

Unilateral agenesis of permanent superior canine in familial peg-shaped lateral incisors: rare case report and literature review

OANA-CELLA ANDREI¹⁾, CĂTĂLINA FARCAȘIU²⁾, ADRIANA BISOC¹⁾, DANIELA IOANA TĂRLUNGEANU¹⁾, LIVIA ALICE TĂNĂSESCU¹⁾, MAGDALENA NATALIA DINA³⁾, MIHAI BURLIBAȘA³⁾, RUXANDRA MĂRGĂRIT⁴⁾

¹⁾Department of Removable Prosthodontics, Faculty of Dentistry, Carol Davila University of Medicine and Pharmacy, Bucharest, Romania

²⁾Department of Pedodontics, Faculty of Dentistry, Carol Davila University of Medicine and Pharmacy, Bucharest, Romania

³⁾Department of Dental Techniques, Faculty of Midwifery and Nursing, Carol Davila University of Medicine and Pharmacy, Bucharest, Romania

⁴⁾Department of Restorative Odontology, Faculty of Dentistry, Carol Davila University of Medicine and Pharmacy, Bucharest, Romania

Abstract

Agenesis of permanent maxillary and mandibular canines is very rare; one to all four can be missing, isolated or in association with other missing teeth or with dental morphology abnormalities. Such cases can present functional, esthetic, and psychological problems, since the canine's role in functional occlusion and in obtaining an aesthetic smile is crucial. Frequently, the canine's absence from the arch is caused by impaction; its congenital absence is extremely rare in patients with no associated syndrome. The aim of this paper was to present a very rare case of a non-syndromic Romanian adult female patient with a unilateral permanent maxillary missing canine, in association with peg-shaped maxillary lateral permanent incisors, which can have a genetic cause, since the same morphological abnormality was present in her father and her younger sister, who also presented a rare situation of bilateral upper transposition between the canines and lateral incisors. The association of a permanent maxillary canine hypodontia with bilateral "peg laterals" and transposition between the maxillary canine and the peg-shaped lateral incisors is also rare as a family character in healthy patients. Future statistical studies are necessary to establish the incidence of permanent maxillary canine agenesis in Romanian population. Also, the article includes a detailed literature review of reported cases of agenesis of permanent maxillary canines.

Keywords: agenesis of permanent maxillary canine, peg-shaped maxillary lateral incisors, transposition.

Introduction

Permanent teeth are subjected to anomalies in number, size or form caused by irregularities in morpho-differentiation. Congenital absence of one or several teeth is called hypodontia and affects most frequently the third molars, upper lateral incisors, second premolars and lower central incisors [1]. Congenital absence of permanent canines is very uncommon, with most occurrence rates of less than 1% in different populations in studies made on large-numbered samples [1–5]. Differential diagnosis must be made with impaction or displacement; canine is the most variable positioned tooth; it can be found palatally or facially displaced or ectopically erupted from the dental arch [6]. The persistence of the deciduous canine is usually a symptom of these situations, agenesis of permanent canine being rarely reported [7]; isolated forms of canine agenesis, which are not associated with other congenitally missing teeth, are even rarer [8]. Asian populations reported a higher percent of cases than Caucasians, which orientated the etiology to the predominance of the genetic factors [9]. Some researchers are considering a polygenic model of inheritance and an influence of the environmental factors during the prenatal period, especially in the intrauterine phase [10]. In a comparative study

conducted on 600 persons belonging to the black population, respectively 1100 persons belonging to the white population in America [11], only two cases of congenitally missing maxillary canines were found in the black population and no case in the white population. For the Romanian population, there is no available data on the prevalence and distribution of permanent canine agenesis in non-syndromic patients.

In most hypodontia cases, a dominant autosomal transmission seems to prevail [12–20]. The genetic or the familial inheritance has been attributed as a more significant etiological factor. Autosomal dominant (AD), autosomal recessive (AR) and X-linked recessive pattern of inheritance have been associated with tooth agenesis; with AD pattern being the most prominent [21]. Environmental factors like tooth bud infection, trauma, nutritional disturbances during pregnancy or infancy, smoking during pregnancy, maternal medications, irradiation at an early stage and somatic diseases (syphilis, scarlet fever, and rickets) are also associated with tooth agenesis [22, 23].

Dental transposition represents the change of the eruption place of two neighboring teeth, frequently between the canine and the first premolar for the upper jaw, and between the canine and the lateral incisor for the lower jaw. Either

unilaterally or bilaterally, the prevalence of transposition is low, with values below 1% in any population. Transposition is more common in women [24] and in the upper jaw than in the mandible; unilateral transposition has a higher prevalence than bilateral [25, 26]. For the maxillary, the most common tooth involved in transposition is the canine; the transposition with the lateral incisor is found in only 20% of all cases, the rest of the cases involving the first premolar [27].

