CASE REPORTS

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A rare case of mandibulary parapremolars in siblings: case report and literature review

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Abstract

Supernumerary teeth in permanent dentition are influenced by hereditary and environmental, having clinical variations such as location, number and morphology. Parapremolars can be found as a single supernumerary tooth or as multiple, usually in the lower jaw, sometimes in association with other types of supernumerary teeth. Familial inheritance often involves more than one generation. This paper presents a case of two brothers, one with only one parapremolar and a good oral status and hygiene, the other with poor hygiene, multiple parapremolars and one mesiodens, causing various complications. The aim of this paper is to show the presence of parapremolars in brothers with no other medical relevant history, located in the same site, and to point the influence of genetic factors in the etiology of supernumerary teeth. Along with a review of the literature, this article shows the importance of an early diagnosis and follow-up using successive panoramic radiographs of such cases, in order to limit or prevent the inflammatory, carious, periodontal and occlusal complications that can seriously affect both the jaws and the normal dentition.

***Keywords*:** supernumerary teeth, parapremolar, mesiodens, siblings, heredity factor.

Introduction

Supernumerary teeth can be found in healthy patients or can be associated with DNA mutations or some general disorders, with a variability also involving age and ethnic characteristics. Although some of them succeed to erupt, in most cases they remain impacted. Their etiology is considered to be multifactorial, as a combination of genetic and environmental factors. Parapremolars are supernumerary teeth found in addition to normal dentition and situated near the normal premolars. The mandibular parapremolar is one of the most frequent supernumerary teeth, along with those appearing on the premaxilla region. In the same patient, the parapremolar can be found as a singular supernumerary tooth, or as two or multiple premolars bilaterally located or even in association with other types of supernumerary teeth (*e.g.*, the mesiodens, situated between the two central incisors). This paper presents a case of siblings, one with a singular parapremolar and the other with an association of three mandibular parapremolars and a maxillary mesiodens. Its aim is to report the presence of parapremolars in brothers with no other medical relevant history, located in the same site, and to point the influence of genetic factors in the etiology of supernumerary teeth. Also, this article advocates for an early diagnosis of such cases, in order to prevent the inflammatory, carious, periodontal and occlusal complications.

Usually, the morphology of the parapremolar is similar to the one of a premolar. A localized hyperactivity of the dental lamina is the most accepted theory for parapremolars; they can also develop at a later age. Mesiodens often remain impacted [1], and can be found either as a single supernumerary tooth, or as multiple supernumerary teeth; in this case, they are called mesiodentes. Mesiodens is generally smaller than the normal central incisors, having a cone shaped crown and a single root, located palatally between the maxillary central incisors. They can be found in normal position or inverted [2]. Some authors found that the inverted mesiodens remain impacted in about 67% of cases [3]. Also, Tay *et al.* reported that the most of the mesiodentes remain impacted [4], and Nazif *et al.* found that about 80% of the impacted mesiodentes are situated in the palatal area [5].

In Romania, there are few studies regarding the variation of the normal number of teeth (supernumerary teeth or anodontia). Bratu *et al.* found a 3% prevalence of supranumerary teeth in a regional study; most cases presented only one supernumerary tooth, frequently erupted in normal position [6]. A study conducted in another region of Romania showed that supernumerary teeth are

rare and most frequently located in incisal area [7]. A genetic study showed that supernumerary teeth are often related to a family history, with phenotypes determined by mutations in different genes [8]. Dumitrescu *et al.* pointed that, in some late diagnosed cases, they can generate important inflammatory complications and need to be extracted [9].

Case presentations

Case No. 1

A 26-year-old male patient with a good dental status and a very good oral hygiene came to the clinic for restorative and orthodontic treatment. On the panoramic X-ray, we discovered the presence of a supernumerary tooth, a third premolar, situated in the right part of the mandible between the roots of the two normal premolars (Figure 1). The patient was not aware of the presence of this tooth, but declared that his brother and also a brother of his grandmother have hyperdontia. This parapremolar was asymptomatic, with no associated complications and in a final stage of development and mineralization. The patient agreed with the treatment plan and signed the informed consent. After finishing the restorative procedures, the patient was referred to an orthodontist who decided, following surgical advice and evaluating the risks and benefits, to remove the third molars but not the supernumerary mandibular premolar. This decision to leave the parapremolar *in situ* was motivated by the reason that the risk of damage for the adjacent structures during its removal would outweigh the benefits. Orthodontic treatment was successfully finished in 20 months. The patient was seen for follow up one year later to monitor the position of the parapremolar and a second set of panoramic and periapical radiographs (Figures 2 and 3) showed that it created no damage to the surrounding tissues. No clinical signs were observed or reported by the patient (Figure 4). The recommendation was to periodically monitor the supernumerary tooth by clinical and radiographic examination.

