



## Demographic Data and Treatment Outcomes of Ameloblastoma in Sudanese Patients Treated at Khartoum Teaching Dental Hospital

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### Abstract

**Background:** Ameloblastoma is the most common clinically significant odontogenic tumor because of its aggressive and local invasion behavior. It is considered the most common odontogenic tumor in Sudan. Familiarity with this type of odontogenic tumor is an absolute necessity for careful management and improvement of outcomes.

**Methods:** A descriptive (retrospective - prospective) cross-sectional study of 209 patients of ameloblastoma was conducted at Khartoum Teaching Dental Hospital. Patients were interviewed and examined. Data were collected and analysed included demography, treatment provided and treatment outcomes.

**Results:** Of all patients, 122 (58.4%) were males and 87 (41.6%) were females. The third and fourth decades being the most commonly affected (29.7% and 22, 5%) respectively. 199 (94.8%) of the cases of ameloblastoma occurred in the mandible, while 11 (5.2%) of the cases occurred in the maxilla. In 129 (50.6%) of the patients ameloblastoma occurred on the left side of the jaws, while 126 (49.4%) of the tumours occurred on the right side with an almost equal distribution. The posterior area of the jaw was commonly involved in (79%) of the cases. Segmental resection was the most common treatment done in (30%) of the cases. Reconstruction plate was the most common means of reconstruction in 107 (77%) patients, followed by bone graft in 19 (13.7%) patients. Follow up revealed 19 (11%) patients who developed recurrence of the ameloblastoma. 127 (73.4%) patients developed teeth-related problems while 29 (16.8%) patients developed difficulty in swallowing and 117 (67.6%) patients developed difficulty in chewing. Speech problem developed in 65 (37.6%) patients. 70 (40.5%) patients developed changes in their appearance and 29 (16.8%) patients developed changes in their psychological status also 10 patients (5.8%) got divorced. 45 (26%) patients developed low daily activity postoperatively while 13 (12.4%) patients lost/stopped their jobs and 20 (11.6%) patients developed negative changes in their socioeconomic status.

**Conclusion:** In this study the demographic data of the presently reported patients are more or less in accordance with those obtained from the literature. Whatever the type of Ameloblastoma radical surgeries showed less recurrence rate rather than conservative one despite the disfigurement that may result. This study showed that there is a direct impact of ameloblastoma disease and treatment provided on patient's quality of life.

**Keywords:** Ameloblastoma; Gender Predilection; Segmental Resection; Teeth Related Problems; Bone Graft

### Abbreviations

SMA: Solid/Multicystic Ameloblastoma; PA: Peripheral Ameloblastoma; UA: Unicystic Ameloblastoma; AC: Ameloblastic Carcinoma; KTDH: Khartoum Teaching Dental Hospital

### Introduction

Ameloblastoma is the most common clinically significant odontogenic tumor that originated from odontogenic epithelium. It may arise from rest of dental lamina, enamel organ, epithelial lining of

odontogenic cyst and from the basal cells of the oral mucosa [1]. Ameloblastoma is a slow growing, locally invasive tumor; it occurs in three different clinicoradiographic situations, conventional solid or multicystic (86%), unicystic (13%), and peripheral-extraosseous (1%) [1]. WHO described the fourth type as desmoplastic ameloblastoma [2]. Ameloblastoma is mainly a lesion of adults. It occurs predominantly in the fourth and fifth decades of life, and the age range is very broad, extending from childhood to late adulthood (with a mean age, of approximately 40 years).

The rare lesions occurring in children are usually cystic and appear clinically as odontogenic cysts. There appears to be no gender predilection for this tumor.

