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Non-Syndromic Multiple Supernumerary Teeth in a child – Management with Literature Review

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Abstract

Multiple supernumerary teeth are rare and mostly associated with syndromes. Aetiology is not fully understood but environmental and genetic factors have been implicated and different theories have been postulated. When supernumerary teeth are present, they pose problems for eruption of normal tooth and development of the jaw. In this report, the case of a non-syndromic child with multiple supernumerary teeth is discussed. Most of the supernumerary teeth were unerupted and seen as incidental findings in the radiographs. A periodic multidisciplinary approach is essential for the successful management of children with super numerary teeth. The report also summarizes the documented dental management in non- syndromic children (<14 years) from a scientific literature search strategy advocated between 1950-2021.

Keywords: Multiple super numerary teeth, non-syndromic child, Hyper dontia, Cone-Beam Computed Tomo graphy, Preventive dentistry.

Introduction

Supernumerary teeth (ST) are formed from tooth germs in excess of normal number, located in any arch regardless of their location, morphology and can be found in both permanent and primary dentition.[1] They may appear as unilateral/bilateral, single / multiple, erupted / impacted or deformed/normal in shape and size. Presence of ST may lead to complications like

malocclusion, midline diastema, delayed or failure of eruption of permanent teeth, periodontal problems, cyst formation and root resorption of adjacent teeth.[1] Multiple ST are usually syndromic and associated with syndromes like Ehlers-Danlos syndrome, Gardner's syndrome, Cleidocranial dysplasia, Ellis-van Creveld syndrome, Cleft lip and palate, Anderson- Fabry disease etc. [2]

ST are most commonly seen in maxilla compared to mandible when one or two supernumerary teeth are present.

Multiple supernumerary (>5) when present are seen most commonly in bicuspid area. So et al., reported single supernumerary tooth occurs in 76%-86%, two supernumeraries in 12%-23% and multiple ST (>5) in <1% of population.[3]

The pathogenesis of ST is not clear and different hypothesis have been proposed by different authors considering the environmental and genetics factors. Studies have shown that genes and signalling pathway responsible for tooth morhogenesis include Want, FGF, BMP, Shh, TNF and RUNX2; however, mechanism of tooth formation in ST still remains unclear. Studies on mice have shown Want and shh activity in dental lamina responsible for the occurrence of ST in them. Massink et al., revealed that human with no systemic syndrome and Lrp6 mutant have ST and Takahashi et al. conducted a whole sequencing in Japanese population with ST without syndromes and found newer genes associated with this condition.[4]

This report describes the dental management of a unique case of multiple ST in a non-syndromic child. It also summarizes the reported scientific incidence and management of ST in the non-syndromic Pediatric age group below 14 years from the last 70 years.

Case report

A 12-year-old male patient reported to the department of Pediatric and Preventive Dentistry with the complaint of additional teeth present in the anterior maxillary region that was aesthetically unpleasing (Figure 1A). The parent explained that the child covered his mouth whenever he had to smile or laugh and tried his best to avoid any social gatherings. However, the patient had no complain of pain, discomfort or infection from the region.

On clinical examination, the dentition was at the mixed stage and two ST were visibly located behind the permanent central incisors (Figure 1B). Karyotyping was withheld as there was no abnormal development in the individual and the familial history was unremarkable. The patient had severe halitosis during oral examination and the oral hygiene index (OHI) by Silness-Loe revealed poor scores.

An orthopantomogram (OPG) was advised to rule out the presence of more ST. The OPG (Figure 2) revealed the presence of 10 ST. In the maxilla and mandible there were five ST each. Cone Bean Computed Tomography (CBCT) was advised thereafter to rule out the spatial position of all ST. CBCT revealed all ST positioned palatally/lingually to the corresponding tooth in the dental arches (Figure 3A, Figure 3B, Figure 3C). In the upper right quadrant, ST were located adjoining the premolar and adjacent to central incisor. In the upper left quadrant, ST was observed adjacent to the central incisor, canine and premolar. ST located palatal to the upper central incisor had exact resemblance to the upper centrals with complete root formation. However, the other ST did not resemble normal tooth structure and root formation was incomplete.

