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## Adenomatoidni odontogeni tumor s tkivom nalik na zub nastao iz odontogene ciste: prikaz jedinstvenog slučaja

### Adenomatoid Odontogenic Tumor with Tooth-like Structure Arising from a Dentigerous Cyst: Report of a Unique Case

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#### Sažetak

Adenomatoidni odontogeni tumor (AOT) rijedak je benigni tumor odontogenog podrijetla sastavljen od odontogenog epitela. Histopatološka slika je različita. U nedavnim klasifikacijama SZO-a uvedeno je nekoliko je promjena u vezi s tom novotvorinom. Prema posljednjoj je klasificirana kao odontogeni tumor epitelnog podrijetla. U britanskoj literaturi nabrojeno je vrlo malo slučajeva toga tumora povezanog s odontogenom cistom. Opisali smo adenomatoidni odontogeni tumor nastao kod dvanaestogodišnjeg dječaka iz odontogene ciste smještene perikoronarno oko donjeg ocnjaka koji još nije izniknuo. Klinički je bila postavljena dijagnoza odontogene ciste. Histološka pretraga pokazala je područja nalik na odontom. Takav isti zabilježen je u samo još dva slučaja. Pretpostavljamo da opisani slučaj predstavlja odontogenu cistu s neoplastičnim promjenama epitelnog i mezenhimalnog podrijetla. Potrebna su daljnja istraživanja kako bi se adenomatoidni odontogeni tumor nastao iz odontogene ciste mogao potvrditi kao zasebna hibridna novotvorina.

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#### Ključne riječi

odontogeni tumori; dentinogena cista

#### Uvod

Adenomatoidni odontogeni tumor rijetka je novotvorina odontogenog podrijetla sastavljena od odontogenog epitela. Histološka slika je različita. Lezija je benigna i neinvazivna te raste sporo, ali progresivno. Pojavljuje se u dva do sedam posto slučajeva svih odontogenih tumora. Epitelni sloj takve ciste može se promijeniti u odontogenu neoplazmu, kao što je ameloblastom ili adenomatoidni odontogeni tumor (1–3). Rijetko koji odontogeni tumor nastaje iz odontogenih cista ili je s njima povezan. Svrha ovog prikaza je predstaviti rijedak slučaj adenomatoidnog odontogenog tumora nastalog iz ovojnice odontogene ciste.

#### Prikaz slučaja

Dvanaestogodišnji dječak upućen je na Stomatološki odjel Kraljevske sveučilišne bolnice u Vadodari zbog bolova i otekline u prednjem dijelu donje čeljusti. Intraoralnim pregledom ustanovljena je tvrda, dobro definirana lezija od donjeg lijevog središnjeg sjekutića do drugog pretkutnjaka na istoj strani. Otekline nije bila mekana na dodir. Sluznica

#### Introduction

Adenomatoid Odontogenic Tumor is an uncommon tumor of odontogenic origin, composed of odontogenic epithelium in a variety of histological patterns. The lesion is benign (hamartomatous) and noninvasive, with slow but progressive growth. It accounts for 2-7% of all odontogenic tumors and is less frequent. The epithelial lining of the odontogenic cyst may transform into an odontogenic neoplasm-like ameloblastoma or AOT (1-3). There are very few reports of odontogenic tumors either arising from or associated with odontogenic cysts. The purpose of this paper is to present a rare case of AOT which originated in the wall of a dentigerous cyst.

#### Case report

A 12 year-old male was referred to KM Shah Dental College & Hospital, Vadodara, with the chief complaint of a swelling in the anterior region of the lower jaw. Intraoral examination revealed a firm well-defined swelling extending from the lower left central incisor to the second premolar of the same side. The swelling was non-tender. The overlying

iznad nje bila je ružičasta i bez ulceracija, intaktna, normalnog izgleda te nije bilo znakova maligniteta. Rentgenska slika pokazala je dobro definiranu, unilokularnu radiolucidentnu leziju povezanu s neizniknutim trajnim očajnikom koja se protezala od apeksa lijevog središnjeg sjekutića do drugog lijevog prekutnjaka. Unilokularna radiolucencija povezana s lijevim neizniknutim trajnim očajnikom bila je vidljiva i na mandibularnoj okluzalnoj slici.

