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## KLINIČKE I RADIOGRAFSKE ODLIKE ADENOMATOIDNOG ODONTOGENOG TUMORA

## CLINICAL AND RADIOGRAPHIC FEATURES OF ADENOMATOID ODONTOGENIC TUMOUR

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### Sažetak

**Uvod:** Adenomatoidni odontogeni tumor (AOT) je neuobičajeni, neagresivni tumor poreklom od odontogenog epitela. On čini 3-7% svih odontogenih tomora.

**Cilj** ovog članka bio je da utvrdi trenutne mogućnosti kliničkog i radiografskog ispitivanja odlike AOT. Koristeći odgovarajuće ključne reči, pretražili smo obilnu literaturu PubMed baze podataka u poslednjih deset godina. Od 57 članaka o AOT, 35 je imalo studijske kriterijume i oni su uključeni u pregled.

**Rezultati** pokazuju da je najčešće mesto pojave AOT u posteriornim delovima mandibule, što je u suprotnosti sa postojećim podacima iz literature, prema kojima se tvrdi da se AOT najčešće pojavljuje u anteriornom regionu maksile. Ovaj članak daje korisne informacije povećane demografske, kliničke, radiografske i histološke odlike AOT.

**Ključne reči:** adenomatoidni odontogeni tumor, odontogeni tumor, intrakoštani

### Abstract

**Introduction:** Adenomatoid odontogenic tumour (AOT) is an uncommon, non-aggressive tumour arising from the odontogenic epithelium. It accounts for 3-7% of all odontogenic tumors.

**The objective** of this systematic review was to determine the recent trends in the clinical and radiographic features of AOT. We conducted a comprehensive literature search regarding AOT in PubMed database over the last ten years using suitable search words. Out of 57 articles of AOT, 35 met the study criteria and were included in the review.

**The results** our review results showed that the mandibular posterior region was the commonest site for the occurrence of AOT. This is contrary to the existing literature data which state that AOT is commonest in the maxillary anterior region. This review provides valuable information and update regarding the demographic details, clinical, radiographic and histologic features of AOT.

**Key words:** adenomatoid odontogenic tumour, odontogenic tumour, intraosseous

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## ***Uvod***

Adenomatoidni odontogeni tumor (AOT) je neuobičajen, neagresivni tumor poreklom od odontogenog epitela<sup>1</sup>. Prvi put ga je opisao Dreibladt<sup>2</sup> 1907. godine kao psedo-adenoameloblastom, a kasnije i Harbitz<sup>3</sup>, kao ameloblastom. Philipsen i Birn<sup>4</sup> su predložili da ne bude posmatran kao ameloblastom na osnovu njegovog kliničkog ponašanja, već su predložili termin „adenomatoidni odontogeni tumor“. Svetska zdravstvena organizacija je 2005. godine definisala AOT kao tumor sastavljen od odontogenog epitela sa raznolikom histoarhitekturom, koji je ugrađen u zrelo vezivno tkivo strome i karakteriše se sporim ali progresivnim rastom<sup>5</sup>.

Tumor se nekada naziva i dvotrećinski tumor, zbog toga što se 2/3 slučajeva javlja u maksili, 2/3 slučajeva kod mladih žena, 2/3 slučajeva je udruženo sa neizniklim zubima i 2/3 slučajeva su očnjaci<sup>6</sup>.

## ***Metodologija***

Sprovedno je sveobuhvatno istraživanje literature PubMed baze podataka, sa terminima „adematoidni odontogeni tumor“, „AOT“, „radiografske odlike“ i „impaktirani zub“ u periodu od 2005. do 2014. godine. Pronađeno je 57 članaka preliminarnom pretraživanju baze podataka, među kojima je 35 imalo uključujuće i isključujuće kriterijume Tabela 1. Dobijeni su izgled pacijenta, klinički, radiološki i histološki podaci i prezentovani u Tabeli 2<sup>2,6-36</sup>. Lokacija ciste je razmatrana u regiji sekutića, očnjaka, premolara, molara i retr/molara. Kada lezija zahvata više regija, smatra se ugroženim centralni deo regije.

## ***Introduction***

Adenomatoid odontogenic tumour (AOT) is an uncommon, non-aggressive tumour of the odontogenic epithelium<sup>1</sup>. It was first described by Dreibladt<sup>2</sup> in 1907 as pseudo adenoameloblastoma and later by Harbitz<sup>3</sup> in 1915 as cystic adamantoma. Philipsen and Birn<sup>4</sup> in 1969 suggested that it should not be considered as ameloblastoma due to its different clinical behaviour and proposed the term ‘adenomatoidodontogenic tumour’. WHO in 2005 defined AOT as ‘a tumour composed of odontogenic epithelium presenting a variety of histoarchitectural patterns, embedded in a mature connective tissue stroma and characterised by slow but progressive growth<sup>5</sup>.

The tumour is sometimes referred to as ‘two-thirds tumour’ because it occurs in the maxilla in about 2/3 of the cases, about 2/3 of the cases arise in young females, 2/3 of the cases are associated with an unerupted tooth, and 2/3 of the affected teeth are the canines<sup>6</sup>.