In the mandible, two ST were present in the right premolar region resembling crown - like structure without complete root formation. In the left quadrant,

there were three ST in the premolar region that also resembled the tooth crown structure without root completion.

Based on the dental findings, familial history and absence of any systemic disorder/syndrome in the child, the possibility of a suitable differential diagnosis was excluded; reaffirming the final impression of Non-Syndromic Multiple Supernumerary Teeth (NSMST).

As the first measure to good oral hygiene maintenance, dental prophylaxis was performed on his first day of visit to the Department of Pediatric and Preventive Dentistry. Orthodontic referral and consultation were planned to decide the fate of existing ST and correction of malaligned teeth. The formulated treatment plan following referral included extraction of the primary mandibular right second molar [Tooth #T] for alignment of permanent mandibular right first premolar [Tooth #28] and extraction of the 2 visible supernumerary teeth palatal to the upper central incisors under local Anesthesia. Other ST were decided to be left untouched as they were asymptomatic, and did not hinder in occlusion.

The extraction of the 2 ST palatal to the upper central incisors was planned and performed under local Anesthesia. As the ST in concern were partially erupted and palatally positioned in less than 0.5-0.9mm to permanent maxillary right central incisor and permanent maxillary right central incisor, the extraction turned out difficult. There was minimal space to engage the upper anterior forceps (Forceps #1102, A-TITAN).

However, following adequate elevation of the mucosal tissues, both the ST were carefully removed without traumatizing the anteriorly positioned central incisors (Figure 4). Post-operative healing and granulation were uneventful and was achieved within the 2-weeks. Following, the extraction of tooth primary mandibular right second molar was carried out and the patient was reassured to use his left quadrant for mastication for the next 2-3 days. The orthodontic intervention has been initiated and is currently under progress. Following orthodontic alignment, long-term follow-up is required to observe for changes in position/development of supernumerary teeth that may alter the stable occlusion achieved in the future.

Discussion

Occurrence of NSMST is rarely cited in literature with the most common location being the mandibular premolar region. The most accepted theory is hyperactivity of the dental lamina suggesting formation of ST as a result of disturbance in independent conditioned hyperactivity. However, the other proposed theories include phylogenetic theory, dichotomy theory and Genetic factors.[3]

Recently many studies done on mice model give insight to genetic factors responsible for the ST. Development of ST was closely related to the signalling pathway of Want, Shh, BMP and FGF signalling. Shh signal is present in dental placodes for early tooth germ development, and Shh over activation leads to ST development. BMPs are also present in developing tooth and present between its epithelium and mesenchyme, BMP4 is responsible for development from bud to bell stage. Msx1 and Shh signal were over activated and extra teeth were formed with the deletion of BMP4. In Wnt pathway, inactivation of USAG1, which binds to Wnt co-receptors Lrp5 and Lrp6, can lead to the overexpression of Wnt signalling and therefore the development of supernumerary teeth. In Runx2 pathway, excess transcription factor Twist1 activated FGF signalling to give rise to supernumerary teeth.[4]

A search was conducted in Scopus, Cochrane, Medline, Google Scholar and Web of Science databases for

literature cited through May 2021. Literature search keywords were 'Supernumerary Teeth', 'Non-Syndromic Multiple Supernumerary Teeth' and 'Hyperdontia'. The published reports were selected independently by two investigators. The title and abstract were pre-screened to exclude the ineligible reports. The retrieved case reports were read well before inclusion in this review. Differences if any between the two investigators were resolved by discussion. All articles were searched using English keywords. However, no restriction were placed on the publication language. A total of 35 case reports were finally listed and a review of clinical findings and course of management in the cases of NSMST teeth has been listed briefly in Table 1 and 2.

