INVITED ARTICLE

Anomalies of Tooth Number in the Age Range of 2–5 Years in Nonsyndromic Children: A Literature Review

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Abstract

Aim and objective: Dental anomalies of number predispose the teeth to malocclusion, caries, periodontal problems, and often compromise aesthetics and function. We present a literature review to assess the distribution of these anomalies and observe associated anomalies of the underlying permanent successor teeth in the age range of 2–5 years in nonsyndromic children.

Review: A literature search was conducted using the "PubMed" database with a manual search of cross-references, published in the years 2000–2018. The following key words were used: "dental anomalies, primary teeth" "hypodontia," "oligodontia," and "hyperdontia." Of the 1,232 records accessed, 41 articles were included in the final review—34 articles (42 clinical cases) and 7 cross-sectional studies.

Results: The overall prevalence of dental anomalies of number ranged from 1.8 to 4.0%. Among all the clinical cases, a predilection for boys (26/42) was observed. Hyperdontia was the most common primary tooth number anomaly; 23.8% cases (10/42) had additional coexisting primary tooth anomalies. In 35.7% cases (15/42), anomalies of the permanent successor teeth were present of which permanent tooth agenesis was most commonly seen in 75% cases of hypodontia (3/4) and 85.7% cases of oligodontia (6/7).

Conclusion: Among all the anomalies, hyperdontia was most common. The anomalies of tooth number were more prevalent in boys and in the maxilla. Apart from dental anomalies of number, those of shape and size also concurrently occurred in the primary dentition and in the permanent successor teeth, stressing upon early diagnosis, radiographic examination, and long-term follow-up visits.

Keywords: Deciduous, Dentition, Permanent, PubMed, Tooth.

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Introduction

A number of dental anomalies occur due to a disruption in the initiation stage of odontogenesis. The etiology is complex and often an interplay of genetic and environmental factors.[1] Although commonly reported in syndromes, these anomalies also exist in nonsyndromic cases.[2,3]

Anomalies that affect the total complement of teeth either show an increase or decrease in number. Anodontia means a complete absence of teeth. Congenital absence of one or more teeth is termed hypodontia, and an absence of ≥6 teeth is termed oligodontia (except third molars).[1] There are some conditions that mimic hypodontia. These include pseudo-anodontia or clinically missing teeth due to impaction and false anodontia or missing teeth following exfoliation or extraction.[3] A detailed history and a thorough clinical and radiographic examination helps differentiate these from true cases of hypodontia. In contrast, hyperdontia refers to those exceeding the normal complement of teeth. Apart from hyperdonts seen in the alveolar bone, extra-teeth occur in extra-gnathic locations such as buccal mucosa or nose and are termed accessory or ectopic teeth.[1]

Anomalies of tooth number can affect either primary or permanent dentition but are often less reported in the former.[4] This may be attributed to the protective environment provided in the prenatal life for development of primary teeth.[4] However, this may also be due to fewer children in this age-group visiting dental clinics.

Anomalies of tooth number often compromise esthetics, function, and occlusion. Apart from dental anomalies of number, those of shape and size such as taurodontism,[5] fusion,[6] dilaceration,[7] talon cusp,[8,9] and peg-shaped teeth may also coexist with these anomalies.[10] In some cases, the underlying permanent dentition may also be involved.[10–13] Hence, it is important to diagnose and detect these anomalies early in the primary dentition.

The aim of the present review is to determine the distribution of anomalies of tooth number and observe associated anomalies in the underlying permanent teeth in the age range of 2–5 years in nonsyndromic children.

Materials and Methods

A review of English literature was conducted for articles indexed in "Pubmed": published between the years 2000 and 2018. The "Pubmed" database was searched along with a manual search of cross-references. The following key words were used: "dental anomalies, primary teeth" '"hypodontia," "oligodontia," and

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"hyperdontia." Those with systemic diseases, syndromes, beyond the age and date range, mentioned in letters, editorials, books, reviews, dissertations, and monographs were excluded.

A total of 1,232 records were accessed (Flowchart 1). Of these, 817 articles were excluded because of syndromes and other anomalies. A total of 305 duplicates were removed. Abstracts of the remaining 110 potential articles were checked for relevance. A total of 75 articles were inaccessible, irretrievable, or did not meet the inclusion criteria and were not considered. Full texts of the remaining articles were evaluated by three independent reviewers, and disagreement, if any, was resolved after consensus. Six new articles were retrieved and added through cross-references. The final list comprised of 41 scientific papers: 7 cross-sectional studies and 34 articles or 42 clinical cases (Flowchart 1).

The type of tooth anomaly, year of publication, country, chief complaint, age, gender, ethnicity/race, method of examination, affected tooth, complications/findings, family history, and treatment were recorded.

Literature Review and Discussion[5–45]

In our review of cross-sectional studies, the prevalence of dental anomalies varied from 1.8 to 4% in the age range of 2–5 years (Table 1).[14–20] The prevalence was less than that reported by Chen et al. (5%)[46] and Yonezu et al. (7.2%)[47] but greater than the ones reported by Whittington and Durward (1.4%)[48] and Magnusson (1.7%).[49] The reasons for this wide range could be differences in genetics, ethnicity, or methodology. In our review, Deolia et al.[20] reported the highest prevalence of 4%. The findings could have been overestimated as the study was conducted in patients visiting the pediatric dental clinic for some dental-related problem whereas others had conducted random examinations in nurseries and schools. Second, these studies employed different methods of examination such as only clinical,[15,17,19,20] a combination of clinical and radiographic examination,[14,18] or radiographic examination with a plaster cast.[16] Studies based only on clinical examination may have resulted in underreporting of anomalies.

