

Shelly Arora<sup>1</sup>, Priya Kumar<sup>2</sup>, Aadithya. B. Urs<sup>3</sup>, Jeyaseelan Augustine<sup>4</sup>

## Unicistični ameloblastom: kliničko-patološka analiza 22 slučaja

### Unicystic Ameloblastoma: Clinical Pathological Analysis of 22 Cases

- <sup>1</sup> Specijalizant na Odjelu za oralnu patologiju Stomatološkog instituta Maulana Azad, New Delhi, Indija  
*Senior Resident, Department of Oral Pathology, Maulana Azad Institute of Dental Sciences New Delhi, India.*
- <sup>2</sup> Asistent na Odjelu za oralnu patologiju Stomatološkog instituta Maulana Azad, New Delhi, Indija  
*Assistant Professor, Department of Oral Pathology, Maulana Azad Institute of Dental Sciences, New Delhi, India.*
- <sup>3</sup> Profesor na Odjelu za oralnu patologiju Stomatološkog instituta Maulana Azad, New Delhi, Indija  
*Professor, Department of Oral Pathology, Maulana Azad Institute of Dental Sciences, New Delhi, India.*
- <sup>4</sup> Asistent na Odjelu za oralnu patologiju Stomatološkog instituta Maulana Azad, New Delhi, Indija  
*Assistant Professor, Department of Oral Pathology, Maulana Azad Institute of Dental Sciences, New Delhi, India.*

#### Sažetak

**Svrha rada:** Uniblastični ameloblastom (UA) vrsta je solidnog ili multicističnog ameloblastoma. To je posebna, manje agresivna varijanta nižeg stupnja rekurencije. Istraživanje je obavljeno usporedbom kliničkih i patohistoloških nalaza niza dijagnoza uniblastičnih ameloblastoma iz arhiva Stomatološkog instituta Maulana Azad sa slučajevima opisanim u literaturi. **Materijali i metode:** Nakon petogodišnje analize 122 dijagnoze ameloblastoma iz arhiva Odjela za oralnu patologiju Stomatološkog instituta Maulana Azad u New Delhiju u Indiji, odabrana su 22 slučaja. Svi su procijenjeni te se bilježila dob pacijenta, spol, radiografska lokacija i podaci o novotvorini, klinička dijagnoza, histopatološka svojstva, terapija i podaci o kontrolama. **Rezultati:** Odabrana su 22 slučaja uniblastičnog ameloblastoma od ukupno 112 izdvojenih dijagnoza unatrag pet godina. Raspon dobi pacijenata bio je od 3 do 65 godina, a najčešće se bolest pojavljivala u trećem desetljeću života. Zastupljenost prema spolu bila je podjednaka. Kod većine oboljelih bila je zahvaćena mandibula (n=19), a samo kod troje maksila. Veličina lezije iznosila je od 5 x 1 centimetar do 7,4 x 2,8 centimetara. Najčešći zajednički simptom bio je bezbolna oteklina, pa se koštana egzostoza vidjela u 18 slučajeva. Jednostruka lezija pojavila se kod 20 pacijenata, a višestruka kod dvoje. Mikroskopski su svi slučajevi zadovoljavali kriterije Philipsena i Reicharta (luminal – 8, luminal i intraluminal – 4, luminal, intraluminal i intramural – 3 te luminal i intramural – 7). Nadene su i višestruke varijacije epitela obloženog oko neoplazme. Tijekom kontrola bolest se kod 16 pacijenata nije ponovno pojavila. **Zaključak:** U istraživanju su se uspoređivali klinički, radiografski i histološki podaci iz 22 dijagnoze UA. Potrebno je dodatno proučiti histogenezu i dijagnostičke nedostatke te treba li produljiti razdoblje kontrola tretiranih lezija.

**Zaprimljen:** 7. svibnja 2012.

**Prihvaćen:** 8. srpnja 2012.

#### Adresa za dopisivanje

Dr Shelly Arora. M.D.S.  
Senior Resident, Department of Oral Pathology, Maulana Azad Institute of Dental Sciences, Bahadur Shah Zafar Marg, New Delhi – 110002, India  
tel.: 91-8800168638  
arorashells76@yahoo.co.in

#### Ključne riječi

ameloblastom; dentinogena cista; donja čeljust

#### Uvod

Ameloblastom je jedan od najčešćih dobroćudnih odontogenih tumora s udjelom od jedan posto među svim novotvorinama i cistama u čeljustima i 10 posto među odontogenim neoplazmama (1,2). Na uniblastični/unicistični ameloblastom (UA) otpada od pet do 15 posto svih ameloblastoma (3). UA se odnosi na one cistične lezije koje imaju klinička i radiografska obilježja odontogene ciste, ali histološka ispitivanja pokazuju karakterističan ameloblastični epitel oko odontogene ciste s proliferacijom ili bez nje u lumen ili u stijenku (4). Obično obolijeva mlađa populacija u usporedbi s uobičajenim ameloblastomom (5, 6, 7, 8).

Potrebno je odvojiti UA od običnih (čvrstih) i perifernih ameloblastoma zbog jedinstvenih morfoloških obilježja i tipičnoga biološkog djelovanja. Histogenetski je podrijetlo

#### Introduction

Ameloblastoma is one of the most common benign odontogenic tumors accounting for approximately 1% of all tumors and cysts of the jaws and 10% of all odontogenic tumors (1,2). Unicystic ameloblastoma (UA) represents 5-15% of ameloblastoma cases (3). UA refers to those cystic lesions that show clinical and radiological characteristics of an odontogenic cyst but in histological examination show a typical ameloblastomatous epithelium lining part of the cyst cavity, with or without luminal and/or mural proliferation (4). It tends to occur in a younger population as compared to patients of conventional ameloblastoma (5-8).

The UA needs to be separated from the conventional (solid) and peripheral ameloblastomas due to its unique clinical morphological features and distinct biological behavior.

uniblastičnog ameloblastoma i dalje predmet rasprava. Mnogi istraživači pronašli su osnovu za njegov razvoj u postojećoj odontogenoj cisti, ali nastaje i kao ponovno razvijeni tumor. No ni jedna hipoteza nije dokazana. Smatra se manje agresivnim od krutoga oblika. Tumor se obično tretira enukleacijom i kiretažom (9, 10).

Ovo istraživanje provedeno je na temelju 22 slučaja kako bi se potanko analizirao klinički morfološki spektrom te bolje odredilo moguće podrijetlo novotvorine i njezino biološko ponašanje.

## Materijali i metode

Nakon analize 112 dijagnoza ameloblastoma iz arhiva, izabrana su 22 slučaja liječena od siječnja 2007. do prosinca 2011. na Odjelu za Oralnu patologiju Stomatološkog instituta Maulana Azad, u tercijarnoj specijaliziranoj bolnici i u Školi dentalne medicine u New Delhiju u Indiji.

Odabir slučajeva i uvrštavanje među UA obavljani su analizom histoloških, kliničkih i radiografskih svojstava. Za istraživanje su odabrani oni koji su zadovoljavali sljedeće kriterije:

1. primarno se radilo o intreaosealnoj lokalizaciji,
2. makroskopski i mikroskopski bile su vidljive monocistične lezije obložene epitelom građenim od cilindričnih bazalnih stanica u palisadnom redu s vakuolama u citoplazmi i hiperkromatičnim jezgrama polariziranim nasuprot bazalnoj membrani; površinski epitel sastavljen je od labavo složenih stanica nalik na stanice zvjezdastoga retikularnog tkiva (*reticulum stellatum*) (11).