Na temelju kliničke i radiološke obrade prihvaćena je diferencijalna dijagnoza odontogene ciste, adenomatoidnog odontogenog tumora te odontogene keratociste.

Cijela je tvorba izvađena zajedno s neizraslim trajnim očajnikom te je poslana na histopatološku obradu. Prvim preliminarnim pregledom ustanovljena je cistična lezija veličine 5x4 centimetra.

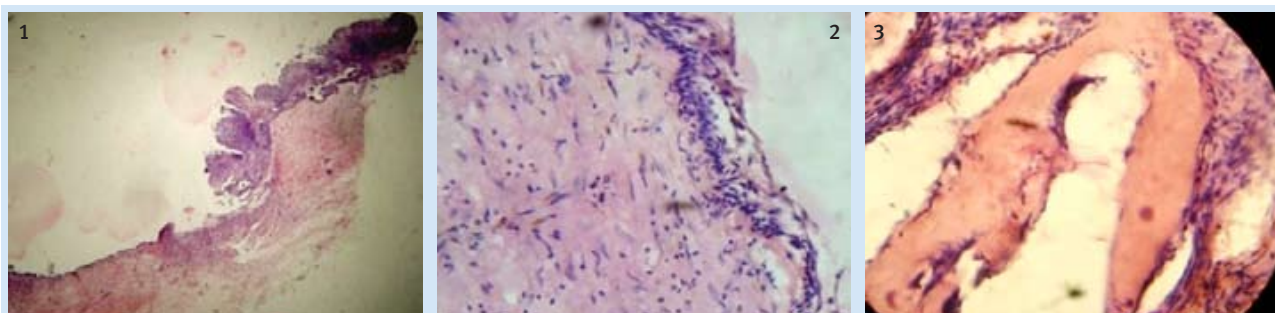
Uzorak je obojen metodom H i E te je ustanovljena cistična tvorba obložena nekeratiziranim višeslojnim pločastim epitelom debljine 2 do 3 sloja stanica. Ponegdje su bila žarišta intraluminalnih proliferativnih otočića stanica cilindričnog oblika. Neke su bile oblikovane poput rozeta (slike 1. i 2.). Sloj tkiva ispod višeslojnog epitela sastojao se od kolagenih vlakana u snopovima s pokojom zaostalim epitelom stanicom i krvnim žilama. U pojedinim dijelovima nalazile su se stanice kubičnog epitela u mrežastom uzorku ili poput čipke, uz vrpce stanica s hrskavičnim tkivom (slika 3.). U nekim dijelovima preparata pronađen je i homogeni eozinofilni materijal ili strukture sa srednjim dijelom obloženim kubičnim stanicama (slika 4.). Epitelni sloj uz eozinofilni materijal sličio je strukturi nalik na zub, a nalazio se u praznom prostoru u sredini tvorbe. Na temelju toga nalaza postavljena je dijagnoza adenomatoidnog odontogenog tumora nastalog iz odontogene ciste.

mucosa was non-ulcerated and pink in color. There was no evidence of any malignancy and the mucosa was normal and intact. Radiographs revealed a well-defined, unilocular radiolucent lesion associated with an unerupted lower left permanent canine extending from the apex of the left central incisor to the distal aspect of the left second premolar. A unilocular radiolucency with an unerupted left permanent canine was also seen on the mandibular occlusal image.

Based on the clinical and radiographic findings, the differential diagnoses of dentigerous cyst, AOT and odontogenic keratocyst were made.

The mass was enucleated completely along with the embedded canine and the specimen was submitted for the histopathological examination. The examination revealed a cystic lesion measuring 5 cm X 4 cm.

