RESEARCH PAPER

Conservative Management of Unicystic Ameloblastoma in Young Patients: A Prospective Single-Center Trial and Review of Literature

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Abstract

Aims The study intends to evaluate the clinical outcome of treating young patients of unicystic ameloblastoma, by various conservative treatment modalities such as marsupialization, enucleation with bone curettage and aid in establishing sound treatment guidelines.

Materials and Methods A case series of fifteen patients is presented of unicystic ameloblastoma, who presented to our Department over the past 6 years (2010–2015) and conservative method of treatment elected in all the patients. Also, a methodical literature review of studies discussing treatment of unicystic ameloblastoma in young age group (<20 years) patients over the last 15 years from 2001 to 2015 is added.

Results Faster osseous regeneration after conservative treatment was noted, owing to the young age and growth potential. Over a mean follow-up of 4 years, uneventful secondary healing, no clinical or radiographic evidence of recurrence and no apparent deformity were noted in any of the cases.

Conclusion Unicystic ameloblastoma is a benign, locally invasive odontogenic neoplasm of young age which can develop during the stage of tooth formation and hence its early inception and vast proliferation is not uncommon in this age group, due to which it can grow into a huge lesion causing significant morbidity. Conservative surgical management may be a viable option to reduce morbidity and

increase the probability of uneventful secondary healing and bone regeneration in the younger population.

Keywords Ameloblastoma · Unicystic · Pediatric · Enucleation

Introduction

Unicystic ameloblastoma refers to those cystic lesions that show clinical and radiographic characteristics of an odontogenic cyst but on histologic examination show a typical ameloblastomatous epithelium lining part of the cyst cavity, with or without luminal and/or mural tumour proliferation. The ratio of mandibular to maxillary unicystic ameloblastoma has been reported to be 13:1 [1].

Unicystic ameloblastoma, first described in 1977 has been considered a special type. The young age at occurrence, unilocular radiographic appearance, macroscopic cystic appearance, and most importantly, the better response to conservative treatment, make it a distinguishable entity. On the basis of histologic features, unicystic ameloblastoma can be classified into luminal, intraluminal and mural subtypes with prognostic and therapeutic significance [2]. Various treatment modalities such as enucleation, marsupialization, segmental or marginal resection are employed; however, more conservative treatments were reported frequently in younger patients [3].

Aims and Objectives

1. To present a case series evaluating the clinical outcome in fifteen young patients with mandibular unicystic ameloblastoma treated conservatively.

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2. To discuss the literature published on studies of treatment modalities employed for unicystic ameloblastoma in young patients from 2001 to 2015.
3. To review the outcome of the various surgical treatment methods for unicystic ameloblastoma in children and adolescents and highlight the importance of establishing a definitive treatment protocol.

Materials and Methods

- A computerized literature search was conducted at our Institute in the Department of Oral and Maxillofacial Surgery using Pubmed for published English articles on unicystic ameloblastoma.
- The search yielded 303 articles, of which 40 were chosen for review.
- The full texts of all these articles were thoroughly examined by all the authors.
- Inclusion criteria included:
 - Age: ≤ 20 years.
 - Case reports, retrospective case series, review studies of unicystic ameloblastoma.
 - Studies involving treatment modalities of unicystic ameloblastoma in pediatric or adolescent population.
 - Articles published from 2001 to 2015.
- Exclusion criteria:
 - Solid, multicystic, peripheral ameloblastomas.
 - Incomplete data studies.

Patients

A study was carried out in Indian patients of age 20 years and below, who were histopathologically diagnosed with unicystic ameloblastoma at our institution in Ahmedabad, India over the past 6 years (i.e., from January 2010 to July 2015). This study was carried out following the approval of the Ethics Committee of our institution. Fifteen patients of unicystic ameloblastoma were found with complete data, presented on follow-up and were hence chosen for the study. Patients' case notes, relevant radiographs, histopathology reports, and treatment charts were reviewed. The incidence of the tumour with respect to the age, sex, site of occurrence, histopathologic type, type of treatment instituted, and the recurrence rate was recorded. All the patients underwent incisional biopsy prior to surgery.