The present review did not observe any significant difference in dental anomalies at different ages.[15,17] However, Deolia et al.[20] observed a significantly greater prevalence in those aged 3 years when compared to 2 years, citing complete eruption of primary teeth by that age. The presence of anomalies at age 2 years, stresses upon the need to create awareness among the parents to be vigilant, and report at an early age for dental visits.

The distribution of dental anomalies and gender predilection is controversial. Although Kramer et al.[15] and Gomes et al.[18] observed no gender difference, yet other studies observed significantly more anomalies in boys[17] and in girls.[20] Such differences may be a consequence of metabolic, environmental, genetic, or individual variations.

Hypodontia

Hypodontia (Table 1), oligodontia, and anodontia are often used to signify the number of missing teeth. A synonym for hypodontia is "tooth agenesis." The latter seems a more appropriate word, and other terms such as "anodontia," "hypodontia," and "oligodontia" are best suited for classification.[50]

Hypodontia is often genetically inherited, and the mode of transmission is autosomal dominant, recessive, or X-linked.[1] A multifactorial etiology, involving an interplay of genetic and environmental factors, is implicated.[1,3] Further, this condition may also be in association with certain syndromes such as Down's syndrome, ectodermal dysplasia, and cleft lip and palate.[1]

Hypodontia is uncommon in the primary dentition with a prevalence of <1%.[1] In the present review, although most studies reported a prevalence comparable to these findings,[15,17–20] Sacal et al.[14] and King et al.[16] reported a high prevalence of 4.8% and 4.1%, respectively. Differences may exist because of study methodology. Both studies were retrospective and conducted in patients who had visited dental clinics previously. However, there may be variations according to region, ethnicity, and genetics.

Hypodontia usually involves anterior teeth, a finding also observed by other authors.[15–17] Either the maxilla or the mandible is involved, and a simultaneous occurrence in both the arches is rare. Missing teeth are often unilateral in distribution.[15,19] Kramer et al.[15] observed hypodontia in eight patients of which six were unilateral. The present review of clinical cases observed that of the four cases

Flowchart 1: Tooth number anomalies in the age range of 2–5 years

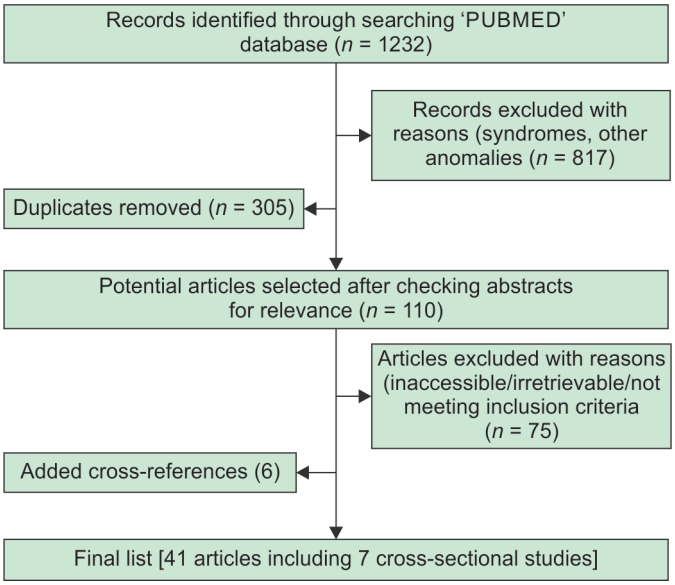

Table 1: Cross-sectional studies and cases of dental anomalies of tooth number

S.no.	Author, year and country of publication	Age, sample size, ethnicity/race (E/R)	Chief complaint or study population and method of examination—C, clinical; R, radiographic	Overall prevalence OP hypo-hypodontia hyper–hyperdontia in% (n), findings according to age, gender, race (AGR)	Involved teeth (FDI notation) and associated anomalies of other primary teeth	Associated anomalies of permanent teeth	Family history	Treatment
Cross-sectional studies (7)								
1.	Sacal et al., 2001[14] USA	3–5 years, 500 E/R NM	1st 500 children examined previously at a pediatric dental clinic at Texas-Houston (maxillary occlusal R)	OP-NM Hypo-4.8 (24) Hyper-0.2 (1) AGR-NM	Could not be determined	Missing teeth with no successors 1.0 (5) and with successors-4.0 (19)	—	—
2.	Kramer et al., 2008[15] Brazil	2–5 years, 1,260 Brazilian children of white and non-white races	28 public nurseries in the city of Canoas, South Brazil (C)	2.5 (32) Hypo 0.6 (8) Hyper 0.3 (4) AGR-NS	Hypo-6 unilateral, 2 bilateral (5 ULI, 7 LLI, 2 CI) Hyper-3 UA, 1 LA (mostly max ant region)	NA	—	—
3.	King et al., 2008[16] China	5 years, 936 Hong Kong children	Randomly selected sample (plaster casts, panoramics; straight anterior occlusal X-ray where needed)	OP-NM Hypo 4.1 Hyper 2.8 AGR-NM	Max LI and mand I	NM	—	—
4.	Kapdan et al., 2012[17] Turkey	2–5 years, 1,149 Turkish children	12 nurseries in the city of Sivas, Turkey (C)	2.0 (23) Hypo 0.2 (2) Hyper 0.3 (3) A-NS; G-Sig. >in boys, R-NM	Predilection for premaxilla	NA	—	—
5.	Gomes et al., 2014[18] Brazil	2–5 years, 1,718 Brazilian children	Residing in 20 nursery schools in Federal district of Brazil (C and R) Control group-OPG of those without dental anomalies	OP-1.8 (31) Hypo/tooth agenesis 0.29 (5) Double tooth and tooth agenesis 0.12 (2) Control group-1.07% (1) Hyper-0.29 (5) Control group-mesiodens (2) A-NM; G-NS; R-NM	Hypo-LI (4 max, 1 mand) Double teeth (mand LI, canine) and tooth agenesis (contralateral LI) Hyper-4 max LI, 1 mand LI	54.8 (17) Hypo-80.0 (4)-all primary max tooth agenesis had agenesis of permanent successors and 1 primary mand unilateral tooth agenesis had a normal dentition Double teeth and tooth agenesis 100.0 (2) had bilateral mandibular LI agenesis Hyper 60.0 (3)-max supernumerary had a permanent supernumerary LI (2 cases) but no anomaly in mand supernumerary tooth	—	—

