Case Report

Sequential Supernumerary Teeth Development in a Non-Syndromic Patient; Report of a Rare Case

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Abstract

Isolated impacted supernumerary teeth are quite rare, but they can be seen associated with several syndromes such as cleidocranial dysostosis or Gardner's syndrome. This article aims to discuss a case of sequential formation of supernumerary teeth with no other associated disease or syndrome.

A 17-year-old Iranian male with 8 impacted supernumerary teeth was referred to the department of pediatric dental clinic at Shahid Beheshti Medical University in Tehran with a history of several impacted unerupted teeth. Repeated and periodical clinical and radiographic examinations revealed newly formed teeth buds in unusual dental ages. All extra teeth were associated with generalized enamel hypoplasia to some degree on their relative permanent adjacent teeth. The patient did not have any record of a systemic disease or any syndromic condition to relate his dental problem to.

This rare condition involved repeated and continued formation of extra teeth out of the normal numbers and dental age evident in serial radiographs.

Key Words: Tooth, Supernumerary; Serial; Syndrome; Child

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INTRODUCTION

Supernumerary teeth (ST) are defined as an extra set of teeth that are counted more than normal count in the component of primary and/or permanent dentition [1] To date several different theories have suggested varying etiologies; factors including: 1. abnormality or hyperactivity of the dental lamina, [2] reactivation of the remaining epithelium after break up in the tooth band (dichotomy theory) [3], and 3. The process of atavism, the return to or appearance of an ancestral condition or type [4]. In addition, familial tendency and racial influences should also be noted when looking at the cause [5].

The formation of multiple supernumerary teeth (MST) does not follow the normal path in Mandelian pattern [6].

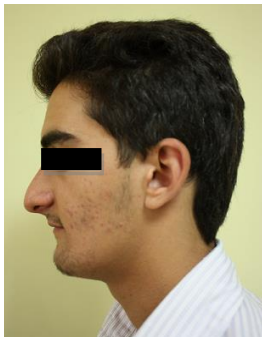

Fig 1. Facial photograph of patient

The anterior maxillary region appears to be the most usual site of occurrence with mesiodense being the most common ST located at and around the palatal midline [7]. The prevalence of ST is reported between 0.3% and 3.8% in America with the male population being affected twice as frequently as the female group [1]. ST may occur in single or multiple patterns, unilateral or bilateral. Multiple ST is counted for 14% of all ST cases, [1] while it is rarely found in individuals with no other associated syndrome [8]. The most known syndrome associated with ST is cleidocranial dysplasia. There are several other syndromes with a lower frequency of association with ST including: Apert syndrome, Gardner syndrome, Down syndrome and Crouzon disease (Table 1 and 2).

The size and shape of ST is different, but in certain cases it is close to the normal adjacent teeth, the so-called supplemental teeth. They may be as variable as those with tuberculates (more than one cusp or tubercle) or even conical (peg in shape) [9]. Several methods have been introduced to classify these extra teeth: 1. Non-sequential in which all ST are developed in a short period of time with no further formation of any tooth bud after a certain age [2]. Sequential where tooth formation will continue for a longer period with newly formed tooth buds appearing in a later stage of life [10, 11-14]. MST has a direct influence on the treatment protocol and its outcome [15].

The purpose of this article was to report a rare case of sequential MST in a non-syndromic Iranian patient.

CASE REPORT

A 16-year-old male patient was referred to the pediatric dental clinic at Shahid Beheshti Medical University, Tehran in fall 2008 with failure of eruption of several upper and lower permanent teeth (Fig 1).

The patient was the second child in his family with no trace and history of any similar condition in other family members. Medical and laboratory test histories revealed no specific medical condition with an overall normal health. The level of intelligence and mental development was assessed as normal based on Wechsler criteria. Physical examination revealed a thin body, slight hyperlaxity in the finger joints raising the possibility of joint problem, later ruled out by a full orthopedic evaluation. Results of a general laboratory test including the thyroid gland function revealed no problem.

The present teeth were as follows in the first visit:

6	E	D	C	B	A	1	B	C	D	E
7	5	4	3	2	1	1	C	5		

Enamel hypoplasia was noted in most of the erupted teeth. Severe caries was noted especially in retained primary teeth. The 1st upper left and1st lower right permanent molar teeth were absent as extracted earlier due to severe caries (Fig 2).

Radiographic evaluation of the chest and wrist showed no sign of abnormality. Panoramic views (three year apart) were also available for evaluation of the patient's development progress (Figure 3, 4 and 5).