Četvero neovisnih patologa određivalo je histološka svojstva koristeći se kriterijima Philipsena i Reicherta (12). Svi su slučajevi analizirani i za varijaciju oblaganja ciste. Tako je epitel ispitan na prisutnost:

1. klasičnoga odontogenog ameloblastičnog epitela,
2. nekeratiniziranog epitela nalik na dentigenu cistu,
3. hiperplastičnog epitela/ stratifikaciju.

Analizirana je i prisutnost ili odsutnost upale u stromalnom tkivu i njezin utjecaj na gornje slojeve epitela. Kod svih odabranih slučajeva procjenjivala se dob pacijenata, spol, lokalizacija, radiografski podaci, klinička dijagnoza, terapija i podaci iz kontrolnog razdoblja.

## Rezultati

### A) Epidemiološki profil

#### Incidencija

Naša studija pokazuje da UA predstavlja 14,4 posto (22 od 122) od svih ameloblastoma zabilježenih tijekom istraživačkog razdoblja.

#### Dob

Zabilježena dob pacijenata u vrijeme dijagnoze bila je od 3 do 65 godina, a približno njih 32 posto bilo je u trećem desetljeću života (tablica 1.).

#### Spol

Gotovo je ravnomjerna raspodjela među spolovima – 10 muškaraca i 12 žena (tablica 1.).

Histogenetically, the origin of UA remains a topic of considerable debate. The origin of tumor from both a preexisting odontogenic cyst and a *de novo* origin have found favor with different researchers but neither hypothesis has been conclusively proven. Generally thought to be less aggressive than its solid counterpart, the tumor is often treated by enucleation and curettage (9,10).

The present study was carried out to analyze and detail the clinical morphological spectrum, possible histogenesis and biological behavior of the tumor in 22 patients.

## Materials and methods

A total of 22 cases of UA were obtained after reviewing 112 cases of ameloblastoma from the archives of the Department of Oral Pathology, Maulana Azad Institute of Dental Sciences, a tertiary specialized dental college and hospital, India, from the period of January 2007 to December 2011. The cases of UA were reconfirmed by reviewing the histology, clinical details and radiographic features. Cases having the following features were included as UA:

1. Cases which were primarily intraosseous.
2. Cases that were macroscopically and microscopically monocystic lesions lined by epithelium composed of columnar basal cells in a palisading arrangement with vacuolated cytoplasm and hyperchromatic nuclei polarized away from the basement membrane. The superficial epithelium was composed of loosely arranged cells resembling the cells of stellate reticulum (11).

The histological features were analyzed independently by four different pathologists, using the criteria proposed by Philipsen and Reichert (12). Furthermore, all cases were analyzed for variations in cyst lining. The lining was examined for the presence of:

1. 'Classical' odontogenic ameloblastomatous epithelium.
2. Non-keratinized lining resembling dentigerous cyst.
3. Hyperplastic epithelium/ stratification.

Presence or absence of inflammation in the stromal tissue and its influence on overlying epithelium was also analyzed. All the 22 cases were further assessed for patient age, gender, location, radiographic features, clinical diagnosis, treatment and available follow-up data.

## Results

### A) Epidemiological profile

#### Incidence

Our study showed that UA variant constituted 14.4% (22 out of 112) of all ameloblastomas seen during the study period.

#### Age

The age ranged from 3-65 years at the time of diagnosis with approximately 32% of the cases presenting during the 3<sup>rd</sup> decade of life. (Table 1)

#### Gender

An almost equal gender distribution was observed in the studied series that consisted of 10 males and 12 females. (Table 1)

**Tablica 1.** Glavna tablica s kliničkim radiografskim i histopatološkim svojstvima 22 slučaja unicističnog ameloblastoma  
**Table 1** Master table showing clinical-radiographic and histopathological features of 22 cases of Unicystic Ameloblastoma

Slučaj broj • Case No.	Dob • Age	Spol • Sex	Lokacija • Location	Veličina • Size	Klinički nalazi • Clinical findings	Mjesto nastanka • Expansion	Rtg. svojstva • Radiographic features
1.	65	M	Maks. 12-16 Sinus • Max 12-16 Sinus	4.5x2.1	Oteklina+pus • Sw+Pus	Palatinalno • Pal	Uni/radiolucencija • Uni/RI
2.	62	Ž • F	Mand. 32-Mand. Ramus • Mand 32-MidRamus	5.4x2.3	Oteklina+parestezija LL • Sw+Pares of LL	Lab+buk+buk. perf. kutno • Lab+Buc+Buc Perf at ang	Multi/radiolucencija • Multi/RI
3.	35	Ž • F	Mand 43-46	3x2	Oteklina+parestezija LL • Sw, Pares of LL	Ne • No	Uni/radiolucencija • Uni/RI
4.	28	Ž • F	Mand 33-43	3.5x1.5	Bolovi • Pain	Ne • No	Uni/radiolucencija • Uni/RI
5.	27	M	Mand. R kut • Mand R Angle	2.5x1.5	Oteklina+bolovi • Sw+Pain	Bukalno • Buc	Uni/radiolucencija • Uni/RI
6.	24	Ž • F	Mand 35-38	2.5x2	Oteklina+bolovi • Sw+Pain	Bukalno • Buc	Uni/radiolucencija • Uni/RI
7.	40	M	Mand 47- kut • Mand 47- Angle	3x1.5	Oteklina+bolovi • Sw+Pain	Bukalno • Buc	Uni/radiolucencija • Uni/RI
8.	13	M	Mand. L Kut.-Ram • Mand L Ang-Ram	3x2.5	Oteklina • Sw	Bukalno • Buc	Uni/radiolucencija • Uni/RI
9.	39	M	Mand 31-36	4.5x2.5	Oteklina • Sw	Buc+Lin	Multi/radiolucencija • Multi/RI
10.	21	Ž • F	Maks. 11-14 Sinus • Max 11-14 Sinus	3.2x2	Oteklina • Sw	Ne • No	Uni/radiolucencija • Uni/RI
11.	44	M	Mand 33-36	2.7x1.5	Oteklina • Sw	Bukalno • Buc	Uni/radiolucencija • Uni/RI
12.	23	Ž • F	Mand 45-47	3.2x2.5	Oteklina • Sw	Ne • No	Uni/radiolucencija • Uni/RI
13.	13	M	Mand 47	1.5x1	Oteklina • Sw	Ne • No	Uni/radiolucencija • Uni/RI
14.	3	Ž • F	Max 61-63	2.7x2	Oteklina • Sw	Lab+Pal	Uni/radiolucencija • Uni/RI
15.	28	Ž • F	Mand. R kut • Mand R Angle	2.5x1.5	Oteklina+bolovi • Sw+Pain	Bukalno • Buc	Uni/radiolucencija • Uni/RI
16.	20	M	Mand 34-37	3.8x2.4	Oteklina+bolovi • Sw+Pain	Bukalno + lingvalno + bukalno perf. • Buc+Lin+Buc Perf	Uni/radiolucencija • Uni/RI
17.	16	M	Mand 43-36	7.4x2.8	Oteklina • Sw	Labijalno + bukalno • Lab+Buc	Uni/radiolucencija • Uni/RI
18.	50	M	Mand. 42-kut • Mand 42-Angle	5.8x3.1	Oteklina • Sw	Bukalno • Buc	Uni/radiolucencija • Uni/RI
19.	13	Ž • F	Mand 44-46	2.5x1.5	Oteklina+bolovi+pus • Sw+Pain+pus	Ne • No	Uni/radiolucencija • Uni/RI
20.	50	Ž • F	Mand 33-35	3x2.5	Oteklina • Sw	Bukalno • Buc	Uni/radiolucencija • Uni/RI
21.	43	Ž • F	Mand 34-45	6.8x3.2	Oteklina+bolovi • Sw+Pain	Labijalno + bukalno • Lab+Buc	Uni/radiolucencija • Uni/RI
22.	28	Ž • F	Mand 46-48	2.4x1.7	Oteklina • Sw	Bukalno • Buc	Uni/radiolucencija • Uni/RI

