CASE REPORTS

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Unilateral supplemental maxillary lateral incisor: report of three rare cases and literature review

OANA CELLA ANDREI[1)], CĂTĂLINA FARCAŞIU[2)], RUXANDRA MĂRGĂRIT[3)], MIRELA ILEANA DINESCU[4)], LIVIA ALICE TĂNĂSESCU[1)], LUMINIŢA DĂGUCI[5)], MIHAI BURLIBAŞA[6)], CONSTANTIN DĂGUCI[7)]

[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 Restorative Odontotherapy, Faculty of Dentistry, "Carol Davila" University of Medicine and Pharmacy, Bucharest, Romania*

[4)]*Private Orthodontic Practice, Bucharest, Romania*

[5)]*Department of Prosthodontics, Faculty of Dentistry, University of Medicine and Pharmacy of Craiova, Romania*

[6)]*Department of Dental Techniques, Faculty of Midwifery and Nursing, "Carol Davila" University of Medicine and Pharmacy, Bucharest, Romania*

[7)]*Department of Prevention of Oro-Dental Diseases, Faculty of Dentistry, University of Medicine and Pharmacy of Craiova, Romania*

Abstract

Additional teeth to the normal dentition are called supernumerary teeth; they can be found in higher rates in permanent than in deciduous dentition, in both sexes, associated or not with other diseases or syndromes. The aim of this article is to report three rare cases of unilateral supplemental lateral incisors, in Romanian child and adult patients, and to evaluate the treatment options in each case, considering particularities such as age, physiognomy, associated issues and occlusion. The differential diagnosis between a supernumerary lateral incisor in formation and a rudimentary is sometimes difficult to make using only panoramic X-ray, so a cone-beam computed tomography analysis can be indicated. Usually, due to the pathological issues that they can cause, the supplemental maxillary lateral incisors are removed; although, in some cases, it may be necessary to remove the normal tooth, for example in cases of major crowding, in which the normal tooth is more displaced from the line of the arch than the supplemental one, or when there is a lack of periodontal support surrounding the normal tooth. This article highlights that even if, in younger patients, the majority of these teeth is extracted because later in life they can cause difficulties in orthodontic and prosthetic treatment, there are still rare cases which do not necessitate treatment, because they are asymptomatic, rather well aligned and go unnoticed. Also, the article includes a detailed literature review of reported cases of supplemental maxillary lateral incisor.

Keywords: supernumerary teeth, supplemental maxillary lateral incisor, flexible treatment plan.

Introduction

Supernumerary teeth have multiple etiology factors, such as hyperactivity of the lamina, trauma, phylogenetic theory, atavism, environmental or genetic (heredity) linked factors, dichotomy of the dental germ [1]; they are more prevalent in permanent dentition and in the premaxilla region [2, 3]; males are reported by some authors to be more affected than females [2]. Some of them are so similar in form, shape and position with the normal ones that the decision on which one to extract is difficult. Still, early diagnosis is important because, with rare exceptions, they can cause problems in eruption, physiognomy and occlusion [4]. Multiple supernumerary teeth are usually found in various medical conditions and syndromes (cleido-cranial dysostosis, Ehlers–Danlos syndrome, Anderson–Fabry's syndrome or Gardner's syndrome, cleft lip and palate, chondroectodermal dysplasia, Ellis–van Creveld syndrome, Nance–Horan syndrome, Rubinstein–Taybi syndrome and trichorhinophalangeal syndrome) [5, 6]; it is rare to find them in non-syndromic patients. The three cases of deciduous and/or permanent supplemental maxillary lateral incisors presented below are rare, because of their presence in patients with no syndrome.

Supplemental teeth are supernumerary teeth that are similar with the adjacent teeth; they mime their normal anatomy and can be well aligned in the arch [7]. After Primosch [8], there are two types of supernumerary teeth, some of normal size and shape, named supplemental, and the others smaller and of abnormal shape, named rudimentary teeth (conical, tuberculate or molariform). The differential diagnosis between a rudimental and a supplemental tooth is made based on clinical, radiographic and cone-beam computed tomography (CBCT) examination; also, they must be differentiated from the geminated or fused teeth; the treatment plan is made accordingly [9]. The presence of more teeth than in the normal dental formula is discovered most of the time by accident, during mixed dentition period, since parents are frequently in the situation of bringing their children to the first dental examination, following the eruption of the permanent anterior teeth or first molars.

