

# NON-SYNDROMIC FAMILIAL SUPERNUMERARY TEETH: CASE REPORT AND REVIEW OF THEIR PHENOTYPIC CHARACTERISTIC

Case Report

W.N. Wan Hassan, N. Ab. Rahman. *Non-syndromic familial supernumerary teeth: Case report and review of their phenotypic characteristic. Annal Dent Univ Malaya 2012; 19(1): 28-35.*

## ABSTRACT

Supernumerary teeth have a genetic predisposition with a predilection for males. This article reports an uncommon radiological finding in a non-syndromic sibling pair who presented with supernumerary teeth of different morphologies on opposite and different regions of the dental arches. A 14-year-old Chinese male presented with a conical supernumerary palatally placed between the upper right central and lateral incisors. His older brother had unerupted bilateral supplemental supernumerary teeth between the roots of the lower second premolars and first permanent molars. Trends of the phenotypic presentation of familial non-syndromic supernumerary cases are discussed. Familial supernumerary teeth have been suggested to be due to autosomal dominance or recessive traits. Variation in the numeral, spatial and morphological phenotypic expressions suggests a multifactorial model of multiple genetic, epigenetic and environmental influences. Clinicians need to be mindful of the possible phenotypic variations that may present when treating cases with family history of dental anomalies.

Key words: Tooth; Supernumerary; Siblings; Male; Phenotype.

## INTRODUCTION

Supernumerary teeth or hyperdontia are additional teeth to the normal complement of the dentition. They are more common in males (1-2) with a predilection of 2 or 2.2 to 1 (3-4) and reported prevalence of between 0.06 to 0.34% (1, 3, 5-6). The order of location has been reported to be more frequently in the premolar than anterior region (1). Non-syndromic multiple supernumerary teeth have been reported to be more frequently in the mandible than maxilla and more frequently in the canine-premolar region while the incisal and molar regions had similar rates of frequencies (7). In the mandible, the canine-premolar region was more frequently affected while the maxilla was most affected in the molar and incisal regions (7). Complications of supernumerary teeth include that these supernumerary teeth may be impacted or have delayed eruption, may cause resorption of adjacent teeth, malpositioning and rotation of the adjacent teeth

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or movement of the unerupted teeth, enlargement of the follicles or develop as dentigerous cysts, development of further supernumeraries, may be a mechanical or obstructive pathology and may fuse to the adjacent teeth of normal tooth series (1,6,8-11).

The aim of this report is to document a non-syndromic familial case of a sibling pair with single and dual supernumerary teeth respectively, located in the opposing jaw and in different regions of the jaw and to discuss the phenotypic pattern of familial non-syndromic supernumerary teeth.

## CASE REPORT

A pair of siblings visited the Faculty of Dentistry, University of Malaya for orthodontic treatment.

### Case 1

A 14-year-old Chinese male presented with a Class II Division I malocclusion on a Class I skeletal base. The malocclusion was complicated by severe crowding on the maxillary and mandibular arches with buccally unerupted maxillary permanent canines, lingually erupted mandibular right permanent canine and buccally displaced mandibular first premolars. The medical history did not reveal any systemic disease or syndrome. The orthopantomogram and maxillary occlusal view (Figure 1 and 2) showed that all the permanent dentition including the third molars were present and a palatally displaced conical supernumerary tooth between the roots of the maxillary right central and lateral incisors. Treatment plan included surgical removal of the supernumerary tooth prior to orthodontic treatment.



Figure 1. Orthopantomogram of the Case 1 patient showing present of supernumerary between the roots of the maxillary right permanent central and lateral incisors.

