

## CASE REPORT



## Orthodontic treatment of a non-syndromic patient with congenital agenesis of multiple permanent teeth: a rare case

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### Abstract

This article presents the first stage of treatment, respectively the orthodontic management of a non-syndromic, 13-year-old patient, with multiple and asymmetric missing teeth. The difficulty of the case was increased by the association of an impacted premolar and also by the loss, due to extended caries, of three of the first permanent molars. The patient came from a rural area, where access to dental treatment was limited. His parents realized too late that the child had significant damage to his permanent first molars, that he was no longer eating properly and that he had spaces between teeth. The uncertain outcome of endodontic treatment and prosthetic restoration at the level of first permanent molars and the additional costs made the parents decide, together with the dental practitioner, to extract teeth Nos. 16, 36 and 46. Giving the situation, the first phase treatment plan was represented by orthodontic closing of several maxillary spaces and the reduction of edentulous ridge in the mandible, followed by the maintenance of the space for two future implants, each one replacing the first molars in the third and fourth quadrants. Particularly for this growing patient, early intervention to treat and save as much from the permanent teeth as possible, to expose the impacted premolar, to level and align the teeth in order to obtain continuous dental arches and a good occlusion plane, and later to apply space maintainers that will be replaced by prosthetic restorations, represented an enormous step for long-term stability and proper functioning.

**Keywords:** hypodontia, congenital missing teeth, tooth agenesis.

### Introduction

Congenital missing of one or several teeth is called tooth agenesis, being one of the most frequent dental anomalies [1]. Depending on the number of teeth missing, it can be classified as hypodontia, oligodontia or anodontia. Hypodontia is the term used to describe the absence of less than six teeth, not including the third molars, while oligodontia describes the absence of more than six teeth, without counting the wisdom teeth. Anodontia is the term used for describing the absence of all teeth [2]. Other authors, such as Dhanrajani, classified hypodontia based on the severity of the condition, as follows: mild to moderate hypodontia when two to maximum fifth teeth are absent and severe hypodontia when more than six teeth are missing, without counting the third molars [3]. AlShahrani *et al.* further explain the classification by making three categories: mild when one or two teeth are absent, moderate when three to fifth teeth are absent and severe when six or more teeth are missing, except the third molars [4].

Congenital missing teeth can occur both in primary or permanent dentitions [5, 6], but most of the studies describe and evaluate the missing teeth in permanent dentition [5, 7–11]. When a tooth is missing in the temporary dentition, the same condition is usually found in the permanent

dentition, involving its successor [12]. In studies that evaluate tooth agenesis in primary dentition, it was found that this situation is more common in the maxilla and also the most frequently involved tooth is the lateral incisor [13]. The prevalence in primary dentition is low, the frequencies mentioned in several studies being 0.5% in a study on Icelandic population [14] and 2.4% in a study on Japanese population [15]. In the permanent dentition, the prevalence of congenital missing teeth is higher and has more variations. For example, in the Turkish population hypodontia is the most common dental anomaly of development, with a frequency of 2.8% [16]. In a Japanese study, the prevalence was 8.5% [17], and in an Irish study, it was as big as 11.3% [18].

### Aim

In the case that will be presented above, four permanent teeth were congenitally missing without counting two permanent third molars, and also one premolar was impacted. We do not have data about the primary teeth, since the patient came from a rural area, where access to medical treatment is limited. To make the case even more complicated, the parents were not aware that the permanent lateral teeth had big decays and they did not afford an initial treatment

with uncertain prognosis, so together with the general dentist the decision was made that three out of four permanent first molars will be extracted. The lack of dental treatment in the early stages of life put this growing patient in the situation that at 13 years of age, he already had nine missing teeth and an impacted one. Immediate multidisciplinary intervention was mandatory, in order to rehabilitate the case and bring the patient to a stable path for optimal further development.