Cases of supplemental lateral maxillary incisors are rarely reported; they can be found unilateral or bilateral. The treatment plan is based on the information obtained through X-rays and CBCT; also, it is made taking into account factors, such as the age of the patient, the position of the supplemental tooth and the consequences that its presence creates on the arch, such as widened follicular space, cyst formation, resorption or blocking of the eruption of the permanent tooth, crowding, difficult hygiene, esthetic problems or malocclusion [10, 11]. Supplemental teeth are found rarer than dysmorphic supernumerary teeth, especially in non-syndromic patients [12]. These three reported cases are illustrative for the unilateral supplemental lateral incisor, in patients without any associated disorder or syndrome, the first being an extremely rare one, since it is not associated with almost any adverse effect and it was detected at an adult age, by chance, during a dental check-up made for another issue.

Case presentations

Case No. 1

A 29-year-old male patient came to our Office for a fractured amalgam filling on the first permanent left mandibular molar (3.6). On intraoral examination, we noticed a minor anterior crowding, midline shift, poor hygiene and dental caries and the presence of a maxillary supernumerary tooth, a fully erupted right supplemental lateral incisor situated between 12 and 13 (Figure 1).

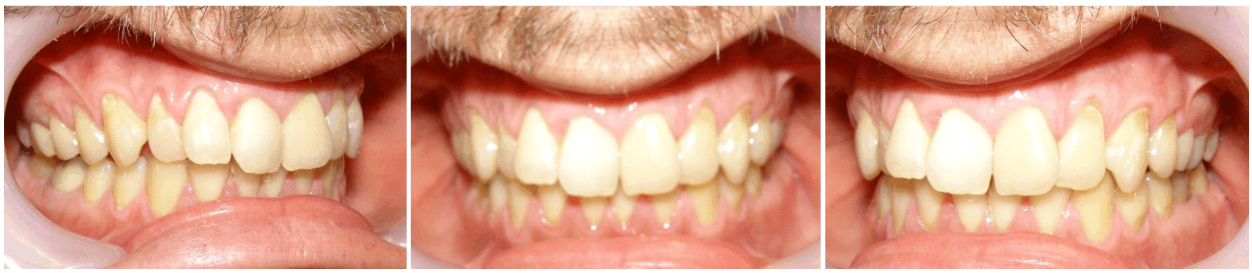

Figure 1 – ***The aspect of supplemental maxillary lateral incisor, Angle Class II occlusion, midline shift and minor crowding.***

All lateral incisors were about the same shape and dimension. On the panoramic X-ray, we discovered that the crown and root were completely formed (Figure 2).

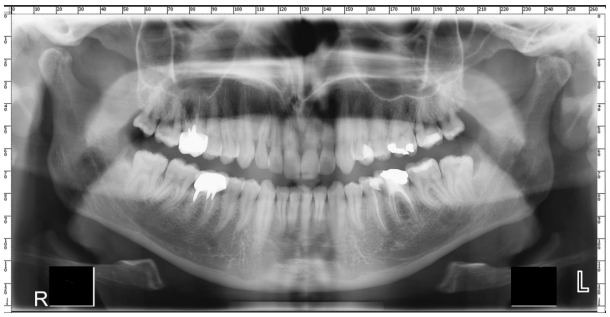

Figure 2 – ***Panoramic radiological investigation showing the presence of one supplemental maxillary lateral incisor normally aligned in the right part of the jaw. L: Left; R: Right.***

The patient presented no other supernumerary tooth and also no medical condition, so an autosomal dominant pattern of inheritance was supposed. Both right lateral incisors had almost the same crown and root dimension; therefore, it was difficult to say which one is the supplemental one, except for the enlargement of the periodontal space of the lateral incisor situated closer to the cuspid. There is a false Angle Class III occlusion on the right side, both in cuspid and molar, due to the presence of the supplemental incisor. There is a pattern of deep-bite with sagital inocclussion and minor crowding in the upper arch. Without any extractions, an attempt to align the teeth will increase the space of inocclusion. Although usually these teeth cause serious problems in occlusion and physiognomy, in this case no such associated factors were present, so the decision to keep the tooth in place was the best option for the patient. Still, there is an interference during lateral shift on the right side, so minor occlusal corrections should be made. This is a rare case of a young adult patient, coming to the Office for other reasons than his unilateral supplemental maxillary lateral incisor, which he completely ignored, and without any serious pathology caused by it.