## ***Methodology***

A comprehensive literature search of the PubMed database with terms such as ‘adenomatoid odontogenic tumour’, ‘AOT’, ‘radiographic feature’ and ‘impacted tooth’ was conducted from 2005-2014. Fifty seven articles were identified in the preliminary database search, among which 35 met the inclusion and exclusion criteria Table 1. Patient demographics, clinical, radiographic and histopathologic information were obtained from these records and are presented in Table 2<sup>2, 6-36</sup>. The location of the cyst was considered as incisor, canine, premolar, molar and retr/molilar areas. When the lesion extended to more than one area, the central part of the lesion was considered as the affected site.

**Tabela 1.** Tabela prikazuje kriterijume uključivanja i isključivanja  
**Table 1.** Table with the inclusion and exclusion criteria

INCLUSION CRITERIA UKLJUČUĆI KRITERIJUMI	EXCLUSION CRITERIA ISKLJUČUĆI KRITERIJUMI
The paper must be in full text and written in English Članak mora biti u punom tekstu na engleskom jeziku	Cases with ambiguous diagnosis Slučajevi sa dvosmislenom dijagnozom
The age and gender of the patient must clearly be mentioned Starost i pol pacijenata moraju biti jasno naglašeni	Cases without any mention of radiographic or histological features Slučajevi bez ikakvog pomena radiografskih ili histoloških karakteristika
The site of the lesion must be described in detail Mesto lezije mora biti detaljno opisano	
The radiographic features of the case must be mentioned along with the presence or absence of an impacted tooth Radiografske odlike slučaja moraju biti pomenute u okviru prisustva ili odsustva impaktiranog zuba	
The histological features of the lesion must be confirmatory of AOT Histološke odlike AOT moraju biti potvrđene	

**Tabela 2.** Podaci o prijavljenim slučajevima adenomatoidnog odontogenog tumora  
**Table 2.** Table with the data of reported cases of adenomatoid odontogenic tumour

SL.NO	AUTHOR AUTOR	AGE / GENDER STAROST/ POL	SITE MESTO	RADIOGRAPHIC FEATURES RADIOGRAFSKE ODLIKE	ASSOCIATION WITH IMPACTED TOOTH UDRUŽENOST SA IMPAKTIRANIM ZUBOM	HISTOLOGICAL FEATURES HISTOLOŠKE ODLIKE
1.	Batra P et al (2005) <sup>2</sup>	17/F 17/Ž	Right mandibular canine region  Regija mandibularnog očnjaka	Well circumscribed radiolucency  Jasno okruglo rasvetljenje	Yes, canine  Da, sa očnjakom	Ductal lamina surrounded by epithelial cells filled with eosinophilic material  Duktalna lamina, okružena epitelijalnim ćelijama, prekrivena eozinofilnim materijalom

2.	Garg D et al (2009) <sup>6</sup>	20/F 20/Ž	Right maxillary incisor-premolar region  Regija desnih i maksilarnih sekutića i pretkutnjaka	Ill-defined radiolucency with radiopaque foci, root resorption  Jasno definisano rasvetljenje, sa fokalnim zasenčenjima, resorpcija korena	Yes, canin  Da, sa očnjakom	Cells arranged in whorl-like, ductular, ring-like and ribbon-like patterns with bands of eosinophilic material  Ćelije aranžirane u spiralne, duktusne, prstenaste i trakaste strukture sa grupisanim eozinofilnim materijalom
3.	Handschei J et al (2005) <sup>7</sup>	23/M 23/M	Right mandibular canine region  Regija desnog mandibularnog očnjaka	Unilocular radiolucency  Unilokularno rasvetljenje	Yes, canine  Da, sa očnjakom	Whorles, sheets and plexiform arrangement of cells, duct like structures with calcifications  Spiralni, listasti i peksiformni aranžmani, duktolike strukture sa kalcifikacijama
4.	Motamedi MHK et al (2005) <sup>8</sup>	13/F 13/Ž	Left mandibular canine region  Regija levog mandibularnog očnjaka	Well defined radiolucency  Jasno definisano rasvetljenje	Yes, canine  Da, sa očnjakom	Rosettes of spindle shaped epithelial cells with duct like structures  Rozete igličastih epitelijalnih ćelija sa duktolikim strukturama
5.	Nigam S et al (2005) <sup>9</sup>	15/F 15/Ž	Left maxillary incisor to premolar region  Regija od levog maksilarnog sekutića do pretkutnjaka	Well defined radiolucency, root resorption  Jasno definisano rasvetljenje i resorpcija korena	Yes, canine  Da, sa očnjakom	Nests, cords and ducts of epithelial cells with eosinophilic amyloid like material  Ćelije koje liče na ameloblaste sa listastim retikulum stelatumom i glednim matriksom koji prekriva dentin
6.	Vargas PA et al (2006) <sup>10</sup>	16/M 16/M	Left mandibular molar  Region levih mandibularnih kutnjaka (molara)	Well defined unilocular radiolucency with discrete areas of radiopacities  Jasno definisano unilokularno rasvetljenje sa zonama diskretnog zasenčenja		

