

Non-syndromic Concomitant Hypo-Hyperodontia in the Anterior Region of Mandible- An Extremely Rare Phenomenon: Literature Review with Recent Updates

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ABSTRACT

Introduction: Hypo hyperdontia is a numeric mixed anomaly in which teeth may be supernumerary or absent, relative to the normal complement. The occurrence of this condition in the same individual is termed "Concomitant Hypo-Hyperdontia" (CHH). The presence of CHH in the same dental arch is uncommon, especially when it occurs in the front of the mandible in a Non-syndromic Concomitant Hypo-Hyperdontia (NCHH).

Aim: To conduct a literature review of the published articles in the English language, regarding NCHH in the anterior region of the mandible covering the period from October 1977 to December 2021.

Materials and Methods: An extensive electronic search was conducted in February 2022, from Google Scholar, PubMed, and Elton B. Stephens Company (EBSCO) databases, using keywords such as mandible, hypodontia, hyperdontia and non-syndromic. Manual sorting of the preselected literature, revealed

only 19 published papers concerning NCHH in the frontal region of the mandible, spanning a period of 44 years.

Results: The review analysed the data from 19 published studies, which presented 24 affected patients with a mean age of 12.36 years, with a male/female ratio of 1:1, involving a total of 41 missing teeth, including bilateral mandibular central incisors at 66.66%, followed by missing unilateral central incisor at 29.16% and 1 case (4.16%) displaying absence of both lateral incisors. Regarding 25 presented supernumerary teeth (ST), 72% of cases presented single midline ST, 20% were single unilateral ST, and one case presented double ST (8%) located bilaterally. The majority of ST (84%) were erupted conical-shaped.

Conclusion: The study confirmed the exceptionally rare occurrence of NCHH in the anterior region of the mandible. Due to its interference with esthetics and function, the clinician should possess sufficient knowledge regarding NCHH diagnosis and management.

Keywords: Frontal region, Missing teeth, Supernumerary

INTRODUCTION

Dental abnormalities of teeth number presenting as Supernumerary Teeth (ST) and hypodontia are fairly frequent and most commonly occur as mutually exclusive conditions [1]. The occurrence of both these conditions in the same individual was termed "concomitant hypodontia and hyperdontia" by Camilleri GE, now replaced by the simpler term "concomitant hypo-hyperdontia" as suggested by Gibson [2,3]. Concomitant Hypo-Hyperdontia (CHH) on very rare occasions can arise in the same individual or the same dental arch, but seldom in the same area of an arch [1-6]. Etiopathogenesis of CHH hitherto remains unknown [6]. However, based on literature, the occurrence of CHH appears to be in correlation with some syndromes such as Marfan syndrome and Down syndrome [7,8]. A limited number of reports from literature have shown that the occurrence of concomitant hypo-hyperdontia in non-syndromic patients (NCHH) is the rarest entity [3,6,9]. CHH may affect both jaws (maxilla and mandible) in the primary or permanent dentition [10-15].

Results of Mallineni SK et al., indicate that the most commonly reported type of CHH is bimaxillary hypo-hyperdontia (65%), followed by maxillary (21%) and mandibular entity (14%) [9]. Zadurska M et al., reported the occurrence of CHH in both jaws or solely in the maxilla, but the authors stated that CHH never occurs solely in the mandible [13]. Nonetheless, few case reports have documented the co-existence of both hypodontia (agenesis of central incisor) and hyperdontia (mesiodens) in the anterior region of the mandible [3-5,11,12,14,16-28].

The scarcity of reported cases in English literature concerning the simultaneous occurrence of the two anomalies (hypodontia and hyperdontia) in the anterior region of the mandible in non-syndromic

patients, prompted us to undertake the research regarding this condition. Therefore, the objective of this report was to conduct a literature review of the published articles in the English language, regarding NCHH in the anterior region of the mandible, covering a 44-year period, from the time when the first article concerning NCHH was published, October 1977 to December 2021. The study aimed to summarize previous data regarding this condition and offer an update with the most recent data obtained from research, thus confirming or denying previous findings regarding the rarity of NCHH in the anterior region of the mandible.