Case No. 2

An 8-year-old male patient came to the Dental Office accusing pain after fracturing a filling in the temporary left first molar area (6.4). On the intraoral examination, we discovered on the right part of the maxillary arch a supplemental permanent lateral incisor; there were two permanent lateral incisors, one aligned and the other one mesiopalatally rotated by 90º and situated between the first one and the central upper incisor, disturbing the occlusion. On the radiographic examination, we could see crowding in the upper arch, with the right cuspid situated high in the bone and pressing on one of the lateral incisors root. Both lateral incisors had open apices, so the pressure from the cuspid eruption would surely affect the root development, the major risk being external root resorption (Figure 3, a and b).

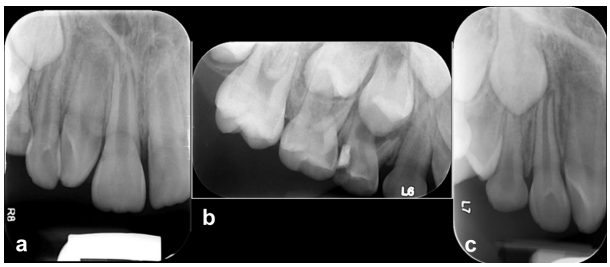

Figure 3 – (*a–c) Retroalveolar images from different incidences showing the supplemental's position and its consequences on the adjacent teeth during the two years of follow-up period.*

The parents did not accept any intervention, agreeing only to monitor the evolution. During the first two years of follow-up period, the tooth was asymptomatic; periodical radiographic observation and careful examinations took place in order to avoid the risk of affecting the normal teeth eruption. After this period, the supplemental lateral incisor was still in an ectopic position (Figures 3c and 4); because both teeth were healthy, the decision to remove one or the other of the incisors was made taking into account the position on the arch, especially the rotation, the dimension and the shape of both teeth comparing with the left lateral incisor, and also the periodontal support. Parents understood the complications and risks related to the dental crowding, the periodontal consequences and the malocclusion. It was also explained to them that a fixed orthodontic treatment can be initiated when the eruption of all premolars and permanent canines will take place. The medical team treatment plan, agreed with the parents, was to extract the rotated incisor due to esthetical reasons related to dental crowding (Figure 5), associated with parent's non-compliance with the consecutive orthodontic treatment.

Figure 4 – ***Panoramic radiological investigation showing the presence of one supplemental maxillary lateral incisor mesiopalatally rotated in the right part of the jaw. L: Left; R: Right.***

Figure 5 – ***Panoramic radiological investigation showing the progressive closure of frontal space after extraction and the progress in the canine's eruption. L: Left; R: Right.***

Case No. 3

A 5-year-old female patient reported with the main complaint of unerupted upper front right central incisor; no other medical conditions were present. Parents' concern was focused on this supplemental lateral incisor (5.2), because it was affecting the developing dentition, creating a disturbance in erupting adjacent permanent tooth. Oral examination showed the presence of the maxillary left incisor with the absence of the right one, and the presence of two deciduous upper right lateral incisors. Radiographic examination (orthopantomography) detected a supernumerary tooth in formation superimposed inside the jaw upon the permanent lateral right incisor (12) (Figure 6); a differential diagnosis must be made between a supplemental permanent lateral incisor and a rudimentary tooth.