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6.	Lochib et al., 2015[19] India	3–5 years, 1000 E/R NM	One school in Faridabad (C)	OP-NM Hypo 0.4 (4) Hyper-Not assessed AGR-NM	Hypo-3 mand CI (1 bilateral) Hypo-Not assessed	NA	—	—
7.	Deolia et al., 2015[20] India	2–5 years, 1398 E/R NM	Visiting pediatric dental clinic at Jodhpur Dental hospital (C)	OP-4.0 (56) Hypo 0.64 (9) Hyper-0.36 (5) A-sig >at 3 years than 2 years; G-sig> in girls; R-NM	Hypo-5 unilateral, 4 bilateral (6 upper arch, 3 lower arch) Hyper-1 unilateral, 4 bilateral (3 upper arch, 2 lower arch)	NA	—	—
Hypodontia (4 articles, 4 cases)								
1.	Pinho et al.,[21] Portugal, 2005	3 year, gender NM, Portuguese	16,771 OPGs screened of archival cases of 3–71 year olds (relevant 1) C and R	—	52, 62	12, 22 and one mandibular incisor missing	NM	NM
2.	Swinnen et al.,[10] Belgium, 2008	5 year F, Caucasian	Congenitally missing deciduous and permanent teeth C and R	—	52, 62; peg-shaped 71, 72, 81, 82 with interdental spacing	12, 22, 13, 23, 31, 41, 32 missing	Father-35, 45 missing; mother's one sister-peg-shaped 12, 22; females from father's side had oligodontia	NM
3.	Surendar et al.,[5] India, 2013	5 year M, Indian	Pain, swelling in mandibular right lower back tooth C and R	—	52; Taurodontism: 74, 84	12 missing	FH –ve	Carious 84 extracted; band and loop space maintainer
4.	Anthonappa and King[22] Australia, 2016	4 year F, Southern Chinese	Missing teeth C and R	—	53, 63, 73, 83	All permanent teeth present except 3rd molars	FH –ve	NM

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Oligodontia (7 articles, 7 cases)								
1.	Shashikiran et al.,[23] India, 2002	3-year-old M, Asian	Missing primary teeth C and R	—	52, 53, 62, 63, 71–73, 82–83	12, 22, 31, 32, 41, 42 missing	FH –ve	Long-term preventive and prosthodontic management
2.	Venkataraghavan et al.,[24] India, 2007	4-year-old M, Asian	Missing primary teeth and improper speech C and R	—	51–54, 61–64, 71–75, 81–85	All permanent teeth present with defective dentin and root formation-16, 26, 36, 46, 31, 41	FH –ve	Complete dentures fabricated with openings for 55, 65
3.	Shilpa et al.,[25] India, 2007	2.5-year-old M, E/R NM	Several missing teeth C and R	—	52–54, 62–64, 71–74, 81–84	31, 32, 41, 42 missing All permanent first molar teeth visible	FH –ve	Restoration of carious 51, 61 and partial dentures for missing teeth
4.	Shilpa et al.,[11] India, 2010	2.5-year-old F, Indian	Several missing teeth C and R	—	51–54, 61–64, 71–72, 81–84	Absence of all permanent teeth except 16, 26, 36, 46	FH –ve	Removable partial denture recommended
5.	Moses et al.,[26] India, 2013	3-year-old M, Indian	Missing primary teeth C and R	—	52, 53, 62, 63, 71–73, 75, 81–83,85 at age 3 years 75, 85 erupted at age 6 years	At age 3 years-absence of 12–15, 22–25, 31–35, 41–45 At age 6 years-developing 24, 34, 44, 15, 25, 45, 47 At age 8 years-developing 37, 47 at an unusual site	NM	Removable partial denture
6.	Correia et al.,[27] Brazil, 2013	4-year-old F, E/R NM	Esthetic concern due to missing mandibular incisors C and R	—	52, 62, 71, 72, 81, 82	Absence of 12, 22, 31, 32, 41, 42, 15, 25, 35, 45	NM	Adhesive partial denture
7.	Zhang et al.,[28] China 2015	2-year-old 9 month M, Chinese	Many missing teeth C and R	—	51–54, 61–64, 71–74, 81–84	Absence of all permanent teeth except 16, 26, 36, 46	NM	removable partial denture