Kratice: OKC – odontogena keratocista, CGCG– središnji gigantocelularni granulom, AOT – adenomatoidni odontogeni tumor  
M–muško, F–žensko, mand –mandibula, max – maksila, Sw – oteklina, pares–parestezija, LL – donja usnica, pal – palatinalno, lab – labijalno, buc – bukalno, lin – lingvalno, perf – perforacija, ang– kut, uni–unilokular, multi– multilokular, RI –radiolucencija, RR–resorpcija korijena, Ab – nedostaje, disp – razmaknuto, imp – impaktirano, uni amelo – unicistični ameloblastom, amelo – ameloblastom, DC – dentigena cista, recurr DC–rekurentna dentigena cista, Rad cyst –radikularna cista, , sm– glatko, fri – odvojeno, var – varijabilno, nod excr – ekscizija čvorova, enuc– enukleacija, resect – resekcija, curr – kiretaža, NR–nema povratka tumora, NA– nema podataka

Zub • Tooth	Klinički profil • Clinical impression	Makroskopski nalazi • Macroscopic findings	Mikroskopski nalazi • Microscopic findings	Liječenje • Treatment	Ponovna pojava/trajanje • Follow up/duration
Ne • No	Uni Amelo	Galtko+thin+odvojeno • Sm+Thin+Fri	1	Enukleacija • Enucl	Nema povratka tumora/5,2 • NR/5.2
Resopr. Korijena 33,34,35,38; Nedostaju 36,37; Razmaknut 38 • RR33,34,35,38; Ab 36,37; Disp 38	Multi Amelo/OKC	Thin cystic cavity	1	Resekcija • Resect	Nema podataka • NA
Impakt. 44 • Imp 44	OKC/DC	Glatko+thin • Sm+thin	1	Enukleacija • Enucl	Nema podataka • NA
Ne • No	Rad cyst	Thick+ focal + eksciz. čvorova+ recd in bits • Thick+ focal + Nod Excr+ Recd in bits	1.2	Kiretaža • Curr	Nema podataka • NA
Ne • No	OKC/Rad cyst	Glatko, collapsed Cystic sac • Sm collapsed Cystic sac	1	Enukleacija • Enucl	Nema povratka tumora/4,3 • NR/4.3
Resopr. korijena 37; Impak. 38 • RR 37; Imp 38	DC	Sm+ Var (Thick+Thin)	1.3	Enukleacija • Enucl	Nema povratka tumora/3,6 • NR/3.6
Resopr. korijena 47; Impak. 48 • RR 47; Imp 48	DC/Uni Amelo	Sm +Thin+Focal Nod Excr	1.2	Enukleacija • Enucl	Nema podataka • NA
Ne • No	OKC	Sm+Thin+Fri+ Recd in bits	1	Kiretaža • Curr	Nema povratka tumora/3,3 • NR/3.3
Nedostaje 34-36 • Ab 34-36	CGCG/OKC	Sm+Thick	1.3	Resekcija • Resect	Nema povratka tumora/2,5 • NR/2.5
Impak. 13 • Imp 13	AOT	Thick Cyst wall+Nod Excr	1.2	Enukleacija • Enucl	Nema podataka • NA
Ne • No	Rad cyst	Sm+Thin+Fri	1	Enukleacija • Enucl	Nema povratka tumora/2,3 • NR/2.3
Ne • No	Rad cyst	Yellow brown Nod Excr + Var (Thick+Thin)	1.3	Enukleacija • Enucl	Nema povratka tumora/1,8 • NR/1.8
Ne • No	DC	Sm+Thin+ Recd in bits	1	Kiretaža • Curr	NR/1.2
Impak. djelomice formiran 25 • Imp partially formed 25	DC	Multiple Nod Excr+Var (Thick+Thin)	1.2.3	Kiretaža • Curr	Nema povratka tumora/1 • NR/1
Ne • No	Residual Cy	Thick Cyst wall+Nod Excr	1.2	Enukleacija • Enucl	Nema povratka tumora/0,9 • NR/0.9
Impak. 35; Razmaknut 34; Resopr. korijena 36 • Imp 35; Disp 34; RR 36	Recurr DC/Uni Amelo	Var (Thick+Thin)	1.3	Resekcija • Resec	Nema povratka tumora/0,6 • NR/0.6
Razmaknuti 43-34 • Disp 43-34	Uni Amelo	Loculated Thick cyst+ Nod Excr	1.2.3	Enukleacija • Enucl	Nema povratka tumora/0,4 • NR/0.4
Ne • No	Uni Amelo	Sm+Thick	1.3	Resekcija • Resec	Nema povratka tumora/4,5 • NR/4.5
Impak. 45 • Imp 45	DC	Thin + Sm+Recd in bits	1	Resekcija • Resec	Nema podataka • NA
Impak. 33,34,35; Razmaknut 43 • Imp 33,34,35; Disp 43	DC/Uni Amelo	Var (Thin+Thick)+ Nod Excr	1.3	Resekcija • Resec	Nema povratka tumora/0,2 • NR/0.2
Ne • No	Uni Amelo	Thick + Hemorrhagic	1.3	Kiretaža • Curr	Nema povratka tumora/0,2 • NR/0.2
Nedostaje 46 • Ab 46	Recurr DC/Uni Amelo	Var (Thin+Thick)+ Nod Excr	1.2.3	Enukleacija • Enucl	Nema povratka tumora/0,1 • NR/0.1

Abbreviations: M-Male, F-Female, Mand –Mandible, Max – Maxilla, Sw- Swelling, Pares-Paresthesia, LL- Lower Lip, Pal- Palatal, Lab– Labial, Buc – Buccal, Lin- Lingual, Perf- Perforation, Ang- Angle, Uni- Unilocular, Multi- Multilocular, RI- Radiolucency, RR- Root Resorption, Ab- Absent, Disp- Displaced, Imp- Impacted, Uni Amelo- Unicyclic ameloblastoma, Amelo- Ameloblastoma, DC- Dentigerous cyst, Recurr DC- Recurrent Dentigerous cyst, Rad cyst-radicular cyst, OKC- Odontogenic Keratocyst, CGCG- Central giant cell Granuloma, AOT- Adenomatoid odontogenic tumor, Sm- Smooth, Fri- Friable, Var- Variable, Nod Excr- Nodular Excrescences, e, Enucl- Enucleation, Resect- Resection, Curr- Curettage, NR- No Recurrence, NA- Not Available.

### Mjesto tumora

Većina novotvorina zahvaćala je mandibulu (n=19) – 15 je pronađeno u njezinu stražnjem dijelu, dvije u prednjem, a dvije su se pružale u oba područja. U gornjoj čeljusti dva su tumora zabilježena u prednjem području, a jedan je obuhvaćao i stražnje i prednje područje maksile (tablica 1.).

### Veličina

Lezije su varirale od 1,5 x 1 centimetar do 7,4 x 2,8 centimetara (tablica 1.).

## B] Klinički profil

### Simptomi nakon dolaska liječniku

Najčešći simptom bila je bezbolna otekline kod 14 od 22 pacijenta. Osam pacijenata navelo je bol, 2 su imala gnojni eksudat iz zahvaćenog područja, a 2 su navodila paresteziju donje usnice.

### Kortikalna ekspanzija

U mandibuli je 14 od 19 tumora imalo znakove ekspanzivnog rasta bukalnih kortikalnih ploča. U dva je slučaja osim bukalne rasla i lingvalna kortikalna ploča, a kod dvoje pacijenata je perforirala bukalna kortikalna ploča. Među maksilarnim tumorima jedan je imao samo palatinalni rast, a jedan je rastao s obje strane – i palatinalne i bukalne (tablica 1.).