Figure 6 – ***Panoramic radiological investigation showing the oblique supplementary tooth blocking the eruption of the central right incisor.***

For the differential diagnosis and especially for the determination of the precise location of the tooth and the visualization of its relationships with the adjacent structures, a CBCT was indicated considering that three-dimensional volumetric data will help to establish the optimal therapeutic option and will provide safety during the surgical intervention (Figure 7). Together, CBCT images and panoramic radiograph revealed the location, position, orientation and morphology of the supernumerary tooth. The CBCT images show a 45º inclined upper lateral right supernumerary tooth, with the incisal margin facing down and mesially, with the crown completely mineralized and which blocks the normal eruption of the central and lateral right permanent incisors (11 and 12) (Figure 8). The treatment plan included the extraction of the deciduous right central, lateral and supplemental lateral incisors (teeth 5.1, 5.2, 5.2 supplemental) and of the permanent supplemental lateral incisor (1.2 supplemental). Giving the lower position of tooth number 1.2, that will block the spontaneous eruption of the right permanent central incisor, it is better that, for the proper eruption and alignment of both upper central incisors, active orthodontic treatment is initiated.

Discussions

The supplemental lateral incisor is rare, difficult to differentiate from normal lateral incisor and the therapeutic attitude must be adapted to the specific clinical situation of each patient. For most cases, it is necessary to have an early diagnosis in order to prevent complications by initiating the treatment; although, in some rare cases, these teeth can be discovered accidentally, later in life, if they do not interfere with the development or eruption of the permanent adjacent teeth. The three cases without any systemic pathology presented here are different and highlight the possible clinical variations and the flexibility of the treatment plan. In our young adult's case, the permanent supplemental lateral incisor was erupted and caused only a minor rotation of the adjacent lateral incisor, causing no symptoms; the patient was unaware of having it. No extraction was needed for this case. In the 8-year-old male patient's case, the

permanent supplemental lateral incisor was erupted, but was not aligned and caused a major crowding. The treatment plan was to extract the lateral incisor from the normal dentition and to keep the supernumerary, since both had the same root development, but the last one was better positioned inside the dental arch. In the 5-year-old female patient's case, the same anomaly of number repeated in both dentitions on the same position inside the arch, but in the permanent one, the supplemental tooth was smaller and mispositioned, hence causing the impaction of the central permanent right incisor (tooth number 1.1).

