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Transmigration of Maxillary Canine in Association with Bilateral Agenesis of Permanent Mandibular Canines-Report of a Rarest Case with Literature Review

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ABSTRACT

The purpose of this article is to present a rarest case, in which the permanent maxillary left canine was impacted and transmigrated from its original position along with presence of idiopathic congenital agenesis of permanent mandibular bilateral (both right and left) canines in a normal healthy Indian female patient. Patient also exhibited other dental anomalies like severe root dilaceration (30-degree bend) in the maxillary left first premolar and short root anomaly or rhizomic involving all third molars. Reporting such unusual dental variations will boost the existing knowledge of the dental anomalies in the web of science.

Keywords: Congenital tooth agenesis, Canine impaction, Maxillary permanent canine, Root dilacerations, Short root anomaly, Transmigration

Abbreviations: MxC; Maxillary Canine, MnC: Mandibular Canine, TrM: Transmigration, BAMnCs: Bilateral Agenesis of Mandibular Canines

INTRODUCTION

Pre-eruptive migration of a tooth across the dental midline without the influence of any pathological factor is termed as 'Transmigration' [1]. Transmigration of maxillary permanent canines is a rarest dental phenomenon characterized by a part of dental tooth structure crossing the mid-palatal suture (dental midline) in the maxilla [1]. The reported prevalence of transmigrated maxillary canines is comparatively very low in contrast to mandibular transmigrated canines [2]. This may be attributed to the fact that the shorter the distance between the roots of maxillary incisors and the floor of the nasal fossa and restriction of the path of tooth movement by the roots of adjacent teeth [1]. This dental variation has been reported only in permanent dentition and hence reporting of such rarity is treasure for inculcating knowledge. Literature search revealed paucity of dental literature on the presentation of permanent maxillary canine transmigration (Table 1) [1, 3-12].

Another dental anomaly like tooth agenesis is the frequently seen clinical entity affecting single or multiple teeth and may be observed as isolated finding or encountered in syndromic patients [13]. Exclusive congenital agenesis of bilateral permanent mandibular canines with no other teeth missing is a sporadic occurrence and to date only countable number of individual cases or prevalence studies has been reported in the dental literature (**Tables 2 & 3**) [13]. This clinical entity when it is present is usually accompanied with morphological and growth-related changes in the teeth and oro-dento-maxillofacial complex. It is also showed that this rare variation occurs in association with other dental abnormalities such as microdontia, retained primary teeth, malocclusion, agenesis of other teeth and supernumerary teeth. The developmental agenesis of permanent canines is reported higher in females compared to males, most frequently affecting the maxillary arch with unilateral predilection, being left side more affected than right side [13-31].

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Table 1. Published articles (including both prevalence studies and case reports) on transmigration of permanent maxillary canines in the web of knowledge.

Sl. No.	Author	Year	Transmigrated maxillary canine Unilateral/Bilateral	Associated other anomalies
1	Present case	2023	Unilateral, Right canine	Congenital agenesis permanent mandibular right and left canines
2	Sharma & Nagpal [3]	2014	5 cases (0.16%) out of 3000 patients All unilateral (2-Right canine)	-
3	Kamiloglu & Kelahmet [4]	2014	2 cases (0.44%) out of 453 patients Unilateral (Left canine)	-
4	Narsapur [5]	2014	Unilateral (Right canine)	-
5	Kumar [6]	2012	3 cases All Unilateral (1-Right 2-Left)	Transmigration of left lateral incisor
6	Mazinis [7]	2012	2 cases out of 3586 patients Both Unilateral	-
7	Aras [8]	2008	12 cases out of 6000 patients All Unilateral 6-Right canine 6-Left canine	-
8	Kurol J [9]	2006	Unilateral	-
9	Auluck & Mupparapu [10]	2006	Unilateral	-
10	Ryan [11]	2005	Unilateral, Left canine	Hemifacial microsomia, cleft palate
11	Shapira & Kuftinec [12]	2005	Unilateral	-
12	Aydin & Yilmaz [1]	2003	Unilateral, Left canine	Retained primary canine
13	Aydin [13]	2003	6 cases out of 4500 patients (0.13%) All Unilateral 3-Right canine 3-Left canine	Impacted canines, microdontia of maxillary lateral incisors, third molars, retained primary teeth, agenesis of maxillary lateral incisor

Sl. No.	Author	Year	Ethnicity/Country	Prevalence (%)
1	Dolder E [14]	1937	-	out of 10,000 children
2	Rose JS [15]	1966	-	0.02
3	Muller [16]	1970	North America	Only 3 cases found out of 14940 children
4	Bergstrom [17]	1977	-	0.08
5	Davis PJ [18]	1987	Hong Kong	0
6	Hokari [19]	2000	-	0.07
7	Fukuta Y [20]	2004	Japan	0.06
8	Rozsa [21]	2009	Hungary	0.09

Table 2. Reported prevalence studies on congenital agenesis of permanent mandibular canines.

