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Obostrana prekobrojnost zuba u donjoj čeljusti povezana s geminacijom: prikaz slučaja

Bilateral Mandibular Supernumerary Teeth Associated with Gemination: a Case report

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Sažetak

Višestruka hiperdoncija rijetka je i obično povezana s kompleksnim sindromima ili geminacijom. Najčešći tretman koji se u tom slučaju primjenjuje jest uklanjanje prekobrojnih zuba iz profilaktičkih razloga. Svrha je ovog prikaza opisati izoliranu pojavu prekobrojnih zuba na više lokacija u čeljusti, no nepovezanih s bilo kojim sindromom. Jedan od zuba bio je rijedak i kao fenomen jer je imao anatomiju korijenskih kanala specifičnu za geminaciju. Ekstropični zubi izvađeni su jer su uzrokovali nelagodu, bol i otekline u ustima. Ekstrahirana su tri prekobrojna zuba bez postoperacijskih komplikacija. Kontrolni pregled bio je dogovoren za godinu dana i tada nije bilo recidiva.

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Ključne riječi

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Uvod

Hiperdoncija je, prema definiciji, povećani broj zuba kod jedne osobe (1). Nalaze se pritom na bilo kojem mjestu u zubnom luku te se mogu pojaviti u trajnoj i mliječnoj denticiji, no u mliječnoj su pet puta rjeđi nego u trajnoj (2).

Etiologija hiperdoncije još nije jasna, ali u epidemiološkim studijama nekoliko je teorija (3). Tako Gardiner (4) predlaže tri mehanizma nastanka: abnormalnu proliferaciju dentalne lamine; dodatni zubni pupoljak prije razvoja trajnih zuba te produžetak dentalne lamine nakon razvoja mliječnih i trajnih zuba (postpermanentni oblik). Većina tih teorija je hipotetska zbog toga što je nemoguće nabaviti dovoljno embriološkog materijala kojim bi se dokazao izvor prekobrojnih zuba, iako literatura uglavnom podupire teoriju dentalne lamine (5).

Prisutnost prekobrojnih zuba na više mjesta u čeljusti vrlo je rijetka. Češće je povezana i pojavljuje kod sindromskih fenotipova, kao što su kleidokranijalna displazija, zatim Gardnerov, Fabry-Andersonov i Ehler-Danlosov sindrom te fisure lica (7). Isto tako može se pojaviti uz ostale sindrome i sistemska stanja (8).

Za dijagnozu i plan terapije prekobrojnih zuba najpouzdanije su klasične rendgenske snimke, a najčešće se koristi-

Introduction

A supernumerary tooth (or hyperodontia) is defined as an increase in the number of teeth in a given individual (1). They have been found in all areas of the dental arches and, although present in both the permanent and primary dentitions, are five times less frequent in the latter (2).

The etiology of hyperodontia remains unclear, but various theories have been presented based on epidemiological studies (3). Gardiner (4) suggested three possible mechanisms: an abnormal proliferation of the dental lamina; an additional follicle prior to the development of the permanent tooth and an extension of the dental lamina after both the deciduous and permanent follicles have developed (post-permanent type). The most of the theories are hypothetical in nature as it has not yet been possible to obtain sufficient embryologic material on the origin of supernumerary teeth, although most of the relevant literature supports the dental lamina theory (5).

The very presence of multiple supernumerary teeth is a rare condition and its occurrence is more frequent in syndromic phenotypes, such as cleidocranial dysplasia, Gardner's syndrome (6), Fabry-Anderson syndrome, Ehler-Danlos syndrome, and facial fissures (7), but can also be identified with-

mo ortopantomogram te periapikalnim i okluzalnim snimkama (9).

Svrha ovog prikaza jest opisati izoliranu pojavu prekobrojnih zuba na više lokacija u čeljusti, no nepovezanu s bilo kojim sindromom. Jedan od zuba predstavljao je rijedak fenomen jer je imao anatomiju korijenskih kanala specifičnu za geminaciju.

Opis slučaja

Dvadesetogodišnji muškarac došao je u Zavod za oralnu kirurgiju Stomatološkog fakulteta Federalnog sveučilišta Pernambuco u Brazilu. Pacijent je upućen zbog oteklina i bolova uzrokovanih izrastanjem triju zuba lingvalno. Iz medicinske anamneze nije se vidjelo je li imao bilo kakvu sistemsku ili težu bolest u djetinjstvu. Općim i ekstraoralnim pregledom nisu bile ustanovljene abnormalnosti.