7.	Sempere FJV et al (2006) <sup>11</sup>	9/F 9/F	Left mandibular canine region  Regija levih mandibularnih očnjaka	Well defined radiolucency  Jasno definisano rasvetljenje	Yes, canine  Da, sa očnjakom	Whorled nests of odontogenic epithelium with calcified basophilic spherules, irregular hyaline and amorphous deposits  Spiralna gnezda odontogenog epitelja sa kalcifikovanim bazofilnim sferama, i regularnim hijalinim i amorfnim depozitima
8.	Jivan V et al (2007) <sup>12</sup>	40/M 40/Ž	Right mandibular premolar to molar region  Regija od desnog mandibularnog pretkutnjaka do kutnjaka	Well defined radiolucency with root resorption  Jasno definisano rasvetljenje i resorpcija korena	No Ne	Clusters and strands of cuboidal cells arranged in a lace-like pattern with hyaline material  Bokori i trake kuboidnih ćelija aranžiranih u strukture slične čipki sa hijalinim materijalom
9.	Nonaka CFW et al (2007) <sup>13</sup>	13/F 13/Ž	Left maxillary canine to pre-molar region  Regija levog maksilarnog očnjaka do pretkutnjaka	Well defined radiolucency with radiopaque areas  Jasno definisano rasvetljenje sa zonom zasenčenja	Yes, canine  Da, sa očnjakom	Islands and sheets of globular cells with duct like structures, amorphous eosinophilic material and calcified areas  Ostrvca, listasti i globularni ćelijski infiltrati, sa strukturama sličnim duktusima, amorfnim eozinofilnim materijalom i kalcifikovanim poljima
10.	Chuan Xiang Z et al (2007) <sup>14</sup>	16/M 16/Ž	Right maxillary canine region  Regija maksilarnog očnjaka	Well defined unilocular radiolucency  Jasno definisano unilokularno rasvetljenje	Yes, canine  Da, sa očnjakom	Nests or rosette like structures and tubular or duct like appearance of odontogenic epithelium with eosinophilic amorphous material and globular calcifications  Odontogeni epitel koji se javlja u vidu gnezda, rozete, tubula ili duktusa, sa amorfnim eozinofilnim materijalom i tubularnim kalcifikacijama
11.	Kemp S et al (2008) <sup>15</sup>	46/M 46/M	Left Mandibular molar region  Regija levog mandibularnog kutnjaka	Unilocular radiolucency with radiopaque foci  Unilokularno rasvetljenje sa fokalnim zasenčenjima	No Ne	Sheets of spindle and epithelial cells, duct like spaces, areas of amorphous calcification  Listaste i igilčaste epitelne ćelije, duktusni prostori, polja amorfne kalcifikacije

12.	Bartake AR et al (2009) <sup>16</sup>	14/F 14/Ž	Maxillary anterior to left posterior region  Od prednjeg dela do zadnje leve maksilarne regije	Multilocular radiolucency with radiopaque foci  Multilokalno rasvetljenje sa fokalnim zasenčenjima	Yes, canine  Da, sa očnjakom	Rosette like structures containing eosinophilic droplets, tubular duct like spaces, focal areas of calcification  Strukture u vidu rozete koje sadrže eozinofine kapi kanalikularne prostore i polja fokalne kalcifikacije
13.	Carlos-Bregni R (2009) <sup>17</sup>	19/F 19/Ž	Left mandibular molar region  Regija levog mandibularnog kutnjaka	Unilocular radiolucency  Unilokalno rasvetljenje	No  Ne	Cuboidal cells resembling ameloblasts and stellate reticulum, presence of enamel matrix overlying dentin  Kuboidalne ćelije nalik ameloblastima i retikulumom stelatum, prisustvo glednog matriksa koji prekriva dentin
14.	Carlos-Bregni R (2009) <sup>17</sup>	13/F 13/Ž	Left mandibular molar region  Regija levog mandibularnog kutnjaka	Unilocular radiolucency  Unilokalno rasvetljenje	No  Ne	Globular dentin, enamel matrix, stellate reticulum, odontogenic epithelium  Globularni dentin, gledni matriks, retikulum stelatum, odontogeni epitel
15.	Ide F (2009) <sup>18</sup>	22/F 22/Ž	Right mandibular pre-molar region  Regija desnog mandibularnog pretkutnjaka	Unilocular radiolucency with snow-flake like radiopacities  Unilokalno rasvetljenje sa pahuljičastim zasenčenjima	No  Ne	Tubular and duct like structures, globular calcifications  Tubularne i duktusne strukture, globularne kalcifikacije
16.	Martinez A et al (2009) <sup>19</sup>	10/F 10/Ž	Left mandibular molar region  Regija levog mandibularnog kutnjaka	Unilocular radiolucency with radiopaque mass  Unilokalno rasvetljenje sa masom zasenčenja	No  Ne	Rosette-like and duct-like structures with intercellular eosinophilic material, calcified masses of tubular dentin and dentinoid material with enamel matrix foci  Ružoloke i duktusne strukture sa intracelularnim eozinofilnim materijalom, kalcifikovanom masomubularnog dentina i dentoidnog materijala sa fokusim glednog matriksa