H & E stained sections of the excised specimen showed a cystic lining of non-keratinized stratified squamous epithelium of 2-3 cell thickness with focal areas of intra-luminal proliferating islands with spindle shaped cells, some arranged in a rosette pattern (Figures 1 and 2). The underlying connective tissue showed collagen fibers arranged in bundles with few epithelial rest cells and blood vessels. In certain areas, cubical cells were arranged in a cribriform or lace-like pattern showing cords of cells with associated hyaline material (Figure 3). In other areas, homogenous eosinophilic material or structure with a central space lined by columnar cells was also seen (Figure 4). The epithelial lining along with the eosinophilic material resembled 'tooth-like' structure encircling the empty space in the centre. A diagnosis of AOT arising from the dentigerous cyst was made.



**Slika 1.** Fotomikrograf pokazuje tanki nekeratizirani višeslojni pločasti epitel s proliferacijom u lumen (metoda H i E obojenje X 50).

**Figure 1** Photomicrograph showing thin non-keratinised stratified squamous epithelium lining with proliferation of the lining into the lumen (H & E stain X 50).

**Slika 2.** Fotomikrograf pokazuje nekeratizirani višeslojni pločasti epitel tipičan za odontogenu cistu (metoda H i E, obojenje X 100).

**Figure 2** Photomicrograph showing the non-keratinised stratified squamous epithelium lining typical of dentigerous cyst (H & E stain X 100).

**Slika 3.** Fotomikrograf pokazuje žarišnu proliferaciju epitela u lumen sa strukturom nalik na ameloid sličan adenomatoidnom odontogenom tumoru (metoda H i E obojenje X 100).

**Figure 3** Photomicrograph showing focal luminal proliferation of lining epithelium with amyloid-like material, mimicking adenomatoid odontogenic tumor (H & E stain X 100).

**Slika 4.** Fotomikrograf pokazuje strukturu nalik na zub koja okružuje prazan prostor u sredini (H i E obojenje X 100).

**Figure 4** Photomicrograph showing a tooth-like structure encircling the empty space in the centre. (H & E stain X 100).

## Rasprava

Adenomatoidni odontogeni tumor prvi je opisao Gosh (4) kao adamantinom maksile, a Staphne (5) ga je prvi, još 1948. godine, smatrao zasebnim patološkim entitetom. Prema drugom izdanju "Histološke tipizacije odontogenih tumora" SZO-a (6), ta je novotvorina definirana kao "tumor nastao iz odontogenog epitela sa cjevastim strukturama i različitog stupnja promjena u vezivnom tkivu".

Adenomatoidni odontogeni tumor benigna je lezija koja nastaje iz kompleksnog sustava dentalne lamine i njezinih ostataka (4). U literaturi se u nekim radovima navodi da neke odontogene ciste nastaju zajedno s odontogenim tumorima (7). Zbog toga što se neoplastične i hamartomatozne lezije pojavljuju u bilo kojem stadiju odontogeneze, te odontogeni tumori s kombiniranim epitelnim i mezenhimalnim komponentama mogu nastati iz odontogenih cista (8).

U ovom slučaju se adenomatoidni odontogeni tumor i odontogena cista nalaze u istoj leziji. Naše je stajalište da se najprije razvila cista, a zatim iz njezine ovojnice adenomatoidni odontogeni tumor. Opisano je malo slučajeva povezanih s odontogenom cistom. Tako su Garcia-Pola i suradnici (1) predstavili proliferaciju adenomatoidnog odontogenog tumora iz epitelne ovojnice odontogene ciste. Tajima i njegovi kolege (9) opisali su takvu novotvorinu lociranu na gornjem dijelu maksilarnog sinusa te pretpostavili da se razvila iz odontogene ciste. Philipsen i suradnici (10–12) zaključili su da se folikularni oblik adenomatoidnog odontogenog tumora razvija iz gnijezda stanica unutar dentalne lamine te zbog toga okružuje zub. Vrlo je malo podataka o nastanku tumora iz odontogene ciste. Tijekom sustavne pretrage medicinske literature na engleskom jeziku nađeno ih je ukupno sedam, a samo je nekoliko bilo povezano s pojavom tumora u maksilarnom sinusu (1, 7–9, 13, 14).