The prevalence of ST have shown differences among the racial and cultural groups. In primary dentition, for Caucasians it was between 0.1% to 1.8%, for Japanese 0.1% and Chinese children in Taiwan 7.8%. In permanent dentition Caucasians had prevalence of 0.4% to 2.1%, 2.4% southern Chinese, 3.4 % for Japanese and 6 % in American blacks.[3]

The most common ST are the mesiodens followed by bicuspids, lateral incisors or distomolars; but in a different report, the most common ST was the distomolars followed by mesiodens and bicuspids.[5] Studies by Yusof and Acikgoz et al., found that multiple ST were more frequent in the mandible with a frequency of 60.9% and 56.8% respectively.[4]However, other studies report the commonest location for multiple ST to be the upper anterior region followed by molar area.[4] Nazif et al., in a report mentions of 50 patients with 14% prevalence of multiple ST that were located in the upper anterior region.[5] However, based on the review of cited literature in this report, the authors confirm that the most common location of ST was the mandibular premolar area followed by the maxillary premolar area, mandibular and maxillary molar region and in certain cases, the incisor and canine region were also involved.

Alvira Gonzalez and Gay-Escoda has reported this disorder to occur in both genders on an equal basis with no difference in its location.[6] However, Rajab and Hamdan found that males show a higher prevalence of ST than females with a sex ratio of 2:1.1. The bilateral presence of supernumerary teeth was another feature reported by the co-workers.[6]

ST may lead to disturbed tooth eruption, tooth rotation, bodily displacement, crowding, spacing or diastema, dilacerations, root resorption of normal teeth. Halitosis in the current report may have been due to poor oral hygiene maintenance due to the crowded dentition with supernumeraries. Diagnosis of such occurrences is usually incidental and most commonly by using an OPG. Mallineni recommends the use of CBCT for better clinical assessment.[3]

The management of ST has mostly been extraction or periodic observation, if not associated with any pathology. Munns recommends extraction to be carried out at the earliest opportunity for better prognosis. Rotberg and kopel, Hog strum and Andersson advise to intervene only until the root formation of adjacent teeth is complete. Other authors, Koch et al., Scanlan and Hodges, Garvey et al., and Shah et al., suggested to avoid immediate removal if no pathology is associated with ST. Omer et al., suggest the optimal time for removal of ST to be 6-7 years for anterior teeth, as minimal disturbance occurson adjacent teeth.[3]

Conclusion

Clinicians should thus possess sound knowledge on the potential ramifications involving multiple ST. Early diagnosis and a tailor-made treatment plan is essential in

the successful management of cases of NSMST. Of utmost importance is a periodic recall and review, both clinically and radiographically, to exclude the possibility of future malocclusions.

Table 1: Reported cases of NSMST in children up to 14 years [1950 -2005]

