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CASE REPORT

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Dentinogena cista upalnog podrijetla: dijagnostički izazov

Dentigerous Cyst of Inflammatory origin: a Diagnostic Challenge

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Sažetak
Odontogene ciste obično se otkrivaju tijekom rutinskoga pregleda u stomatološkoj ordinaciji, a prema podrijetlu mogu biti razvojne ili upalne. Obično su destruktivne za okolne strukture - kako za kost tako i za zube. Moramo istaknuti da različite vrste cista vrlo često radiografski i histopatološki daju sličnu kliničku sliku. U takvim slučajevima točna dijagnoza ovisi o korelaciji svih navedenih nalaza. Dentinogene ciste su one koje zatvara kruna neniklih zuba širenjem folikula i - kako je prirasla na vrat zuba - obično je u vezi s nekim od zuba koji još nisu niknuli. Zna se također da progresija upale od apeksa mliječnog zuba potiče nastanak dentinogenih cista oko nenikloga trajnog zuba. U ovom je članku opisan jedan takav rijedak slučaj upalne dentinogene ciste (IDC-a) u području lijevoga mandibularnog očnjaka kod dvanaestogodišnjeg dječaka. Terapija je bila enukleacija ciste zajedno sa zubom.

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Ključne riječi
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Uvod

Dentinogene ciste vrlo su česte lezije u čeljusti i obično su povezane s impaktiranim trećim molarima i trajnim maksilarnim očnjakom. Među svim čeljusnim cistama njihov je udjel oko 20 do 24 posto, a javljaju se u velikom vremenskom rasponu s vrhuncem incidencije u drugom i četvrtom desetljeću života (1-4). Klinički nema nikakvih bolova ni bilo kakve nelagode u vezi s cistom, osim ako se ne pojavi upalna egzacerbacija. Dentinogene ciste obično se otkrivaju kada se radiološki istražuje nenicanje zuba, loše položen zub ili manjak zuba (5). Histološki je kod tipične neupalne dentinogene ciste šupljina obložena tankim nekeratiniziranim višeslojnim skvamoznim epitelom. Debljina obložnog epitela varira ovisno o vrsti i težini upale (1, 2).

Nije razjašnjeno zašto nastaje IDC. Stručnjaci ističu da se upala na apeksu mliječnoga zuba može proširiti na zametak trajnoga, a posljedica je stvaranje dentinogene ciste (6). Svrha ovog prikaza bila je izvijestiti o slučaju IDC-a vjerojatno nastalog zbog upale mliječnog zuba prethodnika.

Introduction

Dentigerous cysts are one of the most common lesions of the jaws and are often associated with impacted third molars and permanent maxillary canine. They comprise approximately 20%-24% of all jaw cysts and occur over a wide age range with peak incidence in the 2nd to 4th decades (1-4). Clinically, no pain or discomfort is associated with the cyst unless there is acute inflammatory exacerbation. Dentigerous cysts are usually discovered when radiographs are taken to investigate a failure of tooth eruption, mal-aligned or missing tooth (5). Histologically, typical non-inflammatory dentigerous cysts are lined by thin nonkeratinized stratified squamous epithelium. However, many variations in the thickness of the lining epithelium may be noted depending on type and severity of inflammation present (1,2).

The mechanism of development of an IDC is unclear. It is reported that the inflammation present at the root apex of the overlying deciduous tooth may spread around the crown of an unerupted permanent tooth and result in the formation of a dentigerous cyst (6). The aim of the present article is to report an additional case of IDC, possibly associated with inflammation of the overlying deciduous tooth.

Opis slučaja

U ordinaciju je došao dvanaestogodišnji dječak žaleći se na kompresije zuba u gornjoj i donjoj fronti. Pregledom je ustanovljeno da su gornji lateralni sjekutići postavljeni lingvalno, a očnjaci bukalno na objema stranama zubnog luka. Blaga kompresija bila je oko zadržanog mliječnog mandibularnog i trajnog lijevog očnjaka koji je nedostajao. U istom području bila je i mala bezbolna izbočina za koju se kasnije ustanovilo da je intraosealna cista povezana s impaktiranim trajnim mandibularnim lijevim očnjakom (Slika 1.). Nije bilo nikakvih povezanih simptoma.