### Radna dijagnoza

Za 20 pacijenata radne su dijagnoze glasile *specifične ciste čeljusti* (u 9 slučajeva – dentinogena cista; u 4 slučaja – radikularna cista; u 3 slučaja – odontogena keratocista; i u jednom slučaju – rezidualna), za njih osam pretpostavljalo se da je riječ o unicističnom ameloblastomu, za dvoje o multicističnom, za jednoga o adenomatoidnom odontogenom tumoru, te, također za jednoga, o središnjem gigantocelularnom granulomu. U većini slučajeva bilo je predloženo više radnih dijagnoza, a ne samo jedna (tablica 1.).

## C] Radiogramski profil

Svi su pacijenti imali predoperativne radiograme. Lezije su bile dobro ograničene radiolucencije. Unilokularna radiolucencija bila je prisutna u 20 slučajeva, a multilokularna u dva (tablica 1). U devet slučajeva bio je uključen i impaktirani zub. Jedan mandibularni UA imao je uključena tri takva zuba. Resorpcija korijena uočena je u četiri slučaja, pomicanje korijena u tri, a nedostajući zubi bili su povezani s tri slučaja (slika 1.).

## D] Patološki izgled

### Makroskopski

Većina uzoraka bila je u komadićima, nekoliko ih je dobiveno u jednom komadu, a ostali su bili zajedno s dijelom resecirane čeljusti. Većina lezija bila je izgledom monocistična (posebice intaktni uzorci UA) s promjenjivom debljinom stijenke. Intraluminalni nastavci opaženi su u nekoliko slučajeva (n=8) – bilo fokalni ili ravnomjerno raspoređeni prema lumenu. U dobivenim uzorcima u sedam je slučajeva bio uključen jedan zub ili više njih (tablica 1.).

### Site

Majority of the cases involved the mandible (n=19). Of these, 15 were seen in the posterior mandible, 2 in the anterior mandible and 2 were seen extending from anterior to posterior mandibular region. In the maxilla, 2 cases were seen in the anterior region involving the sinus and 1 involved both anterior and posterior maxilla. (Table 1)

### Size

The lesions ranged from 1.5x1cm to a maximum of 7.4x2.8cm. (Table 1)

## B] Clinical profile-

### Presenting Symptoms

A painless swelling was the most common presenting symptom experienced by 14 of 22 patients. 8 patients complained of pain, 2 had purulent discharge from the affected area and 2 complained of paresthesia of the lower lip.

### Cortical Expansion

14 out of 19 mandibular cases showed expansion of buccal cortical plates. 2 cases showed lingual expansion along with buccal and 2 had a perforated buccal cortical plate. Amongst the maxillary cases, one showed both palatal and labial expansion, whereas one case showed only palatal expansion. (Table 1)

### Provisional Diagnosis

In 20 patients, provisional diagnosis of specified jaw cyst (9 cases- dentigerous cyst, 4 cases- radicular cyst, 3 cases- odontogenic keratocyst and 1 case-residual cyst) was suggested, whereas 8 and 2 patients were provisionally diagnosed as unicystic and multicystic ameloblastoma respectively, 1 with adenomatoid odontogenic tumor and 1 with central giant cell granuloma. More than one presumptive diagnosis had been suggested in many cases. (Table 1)

## C] Radiographic profile

Radiographs were available for all 22 patients. All the lesions were well defined radiolucencies. Unilocular radiolucency was encountered in 20 and multilocular radiolucency in 2 cases (Table 1). An impacted tooth was found associated with 9 cases. One mandibular UA was found associated with 3 impacted teeth. Root resorption was noted in 4 cases, displaced roots in 3 cases and missing teeth were associated with 3 cases (Fig. 1).

## D] Pathologic Appearance

### Macroscopic

Many samples were received in bits, few were enucleated in to and others were a part of resected jaw. Most of the cases were monocystic in appearance (especially intact specimens of UA) having variable wall thickness. Intraluminal excrescences either focal or projecting uniformly into the lumen were observed in few cases (n=8). One or more impacted teeth were found embedded in the received sample in 7 cases (Table 1).



### Mikroskopski

Svi slučajevi klasificirani su prema kriterijima Philipsena i Reicharta (12). Tako su određena četiri podtipa UA i zabilježena su sljedeća zapažanja:

### Microscopic

All the 22 cases were classified using the criteria proposed by Philipsen and Reichart (12). Based on this, four histological subtypes of UA were recognized and the following observation was made:

Histološki podtip	Broj slučajeva	Histological subtype	Number of cases
luminalni	8	Luminal	8
luminalni i intraluminalni	4	Luminal and intraluminal	4
luminalni, intraluminalni i intramuralni	3	Luminal, intraluminal and intramural	3
luminalni i intramuralni	7	Luminal and intramural	7

Svi su slučajevi nakon toga analizirani kako bi se doznale varijacije epitela obloženog oko tumora. U 12 slučajeva epitel je imao karakteristična ameloblastomna svojstva s bazalno palisadno poredanim cilindričnim stanicama, vakuoliziranim citoplazmama i hiperkromatičnim jezgrama polariziranim suprotno od bazalne membrane. U površinskom sloju bile su nepravilno raspoređene stanice nalik na zvjezdoliki retikulum (slika 2A). Oblaganje je bilo neprekinuto, s pločastim nekeratiniziranim epitelom povremeno sličnim epitelu obloženom oko dentinogene ciste (slika 3A). Takva varijacija obložnog epitela zabilježena je uglavnom kod subgrupa luminarnog i intraluminalnog tipa (n=12). Sve deblji epitel opažen je u sedam slučajeva i većina je pripadala intramuralnom tipu (slika 3B). U pet slučajeva bio je u rasponu od nekeratiniziranoga do klasičnoga ameloblastičnog zadebljanog epitela koji oponaša stratifikaciju. Ti se slučajevi ubrajaju u podskupine 1.2.3 (n=1) i 1.3 (n=4). U svim slučajevima sa stratificiranim epitelom (zadebljanje epitela) nastale su stanične reakcije u cističnoj stijenci. Jukstaepitelijalna hijelinizacija pojavila se u jednom slučaju.

All cases were further assessed to analyze variations in the lining epithelium. 12 cases showed the characteristic ameloblastomatous lining composed of columnar basal cells in a palisading pattern with vacuolated cytoplasm and hyperchromatic nuclei polarized away from the basement membrane. The superficial layer showed loosely arranged cells resembling the cells of stellate reticulum (Fig 2A). This lining was continuous with flattened non-keratinized epithelium resembling the lining of dentigerous cyst at areas (Fig 3A). This variation in the lining was observed mainly in the subgroups of luminal and intraluminal type (n=12). An increase in the thickness of lining epithelium was observed in 7 cases, most of which were associated with intramural type (Fig 3B). In 5 cases, lining epithelium ranged from non-keratinized epithelium to classical ameloblastomatous lining to thickened epithelium mimicking stratification. These cases belonged to subgroup 1.2.3 (n=1) and subgroup 1.3 (n=4). All cases showing epithelium stratification (thickened epithelial lining) were associated with inflammatory cell reaction in the cyst wall. Juxtaepithelial hyalinization was observed in 1 case.

### E] Terapija i kontrole

U ovo istraživanje bilo je uključeno 11 pacijenata kod kojih je obavljeno kirurško ljuštenje – kod njih šestero učinjena je rubna i segmentalna resekcija čeljusti, a pet je bilo kiretirano. Dva slučaja s resekcijom/izrezana slučaja (slučajevi #16 i 22) primarno su dijagnosticirana kao dentogene ciste. Kod tih pacijenata bolest se ponovno pojavila nakon 2,3 i 1,2 godine te je konačna dijagnoza glasila – UA. Podaci tijekom kontrola bili su na raspolaganju za 16 slučajeva, a za šest pacijenata nije ih bilo. U 16 slučajeva s podacima o kontrolama, duljina kontrolnog razdoblja bila je od jednog mjeseca do pet godina. Od pacijenata s podacima o kontrolnim pregledima do danas se ni kod jednoga nije ponovno pojavio UA (tablica 1). Kontrolni pregledi nastavljaju se i tijekom pisanja ovog članka.