Clinical Findings

Patients between age group of 9–18 years with a mean age of 13.2 years; 9 males and 6 females, with the male to

female ratio of 1.5:1 and having unicystic ameloblastoma, confirmed histopathologically by incisional biopsy. All the lesions were located in the mandible: 5 lesions on the right, 9 lesions on the left side and one noted in the symphysis region. Also, 10 lesions occurred in the angle and/or ramus region, 4 involved the body region and one involved the symphysis. Clinical examination revealed a large, expansile mass with soft to hard swelling, painless on palpation and covered by normal mucosa. No anesthesia or paresthesia was reported. All the patients were identified with luminal or intra-luminal variant of unicystic ameloblastoma and one was reported to have the mural variety.

Radiologic Findings

In almost all cases, plain radiography by an O.P.G. showed a single well defined, unilocular radiolucency with associated involvement of one or more teeth or teeth buds. In two cases, a large multilocular radiolucency with typical soap bubble appearance was found. Condylar and coronoid processes were intact, no fracture was observed. Coronal computed tomography (CT) showed a large expansile lesion with cortical thinning of buccal and lingual cortical plates in majority of cases. Expansion of the lesion had caused displacement of the adjacent teeth. Root resorption was not observed.

Case 1

See Figs. 1, 2, 3.

Case 2

See Figs. 4, 5.

Fig. 1 Pre-operative O.P.G. of an 18 year old male showing a well defined unilocular radiolucency at left angle ramus region of mandible with impacted 37

Fig. 2 Pre-operative 3D-C.T. scan of an 18 year old male showing an expansile lesion involving left angle–ramus region with cortical expansion

Fig. 3 After enucleation and curettage of the lesion, O.P.G. at 6 months follow-up showing bony remodelling of the lesion at left mandible

Treatment

All the cases were treated conservatively either with enucleation with or without bone curettage or marsupialization followed by enucleation under General Anesthesia.

Marsupialization was chosen in two patients due to young age (10 and 9 years), involved erupting teeth and facial asymmetry. After 14 and 20 months respectively, sufficient regression of lesion and eruption of involved teeth in the lesion was noted in both the cases and they were enucleated [4].

A medicated ribbon gauze was used to pack large lesional cavities for permitting secondary closure. The ribbon gauze consisted of a medicated pack of a mixture of

Fig. 4 Pre-operative O.P.G. of a 16 year old male showing a well defined unilocular radiolucency at right angle ramus region of mandible with impacted 47, 48

Fig. 5 O.P.G. at 6th month follow-up, after enucleation, showing rapid remodelling and restoration of normal bony architecture at right angle ramus region of mandible

Betadine ointment, Neosporin ointment and Placentrex ointment. The pack was changed after every 3–4 days till sufficient shrinkage of the lesion was noted to permit closure.

Recurrence was not seen in any of the cases and we are monitoring these patients on follow-up for the same (Table 1).

Review

A world review of the literature over the last 15 years (2001–2015) identified a total of 74 patients exhibiting unicystic ameloblastoma in maxilla or mandible in 40 reports. Analysis of this data is provided in Table 2.

Of the total 74 patients identified with unicystic ameloblastoma, 37 are females (57.8%) and 27 are males (42.1%) with a ratio of 1.37:1. The mean age is 13 years. With regard to site, 89.1% occurred in the mandible, 10.8% in maxilla. This inference supports the studies stating mandible to be the commonest site followed by maxilla [2].

The articles were reviewed for radiological appearance and histological subtypes of unicystic