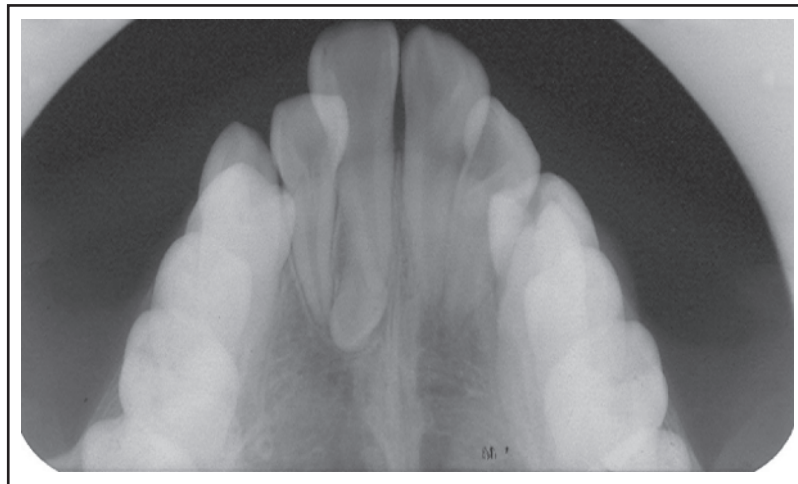


Figure 2. Upper occlusal view of the Case 1 patient showing conical supernumerary tooth between root of maxillary right central and lateral incisor.

### Case 2

The patient's brother, a 16-year-old Chinese male presented with a Class III malocclusion on a mild Class III skeletal base. The malocclusion was complicated by moderate crowding on the maxillary and mandibular arches with palatally erupted maxillary left permanent lateral incisor, mesio-lingually rotated mandibular left permanent lateral incisor and disto-bucally rotated mandibular right permanent central incisor. The medical history did not reveal any systemic disease or syndrome. The orthopantomogram (Figure 3) showed that all permanent dentitions including the third molars were present. The mandibular right and left third permanent molars were mesio-angularly impacted. Developing supplemental supernumerary teeth were detected bilaterally on the mandibular right and left premolar regions between the roots of the mandibular second premolars and the first permanent molars. Treatment

plan also included surgical removal of the supernumerary teeth prior to orthodontic treatment. However, the patient refused to undergo surgical removal of the supernumerary teeth and therefore did not proceed to commence orthodontic treatment.

### Family history

The mother had an orthopantomogram taken for another dental investigation which did not show the presence of any supernumerary teeth (Figure 4). Based on the family history, it was not known if any other family members of this case study had supernumerary teeth. The family was informed of a possibility for the presence of supernumerary teeth in their family members and had invited the patients' father and eldest brother for a dental check up. However, they were not interested to attend for an appointment. Therefore, no further investigation was made.

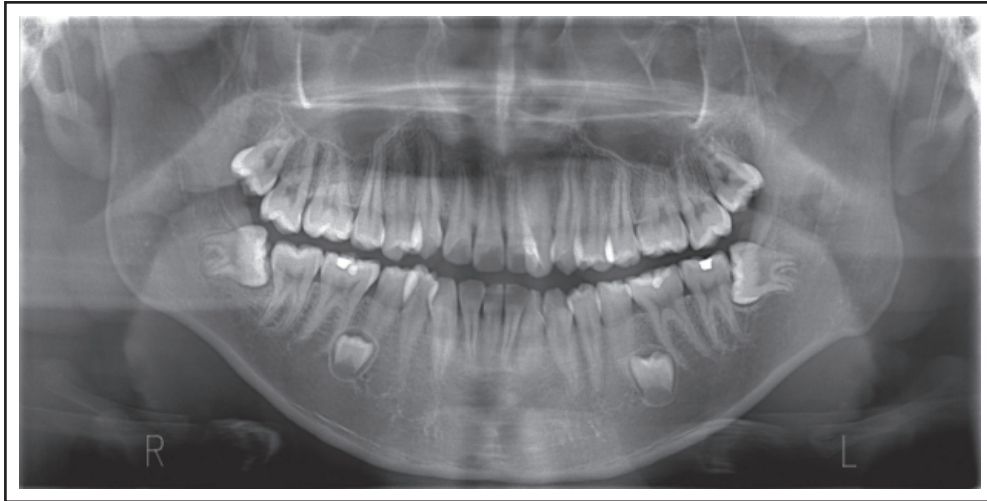


Figure 3. Orthopantomogram of the Case 2 patient showing presence of developing bilateral supplemental mandibular premolar supernumerary teeth.