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Hyperdontia (16 articles, 21 cases, 25 supernumerary teeth)								
1.	Miyoshi et al.[6] Japan, 2000 conducted a survey on 8122 Japanese kindergarteners, aged 3–6 years, from cities of Fukuoka and Sasebo in Kyushu, Japan (relevant 3)	5-year-old M, Japanese	Routine dental checks C	—	Supplemental tooth between 51, 52 resembling 52	NA	NM	NM
		5-year-old M, Japanese	Routine dental checks C	—	Bilateral supplemental tooth between 51, 52 and between 61, 62 resembling lateral incisor with a cusp	NA	NM	NM
		5-year-old M, Japanese	Routine dental checks C	—	Supplemental tooth between 61, 62 resembling 62	NA	NM	NM
2.	Aguilo L et al.,[12] Spain, 2001	3-year-old F, Caucasian	Routine dental checks C and R	—	Triple tooth (fusion of 61, 62 with supernumerary tooth)	All permanent successors present	FH −ve	Fractured following trauma and was extracted
		2-year-old M, Caucasian	Abscesses above right maxillary triple tooth C and R	—	Triple tooth (fusion of 51, 52 with supernumerary tooth)	Permanent successor lateral incisor missing	FH −ve	Extracted
3.	Lehl and Kaur[29] India, 2002	5-year-old M, E/R NM	Pain in upper front tooth C and R	—	Cone-shaped, short-rooted mesiodens between 51, 61	NM	NM	Extracted
		5-year-old M, E/R NM	Tooth erupting behind upper front tooth C and R	—	supplemental tooth resembling primary lateral incisor palatal to 51, 52	NM	FH +ve (mother, maternal grandmother)	Extracted
4.	Tatel,[30] USA, 2003	3-year-old 3 month F, White	'loose' front tooth C and R	—	Mesiodens erupted beneath 61 and had caused its root resorption	No other abnormalities	NM	After 61 exfoliated, mesiodens was recontoured
5.	Chevitarese et al.,[31] Brazil, 2003	4-year-old M, Brazilian	Referral after diagnosis of supernumerary tooth C and R	—	Mesiodens erupted in 51 area, causing pathologic resorption of 51	Permanent successors present	NM	Mesiodens restored with celluloid crown and composite

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6.	Yeung et al.,[7] Hong Kong, China, 2003	2-year 5-month-old M, Chinese	Unerupted 51 C and R	—	Unerupted inverted conical supernumerary crown around developing crown of 21 Dilaceration with compound odontome impeding eruption of 51	Permanent successors present	−ve for unerupted teeth/ hypodontia	Removal of 51, 61, supernumerary tooth and compound odontome
7.	Ray et al.,[32] India, 2005	4-year-old F (Bengali) Indian	Odd appearance of teeth C and R	—	Conical mesiodens between 51 and 61	NM	FH −ve	Mesiodens extracted
8.	Roberts et al.,[33] UK, 2005	22-month-old M, Caucasian	Referral because of habitual biting of inanimate objects and occasional siblings C	—	Erupted unilateral supernumerary primary maxillary right lateral incisor and bilateral supernumerary primary maxillary canines	NA	FH −ve	Regular follow-up
9.	Siraci et al.,[8] Turkey, 2006	3.5-year-old M, E/R NM	Carious tooth (had undergone cleft lip and palate surgery) C and R	—	Partially erupted, rotated supernumerary tooth between 61 and 62, interfered with occlusion Talon on facial and palatal part of crown	NM	NM	Supernumerary tooth extracted
10.	Batra et al.,[9] Sweden, 2006	5-year-old F, E/R NM	Routine checks (H/o cleft lip repair, present cleft alveolus) C and R	—	Unerupted supernumerary tooth distal to 61. Talon cusp: facial talon cusp wrt 62 that was mesially titled	Very small peg-shaped permanent lateral incisor	FH −ve	No immediate intervention
11.	Webb and Unkel[34]USA, 2007	5-year-old F, White	Emergency visit due to fall at a skating party C	—	Fractured 51, 52, 61, 62, carious 53, 63 incidental finding of mesiodens between 51 and 61	NA	NM	53, 63 restored and 51, 52, 61, 62, mesiodens extracted
12.	Raupp et al.,[35]Brazil, 2008	5-year-old M, E/R NM	Referred for removal of supernumerary teeth R	—	2 supernumerary teeth lingual to tooth germs of 11, 21	Tooth germs of 11, 21 present	NM	Surgically removed
13.	Bahadure et al.,[36] India, 2012	5-year-old M, E/R NM	Routine dental checks C and R	—	Rotated mesiodens between 51, 61	21 present	NM	Regular follow-up

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		4-year-old F, E/R NM	Unerupted lower front teeth C and R	—	71, 72, 73, 81, 82, 83 embedded mesiodens in the lower arch	Missing 31, 32, 33, 41, 42, 43	FH –ve	Supernumerary tooth extracted
14.	Shilpa and Nuvvula,[37] India, 2013	5-year-old M, E/R NM	Abnormal upper front tooth C and R	—	Supernumerary tooth between 61, 62 Fusion: triple teeth showing fusion of 61, 62 and supernumerary tooth	Agenesis of 22, central incisor normal	FH –ve	1 year follow-up
15.	Mohan et al.,[38]India, 2014	5-year-old M, E/R NM	Reported with cleft upper lip since birth C and R	—	Supplemental tooth between 61, 62	NM	FH –ve for clefts and congenital anomalies	6 months follow-up; referred for cleft repair
16.	Indira et al.,[39] India, 2014	5-year-old F, E/R NM	Unaesthetic smile C and R	—	Molariform mesiodens between 51, 61	11, 21 developing	NM	Carious teeth 51, 61 and mesiodens extracted
Hypohyperdontia (3 articles, 4 cases)								
1.	Miyoshi et al.,[6]Japan, 2000 conducted a survey on 8122 Japanese kindergarteners, aged 3–6 years, from cities of Fukuoka and Sasebo in Kyushu, Japan (relevant 1)	3-year-old M, Japanese	Routine dental checks C and Cast analysis	—	82 missing; Supernumerary between 62, 63, wider than lateral incisor, with a cusp; Fusion 71, 72	Not assessed	NM (Not mentioned)	NM
2.	El-Bahannasawy and Fung,[40] Glasgow, UK 2004	4.5-year-old M, Caucasian	Failure of 53 to erupt C and R	—	53 missing; erupted supplemental tooth 54	Supplemental premolar in upper right quadrant seen at age 12 years	FH –ve	Supplemental 54 extracted at 7 years to relieve crowding