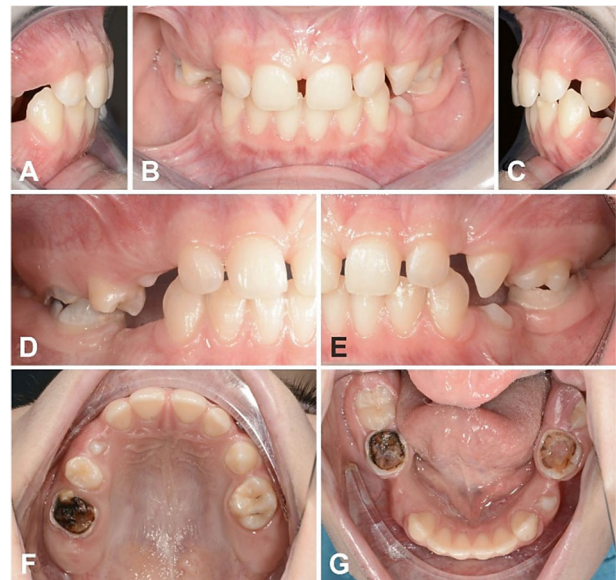
### ☞ Case presentation

The male patient, aged 13, came to our Clinic with one of his parents, with the main concern of multiple unerupted teeth. Our initial intraoral evaluation revealed the absence of teeth 15, 24, 25, 35 and 45. No deciduous teeth were present on the arches. Several permanent teeth had important crown alterations (Figure 1, A–G). Anterior diastema was caused by the absence of several teeth in the maxilla. There was also a slight deep bite at the frontal level. The vertical dimension was unsupported on the lateral sides due to the important molar destruction, especially on the right side, and some of the teeth were over erupted (first right upper premolar and left upper canine). The panoramic X-ray confirmed that no dental buds were forming for teeth 24, 25, 35, 45, nor for the upper third molars (Figure 2A). The tooth No. 15 was impacted in a mesial position, just above tooth No. 14 (Figure 2A). A lateral cephalogram (Figure 2B) and regular dental photographs (Figure 1, A–G) were performed before starting the orthodontic treatment. The occlusion was class I at the canine level and class II at the molar level, on a normodivergent pattern (Figure 2B). The face was well developed, the profile was harmonious, and the dental midlines coincided with the face midline (Figure 1B; Figure 3, A and B). Following rigorous evaluation and considering the uncertain prognosis and high costs of endodontic treatment, it was decided together with the parents that the teeth Nos. 16, 36 and 46 cannot be kept and their extraction was performed, making it even more difficult to manage the absence of multiple dental units. The advantage was that their extraction was made after the eruption of the second molars and their intercuspation maintained the vertical occlusion to some extent.

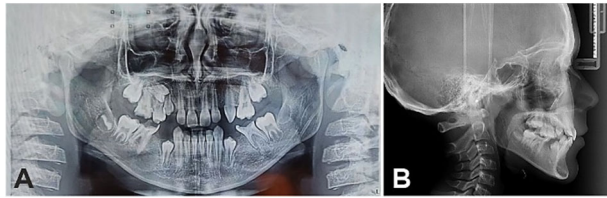
The orthodontic treatment was performed using multi-attachment fixed upper and lower appliances in MBT 022 slot prescription. There was no need for other enlargement devices, since the maxilla and the mandible were developed enough to be able to incorporate this number of teeth. The general objectives were to achieve a good and stable occlusion, to keep all the remaining teeth in good function, to create space, expose and align on the arch the second right upper premolar, to keep the dental midline coincident with the facial one, to close all the residual spaces in the upper arch and to obtain and keep space for two dental implants in the third and fourth quadrants. We started the treatment with the upper appliance and after three months the lower one was associated. After initial levelling and alignment, space was created in the first quadrant for tooth No. 15 (Figure 4). It was exposed by using the laser machine and a traction eyelet was bonded on it, after evaluating the position on a cone-beam computed tomography (CBCT). Its position inside the bone was with the root blocked at the level of the buccal alveolar bone and the accessible face for bonding was the lingual one (Figure 5). Due to the high rise of fenestrations during rotation, it was decided

that it will be kept in a 180°-rotated position, so with the lingual face on the buccal side and that some selective grinding will be performed at the tip of the buccal cusp to remove interferences when the prosthetic restoration will be made in the fourth quadrant. Class II elastics were used to maintain class I at the canine level and to mesialize the lower second molars, in order to reduce the edentulous ridges.