Aim

The aim of this case report was to highlight a case of non-syndromic unilateral agenesis of left permanent maxillary canine, along with peg-shaped bilateral maxillary permanent lateral incisors in a healthy 34-year-old Romanian female patient; her father and sister had all permanent canines present on the arches, but the same peg-shaped lateral incisors, in contrast to her mother, who presented no number or shape dental modifications. Also, the sister presented a rare situation of bilateral upper transposition of the permanent canines and peg-shaped lateral incisors, suggesting a genetic etiology. Detailed dental and general history data were obtained from each of all four patients; for the first patient, any possibility of extraction or avulsion by traumatism of the maxillary left permanent canine was excluded. This article also presents a literature review of reported cases of agenesis of permanent maxillary canines,

and also of prevalence, etiology and clinical implications of the condition.

Case presentations

Case No. 1

A 34-year-old Romanian female patient having no general medical condition came to our Clinic accusing pain on the level of the left mandibular first molar, 3.6. On intraoral examination, we noticed the retention of the left primary canine and the peg-shaped lateral permanent incisors (Figure 1, a and b). On the panoramic and periapical X-rays, we discovered the agenesis of the left permanent canine, 2.3; the degree of the root resorption of the persistent primary canine was minimal (Figure 2, a and b). The patient also presented no other agenesis but of all third molars, confirmed both radiologically and through anamnesis, the other missing teeth being extracted at different ages, due to the carious complications. Occlusion and physiognomy were affected. The patient did not benefit of specialized treatment during childhood; the retained primary canine with minimal root resorption was positive, preserving the dental arch integrity and providing a good clinical situation for a later implant–prosthetic rehabilitation. Considering the age, extraction of the deciduous canine with orthodontic space closure and premolar crown-plasty was not an option. The treatment planning agreed with the patient was temporization until the loss of the deciduous canine.

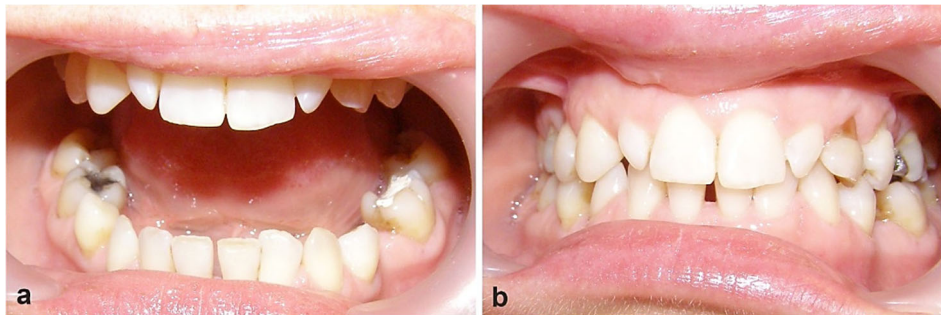
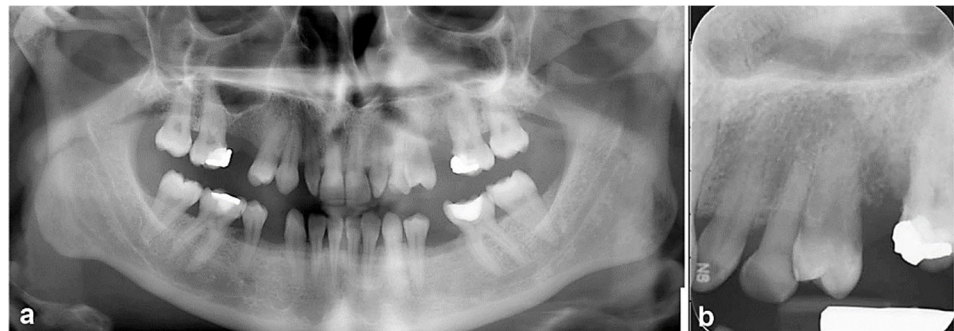


Figure 1 – (a and b) Aspect of the mandibular arch at presentation; aspect of peg-shaped maxillary lateral incisors, persistence of the primary maxillary left superior canine.

Figure 2 – (a and b) Panoramic radiological investigation showing the absence of the permanent maxillary left canine; retroalveolar radiograph showing the minimal degree of the root resorption of the persistent primary canine.