Ortopantomogram (OPG) i intraoralni periapikalni radiogram (IOPA) pokazali su malo unilokularno radiolucentno područje cistične lezije sa skleroziranim rubovima u promjeru 2x2 centimetra, a u vezi s krunom impaktiranog trajnog mandibularnog očnjaka. Pružalo se od cemento-caklinskog spoja do apeksa zuba 31 i 32 (Slike 2. i 3.). Apeks mliječnog zuba iznad nije bio u komunikaciji s translucencijom.

Bila je postavljena privremena dijagnoza "dentinogene ciste". Mliječni zub tijekom prvog posjeta bio je ekstrahiran i planirano je kirurški ukloniti cistu. Cista je kasnije enukleirana i više tkivnih fragmenata različitih dimenzija poslano je na histopatološku analizu.

Histopatološkom analizom mekoga tkiva ustanovljeno je da je uzorak sadržavao upaljen nekeratinizirani višeslojni rožnati epitel različitih debljina (Slika 4.).

Pronađen je bio i upalni stanični infiltrat u obložnom epitelu i subepitelialnom vezivnom tkivu. Uočeni su u mnogim područjima označeni hiperplastični epiteli s anastomoziranim mrežnim mostovima koji su oponašali uzorak sličan radikularnoj cisti (Slika 5.).

Konačna dijagnoza - "dentinogena cista upalnog podrijetla" bila je postavljena na temelju kliničkih, radiografskih i histopatoloških nalaza.

Rasprava

Dentinogena cista razvojna je šupljina koja uključuje krunu neniknuloga zuba na cemento-caklinskom spoju i obložena je epitelom. Te su lezije druge najčašće odontogene ciste, odmah nakon radikularnih (4). Asimptomatske su i najčešće se otkrivaju slučajno tijekom rutinskoga radiografskog pregleda (5). Na dentinogenu cistu može se posumnjati ako je folikularni prostor veći od pet milimetara, budući da je normalni u prosjeku 3,4 milimetra (5,6). No, dijagnoza se ne može postaviti isključivo na temelju kliničkog i radiografskog nalaza.

Upalne dentinogene ciste (IDC-i) nalaze se isključivo u miješanoj denticiji. Radiografski se vide kao okrugle ili ovalne dobro ograničene unilokularne tranlucencije sa sklerotičnim rubom unutar mandibule (7).

Obično su povezane s korijenom ili korijenima avitalnih mliječnih zuba i krunom neniknulih trajnih zuba.

U opisanom slučaju smatramo da se upala iz periapikalnog područja mliječnog očnjaka proširila na trajni mandibularni očnjak ispod, te rezultirala stvaranjem cistične lezije.

Benn i Altini (8) smatraju da postoje barem dvije vrste dentinogenih cista. Prva je čisto razvojna i javlja se kod trajnih zuba obično zbog impakcije. Caklinska hipoplazija im-

Case report

A 12 year old boy reported with a complaint of crowding of teeth in upper and lower front region. On examination, permanent maxillary lateral incisors and canines were lingually and buccally placed, respectively, on both sides of the arch. Mild crowding was noted with an over retained right deciduous mandibular canine and missing permanent mandibular left canine. A small painless swelling was also observed in the same region that was later found to be an intraosseous cystic lesion associated with the impacted permanent mandibular left canine (Figure 1). There were no other associated complaints.

Orthopantomograph (OPG) and intra-oral periapical (IOPA) radiographs showed a small unilocular radiolucent cystic lesion with sclerotic border measuring about 2x2 cm, associated with the crown of the impacted permanent mandibular canine at the cemento-enamel junction, and extending up to apices of teeth 31 and 32 (Figures 2 and 3). The apex of the overlying deciduous tooth root was involved with the radiolucency.