17.	Friedrich R et al (2009) <sup>20</sup>	16/M 16/M	Left maxillary canine region  Regija levih maksilarnih očnjaka	Slight radiopacity of the sinus with punctate radiopacities  Blago zasenčenje sinusa sa tačkastim senkama	Yes, canine  Da, očnjak	Rosette-like and tubular structures with eosinophilic cellular material  Ružolike i tubularne strukture sa eozinofinim čelijskim materijalom
18.	Yilmaz N et al (2009) <sup>21</sup>	15/F 15/Ž	Mandibular anterior region  Regija prednje mandibule	Periapical radiolucency with fine calcifications  Periapikalno rasvetljenje sa finim kalcifikacijama	No Ne	Ameloblast like cells forming duct like structures, eosinophilic droplets and odontogenic calcifications  Ćelije slične ameloblastima koje formiraju kanalne strukture, eozinofilne kapljice i odontogene kalcifikacije
19.	Friedrich RE et al (2010) <sup>22</sup>	23/M 23/M	Right mandibular canine region  Regija desnog mandibularnog očnjaka	Unilocular radiolucency with punctuate radiopacities  Unilokularno rasvetljenje sa tačkastim zasenčenjima	Yes, canine  Da, sa očnjakom	Rosette like or tubular structures with eosinophilic cellular material  Ružolike i tubularne strukture sa eozinofinim čelijskim materijalom
20.	Tejasvi A et al (2010) <sup>23</sup>	15/M 15/M	Left maxillary incisor region  Regija levih maksilarnih sekutića	Well defined unilocular radiolucency  Jasno definisano unilokularno rasvetljenje	No Ne	Odontogenic epithelium in duct like pattern, eosinophilic coagulum  Odontogeni epitel u formi duktusa sa eozinofinim koagulumom
21.	Sandhu SV et al (2010) <sup>24</sup>	25/F 25/F	Right maxillary incisor to pre-molar region  Regija levog i maksilarнog sekutića i pretkutnjaka	Expansion and thinning of bony sinus (CT)  Ekspanzija i istanjenje koštanih zidova sinusa (CT)	Yes, canine  Da, sa očnjakom	Nests, rosettes, cribriform and lace like pattern of cuboidal cells with hyaline material  Gnezdaste, ružolike, korpaste i čipkaste forme kuboidalnih ćelija sa hijalinim materijalom

22.	John JB et al (2010) <sup>25</sup>	39/F 39/Ž	Left maxillary pre-molar to molar region  Regija levog maksilarnog kutnjaka i pretkutnjaka	Well defined unilocular corticated radiolucency  Jasno definisano unilokularno rasvetljenje sa zonom kompakte	Yes, molar  Da, sa molarom	Whorls, rosettes and duct like pattern of epithelial cells with calcification  Epitelne ćelije sa kalcifikacijama u obliku spirale, rozete, i duktusa
23.	Sekia R et al (2011) <sup>26</sup>	21/F 21/Ž	Right maxillary incisor to molar region  Regija desnog maksilarnog sekutića i pretkutnjaka	Well defined radiolucency with calcified structures  Jasno definisano rasvetljenje sa kompaktnim strukturama	Yes, canine  Da, sa očnjakom	Pseudo-tubular structures, acidophilic hyaline like material, dystrophic calcifications  Pseudo tubularne strukture, acidofilni, hijalini materijal, distrofične kalcifikacije,
24.	Bhaskaran P et al (2011) <sup>27</sup>	34/F 34/Ž	Left maxillary canine region  Regija levog maksilarnog očnjaka	Well circumscribed sclerotic radiolucency, root resorption  Jasno, okruglasto sklerotično rasvetljenje, korenska resorpcija	Yes, supernumerary tooth  Da, preko brojnih zuba	Whorls, sheets and duct like structures lined by epithelial cells with evidence of calcification  Epitelne ćelije u vidu spirale, lista ili duktusa, sa prisutnom kalcifikacijom
25.	Bhaskaran P et al (2011) <sup>27</sup>	15/F 15/Ž	Left maxillary canine region  Regija levog maksilarnog očnjaka	Well circumscribed unilocular radiolucency with flecks of calcifications  Jasno, okruglasto unilokularnorasvetljenje, sa pahuljičastom kalcifikacijom	Yes, canine  Da, sa očnjakom	Focal areas of solid tumours showing whorls, sheets and duct like structures  Fokalna polja solidnih tumora spiralne, listaste i duktusne strukture