Table 1 Patients treated from 2010 to 2015 of Unicystic Ameloblastoma

Sr. no.	Age/sex	Reported in	Region of mandible	Maximum size of lesion (mm)	B/L/BL expansion	Radiologic appearance	Histopathological variant	Treatment	Followup period (years)	Recurrence
1.	14/F	Sept. 2011	Left angle-ramus	55 × 45	BL	UR	Intraluminal	EC	4	–
2.	10/M	Dec. 2011	Left body	35 × 25	BL	UR	Luminal	M + EC (EC done after 14 months of M)	4	–
3.	11/F	Jan. 2010	Right angle-ramus	40 × 50	BL	MR	Luminal	EC	5	–
4.	16/M	Jan. 2012	Right angle-ramus	35 × 45	BL	UR	Intraluminal	E	3	–
5.	10/F	Sept. 2012	Left angle-ramus	55 × 40	BL	UR	Intraluminal	EC	3	–
6.	15/M	Aug. 2011	Left ramus	25 × 30	B	UR	Luminal	EC	4	–
7.	13/F	March 2012	Right body	35 × 40	BL	UR	Mural	EC	3	–
8.	9/M	Apr. 2012	Symphysis	28 × 35	BL	UR	Intraluminal	M + EC (EC done after 20 months of M)	3	–
9.	16/F	Jan. 2011	Right angle	45 × 50	BL	UR	Luminal	EC	5	–
10.	10/F	May 2011	Left angle ramus	35 × 40	BL	MR	Intraluminal	EC	4	–
11.	12/M	Nov. 2010	Left body	35 × 25	BL	UR	Intraluminal	EC	5	–
12.	13/F	June 2010	Left angle-ramus	25 × 30	BL	UR	Intraluminal	EC	5	–
13.	16/F	Jan 2011	Right body	25 × 35	BL	UR	Luminal	EC	4	–
14.	18/M	Feb. 2010	Left angle ramus	20 × 30	B	UR	Intraluminal	EC	5	–
15.	15/F	Nov. 2012	Left angle	20 × 25	B	UR	Luminal	EC	3	–

M Male, *F* Female, *B* Buccal cortical plate expansion, *L* Lingual cortical plate expansion, *BL* Bucco-lingual expansion, *UR* Unilocular radiolucency, *MR* Multilocular radiolucency, *E* Enucleation, *M* Marsupialization, *EC* Enucleation with curettage

ameloblastoma, according to Ackermann classification [2]. Of the data available on 67 patients, only 3 studies presented a diagnosis of unicystic ameloblastoma appearing radiographically as multilocular radiolucency, while 64 reports presented a unilocular radiolucent lesion, suggesting that unicystic ameloblastoma radiographically also appears as a single cystic radiolucency. Type 2 (intraluminal) unicystic ameloblastoma—43.7% were found to be the highest, followed by Type 3 (mural)—37.5% and Type 1 (luminal)—18.7%. Of the

40 articles reviewed (74 patients), 63 case reports presented on followup of patients, of which 4 cases reported recurrence of lesion.

Review of the treatment protocols in the evaluated studies shows that enucleation was the most commonly performed procedure in 49 out of the 74 cases (66.21%) followed by marsupialization with enucleation (10.8%). Conservative procedures were performed in 65 out of 74 patients. (87.8%) while radical treatment was done in 9 (12.1%) patients (Table 3).

Table 2 Review of cases of pediatric and adolescent unicystic ameloblastoma from 2000 to 2015