Intraoralnim pregledom pronađena su tri prekobrojna zuba u mandibuli s lijeve i desne strane u području premolara. Morfološki su sličili premolarima. Na desnoj strani čeljusti u području prekobrojnog zuba uočen je edem sluznice lingvalne strane alveolarnog grebena (Slika 1.). U povijesti bolesti nije bilo dokaza o dentalnoj patologiji, kongenitalnim anomalijama ili genetskim promjenama i sindromima koji bi mogli biti povezani s pojavom prekobrojnih zuba. Pacijent u povijesti bolesti nije naveo ni jednu nasljednu etiologiju.

Predoperativni ortopantomogram (Slika 2.) i intraoralna okluzalna rendgenska slika (Slika 3.) pokazali su tri prekobrojna zuba s lingvalne strane zubnog luka. U tom je slučaju jedini izbor bio kirurški zahvat zato što su prekobrojni zubi otežavali pacijentu govor te uzrokovali nelagodu, bol i otekline. Predloženi zahvat sastojao se od ekstrakcije triju posteriornih donjih ektopičnih zuba.

Svi uobičajeni predoperativni laboratorijski testovi (kompletna krvna slika i PV) pokazali su normalne vrijednosti. U dodatnim rendgenskim nalazima vrijednosti su također bile normalne.

Nakon što je pacijent potpisao pristanak, dobio je anestetik u oba donja alveolarna, lingvalna i mentalna živca, no bila je upotrijebljena i intraligamentarna anestezija. To je učinjeno Mepivacain hidrokloridom s epinefrinom razrijeđenim u omjeru 1:100000. Nakon toga odignut je režanj oko prekobrojnih zuba.

Tri su zuba bila izvađena Cryerovim polugama. Prekrivni zub ekstrahiran je nakon podizanja režnja. Rana je zaživela nakon ispiranja fiziološkom otopinom (Slika 4.). Nakon ekstrakcije uočeno je da jedan od ektopičnih zuba ima promijenjeni oblik, što može biti posljedica geminacije ili fuzije (Slike 5. i 6.). Pacijent je bio naručen na postoperativne preglede nakon sedam, petnaest i trideset dana (Slika 7.). Tjedan dana nakon zahvata rana je zacijelila bez komplikacija te su uklonjeni šavovi. Na kontrolama nakon šest i dvanaest mjeseci radiološkim pretragama nisu ustanovljene patološke tvorbe.

out other systemic conditions or associated syndromes (8).

Conventional radiographs are the most reliable and definite means for the diagnosis of supernumerary teeth and planning their treatment. The most commonly used radiographs are orthopantomogram, periapical and occlusal radiographs (9).

The aim of this case report was to present an isolated nonsyndromic occurrence of multiple supernumerary teeth, one of which had a root canal anatomy of gemination, representing a rare phenomenon.

Case report

A 20-year-old male was referred to the Department of Oral Surgery, at the School of Dentistry of the Federal University of Pernambuco, in Brazil, due to the eruption of three teeth on the lingual edge, as well as a complaint of pain and swelling in this region. The medical history did not reveal any history of serious childhood illness or systemic abnormality. The general physical and extraoral examination also did not show any abnormality.

An intraoral examination revealed the presence of three supernumerary premolars in the region of the mandibular left and right premolars. The supernumerary teeth had morphology similar to that of the premolars. In addition, an edematous area was also detected on the lingual edge, next to the ectopic tooth on the right side (Figure 1). The case history excluded dental pathologies, congenital anomalies and genetic or syndromic alterations associated with the supernumerary teeth. The patient reported no hereditary etiology.

The preoperative panoramic radiograph (Figure 2) and intraoral occlusal radiograph (Figure 3) revealed the presence of three bilateral supernumerary teeth at the level of the lingual edge. Since the impacted and erupted supernumerary teeth located on the mandibular lingual edge were associated with discomfort, pain and swelling, and considering the fact that this situation was impairing the patient's speech, surgical treatment was required. The proposed treatment, therefore, consisted of the excision of the three lower posterior ectopic teeth.

All the routine preoperative laboratory tests, such as complete blood count, coagulation studies and blood chemistry were requested and the results were all within the normal range. The additional radiographic images were likewise normal.