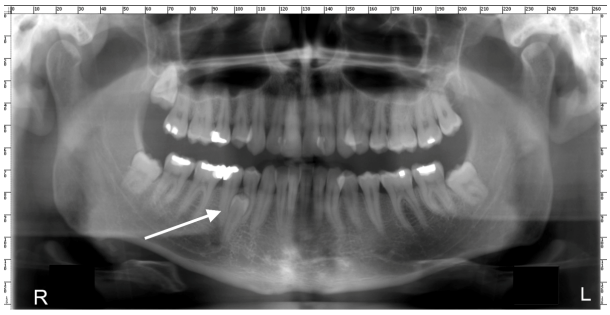

Figure 1 – ***Case No. 1: Panoramic radiological investigation at presentation showing the presence of the parapremolar in the right part of the lower jaw. R: Right; L: Left.***

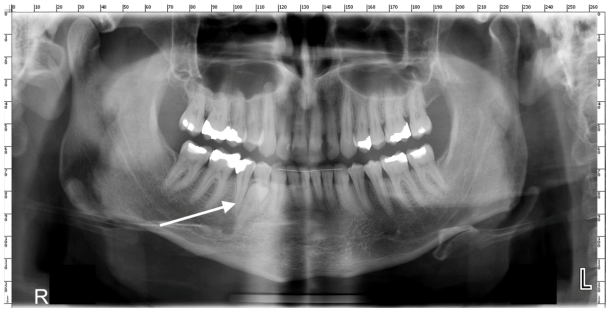

Figure 2 – ***Case No. 1: Panoramic radiological investigation after the orthodontic treatment showing the same position of the parapremolar in the right part of the lower jaw. R: Right; L: Left.***

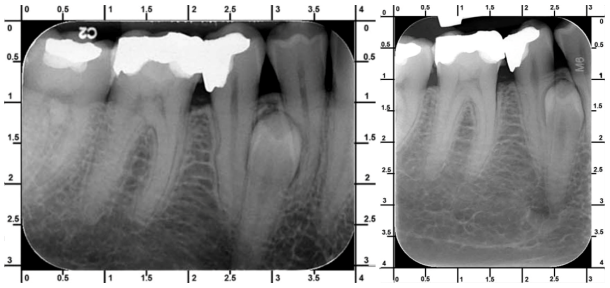

Figure 3 – ***Case No. 1: Retroalveolar images showing the parapremolar's position before (left) and after (right) the treatment.***

Figure 4 – ***Case No. 1: Clinical aspect of the patient after the restorative and orthodontic treatment.***

Case No. 2

A 23-year-old male patient, the first patient's younger brother, came to the office having a much different oral health situation, with very poor hygiene, a lot of caries and tartar. The radiographic examination revealed the presence of a parapremolar located in the same area as his brother, the mandibular right area, but also another two parapremolars, located in the mandibular left premolar area, and a maxillary mesiodens (Figures 5 and 6). One of the parapremolars situated in the left area was erupted in a lingual position between the second premolar and the first molar, while the other seemed to be blocked in eruption between the two premolars, on the buccal side (Figures 7 and 8). These teeth caused various complications, such as crowding, root resorption, impaction and cyst formation. Considering that, we should maintain the healthiest teeth from the normal or supernumerary series, with the longest root and, if possible, caries-free, and in the best position for intercuspation, we recommended

surgical removal of 35 and 45, and also of two of the supernumerary parapremolars, situated parallel to the dental arch. Also, restorative, periodontic and orthodontic treatment were indicated. For the mesiodens, a regular follow-up including a local X-ray was advised. The patient refused the surgical treatment for psychological (fear related) reasons.