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3.	Anthonappa et al.,[13] Hong Kong, China 2008 (records of children visiting the pediatric dental clinic between 2005–2007 were identified. Out of 7 cases of hypohyperdontia, 2 were relevant)	5-year-old M, E/R NM	Referred for management of erupted mesiodens C and R	—	71, 81 missing; erupted mesiodens in the maxillary region	42 missing	NM	NM
		5-year-old F, E/R NM	Regular dental checks C and R	—	72 missing; inverted mesiodens in 21 regions	32 missing	NM	NM
Ectopic hyperdontia/odontogenic choristoma/accessory teeth (5 articles, 6 cases, 7 teeth)								
1.	Lee,[41] Taiwan, 2001 (a review of 13 cases of intranasal teeth treated in the department of a Taiwan hospital in patients aged 4–39 years, relevant 2)	4-year-old F, E/R NM	Could not be determined C and R	—	11 mm intranasal supernumerary tooth in the floor of the left nasal cavity. Mucosal cover −ve	NM	NM	Extracted under rigid endoscope
		5-year-old F, E/R NM	Could not be determined C and R	—	7 mm intranasal supernumerary tooth in the floor of the left nasal cavity. Mucosal cover +ve	NM	NM	Extracted under rigid endoscope
2.	Noroozi and Arora[42] USA, 2011	2-year-old M, E/R NM	Swelling inside the mouth C and R	—	2.5 × 2 cm tooth-like lesion in the right buccal mucosa which had a bony stalk fused to the right zygomatic arch. All primary teeth present	NM	FH −ve	Excisional biopsy through intraoral approach
3.	Nagarajappa and Manjunatha[43] India 2011	4-year-old M, E/R NM	Pain, discomfort during swallowing for 1 week C	—	Supernumerary molar-like structure posterior to soft palate in the oropharynx. All primary teeth present	NM	NM	Tooth fell off on its own within 10 days

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4.	Liu et al.,[44] China, 2013	4-year-old F, E/R NM	2 teeth erupted from buccal mucosa C and R	—	Two molariform teeth found in left buccal mucosa CBCT revealed 5 developing or developed teeth in the left cheek surrounded by osseous structure fusing with left zygomatic arch. All primary teeth present	All permanent tooth germs present	NM	Surgery advised but refused due to financial constraints
5.	Ogane et al.,[45] Japan, 2016	2-year-old M, Japanese	No subjective symptoms C and R	—	Inverted supernumerary tooth in the right nasal cavity, about 10 mm in length. Crown resembled deciduous canine. All primary teeth present	All permanent tooth germs present	NM	Extracted using mosquito clamp

NM, not mentioned; NA, not assessed; NS, not significant; I, incisor; ULI, upper lateral incisor; LLI, lower lateral incisor; CI, central incisor; UA, upper arch; LA, lower arch; FDI, federation dentaire internationale

of hypodontia, three were bilateral (2 involved maxillary lateral incisors and one involved maxillary and mandibular canines),[10,21,22] and one was unilateral.[5]

In hypodontia, often, a maxillary lateral incisor or a mandibular incisor is involved. This finding was also depicted in our review, where three of the four cases reported a missing maxillary lateral incisor.[5,10,21] Kramer et al.[15] reported that of the 14 missing teeth, 12 were lateral incisors and 2 were central incisors. Kapdan et al.[17] reported missing mandibular central incisors in all the three cases. Congenital agenesis of primary molars, canines, and maxillary central incisors is very rare.[22] A rare case of hypodontia with bilateral involvement of canines in both the arches was also found in our review.[22]

Hypodontia may not exist as a single entity. Other dental anomalies of size and shape, such as double teeth, microdontia, and taurodontism, may coexist in the primary dentition.[50] Gomes et al.[18] observed a simultaneous occurrence of double teeth with tooth agenesis in two cases. The present review observed taurodontism[5] and peg-shaped teeth/microdonts[47] in cases of hypodontia, and interestingly, both teeth were of the opposing arch. A more frequent occurrence of taurodontism in permanent first molars was found in association with hypodontia (29%) when compared to controls (10%).[50] According to Brook,[51] both microdontia and hypodontia were genetically related and if a tooth bud failed to reach a specific size and thresholds of number, it would not develop. This finding justifies the need to perform a complete dental examination.