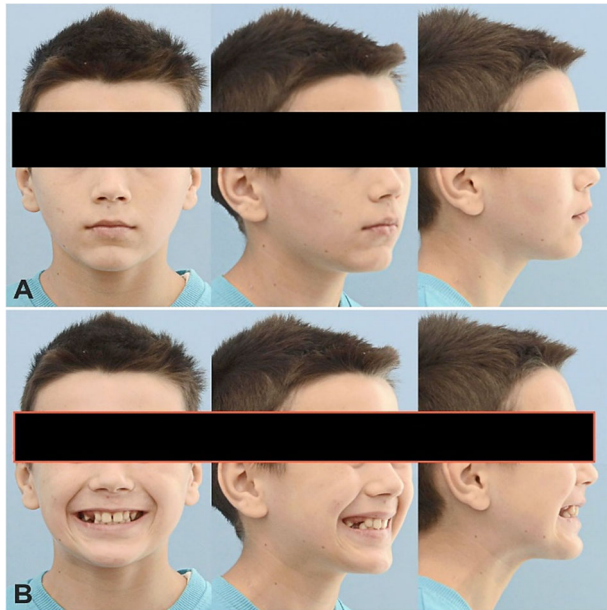
The most difficult situation was found in the second quadrant, where both the first and second premolars were missing. Fortunately, the first molar was kept by the general dentist due to less extended destruction so by its mesialization all the spaces were closed in the upper arch. We were able to achieve good interproximal contact between the upper canine and the upper first molar. Two spaces remained in the lower arch at the level of teeth Nos. 36 and 46, that will be restored after the end of growth (Figure 6, A and B). The final established occlusion was class I at the canine level, class I molar on the right side and class II molar on the left side. The deep bite was reduced and the curve of Spee levelled (Figure 7, A–C). For the contention phase, a thermoformed 1 mm retainer to be worn at night was given for the upper part and a removable appliance with two plastic teeth for the lower part. The artificial teeth were designed to maintain the created spaces inside the dental arch and the vertical space (Figure 8). Also, teeth 37 and 47 will not be allowed to mesially incline. This innovative method could be performed due to the fact that both edentulous spaces were on the lateral sides, so the aesthetic demands were not very high. The patient must wear both appliances during the night. Future implants will be placed once the growth process finishes.



**Figure 1 – Initial occlusion and arches:** (A) Right view: class I at the canine level; (B) Frontal view: upper diastema, coincident upper and lower midlines, slight deep bite; (C) Left view: class I at the canine level; (D) Lateral occlusion right view: over eruption of tooth 14, unsupported lateral vertical dimension due to important destructions of right permanent first molars; (E) Lateral occlusion left view: over eruption of tooth 23, pronounced curve of Spee, unsupported lateral occlusion; (F) Upper arch: absence of teeth 15, 24, 25, important destruction of tooth 16; (G) Lower arch: absence of teeth 35, 44, 45, important caries on lower first permanent molars.



**Figure 2 – Initial radiological examination:** (A) Panoramic X-ray: the congenital absence of teeth number 18, 24, 25, 28, 35 and 45 is confirmed, together with the impaction of upper right second premolar; (B) Cephalometric X-ray: class I skeletal relationship between maxilla and mandible, on a normodivergent pattern.



**Figure 3 – Initial extraoral appearance of the patient:** (A) During resting position; (B) During smile. The face is well developed and symmetric and the profile is harmonious, the intraoral midlines are coincident with the face midline.

**Figure 6 – Final aspect of upper arch (A) and lower arch (B).** In the maxilla, the arch is continuous, and all the spaces were closed using the natural permanent teeth of the patient, while in the mandible the big edentulous ridges were orthodontically reduced for two future implant supported crowns.

