

Multiple Supernumeraries in a Non-Syndromic Patient

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Presence of supernumerary teeth (ST) in non-syndromic patients is an exceptional event. The aim of this article is to present an unusual case of a non-syndromic 12 years old girl with 12 ST. In the reported case supernumeraries were found in three quadrants and the most common ST were bicuspid. All ST were surgically excised and occlusion and functionality was restored with prosthetic appliances. ST are an exceedingly uncommon event in the Pediatric Clinic. An extensive review of the literature dealing on non-syndromic cases comprising 10 or more ST and a discussion about the origin of the ST is presented.

Keywords: Supernumerary teeth, origin, radiographs, epidemiology, surgery.

INTRODUCTION

Abnormalities in the number of teeth are occasionally noted during the Pediatric Clinical Practice. Despite their rarity, supernumerary teeth (ST) are considered one of the most common developmental dental anomalies. Frequency of ST is widely variable depending on the method employed to collect data, country and analyzed ethnic group. Several studies showed that frequency of these teeth varied from 0.06% to 1.05%^{1,2} and frequency of ST in pediatric population varied from 0.36%³ to 1.53%.⁴ It is well known that ST appear in children affected by a wide variety of syndromes and that their presence in non-syndrome affected patients is a rare event.⁵ Cases with more than 10 ST are a very unusual finding in the Private and Institutional Pediatric Dental Practice. We reviewed those cases previously published including those mentioned in some reviews of the literature.⁵⁻²⁴

More than 200 genes have been implicated in odontogenesis; some of them are associated with codification of transcription

factors, others with growth factors and others with extracellular matrix protein production and secretion.²⁵ The etiology for the development of ST is unknown to date and since several years ago, different theories as hyperactivity of the dental lamina, splitting of a tooth bud, a dichotomy mechanism and atavism have been proposed to explain the development of ST.^{17,18,26,27}

An extensive review of the literature on ST was made, selecting only clearly described cases with no clinical or radiographic signs of any syndrome and dealing with 10 or more erupted or non-erupted ST. As it is shown in table 1, 18 cases were included.^{5,7-11,13-23} The main clinical features of the included cases are summarized in table 1.

We found 12 boys (66.7%) and six girls (33.3%). Age of the patients varied among nine to 36 years (mean age=17.06 years). It should be noted that 61% (n=11 cases) of the sample were 18 years old or younger patients.

In this review, a total of 237 teeth were recorded. The number of teeth *per* patient varied from 10 to 22 teeth. 113 ST (47.7%) were found in maxilla and 124 ST (52.3%) in mandible. The most common ST were bicuspid (66.67%) followed by incisors (9.28%) and distomolars (7.17%); frequency of mesiodens was very low (3.38%).

In mandible, bicuspid were the most common ST (79.03%) followed by incisors (8.06%) and canines (6.45%). In maxilla, they were bicuspid (53.1%), distomolars (11.5%) followed by incisors and molars (10.62% respectively). A detailed location and number of the studied teeth is shown in table 2.

As it is shown in table 3, 158 ST (66.67%) appeared in boys and 79 ST (33.33%) in girls. In boys, the most frequent ST were bicuspid (63.9%) followed by canines and incisors (9.49% and 8.86% respectively). In girls, the most common ST were bicuspid, incisors and distomolars (72.15%, 10.12% and 7.59% respectively).

The aim of this article is to present an uncommon case of a 12 years old girl with no clinical or radiographic signs of any syndrome with 12 unerupted ST and the results of an extensive review of the literature concerning previously reported cases of non-syndromic patients with 10 or more ST.