A provisional diagnosis of *'dentigerous cyst'* was made. At the first visit the deciduous tooth was extracted and surgical removal of the cyst was planned. Later, the cyst was enucleated and multiple soft tissue fragments of varying dimensions along with impacted tooth were sent for histopathological examination.

The histopathological examination of tissue specimen showed inflamed nonkeratinized stratified squamous epithelial lining of varying thickness (Figure 4). The presence of inflammatory cell infiltration in the epithelial lining and sub-epithelial connective tissue were also observed. Marked hyperplastic epithelium with anastomosing rete ridges exhibiting 'arcading pattern' similar to 'radicular cyst' was noted in many areas (Figure 5).

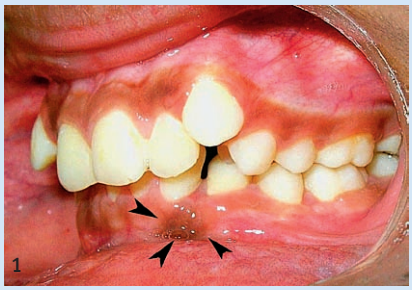
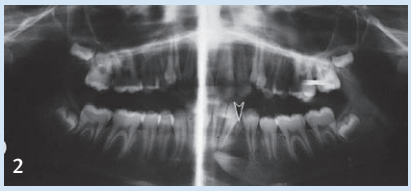
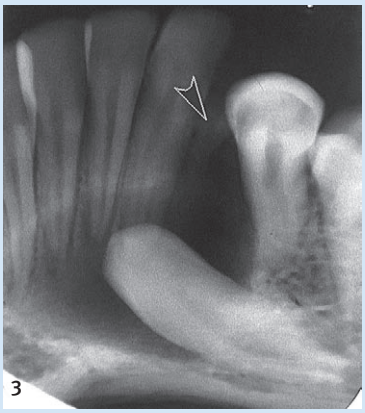
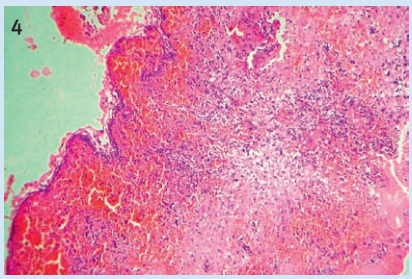
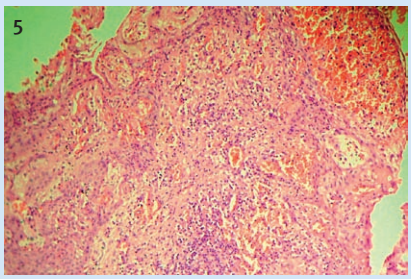
The final diagnosis of 'dentigerous cyst of inflammatory origin' was given considering clinical, radiographic and histopathologic features.

Discussion

A dentigerous cyst is an epithelial-lined developmental cavity that encloses the crown of an unerupted tooth at the cemento-enamel junction. Dentigerous cysts are the second most common odontogenic cyst after the radicular cyst (4). The cyst remains asymptomatic and frequently found only by chance during routine radiographic examination (5). A dentigerous cyst can be suspected when the follicular space is more than 5 mm, as the normal space is 3.4 mm (5,6). Hence, the diagnosis should not be made only from clinical and radiographic features.

Inflammatory dentigerous cysts (IDC) are only found in the mixed dentition. Radiographically, they appear as round or ovoid well demarcated unilocular radiolucencies with a sclerotic border within the mandible (7). IDCs are usually associated with the root or roots of a non-vital primary teeth and the crown of an unerupted permanent tooth.