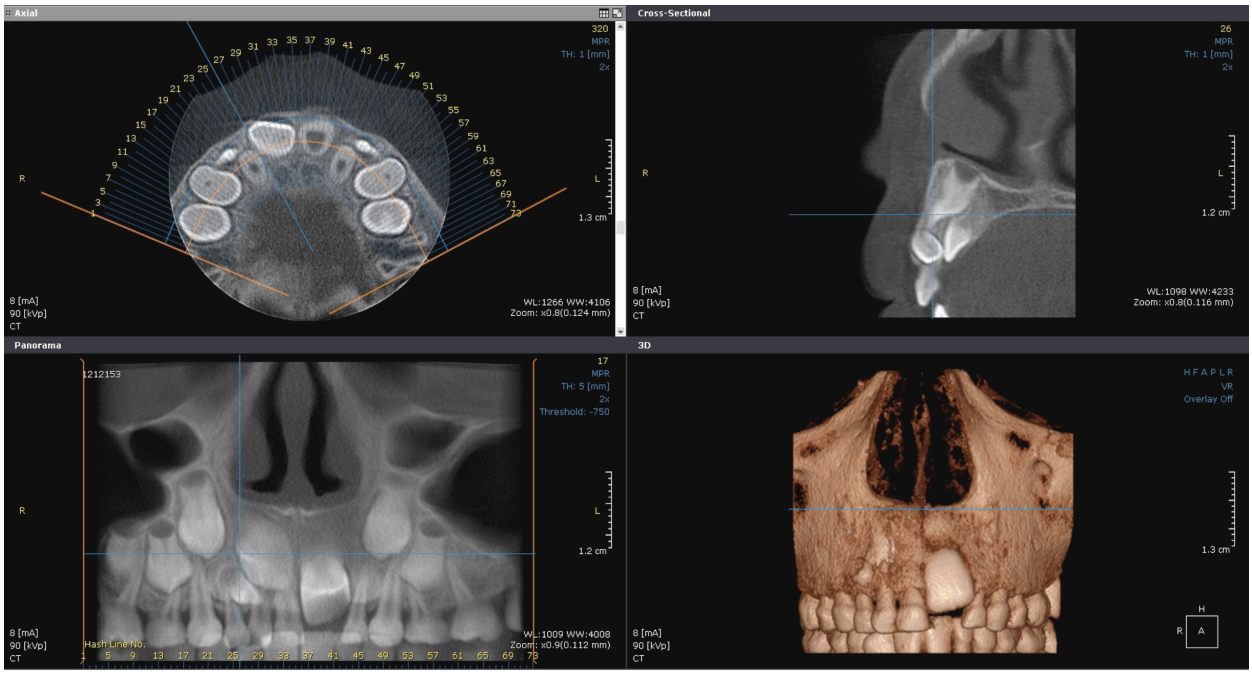

Figure 7 – ***CBCT image with axial, sagittal and coronal views and indirect volumetric rendering image showing the supranumerary tooth in the right upper jaw erupting and blocking the eruption of the central incisor. CBCT: Cone-beam computed tomography.***

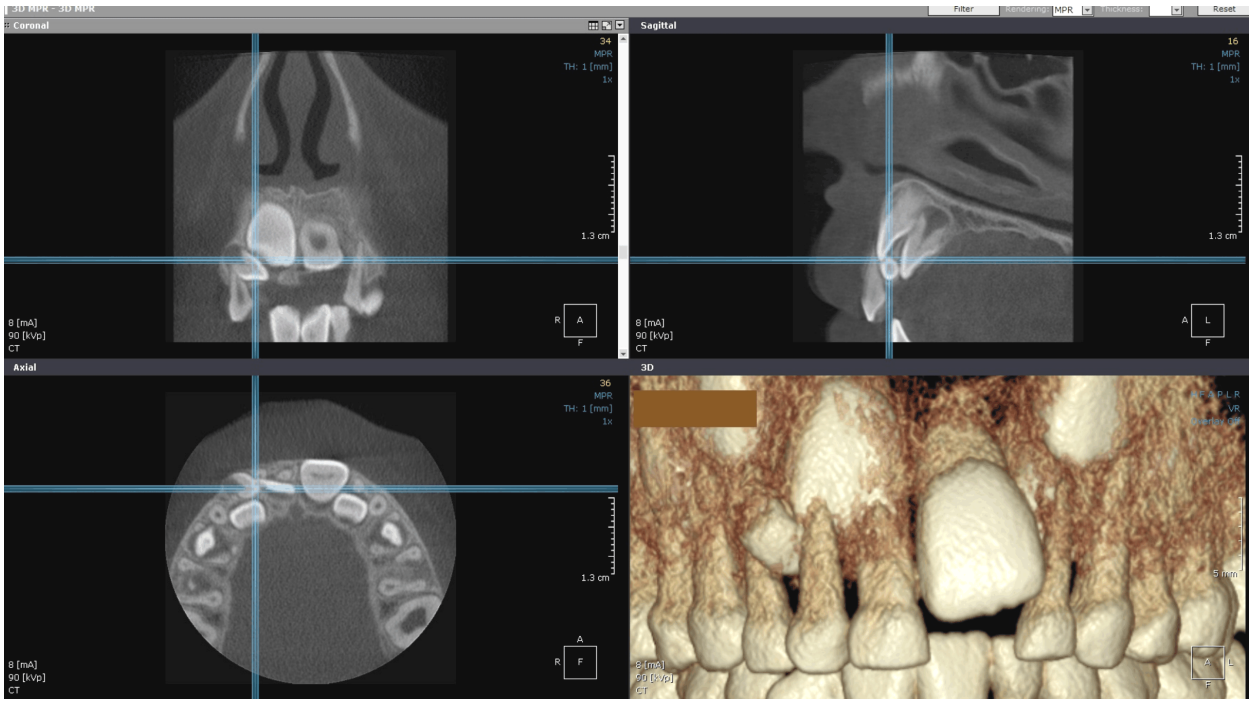

Figure 8 – ***CBCT image offering more details about the position of the supranumerary and clarifying the necessity of its extraction. CBCT: Cone-beam computed tomography.***

Supernumerary canines and lateral incisors have a low prevalence; some authors found 2.8% from all supernumerary teeth [13]. Supplemental teeth are the duplicates for the teeth from the normal dental formula; they can be molars, premolars or incisive [14]. Although reported data of supplemental teeth exists in literature, for the supplemental lateral incisors, and especially for the deciduous ones, this is rather scarce [1]. In a study made by Jung *et al.*, from 193 patients with 241 impacted supernumerary teeth, only seven were supplemental [15].