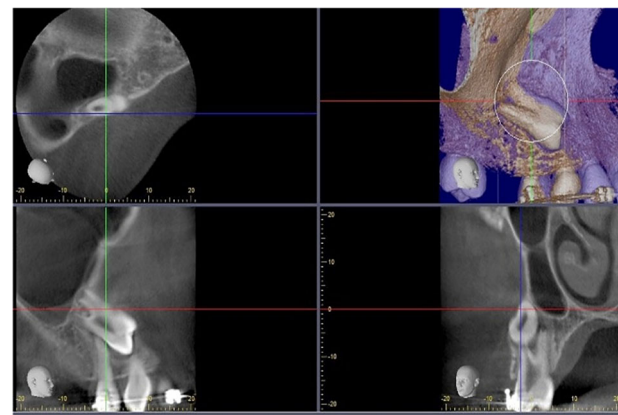


**Figure 7 – Final occlusion:** (A) Lateral right view; (B) Frontal view; (C) Lateral left view. Upper diastema was closed, and the midlines were kept coincident. Class I canine relationship was maintained on both sides and a stable occlusion was achieved on lateral sides (therapeutic class III on the right side and therapeutic class II on the left side).

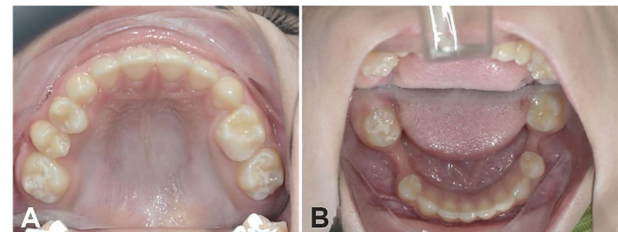
All the initial objectives were obtained, with the biggest achievement being the meeting of the function and aesthetic demands. The immediate start of the initial orthodontic phase was the key for a good result and also making the patient and the parents realize the importance of the treatment, starting from treating all the cavities on the remaining teeth, ensuring a good oral hygiene during the entire life,



**Figure 4 – Mid-treatment panoramic X-ray:** the progress of orthodontic treatment with upper and lower fixed appliances, with space maintained in the first quadrant for the traction of impacted upper right second premolar. The lower third molars continue to develop, while in the maxilla no other dental buds appear. L: Left; R: Right.



**Figure 5 – The position of the impacted tooth 15:** section from CBCT, with the root blocked at the level of the buccal alveolar bone and with the lingual face accessible for exposure and eyelet bonding. CBCT: Cone-beam computed tomography.



ending with wearing the space maintainer and the upper retainer in order to keep a good occlusion until the end of growth. This first phase represents only the preparation for later treatments, such as replacement of space maintainers with prosthetic restorations or extraction of lower third molars if they block the occlusion, so the multidisciplinary treatment is mandatory.



**Figure 8 – Contention appliance for lower arch with space maintainers. This device will be worn by the patient until the end of growth, when two implant-supported crowns will be placed in the third and fourth quadrants.**

## ☒ Discussions

Treating patients with several missing teeth represents a challenge for every practitioner. Besides the aesthetic concerns, patients with hypodontia present malposition of teeth, malocclusions, periodontal disease, lack of sufficient bone growth in the areas without dental buds. Sometimes, when the number of missing teeth is very high, the patient can also have digestive or phonetic problems. Every case requires multidisciplinary treatment, in order to establish proper functioning of the stomatognathic system and aesthetics, and also to obtain a stable result, which is difficult in a growing patient. Also, in cases having asymmetric teeth are missing, it is furthermore difficult to achieve a good occlusion. Several treatment options exist in these cases, such as maintaining temporary teeth when they are intact, closing spaces when permanent teeth are absent, either by spontaneous movement of teeth or by orthodontic treatment or, in mature patients, replacement of missing teeth by pontics, implants or even autotransplantation.