Sr. no.	Author	Year	Country	No. of patients	Patient age (years)	Patient gender	Tumour location	Radiologic appearance	Histologic type	Treatment	Followup period (years)	Recurrence
1.	Acar et al. [5]	2015	Turkey	1	1.6	M	MAND	UR	3	ENUC	4.5	None
2.	de Paulo et al. [4]	2014	Brazil	1	7	F	MAND	UR	2	MARS + ENUC	3	None
3.	Park HS et al. [6]	2014	Korea	5	18.6 (mean)	1M 4F	MAND	UR	NA	MARS + ENUC	NA	–
4.	Saluja HM [7]	2014	India	1	16	F	MAND	UR	NA	ENUC	NA	–
5.	Deore SS [8]	2014	India	1	15	M	MAND	UR	2	RES + ILIAC	NA	–
6.	Sasaki et al. [9]	2014	Japan	1	20	M	MAND	UR	3	ENUC	2.5	None
7.	Naidu et al. [10]	2014	India	1	11	F	MAND	UR	2	ENUC	NA	–
8.	Bhushan et al. [11]	2014	India	1	14	M	MAND	UR	2	ENUC	0.5	None
9.	Saravana kumar [12]	2014	India	1	16	F	MAND	UR	3	MARS + ENUC + ILIAC	2	None
10.	Shi S et al. [13]	2014	China	5	17	M	MAND	UR	NA	ENUC	9.7	None
					19	F	MAND	UR	NA	ENUC	4	+
					18	F	MAND	UR	NA	ENUC	3.5	None
					13	M	MAND	UR	NA	ENUC	2.5	None
					20	F	MAND	UR	NA	ENUC	2	None
11.	Urs et al. [14]	2014	India	7	NA	3 M 4F	3MAX 4MAND	–	NA	ENUC	6	None
12.	Xavier et al. [15]	2014	Brazil	1	19	F	MAND	UR	3	MARS + ENUC and CARN	3	None
13.	Bhutia et al. [16]	2013	India	1	5	M	MAND	UR	1	ENUC and CARN	2	None
14.	Arora et al. [17]	2013	India	1	3	F	MAX	UR	2	ENUC	1.25	None
15.	Andrade NN et al. [18]	2013	India	10	NA	NA	MAND	UR	NA	ENUC	15	None + (1 patient)
16.	Rajeshkumar et al. [19]	2013	India	1	14	F	MAND	UR	NA	ENUC and CARN	1	None
17.	Sengüven et al. [20]	2013	Turkey	1	9	M	MAND	UR	3	ENUC	8	+
18.	Sarode et al. [21]	2013	India	1	18	M	MAND	MR	2	ENUC and CARN	NA	–
19.	Chaudhary et al. [22]	2013	India	1	3	F	MAX	UR	3	ENUC and CARN	0.5	None
20.	Tomita Y [23]	2013	Japan	1	14	F	MAND	UR	NA	MARS + ENUC	10	None
21.	Bates et al. [24]	2013	U.K.	1	15	F	MAND	UR	NA	ENUC	NA	–
22.	Nagalaxmi et al. [25]	2013	India	1	18	F	MAND	MR	2	ENUC	0.75	None
23.	Stoeltinga PJ [26]	2012	Netherlands	1	18	F	MAND	UR	NA	ENUC	NA	–
24.	Scariot et al. [27]	2012	Brazil	1	9	F	MAND	UR	2	ENUC	2	None
25.	Ponniiah I. [28]	2011	India	1	8	F	MAND	UR	NA	ENUC	4	+
26.	Gupta et al. [29]	2011	India	1	18	F	MAND	UR	1	HEMI + FIBULA	1	None

Table 2 continued

Sr. no.	Author	Year	Country	No. of patients	Patient age (years)	Patient gender	Tumour location	Radiologic appearance	Histologic type	Treatment	Followup period (years)	Recurrence
27.	Dolanmaz D et al. [30]	2011	Turkey	1	17	M	MAND	UR	1	MARS + ENUC	2.5	None
28.	Kalaskar et al. [31]	2011	India	2	9	M	MAX	UR	2	ENUC and CARN	0.5	None
29.	Khare et al. [32]	2011	India	1	12	M	MAND	UR	2	ENUC and CARN	NA	–
30.	Chacko et al. [33]	2011	India	1	13	M	MAND	UR	1	ENUC and CARN	5	None
31.	Menditti et al. [34]	2011	Italy	1	9	M	MAND	NA	NA	ENUC	5	None
					14	M	MAND	UR	1	ENUC	1	None
32.	Pitak-Arnop P et al. [35]	2010	France	1	19	M	MAND	UR	1	ENUC	5	None
33.	Yavagal et al. [36]	2009	India	1	13	F	MAND	UR	2	ENUC	NA	–
34.	Qureshi et al. [37]	2008	India	1	10	F	MAND	UR	NA	ENUC	2	None
35.	Patel et al. [38]	2007	U.K.	1	14	M	MAND	UR	2	ENUC	1	None
36.	Navarro CM et al. [39]	2004	Brazil	1	17	M	MAX	UR	2	ENUC	NA	–
37.	Al-Khateeb et al. [40]	2003	Jordan	6	18	M	MAND	UR	3	ENUC	13	None
					14	F	MAND	UR	3	ENUC	NA	–
					18	F	MAND	UR	3	ENUC	9	None
					19	F	MAND	MR	3	RESECTION	7	None
					13	M	MAND	UR	3	ENUC	6	None
					9	F	MAND	UR	3	ENUC	4	None
38.	Ord et al. [41]	2002	U.S.A.	6	16	M	MAND	UR	NA	MARG RES	7	None
					16	F	MAND	UR	NA	MARG RES	6	None
					12	M	MAND	UR	NA	ENUC	0.5	None
					13	F	MAND	UR	NA	ENUC	1.5	None
					15	F	MAX	UR	NA	ENUC	0.5	None
					18	F	MAND	UR	NA	RES + ILIAC	0.5	None
39.	Gordon SC et al. [42]	2001	U.S.A.	1	19	F	MAND	UR	2	PART MAND + ILIAC	NA	–
40.	Matsumoto et al. [43]	2001	Japan	1	19	M	MAND	UR	NA	MARS + ENUC	2.5	None