Some authors reported in a study of 739 7-year-old Caucasian children only two cases presenting supplemental maxillary lateral incisors (less than 1%) [16].

Some authors are stating that hyperdontia in primary dentition should alert the clinician to the possibility of repeating itself in the permanent one [4, 17]; still, it is frequently overlooked, partly because of their quiet eruption and normal alignment, partly because of the fact that most children do not visit the dental office prior to the eruption of their first permanent teeth [18]. A study made on 62 cases of supplemental permanent maxillary lateral incisors found that 30% of them have had supplemental deciduous lateral incisors [19]. A case reported by Bargale *et al.* presents a 4-year-old female patient with bilateral supplemental primary maxillary lateral incisors, decided to be kept under observation because they were completely erupted and causing no issues [20]. Hekmatfar *et al.* present a similar case of a non-syndromic 9-year-old boy, having non-erupted yet bilateral supplemental primary maxillary lateral incisors discovered during radiographic examination, with mixed dentition and no disturbing effect on the eruption of the adjacent teeth; parents and siblings were examined and did not present any supernumerary teeth; no treatment was performed, due to lack of economic means [21]. Lehi & Kaur present a case of a 5-year-old boy having a deciduous supplemental lateral incisor; his mother declared that she and her mother also had the same [22]. Beere *et al.* reported a case of 5-year-old twin boys from a rural area of northern Namibia with mirror supplemental primary lateral incisors, one right and one left; on the radiographic examination, also mirror supplemental permanent lateral incisors were discovered along with symmetrical caries pattern, suggesting a homozygotic relationship that due to local situation could not be confirmed by further investigations [23]. Also, a case reported by Shanmugha *et al.* present an 8-year-old female patient with a unilateral supplemental primary maxillary lateral incisor and an unerupted permanent supernumerary, discovered on the radiological examination, related with the permanent lateral incisor [24], while Bhat *et al.* present a 10-year-old male patient with the same situation, causing anterior crossbite [25]. Bala *et al.* present an 8-year-old boy with two deciduous lateral incisors in the right maxillary quadrant, one of them mobile; orthopantomography revealed the presence of the supplemental successor and of another supernumerary teeth in the molar region [26]. Other authors reported a case of unilateral supplemental primary maxillary lateral incisor without a permanent supernumerary, which further complicates the question not yet understood of etiology [27].

Wedrychowska-Szulc & Janiszewska-Olszowska made a study on panoramic X-rays and casts in order to assess the morphology of these teeth and the malocclusions. The dimensions found were slightly smaller than the normal ones; this being an argument for the dichotomy theory etiology. The consequences of their presence were midline shifts, crowding, space deficiency, ectopic eruption, excessive overjet. Some of them were not erupted; the frequency was the same in both sexes [28]. Aguiló *et al.* opiniate that geminated primary teeth are predicting supernumerary teeth in permanent dentition [29]. Other authors reported in a series of three cases of Chinese children the associations of supplemental permanent maxillary lateral incisors with talon cusp on the primary predecessors [30]; they agree with the use of a conservative approach in some specific cases in which the supplemental tooth can be used, for example, as a donor tooth in autotransplantation and recommend a regular follow-up every four to six months to monitor the degree of a possible impaction. They opiniate that a decision regarding which tooth is better to be extracted is more appropriate to be taken when both the supplemental tooth and the normal one are erupted, in order to comparatively evaluate their morphology, size and alignment. Also, this approach avoids the extraction of an unerupted tooth under a general anesthesia.