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Table 1: Previously published cases of 10 and more supernumeraries

Author	Age	Gender	Number of teeth	Location	
				Maxilla	Mandible
Wang <i>et al</i> (5)	11	Girl	16	6 bicuspid 2 distomolars	8 bicuspid
Arcuri <i>et al</i> (7)	9	Girl	13 ¹	1 mesiodens 3 bicuspid 2 molars 2 distomolars	1 incisor 4 bicuspid
Rizzuti & Scotti (8)	10	Boy	22	1 temporal incisor 2 incisors 2 canines 5 bicuspid 1 distomolar	2 incisors 2 canines 5 bicuspid 2 distomolars
Arathi & Ashwini (9)	14	Boy	12 ²	1 incisor 4 bicuspid	2 incisors 5 bicuspid
Nayak & Mathian (10)	18	Boy	13	2 molars 4 bicuspid	1 incisor 6 bicuspid
Desai & Shah (11)	36	Boy	16	1 mesiodens 1 incisor 1 canine 1 bicuspid 4 molars 1 distomolar	5 bicuspid 1 molar 1 distomolar
Foley & del Rio (13)	22	Boy	16 ³	1 canine 6 bicuspid 1 molar 2 distomolars	3 canines 2 bicuspid 1 molar
Stevenson & McKechnie (14)	10	Boy	12 ⁴	4 mesiodens 1 canine 1 bicuspid	6 bicuspid
Fitzgerald & Zallen (15)	20	Boy	12 ⁵	1 incisor 3 bicuspid 1 distomolar	2 canines 5 bicuspid
Mercury & O'Neil (16)	17	Girl	11 ⁶	1 mesiodens 2 distomolars	8 bicuspid
Yusof & Awang (17)	22	Girl	10	2 bicuspid 1 molar	6 bicuspid 1 molar
Hopcraft (18)	18	Boy	10	3 bicuspid 2 molars	5 bicuspid
Mason <i>et al</i> (19)	15	Boy	16	2 incisors 2 bicuspid 2 distomolars	9 bicuspid 1 distomolar
Bartleman (20)	18	Boy	12	2 incisors 4 bicuspid	6 bicuspid
Batra <i>et al</i> (21)	17	Girl	11	1 mesiodens 2 bicuspid	2 incisors 6 bicuspid
Duffy (22)	23	Boy	13	6 bicuspid	1 canine 6 bicuspid
Sharma (23)	12	Girl	11	3 incisors 4 bicuspid	2 incisors 1 bicuspid 1 molar
So (24)	15	Boy	11 ⁴	2 canines	4 bicuspid 5 bicuspid
Ledesma-Montes <i>et al.</i> This report	12	Girl	12	1 canine	1 incisor 2 canines 6 bicuspid 2 molars

¹One mesiodens the authors did not take in count. ²Three anterior supernumeraries previously extracted with unknown morphology (none included in this tabulation). ³One ectopic supernumerary canine not taken in count. ⁴The maxillary odontoma was not considered as supernumerary and only five supernumerary lower bicuspid were found. ⁵12 supernumeraries from the 16 mentioned were located. ⁶Of the 27 impacted and supernumerary teeth, 16 teeth were of the normal dentition.

Table 2: Frequency of supernumerary teeth in this study

MAXILLA						
Type of Teeth	Maxilla	%	Mandible	%	Total	%
Mesiodens	8	7.08	0	0	8	3.38
Incisors	12	10.62	10	8.06	22	9.28
Canines	7	6.19	8	6.45	15	6.33
Bicuspid	60	53.1	98	79.03	158	66.67
Molars	12	10.62	4	3.23	16	6.75
Distomolars	13	11.5	4	3.23	17	7.17
Temporal Incisor	1	0.89	0	0	1	0.42
Total	113	100	124	100	237	100

Case Report

A 12 years old Mexican Mestizo girl was received in the Outpatient and Diagnosis Clinic of the División de Estudios de Posgrado e Investigación (Facultad de Odontología, UNAM) for treatment of primary teeth with extensive carious lesions. Clinical oral examination revealed that several teeth were absent including all permanent canines, both permanent mandibular lateral incisors and both mandibular second bicuspid with retention of all four primary canines and both primary mandibular second molars. Extensive carious lesions in both primary mandibular second molars with radiolucent periapical lesions were also found (Fig. 1). In total, 20 unerupted teeth (12 supernumerary and 8 of the permanent formula) and two extensively carious teeth (primary mandibular second molars) with periapical radiolucent areas were recorded. The clinical history of the patient showed no data for any local or general illness and no signs or symptoms of an associated syndrome were found. Also, clinical history and clinical-radiological review of the parents and brothers of our patient showed any familial history of ST. First, both carious primary mandibular second molars were