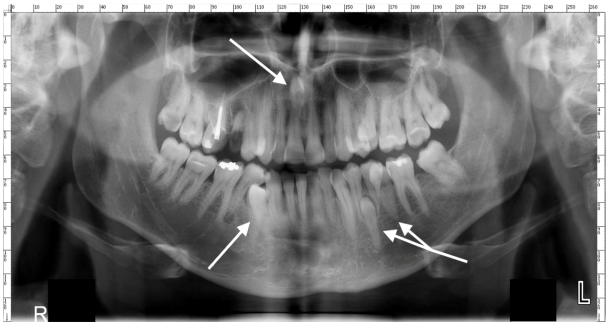

Figure 5 – ***Case No. 2: Panoramic radiological investigation at presentation showing the presence of one parapremolar in the right part of the lower jaw, two on the left part and mesiodens. R: Right; L: Left.***

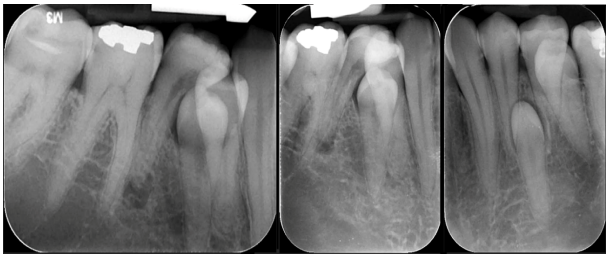

Figure 6 – ***Case No. 2: Retroalveolar images showing the parapremolar's position on the left and right part of the lower jaw and the status of the adjacent teeth.***

Discussion

There are few studies on Romanian population regarding the supernumerary teeth, while those regarding the prevalence of the supernumerary premolars are even harder to find. In other populations, studies showed that supernumerary teeth can be singles or multiples, unilaterally or bilaterally located, in the upper or lower jaw or both [10]. Some authors pointed that supernumerary teeth can be found as single supernumerary in 76–86% of the cases, as double (two supernumerary) in 12–23% of the cases, and as three or more in 2–8% of cases [11]. Rajab & Hamdan showed in their study that five or more supernumerary teeth in the same patient can be found in about 1% or less. Also, they found that about 73% of primary supernumerary teeth erupt in the oral cavity, the rest remaining impacted. Regarding the permanent supernumerary teeth, Rajab & Hamdan reports that 13–34% of them can be observed in the oral cavity [12]. Another study conducted by Açikgöz *et al.* showed that the prevalence of multiple supernumerary teeth is about 0.06% [13].

The etiopathogeny of supernumerary teeth is not completely understood; heredity seems to be an important factor, because supernumeraries are often found among the relatives of affected patients [14]. The literature presents some cases of supernumerary teeth in healthy patients, without any general diseases associated [11]. There are studies reporting that in the upper jaw the supernumerary teeth are found in 8.2 to 10 times more than in the lower jaw [15, 16]. An area where we can frequently find supernumerary teeth is the midline of maxilla (mesiodens). Another location is the palatal area of upper incisors, the mandibular premolar area (parapremolar), the area located buccal or oral of a molar (paramolar), and the distal area of the third molar (distomolar) [17, 18].

Figure 7 – ***Case No. 2: Clinical aspect of the patient at presentation showing poor hygiene, caries, tartar, periodontitis, crowding and malocclusion. R: Right; L: Left.***

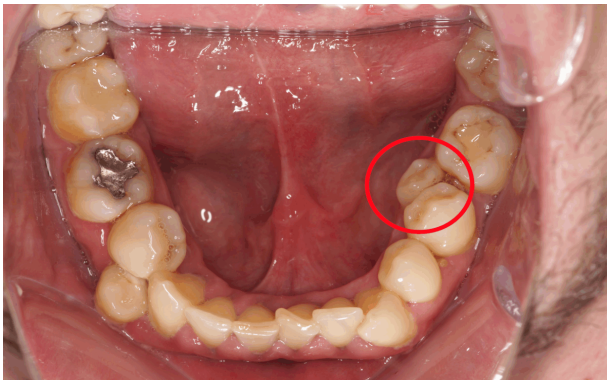

Figure 8 – ***Case No. 2: Clinical aspect of the lower jaw at presentation showing the malpositioned normal premolars and one parapremolar lingually erupted in the left part of the arch.***