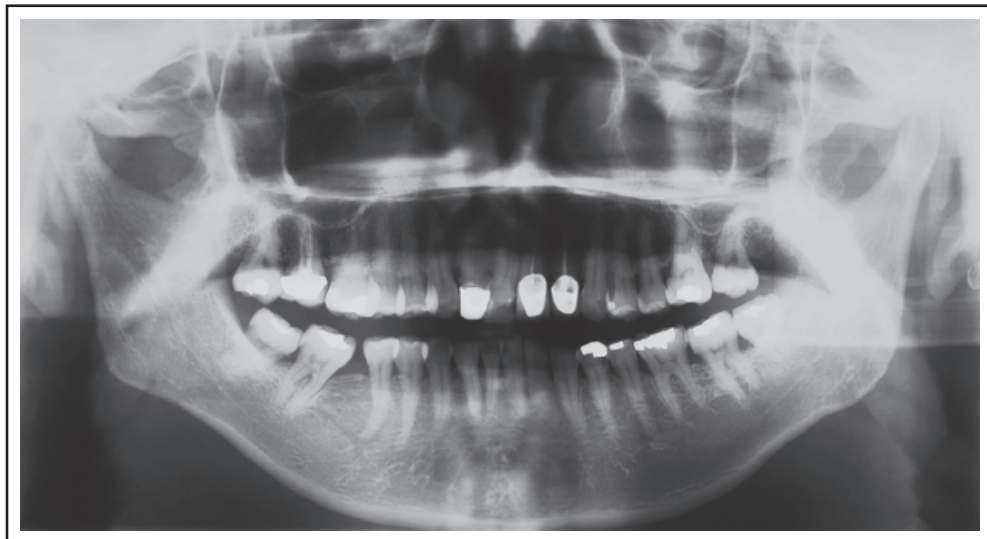


Figure 4. Orthopantomogram of the patient's mother indicating absence of supernumerary teeth.

## DISCUSSION

There have been several proposals for the etiology of the development of supernumerary teeth: atavism of a phylogenetic reversion to extinct primates; dichotomy of the tooth bud to split which later develops into separate teeth; an independent, locally conditioned hyperactivity of the dental lamina; and genetic linkage to the dental anomaly (3-4, 12-13). Several candidate genes have been identified to play a role in the development of supernumerary teeth in humans including the Hedgehog, FGF, Wnt, TNF and BMP families (13). The outcome of the inappropriate regulation of the molecular signaling pathway in the normal development of the tooth germ is observed in the phenotype. Townsend *et al.* (2009) proposed a multifactorial model linking tooth size and number to account for the different patterns of expression observed in supernumerary teeth cases (14). Tempo-

spatial variations in the signaling pathways during development have been suggested as the potential etiology to the variation in the phenotypes. Such multifactorial model, with multiple genetic, epigenetic and environmental influences would provide the explanation for the observation seen in familial cases of supernumerary teeth particularly in cases of discordance in monozygotic twin pairs (14).

Supernumerary has been suggested to be due to an autosomal dominant (15-16) or autosomal recessive trait (17) with variable penetrance. Since the anomaly does not follow a simple Mendelian pattern, it would be prudent to suspect family members of patients presenting with supernumerary teeth to be susceptible to have this dental anomaly (18). The timing of development of supernumerary teeth may be later than that of the dental series (19). Therefore, patients or their family members need to be advised of the risks of developing supernumerary teeth and the risks that may

present if the dentition were moved for orthodontic treatment should a supernumerary tooth develop during treatment. Supernumerary teeth are usually removed prior to orthodontic treatment if they risk obstructing tooth movement or predispose to root resorption of the adjacent teeth. However, they may be left *in situ* during orthodontic treatment following surgical advice of the risk of damaging adjacent structures outweighs the removal benefits without causing significant damage to the adjacent teeth (20).