extracted and the underlying bone curetted. Later, under local anesthesia; both upper primary canines and the right supernumerary canine were extracted without complications. Two weeks later, after completion of the blood tests, the patient was admitted in the operating room and all the mandibular supernumeraries were surgically extracted under general anesthesia. The complete list of all 12 supernumeraries extracted is as follows: one right upper canine; one left mandibular molar; three left mandibular bicuspid one left mandibular canine; one right mandibular lateral incisor; one right mandibular canine; three right mandibular bicuspid and one right mandibular molar.

The patient was discharged with no postoperative complications. She was sent to the Pediatric Dentistry Department for evaluation and building prosthetic space maintainer. To date, no new ST have been detected in radiographs taken each 6 months and ten years later, she is in good health wearing her partial prosthesis.

DISCUSSION

In a survey of 200 patients with ST, it was found that 90% of them had a definite genetic influence. Familial occurrence^{11,24,27-29} supports an autosomal dominant trait^{5,20} and inheritance does not follow a simple Mendelian pattern. Case reports support a familial tendency^{14,28-31} and lack of penetrance in some generations has been suggested.²⁹ Because ST appear more frequently in males a sex-linked inheritance was proposed by several authors;³²⁻³⁴ others suggested that ST development is associated to multifactorial inheritance^{35,36} and some proposed that ST is a kind of post permanent dentition.^{18,37}

The most accepted theory is hyperactivity of the dental lamina; it suggests that ST are formed as a result of a disturbance of locally independent conditioned hyperactivity.²⁵ It includes unknown disturbances of this structure with formation of multiple budding which results in additional tooth germs.¹⁶ Also, splitting of a tooth bud with formation of a supplemental tooth is a possibly involved mechanism.¹⁸ Theory of the dichotomy proposes that ST arise from

Table 3: Frequency of supernumeraries by location and gender

Type of Teeth	Location	Boys	%	Girls	%	Total	%
Mesiodens	Maxilla	5	6.76	3	7.69	8	7.08
	Mandible	0	0	0	0	0	0
Incisors	Maxilla	9	12.16	3	7.69	12	10.62
	Mandible	5	5.95	5	12.5	10	8.06
Canines	Maxilla	7	9.46	0	0	7	6.2
	Mandible	8	9.52	0	0	8	6.45
Bicuspid	Maxilla	36	48.65	24	61.54	60	53.1
	Mandible	65	77.38	33	85.2	98	79.03
Molars	Maxilla	9	12.16	3	7.69	12	10.62
	Mandible	2	2.38	2	12.5	4	3.22
Distomolars	Maxilla	7	9.46	6	15.38	13	11.5
	Mandible	4	4.76	0	0	4	3.22
Temporal Incisor	Maxilla	1	1.35	0	0	1	0.88
	Mandible	0	0	0	0	0	0
TOTAL	Maxilla	74	46.83	39	49.36	113	100
	Mandible	84	53.17	40	50.64	124	100



Figure 1. Panoramic radiograph showing the presence of twelve supernumerary teeth in three quadrants.

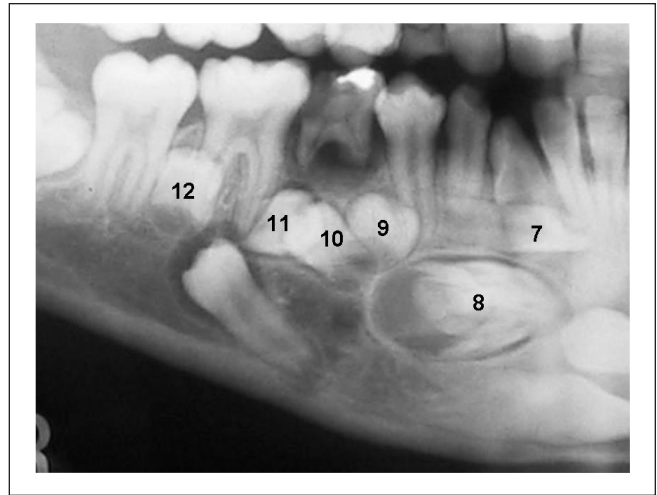


Figure 2. c) Right mandible. Six supernumeraries are observed in this zone.