4	1	1
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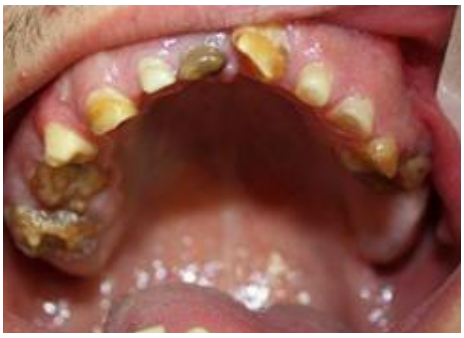
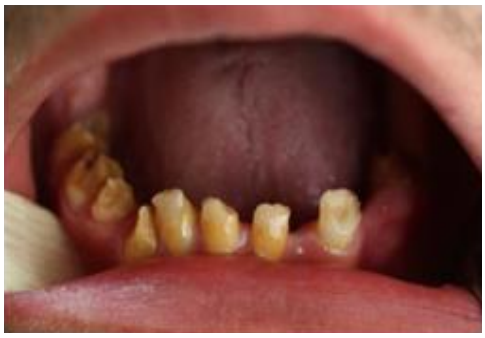

Fig 2. a,b: Intra oral photographs showing present teeth with enamel hypoplasia

The first radiograph was taken when he was 11 years old, at which three supernumerary teeth were located at the upper anterior and right premolar regions namely (Fig 3).

4'	1"	1'	1'	4'	
4'				4'	5'

The second panoramic radiograph was taken at 14 years of age showing four other supernumerary teeth bilaterally in the mandibular premolar and left maxillary premolar areas as indicated below:

	4'		
4'		4'	5'

Interestingly, no signal of any of these teeth could be traced in earlier radiographs.

The patient's history revealed that the right and left upper 6 was extracted due to severe caries (Fig 4).

New supernumerary teeth were observed in the third radiograph taken at 17 years of age that did not have any trace at earlier radiographs.

The locations of these extra teeth were defined between 1 and 1'. This newly appeared tooth was named as 1".

The upper left maxillary central incisor was present at the time of oral examination while the right central and both upper lateral insisors remained unerupted.

In addition, the lower left lateral incisor was also present in the oral cavity (Fig 5).

There was a total number of 8 extra teeth in the jaw. The collective figure of these extra teeth are shown as:

4'	1"	1'	1'	4'	
4'				4'	5'

Consultations were sought from oral surgery and orthodontic departments in order to plan the best treatment option for the patient. An overall agreement was achieved to remove all the malformed and malposed teeth including: several unsavable permanent teeth as listed below:

4		1	B	6	7	
		2	4'	5'	6	7

The treatment process included a phase of multiple extractions under general anesthesia as several surgical sites needed intervention. This was later followed by an initial step of orthodontic forced tooth movement in order to encourage the eruption of impacted teeth including:

32	34
	4

The patient was discharged the day after the operation with his orthodontic visits set along with a full mouth follow up at 6 and 12 months.

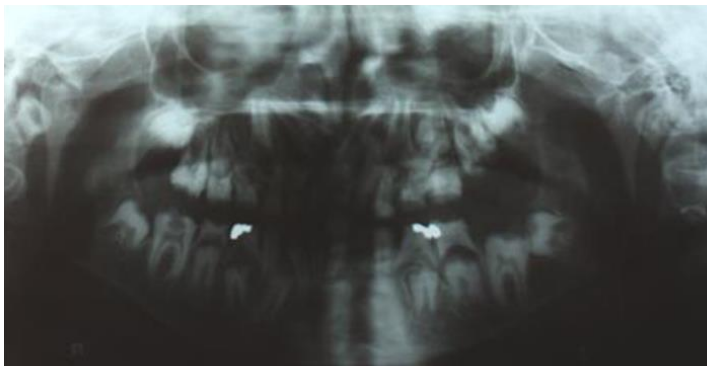

Fig 3. First panoramic radiograph with teeth present

The patient's current status is as close to normal with an appropriately aligned set of teeth in order as shown in Fig 6.

DISCUSSION

Supernumerary teeth are usually seen along with and at the same time as the normal teeth. They can also be seen associated with certain syndromes.

The rate of multiple supernumerary teeth has been reported as 23% among all supernumerary cases [4,16].

An important point is that the development of 5 supernumerary teeth of this report was noticeable over a 5-year period (sequential supernumerary teeth formation.

Multiple nonsequential, non-syndromic supernumerary teeth tend to occur more in the mandibular premolar area, as recently reported [17-21]. (Table 3).