The presence of supernumerary parapremolars was reported in many populations. Studies show that from all the supernumerary teeth, a percentage of 8–10% are represented by parapremolars. King *et al.* reported a prevalence of 8% of supernumerary premolars, in an Australian population [19]. Nazif *et al.* [5] and Grahnén & Lindahl [20] studied the prevalence of supernumerary premolars in American population and reported a percentage between 8% and 9.1% of all supernumerary teeth. Also, they showed that parapremolars are found more often in the mandible than in the maxilla. Singh *et al.* reported a case of unilateral impacted supernumerary premolar impacted in the upper left premolar region [21]. Cases of late development of parapremolars have been reported in the literature by Rubenstein *et al.* [22], Chadwick & Kilpatrick [23], Hegde & Munshi [24],

Breckon & Jones [25], Shapira & Haskell [26], Gibson [27] and Kocadereli *et al.* [28]. In their study, Rubenstein *et al.* [22] showed a parapremolar incidence of 0.64%. Regarding the impacted mandibular parapremolar, its presence was also highlighted in a study conducted by Poyton *et al.* [29].

Kaya *et al.* showed that the prevalence of parapremolars in Turkish population is between 0.075% and 0.26% and they represent about 10% of all the supernumerary cases [30]. They are more common in permanent dentition that in primary one. The results of the study conducted by Shah *et al.* for a population in Great Britain showed that 60.9% of the supernumerary teeth are found in the mandible, while 44.8% of them are found in the mandibular premolar region [15]. In a study conducted by Kantor *et al.*, on American population, it was shown that the incidence of supernumerary premolars is about one in 10 000 individuals [31]. Solares & Romero, in a literature review, showed that a percentage of 74% of supernumerary teeth are mandibular parapremolars. They occur three times more in males than in females, showing a possible sex-linked inheritance, as it was suggested by our patient's family history too. The authors found that patients with supernumeraries in the anterior region have 24% chances of developing parapremolars at a later age [32]. Studying the prevalence of the parapremolars on patients of Mexico, Salcido-García *et al.* found a percentage of 1.7% [33].

Regarding the mandibular parapremolar, Grahnén & Lindahl showed that these teeth are seen in about 0.29% of the general population [20]. In a study conducted by Parry & Iyer, which analyses mandibular parapremolar, only one case was found, representing 0.05% of all patients included in the study [34]. Hyun *et al.* showed a prevalence of 0.029% for supernumerary mandibular premolars in cases of patients without other syndromes associated [18]. In a study conducted by Esenlik *et al.* on a Turkish population, it was shown that the prevalence of the maxillary parapremolars is 0.2% and that of mandibular parapremolars is 0.5% [35].

Multiple supernumerary teeth can be associated with different syndromes or not. When they are present in non-syndromic patients, some studies found that their most common location is on the mandible, especially in the anterior region of the lower jaw [16]. In the literature, there are reports of unilateral mandibular parapremolars, while the bilateral location is rarer. A higher percentage of parapremolars are found in mandible then in maxilla; they can be present in more than one quadrant. Bilateral parapremolars situated both in upper and lower jaws are also not seen very often. Like other supernumerary teeth, parapremolars are found more often in males than in females. In a case report, Chanagay *et al.* describe the presence of a 35-year-old male non-syndromic Indian patient with six parapremolars and complete dentition [36]. A 35-year-old man presented four erupted parapremolars in the mandible. Two of them were situated lingually to 34 and 35, one was situated lingually between 44 and 45 and the fourth one between 45 and 46. On the X-ray investigation, appeared multiple supernumerary teeth that were impacted. It was reported that supernumerary premolars occur three times more in males than in females and the most frequent area where they were found is represented by the mandibular area (74%) [15, 37]. Also, it was shown that the supernumerary premolars are located predominantly lingual. In cases when they are situated buccally, it seems these teeth remain partial impacted [16, 37, 38], as it is the case in our younger patient. Masih *et al.* showed that a percentage of 75% of parapremolars remain impacted, but they are usually asymptomatic, as it is the case of our older patient [37].

Shapira & Haskell [26], Chadwick & Kilpatrick [23] and Poyton *et al.* [29] showed that, in some cases, the anterior supernumerary teeth (mesiodens) coexists with mandibular parapremolars, similar to the case of our younger patient. In some cases, the presence of anterior supernumerary teeth in late childhood can associate with the presence of mandibular parapremolars in adolescence. The period of time when the premolars are forming is between 1.5 and 2.5 years of age and they became visible on radiographs around 3–4 years [39]. The parapremolars supernumerary teeth begin their forming about 7–11 years after normal premolars [22, 31]. Regarding the shape of the parapremolars, Masih *et al.* found that they resemble the normal premolars [37]. The morphology of the mesiodens is usually conical and it can be situated between the maxillary central incisors or in the palatal area behind the central incisors [12, 40, 41]. Studying the prevalence of mesiodens, the data available in literature showed a percentage between 0.09% and 3.9% in general population [10, 42–44]. This tooth is considered one of the most common supernumerary teeth found in permanent dentition; it is not often seen in primary dentition [45]. Frequently, in about 80–90% of cases, its location is the upper jaw; a high percentage is seen in the anterior region of the maxilla [46, 47]. Mesiodens is more often seen in male population, a ratio of 2:1 being reported in the literature [48].