In our case, we believe that inflammation from the periapical area of the deciduous canine that spread to involve the underlying permanent mandibular canine, resulted in a cystic lesion. Benn and Altini (8) indicated that at least two types of dentigerous cysts occur. The first type is purely developmental in origin and occurs in mature teeth usually as result of impac-

Slika 1. Trajni mandibularni lijevi očnjak koji nedostaje, uz blago otvoreni prostor (strelice pokazuju granice otekline).
Figure 1 Photograph showing the missing permanent mandibular left canine with slight spacing (arrows indicate borders of the swelling).
Slika 2. Predoperativni radiogram pokazuje dobro ograničenu radiolucenciju u blizini mandibularnog lijevog očnjaka (strelica pokazuje mliječnog prethodnika iznad)
Figure 2 Pre-operative panoramic radiograph showing a well defined radiolucency involving permanent mandibular left canine (arrow indicates the overlying deciduous tooth).
Slika 3. IOPA koja pokazuje cističnu leziju s mliječnim lijevim mandibularnim očnjakom i impaktiranim trajnim mandibularnim lijevim očnjakom (strelica pokazuje ležeći mliječni očnjak)
Figure 3 IOPA showing the cystic lesion associated with the primary left mandibular canine and impacted permanent mandibular left canine (arrow indicates the overlying deciduous tooth).
Slika 4. Mikrofotografija pokazuje cističnu leziju obloženu nekeratiniziranim višeslojnim orožnjelim epitelom (Hematoxylin i Eosin, originalno povećanje X100).
Figure 4 Photomicrograph of the cystic lesion lined by nonkeratinized stratified squamous epithelium (Hematoxylin and Eosin, Original magnification X100).
Slika 5. Mikrofotografija koja pokazuje infiltraciju upalnih stanica u epitel i okolno vezivno tkivo oponašajući lučni uzorak (Hematoxylin i Eosin, originalno povećanje X100).
Figure 5 Photomicrograph showing inflammatory cell infiltration in the epithelial lining mimicking arcading pattern and subjacent connective tissue. (Hematoxylin and Eosin, Original magnification X100).

paktiranog zuba može biti važan čimbenik u nastanku ciste. Druga vrsta je upalnog podrijetla i javlja se kod nezrelih zuba poslije periapikane upale nevitalnog mliječnog zuba ili nekog drugog uzroka, te se nakon toga proširila i uključila zubni folikul (vrećicu) trajnog zuba. Benn i Altini (8) opisali su tri moguća mehanizma histogeneze upalne dentinogene ciste:

1. intrafolikulani razvoj oko sekundarno upaljenih kruna trajnih zuba, a zbog periapikalne upale koja se proširila s avitalnog mliječnog zuba prethodnika;
2. radikularne ciste na apeksu avitalnih mliječnih zuba koji se spajaju s folikulom (vrećicom) neniknuloga trajnog nasljednika. "Nicanje" nasljednika u cističnoj šupljini rezultira ekstrafolikularnom dentinogenom cistom;
3. periapikalna upala iz bilo kojeg izvora, ali uglavnom iz avitalnoga mliječnog zuba koja se širi i zahvaća folikul neniknuloga trajnog nasljednika.

Main (9) je uveo pojam - "upalna folikularna cista" i postavio hipotezu da se ona razvija oko djelomice stvorene krune trajnog zuba kao rezultat intra-folikularnog širenja periapikalne upale od oboljeloga mliječnog zuba smještenog iznad trajnoga. Wood i suradnici izvijestili su 1988. (10) o trima slučajevima upalne dentinogene ciste ekstra-folikularnog podrijetla.

Terapija upalnih dentinogenih cista (IDC-a) uključuje ekstrakciju avitalnoga mliječnog zuba i masupijelizaciju cistion. Enamel hypoplasia of an impacted tooth may be a significant factor in the formation of the cyst. The second type is inflammatory in origin and occurs in immature teeth as a result of periapical inflammation from a non-vital deciduous tooth or other source, spreading to involve the tooth follicle. Benn and Altini (8) considered three possible mechanisms in the histogenesis of inflammatory dentigerous cysts:

1. Intra-follicular developmental cysts formed around the crowns of permanent tooth that become secondarily inflamed, as a result of periapical inflammation spreading from non-vital deciduous predecessors.
2. Radicular cysts at apices of nonvital deciduous teeth that fuse with the follicles of unerupted permanent successors. "Eruption" of successor teeth into the cystic cavity results in the formation of extra-follicular dentigerous cyst.
3. Periapical inflammation from any source, but usually from nonvital deciduous teeth spreading to involve follicles of unerupted permanent successors.