A search through online databases (Pubmed®, Web of Science<sup>SM</sup>, Scopus) and individual papers of English written case reports up to December 2011 was made to identify the common pattern of phenotypic locations of supernumerary teeth in familial non-syndromic cases based on the number of supernumerary in the individuals within the family (Tables 1 to 3).

A general observation seemed to suggest that the trend for familial non-syndromic single supernumerary teeth from within the nuclear family, whether they were in the permanent or deciduous dentition, tend to be located in the maxillary anterior region (Table 1).

Seddon *et al.* (1997) suggested the numbers of mesiodentes in monozygotic twins had a high degree of concordance (21). However, this finding was based on literature search. In contrast, Townsend *et al.* (2005) found a higher degree of discordance (88.9%) in their monozygotic twin pair sample with mesiodentes (22). The mesiodentes could be found in the same side or as mirror images. Familial non-syndromic single supernumerary teeth in other locations were rarely reported with only one case report of a fifth mandibular incisor where the subjects were related to each other as the eighth generation of a consanguineous pedigree family (17, 23). It was suggested that fifth mandibular incisor is an autosomal recessive trait due to a mutation in the homozygous region on chromosome 16q12.2 (17).

The phenotypic locations of familial non-syndromic multiple supernumerary teeth may include other regions of the jaw. The more number of supernumeraries found in a family member, the spatial distributions tended to be less predictable than in the single supernumerary cases (Table 3). In cases where

**Table 1.** Reported spatial phenotype of familial non-syndromic single supernumerary cases

First Author (Year)	Ethnic	Proband's gender; Family member	Number of supernumerary teeth reported within the spatial region of the maxilla and mandible									
			Incisor		Premolar		Molar		MdR	Md L		
			Mx	Md	MxR	MxL	MdR	Md L				
Witanowska (2011) <sup>21</sup>		Monozygotic twin females	1									
Babacan (2010) <sup>22</sup>		Monozygotic male twins	1									
Wicomb (2009) <sup>23</sup>	Caucasian	Male Brother	1 <sup>§</sup>									
Cassia (2004) <sup>24</sup>	Lebanese <sup>¶</sup>	Male		1								
		Female		1								
		Female		1								
		Male		1								
Sharma (2003) <sup>25</sup>		Female Father	1									
Atwan (1998) <sup>26</sup>		Female	1									[Father and maternal uncle had history of a mesiodens]
Almeida (1995) <sup>27</sup>	Caucasian	Male	1									
		Sister	1									
		Sister	1									
Mason (1995) <sup>28</sup>	*Family 1	Male	1									
		Mother	1									
	*Family 2	Male	1									
		Father	1									
Beere (1990) <sup>29</sup>	African	Twin males	1 <sup>§</sup>									
			1 <sup>§</sup>									
Carton (1987) <sup>30</sup>		Twins (gender NA)	1									
			1									
Sedano (1969) <sup>31</sup>		Female	1									
		Brother	1									

\*Location (Region) legend: (Mx) Maxillary arch; (Md) Mandibular arch; (R) Right side of the arch; (L) Left side of the arch; [square brackets] based on patient's history; (§) deciduous teeth; (¶) Patients are related as the 8<sup>th</sup> generation of a consanguineous pedigree family; (NA) Information not available

the family member had at most two supernumerary teeth, the trend for the phenotypic locations of the supernumeraries were similar to single supernumerary cases where they were generally found in the maxillary anterior regions (Table 2). There were few exceptions including the current case and another report in which the twin pair with mesiodens had their second supernumerary in the mandibular premolar region as mirror images to each other (24). Another case reported of monozygotic twins with bilateral mesiodens and a family history where the paternal grandmother had a mesiodens removed in childhood but father had a supplemental premolar (21).

In cases where at least one of the family members had more than two supernumerary teeth, the phenotypic locations were generally more sporadic, with at least one family member with supernumerary teeth in the posterior region (premolar and/or molar regions) (Table 3). The family member may also have them in the same region (anterior, canine-premolar or molar region) of the same jaw or in the same region of the opposite jaw, in different regions of the same jaw or in different regions of the opposing jaw.