After obtaining the patient's consent, he was anesthetized by bilateral blockage of the inferior alveolar and lingual nerves, as well as the intraligament and bilateral mental nerve, with 2% mepivacaine hydrochloride 1:100000 epinephrine. Reflection of the mucoperiosteal flap was performed around the supernumerary teeth.

Using Seldin's straight elevator, the three teeth were extracted. As for the enclosed tooth, following reflection of the mucoperiosteal flap, it was exposed and then removed using Seldin's elevator number 3. After rinsing with physiological saline, the suture was made (Figure 4). After extraction, it was noticed that one of the ectopic teeth was associated with a shape alteration which may have been the result of ei-



Slika 1. Kliničko stanje pacijenta prije kirurškog zahvata; vide se prekobrojni zubi u mandibuli
 Figure 1 Clinical appearance of the patient prior to the final surgical procedure showing supernumerary teeth in the mandible.
 Slika 2. Ortopantomogram pokazuje prekobrojne zube i geminaciju premolara u mandibuli
 Figure 2 The panoramic radiograph showing supernumerary teeth and geminated premolar in the mandible.
 Slika 3. Prekobrojni zubi na okluzalnoj snimci
 Figure 3 Appearance of supernumerary teeth in an occlusal radiograph.
 Slika 4. Kliničko stanje pacijenta odmah nakon kirurškog zahvata
 Figure 4 Clinical appearance of the patient immediately after surgery.
 Slika 5. Izgled prekobrojnih zuba nakon ekstrakcije; kod jednoga se vidi promjena oblika
 Figure 5 Appearance of supernumerary teeth after extraction, showing shape alteration associated with one of the supernumerary teeth.
 Slika 6. Periapikalna rtg. snimka pokazuje promjenu oblika zuba
 Figure 6 Periapical radiograph showing shape alteration.
 Slika 7. Kliničko stanje pacijenta trideset dana nakon kirurškog zahvata
 Figure 7 Clinical appearance of the patient 30 days after the procedure.

ther gemination or fusion (Figures 5 and 6). The patient was examined postoperatively at the intervals of 7, 15, and 30 days (Figure 7). One week following extraction, the wound was observed to be healing well, no complications were noted, and residual sutures were removed after 7 days. At both 6-month and 1-year follow-ups, no pathologic changes were observed on the radiographs.

Rasprava

U ovom prikazu slučaja dijagnosticirana su tri prekobrojna zuba. Od dva locirana na desnoj strani čeljusti, jedan je bio morfološki oblikovan geminacijom, a drugi prekriven desnima. Treći zub bio je smješten lingvalno na lijevoj strani između prvog i drugog premolara. Fuzija i geminacija razvojne su anomalije tvrdih zubnih tkiva. Fuzija se definira kao spajanje dvaju zubnih zametka koji izrastu u obliku jed-

Discussion

In this case report a total of three supernumerary teeth were diagnosed, including an enclosed one and another associated with the morphological alteration known as gemination, both of which were located on the right side of the mouth. The third one was located in the mandibular premolar region on the left side, between the first and the second premolars. Fusion and gemination are developmental anom-

noga velikog zuba, a geminacija se može definirati kao pokušaj jednoga zubnog zametka da stvori dva zuba. Klinički ih je teško razlikovati ako je prekobrojni zub srašten s trajnim zubom. Kad je riječ o terapiji, nije nužno razlikovati fuziju od geminacije (10).

Prekobrojni zubi pojavljuju se u 1,05 posto slučajeva i najčešći su u gornjoj distomolarnoj regiji (1). No, u skladu s dosadašnjim tvrdnjama, većina se nalazi u premaksilarnoj regiji, koničnog su oblika i ne niču. Ovaj prikaz slučaja nije u skladu s dosadašnjim istraživanjima, ali jest sa studijom (13) u kojoj se ističe da je najčešća lokacija prekobrojnih zuba u mandibularnoj premolarnoj regiji. Kad je riječ o spolovima, ovaj je prikaz u skladu s ostalim istraživanjima u kojima se navodi da su prekobrojni zubi češći kod muškaraca (1, 2, 6). No, neki istraživači nisu uočili razlike među spolovima (14).

U retrospektivnoj studiji (2) omjer između muškog i ženskog spola iznosio je 2:1 za bijelu rasu, 5,5:1 za Japance i 6,5:1 za osnovnoškolsku djecu u Hong Kongu (15). U ostalim istraživanjima (8) ističe se omjer 4,3:1 i sugerira da su razlike između omjera u dosadašnjim studijama posljedica načina istraživanja ili genetskog podrijetla pacijenata.