Figure 2. a) Right maxilla. We can see the presence of an included right upper supernumerary canine.

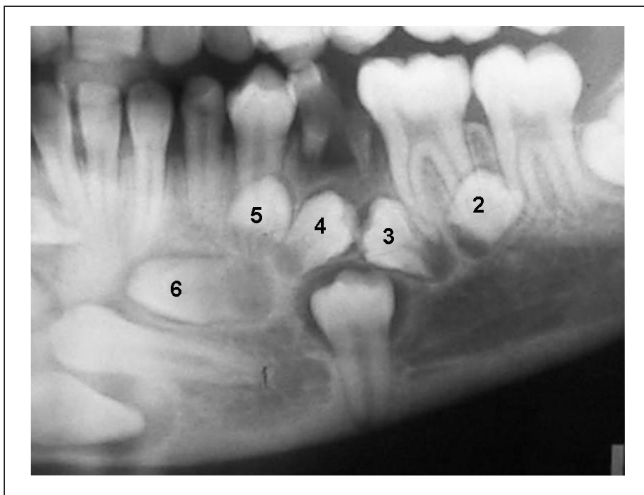


Figure 2. b) Left mandible. Five supernumeraries are observed in this magnified area of the original radiograph.

clumps of epithelium remaining after breaking up of the tooth band that later are activated to tooth formation.²⁶ Atavism is the expression of a series of clinical or pathological features coming from an ancestor who remained hidden during long time or multiple intermediate generations as a result of a chance in recombination of genes and this mechanism is proposed as the cause for development of ST.¹⁵ Trauma has been related to the development of ST since it has been reported that two patients developed ST near to a fracture line³⁸ and experimental evidence supports this possibility.^{26,30,40}

Some studies found that prevalence of ST ranges from 0.1% to 3.8%.^{2,41,42} ST are more frequent in males and it has been reported very variable M:F rates: 2:1;^{43,44} to 9:2¹³ and in the Rajab and Hamdam study, all patients were males.⁴³ The most common ST is the mesiodens followed by bicuspid, lateral incisors or distomolars;^{6,45} but in other series, the most common ST was the distomolar followed by mesiodens and bicuspid.⁴⁶ It has been reported that ST were more common in maxilla.^{1,13,47}

Frequency of multiple ST varies widely from 11.1% to 1%.^{48,49} Yusof¹³ found that multiple ST were more frequent in the mandible (60.9%) and in the Acikgoz *et al* study,¹ a rate of 56.8% for multiple mandibular ST was found. According to several authors, the main location for multiple ST is the upper anterior region followed by molar zone.^{47,50} Nazif *et al*² reported that multiple ST were 14% and all their cases were located in the upper anterior region. In another report Yusof¹³ stated that the most common location for multiple ST was the bicuspid area (62.1%), pointing out that the lower premolar region was a characteristic location for non-syndrome associated multiple ST.

It is well known that ST are diagnosed during routine radiographic examination, for this reason, it is important the radiographic study in all pediatric patients in order to diagnose the presence of unerupted ST and to reduce the risk of complications as delaying or prevention of eruption of the permanent teeth, displacement, root resorption and cystic lesions.^{1,5,7,18,43} We like to point out that the most important risk associated to non-erupted ST is the risk for development of odontogenic cysts and tumors as ameloblastoma, adenomatoid odontogenic tumor and odontogenic keratocystic

tumor (odontogenic keratocyst). We decided to extract all ST present in our patient after several discussion sessions by a team formed by Oral and Maxillofacial Pathologists, Oral and Maxillofacial Surgeons, Pediatricians and Orthodontists. We considered that risks during surgery outweigh the benefits of their removal. But, when clinicians consider that unerupted ST should be left in situ long-term clinical and radiographic monitoring is mandatory.

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