The causes for missing teeth can be local or general [4]. Agenesis can be syndromic or non-syndromic [1, 19]. Non-syndromic agenesis is more frequent than the syndromic one [19]. Syndromic agenesis is associated with problems in other ectodermal derivatives [19] and is a common finding in oral clefts, Down syndrome, Book syndrome, hemifacial microsomia, Rieger syndrome, ectodermal dysplasia and many others [1, 6, 13, 20–22]. The non-syndromic agenesis can be familiar (inherited) or sporadic [13] and is usually the only clinical finding [1]. For the inherited type, the genes involved can be autosomal dominant, autosomal recessive or with X-linked inheritance [23–26]. For the sporadic cases, the environmental causes can be orofacial trauma, infection, endocrine problems, maternal rubella during pregnancy, the use of certain medication (*e.g.*, Thalidomide), chemotherapy or radiotherapy during odontogenesis and many others [2, 4, 27–31]. The sex distribution varies with different studies, but most of them found that females are usually more affected, with a male-to-female ratio of 2:3 [11, 13, 32].

In sporadic agenesis, the missing units usually concern the most distal tooth of each group of teeth, such as the lateral incisor, the second premolar and the wisdom tooth [11, 13]. On the other hand, the central incisors, the canine, the first premolars and molars are more stable, and they

are usually involved when the number of missing teeth is more important [17, 31, 33]. But as a general rule, any tooth might be congenitally missing [13, 34]. The most frequent missing tooth is the third molar, followed by mandibular second premolar and maxillary lateral incisor [2, 4, 32]. In studies that don't consider the third molar, like the one from Polder *et al.*, the mandibular second premolar was missing in 41% of cases, the maxillary lateral incisor in 23% of cases, the maxillary second premolar in 21% of cases and the mandibular incisors in 6% of cases [32]. In Caucasian population, the most involved teeth when analyzing the agenesis are the second mandibular premolar and the maxillary lateral incisor [4], while Davis found in Asian population that the mandibular lateral incisor was the most involved tooth [35]. In a study on Iranian population, the most common missing tooth was the upper lateral incisor [11].

Unilateral agenesis is more common than the bilateral one [11, 32]. The maxillary lateral incisor agenesis is usually bilateral [32]. Other studies found that the most common symmetrical agenesis is the one of mandibular second premolar [7, 17]. If we talk about the severity of the agenesis, population studies suggest that most of the cases (around 80%) will have one or two missing teeth, 10% will present four or more congenital missing teeth and less than 1% will have six or more missing teeth [9, 22, 31].

In the case presented above, the congenital missing teeth were asymmetric in the maxilla (one first premolar and two second premolars) and symmetric in the mandible (two second premolars). Also, all the upper third molars were missing. After the decision of the general dentist to extract three out of the four first molars, the case became more symmetrical. The total number of missing teeth was four without counting the third molars and six if including them. For patients with one or more missing teeth, there are several problems that can occur. The aesthetic concern is of major importance, especially when the agenesis involves the frontal teeth or when it is asymmetric, deviating from the midline. If there are several units missing, the patient can present lack of bone growth which impacts the aesthetics (one side can be more prominent than the other), making it more difficult to orthodontically close the space or place an implant. Also, it can lead to malocclusions such as class II by Angle when the mandible is underdeveloped or class III by Angle when the missing teeth are located in the maxilla. As the number of missing dental units increases, the functions of the oral cavity suffer, like mastication or phonation. This can lead to digestive problems and social implications. When a tooth is missing and the arch is discontinued, malpositions occur at the level of adjacent and antagonistic teeth. These malpositions can lead to malocclusion and also periodontal problems.

Most of the time agenesis is diagnosed around the age of 10, when the patient is still growing. The treatment must take this aspect into consideration, therefore most of the time it is multidisciplinary. It can involve every member of the dental specialties, depending on the severity of the case, the localization of the missing teeth inside the dental arch and also the type of malocclusion associated, where applicable [8]. Many factors must be included in the treatment decision: the age of the patient, the number of