Author	Year	Country	Age	Sex	Number of supernumeraries teeth	Course of management
Berendt [3]	1951	Israel	12	M	Maxilla – 5 I, 1 Pm	Extraction of ST in multiple visits followed by orthodontic alignment
Poyton et al. [4]	1960	Canada	11	F	Maxilla - 2 Pm Mandible – 6 Pm	Extraction of ST over a span of 5 years
Ruhlman, Neely [2]	1964	USA	14	M	Maxilla – 6 I Mandible – 3 Pm	Extraction of ST (except supernumerary maxillary central incisor) that was left as it was less carious than the permanent teeth
Stevenson [1]	1964	Scotland	12	M	Maxilla – 5 Pm Mandible – 3 Pm	Management not disclosed
Santangelo MV [7]	1968	USA	14	F	Maxilla – 4 Pm Mandible – 3 Pm	Management not disclosed
Barnett [8]	1974	UK	12	M	Maxilla – 2 M Mandible – 2 Pm, 2 M	Extraction of all ST
Stevenson, McKechnie [6]	1975	Scotland	10	M	Maxilla – 5 I, 1 Pm Mandible – 5 Pm	Extraction of ST under General Anesthesia followed by orthodontic alignment
Shusterman et al. [9]	1978	USA	7	F	Maxilla – 2 I, 2 Pm Mandible – 2 Pm	Management not disclosed
Becker et al. [1]	1982	Israel	12	M	Maxilla – 3 Pm Mandible – 2 Pm	Pedodontics, orthodontist and Oral surgeon were part of team from preventive procedure, exposure of permanent teeth or supernumerary teeth extraction and final alignment by an Orthodontist.
Ohmori et al. [8]	1984	Japan	11	F	Maxilla – 2 C, 2 Pm Mandible – 4 Pm	Extraction of all ST
Kantor et al. [1]	1988	USA	9	M	Maxilla – 4 Pm Mandible – 4 Pm	Surgical extraction of ST followed by space maintenance
King et al. [8]	1993	Hong Kong	13	F	Maxilla – 3 Pm Mandible – 3 Pm	Management not disclosed
Mason et al. [8]	1996	UK	12	F	Maxilla – 2 Pm, 1 Dm Mandible – 4 Pm	Extraction of mesially located supernumeraries, while distal were left in situ
Rizzuti, Scotti [8]	1997	Italy	10	M	Maxilla – 4 I, 5 Pm, 1 M Mandible – 4 I, 5 Pm, 2 M	Extraction of all teeth including deciduous in 2 stages - First mandible, followed by maxilla after 2 months. Complete denture prosthesis was given, followed by orthodontic treatment
Sharma [5]	2001	India	12	F	Maxilla – 3 I, 4 Pm Mandible – 2 I, 2 Pm	Extraction of all ST teeth except in relation to 22 which was converted to canine followed by orthodontic alignment
Arcuri et al. [5]	2002	Italy	9	F	Maxilla – 1Me, 3 Pm, 2 M, 2 Dm Mandible – 1 I, 4 Pm	Extraction of ST over period of years
Guthua SW [6]	2002	Kenya	14	M	Maxilla – 4 Pm Mandible – 4 Pm	Case series showed extraction of all ST under general Anesthesia followed by orthodontic alignment
			13	F	Maxilla – 2 Dm Mandible – 5 Pm	
Mopager et al. [8]	2002	India	13	M	Maxilla – 4 Pm Mandible – 3 Pm	Surgical extraction of all ST
Manrique Mora et al.[6]	2004	Spain	12	M	Maxilla – 1 I, 1 Pm Mandible – 3 Pm	First visit extraction of lower primary molar followed by space maintainer for the eruption of lower ST that was extracted later. Next visit, lower left supernumerary was extracted. Extraction of other ST was planned for later appointments.
Miura et al. [9]	2004	Japan	14	F	Maxilla – 1 Pm Mandible – 7 Pm	Management not disclosed
Arathi, Ashwini [5]	2005	India	14	M	Maxilla – 1 I, 4 Pm Mandible – 2 I, 5 Pm	Extraction of all ST followed by orthodontic alignment
Ramsaran et al.[10]	2005	Jamaica	14	F	Maxilla – 5 Pm Mandible – 7 Pm	Extraction of one erupted ST and long-term follow up for other ST

###Me - Mesiodens I – Incisors, C – Canine, Pm – Premolar, M – Molar, Dm – Distomolar,

Table 2: Reported cases of NSMST in children up to 14 years [2006-2021]