Ameloblastoma may occur anywhere in the mandible or maxilla, although the mandibular molar-ramus area is the most common site. In the maxilla, the molar area is more commonly affected than the premolar and anterior regions. Lesions usually are asymptomatic and are discovered during routine radiographic examination or because of asymptomatic jaw expansion, tooth movement or malocclusion may be the initial presenting signs [3]. Radiographic presentations of ameloblastoma varied from small unilocular radiolucency to large multilocular radiolucency, it may associate with root resorption and unerupted tooth. It may vary from small loculation (honey comb) to large loculation (soap bubble). Buccal and lingual cortical expansion is frequently present. Follicular and plexiform patterns are the most common variants of conventional ameloblastoma. Less common histopathologic patterns include the acanthomatus, granular cell, and desmoplastic cell types. Luminal, intra luminal and mural are the common variants of unicystic ameloblastoma. Peripheral ameloblastoma has same histopathological features as the intraosseous form of this tumor [1,3]. Management of ameloblastoma ranges from conservative, simple surgical to radical surgical treatment according to clinical, radiological and histopathological presentations. The surgical management of ameloblastoma ranges from simple enucleation plus curettage or bone skimming, segmental resection, or wide resection with or without reconstruction. If neighboring soft tissues are confirmed to be infiltrated have to be excised including healthy margins. If the tumor remains untreated for a long time, it may reach a large size, compress and infiltrate the neighboring soft tissues, obstruct the air way, affecting nutrition by interfering with mastication and swallowing, erode major arteries and reach distant parts of body or by aspiration or inhalation may deposit in the lungs [4].

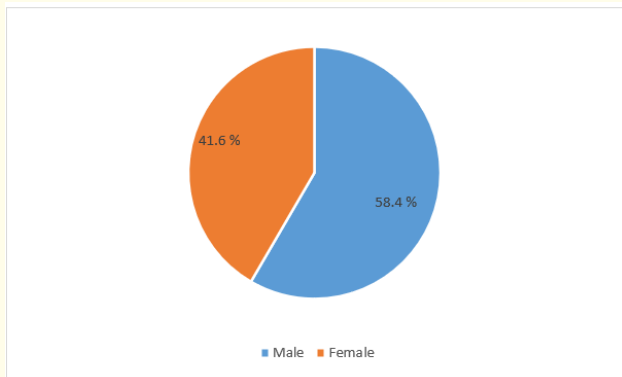
## Materials and Methods

Descriptive cross sectional hospital based study conducted at Khartoum Teaching Dental Hospital the main referral oral and maxillofacial center in Sudan included all patients who diagnosed with ameloblastoma and operated during a 10 years' period from January 2006 to January 2016. Of 235 files of ameloblastoma patients were reviewed during the period of the study, 209 files were included and 26 files were excluded due to incomplete data. Patients attended to (KTDH) interviewed and examined using mask and Gloves for each one. Histopathology records and radiographs reviewed again and surgical site examined by the researcher. Patients who could not attend to KTDH were interviewed over the phone. Data were collected from patients' files, radiographic records, histopathological records and patient's interviews. For the analysis of mandibular ameloblastoma, the site of occurrence was categorized as anterior, premolar, molar, and posterior based on the region affected. For the mandible, the posterior region was defined to include (the ramus, angle, coronoid process condyle). For the analysis of maxillary ameloblastoma, the site of occurrence was categorized as anterior, premolar, molar regions. The treatment methods classified into groups: Conservative treatment (enucleation, marsupialization bone curettage), resection (marginal or segmental) mandibulectomy, partial and total maxillectomy. For the analysis of patient's satisfaction, a score of 1- 10 was used divided in to three categories, 1 - 3, 4 - 6 and 7 - 10. All data collected by the researcher in data collection sheets. Data analyzed by using computerized method (SPSS version 22) and statistical analysis set at 95% confidence level with 0.05 level of significance. Data presented in form of graphs and tables. Approval was obtained from Sudanese Medical Specialization Board and from the general Director of KTDH. The patients included in the study wrote informed consent and those in distant area consented verbally by Telephone calls.