Both premaxillary and mandibular premolar regions were equally affected in the current case, followed by maxillary premolar region with a lower incidence (less affected teeth). Gomes et al. (2008) reported three cases of sequential supernumerary teeth in mandibular premolar and premaxillary regions considered as the most frequent areas for these teeth to be detected respectably. The maxillary premolar region was, however, the site least affected. Supernumerary teeth of the current case were more frequent in themaxilla (five in the maxilla and 3 in the mandible) as seen in other reports [9].

Non-syndromic multiple supernumerary teeth are more likely to occur in patients whose other relatives are also involved [2]. However, the patient in the current case did not have a positive family history similar to the case reported by Gomes et al. (2008) [9].

Table 1. Classic syndrome with multiple supernumerary teeth[3]

Syndrome's Name	Clinical Manifestation
Apert syndrome	Scaphocephaly,craniosynostosis,bilateral syndactyl,midface hypoplasia
Cleidocranial dysplasia	Aplastic clavicles,frontal bossing,hypoplastic midface
Gardner syndrome	Osteomas,epidermoid cysts,odontomas,intestinal polyps
Down syndrome	Brachycephay,mental retardation,epicanthal folds
Crouzon disease	Craniosynostosis, exophtalmos, hypoplastic midface
Sturge-weber syndrome	Angiomatosis and calcification of leptomeninges,seizures,port-wine nevi of face
Oral-facial-digital syndrome	Hypoplastic alar cartilage, cleft tongue,clinodactyly
Hallermann-streiff syndrome	Dyscephaly,mandibular hypoplasia,hypotrichosis

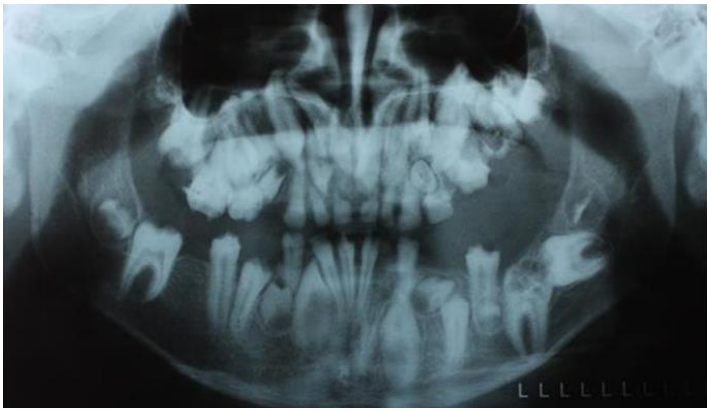

Fig 4. Second panoramic radiograph, appearance of new teeth buds

Continuous supernumerary teeth formations are thought to be due in part to reactivation of the tooth follicle with no clear explanation.
Another possible mechanism is unresorbed remained dental lamina. This could lead to reactivation of the normal permanent teeth at the time of crown completion. Lastly it is thought that the crypts of the supernumerary teeth may have been present but were not detectable on earlier radiographs [2].
The Wnt gene has been considered another likely etiology for supernumerary teeth [22].
Jarvinen et al. (2006) stated that Wnt signaling has been associated with tooth renewal in the human. Tooth bud expressing stabilizes beta-catenin (intracellular mediator of Wnt signaling) in the epithelium, leading to the formation of dozens of teeth.
Wang et al. (2009) stated that even adult dental tissues could lead to the formation of new teeth in response to either loss of function of epithelial APC (adenomatous polyposis coli gene protein) or Wnt/beta-catenin activation [22].

Table 2. Rare syndrome with multiple supernumerary teeth and their characteristics

Syndrome's Name	Clinical Manifestation
Robinow[9]	Short stature,hypertelorism,high nasal bridge,midface hypoplasia,micropenis
Ehler-Danlos[10]	Skin hyperelasticity,Articular hypermobility,Vascular fragility
Nance-Horan[11]	Congenital cataract, Malformed teeth
G/BBB[12]	Eye anomalies, Laryngotracheoesophageal cleft, Congenital heart disease, Genitourinary anomalies , Gastrointestinal disorders
Zimmermann-Laband[13]	Gingival fibromatosis, Absent or dysplastic distal phalanges, Vertebral defects, Hepatosplenomegaly, Ear, Nose, Bone, and Nail defects
Leopard[14]	Multiple lentigines, Ocular hypertelorism, Delayed secondary sexual characteristics, Mild cardiac abnormalities
Enamel Dysplasia with Hamartomatous atypical Follicular Hyperplasia(EDHFH)[15]	(only In black south Africans)Hamartomatous follicular hyperplasia,Central odontogenic fibroma(WHO type),Enamel Dysplasia
Tricho-Rhino-Phalangeal[16]	Slowly growing sparse hair, Medially thick and laterally thin eyebrows, Bulbous tip of the nose, Long flat philtrum , Thin upper lip with vermilion border, Protruding ears, Cone-shaped epiphyses
Kabuki make up[17]	Cleft lip/palate high-arched palate, Bifid tongue and uvula, anterior open bite, overretention of primary teeth, Long palpebral fissure, hypertelorism, palpebral ptosis, broad, depressed nasal root with a flat nasal tip, mild neuropsychomotor developmental deficits