Hypodontia may impair mastication, esthetics, and occlusion and, importantly, may be associated with missing permanent successor teeth, ectopic eruption, short roots, enamel hypoplasia, over-retained primary teeth, and delayed tooth development. Gomes et al.[18] observed that although hypodontia of primary maxillary teeth had missing permanent counterparts, yet unilateral hypodontia of primary mandibular teeth had a complete permanent dentition. The finding was consistent with our review, where all missing primary maxillary lateral incisor teeth had a missing permanent successor.[5,10,21]

Sacal et al.[14] observed agenesis of permanent tooth in 20.8% (5/24) cases of hypodontia. Gomes et al.[18] observed similar findings in 80% (4/5) cases, and if associated with double teeth, agenesis could be as high as 100% (2/2). In bilateral hypodontia of primary teeth, agenesis of permanent successors is 100%, and this is consistent with the reports by Ravn,[52] Gellin,[53] and Gomes et al.[18] This finding was also evident in our review, where bilateral hypodontia of primary maxillary lateral incisors had missing permanent counterparts.[10,21] However, our single case of bilateral hypodontia of primary canines, in both maxilla and mandible, had all the permanent successors present.[22]

Oligodontia

Oligodontia (Table 1) may be attributed to a viral infection during pregnancy, genetics, metabolic imbalances, and developmental abnormalities.[11] In addition, environmental factors (especially

maternal), trauma, infection, radiation, syndromes, and idiopathic causes have been implicated.[11,27]

Oligodontia impairs development of bone height of maxilla and mandible, causes alveolar ridge resorption, decreases lower facial height, and affects speech, function, and esthetics.[11,27,28] The primary lateral incisor, mandibular left central incisor, and mandibular left lateral incisor were notably absent in all our cases, compromising speech, function, and esthetics.[11,23–28]

Oligodontia is often associated with ectopic eruption, delayed eruption, rotations, spacing, or more commonly, missing permanent successor teeth.[11,23–28] Ravn[52] observed that in aplasia of primary teeth, agenesis of permanent teeth occurs in 80% cases. In our review, four of the seven cases had all missing permanent successors and missing additional permanent teeth.[11,26–28] Delayed tooth development, extending over years, was observed in one case, recommending long-term follow-up visits.[26] In two cases, few permanent successors were missing[23,25] of which one had an additional permanent tooth missing.[23] In our one case,[24] all permanent successor teeth were present which was consistent with the observations by Ooshima et al.[54] An important finding was that permanent first molars were evident in all our cases; these were either unaffected[11,23,25–28] or had defective roots.[24]

Hyperdontia

Supernumerary teeth or hyperdonts (Table 1) are teeth in addition to the normal complement. Several etiological theories are proposed such as atavism, dichotomy of the tooth germ, and local hyperactivity of dental lamina, the last one being the most popular.[55]

The present review on hyperdontia reported a prevalence of 0.20–2.8%.[14–18,20] The findings are comparable to the previous reports of 0.07–0.6%,[47,48,52] but the present review also observed a high occurrence of 2.8% in 5-year-old south Chinese children.[16] Differences in study design, ethnicity, and/or genetics could be some of the implicating factors. Nevertheless, a gender predilection was observed among all our 21 clinical cases (Table 2) with more males being affected when compared to females (14 vs 7).[6–9,12,29–39]

Hyperdontia is most prevalent in the anterior maxilla, and lateral incisor is often involved.[1,52,53] These findings were also confirmed in our review, where of the 21 cases, 20 were in the anterior maxilla[6–9,12,29–39] and 1 in the anterior mandible.[36] Of the 25 supernumerary teeth, 12 were in the lateral incisor region,[6,8,9,12,29,33,37,38] 6 in the midline,[29,32,34,36,39] 5 in or around the central incisor region,[7,30,31,35] and 2 in the canine region.[33] Such teeth are often unilateral, but a frequent bilateral occurrence is also reported. In the present review, 16 cases had a unilateral/midline distribution[7–9,12,29–32,34,36–39] and 3 had a bilateral distribution.[6,33,35]

Supernumerary teeth may be of different shapes, but conical form is the most common.[7,29,32] Supernumerary teeth resembling normal morphology of teeth are termed supplemental teeth and are almost always erupted. This was consistent with our observation, where all the nine supplemental teeth had erupted.[6,29,33,38] Kapdan et al.[17] observed that hyperdonts in the central incisor area are peg-shaped and those in the lateral incisor area are normal. This was further confirmed by Saarenmaa.[56] The present review observed nine supplemental teeth,[6,29,33,38] of which six resembled maxillary lateral incisors,[6,29,33] two resembled maxillary canines,[33] and in one case, the shape of the tooth was not mentioned.[38] The rest presented as conical mesiodens in eight cases,[7,29–32,34,36] triple teeth in three cases,[12,37] and a molariform shape in one case.[39]

The association between racial preference and hyperdontia is controversial. A predilection for occurrence in non-White races was highlighted by Kramer et al.,[15] yet a study by King et al.[16] observed no racial difference. However, the Japanese children reported a very low prevalence (0.05%)[6] when compared to the Chinese (0.44%),[57] Caucasians (0.64%),[58] and natives of Finland (0.4%).[59] Miyoshi et al.[6] observed a significant difference among the Japanese and Chinese despite both being of Mongoloid ancestry. Again, both ethnicity and race are confusing terminologies and often used interchangeably.[15] More such studies should be conducted for any conclusive evidence.