MAND Mandible, MAX Maxilla, UR unilocular radiolucency, MR multilocular radiolucency, NA not available, M male, F female, ENUC enucleation, MARS marsupialization, RES segmental resection, ILIAC Iliac crest graft, CARN Carnoy's solution (chemical cauterization), HEMI hemimandibulectomy, FIBULA free fibula graft reconstruction, PART MAND partial mandibulectomy, MARG RES marginal resection

Discussion

Gardner [44] has pointed out that there is a difference in pathological behaviour between those lesions that are simply cystic or show intraluminal proliferation (Types 1 and 2) and those in which the epithelium penetrated and breached the fibrous wall, therefore having the capacity to invade the cancellous bone (type 3). This classification has serious implications for the clinician, since types 1 and 2 favoured successful outcome with conservative management [2].

In children, the treatment of unicystic ameloblastoma is complicated by three factors [27]: (1) Continuing facial growth, different bone physiology (greater percentage of cancellous bone, increased bone turnover and reactive periosteum) and presence of unerupted teeth; (2) Difficulty in initial diagnosis; and (3) Predominance of the unicystic type of ameloblastoma. There is a general consensus in various literatures that ameloblastoma has to be treated aggressively to avoid recurrences, but there is a dilemma on the applicability of an initial radical, extensive surgery in children.

Recent advancements in the understanding of the biological behaviour of ameloblastoma have revealed that unicystic lesions are well-localized by the fibrous capsule of the cyst, with few tumours breaching peripheral tissues, Gardner [44] discussed the treatment of unicystic ameloblastoma on the basis of pathological and anatomical considerations and recommended curettage. Overall health, tumour size, location, duration, psychological impact, periodic follow-up examinations should all be considered when formulating the surgical treatment plan.

In a case presented by de Paulo et al. [4], marsupialization followed later by enucleation was adopted as treatment of choice for an extensive unicystic ameloblastoma in mandible involving body, ramus and condyle. Among the pediatric and adolescent population, the conservative line of treatment plays an excellent role as it is associated with a faster ‘bone fill’ and efficient restoration of normal bony architecture, which is attributed to the pliability of the young bones and hence lesions of a huge expanse can be successfully treated. Unicystic ameloblastoma is biologically less aggressive and has a better response to enucleation or curettage than the solid ameloblastoma [45].

Often, unicystic ameloblastomas involving the ramus of the mandible do not require resection and respond well to careful enucleation and chemical cauterization [3]. Samman and Lau [3], reviewed that large unicystic ameloblastomas are usually approached extraorally with decortication followed by enucleation and application of Carnoy’s solution. The wound is then either closed primarily or packed with ribbon gauze to facilitate healing.

Khare and Kumar [32] modified the enucleation procedure for unicystic ameloblastoma lesions involving ramus region, by employing a pedicled lateral ramal osteoperiosteal flap. This facilitates healing and resists infection. Moreover, the flap has the potential for regeneration, obliterates the dead space left behind after excavation of lesion and prevents collapse of the facial soft tissues and minimizes postoperative deformity. Re-entry surgery with liquid nitrogen application (cryotherapy) following enucleation is another novel treatment modality for large lesions of unicystic ameloblastoma and which may play a great role in prevention of recurrence of this tumour, as discussed by Ogunsalu et al. [46]

Sasaki et al. performed enucleation according to a “dredging method,” as reported by Kawamura et al. in 1991 [47], for a large unicystic ameloblastoma lesion in mandible. In their method, after the deflation (marsupialization) and enucleation or only enucleation, enucleation is repeated five to six times within 2–3 months until no tumour cell nest is identified in the pathological findings of scar tissues removed by two consecutive dredging. This method gives a low recurrence rate and complication rate with no dysfunction and deformity.

Unicystic ameloblastoma is best treated in children and adolescents conservatively with decompression, enucleation and peripheral ostectomy and periodic long-term follow up maintained. A more aggressive surgical approach may be considered when the condition recurs more than twice or according to the patient’s wishes [48]. In order to avoid the high recurrence rates, especially in type 3 unicystic ameloblastoma, a biopsy is recommended to rule out possible mural involvement.