Table 3. Detailed description of reported case reports pertaining to congenital agenesis of permanent mandibular canines.

Sl. No.	Author/Year	Location of the study	Missing Permanent Mandibular Canine Unilateral/Bilateral	Associated Anomalies
1	Present case, 2023	India	Bilateral (33, 43)	Transmigration of maxillary right canine
2	Marya [22]	Combodia	Bilateral (33, 43)	Congenital agenesis permanent maxillary both canines
3	Shirolkar [23]	India	Bilateral (33, 43)	Agenesis of both maxillary canines, congenital absence of mandibular central incisors and shovel-shaped maxillary incisors
4	Garcia-Marin C [24]	Isfahan	Unilateral (43)	Sella turcica bridging
5	Yadav [25]	India	Bilateral (33, 43)	Congenital agenesis of maxillary both canine, and mandibular both central incisors
6	Trophimus GJ [26]	India	Bilateral (33, 43)	-
7	Jindal [27]	India	Bilateral (33, 43)	-
8	Kohli [28]	India	Bilateral (33, 43)	-
9	Arora [29]	India	Bilateral (33, 43)	-
10	Guna Shekhar [30]	India	Unilateral (43)	Congenital missing mandibular permanent central incisors and microdontic maxillary lateral incisors
11	Ramaraj & Mirza [31]	Bahrain	Bilateral (33, 43)	Supernumerary supplemental lower incisor

Published literature shows individual case reports pertaining to the above uncommon dental conditions described. Concomitant occurrence of both these different dental variations in a non-syndromic single patient is an infrequent dental phenomenon. Publishing such unusual combination of tooth anomalies, throws enormous light on the existing literature and increase the validity of web of knowledge. Therefore, the purpose of this paper is to report an uncommon idiopathic development of two different tooth variations like transmigration of maxillary permanent canine and congenital agenesis of permanent mandibular bilateral canines (both right and left) in an Indian female patient.

CASE REPORT

A 35-year-old female patient reported to a private dental clinic complaining of pain in the upper right back tooth region from past few months. Patient was apparently normal, healthy and did not show any features of syndrome or systemic or metabolic disorders. Patient gave a history of few milk teeth not exfoliated at the regular time as other teeth and few permanent teeth not come into the cavity. On intra oral examination, complete permanent dentition was observed with all third molars erupted. On further observation of the oral cavity, the primary canines including left canine in the maxilla and both right and left canines in the mandible were retained with absence of mobility in those teeth. The crown morphology was studied in detail to rule out from its permanent successors. The crown dimensions were small in all three canines and anatomical features were suggestive of primary canines. Suspecting the impaction of permanent canines, an orthopantomography radiograph was advised. On radiographic examination, an impacted permanent maxillary left canine was observed. On careful examination, the left canine was located mesio-angularly about 30-35-degree inclination with the midline and part of the crown was crossing the mid-palatal suture and located above the level of root tip of the left central incisor (Figure 1). In the mandibular arch, presence of retained both right and left primary canines was confirmed as the roots of both primary canines were very short as compared to permanent canines where they exhibit long roots. Both right and left permanent canines were congenitally missing. Other dental variations were on this radiograph like severe root dilaceration (30-degree bend) in the left maxillary first premolar and short root anomaly or rhyzomicroly involving all third molars (Figure 1). Therefore, based on all clinical, radiographic and literature findings the case was diagnosed as idiopathic, non-syndromic, congenital agenesis of bilateral mandibular permanent canines, transmigration of permanent maxillary canine in association with other dental anomalies. As patient had pain because of impacted canine, the tooth was surgically removed through labial approach under local anesthesia. As all three retained primary canines were intact with no evidence of mobility or root resorption and all permanent teeth were well aligned, no other treatment was advised and patient was kept under periodic follow up until exfoliation of primary canine teeth.



Figure 1. Orthopantomograph radiograph showing transmigration of maxillary left canine, congenital agenesis of permanent mandibular both right and left canines, retained primary maxillary left canine (blue arrow) and mandibular both right and left canines (yellow arrows). Other anomalies like dilaceration (30-degree bend) in root of maxillary left first premolar and short roots involving all third molars were present.