Višestruki prekobrojni zubi česti su kao simptom u mnogim sindromima, primjerice u slučaju kleidokranijalne displazije, Gardnerova sindroma, ili rascjepa usnice i nepca (2, 3, 6, 12, 17, 18). Suprabha i suradnici (19) opisali su neobičnu kombinaciju dentalnih anomalija kod naizgled zdrave dvanaestogodišnje pacijentice, a sastojala se od mnogostrukih dens invaginatus, generalizirane hipoplazije cakline i mikrodoncije, resorpcije korijena, mnogostrukih periapikalnih lezija, polumjesečastih sjekutića, batičastih premolara, taurodontizma, hipodontije i prekobrojnih zuba. Prava je rijetkost pronaći prekobrojne zube kod pacijenata koji ne pokazuju druge sistemske bolesti ili sindrome, pa su takvi slučajevi zabilježeni u literaturi (3, 6, 10, 13, 15, 20). Yagüe-García i kolege (7) napisali su malu retrospektivnu studiju o osmero pacijenata s nesindromskom višestrukom hiperdontijom. Naš pacijent u povijest bolesti nije naveo sindrom koji bismo mogli povezati s višestrukom hiperdontijom i naša su nalazi u skladu s rezultatima dosadašnjih studija (3, 6, 7, 10, 13, 15, 20).

Prekobrojni zubi češći su kod pacijenata u srodstvu s ljudima za koje se zna da su imali prekobrojne zube (6), ali to nije bio slučaj u ovom prikazu. Inchingolo i njegovi suradnici (21) zabilježili su rijedak oblik hiperdontije s obostrano višestrukim prekobrojnima zubima, no nije bio povezan sa sindromima nego s genetskim probijem u obiteljski fenotip. U pregledu literature navedeni su slučajevi višestrukih prekobrojnih zuba u obitelji, no bez zabilježenih sistemskih poremećaja ili sindroma (22-27). Ti slučajevi nisu u skladu s našim slučajem.

Prikazi slučaja iz 2007. godine (17) opisuju pacijenta s četrnaest prekobrojnih zuba – devet je bilo locirano u mandibularnoj premolarnoj regiji s lingvalne strane, no nije mu bio dijagnosticiran ni jedan sindrom.

U istraživanju iz 2008. godine zabilježeno je da se prekobrojni zubi u premolarnoj regiji najčešće nalaze lingvalno te da postoje samo četiri iznimke od ukupno devedeset i četiri prekobrojna mandibularna premolara. Autori tih studija za-

alies of the dental hard tissue. Fusion is defined as the joining of two developing tooth germs resulting in a single large tooth structure. Geminatio is defined as attempted formation of two teeth from a single enamel organ. Clinically, it may be difficult to differentiate between fusion and geminatio when a supernumerary tooth is fused with a permanent tooth. Concerning treatment, differentiation between fusion and geminatio may not be critically important (10).

The prevalence of supernumerary teeth was 1.05%, the most frequent location being at upper distomolar level (1). However, in accordance with previous studies (1, 2, 11, 12), the majority of supernumerary teeth were located in the premaxillary region, they were of conical shape, and remained unerupted. The present case report is not in accordance with the previous study, but complies with the other study (13) which stated that the most common site is the mandibular premolar region where the supernumerary teeth are of the supplemental type. Regarding gender distribution, this case report is in agreement with many studies that showed that males are more commonly affected than females (1, 2, 6). However, some other researchers (14) have observed no differences between genders.

In a retrospective study (2), a male:female ratio of 2:1 was found in the permanent dentition for Caucasians, while other authors reported ratios of 5.5:1 and 6.5:1 in Japanese (15) and Hong Kong (16) schoolchildren, respectively. Results of other studies (8) revealed a male:female ratio of 4.3:1 and suggest that differences in prevalence regarding previous studies might be due to the study design or patients' genetic background.

Multiple supernumerary teeth are frequently associated with a number of syndromes, namely cleidocranial dysplasia, Gardner's syndrome, cleft lip and palate, and are recognized as a part of their characteristics (2, 3, 6, 12, 17, 18). Suprabha et al (19) described an unusual combination of dental anomalies in an apparently normal and healthy 12-year-old female patient. The dental anomalies were multiple dens invaginatus, generalized enamel hypoplasia, generalized microdontia, root resorption and multiple periapical lesions, shovel shaped incisors, cup shaped premolars, taurodontism, hypodontia and supernumerary teeth. It is rare to find multiple supernumerary teeth in individuals without other systemic conditions or associated syndromes, although some cases were reported in literature (3, 6, 10, 13, 15, 20). Yagüe-García et al. (7) conducted a small retrospective observational study of 8 patients diagnosed with nonsyndromic multiple hyperdontia. The present case had no history of combined diseases or syndromes and was consistent with previous findings (3, 6, 7, 10, 13, 15, 20).