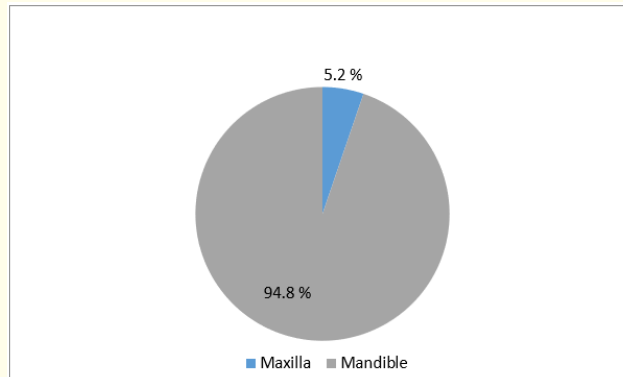
## Results

This study was conducted among Sudanese patients who were diagnosed with ameloblastoma and operated in KTDH. A total of 209 patients were enrolled in this study from (January 2006 to January 2016), 122 patients (58.4%) were males and 87 (41.6%) were females (Figure 1).

The mean age of patients in this study was 49 years, with an age range of 9 - 89 years. Ameloblastoma was encountered in almost

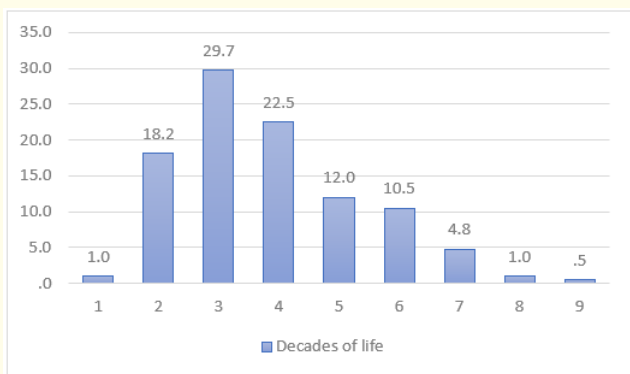


**Figure 1:** Gender distribution of ameloblastoma.

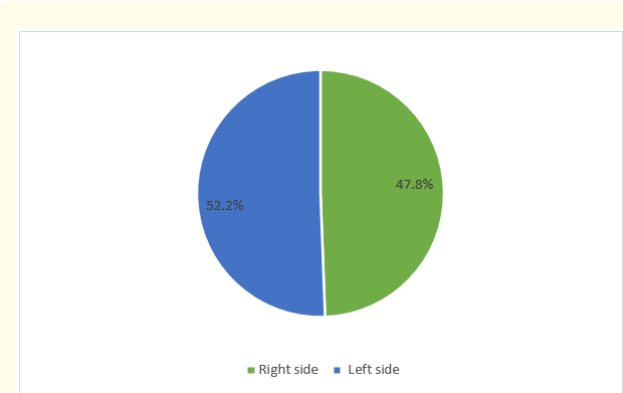


**Figure 3:** Maxillo- mandibular distribution of ameloblastoma.

all decades of life with the third and fourth decades being the most common (29.7% and 22.5%) respectively while 92.9% of lesions occurred between the second and the sixth decades (Figure 2).



**Figure 2:** Age percentages of ameloblastoma.

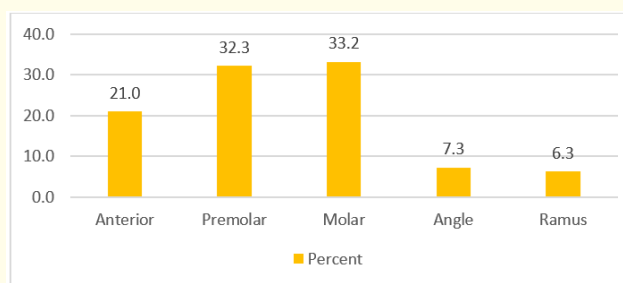


**Figure 4:** Side distribution of ameloblastoma

Out of 209 patients, 199 (94.8%) of the cases of ameloblastoma occurred in the mandible, while 11 (5.2%) of the cases occurred in the maxilla (Figure 3).

One hundred nine (52.2%) of the patients had ameloblastoma occurring on the left side of the jaws, while one hundred (47.8%) of the tumors occurred on the right side with almost equal distribution (Figure 4).

The molars area was the most frequent area that was encountered (33.2%) followed by premolars area (32.3%), ramus area showed less frequency (6.3%) (Figure 5).