In terms of the morphology, supernumerary teeth can present as conical or tuberculate form, which are usually found in the maxillary anterior region, as supplemental teeth with similar morphology to the teeth of the normal tooth series of the region or as odontome, which may be disorganized or bear some anatomical similarity to a normal tooth (18). Tuberculate supernumerary has also been known as molariform supernumerary because it has more than one cusp or tubercle (25). Few familial cases reported variation in the morphological expression of the supernumerary teeth. Sharma *et al.* (2003) reported molariform mesiodens single supernumerary teeth presented in their female patient but the patient's father had conical mesiodens in the maxillary anterior region (26). Verma *et al.* (2010) described of a case where a male patient had a tuberculate mesiodens and the father had two fused mesiodens (25). Marya and Kumar (1998) reported of a male patient with two conical mesiodens but his brother had one conical mesiodens and a molariform mesiodens (27). Seddon *et al.* (1997) found incisiform mesiodens on the left side of their twin patients but on the right side, one twin had

**Table 2.** Reported numerical and spatial phenotype of familial non-syndromic supernumerary cases with at least one family member with at most two supernumerary teeth

First Author (Year)	Ethnic	Proband's gender; Family member	Number of supernumerary teeth reported within the spatial region of the maxilla and mandible									
			Incisor		Premolar		Molar					
			Mx	Md	MxR	MxL	MdR	Md L	MxR	MxL	MdR	Md L
Current case report	Chinese	Male Brother	1					1	1			
Verma (2010) <sup>32</sup>	*Family 1	Female Brother	1									
		2										
	*Family 2	Male Father	1									
		2 <sup>§</sup>										
Townsend (2005) <sup>33</sup>	*Family 1	Twin males	1									
		2										
	*Family 2	Twin males	1									
		2										
	*Family 3	Twin males	2									
		2										
Gallas (2000) <sup>34</sup>		Female Sister	2									
		2										
Brand (2000) <sup>35</sup>		Male twins	1						1			
		1					1					
Marya (1998) <sup>36</sup>		Male Brother	2									
		2										
Seddon (1997) <sup>37</sup>	Afro-Caribbean	Monozygotic male twins	2									
		2										
		Father Grandmother								[History of a supplemental premolar]		
Choi (1990) <sup>38</sup>	Chinese	Male twins	2									
		2										
		Brother									[History of an extracted mesiodens]	
Schon (1974) <sup>39</sup>		Twin males	2									
		2										

\*Location (Region) legend: (Mx) Maxillary arch; (Md) Mandibular arch; (R) Right side of the arch; (L) Left side of the arch; [square brackets] based on patient's history; (§) fused supernumerary teeth.



**Table 3.** Reported numerical and spatial phenotype of familial non-syndromic supernumerary cases with at least one family member with more than two supernumerary teeth