26.	Singh V et al (2012) <sup>28</sup>	18/F 18/Ž	Right maxillary incisor to molar region  Regija od desnog maksilarног sekutića do kutnjaka	Irregular corticated radiolucency  Iregularno kortikalno rasvetljenje	Yes, canine  Da, sa očnjakom	Islands and ducts lined by cuboidal cells with amorphous calcified material  Ostrvca i kanalići kiuboidalnih ćelija sa amorfim kalcifikonim materijalom
27.	Mutalik VS et al (2012) <sup>29</sup>	14/F 14/Ž	Left maxillary canine region  Regija levog maksilarног očnjaka	Well defined radiolucency, root resorption  Jasno, definisano rasvetljenje, korenska resorpcija	Yes, canine  Da, sa očnjakom	Whorls, rosettes, duct like and tubular arrangement of epithelial cells with areas of calcification, eosinophilic material and leisegang rings  Spiralni, ružoliki, kanalni i tubularni aranžmani epitelnih ćelija sa polljima kalcifikacije, eozinofinim materijalom
28.	Gomez RS et al (2013) <sup>30</sup>	32/M 32/M	Left mandibular retromolar area  Leva mandibularna površina iza kutnjaka	Well circumscribed unilocular radiolucency  Jasno okruglo unilokularno rasvetljenje	No  Ne	Nodules, cords and strands of epithelial cells, swirls and ribbons of ameloblast cells, islands of odontogenic epithelium with enamel matrix deposits  Čvorići, vrpe i niti epitelnih ćelija upletene trake ameloblasta, ostrvca odontogenog epitelia sa depozitima glednog matriksa
29.	Narayan VS et al (2013) <sup>31</sup>	17/F 17/Ž	Left mandibular molar to right mandibular molar region  Regija od desnog kutnjaka	Multilocular radiolucency with radiopaque foci and septae, root resorption  Multilokularno rasvetljenje sa fokalnim zasećenjima, septumima i korenskom resorpcijom	Yes, incisor  Da, sa incizivom	Whorls and duct like arrangement of epithelium with foci of basophilic calcifications  Epitel u vidu duktusa i spirala, sa fokalnim bazofilnim kalcifikacijama
30.	Shivali V et al (2013) <sup>32</sup>	18/M 18/M	Left mandibular ramus and retromolar area  Levi ramus mandibule i predeo iza kutnjaka	Well defined unilocular sclerotic radiolucency  Jasno definisano unilokularno rasvetljenje sa zonom skleroze	No  Ne	Ductal, rosettes and convoluted pattern of odontogenic epithelial cells with globular irregular calcifications  Duktusi, rozete odontogene epitelne celije uvijene u rozete i kanaliće, sa iregularnim globularnim kalcifikacijama

31.	Marelli M et al (2014) <sup>33</sup>	18/F 18/Ž	Right maxillary canine region  Regija desnog maksilarnog očnjaka	Well circumscribed radiolucency  Jasno okruglasto rasvetljenje	Yes, canine  Da, sa očnjakom	Odontogenic epithelium with formation of follicles, spindle cells, psammoma bodies  Odontogeni epitel koji formira iglice, i folikule
32.	Sharma N et al (2014) <sup>34</sup>	14/F 14/F	Right mandibular canine to molar region  Desni mandibularni očnjak	Well defined radiolucency with flecks of calcification  Jasno definisano rasvetljenje sa pahuljičastim kalcifikacijama	Yes, canine and premolars  Da, očnjak i premolar	Duct like pattern with foci of calcifications  Duktolike forme i fokalne kalcifikacije
33.	Bhullar RPK et al (2014) <sup>35</sup>	24/F 24/F	Right mandibular incisor to molar region  Regija desnog mandibularnog sekutića do kutnjaka mandibule	Well defined unilocular corticated radiolucency  Jasno definisano unilokularno kortikalno rasvetljenje	Yes, premolar  Da, sa premolarom	Nests, whorls and rosette like structures with eosinophilic hyaline droplet material and calcifications  Strukture u vidu gnezda, spirale i rozete, sa eozinofinim hijalinim kapljicama i kalcifikacijama
34.	Vasudevan K et al (2014) <sup>36</sup>	14/F 14/Ž	Right maxillary anterior region  Desna maksilarna anteriorna regija	Well defined radiolucency with flecks of calcifications  Jasno definisano rasvetljenje sa pahuljičastim kalcifikacijama	Yes, canine and premolar  Da, sa očnjakom i premolarom	-

## Diskusija

Adenomatoidni odontogeni tumor je benigni, sporo rastući, neinvazivni odontogeni tumor, koji čini 3-7% svih odontogenih tumora<sup>2</sup>. Poreklo tumora je kontraverzno, dok neki autori tvrde da se radi o hamartromu<sup>6</sup>. Većina autora prihvata da je odontogenog porekla, jer se nalazi u vilicama u predelu zuba i povezan je često sa impaktiranim zubima. Postoji dokaz da tumorske ćelije vode poreklo od epitela gleđnog organa; istraživači su takođe sugerisali da tumorske ćelije vode poreklo od ostataka dentalne lamine<sup>27</sup>. Kako je histogeneza još uvek nesigurna, dugo se raspravljaljalo o tome da li ima hamartomazni rast ili je prava benigna neoplazma, koja ima citološke odlike slične raznim komponentama gleđnog organa, dentalne lamine i redukovanih gleđnih epitela<sup>6</sup>. Karakteristične odlike ove lezije vazane su i za visoku sposobnost diferencijacije epitelnih komponenti i gubitak prijemčivosti mezenhimalnog tkiva za epitel<sup>1</sup>.

Martinez A. i sar.<sup>19</sup> utvrdili su da se AOT obično javlja kod mladih pacijenata, u drugoj deceniji života i neuobičajeno je kod

## Discussion

The adenomatoid odontogenic tumour is a benign, slow growing, non-invasive odontogenic lesion that accounts for 3%-7% of all odontogenic tumours<sup>2</sup>. The origin of AOT is controversial with some authors claiming it to be a hamartoma<sup>6</sup>. However, most authors accept its odontogenic source as it occurs within the tooth bearing areas of the jaws and is often found in close association with impacted teeth. There is evidence that the tumour cells are derived from enamel organ epithelium; investigators have also suggested that the lesion arises from remnants of dental lamina<sup>27</sup>. As the histogenesis is still uncertain, there has long been a debate as to whether it represents a hamartomatous growth or a true benign neoplasm, having cytological features similar to various components of enamel organ, dental lamina and reduced enamel epithelium<sup>6</sup>. The characteristic features of this lesion are due to both the highly differentiating ability of the epithelial component and the lack of mesenchymal tissue receptive to the influence of the epithelium<sup>1</sup>.