Case No. 2

A 62-year-old male patient, the father of the first patient, came to the our Clinic soliciting treatment for his partially edentulous state, accusing affected esthetics and difficulties in mastication; the maxillary arch was in a second class Kennedy edentulous state with two modifications, while on the mandibular arch only the second left molar was missing; on the maxillary arch the teeth were in a severe state of attrition; the interarch space was limited and the vertical dimension of occlusion severely reduced (Figure 3, a–c). The space available for fabricating the acrylic partial

denture was very limited, making the prosthetic restoration a challenging task. For better esthetic results, the patient was asked to bring old photos; on these images the peg-shaped maxillary lateral incisors were clearly observed, but also the presence of both permanent canines (Figure 4, a and b).

Case No. 3

The third patient, the 30-year-old first patient's sister, presented the same shape and aspect of the first patient's permanent canine and lateral incisors, but in a rare situation of bilateral transposition, which was confirmed on the panoramic X-ray (Figure 5). She also presented no other

agenesis; three of the four of her third molars were still on the arches, while the fourth was extracted six years ago. All her other missing teeth were also extracted at different ages, due to the carious complications. Occlusion and physiognomy were affected through tooth shape, bilateral transposition between upper canines and lateral incisors and deficient space closure caused by absence of prosthetic or orthodontic treatment consecutive to early extractions. Only one of the edentulous spaces was restored using a three-unit bridge, in the adult age.

Case No. 4

The fourth patient, the 58-year-old first and third patient's mother, came to our Clinic soliciting the replacement of her old upper bridges, accusing bad breath and esthetic issues. The clinical shape, aspect and alignment of her permanent canines and lateral incisors were normal; also, on her panoramic X-ray no sign of hypodontia or morphological variation of any tooth was detected (Figure 6).

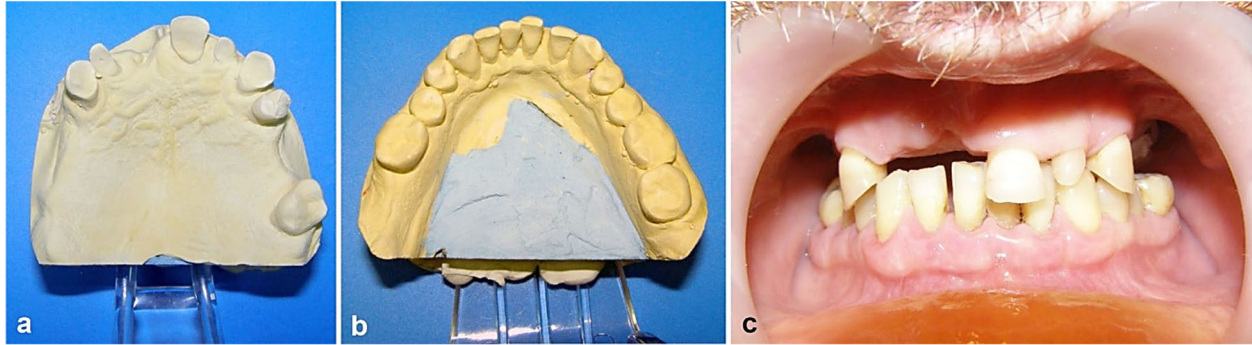


Figure 3 – (a–c) Present clinical situation of the second patient: diagnostic cast of the maxillary arch, diagnostic cast of the mandibular arch and occlusion, showing the pronounced abrasion of the remaining maxillary teeth.

Figure 4 – (a and b) Old photograph of the father showing peg-shaped maxillary lateral incisors and the presence of both permanent canines; similar shape and aspect of the first patient's permanent canine and incisors.

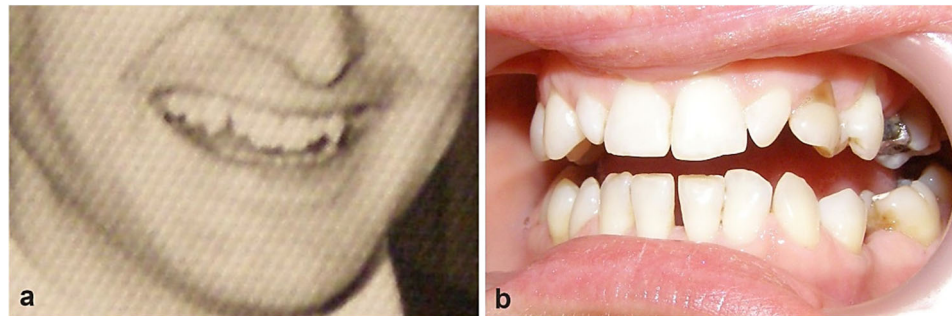


Figure 5 – Panoramic radiological investigation showing the third patient's peg-shaped maxillary lateral incisors and the presence of both permanent canines, having similar shape with her father's and sister's.