### E] Treatment and follow up

11 patients included in the studied series were treated by surgical enucleation. Six patients were treated with marginal or segmental resection of the jaw and five were curetted. 2 excised cases (case no. 16, 22) were diagnosed as dentigerous cyst. These cases recurred after a period of 2.3 and 1.2 years, respectively, post treatment and were subsequently diagnosed as UA. The follow-up data were available for 16 cases and 6 patients were lost to follow-up. For these 16 cases, the length of the postoperative observation ranged from one month to 5 years and none of these patients have manifested recurrence till date (Table 1). A continuing follow-up is being pursued as an ongoing basis at the time of submission of this manuscript.

### Rasprava

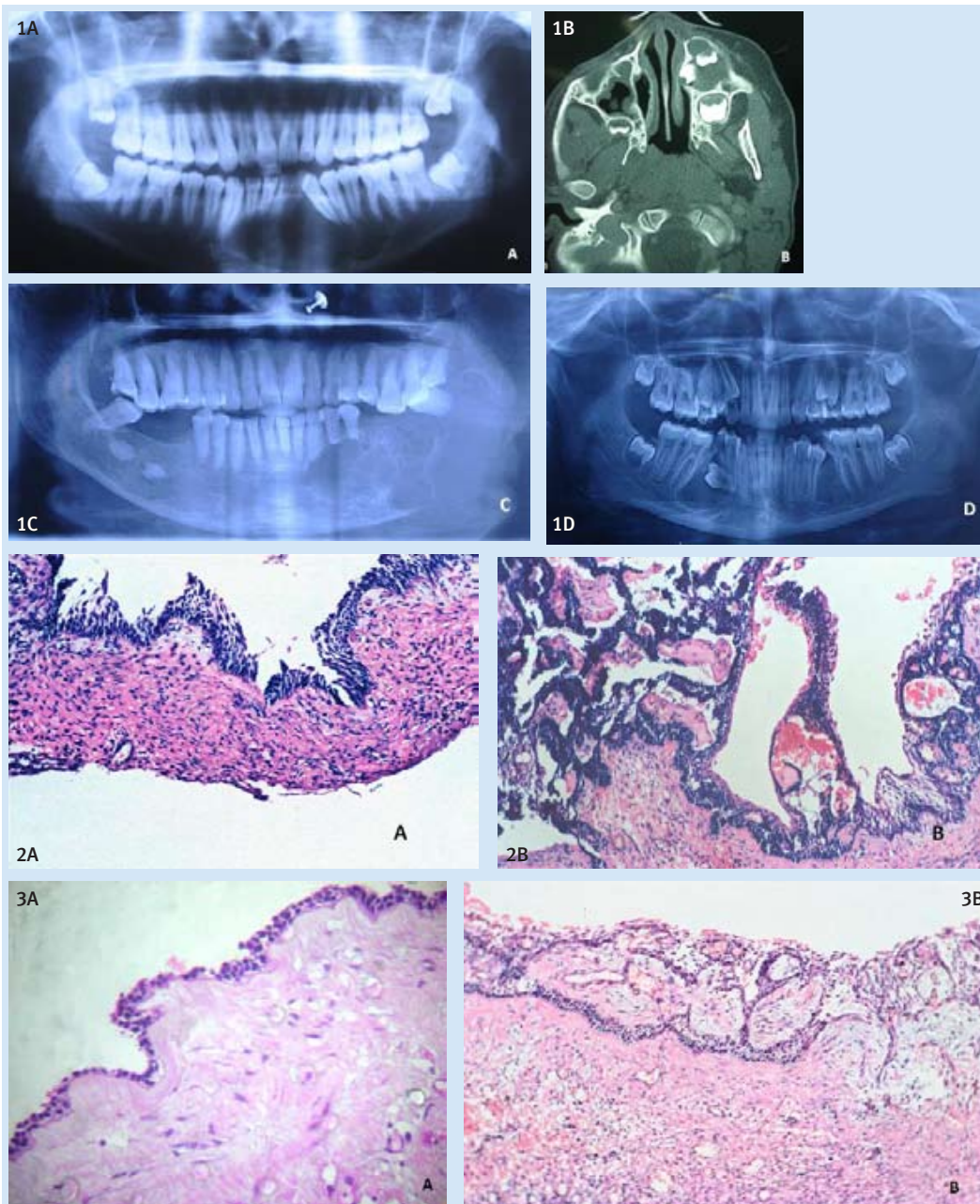
U odontogene tumore ubrajaju se skupine lezija s različitim histopatološkim tipovima i kliničkim tijekom (12). Ameloblastom je najčešća klinički važna novotvorina. Njezina relativna učestalost jednaka je ukupnoj učestalosti svih ostalih odontogenih neoplazmi.

UA predstavlja pet do 15 posto svih intrakoštanih ameloblastoma (3). Prvi su ga opisali Robinson i Martinez još 1977. godine (13). Do tada je bio poznat pod različitim nazivima kao cistični ameloblastom, ameloblastom povezan s dentige-

### Discussion

Odontogenic tumors comprise a complex group of lesions of diverse histopathological types and clinical behavior (12). The ameloblastoma is the most common clinically significant odontogenic tumor. Its relative frequency equals the combined frequency of all other odontogenic tumors.

UA represents 5-15% of all intraosseous ameloblastomas (3). This variant of ameloblastoma was first described by Robinson and Martinez in 1977 (13). Prior to 1977, it was known under various names such as cystic ameloblastoma,



**Slika 1A** Velika multilokularna radiolucencija u području prednjeg dijela mandibule s pomicanjem zahvaćenih zuba (slučaj#17)

**Figure 1A** Large multilocular radiolucency involving anterior mandible with displacement of involved teeth. (Case#17)

**Slika 1B** CECT pokazuje kod trogodišnje djevojčice ekspanzivnu dobro ograničenu cističnu leziju u koju je uključen djelomice formiran lijevi maksilarni pretkutnjak (slučaj#14)

**Figure 1B** CECT showing expansile well corticated cystic lesion involving partially formed maxillary left premolars in a 3 year old girl (Case#14)

**Slika 1C** Ortopantogram pokazuje opsežnu multilokularnu radiolucenciju koja je uzrokovala resorpciju više zubnih korijena (slučaj #2)

**Figure 1C** Orthopantogram showing extensive multilocular radiolucency causing root resorption of multiple teeth. (Case#2)

**Slika 1D** Dobro ograničena unilokularna radiolucencija s impaktiranim zubom 45 – lezija se najprije smatrala dentigenom cistom te je izluštena (slučaj # 19)

**Figure 1D** Well defined unilocular radiolucency with impacted 45. The lesion was presumed to be a dentigerous cyst and enucleated. (Case # 19)

**Slika 2A** Fotomikrografija s prikazom luminalnog UA s visokim bazalnim cilindričnim stanicama poput ameloblasta i površinskim stanicama poput zvjezdolikog retikuluma (slučaj #2) (H&E X 100)

**Figure 2A** Photomicrograph showing Luminal UA with tall columnar ameloblast like basal cells and stellate reticulum like superficial cells. (Case#2) (H&E X 100)

**Slika 2B** Intraluminalni UA s nepravilnim proliferacijama u cističnu šupljinu (slučaj # 4) (H&E X 100)

**Figure 2B** Intraluminal UA displaying plexiform pattern of proliferation into the cystic lumen. (Case# 4) (H&E X 100)

**Slika 3A** Fotomikrografija s prikazom nekeratiniziranog obložnog epitela sličnog dentigenoj cisti (slučaj # 13) (H&E X 100)

**Figure 3A** Photomicrograph showing non-keratinised epithelial lining resembling dentigerous cyst. (Case# 13) (H&E X 100)

**Slika 3B** Luminalni UA sa značajkama hiperplastičnog epitela sa sličnom slojevitom strukturom kao kod radikularne ciste (slučaj # 1) (H&E X 100)

**Figure 3B** Luminal UA showing hyperplastic epithelium reminiscent of epithelial arcading seen in radicular cyst. (Case# 1) (H&E X 100)

nom cistom, cistogenetski ameloblastom, opsežna dentigena cista s intracističnim ameloblastičnim papilama, muralni ameloblastom, dentigena cista s ameloblastnom proliferacijom i razvojni ameloblastom iz rezidualne ciste (12).