pacijenata koji imaju preko 30 godina. Žene su češće pogodjene od muškaraca u odnosu 1,9:1<sup>37,38</sup>. Batra i sar<sup>2</sup> otkrili su da je 69% AOT dijagnostikovano u drugoj deceniji života, a više od polovine slučajeva javlja se u tinejdzerskom periodu. U prisutnoj analizi utvrđeno je da je starost pacijenta između 9 i 46 godina sa srednjim dobom od 19 godina. Prijavljeno je 22 slučaja u drugoj dekadi života i samo jedan (2,8%) slučaj u petoj deceniji života. Na ženski pol (71,4%) odnosi se dvadeset pet slučajeva dok su desetorica bili muškarci (28,5%), što ukazuje na odnos 2,5:1.

Lezija je retko veća od 3 cm i ima upečatljivu tendenciju da se javlja u prednjim delovima vilica. Dva puta češće se nalazi u maksili nego u mandibuli<sup>7,19</sup>. Leon i sar.<sup>39</sup> u njihovoј studiji od 39 slučajeva AOT utvrdili da je najčešće mesto pojave prednji deo maksile.

U svakom slučaju, među 35 slučajeva koje smo mi razmatrali u ovoj studiji, 19 slučajeva je bilo u mandibuli se (54,3%) dok je 16 bilo u maksili (45,7%). Posteriorne regije u mandibuli, su zahvaćene u 57,8% slučajeva, dok su prednje regije maksile pogodjene u 42,2%. Topografska podela u maksili pokazuje da su prednje regije najčešće pogodjena mesta sa frekvencijom od 93,7%, dok posteriorne regije pokazuju najmanje afiniteta za AOT (6,3%). Tako da je prema prisutnim podacima posteriorna mandibula predominantno mesto pojave. Ovaj podatak je u suprotnosti sa postojećim podacima iz literature<sup>13,15,16,19,39</sup>.

Uzrok ovih rezultata može biti sledeći: ili je on to pravi pokazatelj nedavnog trenda u nastanku AOT, ili je odraz pristrasnosti zbog čega se objavljuju samo slučajevi koji pokazuju retku prezentaciju AOT. Neprijavljinje slučajeva AOT koji se javljaju u maksilarnog prednjem području može biti još jedan od uzroka.

Adenomatoidni odontogeni tumor se može javiti intraosealno i ekstraosealno<sup>27</sup>. Intraosealni AOT ima ekpanzivni rast i može biti radiografski podeljen na dva tipa: folikularni (perikoronarni) i ekstrafolikularan (ekstrakoronarni). Fridrih i sar.<sup>20</sup> ukazuju da je folikularni tip češći tip AOT. Obično se manifestuje kao perikoronarno, jasno ograničeno, kružno, unilokularno rasvetljenje ili mešovita senka sa jasnom granicom kompaktne ili sklerotične kosti koja okružuje neiznikli Zub, dok se ekstrafolikularna varijanta javlja kao jasno definisano unilokularno rasvetljenje, između, iznad zuba ili kao rasvetljenje koje se superponira sa korenom zuba<sup>6</sup>.

Martinez A et al.<sup>19</sup> stated that AOT is usually encountered in young patients, usually in the second decade of life and is uncommon in patients over 30 years of age. Females are affected more often than males with a female to male ratio of 1.9:1<sup>37,38</sup>. Batra et al.<sup>2</sup> found that 69% of AOTs were diagnosed in the second decade of life and more than half occur during the teenage years. In the present analysis, it was seen that the age of the patients ranged from 9 to 46 years with a mean age of 19 years. Twenty two (62.8%) of the cases were reported in the second decade of life and only 1 (2.8%) case was seen in the fifth decade. Twenty-five of the cases affected females (71.4%), while 10 were males (28.5%) bringing the female to male ratio to 2.5:1.

The lesion is seldom more than 3 cm in size and has a striking tendency to occur in the anterior portions of the jaws. It is found twice as often in the maxilla as in the mandible<sup>7,19</sup>. Leon et al.<sup>39</sup> in their study of 39 cases of AOT found that anterior maxilla is the commonest site.

However, among the 35 cases that were considered in the present review, 19 cases affected the mandible (54.8%) while 16 cases affected the maxilla (45.7%). In the mandible, the posterior region was affected in 57.8% of the cases while the anterior region was affected in 42.1% of cases. In the maxilla, topographical division showed that the anterior region was the most commonly affected site with a frequency of 93.7% while the posterior region showed the least affinity for AOT (6.2%). Thus, according to the present review results posterior mandible is the predominant site of occurrence. This is contrary to the existing literature data<sup>13,15,16,19,39</sup>.

This result may be due to the following reasons: either it is a true indication of the recent trend in occurrence of AOT, or it is a reflection of publication bias wherein only cases which show a rare presentation are being published. Underreporting of cases of AOT occurring in the maxillary anterior region may be another cause.

AOT can occur both intraosseously and extraosseously<sup>27</sup>. Intraosseous AOT occurs as an expansile growth and can be radiographically divided into two types: follicular (pericoronal) and extrafollicular

Ekstraosealna AOT se prezentuje kao sesilna masa na gingivi<sup>27</sup>.