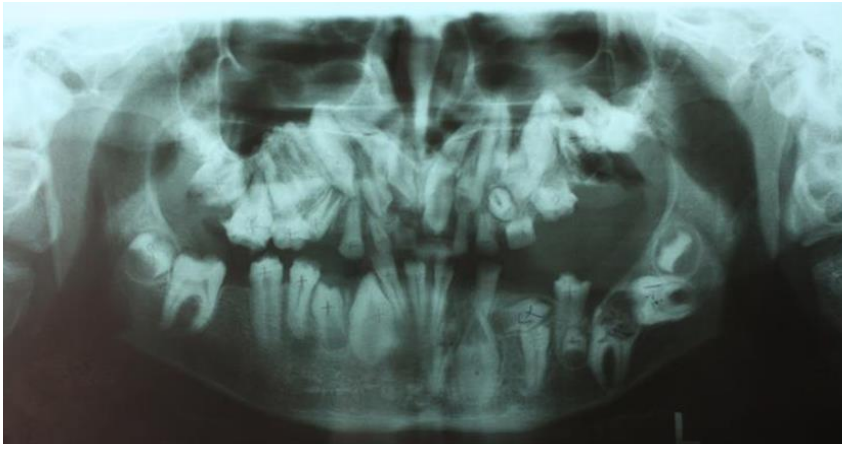

Fig 5. Third panoramic radiograph showing further teeth formation at this stage

It is concluded that a small number of APC deficient cells is enough to induce the surrounding wild-type epithelial and mesenchymal cells to participate in the formation of new teeth [23].

The presence of general enamel hypoplasia of supernumerary and permanent teeth was another important finding in the current case with no report of such findings by other investigators. The presence of multiple supernumerary teeth would routinely suggest an underlying genetic disorder [22, 23]. It is important to mention that some eruption disturbances may be seen with the presence of supernumerary teeth.

Any orthodontic force effect needs to be fully investigated in such individuals.

When supernumerary teeth are found a prompt decision should be made in order to decide their removal. Uncontrolled removal of supernumerary teeth could lead to further complications affecting adjacent structures. It is therefore important to determine if the risks of surgery outweigh the benefits of removal. Due to the high number of supernumeraries and impacted teeth, a stepwise therapy was planned in this case with extraction of the selected number of supernumerary teeth being the starting point followed by forced eruption as the next phase.

Table 3. Recently Reported non Syndromeical Multiple Supernumerary teeth

Author	*year*	*Age / Gender*	*Number*	*location*
Batra et al[8]	2004	17/ girl	11	One mesiodens,two upper premolar,two lower anteriors,six lower premolars
Arathi & Ashwini[17]	2005	14/ boy	12	One lower anterior, five lower premolars, three upper anteriors,three upper premolars
Sivapathasundharam & einstein[18]	2007	20/ boy	14	Nine lower premolars,three upper premolars,one upper anterior, one lower anterior
Aravindha &Srivatsa[19]	2007	19/ girl	10	One mesiodens, one upper anterior,two lower canines,,four lower premolars,two lower distomolars(one in each side
Rasi Beyer G[20]	2008	21/ boy	5	One upper anterior, Four upper premolar
Diaz et al[21]	2009	20/ girl	17	twelve upper(premolars and distomolar), five lower premlars

CONCLUSSION

A case of repeated development of several supernumerary teeth was noted with no history of any syndrome or medical condition. Periodical routine radiographic images confirmed the case as a rare but non-syndromic individual with sequential supernumerary teeth formation. Treatment includes elimination of these supernumeraries in order to help retain the teeth and the surrounding structures.

REFERENCES

1- Taner T, Uzamis M. Orthodontic treatment of a patient with multiple supernumerary teeth and mental retardation. J Clin Pediatr Dent. 1999 Spring;23(3):195-200.

2- Brook AH. A unifying aetiological explanation for anomalies of human tooth number and size. Arch Oral Biol. 1984;29(5):373-8.