Običan ameloblastom pojavljuje se u svim dobnim skupinama, a najčešće u trećem i četvrtom desetljeću života (14). Suprotno tomu UA ima tendenciju pojavljivati se u mlađoj populaciji s prosjekom od 22,1 godine (15). U našem istraživanju zabilježen je kod pacijenata u dobi od 3 do 65 godina (srednja vrijednost 29,9 godina). Vrhunac incidencije bio je u trećem desetljeću. Vrlo je malo slučajeva te bolesti opisano u prvih deset godina života. Temeljito pretraživanje literature pokazalo je da je naš trogodišnji pacijent bio najmlađi opisani slučaj s UA-om u britanskoj literaturi.

Odnos muških i ženskih pacijenata (10 muškaraca i 12 žena) koji smo mi pronašli u našem istraživanju slaže se s rezultatima ostalih studija (9, 11, 12).

Naši nalazi prema kojima je mandibularni ramus najčešće mjesto, a bezbolna oteklina najčešći klinički simptom UA, isti je kao u opisima ostalih stručnjaka (4, 10, 13). Osam se pacijenata žalilo na bolnost, a dva na gnoj iz pogođenih područja. Bol je najvjerojatnije bila posljedica superinfekcije. Parestezija donje usne nastala je u dvama slučajevima i rezultat je rasta tumora i pritiska na mandibularni živac (n. alveolaris inf.).

Radiografski je za UA rečeno da se ponajprije pojavljuje kao unilokularna radiolucencija, ali ponekad može izgledati i multilokularno (12). Kod više od 50 posto pacijenata bila je riječ i o impaktiranom zubu te je novotvorina obično nastala oko njegove krune, slično kao dentigena cista (16). U ovom istraživanju je u 20 slučajeva imala izgled unilokularne radiolucencije i u dva slučaja multilokularne (tablica 1.). Radiolucencija je uglavnom bila na perikoronarnom mjestu.

Temeljito proučavanje dobivenih velikih uzoraka dalo je vrlo važne podatke o mikroskopskoj prirodi tumora. Slučajevi kod kojih varira debljina cistične stijenke te postoje područja bez praznog sadržaja i intraluminalni produžeci koji nisu žuti, moraju biti sumnjivi i upućuju na odontogene tumore prije negoli na ciste. U našem je istraživanju jednostavna cistična šupljina bila povezana s tipom I UA (luminalnim). Prisutnost nodularnih produžetaka odgovarala je intraluminalnom tipu, dok su debeli obložni epitel i puna područja bez šupljina histološki korelirala s UA-om koji se protezao u cističnu stijenku. Histološka svojstva UA opisali su Ackerman (11), te Philipsen i Reichert (12). Svi su ga klasificirali u različite podgrupe (kao što je dolje navedeno), ovisno o proliferaciji ameloblastičnog epitela u odnosu na cističnu stijenku.

Ackerman(11) je klasificirao UA na sljedeći način:  
 tip 1 – unilokularni, unicistična lezija obložena epitelom  
 tip 2 – pleksiformna raznolikost  
 tip 3 – povremene oteklina ameloblastomnog epitela  
 tip 3a – oteklina odvojene od cistične stijenke  
 tip 3b – oteklina spojene s cističnom stijenkom

Histopatološka podjela prema Philipsenu i Reichartu (12):

podgrupa 1 – luminalni UA

podgrupa 1.2 – luminalni i intraluminalni UA

ameloblastoma associated with dentigerous cyst, cystogenic ameloblastoma, extensive dentigerous cyst with intracystic ameloblastoma papilloma, mural ameloblastoma, dentigerous cyst with ameloblastomatous proliferation and ameloblastoma developing in a residual cyst (12).

Conventional ameloblastoma occurs in all age groups with peak incidence in the third and fourth decade of life (14). However, UA tends to occur in a younger population with average age being 22.1 years (15). In the present study, UA was observed in patients falling under a wide range i.e. from 3-65 years with a mean of 29.9 years. Peak incidence was noted in the third decade. Very few cases of UA have been reported in the first decade. A thorough search of available literature revealed that our 3-year-old patient with UA is probably the youngest reported patient of UA in the English literature.

The male to female ratio (10 males and 12 females) in our reviewed cases was found to be in agreement with other authors (9,11,12).

Our findings of mandibular ramus as the favored site and a painless swelling being the most common clinical presentation of UA, agree with that of previous reports (4,10,13). Eight patients complained of pain and two of purulent discharge from the affected area. Pain was most likely a consequence of superimposed infection. Lower lip paresthesia observed in 2 cases may result from the enlarging tumor impinging on the mandibular nerve.

Radiographically, UA is said to present primarily as a unilocular radiolucency but may occasionally exhibit multilocular appearance (12). More than half of the cases are associated with impacted teeth and the tumor is usually seen surrounding the crown of the impacted tooth similar to a dentigerous cyst (16). In the present series, 20 were unilocular radiolucency and 2 were multilocular radiolucency (Table 1). The radiolucency was in pericoronal position in almost all cases.

A thorough examination of the received gross specimen may yield vital information regarding the true microscopic nature of the tumor. Areas of varying thickness in cyst wall, solid areas, intraluminal excrescences that are not yellow must be treated with suspicion as they may prove to be odontogenic tumors rather than cysts. In the present review, simple thin cystic sacs were usually associated with Type 1 (luminal) UA. Presence of nodular excrescences corresponded with intraluminal type, whereas thick linings and solid areas correlated mostly histologically with UA extending into cyst wall.

The histological features of UA have been established by Ackerman (11), Philipsen and Reichert (12). All of them have classified UA into various subgroups (as listed below) depending on the proliferation of the ameloblastic lining with respect to cyst wall.

Ackerman has classified UA as (11):

Type 1 – unilocular, unicystic lesion lined by epithelium

Type 2 – plexiform variety

Type 3 – invariant islands of ameloblastomatous epithelium

Type 3a – islands not connected to cyst lining

Type 3b – islands connected to cyst lining



podgrupa 1.2.3 – luminalni, intraluminalni i intramuralni UA

Podgrupa 1.3 – luminalni i intramuralni UA

U ovom smo istraživanju za klasifikaciju primijenili Philipsenovu i Reichartovu (12) podjelu (tablica 1). Većina naših slučajeva ubraja se u podgrupe 1 (n=8) i 1.3 (n=7). Analizirana je i morfološka raznolikost obložnog epitela te je u svim slučajevima uočeno ameloblastično tkivo (slika 2A). Bilo je i dijagnostičkih dvojbi, posebice ako je hiperplastični epitel urastao u okolno vezivno tkivo i bio povezan s kroničnom staničnom upalnom reakcijom. Takvi slučajevi teško su se razlikovali od radikularne ciste (slika 3B). Slojevita proliferacija UA podsjeća na uzorak radikularne ciste, kao odgovor na upalu. Upale u slučaju UA i raspoloživost samo dijela ciste tijekom incizijske biopsije, dodatno kompliciraju i otežavaju dijagnostiku. Sličan dijagnostički problem uočili su i opisali Li i njegovi suradnici (4). Isti autori raspravljali su o jukstaglomerularnoj hijalinizaciji kod polovice od 33 opisana slučaja UA i predložili da taj nalaz upozorava patologe na UA u malim biopsijskim uzorcima (4). Mora se ipak istaknuti da je samo jedan od 22 slučaja UA imao značajke jukstaglomerularne hijalinizacije i da u našem istraživanju nije potvrđena važnost ovog nalaza u postavljanju dijagnoze.