In the present case series, the association with an impacted and/or erupting tooth and presence of non-specific thin epithelium in focal areas of cystic tumour supports the hypothesis [49] i.e. ameloblastoma arising from preexisting dentigerous cyst. Hence, the choice of conservative treatment of enucleation with curettage and the extraction of involved teeth proved effective. Ameloblastoma being an aggressive condition, the extraction of involved teeth is a way of minimizing the possibility of recurrence. In the case series reported here, the conservative surgical treatment of mandibular ameloblastoma yielded excellent postoperative function and aesthetics with no recurrence, which is in accordance with the results from the review as well. Regardless of the kind of treatment plan employed, a strict follow-up regime of monthly evaluation during the first year of surgery and every 3 monthly, 6 monthly or yearly follow-up thereafter should be strictly adhered to for at least a period of 15 years, due to the late recurrence of the tumour noted in adulthood.

Table 3 Review of treatment protocols for unicystic ameloblastoma in young patients

Sr. no.	Treatment	Number of cases (out of 74)	Percentage (%)
1.	Enucleation	49	66.21
2.	Marsupialization + enucleation	8	10.81
3.	Enucleation with use of Carnoy's solution	7	9.45
4.	Resection with Iliac crest graft reconstruction	2	2.7
5.	Marginal resection	2	2.7
6.	Marsupialization + enucleation with Iliac crest graft reconstruction	1	1.35
7.	Marsupialization + enucleation with use of Carnoy's solution	1	1.35
8.	Hemimandibulectomy with free fibula graft reconstruction	1	1.35
9.	Segmental resection	1	1.35
10.	Segmental resection with iliac crest graft reconstruction	1	1.35
11.	Partial mandibulectomy with iliac crest graft reconstruction	1	1.35

Conclusion

In the study, we show that among the younger population, unicystic ameloblastoma can be successfully treated by a conservative line of treatment viz. enucleation with bone curettage. We preferred conservative surgery in the treatment of such cases primarily because of the young age of the patients. In all cases, long-term follow-up with radiography is important. We are still monitoring our patients annually using radiography and biopsy if required.

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Compliance with Ethical Standards

Conflict of interest The authors hereby wish to state that this paper does not have any financial and personal relationships with other people or organisations that could inappropriately influence (bias) their work.

Informed consent Informed consent was obtained from all individual participants included in the study.