Author	Year	Country	Age	Sex	Number of supernumerary teeth	Course of management
Nayak, Mathian [5]	2006	India	13	M	Maxilla – 1 I, 1 Pm, 1 in anterior region Mandible – 1 C, 1 Pm	Extraction of upper ST, while for other supernumeraries, no treatment was rendered as it was asymptomatic
Hyun et al. [6]	2008	South Korea	13	M	Maxilla – 2 Pm Mandible – 4 Pm	Prevalence study, management not disclosed
Brauer [11]	2010	Germany	14	M	Maxilla – 4 I, 2 Pm, 2 Dm Mandible – 5 Pm	Surgical extraction under general anaesthesia (GA) in 2 stages, followed by orthodontic alignment
Ferriol et al. [6]	2011	Spain	12	F	Maxilla – 1 I, 2 Pm, 2 Dm Mandible – 1 I, 6 Pm, 2 Dm	Management not disclosed
Ledesma et al. [5]	2012	Mexico	12	F	Maxilla – 1 C Mandible – 1 I, 2 C, 6 Pm, 2 M	Extraction of upper ST under LA and the remaining under GA
Ansari et al. [12]	2013	India	14	M	Maxilla – 4 Pm Mandible – 4 Pm, 5M	Extractions followed by delivery of a functional space maintainer, fixed space maintainer bilateral spring open coil and lingual arch in subsequent visits.
Santos JD et al. [13]	2014	Brazil	12	F	Maxilla – 2 Pm Mandible – 6 Pm, 2 M, 4 in anterior region	Extraction in a single intervention under GA
Bozkurt et al. [14]	2015	Turkey	10	F	Maxilla – 2 Me, 1 I, 4 Pm Mandible – 4 Pm	Extractions done under GA, removable space maintainer followed by orthodontic alignment
			13	M	Maxilla – 1 C, 1 Pm	
					Mandible – 4 Pm	
Katsu Takashi et al. [4]	2016	Japan				Case series, management not
			10	M	Maxilla – 1 Pm, 2M	discussed
					Mandible – 1 Pm, 1M	
Riberio MC et al. [15]	2017	Brazil	13	F	Maxilla – 2 Pm, 1 M, 2 Dm Mandible – 2 I, 5 Pm, 2 Dm	Surgical extraction of all ST
			14	M	Maxilla – 2 I, 3 Pm	
					Mandible – 6 Pm, 1 M	
Daniel Rey et al. [16]	2019	Colombia				Case series reported surgical
			14	M	Maxilla – 2 I, 3 Pm, 1 M	extraction of all ST under GA
					Mandible – 2 I, 7 Pm	
Priyanka, Sagar [17]	2019	India	14	M	Maxilla – 4 Pm Mandible – 3 Pm	Management not discussed
Jayaraman [18]	2021	Malaysia	10	M	Maxilla – 52 denticles posterior maxilla	Surgical extraction of all ST under nitrous oxide followed by orthodontic traction of impacted maxillary canine

*I – Incisors, #C – Canine, **Pm – Premolar, ##M – Molar, ***Dm – Distomolar, ###Me - Mesiodens

Figure legends

Figure 1: A. Intraoral anterior view in occlusion; B. Maxillary teeth with two palatally erupted ST

Figure 2: Pre-operative OPG showing multiple ST. The arrows show ten ST in both jaws.

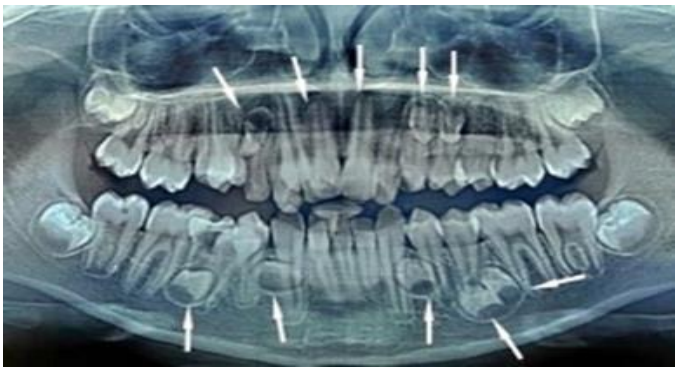
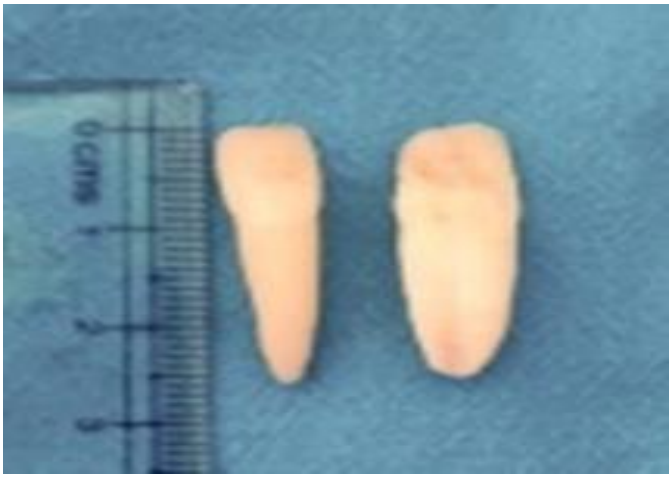


Figure 3: CBCT showing the presence of ST in the: A. Upper anterior palatal region; B. Right; C. Left quadrants of maxilla and mandible

Figure 4: Extracted supernumerary teeth of the upper anterior palatal region



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