characteristics.

Identification of the Taurodont teeth can only be made by radiographic examination as the external morphology of the teeth is within normal configurations.

Shifman and Chanannel [26] proposed a radiographic criteria for evaluating the presence of Taurodontism, if the distance between the CEJ and the floor of the pulp chamber is ≥ 2.5 mm and if the distance from the lowest point at the occlusal end of the pulp chamber (A) to the highest point at the apical end of the chamber (B) divided by the distance from A to the apex is 0.2 or greater [27].

The treatment of choice for taurodontism is root canal treatment and the presented case did not require treatment as all the first permanent first molars were caries free hence only preventive measures were done.

In cases of primary double teeth with associated successional hypodontia/anodontia every possible step should be considered to retain the Primary double tooth in the arch by performing appropriate endodontic treatments [28].

Primary Double Tooth associated with further problems in the permanent successors such as hypodontia, double teeth, and peg-shaped teeth. These problems may affect tooth alignment, arch symmetry and facial appearance. In addition, double primary teeth may cause delayed resorption of the root because of a big root mass, resulting in delayed or ectopic eruption of permanent successors. In order to intercept future malocclusion, further treatment, including extraction, partial removal, or separation of double teeth, should be considered. In the presented case double teeth were exfoliated normally.

Orthodontic and Prosthodontic management should be considered to ensure functional occlusion and improve esthetics. In a preventive concern, the labial and lingual vertical grooves of the double primary teeth may be pronounced and difficult to clean, and are highly susceptible to caries so Sealing the grooves with sealant or resin may decrease the risk of caries.

Few reported latest cases of double teeth with associated anomaly have been given in the table 2.

Conclusion

The existence of double primary teeth due to bilateral gemination which is associated with underlying permanent successor with a talons cusp is a rare finding along with congenitally missing mandibular second premolar as well as taurodontism of four permanent first molars is very rare in the same child. Appropriate diagnosis of primary double teeth and its etiology can be a valuable aid to device correct treatment plan.

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