Nejasno je ima li ta pojava (adenomatoidni odontogeni tumor iz odontogene ciste) agresivniji potencijal. Adenomatoidni odontogeni tumor i odontogena cista benigne su inkapsulirane lezije i liječe se konzervativnom kirurškom metodom enukleacije ili kiretažom. Pretpostavljamo da se u ovom slučaju radi o odontogenoj cisti s neoplastičnim razvojem te epitelnim i mezenhimalnim komponentama. Detaljna histopatološka pretraga bitna je u procesu obrade svake enukleirane ciste te pridonosi dijagnosticiranju sličnih slučajeva (15).

I na kraju, ovaj slučaj je rijedak primjer adenomatoidnog odontogenog tumora povezanog s odontogenom cistom i zbog toga se ističe važnost histopatološke obrade cističnih lezija u čeljusti.

## Sukob interesa

Autori izjavljuju da nisu bili u sukobu interesa dok su se pripremali za objavljivanje ovog slučaja.

## Discussion

Adenomatoid odontogenic tumor was first described by Ghosh (4) as an adamantinoma of the maxilla and was first recognized as a distinct pathological entity by Staphne (5) in 1948. According to the second edition of the WHO "Histological Typing of Odontogenic Tumors" (6), AOT is defined as "A tumor of odontogenic epithelium with duct-like structures and with varying degrees of inductive change in the connective tissue". Adenomatoid odontogenic tumor is a benign lesion derived from the complex system of dental lamina or its remnant (4). It has been reported that some odontogenic cysts occur in association with odontogenic tumors (7). Since neoplastic and hamartomatous lesions can occur at any stage of odontogenesis, odontogenic tumors with combined features of epithelial and mesenchymal components may arise within the odontogenic cyst (8).

In the present report, AOT and dentigerous cyst are found in the same lesion. We believe that the dentigerous cyst developed first followed by the AOT in the cyst wall. Very few cases have been described that arise in association with a dentigerous cyst. Garcia-Pola et al. (1) have described the proliferation of an AOT in the epithelial lining of a dentigerous cyst. Tajima et al. (9) described an AOT located in the superior portion of the maxillary sinus and speculated that the tumor was derived from a dentigerous cyst. Philipsen et al. (10–12) also postulated that the follicular type of AOT develops from nests of cells within the dental lamina and, therefore, as a result, surrounds the tooth. Very few cases have been described that arise in association with a dentigerous cyst. A systematic search of the English language medical literature revealed only seven such cases, and only a couple of cases of its occurrence in the maxillary sinus (1, 7–9, 13, 14).

It is unclear whether this entity (AOT arising from dentigerous cyst) has a more aggressive potential. The AOT and dentigerous cyst are both benign, encapsulated lesions and conservative surgical enucleation or curettage is the treatment of choice. We believe that the present case represents an odontogenic cyst with neoplastic development, containing both epithelial and mesenchymal components. Meticulous histopathological evaluation is thus required of all enucleated cysts, which could contribute to the diagnosis of similar cases as reported in the present study (15).

To conclude, the present report describes a rare case of AOT associated with a dentigerous cyst, highlighting the importance of the histopathological examination in the cystic lesions of the jaw bones.

## Conflict of interest

The authors declare that they have no conflict of interest in preparation and publication of this case.

**Abstract**

Adenomatoid odontogenic tumor (AOT) is an uncommon benign tumor of odontogenic origin composed of odontogenic epithelium in a variety of histopathological patterns. Recently, the WHO classification of odontogenic tumors has been revised and few changes have been made. According to WHO, the AOT is reclassified as an epithelial odontogenic tumor. Very few cases of AOT associated with a dentigerous cyst have been reported in the available literature. We present an additional case of an AOT arising from a dentigerous cyst around the crown of an unerupted lower canine in a 12-year-old boy, which was clinically diagnosed as a dentigerous cyst. Histologically, our case showed 'odontoma-like' areas which have been described in only two other previous case reports. We believe that this case represents an odontogenic cyst with neoplastic change containing both epithelial and mesenchymal components. Further studies to determine whether the AOTs derived from an odontogenic cyst could represent a distinct 'hybrid' variant (different from the three variants described so far) are to be done.

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**Key words**

Odontogenic Tumors; Dentigerous Cyst

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