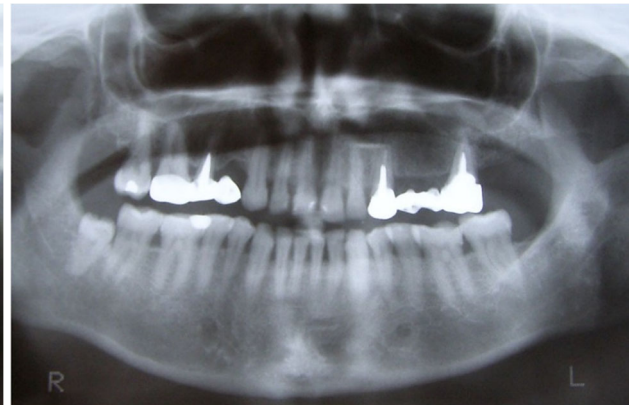


Figure 6 – Panoramic radiological investigation of the mother, showing the presence of both permanent maxillary canines and no similarity in incisor's or canine's shape.

☒ Discussions

Some of the studies in literature reported a more frequent occurrence of unilateral tooth agenesis than bilateral, in maxilla than in mandible, in female patients than in male patients [3, 28]. Still, other authors found that bilateral form is more frequent [2]. In the present article, we report a case of a non-syndromic young female patient presenting a unilateral permanent maxillary canine agenesis. Bailit

classification [10] considers the upper canine as stable, along with the upper central incisors, the first premolars and the first molars, while another study [29] found a very high prevalence of 3.7%, which can only be explained by the fact that the studied sample was very small (only 27 cases). In a study on the Chinese population made by Cho *et al.* [9], the absence of upper permanent canines was shown in 32 cases among 69 852, while another study led by Davis [30], also on the Chinese population, showed the

absence of upper permanent canines in five cases among 1093. For the Japanese population, a percentage of 0.11% upper permanent canines missing was found [3]. For the European population the studies show a prevalence of permanent maxillary canine agenesis of 0.27%, a value found by Rózsa *et al.* [2] in the Hungarian population; a 0.37% value was found by Sisman *et al.* [31] in the Turkish population and a 2.1% value was found by Fekonja [32] in a study on Slovenian orthodontically treated children. The prevalence of permanent maxillary canine agenesis has been shown to be generally very low in the population, with a prevalence from 0.07% to 0.13% [33].

Rózsa *et al.* found various complications of dental anomalies associated with permanent canine agenesis, such as persisting primary canines, agenesis of permanent germs, presence of other primary supernumerary teeth or of a supernumerary cusp, and frequent occlusal disturbances [2]. Sharma reported a non-syndromic case of concomitant multiple supernumerary teeth and partial anodontia [34]. In our first case, besides the persistence of the primary left canine, we also found microdontia affecting the maxillary lateral permanent incisors of the patient, of the patient's father and sister, while the mother presented no hypodontia or morphological variation. In association with the peg-shaped laterals, the sister also presented an upper bilateral transposition between the canines and lateral incisors, which is a very rare situation as well. In a study conducted by Fukuta *et al.* [3], three permanent canines were absent in two of the patients. Bazan reported two cases where he found the association between congenitally missing maxillary permanent canines and peg-shaped maxillary lateral incisors [35]. Similar to this study, Cho *et al.* [9] showed the association between congenitally missing maxillary permanent canines and microdontic bilateral maxillary permanent lateral incisors, reporting three such cases. Peck *et al.* [25] showed the association between transposition of the canine and first maxillary premolar and hypodontia of maxillary lateral incisor.

Microdontia affecting the maxillary lateral permanent incisors, known as "peg laterals", is one of the most common localized microdontia, with a prevalence between 0.8–8.4% [36]. Teeth size is predominantly genetically determined; it depends also on the race and endocrinal factors. Unilateral peg laterals are more commonly found; their bilateral presence is rare [37]. In our cases, three members of the same family (the father and his two daughters) share this trait, while the mother has normal lateral permanent incisors; none of them are suffering from any associated syndrome or systemic disease. Some authors reported an X-linked recessive heredity in five families (affected boys and their mothers) with hypohidrotic ectodermal dysplasia; the analysis of dental traits showed that the mothers, gene carriers, had a normal appearance but had either hypodontia or peg shape incisors [38]. A study comparing transposition between maxillary canine and first premolar with transposition between maxillary canine and lateral incisor showed that in both groups there is an increased percentage of association with peg-shaped lateral incisors or with other congenitally missing teeth [39]. In our case, no member of the family presented general medical conditions or syndromes; peg laterals appeared bilaterally and were present only in the father and daughters, the mother having normal shaped

incisors. Galluccio *et al.* reported that more than 300 genes are involved in different phases of teeth development, sustaining the necessity to develop tests for early diagnosis of anomalies in teeth organogenesis [40]. In our case, the non-syndromic female first patient associates two anomalies, one of number (alterations of the dental lamina or the germ) and the other of shape, that appeared in two different development stages.