References

- Philipsen HP, Reichart PA (1998) Unicystic ameloblastoma; a review of 193 cases from the literature. *Oral Oncol* 34:317–325
- Ackermann GL, Altini M, Shear M (1988) The unicystic ameloblastoma: a clinicopathological study of 57 cases. *J Oral Pathol* 17:541–546
- Lau SL, Samman N (2006) Recurrence related to treatment modalities of unicystic ameloblastoma: a systematic review. *Int J Oral Maxillofac Surg* 35:681–690
- de Paulo LF, Oliveira MT, Rodrigues ÁR, Zanetta-Barbosa D (2015) Treatment of an extensive unicystic ameloblastoma in a 7-year-old child: the best approach? *Br J Oral Maxillofac Surg* 53(3):292–294
- Acar AH, Yolcu Ü, Erdem NF, Asutay F (2015) Is conservative surgical treatment sufficient to treat unicystic mural ameloblastoma in infant? *J Craniofac Surg*. 26(4):e297–e299
- Park HS, Song IS, Seo BM, Lee JH, Kim MJ (2014) The effectiveness of decompression for patients with dentigerous cysts, keratocystic odontogenic tumours, and unicystic ameloblastoma. *J Korean Assoc Oral Maxillofac Surg* 40(6):260–265
- Saluja HM, Rudagi BM, Mahindra UR, Gaikwad PT, Dehane VV (2014) Retrieval of foreign body from a postoperative defect in the mandible during the follow-up period: a bizarre occurrence. *Natl J Maxillofac Surg* 5(1):67–69
- Deore SS, Dandekar RC, Mahajan AM, Patil R, Prakash N (2014) Plexiform unicystic ameloblastoma: a rare variant of ameloblastoma. *Case Rep Dent* 2014:146989
- Sasaki R, Watanabe Y, Ando T, Akizuki T (2014) Two stage enucleation and deflation of a large unicystic ameloblastoma with mural invasion in mandible. *Craniofac Trauma Reconstr* 7(2):139–142
- Naidu SR, Hegde RJ, Devrukhkar VN, Patel AR (2014) Conservative management of unicystic ameloblastoma in a young child: a case report. *J Ind Soc Pedod Prev Dent* 32(3):251–254
- Bhushan NS, Rao NM, Navatha M, Kumar BK (2014) Ameloblastoma arising from a dentigerous cyst—a case report. *J Clin Diagn Res* 8(5):ZD23–ZD25
- Saravanakumar B, Parthiban J, Aarthi Nisha VA, Sarumathi T, Prakash CA (2014) Unicystic ameloblastoma of the mandible—report of two cases with review of literature. *J Clin Diagn Res* 8(5):07–09
- Shi S, Liu Y, Shan Y, Fu T, Zhao S (2014) Enucleation combined with peripheral ostectomy: its role in the management of large cystic ameloblastomas of the mandible. *J Craniofac Surg* 42(8):1659–1663
- Urs AB, Arora S, Singh H (2014) Intra-osseous jaw lesions in paediatric patients: a retrospective study. *J Clin Diagn Res* 8(3):216–220
- Xavier SP, de Mello-Filho FV, Rodrigues WC, Sonoda CK, de Melo WM (2014) Conservative approach: using decompression procedure for management of a large unicystic ameloblastoma of the mandible. *J Craniofac Surg* 25(3):1012–1014
- Bhutia O, Roychoudhury A, Arora A, Mallick S et al (2013) Management of unicystic ameloblastoma of the mandible in a 5-year old child. *Natl J Maxillofac Surg* 4(2):232–234
- Arora S, Kumar P, Urs AB, Augustine J (2013) Unicystic ameloblastoma in 3 year old paediatric patient—a rare entity. *J Clin Exp Dent*. 5(1):e54–e57
- Andrade NN, Shetye SP, Mhatre TS (2013) Trends in pediatric ameloblastoma and its management: a 15 year Indian experience. *J Maxillofac Oral Surg* 12(1):60–67