Radiografska prezentacija AOT obično pokazuje jasno definisano rasvetljenje sa kompaktnom ili sklerotičnom granicom u očnjačkoj regiji. Viđa se dislokacija susednog zuba, ali retko resorpcija korena zuba.<sup>1</sup> Od 35 slučajeva uzetih na analizu, resorpcija korena susednog zuba pronađena je u 6(17,1%) slučajeva. Uočeno je da su udruženi sa impaktiranim zubima u 65,7% slučajeva. Batra i sar<sup>2</sup> prijavili su da se impaktirani očnjaci nalaze u 59% slučajeva. Slično tome, i u ovoj studiji, pronađeno je da je među impaktiranim zubima najviše očnjaka, sa prevalencijom od 82,6%, dok je kod premolara bila 13%, a sekutići, molari i prekobrojni zubi pokazuju minimalnu prevalenciju od 4,3%. Adenomatoidni odontogeni tumor mogu sadržati i više malih fokusa različitog oblika i transparencije ili kalcifikacije koje se mogu manifestovati kao grupice peska ili kao kalcifikovane pahuljice<sup>27</sup>. Ovi kalcifikovani depoziti mogu se videti u oko 70% slučajeva<sup>40,41</sup>. Za detekciju ovakvih kalcifikata indikovana je intraoralna periapikalna radiografija. Iz Tabele 2 vidi se da se AOT prezentuje kao unilokularno rasvetljenje u 18 (51,4%) slučajeva, dok se pahuljičasto zasenčenje kalcifikovanog materijala javlja u 14 (40%) slučajeva. Samo u dva slučaja (5,6%) manifestuje se kao multilokularno rasvetljenje sa zasenčenim fokusima.

Radiografski sagledano, diferencijalna dijagnoza AOT može biti: folikularna cista, kalcifikovana odontogeni cista, kalcifikovani epitelnii odontogeni tumor, unilokularni ameloblastom, keratocistični odontogeni tumor. Lateralna periodontalna cista i keratocistični odontogeni tumor se mogu dijagnostički razmatrati u određenom broju slučajeva. Makroskopski, AOT je jasno definisana lezija koju okružuje tanka fibrozna kapsula. Mikroskopski, tumor je sastavljen od vretenastih epitelialnih ćelija koje formiraju listasate, duguljaste ili spiralne mase u oskudnoj fibroznoj stromi. Epitelijalne ćelije mogu formirati ružolike strukture u centralnom delu promene<sup>42</sup>. Krarakteristične kanalne strukture okružene su jednim slojem epitelnih ćelija, čija su jedra izmeštena iz centralnog položaja<sup>37</sup>. Mehanizam formiranja ovih tubularnih struktura može biti rezultat sekretorne aktivnosti ćelija, koje su izgleda preameloblasti<sup>38</sup>.

(extracoronal). Friedrich et al.<sup>20</sup> found that the follicular variety is by far the most frequent type of AOT. It usually appears as a pericoronal well circumscribed unilocular radiolucency or radiopaque-radiolucent mixed lesion with well-defined corticated or sclerotic border, usually surrounding an unerupted tooth while the extra-follicular variant presents as a well-defined unilocular radiolucency located between, above or superimposed on the root of a tooth<sup>6</sup>. Extraosseous AOT presents as a sessile mass on the facial gingiva<sup>27</sup>.

The usual radiographic appearance of AOT is a well-defined radiolucency with a corticated or sclerotic border at the cuspid region of maxilla. Displacement of adjacent teeth is seen but root resorption is rare<sup>1</sup>. Of the 35 cases taken up for analysis, root resorption of the adjacent teeth was seen in 6 (17.1%) cases. It was noted that there was an associated impacted tooth in 65.7% of the cases. Batra et al.<sup>2</sup> reported that impacted canines accounted for 59% of the cases. Similarly, in the present study, it was found that among the teeth that were impacted, canine showed the highest prevalence (82.6%), while premolar was associated in 13% of the cases and incisors, molars and supernumerary teeth showed minimal prevalence at 4.3%. AOTs may contain multiple minute variably shaped radiopaque foci or calcifications which may appear as a cluster of pebbles or may show fine snowflake like calcifications<sup>27</sup>. These calcified deposits are seen approximately in 78% of the cases<sup>40,41</sup>. Intraoral periapical radiograph is the indicated radiograph for the detection of calcifications. From Table 2, it is seen that AOT presented as a unilocular radiolucency in 18 (51.4%) of the cases, while radiopaque flecks of calcified material were seen in 14 (40%) of the reported cases. Only 2 (5.7%) cases presented as a multilocular radiolucency with radiopaque foci.

Generally, the radiographic differential diagnosis of an AOT includes dentigerous cyst, calcifying odontogenic cyst, calcifying epithelial odontogenic tumour, unilocular-meloblastoma and keratocystic-odontogenic tumour. In some cases, lateral periodontal cyst and keratocystic odontogenic tumour can be considered in the diagnosis. Macroscopically, AOT is a well-defined lesion surrounded by a thick fibrous capsule.