**Figure 5:** Locations distribution of ameloblastoma.

Out of 11 cases that occurred in the maxilla, 6 (54.5%) cases occurred at right side, 3 (27.3%) cases occurred at left side while 2 (18.2%) cases crossed the midline of the maxilla (Figure 6A).

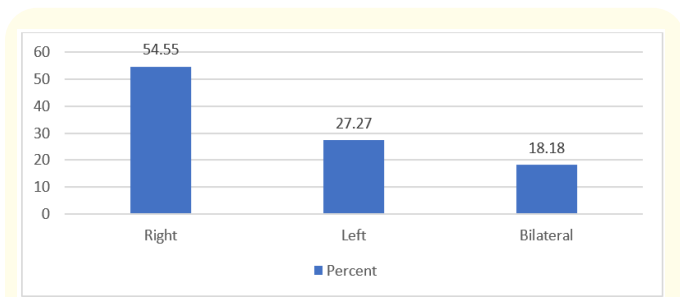


Figure 6A: Showing tumor location within the maxilla.

Regarding the mandible, 87 (44%) of the cases of ameloblastoma occurred at left side, 74 (37.3%) cases occurred at right side while 37 (18.7%) of the cases crossed the midline (Figure 6B).

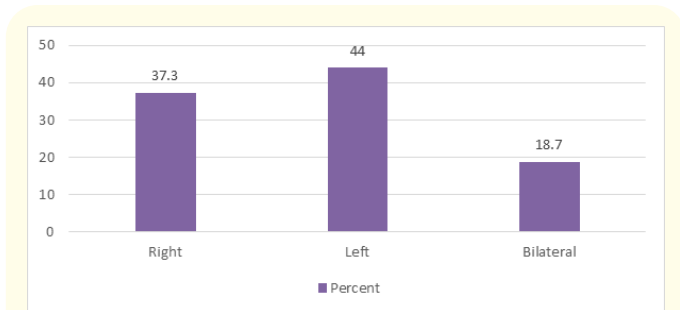


Figure 6B: Showing tumor location within the mandible.

Regarding the radiographic presentations, 152 (72.7%) of the lesions showed multilocular radiolucencies, while 57 (27.3%) presented unilocular radiolucencies (Figure 7).

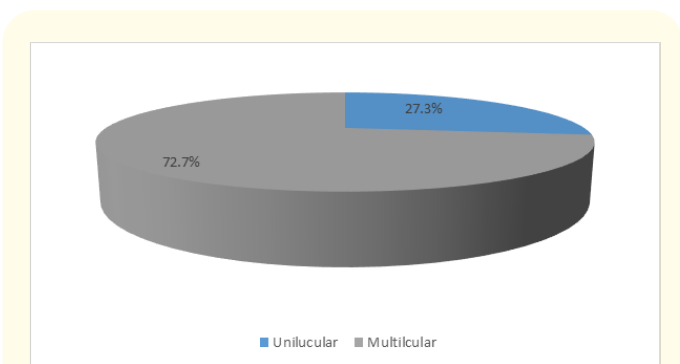


Figure 7: Radiographic presentations of ameloblastoma.

In radiographic involvement, 178 (86%) cases of ameloblastoma showed root resorption (Figure 8A).

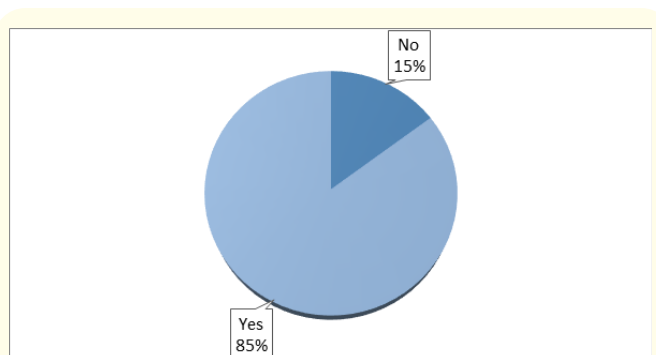


Figure 8A: Percentage of root resorption in cases of ameloblastoma.