The etiology of congenital absence of the teeth is not fully known. Non-syndromic congenital absence of teeth can be sporadic or familial. Several studies have demonstrated that dental development is under strict genetic control of position, number, dimension, and shape of the tooth. Genetic transmission can be X-linked, recessive or dominant. Various diseases with genetic transmission, such as ectodermal dysplasia, Rieger syndrome, Down syndrome, or Witkop syndrome, associate congenital absence of the teeth [41, 42]. Still, cases of congenital absence of the teeth have been also described in healthy patients. In such situations, the environmental factors and subsequently the genetic transmission must be considered [43]. Mutations in *msh* homeobox 1 (*MSX1*), paired box 9 (*PAX9*), ectodysplasin A1 (*EDA1*), and axis inhibition protein 2 (*AXIN2*) genes are involved in the etiology of isolated hypodontia [44]. Regarding the etiology of isolated hypodontia of the maxillary permanent canines, the study presented by Kantaputra *et al.* [44] showed that mutations in the Wnt family member 10A (*WNT10A*) gene are involved. It has also been shown that hypodontia of the maxillary permanent canines may be associated with microdontia of the maxillary permanent lateral incisors, but also with hypodontia of the maxillary permanent lateral incisors [9, 35]. A study made by Han *et al.* in two Chinese families with non-syndromic X-linked hypodontia has shown that a *Thr338Met* mutation of the *EDA* gene was responsible for the congenital absence of maxillary and mandibular central incisors, lateral incisors and canines, with the high possibility of persistence of maxillary and mandibular first permanent molars [45]. The genetic etiology of transposition has been shown in specialized studies [46], and its association with other changes, such as hypodontia [25, 47] or peg-shaped maxillary lateral incisor teeth [26], has been described. Tooth agenesis is frequently associated with other dental disorders, such as hypodontia, different ectopias or transpositions [48], indicating the influence of genetic transmission. The determination of the eruption site is made genetically by means of the *MSX1* and *MSX2* genes [49]. The presence of mutations in these genes will lead to changes in the eruption and position. At the same time, a mutation of the *AD MSX1* gene in a family has been shown to generate hypodontia [13, 50]. The studies showed that the frequently encountered association between changes in the structure or number of the teeth and changes of the eruption and position is generally genetic in nature [51].

☐ Conclusions

The canine's role in functional occlusion and in obtaining an aesthetic smile is crucial; its absence has serious consequences. Frequently, the canine's absence from the arch is caused by impaction; its congenital absence is very rare. Although any permanent tooth can be congenitally absent, patients with missing upper permanent canine and

no associated syndrome are extremely rare. In our country, no studies specially made on missing permanent canines have been made yet. The cases presented here are highlighting the possible association of a rare permanent maxillary canine hypodontia with other rare situations, such as bilateral morphological variations of maxillary lateral permanent incisors (“peg laterals”) and transposition between the maxillary canine and the peg-shaped lateral incisor, all having a family character. Further investigations are necessary to prove the genetic association between peg-shaped maxillary lateral incisors and maxillary canine agenesis or transposition. Also, further studies are necessary to offer data on the prevalence and distribution of permanent canine agenesis or transposition in the Romanian population, especially in patients with no associated syndrome or systemic disease.

Conflict of interests

The authors declare that they have no conflict of interests.

Authors' contribution

Authors #1 (OCA) and #2 (CF) have equal contributions to this paper.

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Corresponding authors

Livia Alice Tănăsescu, Assistant, DMD, PhD, Department of Removable Prosthodontics, Faculty of Dentistry, Carol Davila University of Medicine and Pharmacy, 37 Dionisie Lupu Street, Sector 2, 020021 Bucharest, Romania; Phone +40723–980 203, e-mail: tanasescualice@gmail.com

Magdalena Natalia Dina, Assistant, DMD, PhD, Department of Dental Techniques, Faculty of Midwifery and Nursing, Carol Davila University of Medicine and Pharmacy, 37 Dionisie Lupu Street, Sector 2, 020021 Bucharest, Romania; Phone +40723–198 709, e-mail: stomatologie@instantprint.ro

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