Patogeneza UA ostaje kontroverzna. Neki istraživači tvrde da tumor nastaje iz odontogene ciste, a drugi smatraju da se od početka samostalno razvija (9, 10, 11, 17). Prvo je stajalište potkrijepljeno činjenicom da se UA često nalazi u bliskom kontaktu s impaktiranim zubom, a određena područja novotvorine podsjećaju također na oblaganje dentogene ciste. U našem istraživanju devet slučajeva bilo je u bliskom doticaju s impaktiranim zubom, a 12 s tankim nekeratiniziranim epitelom koji se nalazi kod dentogenih cista. Nadalje, u dva slučaja bile su nakon biopsije dokazane i izljučene dentogene ciste, ali se su lezije ponovno pojavile te su naknadno dijagnosticirane kao UA. U 13 slučajeva UA nije bilo povezanosti s impaktiranim zubima te se njihov razvoj iz postojeće dentogene ciste zbog toga ne može potvrditi, pa to podupire hipotezu o izvornom nastanku bolesti. Diferencijalnodijagnostički pokušalo se za dokazivanje je li riječ o odontogenoj cisti ili o UA-u uporabiti različite biokemijske biljege poput lecitina, ugljikohidrata krvnih stanica, receptora epidermalnog faktora rasta (EGF-R), antigena stanične proliferacije i Ki-67, ali do danas nema konačnih zaključaka (18–22).

Pitanje treba li se u slučaju sumnje na UA uopće obavljati incizijska biopsija, ostaje otvoreno za raspravu. Autori koji su protiv potkrepljuju svoje stajalište time da se tijekom incizijske biopsije dobiju samo mali dijelovi lezije, pa je čak i iskusnom patologu teško procijeniti kakva je (4).

Pravu prirodu lezije moguće je procijeniti samo ako se ekscidira kompletno tkivo i histopatološki analizira. Smatramo da je za dijagnozu UA potrebna točna kirurška, radiološka i histopatološka korelacija.

Plan terapije ovisi o dobi pacijenta, veličini tumora, lokalizaciji, radiografskim značajkama (unilokularni ili multilokularni), konačnom histopatološkom nalazu i činjenici je li lezija primarna ili rekurentna. U slučaju luminarnog i intraluminarnog UA najprihvatljivije su terapije enukleacija (iz-

Histopathological grouping by Philipsen and Reichart is as follows (12):

Subgroup 1 – Luminal UA

Subgroup 1.2 – Luminal & intraluminal UA

Subgroup 1.2.3 – Luminal, intraluminal & intramural UA

Subgroup 1.3 – Luminal & intramural UA

The present study adopted Philipsen and Reichart (12) criteria to classify the cases (Table 1).

Most of our cases fell into subgroups 1 and 1.3 (n=8 and n=7 respectively). Furthermore, the morphological diversity in the lining epithelium was also analyzed. The characteristic 'classical' ameloblastomatous lining was seen in all cases (Fig 2A). A diagnostic dilemma was encountered especially in those cases where hyperplastic epithelium was growing into the underlying connective tissue and was associated with chronic inflammatory cell reaction. Such cases became difficult to differentiate from radicular cyst (Fig 3B). Arcading proliferation of UA in particular is reminiscent of the arcading pattern seen in radicular cysts in response to inflammation. The presence of inflammation in UA and the availability of only a part of the cyst during an incisional biopsy further complicate the issue. A similar diagnostic pitfall has been discussed by Li et al. (4).

The same authors have described the presence of juxta-epithelial hyalinization in half of their reported 33 cases of UA and suggest that this finding should alert the pathologist to the finding of UA in small biopsy samples (4). However, in the present case series, only one case out of 22 displayed juxta-epithelial hyalinization and the importance of this finding in making a diagnosis of UA remains unsubstantiated.

The pathogenesis of UA remains controversial. Some researchers believe that the tumor arises from pre-existing odontogenic cyst while others consider that it arises *de novo* (9-11,17). The former argument is primarily based on the fact that UA are very often associated with impacted teeth. Also, areas of tumor resemble dentigerous cyst lining. In our study, 9 cases were associated with an impacted tooth and 12 were associated with thin non-keratinized epithelium as seen in dentigerous cyst. Moreover, 2 cases were biopsy proven cases of dentigerous cyst that had been enucleated. These lesions recurred and were subsequently diagnosed as UA. However, 13 cases were not tooth associated; hence their development from a pre-existing dentigerous cyst cannot be substantiated, thus supporting the *de novo* origin hypothesis. Various immunocytochemical markers such as lectins, blood cell carbohydrates, epidermal growth factor receptor (EGF-R), proliferating cell nuclear antigen and Ki-67 have been tried to differentiate between odontogenic cyst and UA but no consistent findings have been obtained till date (18-22).

Whether an incisional biopsy for a suspected UA should be done or not is a subject open to debate. Authors who are not in favor argue that an incisional biopsy consists of only small fragments and true nature of the lesion is difficult to assess even by trained oral pathologists (4). Moreover, the true nature of the lesion becomes evident only when the completely excised tissue is available for histopathological examination. The current authors suggest that diagnosis of UA

ljuštenje) i kiretaža (3, 10, 13). Ako novotvorina urasta u stijenke ciste (muralni oblik), tada je tehnički riječ o solidnom (čvrstom) tipu te se pri odabiru terapije (3) preporučuje radikalni kirurški pristup. Istog su mišljenja Black i suradnici (3), te Kessler (23) i Gardner (24).

Kod većine naših pacijenata (n=11) obavljeno je izljuštenje, većinom zbog njihove mladosti. U našem istraživanju ni u jednom od 16 slučajeva za koje postoje podaci nakon kontrole, nije bilo rekurencije, iako je primijenjena konzervativna kirurška terapija. Prosječni interval između terapije i rekurencije UA približno je sedam godina, a u gotovo svim opisanim slučajevima tumor se ponovno pojavio nakon četiri godine (4). Budući da ponovni UA nastaje kasno u odnosu na obični ameloblastom (rekurencija 50 % unutar 5 godina od terapije), potrebno je produljiti kontrolna razdoblja i povremeno kontrolirati sve oboljele od te podgrupe ameloblastoma (25).

## Sukob interesa

Tijekom istraživanja i pisanja ovoga rada nije bilo sukoba interesa.

requires strict surgical, radiological and histopathological correlation.

Treatment planning depends on the patient age, tumor size, location, radiographic appearance (unilocular or multilocular), final histopathological diagnosis and assessment of whether it is an initial presentation or a recurrence. However, for luminal and intraluminal UA enucleation and curettage is the most acceptable treatment modality (3,10,13). If tumor invades into the wall of the cyst (mural variant), then it technically has a solid component which outweighs the cystic component regarding treatment planning (3). In such cases, we recommend a radical surgical approach and the same has been stated by Black et al (3), Kessler HP (23) and Gardner (24). Most of our cases (n =11) were treated by enucleation according to the patients' age.