missing teeth and of those present inside the dental arch, the existence of impacted teeth, the condition of the remaining teeth and of the support structures, the occlusion [3, 4]. The treatment can be represented by one simple restoration or by surgery and complex restorations [36]. AlShahrani *et al.* recommends closing the remaining space inside the dental arch using the neighboring teeth when there is a small number of missing units and orthodontic redistribution of spaces and prosthetic replacement in more complex cases [4]. Rosa *et al.* concluded that in cases of missing maxillary lateral incisor, space closure and the extrusion of the canine and intrusion of the premolar ensures a good gingival level and aesthetics and does not compromise the periodontal health [37]. In cases with missing premolars, the decision on whether the treatment plan involves space closure or maintaining or opening the spaces depends on factors such as: patient's age, the condition of the remaining teeth, if the deciduous molars are present, their condition, the crowding inside the dental arch, type and severity of malocclusion, personal factors related to the patient [38]. For example, in young patients with some anterior crowding and missing second premolars, if the second primary molars are extracted at an optimal age, before the growing spurt peak, the crowding can be relieved and the lateral space can close spontaneously, without orthodontic treatment [39].

In cases where spaces are kept, prosthetic replacement can be represented by removable partial dentures, fixed partial dentures, implant supported dentures, resin bonded bridges. Redistribution of the spaces by the aid of orthodontic mechanics helps to create an adequately sized area for prosthetic replacement. In these cases, fixed appliances are preferred because they ensure parallel teeth and parallel roots, which are needed for an optimal prosthetic restoration [31]. The resin bonded fixed partial dentures can be used only when a few teeth are missing, but they are the most conservative method of restoration since minimal neighboring teeth reduction is needed [40]. Conventional fixed partial dentures can be used when more teeth are missing, but with the main disadvantage of important reduction of healthy tooth structures [41]. In these cases, the parallelism created by the orthodontist with the pre-prosthetic treatment can reduce the quantity of dental structure reduction [31, 40, 41]. Implant supported dentures can be used both in anterior and posterior areas and they don't require healthy tissue removal of the adjacent teeth. Their main disadvantage is that they must be applied only after the end of the growth, because if applied earlier, they might remain infraocclusally [31].

In recent years, doctors have tried to find solutions to utilize one's teeth where they are needed. For example, in cases with missing premolars, premolars from other quadrants where crowding is present, or third molars can be autotransplanted. It is important that the extraction is made with low forces and with minimum impact on periodontal and cement structures. The transplanted tooth should have at least three quarters of the root already developed. Also, better responses by pulpal revascularization are obtained if the apex is open [31]. Other surgical adjuvant procedures may be needed, especially in complex agenesis cases, since bone is formed during tooth eruption. If many

teeth are missing, the bone might be insufficient in width or height and bone grafts must be performed. Other procedures like widening the alveolar crest, alveolar distraction, bone augmentation can be applied [40]. In cases with missing second premolars, like in our case, the treatment should be started in early adolescence, when most of the growth has happened and all the permanent teeth are erupting [42].

## ☐ Conclusions

Complex agenesis cases require multidisciplinary treatment. The orthodontic treatment with fixed appliances can help to close spaces when this is possible or ensure a good distribution of spaces when the edentulous ridges are too long. It can create a sufficient amount of space for future prosthetic restorations and also the parallelism of crowns and roots. When optimal spaces are obtained, a decision must be made for the type of restoration to be performed. If implants are the chosen solution, then spaces must be obtained and kept until the end of growth. All this must be done with the main objective of ensuring the patient's aesthetics, occlusion stability and function. In our case, we managed to bring one impacted premolar inside the dental arch, close all the remaining spaces in the maxilla and reduce the big edentulous lateral ridges to only two spaces for future implants in the lower quadrants, at the same time ensuring long-term stability, aesthetics and proper conditions for all functions. Maintaining spaces until the bone achieves maturity was done using a removable orthodontic appliance that incorporates two artificial teeth. Immediate intervention was very important in this case, since six teeth were congenitally missing and three others were extracted, so a growing patient was deprived by nine centers of development. Orthodontic movements for closing spaces that were performed on both arches, stimulated bone production and helped to create a perfect path for later multidisciplinary interventions, simplifying or eliminating the surgical procedures that will later have to be performed.

## Conflict of interests

The authors declare that there is no conflict of interests.

## Authors' contribution

Oana Cella Andrei and Mirela Ileana Dinescu equally contributed to this article.

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