The supernumerary teeth are more likely to be present in patients whose relatives also had supernumerary teeth (6), but this has not been found in the present case. Inchingolo et al. (21) reported a rare form of hyperdontia, with bilateral nonsyndromic multiple supernumerary teeth, with evident penetrance of the phenotype in the family unit. In the reviewing of the literature some cases of familial occurrence of multiple supernumerary teeth were found without any associated systemic disorders or syndromes (22-27). These find-

ključili su da bukalno smješteni zubi uvijek ili djelomice niknu, a lingvalno smješteni zubi pretežno ne izniknu.

Klinički i radiološki pregledi vrlo su važni u odabiru terapije koja se može sastojati od jednostavne ekstrakcije pa sve do onih praćenih ortodontskom terapijom kako bi se osigurala pravilna okluzija (6). Prekobrojne premolare bilo bi vrlo teško pronaći bez radiološke dijagnostike.

Odluka za kirurško uklanjanje prekobrojnih zuba mora se temeljiti na mogućem razvoju patoloških tvorbi vezanih za prekobrojne zube. Jedan prikaz (13) pokazao je da su najčešće patološke tvorbe povezane s prekobrojnima zubima ciste u premolarnoj regiji (9%), a kao komplikacija navodi se oštećenje susjednih zuba (13%). U našem slučaju pacijent nije razvio cistu, a ni susjedni zubi nisu bili oštećeni. Bol, oteklina i otežani govor jedine su komplikacije u ovom slučaju koje su zahtijevale ekstrakciju.

Zaključak

Protokol liječenja prekobrojnih zuba nije standardiziran. Svi prekobrojni zubi ne trebaju se vaditi. Ako zub ne čini teškoće može ostati *in situ* i u promatranju, a ako uzrokuje lokalne simptome ekstrakcija je indicirana i pacijent mora biti obaviješten o svim rizicima zahvata.

ings are not in accordance with our report.

The series of case reports in 2007 (17) also presented a case of a nonsyndromic male patient with fourteen supernumerary teeth, nine of which were located in the mandibular premolar region; all of them were on the lingual edge.

In 2008 (8), it was reported that supernumerary premolar locations were predominantly lingual with only four exceptions among 94 mandibular supernumerary premolars. The authors also concluded that the buccally located ones were partially or completely erupted, as in this report, in contrast to the majority of lingually located ones, which were in the alveolar bone.

The clinical and radiographic examinations are of vital importance for the correct treatment plan, which may range from simple extractions to extractions followed by orthodontic treatment in order to obtain correct occlusion (6). Without radiographic examination, it is unlikely that the development of supernumerary premolars could be diagnosed.

The surgical removal of supernumerary teeth should always be based on the possible pathological sequel associated with supernumerary teeth. The case report (13) showed that the most common complications associated with supernumerary teeth in the premolar area are cyst formation (9%) and damage to adjacent teeth (13%). In the present case report the patient presented neither of these complications, although there were other associated complications, such as pain, swelling and difficulties in speech, making extraction necessary.

Conclusions

The treatment of supernumerary teeth has no standardizations. Not all supernumerary teeth require extraction. If the tooth is asymptomatic, it can be left *in situ* and kept under observation; if, however, it is associated with local disorders, extraction is recommended and the patient should be made aware of the risks involved in this treatment.

Abstract

Multiple hyperodontia rarely occurs without being associated with complex syndromes or gemination. The prophylactic surgical removal of the supernumerary teeth is generally the treatment of choice. The aim of this case report was to present an isolated nonsyndromic occurrence of multiple supernumerary teeth, one of which had a root canal anatomy of gemination, representing a rare phenomenon. However, since the ectopic teeth were associated with discomfort, pain and swelling, surgical treatment was necessary. The chosen treatment was surgery, with the extraction of the three supernumerary teeth and no evidence of any postoperative complications. No recurrences occurred in one year of follow-up.

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Key words

Tooth, Supernumerary; Tooth Eruption; Odontogenesis

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