DISCUSSION

Congenital agenesis of bilateral permanent mandibular canines is extremely rare and the prevalence varies from 0.18% to 0.45% in different population like Hong Kong, Hungary and Japan [13-25]. The extensive review of available literature revealed very few publications on prevalence studies and case reports regarding congenital agenesis of permanent mandibular canines and is elaborated

in **Tables 2 & 3** [13-31]. It is evident from the reported studies that, the prevalence of missing canines is more in females compared to males and mostly reported in the maxilla compared to mandible. Even in the present case, it occurred in female patient thereby strongly supporting the existing literature. The exact etiology behind occurrence of agenesis of permanent canines is not stated. However, a more significant factor has been linked with familial or genetic inheritance. In genetic etiologic basis, the autosomal

dominant inheritance pattern is the most reported factor with identification of regulatory homeo-box genes like AXIN2, EDA, PAX9, MAX1 in association with tooth agenesis. The etiology of dental agenesis has also been suggested to be multifactorial, which combines genetic, epigenetic and environmental factors. Sometimes, environmental factors like tooth bud infection, trauma, maternal medication, nutritional disturbances during pregnancy or infancy, irradiation at early stage and somatic diseases such as scarlet fever, syphilis and rickets are also reported with tooth agenesis [13-31].

There is no particular treatment for the agenesis of permanent canines. However, researchers have stated different treatment options like timely extraction of the primary canines to facilitate spontaneous closure with or without additional orthodontic treatment, replacement with removable or fixed partial dentures, and closure of spaces by orthodontic movement of teeth, coronoplasty for lateral incisor and first premolar and retaining the primary canines by restoring with a suitable prosthesis when they exfoliate [16-20]. There are reports showing presence of retained primary canines in an individual's age ranging from 35 to 50 years and is usually associated with either impaction of permanent successors or their agenesis [17-20]. This factor is evident in the present case showing presence of three retained primary canines. This may be due to the lack of root resorption phenomenon because of absence of underlying teeth. It is been suggested that retaining the primary predecessor is more advantageous rather than going for its extraction followed by replacement. By doing this resorption of alveolar bone is avoided till patient reaches appropriate age for placement of implant-based prosthesis without need for bone grafting [18-22]. In the case presented here, as the retained primary teeth were found with absence of root resorption and tooth mobility, and there were well aligned teeth without any existing spaces no treatment was advised for the patient. A definite treatment can be performed when the primary canines exfoliate. Therefore, patient was informed about the condition and kept under observation.

The transmigration of permanent canines happens not only in the mandibular arch but also found in the maxilla. The first case of maxillary canine transmigration was shown by Aydin and Yilmaz in the year 2003 [1]. Pertaining to mandibular canine transmigration, literature shows different classification systems given by Mupparapu [2] based on position of the transmigrated canines. However, in the maxilla as permanent canines rarely transmigrate, the literature on its etiology, incidence, demographic factors, and classification and on mechanism behind how transmigrated maxillary canines cross the mid-palatal suture and pass onto the contralateral side of the midline needs to be studied extensively in the future. The presence of retained primary teeth is the most frequently encountered dental condition in agenesis of the permanent canines [13-31]. Even in the present case, retained primary canines in the maxillary right region, and mandibular both right and left retained primary canines were found. On both clinical and radiographic examination, the anatomical features of the primary canines were ruled out from the permanent canines.

In the case presented here, the permanent maxillary left canine was impacted and oriented in mesio-angular direction with its part of the crown structure crossing the mid-palatal suture above the roots of central incisors. Apart from original definition on transmigration of canines, it is also stated that, when a part of crown an impacted tooth has crossed the midline should be considered as transmigrated [1]. Therefore, the more important factor is the tooth crossing the midline not the distance of the migration. Even Mupparappu's nine patient series and most of the reports published so far shows the teeth with only the part of the crown crossing the midline [2-10]. Moreover, the stage of transmigration will depend on the time of diagnosis of the case. Based on these factors, the present case was considered as a 'true transmigration' fitting to the criteria given by original researchers [2-12].

Regarding transmigration of canines, there is no wellestablished treatment modality. Different treatment options have been suggested based on different cases like surgical removal to avoid formation of cysts or any other complications, regular observation and orthodontic movement of the tooth for proper alignment in the oral cavity [4-10]. In this case, the transmigrated canine was surgically removed through labial approach as patient had discomfort from this impacted tooth. The retained primary canine in the left quadrant was firm and did not exhibit any mobility. Moreover, there was no any evidence of root resorption or any other pathological entity observed on the radiograph. Therefore, the primary canine was kept under observation till its complete exfoliation.

In the present case, bilateral agenesis of permanent mandibular canines was observed along with transmigration of permanent maxillary canine and other dental anomalies. This combination of two entirely different dental variations occurring in an Indian patient is not reported so far, thereby making this case as unique and worth to publish to delineate more knowledge. From author's archive, many reports on dental anomalies have been published regarding transmigration of mandibular permanent canines and bilateral congenital agenesis of mandibular central incisors from India [32-34]. From the current case report and other previous published documents [13-31], it is more confirmed that, although transmigration of permanent canines is an unusual phenomenon but still this clinical condition is not mentioned in the textbooks as a distinct entity. Author hereby strongly recommends for the inclusion of this condition as a developmental tooth anomaly in the textbooks and more studies should be undertaken in the coming years to draw out appropriate classification systems on transmigration of maxillary canines.

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