Lumen ćelije može biti prazan ili može sadržati male količine eozinofilnog materijala u vidu ameloidne mrlje. Mali kalcifikovani fokusi mogu biti rasuti duž lezije, koje se mogu interpretirati kao abortivna gledna formacija, dentinska masa ili cement<sup>7</sup>. Analiza histološkog materijala u 35 slučajeva pokazala je spiralne, listaste i pleksiformne aranžmane sa ružolikim ili vretenastim epiteljalnim ćelijama koje okružuju kanalnu arhitekturu. Amorfni eozinofilni ameloid ili hijaline strukture sa distrofičnim poljima kalcifikacije viđaju se u 25 (71,4%) slučajeva. Depoziti glednog matriksa prekrivenih slojem dentina viđaju se u 4 (11,4%) histološke sekcije.

Imunohistohemijske studije ukazuju na pojavu keratina i vimentina u tumorskim ćelijama na periferiji kanala, tubula i spiralnih struktura<sup>43</sup>. Amelogenin i enukleacija u malim tumorskim ćelijama i kapljicama hijalina nađeni su mineralizovani fokusi<sup>44</sup>.

Kako sve varijante tumora imaju kapsulu, trapiski izbor je hirurška enukleacija i kiretaža promene<sup>23</sup>. Stopa recidiva kreće se oko 0,2%<sup>32</sup>. Ako je folikul zuba prisutan, moguće je ukloniti promenu sa prezervacijom zuba, naročito kada se govori o gornjim očnjacima kod mladih pacijenata<sup>2</sup>. Preporučljivo je dugotrajno praćenje pacijenta. Naš pregled je u saglasnosti sa postojećom literaturom u vezi sa AOT, u pogledu starosti, pola, radiografske prezentacije i histopatoloških odlika. Ipak, pronašli smo da se AOT javlja najčešće u posteriornim regijama mandibule. Ovaj podatak je u suprotnosti sa postojećim podacima iz literature koji ukazuju da je uobičajeno mesto za pojavu AOT anteriorna regija maksile.

## Zaključak

1. Adenomatoidni odontogeni tumor je benigni, sporo rastući, ne agresivni odontogeni tumor. U u našem pregledu da se najčešće viđa kod žena u drugoj deceniji života, dok je u starijem dobu redak, naročito posle pedesete godine života. Najčešće se javlja u zadnjim delovima mandibule. Adenomatoidni odontogeni tumor obično se manifestuje kao unilokularno rasvetljenje koje uzrokuje resorpciju ili dislokaciju susednog zuba. Impaktirani zub, najčešće očnjak, nalazi se u najvećem broju slučajeva. Ovim radom predstavili smo trenutne literaturne činjenice, bazirane na demografskim parametrima, kliničkim, radiološkim i histološkim odlikama AOT.

Microscopically, the tumour is composed of spindle-shaped epithelial cells that form sheets, strands or whorled masses in a scant fibrous stroma. The epithelial cells may form rosette-like structures about a central space<sup>42</sup>. The characteristic duct-like structures are lined by single layer of columnar epithelial cells, the nuclei of which are directed away from the central lumen<sup>37</sup>. The mechanism of formation of these tubular structures may be a result of the secretory activity of the cells which appear to be preameloblasts<sup>38</sup>. The lumen may be empty or may contain small amounts of eosinophilic material that may stain for amyloid. Small foci of calcification may be seen scattered throughout the lesion which have been interpreted as abortive enamel formation, dentinoid material or cementum<sup>27</sup>.

Analysis of the histopathological features of the 35 cases revealed that the lesions showed whorls, sheets, and plexiform arrangement of cells with rosettes of spindle-shaped epithelial cells surrounding the ductal architecture. Amorphous eosinophilic amyloid and hyaline-like material with dystrophic areas of calcification were seen in 25 (71.4%) of the cases. Deposits of enamel matrix with the overlying dentin was seen in 4 (11.4%) of the histologic sections.

Immunohistochemistry studies of the lesions suggest the expression of keratin and vimentin in the tumour cells at the periphery of the ductal, tubular or whorled structures<sup>43</sup>. Amelogenin and enamelin in small mineralised foci are found in the tumour cells and in hyaline droplets<sup>44</sup>.

Since all variants of the tumour show encapsulation, surgical enucleation or curettage is the treatment of choice<sup>2</sup>. A recurrence rate of 0.2% has been reported<sup>32</sup>. If the follicle is found to be uninvolved during surgery, it may be possible to remove the lesion while leaving the tooth in place, especially in cases of involvement of the maxillary canine in young patients<sup>2</sup>. A long term follow-up of the patient is recommended. Our review was in accordance with the existing literature of AOT regarding the age, sex, radiographic presentation, and histopathological features. However, we found highest predilection for AOT in the mandibular posterior region. This contradicts the existing literature where the maxillary anterior region is the common site of involvement.

### ***Conclusion***

1.AOT is a benign, slow growing, non-aggressive odontogenic tumour. Our systematic review revealed that AOT was most frequently seen in females and in the second decade of life and was rare in older individuals after the fifth decade. The commonest site of occurrence was the mandibular posterior region. It was found that AOT usually presents as a unilocular radiolucency causing resorption or displacement of the adjacent roots. An impacted tooth, usually a canine was found in a majority of the cases. Thus, this review of AOT compiles the recent literature regarding the demographic parameters, clinical, radiographic, and histological features.

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