MATERIALS AND METHODS

An extensive search was done in February 2022, regarding the occurrence of NCHH in the anterior region of the mandible in electronic databases such as Google Scholar, PubMed, and EBSCO, was performed by using various combinations of keywords such as "mandible", "hypodontia", "hyperdontia", "nonsyndromic", "missing teeth", and "supernumerary".

Initially, 30 papers concerning hypodontia and supernumerary teeth as well as their simultaneous presence, were selected and incorporated into the full text of the current paper. Further selection of the preselected literature was performed by manually sorting only papers related to NCHH in the anterior region of the mandible, based on the title and full content of each paper.

Inclusion criteria: Articles regarding NCHH in the anterior region of the mandible, published in the English language spanning from October 1977 to December 2021, including clinical studies and review articles, were incorporated in the study.

Exclusion criteria: Out of 30 pre-selected papers, 11 of them did not meet the required criteria and did not present data relevant to this investigation such as syndromic CHH, and non-syndromic bimaxillary CHH, hence excluded from the study.

RESULTS

A total of 19 articles were discovered through literature selection, relevant to "Nonsyndromic Concomitant Hypo-Hyperdontia (NCHH) in the anterior region of mandible" published in English language, spanning a period of 44 years (October 1977-December 2021) [3-5,11,12,14,16-28]. Data extracted from the selected literature such as authorship, year of publication, country of origin, age, gender, number of involved teeth (missing teeth), as well as morphology and location of present supernumeraries along with their eruption status, have been presented in separate columns in [Table/Fig-1] [3-5,11,12,14,16-28].

Analysis of 19 reported papers concerning NCHH in the anterior region of the mandible, identified 24 affected patients with a mean age of 12.36 and a male/female ratio of 1:1. The lowest reported age with NCHH was 6 years, while the highest age was 28 years [Table/Fig-1]. This condition was reported more frequently in the mixed dentition in 17 patients (70.83%). All 24 reported patients presented 41 missing teeth and 25 supernumeraries in total.

Literature analysis revealed that the most prevalent missing teeth were bilateral mandibular central incisors in 16 (66.66%) cases, followed by 7 (29.16%) cases with unilateral absence of central incisor and only 1 case (4.16%) displaying absence of both lateral incisors.

Regarding the location of supernumerary teeth (ST), 18 (72%) of them presented as single midline ST (mesiodens), 5 (20%) presented as unilateral single ST, and only 1 case presented bilateral ST [Table/Fig-1].

Concerning eruption status and shape of presented ST, the review showed that 21 (84%) presented ST were erupted conical shaped, 3 (12%) cases were unconfirmed regarding their shape and eruption status, and only 1 (4%) case was confirmed as unerupted conical ST, based on radiography. Regarding the country of origin of the reported cases, the study showed that 17 (89.47%) of the papers were from India.

DISCUSSION

Hypo hyperdontia is a "rare mixed numerical anomaly in which teeth may be supernumerary or absent, relative to the normal complement [9]. Simultaneous occurrence of this condition in the same dental arch is tremendously rare, especially its occurrence in a non-syndromic patient [5,18,19,22]. The occurrence of nonsyndromic concomitant hypo-hyperdontia (NCHH) in the anterior region of the mandible is even more uncommon and exceptionally rare [5,24].

The presence of CHH in the anterior region of the mandible has been seldom reported in English literature until present day. The first case presenting this condition was reported by Low T in 1977 [4], then by Gibson AC in 1979 [3], followed by sparse published case reports and literature reviews over the years [5,11,12,14,16-28]. The rare occurrence of NCHH in the anterior region of the mandible has been documented in reviews by Marya CM et al., and Verma KG et al., in 2012, who confirmed that this condition was only the fourth respectively the fifth case reported in English dental literature until 2012 [17,18].