Supernumerary teeth are frequently associated with crowded or spaced dentition, midline deviation, failure of eruption, ectopic eruption, derangement of occlusion, dental caries along the line of fusion, periodontal problems, or an unesthetic appearance.[1] In some cases, hyperdontia coexists with other dental anomalies in the primary dentition. In our review, fusion was detected in three cases, all of which were triple teeth.[12,37] Talon cusp was detected in two cases, both of which, incidentally, were cleft patients.[8,9] Oligodontia coexisted with hyperdontia in one case[36] and in the other with compound odontoma and dilaceration.[7]

Hyperdontia of primary teeth may be associated with anomalies of the successor permanent teeth.[48,52,53] Nik-Hussein and Majid[60] reported a corresponding supernumerary permanent tooth in 35–60% cases. These findings were confirmed by Gellin[53] in 63% cases. According to the latter, a supplemental successor tooth, a supernumerary permanent tooth, or hypodontia of the permanent successor may result.[53] The permanent successor teeth may be associated with failure of eruption or ectopic eruption, rotation or displacement, dilaceration, or cyst formation. Gomes et al.[18] also found that the chances of finding a supernumerary tooth in the successors was 60% (3/5 cases) if a similar condition occurred in the primary teeth. In their study, hyperdontia of primary maxillary tooth was associated with a supernumerary permanent lateral incisor in two cases, but this was not so in hyperdontia of primary mandibular tooth where no anomaly was present. The present review of cases in Table 2 showed that all successors were present in 8 cases,[7,9,12,31,35,36,39] missing in 3,[12,36,37] and not mentioned/not assessed in 10 cases.[6,8,29,32–34,38] An important observation was that among all the three cases of maxillary triple teeth involving a fusion of a central incisor, lateral incisor, and a supernumerary tooth,[12,37] all the permanent successors were either present or had a missing permanent lateral incisor (Table 1).

Miyoshi et al.[6] reported four cases of supernumerary teeth of which one case had associated hypodontia. In our review, we considered the latter case under the category of "hypohyperdontia" because of a simultaneous occurrence of both hypodontia and hyperdontia in the same patient. A rare co-occurrence of hyperdontia with oligodontia was reported by Bahadure et al.[36] The present review considered this rare case under the category of "hyperdontia" because of limitations in the existing classification. This case highlights the need for another subclassification of dental anomalies of number.

Hypohyperdontia

Hypohyperdontia (Table 1) are two rare developmental anomalies seen concomitantly in the same patient. This anomaly was first described in 1967 by Camilleri[61] as "concomitant hypodontia and hyperdontia." However, Gibson in 1979 discarded the word "concomitant" and replaced it with "hypohyperdontia."[62]

Table 2: A summary of the reviewed 34 articles (42 clinical cases)

Type of anomalies (number of cases)	*Gender*			*Arch affected*			*Associated anomalies of primary teeth*	*Permanent successor teeth*			*Family history*		
	Boys	*Girls*	*NM*	*Maxilla*	*Mandible*	*Bimaxilla*		*Associated anomalies*	*Presence of successors*	*NA/NM*	*FH +ve*	*FH −ve*	*NM*
Hypodontia (4)	1	2	1	3	—	1	Peg shaped (1), taurodontism (1)	Agenesis (3)	1	—	1	2	1
Oligodontia (7)	5	1	1	—	—	7	—	Agenesis (6)	1 all present but with defective dentin and root formation	—	—	4	3
Hyperdontia (21)	14	7	—	20	1	—	Fusion (3), dilaceration with compound odontoma (1), talon cusp (2), oligodontia (1)	Agenesis (3)	8 of which 1 was peg-shaped	10	1	9	11
Hypohyperdontia (4)	3	1	—	1	—	3	Fusion (1)	Agenesis (2), supernumerary (1)	—	1	—	1	3
Ectopic hyperdontia (6)	3	3	—	—	—	—	NM (2)	—	2	4	—	1	5
Total (42)	26 (61.9%)	14 (33.3%)	2 (4.8%)	24	1	11	10 (23.8%)	15 (35.7%)	12 (28.6%)	15 (35.7%)	2 (4.8%)	17 (40.5%)	23 (54.8%)

Ranta[63] suggested that hypohyperdontia could be the result of altered migration, proliferation, and differentiation of neural crest cells or result from faulty epithelial–mesenchymal interactions during the initiation stage of tooth development.[59] Syndromic involvement is also reported.[64]

Anthonappa et al.[13] reported a prevalence ranging from 0.002% to 3.1%[65,66] in their compiled review. Gender differences were not found, but in their presentation of seven cases, five (71.4%) were males. The authors felt that since both the genders were affected by this mixed numerical defect, one must exercise caution in concluding that gender affects hypohyperdontia. A similar predilection for males was observed (3 vs 1) in the present review.[6,13,40]

The present review showed four cases of hypohyperdontia in the anterior teeth.[6,13,40] In three cases, mandibular incisor was the most common missing tooth (lateral incisors in two cases[6,13] and a bilaterally missing central incisor in one case).[13] One rare case of hypodontia involved a primary maxillary canine.[40]

The findings of clinical cases showed that in contrast to hypodontia, hyperdontia typically involved maxillary teeth in all the cases. Of the four cases, two were mesiodens (1 erupted and 1 inverted),[13] one was a supplemental tooth 54[40] and the fourth one was a supernumerary tooth between 62 and 63 (wider than a lateral incisor, with a cusp).[6]

Our review showed that hypohyperdontia in the primary dentition coexisted with fusion involving a central incisor and lateral incisor in one case.[6] The present review of compiled cases observed that the associated anomalies of permanent teeth were present in 3/4 cases (Table 2). These were missing mandibular right lateral incisor in two cases,[13] and a supplemental premolar in one, detected later at age 12 years.[40] The latter highlights the need for long-term regular follow-up visits (Table 1).