19. Rajeshkumar BP, Rai KK, Geetha NT, Shivakumar HR, Upasi AP (2013) Carnoy's in aggressive lesions: our experience. *J Maxillofac Oral Surg* 12(1):42–47
20. Sengüven B, Bariş E, Oygür T, Öztemel A (2013) Recurrent unicystic mural type ameloblastoma in a 9-year-old boy, 8 years follow-up. *Contemp Clin Dent* 4(4):569–572
21. Sarode GS, Sarode SC, Vaidya K (2013) Intraluminal plexiform hemangioameloblastomatous proliferation in unicystic ameloblastoma: an unusual case report. *Ind J Dent Res* 24(3):390–392
22. Chaudhary Z, Krishnan S, Sharma P, Sharma R, Kumar P (2012) A review of literature on ameloblastoma in children and adolescents and a rare case report of ameloblastoma in a 3-year-old child. *Craniomaxillofac Trauma Reconstr* 5(3):161–168
23. Tomita Y, Kuroda S, Takahashi T, Ohura R, Tanaka E (2013) Orthodontic occlusal reconstruction after conservative treatment of unicystic ameloblastoma in an adolescent patient: 10-year follow-up. *Am J Orthod Dentofacial Orthop* 144(3):466–470
24. Bates T, Keith D, Sloan P (2014) Unicystic ameloblastoma presenting in Gardner's syndrome. *Head Neck Pathol* 8(2):239–240
25. Nagalaxmi V, Sangmesh M, Maloth KN, Kodangal S, Vani C, Goyal S (2013) Unicystic mural ameloblastoma: an unusual case report. *Case reports in dentistry*, vol. 2013, Article ID 957418, 6 pages. doi:[10.1155/2013/957418](https://doi.org/10.1155/2013/957418)
26. Stoelinga PJ (2012) The management of aggressive cysts of the jaws. *J Maxillofac Oral Surg* 11(1):2–12
27. Scariot R, da Silva RV, da Silva Felix W Jr et al (2012) Conservative treatment of ameloblastoma in child: a case report. *Stomatologija* 14(1):33–36
28. Ponniah I (2011) Recurrent unicystic ameloblastoma in a child. *J Oral Maxillofac Pathol* 15(2):236–238
29. Gupta N, Saxena S, Rathod VC, Aggarwal P (2011) Unicystic ameloblastoma of the mandible. *J Oral Maxillofac Pathol* 15(2):228–231
30. Dolanmaz D, Etoz OA, Pampu A, Kalayci A, Gunhan O (2011) Marsupialization of unicystic ameloblastoma: a conservative approach for aggressive odontogenic tumours. *Ind J Dent Res* 22:709–712
31. Kalaskar R, Unawane AS, Kalaskar AR, Pandilwar P (2011) Conservative management of unicystic ameloblastoma in a young child: report of two cases. *Contemp Clin Dent* 2(4):359–363
32. Khare G, Kumar S (2011) Application of osteomucoperiosteal flap in large unicystic ameloblastoma to promote early bone healing: an alternative to primary bone grafting. *Contemp Clin Dent* 2(4):283–286
33. Chacko V, Kuriakose S (2011) Conservative management of a case of plexiform ameloblastoma. *Dent Update* 38(5):336–338
34. Menditti D, Laino L, De Marco G, De Rosa A (2011) Unicystic ameloblastoma of the mandible. *In Vivo* 25(1):125–127
35. Pitak-Arnnop P, Chaine A, Dhanuthai K, Bertrand JC, Bertolus C (2010) Unicystic ameloblastoma of the maxillary sinus: Pitfalls of diagnosis and management. *Hippokratia* 14(3):217–220
36. Yavagal C, Anegundi RT, Shetty S (2009) Unicystic plexiform ameloblastoma: an insight for pediatric dentists. *J Ind Soc Pedod Prev Dent* 27(1):70–74
37. Qureshi SS, Qureshi SS, Medhi SS, Kane SV (2008) Unicystic ameloblastoma of the mandible masquerading as carcinoma of the oral cavity in a 10-year-old girl. *Am J Surg* 196(3):e7–e9
38. Patel H, Rees RT (2005) Unicystic ameloblastoma presenting in Gardner's syndrome: a case report. *Br Dent J* 198(12):747–748
39. Navarro CM, Principi SM, Massucato EM, Spoto MR (2004) Maxillary unicystic ameloblastoma. *Dentomaxillofac Radiol* 33(1):60–62
40. Al-Khateeb T, Ababneh KT (2003) Ameloblastoma in young Jordanians: a review of the clinicopathologic features and treatment of 10 cases. *J Oral Maxillofac Surg* 61(1):13–18
41. Ord RA, Blanchaert RH Jr, Nikitakis NG, Sauk JJ (2002) Ameloblastoma in children. *J Oral Maxillofac Surg* 60(7):762–770 (**discussion, 770–1. Review**)
42. Gordon SC, MacIntosh RB, Wesley RK (2001) A review of osteoblastoma and case report of metachronous osteoblastoma and unicystic ameloblastoma. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 91(5):570–575 (**Review**)
43. Matsumoto Y, Mizoue K, Seto K (2001) Atypical plexiform ameloblastoma with dentinoid: adenoid ameloblastoma with dentinoid. *J Oral Pathol Med* 30(4):251–254 (**Review**)
44. Gardner DG (1984) A pathologist's approach to the treatment of ameloblastoma. *J Oral Maxillofac Surg* 42:161–166
45. Rosenstein T, Pogrel AM, Smith RA, Regezi JA (2001) Cystic ameloblastomas: Behavior and treatment of 21 cases. *J Oral Maxillofac Surg* 59:1311–1316
46. Ogunsalu C, West W, Lewis A, Williams N (2011) Ameloblastoma in Jamaica—predominantly unicystic: analysis of 47 patients over a 16-year period and a case report on re-entry cryosurgery as a new modality of treatment for the prevention of recurrence. *West Indian Med J* 60(2):240–246
47. Kawamura M, Inoue N, Kobayashi I et al (1991) “Dredging Method”—a new approach for the treatment of ameloblastoma. *Asian J Oral Maxillofac Surg* 3:81–88
48. Huang Y, Lai ST, Chen CH, Chen CM, Wu CW, Shen YH (2007) Surgical management of ameloblastoma in children. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 104:478–485
49. Li TJ, Wu YT, Yu SF, Yu GY (2000) Unicystic ameloblastoma: a clinicopathologic study of 33 Chinese patients. *Am J Surg Pathol* 24:1385–1392