There were 29 (14%) cases with unerupted teeth visible within the radiographs (Figure 8B).

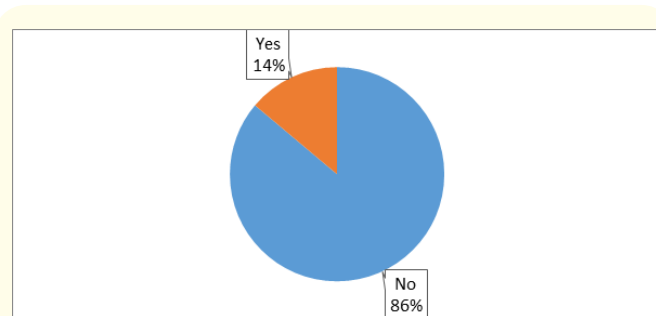


Figure 8B: Percentage of unerupted teeth in cases of ameloblastoma.

With regard to the treatment modalities segmental resection was the most common treatment performed (30%) followed by enucleation and curettage (20%) and partial maxillectomy was the least common treatment performed (1.4%) (Figure 9).

Regarding Reconstruction, of all 20 patients only 139 underwent reconstruction surgery. Reconstruction plate alone was the most common means of primary reconstruction used in 130 (87.2%) patients, followed by bone graft and bone plating 40 (26.8%) patients and soft tissue flap was the least common reconstruction means which was performed in 5 patients (2.7%) (Figure 10).

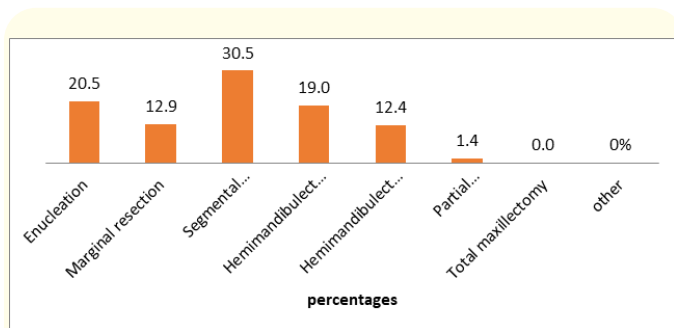


Figure 9: Treatment modalities performed to the patients of ameloblastoma.

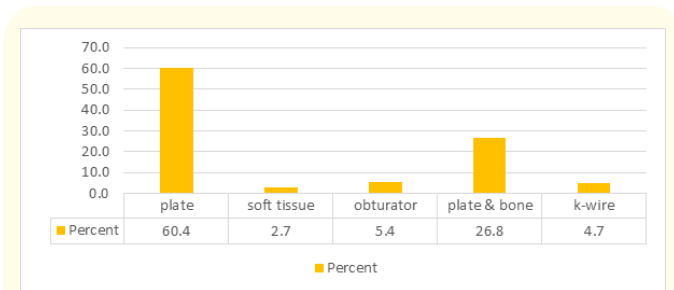


Figure 10: Showing the various materials of reconstruction.

In bone graft reconstruction, the iliac crest was the most common source of bone graft used in 35 (87.5%) of the cases, followed by fibula graft in 3 (7.5%) patients then costochondral graft which was the least bone graft used in 2 (5%) patient, all were free grafts (Figure 11).

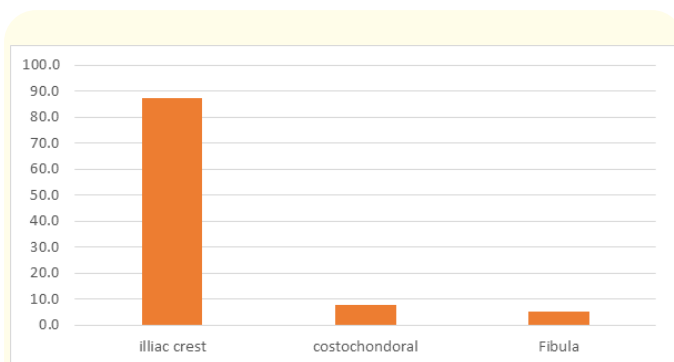


Figure 11: Types of bone graft used in reconstruction.