In the present study, none of the 16 cases for whom the follow-up was available showed recurrence despite being treated by conservative surgical procedures. The average interval between treatment and recurrence of UA is approximately 7 years and almost all reported recurrences occurred after a period of 4 years (4). Since UA is reported to recur late compared to conventional ameloblastoma (recurrence of 50% within a 5 year period), the need for further, prolonged and periodic evaluation of all cases is mandatory for this subgroup of ameloblastoma (25).

## Conflict of interest

No conflict of interest.

### Abstract

**Objective:** Unicystic ameloblastoma (UA) is a variant of solid or multicystic ameloblastoma. It is characterized as a distinct variant exhibiting less aggressive behavior and a lower rate of recurrence. The current study was carried out to correlate the clinical pathological findings of the studied series of UA with the other reviewed cases in the literature. **Materials and Methods:** A total of 22 cases of UA were obtained after reviewing 112 cases of ameloblastoma from the archives of Department of Oral Pathology, Maulana Azad Institute of Dental Sciences, India over a five year period. All the cases were assessed for patient age, gender, location, radiographic features, clinical diagnosis, histopathological features, treatment and available follow-up data. **Result:** The series represents 22 cases of UA out of 112 cases of ameloblastoma seen during a 5 year period. The age at diagnosis ranged from 3-65 years and peaked during the 3<sup>rd</sup> decade of life with an almost equal gender predilection. The majority of the cases involved the mandible (n=19) and only 3 were seen in the maxilla. The size of the lesion ranged from 1.5x1cm to 7.4x2.8cm. A painless swelling was the most common presenting symptom and bony expansion was evident in 18 cases. Unilocular radiolucency was encountered in 20 and multilocular in 2 cases. Microscopically, all the 22 cases satisfied the criteria proposed by Philipsen and Reichart (Luminal-8, Luminal and intraluminal-4, Luminal, intraluminal and intramural- 3 and Luminal and intramural-7). Numerous variations in the lining epithelium were observed. The follow-up data were available for 16 patients and revealed no recurrence. **Conclusion:** The study has collected clinical, radiographic and histological data for 22 cases of UA. The histogenesis, diagnostic pitfalls as well as the need for prolonged follow-up of treated lesions have been emphasized.

**Received:** May 14, 2012

**Accepted:** July 23, 2012

### Address for correspondence

Dr Shelly Arora, M.D.S.  
Senior Resident, Department of Oral Pathology, Maulana Azad Institute of Dental Sciences, Bahadur Shah Zafar Marg, New Delhi - 110002, India.  
Phone: 91-8800168638  
arorashells76@yahoo.co.in

### Key words

Ameloblastoma; Dentigerous Cyst; Mandible

## References

1. Olaitan AA, Adekeye EO. Clinical features and management of ameloblastoma of the mandible in children and adolescents. *Br J Oral Maxillofac Surg.* 1996 Jun;34(3):248-51.
2. Rapidis AD, Andressakis DD, Stavrianos SD, Faratzis G, Arngianaki-Liappi N, Lagogiannis GA et al. Ameloblastomas of the jaws: clinico-pathological review of 11 patients. *Eur J Surg Oncol.* 2004 Nov;30(9):998-1002.
3. Black CC, Addante RR, Mohila CA. Intraosseous ameloblastoma. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod.* 2010 Nov;110(5):585-92.
4. Li TJ, Wu YT, Yu SF, Yu GY. Unicystic ameloblastoma: a clinicopathologic study of 33 Chinese patients. *Am J Surg Pathol.* 2000 Oct;24(10):1385-92.
5. Kessler HP, Schwartz-Dabney C, Ellis E 3rd. Recurrent left mandibular enlargement. *J Contemp Dent Pract.* 2003 Nov 15;4(4):127-37.
6. Ord RA, Blanchaert RH Jr, Nikitakis NG, Sauk JJ. Ameloblastoma in children. *J Oral Maxillofac Surg.* 2002 Jul;60(7):762-71.
7. Gardner DG. A pathologist's approach to the treatment of ameloblastoma. *J Oral Maxillofac Surg.* 1984 Mar;42(3):161-6.
8. Feinberg SE, Steinberg B. Surgical management of ameloblas-

- toma. Current status of the literature. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod.* 1996 Apr;81(4):383-8.
9. Gardner DG, Corio RL. Plexiform unicystic ameloblastoma. A variant of ameloblastoma with a low-recurrence rate after enucleation. *Cancer.* 1984 Apr 15;53(8):1730-5.
  10. Leider AS, Eversole LR, Barkin ME. Cystic ameloblastoma. A clinicopathologic analysis. *Oral Surg Oral Med Oral Pathol.* 1985 Dec;60(6):624-30.
  11. Ackermann GL, Altini M, Shear M. The unicystic ameloblastoma: a clinicopathological study of 57 cases. *J Oral Pathol.* 1988 Nov;17(9-10):541-6.
  12. Philipsen HP, Reichart PA. Unicystic ameloblastoma. A review of 193 cases from the literature. *Oral Oncol.* 1998 Sep;34(5):317-25.
  13. Robinson L, Martinez MG. Unicystic ameloblastoma: a prognostically distinct entity. *Cancer.* 1977 Nov;40(5):2278-85.
  14. Takahashi K, Miyauchi K, Sato K. Treatment of ameloblastoma in children. *Br J Oral Maxillofac Surg.* 1998 Dec;36(6):453-6.
  15. Reichart PA, Philipsen HP, Sonner S. Ameloblastoma: biological profile of 3677 cases. *Eur J Cancer B Oral Oncol.* 1995 Mar;31B(2):86-99.
  16. Ueno S, Nakamura S, Mushimoto K, Shirasu R. A clinicopathologic study of ameloblastoma. *J Oral Maxillofac Surg.* 1986 May;44(5):361-5.
  17. Gardner DG. Plexiform unicystic ameloblastoma: a diagnostic problem in dentigerous cysts. *Cancer.* 1981 Mar 15;47(6):1358-63.
  18. Saku T, Shibata Y, Koyama Z, Cheng J, Okabe H, Yeh Y. Lectin histochemistry of cystic jaw lesions: an aid for differential diagnosis between cystic ameloblastoma and odontogenic cysts. *J Oral Pathol Med.* 1991 Mar;20(3):108-13.
  19. Gardner DG, O'Neill PA. Inability to distinguish ameloblastomas from odontogenic cysts based on expression of blood cell carbohydrates. *Oral Surg Oral Med Oral Pathol.* 1988 Oct;66(4):480-2.
  20. Li TJ, Browne RM, Matthews JB. Expression of epidermal growth factor receptors by odontogenic jaw cysts. *Virchows Arch A Pathol Anat Histopathol.* 1993;423(2):137-44.
  21. Li TJ, Browne RM, Matthews JB. Expression of proliferating cell nuclear antigen (PCNA) and Ki-67 in unicystic ameloblastoma. *Histopathology.* 1995 Mar;26(3):219-28.
  22. Li TJ, Browne RM, Matthews JB. Epithelial cell proliferation in odontogenic keratocysts: a comparative immunocytochemical study of Ki67 in simple, recurrent and basal cell naevus syndrome (BCNS)-associated lesions. *J Oral Pathol Med.* 1995 May;24(5):221-6.
  23. Kessler HP. Intraosseous ameloblastoma. *Oral Maxillofac Surg Clin North Am.* 2004 Aug;16(3):309-22.
  24. Gardner DG. Some current concepts on the pathology of ameloblastomas. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod.* 1996 Dec;82(6):660-9.
  25. Olaitan AA, Arole G, Adekeye EO. Recurrent ameloblastoma of the jaws. A follow-up study. *Int J Oral Maxillofac Surg.* 1998 Dec;27(6):456-60.