Author	Year	Country of origin	Patient Gender/Age (years)	Hypodontia	Agenesis of other teeth	Hyperodontia	Eruption status erupted/unerupt
Low T [4]	1977	Malaysia	M/7	31, 41		Mesiodens	Unknown
Gibson AC [3]	1979	U.K	F/6	31, 41	/	Mesiodens	Unknown
Das G et al., [5]	2006	India	F/8	31, 41	/	Mesiodens	Unerupted/conical
Raghavan V H [11]	2009	India	F/8.5	31,41	/	Mesiodens	Unknown
Nuvula S et al., [12]	2010	India	F/15	31,41	18, 28,48	Mesiodens	Erupted/conical
Nayak AG et al., [14]	2010	India	M/28	32,42		Mesiodens	Erupted/ conical
Venkataraghavan K. et al., [16]	2011	India	F/9	31,41		Mesiodens	Erupted conical
Marya CM et al., [17]	2012	India	M/20	31,41	18, 28, 48	Mesiodens	Erupted/ conical
Verma KG et al., [18]	2012	India	M/15.6	31,41		Mesiodens	Erupted/ conical
Verma KG et al., [19]	2013	India	M/9	31,41	18,28,38,48	Mesiodens	Erupted/ conical
Nirmala S.V.S.G et al., [20]	2013	India					
	1.		F/8.5	41	18,28	Unilateral 41 st.	Erupted/ conical
	2.		M/10.9	41	18,28,48	Unilateral 41 st	Erupted/ conical
	3		M/9.2	41	18	Unilateral 41 st	Erupted/ conical
Nirmala S.V.S.G et al., [21]	2014	India	M/8	41		Unilateral 41 st	Erupted/ conical
Nagaveni NB et al., [22]	2014	India					
	1.		M/9	31,41		Bilateral mesiodens	Erupted/ conical
	2.		F/13	31,41		Mesiodens	Erupted/ conical
Tamrakar AK and Rathee M [23]	2014	India	F/21	31,41		Mesiodens	Erupted/ conical
Acharya S [24]	2015	India	M/10	31.41	18, 28,48	Mesiodens	Erupted/ conical
Mandal PK et al., [25]	2015	India					
	1		F/7	31		Mesiodens	Erupted/conical
	2		M/10	31,41	18,28,38,48	Mesiodens	Erupted/conical
Sawai MA [26]	2016	India	F/26	31,41		Mesiodens	Erupted/ conical
Bhadran D et al., [27]	2016	India					
	1		M/7	31		Mesiodens	Erupted/conical
	2		F/9	31		Mesiodens	Erupted/conical
Gupta N et al., [28]	2017	India	F/20	31,41	17,27,18,28,38,48	Unilateral 31 st	Erupted/conical

[Table/Fig-1]: Summary of articles concerning "Nonsyndromic Concomitant Hypo-hyperdontia (NCHH) in the anterior region of mandible", published in English literature covering the period from October 1977 to December 2021 [3-5,11,12,14,16-28].

Verma KG et al., and Nirmala SVSG et al., in their literature reviews of 2013 have reported the presence of NCHH in 9 cases and 11 cases respectively, thus revealing that reviews of Marya CM et al, and Verma KG et al., in the year 2012 did not present the real number of published cases with NCHH in the anterior region of the mandible [17-20]. The last review conducted by Nagaveni NB et al., in 2014, reported 11 published papers with 14 cases (patients) affected with NCHH [22]. As of 2014, few cases have been reported concerning this condition. [21,23-28]. In comparison to previous literature reviews conducted by Marya CM et al, and Verma KG et al., in 2012, as well as the reviews of Verma KG et al., and Nirmala S.V.S.G et al., in 2013, the present study showed more similarity to the review of Nagaveni NB et al., conducted in 2014 [17-20,22].

The similarity can be attributed to the fact that the review of Nagaveni NB et al. in 2014, presented more cases with NCHH in the anterior region of the mandible and additionally more features descriptive of the presented cases [22]. Regarding the relation between NCHH occurrence and gender, the present review showed the same ratio between males and females 1:1, in accordance with the review of Nagaveni NB et al. in 2014, whereas the review of Mallineni SK et al., in 2014 reported a higher propensity of NCHH in males with a 1.3:1 male to female ratio [9,22].