Ectopic Hyperdontia

The word "ectopic" means an abnormal location, and an ectopic tooth refers to a tooth in locations other than the alveolar bone (Table 1). "Choristoma" refers to a growth of normal histology in an abnormal location, composed of tissues derived from 1 or 2 germ layers.[42] The term "osseous choristoma" is a bony growth within the soft tissues of the oral cavity.[67] One such case was reported by Noroozi and Arora,[42] where tissues of both ectodermal (enamel) and mesodermal origin (dentin, pulp) were present in the buccal mucosa, with a bony stalk fused to the zygomatic arch. Since the endodermal tissues were not involved, the term used was "odontogenic choristoma."[42] The etiology is unknown and may be related to entrapment of embryonal tissues *in utero*.

Ectopic hyperdontia is attributed to idiopathic or genetic causes, displacement of the tooth due to trauma, cyst, or infections.[41] A high frequency is seen in syndromes and cleft lip and palate cases.[41]

Teeth have been detected in ectopic areas such as the ramus of mandible,[68] maxillary sinus,[69] nasopharynx,[70] nasal cavity,[45] and oropharynx[43] to name a few. The present review of six cases observed that ectopic teeth were most prevalent in the nasal cavity (3),[41,45] followed by buccal mucosa (2),[42,44] and oropharynx (1)[43] with no significant gender predilection.

The ectopic teeth, if allowed to remain, may have local symptoms and/or serve as a nidus for infection. Intranasal teeth can cause epistaxis, nasal obstruction or discharge, oronasal fistula, abscess, sinusitis, deviated septum or perforation.[41] However, these may also be asymptomatic and remain undetected, particularly if covered by nasal mucosa.[41] The present review of intranasal teeth showed either no subjective symptoms (1 case)[45] or could not be determined (2 cases) because the latter was not specified according to age.[41] Lee[41] observed that of the 13 patients aged 4–39 years, surgically treated for intranasal supernumerary teeth, 5 had no symptoms, 5 presented with nasal obstruction, and 3 had rhinorrhea. Ectopic teeth in buccal mucosa were either erupted[44] or presented with a localized swelling at the site,[42] and one in the oropharynx had pain and discomfort while swallowing.[43]

Ectopic teeth are usually unilateral. Bilateral distribution or occurrence of multiple teeth in one area is rare. The present review reported all unilateral and single cases[41–43,45] except for one which had five molariform teeth on the left cheek of which two had erupted and three were developing.[44]

An ectopic tooth can be a supernumerary, primary, or a permanent tooth. The former is usually deformed and peg shaped.[71] Smith et al.[72] reported that of his 27 intranasal cases, 17 were supernumerary, 2 primary, and rest were permanent teeth. Lee[41] observed that of his 13 surgical cases of intranasal teeth, 11 were supernumerary and 2 were permanent teeth. In our review, all the ectopic teeth (intranasal, buccal, and oropharyngeal locations) were supernumerary teeth.

Our literature reported that the length of an intranasal ectopic tooth was 7–11 mm[41,45] and that of an ectopic molar tooth in the buccal mucosa was 2.5 × 2 cm.[42] In two cases, the shape was molariform.[43,44] In the third case, although the shape was not mentioned, the computed tomography (CT) scan and excisional biopsy specimen showed resemblance to a molar.[42] Of these three cases, two had a bony stalk/osseous structure fused with the zygomatic arch and were reported/suspected as odontogenic choristoma.[42,44] Of the three intranasal teeth, one resembled a primary canine[45] and for the others, shape was not specified.[41]

Extraction of an ectopic tooth is done to relieve symptoms and minimize complications. However, in select cases, extraction of an intranasal tooth may be delayed till root completion of a permanent tooth.[73] When required, removal of this tooth under endoscopic guidance provides for better illumination, visualization, and precise dissection.[41] In the present review, three intranasal teeth were extracted using a rigid endoscope in two cases[41] and a mosquito clamp in the third.[45] For the molariform teeth, excisional biopsy was conducted in one case,[42] the other fell off after 10 days,[43] and surgery could not be conducted for financial reasons in the third case.[44]

To conclude (Table 2), tooth number anomalies were reported in 61.9% or 26 boys when compared to 33.3% or 14 girls. Hyperdontia was the most common primary tooth number anomaly in 2–5-year-old nonsyndromic children. In all, 23.8% (10/42) cases had additional primary tooth anomalies coexisting in the same patient. In 35.7% (15/42) cases, anomalies of permanent successor teeth were observed of which permanent tooth agenesis was seen in 75% (3/4) cases of hypodontia and 85.7% (6/7) cases of oligodontia. However, in significant 35.7% cases (15/42), anomalies of the permanent successor teeth were either not mentioned or not assessed with some cases conducting exclusive clinical examinations with no radiographs. This stresses upon the need for radiographic examination, preferably panoramic radiographs to permit visualization of the complete primary and permanent dentition. Periodic monitoring through long-term follow-up visits is also advised.

The importance of family history also cannot be ruled out. The present review observed that though the family history was positive in 4.8% of cases, negative in 40.5% cases, yet this was not mentioned in significant 54.8% or 23 cases (Table 2). Parents should be made aware of similar such occurrences in their own children and advised to report early for dental visits. The clinician can also benefit by studying the familial inheritance patterns.

Strengths and Limitations

The current review, spanning 19 years, forms a baseline data for future research. The extracted data is limited to "PubMed" database and needs a wider inclusion involving multiple search engines and a longer period to reach a strengthening evidence.

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