In a three years study on 111 293 patients that analyses the clinical and radiological features of supernumerary teeth, the related complications and the treatment solutions, Bereket *et al.* found that 17.16% were parapremolars; 38.36% of all cases presented associated complications [49]. Like other supernumerary teeth, parapremolars are usually asymptomatic and they are discovered by chance during routine radiological exams, done before starting a general oral rehabilitation or an orthodontic treatment [32]. Bodin *et al.* reported that only 2% of the supernumerary premolars can lead to complications [50]. Among those complication, Solares & Romero found especially dentinogenic cyst and root resorption of the adjacent tooth [32]. Parapremolars can generate complications, both if they remain impacted but also when erupting: failure of eruption, esthetic issues, displacement or rotation of adjacent teeth, crowding, malocclusion (crowding, open bite, lateral cross bite), root resorption, dilaceration, delayed or abnormal root development of permanent teeth, nasal cavity eruption, cystic formation, and fistulas [14, 51, 32]. Our two cases are representative for both situation: the older brother having an impacted parapremolar discovered by routine radiological examination has no complication at all, while the younger brother is much more affected by complications as crowding, malocclusion, root resorption.

In consequence, the treatment considerations depend on evaluating each case in particular, by a multidisciplinary approach of a team of specialists. Pediatricians should warn parents about the development of supernumerary teeth if they have a related family history. Still, most frequently, supernumerary teeth are discovered during routine panoramic radiographic investigation; periapical and occlusal radiographs are also useful. When they are discovered later in life, these cases require a comprehensive treatment plan involving a team of specialists in pediatrics, oral surgery, orthodontics, prosthetics. Cases without any clinical complications are usually followed-up. A question arising for the supernumerary teeth is if it is better to extract or retain them, according to location, morphology and complications. The presence of supernumerary teeth can cause serious problems to the normal adjacent teeth [52]. One or more extra-teeth have a big potential to disrupt the normal development of occlusion. Herath *et al.* reported the characteristics and sequelae of erupted supernumeraries in a group of Sri Lankan children and showed that the sequelae of supernumeraries on the dentition are increasing with age, leading to malocclusion [53]. A part of the specialists considers that when we discover this condition early in time, it is better to remove the supernumerary teeth by an early intervention, in order to allow the rest of the teeth to align and to establish a good intercuspal relationship. They are arguing that when we deal with a single, normal developed supernumerary tooth, it can erupt spontaneously, parallel to or inside the dental arch, causing crowding issues; usually the extraction of these teeth is all that it is required [54]. In a study aiming to survey the demographic profile of supernumerary teeth in 305 Brazilian children and adolescents, de Oliveira Gomes *et al.* concluded that evaluating these teeth implies a wide range of factors, and that early detection is essential to avoid complications and assure successful management. Permanent teeth displacement was found most frequently (36% of cases), while most cases were treated by surgical removal followed by orthodontics [55].

Conclusions

Parapremolars are the most frequent supernumeraries in the mandibular arch; they occur single or multiple and they can associate with other type of supernumerary teeth. Data in literature show a sex-linked factor and a high incidence, similar with that of mesiodentes. Screening, preventive and follow-up measures are important for such cases in order to limit the consequences of their presence and the amplitude of possible complications. In Romania, studies on this topic are very few. This case shows the presence of parapremolars in brothers with no other medical relevant history and points the importance of an early diagnosis and follow-up using successive panoramic radiographs of such cases, in order to limit or prevent the inflammatory, carious, periodontal and occlusal complications that can seriously affect both the jaws and the normal dentition; also, it points the importance of further studies on our population.

Conflict of interests

The authors declare that they have no conflict of interests.

Author contribution

Authors #1 (OCA) and #4 (LD) have equal contributions to this paper.

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