Because of their normal shape and size, supplemental teeth are less detected by parents and they are easily overlooked if they are not causing cyst formation, ectopic eruption, delayed eruption or esthetic and hygiene problems, by crowding, rotations or diastema. A short communication was made by Mitchell & Ahluwalia, describing a facial cellulitis in an 8-year-old male patient, caused by an invaginated permanent supplemental right lateral incisor; antecedents reported the existence of another supplemental right lateral incisor in the deciduous dentition, on the same side [31]. The decision to remove a supplemental tooth is made when it is causing pathological issues; however, in very rare cases, such as the young adult male presented in this article, the supplemental lateral incisor on the right side was left in place, since it was mostly aligned on the arch and the occlusion was functional.

Other cases of adults having discovered their supplemental lateral incisors later in life were also reported by Premkumar *et al.* [32]. Also, Rodrigues *et al.* reported a case of a 45-year-old male patient having a unilateral supplemental incisor in which they showed that their treatment plan should be carefully elaborated, the supplemental tooth cannot be considered isolated [33]. Despite the age of the patient, because of crowding, deep bite and midline shift, it was decided to extract the supplemental and to align the teeth through fixed orthodontic treatment. In cases where the teeth are equally formed in both crown and root, Hattab *et al.* [2] recommend to extract the one that is the most displaced. Similar, in a 20-year-old male patient with bilateral supplemental lateral incisors and Class II division 2 malocclusion, Urala & Divya recommended extraction and orthodontic treatment for the closure of the spaces [34].

Yildirim & Bayrak [4] also recommended the extraction of the bilateral supplemental lateral incisors when they prevent the eruption of permanent ones, insisting on the importance of early diagnosis and treatment during the period of mixed dentition. The case reported is similar with our third case, having both primary and permanent supplemental lateral incisors, only bilateral, and the treatment plan was also similar, with extraction and follow-up. Bhullar *et al.* published a case of a 14-year-old female patient with permanent bilateral supplemental lateral incisors having the same form and dimension with the normal ones, both regarding the crowns and the roots. All of them were kept because of existing spaces between her teeth on both jaws; fixed orthodontic treatment was

also advised [35]. Despite the fact that these supernumerary teeth usually cause crowding, in this case, the spaces between the frontal teeth were the reason for keeping them on the arch, helping in closing the spaces and obtaining a better esthetic result. On the contrary, Singla & Negi reported a similar case, but in which the two supplementals caused serious crowding and there was also an arch length discrepancy, which determined the decision to remove them in order to align the remaining teeth [36].

Chalakkal *et al.* reported a case of an 11-year-old boy having two bilateral supplemental incisors with anterior crowding; the treatment plan was to extract them and to perform an orthodontic treatment [37]. Lo Giudice *et al.* reported a patient with multiple supernumerary teeth, among them being also present the supplemental maxillary lateral incisors in association with a mesiodens, which were removed in order to allow the eruption of the normal dentition [38]. Some authors claim that in the presence of two supplemental lateral incisors the treatment plan must include their extraction altogether with that of all first premolars, in order to obtain the best esthetic result [39, 40]. Reports of cases of bilateral supplemental lateral incisors are also found in other articles; sometimes, one supplemental is similar to the normal one, while on the other part they have a fused crown [41].

Conclusions

In absence of any associated syndrome, patients with supplemental lateral incisor are rare. In Romania, no studies especially made on supplemental lateral maxillary incisors have been found yet. Present cases are unusual because are highlighting the great variations in clinical situations and consequently in treatment plan of such patients having such supernumerary teeth, without associated medical conditions, showing the fact that extraction is only one treatment option, but it is not always necessarily the most appropriated one. The therapeutic decision must be flexible, based upon the age, shape, dimension, position, pathological issues, aesthetic, hygiene and existence of any associated problems; thus, it has to be adapted to the particularities of each clinical case.

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

Ruxandra Mărgărit, Assistant, MD, PhD, Department of Restorative Odontotherapy, Faculty of Dentistry, "Carol Davila" University of Medicine and Pharmacy, 37 Dionisie Lupu Street, Sector 2, 020021 Bucharest, Romania; Phone +40747–440 013, e-mail: ruxandra.margarit@gmail.com

Livia Alice Tănăsescu, Assistant, MD, 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

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