First Author (Year)	Ethnic	Proband's gender; Family member	Number of supernumerary teeth reported within the spatial region of the maxilla and mandible											
			Incisor		Premolar		Molar							
			Mx	Md	MxR	MxL	MdR	Md L	MxR	MxL	MdR	Md L		
Inchingolo (2010) <sup>40</sup>	Caucasian	Male											1	
		Sister								1	2	1	1	
		Sister								1	1			
		Mother	[History of avulsed supernumerary in the posterior region of the superior maxillary bone]											
Bhatia (2010) <sup>41</sup>		Female						3	2					
		Mother										3	1	
		Sister								2	2	3	1	1
Location of 3 other supernumeraries were not specified														
Cho (2009) <sup>19</sup>		Male	[1]						1	2				
		Sister							1	2				
Krishnan (2009) <sup>42</sup>	Asian	Male				1			3	3			1	
		Sister							1	2				
		Father									1			
Wang (2007) <sup>16</sup>	Chinese	Female							6 in the Mx premolar region	8 in the Mn premolar region		1	1	
		Mother	Multiple supernumerary teeth on both jaws. Located in most regions but not clearly described. [Maternal grandfather and great grandmother had history of supernumerary teeth. Locations were not specified]											
Orhan (2006) <sup>43</sup>	Caucasian *Family 1	Male				1			3	3			1	
		Mother							2	1				
	Caucasian *Family 2	Male			2		1			2	2			
		Mother							[1]	2 <sup>§</sup>	[2] <sup>§</sup>			
Batra (2005) <sup>15</sup>	Indian	Female	1	2	1	1			3	3				
		Brother		4					2	2				
		Father				2	1							1
Langowska-Adamczyk (2001) <sup>44</sup>		Monozygotic twin males	[1]						1 <sup>§</sup>	1	1	1	1	
		Mother	[1]						1 <sup>§</sup>	1	1	1	1	
[History of treated supernumerary but locations were not specified]														
Desai (1998) <sup>45</sup>		Male						2	1	1	1	2		
		Brother	2		1	1			3	2	3	2	1	1
Babu (1998) <sup>46</sup>	Indian	Female	1	3										
		Mother	8 supernumerary teeth in the mandible. Locations were not clearly described.											
Scanlan (1997) <sup>20</sup>	African	Female								1				
		Brother										1	1	1
Jasmin (1993) <sup>47</sup>	Caucasian	Male twins							2	1				
									1	2				
Konttinen (1984) <sup>48</sup>		Female							1	2	2	2		
		Sister								2	2	1		
		Sister												1
Mercuri (1980) <sup>49</sup>		Female	27 Multiple supernumerary teeth on both jaws. Located in most regions but were not clearly described.											
		Sister	17 Multiple supernumerary teeth on both jaws. Located in most regions but were not clearly described.											
		Father	[History of supernumerary teeth]											
Brodie (1936) <sup>50</sup>		Male	4		[Locations of 6 other supernumeraries were not specified]									
		Sister	1	1										
		Sister	1											

\*Location (Region) legend: (Mx) Maxillary arch; (Md) Mandibular arch; (R) Right side of the arch; (L) Left side of the arch; [square brackets] based on patient's history; (§) Reported as canine-premolar region.

tuberculate and the other twin had conical supernumeraries (21). In the case of the relatives of the eighth generation of a consanguineous pedigree family, two cases had distinct fifth mandibular incisor while the other two had fused mandibular incisor (23). In this case study, the supernumerary teeth presented with morphologies as expected when found in the regions of the arches but their morphology was different to each other; the mesiodens in the younger brother was conical while the older brother had supplemental premolars.

Common complications were also noted in these familial supernumerary cases such as malpositioning of the incisors (26-28) and impaction of the permanent and supernumerary teeth. Few papers have reported unerupted incisors due to the presence of mesiodens in twins (21, 24, 29-32). Families with multiple supernumeraries have shown to commonly have multiple impacted teeth (33-36). The impacted permanent teeth may be of the same tooth series (37) although some cases have shown variation in the teeth that is impacted. Batra *et al.* (2005) found multiple impacted permanent teeth in their female patient but her brother only had an impacted canine (15). Scanlan and Hodges (1997) reported impacted palatal canine in their female patient with a supernumerary in the mandibular region but the brother of the patient had impacted second molars due to the occlusal position of the supernumerary teeth (20). Such complications may require removal of the supernumerary to allow for spontaneous eruption of the permanent teeth or orthodontic alignment. Less commonly reported associated hereditary trait includes retention of the primary set of deciduous teeth (35).

In conclusion, the variations observed in the phenotypic pattern of non-syndromic familial supernumerary teeth highlights the complex genetic association of this dental anomaly which may support the multifactorial model of multiple genetic, epigenetic and environmental influences during development (14). Radiographic guideline stresses the need to restrict the radiation doses to patients as low as reasonably practicable to minimize the amount of radiation exposure (38). However, it may be prudent to take appropriate radiograph views prior to treatment planning, particularly if such patient has a family history of dental anomalies.

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