Concerning NCHH in relation to patient dentition status, the present review showed a higher occurrence of NCHH in mixed dentition in 17 patients (70.83%). Nevertheless, data from literature showed that NCHH was found more frequently in the permanent dentition [29]. Concerning missing teeth, findings of the current review showed a higher tendency of missing bilateral mandibular central incisors in 16 (66.66%) cases, similar to data of Verma KG et al., in 2013 with 8 (88.88%) and Nagaveni NB et al. in 2014 with 11(78.57%) cases [19,22].

Regarding the location of ST, the study of Nagaveni NB et al., in 2014 [22], showed predominance of unilateral supernumeraries with a total of 13 (92.85%) cases, compared to results of the present study, with 5(20%) cases of single unilateral ST. These results of Nagaveni NB et al., in 2014, emerged due to the author having reported all single ST as unilateral mesiodentes. Analysis of the present study of the cases presented by Nagaveni NB et al., in 2014 [22], revealed the presence of single midline supernumeraries in 10 (71.42%) cases, which is in accordance with results of present study with 18 (72%) cases of midline supernumeraries.

The present literature review, as the most recent concerning NCHH in the anterior region of the mandible, in comparison to other reviews, is more comprehensive, because none of the previous reviews described all features of the presented cases. Accordingly, the reviews of Marya CM et al., and Verma KG et al., in 2012 [17,18] did not present any ST in addition to missing teeth, while the review of Nirmala SVSG et al., in 2013 [20] did not record the age of the patients. Also, with regard to ethnic background and country of origin of the reported cases, neither the previously mentioned reviews, nor the review of Nagaveni NB et al., in 2014 have specified the ethnic background and the country of origin of presented patients as well as the eruption status of presented supernumeraries. Analysis of the papers included in the present review showed that 17 (89.47 %) published papers were from India, thus suggesting a correlation may exist between ethnic groups and the prevalence of NCHH in the anterior region of the mandible [22]. Eshgian N et al., in 2021, also suggested a possible correlation between ethnic background and prevalence of CHH, based on their study results showing a high prevalence of CHH in Hispanic/Latino patients [30].

In contrast to previous reviews, the current study incorporated the eruption status of presented supernumeraries. The study showed that 20 (83.33%) of patients presented erupted conical-shaped ST. Based on the analysis of each presented case, the erupted ST were the main reason prompting patients to seek dental care, due to their impact on aesthetics and function.

Unerrupted ST usually do not present any symptoms, therefore they may be detected accidentally during the radiographic examination of the jaws for other causes [11]. However, if erupted, ST in the frontal region of the dental arch, especially in mixed dentition, may pose difficulty in diagnosis, causing a dilemma whether the tooth present in the dental arch is a normal central incisor or mesiodens with a missing permanent incisor [22]. Therefore, management of this condition requires accurate diagnosis by careful clinical and radiographic examination as well as multidisciplinary treatment planning with aim of minimizing immediate and long-term consequences [22,23].

This literature review has updated the data regarding NCHH in the anterior region of the mandible, summarizing the data of previous literature reviews with the most recent obtained from the present investigation. The study confirmed the exceptionally rare occurrence of this phenomenon with only 19 published papers in the English language, spanning a period of 44 years.

Limitation(s)

The limitations of the study can be attributed to the fact that the review included only English language articles, therefore providing a low number of reported cases concerning NCHH. The latter can be considered a consequence of the former. Difficulties encountered during the literature review arose because some previous literature reviews did not provide a comprehensive analysis, failing to include all data available such as the number of published cases with NCHH in the front of the mandible or other details of each presented case. Therefore, in order to obtain more reliable data regarding this condition, it was necessary to analyse each article in detail separately, whether a case report or literature review.

CONCLUSION(S)

The present literature review confirmed the extraordinarily rare occurrence of NCHH in the anterior region of the mandible, showing that only 19 papers with this condition have been reported in English literature spanning a period of 44 years (1977-2021). Due to the rare occurrence of NCHH and its interference in patient esthetics and function, the clinician should possess sufficient knowledge regarding diagnosis through careful clinical and radiographic examination, especially in mixed dentition, as well as institute a multidisciplinary treatment of the immediate and long-term adverse implications.

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