3- Foley MF, Del Rio CE. Supernumerary teeth. Report of a case. Oral Surg Oral Med Oral Pathol. 1970 June;30(7):60-3.

4- Rajab LD, Hamdan MA. Supernumerary teeth: Review of literature and a survey of 152 cases. Int J Pediatr Dent. 2002 July;12(4):244-54.

5- Zhu JF, Marcushamer M, King DL, Henry RJ. Supernumerary and congenitally absent teeth: A literature review. J Clin Pediatr Dent. 1996;20:87-95.

6- Brook AH, Elcock C, Al-Sharood MH, .McKeown HF, Khalaf K, Smith RN. Further studies of a model for the etiology of anomalies of tooth number and size in humans. Connect Tissue Res. 2002;43(2-3):289-95.

7- Pinkham JR, Casamassimo PS, Mctigue DS. Pediatric dentistry, infancy through adolescence. 4th Ed. St. Louis, MI: Elsevier; 2005. p. 61-2, 263.

8- Batra P, Duggal R, Parkash H. Non-syndromic multiple supernumerary teeth transmitted as an autosomal dominant trait: case report.J Oral Pathol. 2005 Nov; 34(10):621-5.

9- Gomes CO, Jham BC, Souki BQ, Pereira TJ, Mesquita RA. Sequential supernumerary teeth in nonsyndromic patients: report of 3 cases. Pediatr Dent. 2008 Jan/Feb;30(1):66-9.

10- Moore SR, Wilson DF, Kibble J. Sequential development of multiple supernumerary teeth in mandibular premolar region:Aradiographic case report. Int J Pediatr Dent. 2002 Mar;12(2):143-5.

11- Nuvvula S, Kiranmayi M, Shilpa G, Nirmala SV. Hypohyperdontia: Agenesis of three third molars and mandibular centrals associated with midline supernumerary tooth in mandible. Contemp Clin Dent. 2010 Jul-Sep;1(3):136-41.

12- 12 Stevenson W, McKechnie AD. Recurring supernumerary teeth: report of a case. Oral Surg Oral Med Oral Pathol. 1975 Jul; 40(1):76-80.

13- Pasha Z, Choudhari S, Rathod A,. Sulabha AN. Bilaterally impacted mandibular supernumerary premolars associated with unusual clinical complications. J Pharm Bioallied Sci. 2013 Apr-Jun; 5(2): 166-9.

14- Panetta G, Favaretti F, Cerulli Mariani G, Gatto R. Multiple and relapsing supernumerary teeth associated with dens invaginations: Review of the literature and case report . Minerva Stomatol 2005 May; 54(5):321-32.

15- Tanaka E, Kawazoe A, Nakamura S. An adolescent patient with multiple impacted teeth. Angle Orthod. 2008 Nov;78(6):1110-8.

16- Fernandez MP, Valmaseda CE, Berini AL, Gay EC. Retrospective study of 145 supernumerary teeth. Med Oral Patol Oral Cir Bucal. 2006Jul;11(4): E339-44.

17- Arathi R, Ashwini R. Supernumerary teeth: A case report. J Indian Soc Pedod Prev Dent. 2005Apr;23(2):103-5.

18- Sivapathasundaram B, Einstein A. Non-syndromic multiple supernumerary teeth: Report of acase with 14 supplemental teeth. Indian J Dent Res. 2007 Aug;18(3):144

19- Sirvatsan P, Aravindha Babu N. Mesiodens with an unusual morphology and multiple impacted supernumerary teeth in a non-syndromic patient. Indian J Dent Res. 2007

Aug;18(3):138-40.
20- Rasit Beyer G, Ortakoglu K, Sencimen M. Multiple impacted teeth: report of 3 cases. Eur J Dent. 2008 Jan;2(1):73-8.
21- Diaz A, Orozco J, Fonseca M. Multiple hyperdontial: report of a case with 17 supernumerary teeth with non syndromic association. Med Oral Cir Bucal. 2009 May;14(5):299-31.
22- Järvinen E, Salazar-Ciudad I, Birchmeier W, Taketo MM, Jernvall J, Thesleff I. Continuous tooth generation in mouse is induced by activated epithelial Wnt/beta-catenin signaling. Proc Natl Acad Sci U S A. 2006 Nov;103(49):18627-32.
23- Wang XP, Lund JJ, O'Connel DJ, Saadi I. Apc inhibition of Wnt signaling regulates supernumerary tooth formation during embryogenesis and throughout adulthood. Development. 2009 Jun;136(11):1939-49.