Main (9) termed such cysts as 'inflammatory follicular cysts' and hypothesized that the cyst develops around the partly formed crown of a permanent tooth as a result of intra-follicular spread of periapical inflammation from an overlying diseased primary tooth. Three cases of inflammatory dentigerous cysts of extra-follicular origin have been reported by Wood et al in 1988 (10).

ste. To omogućuje cijeljenje cistične šupljine i nicanje trajnog zuba, ali uz uvjet da se zahvati obavljaju u normalno vrijeme erupcije (8). Ta se terapija može primijeniti pod lokalnom anestezijom. Naravno, pacijenta treba nadzirati sve dok mu ne nikne zametnuti trajni zub i koštano se ne učvrsti. Ponekad je potrebno maknuti trajni zub ako je zaostao u razvoju i više ne raste (11). U prikazanom slučaju bila je obavljena enukleacija ciste i uklonjen trajni mandibularni očnjak iz sljedećih razloga: nicanje uključenog trajnog zuba kasnilo je više od dvije godine; bilo je završeno stvaranje zuba; zub je bio vodoravno impaktiran u donjoj polovici mandibule; nije bilo prostora za smještaj zuba u zubni luk.

Zaključak

Prikazani slučaj jedan je od nekoliko opisanih u engleskoj literaturi. Raspravlja se o etiopatogenezi i terapiji IDC-a. Za konačnu dijagnozu potrebno je korelirati kliničke, radiološke i histopatološke nalaze. IDC se javlja isključivo u miješanoj denticiji i vrlo je važno da specijalisti dječje dentalne medicine znaju da postoji.

The treatment of inflammatory dentigerous cysts (IDC) includes extraction of the non-vital primary tooth and marsupialization of the cyst. This allows healing of the cystic cavity and eruption of the permanent tooth, provided that these procedures are performed at the normal time of eruption (8). The procedure can be done under local anesthesia. However, the patient must be followed up until eruption of the displaced permanent teeth and bony consolidation. Removal of the permanent tooth may be necessary if it has suffered arrested development or is hopelessly displaced (11). In our case, enucleation of the cyst along with the involved permanent mandibular canine was done for the following reasons:

- The eruption time of involved permanent tooth was exceeded by more than two years.
- The formation of the tooth was complete.
- The tooth was horizontally impacted to the lower half of the mandible.
- There was lack of adequate space to accommodate the tooth in the arch.

Summary & conclusion

The case presented here adds to the existing few cases of IDC reported in English literature. It also discusses the etiopathogenesis and treatment of IDC. Correlation of clinical, radiographic and histopathological findings is necessary for arriving at the final diagnosis. IDCs are only found in the mixed dentition and it is very important for pediatric dentists to know about their existence.

Abstract
Odontogenic cysts are commonly encountered in routine dental practice and can be developmental or inflammatory in origin. These are often destructive to both teeth and jaws. In many instances, different types of cysts exhibit similar findings clinically, radiographically & histopathologically. In such cases, accurate diagnosis depends on co-relation of all these findings. A dentigerous cyst is one that encloses the crown of an unerupted tooth by expansion of its follicle and is attached to the neck of the tooth and it is usually associated with any of the unerupted tooth. It is also been reported that progressing inflammation from the root apex of the deciduous tooth brings about the development of the dentigerous cyst around the unerupted permanent tooth. One such rare case of inflammatory dentigerous cyst (IDC) in left mandibular canine region in a 12 year old boy is presented in this article, which was treated by enucleation of the cyst along with the tooth.

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Dentigerous Cyst; Tooth, Impacted; Odontogenic Cyst

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