Out of 209 patients, 21 (10%) patients could not be followed, our contacts provided information on 188 patients of whom 173 were alive (82.8%) and 15 (7.2%) reported as dead (Figure 12).

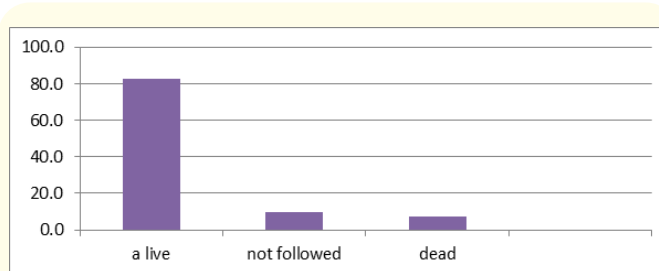


Figure 12: Showing percentage of patient's availability for postoperative follow up.

Follow up periods of ameloblastoma patients in a regular manner divided as table 1 illustrated below.

Period	No. of patients	Percentage
Within the first year	26	12.4 %
Two years	27	12.9 %
Three years	31	14.8 %
Four years	30	14.4 %
Five years	15	7.2 %
Six years	25	12.0 %
Seven years	18	8.6 %
Eight years	17	6.7 %
Nine years	13	6.2 %
Ten years	10	4.8 %

Table 1: Showing periods of postoperative follow up in years.

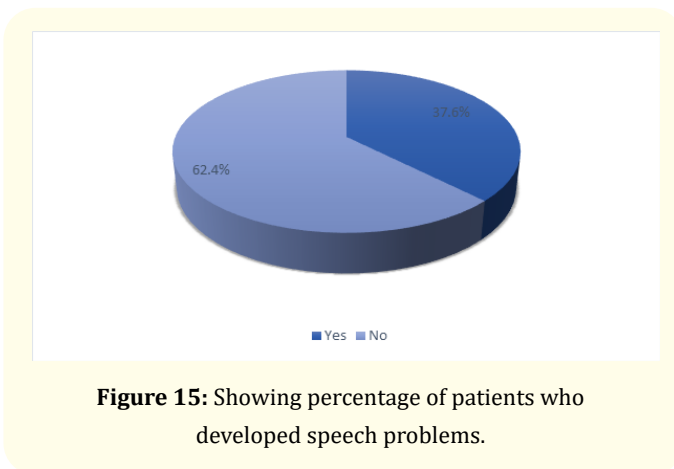
Different reasons were mentioned for lack of follow up distance was the most common cause (33.1%) (Table 2).

Nineteen (11%) patients developed recurrence after surgery in a total of 173 patients (Figure 13).

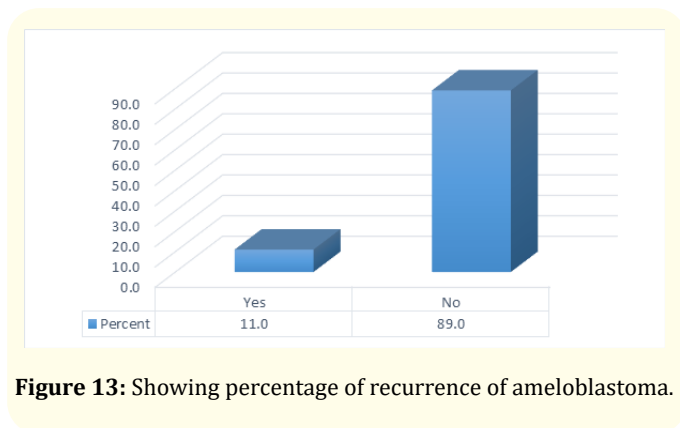
Out of 173 patients, 127 (73.4%) patients developed teeth-related problems, as it was not possible to construct a replacement while 46 (26.6%) patients received a prosthesis (Figure 14).

Reasons for lack of follow up	No. of patients	Percentage
Financial	39	30.7%
Ignorance	21	16.5%
Distance	42	33.1%
Loss of motivation	25	19.7%

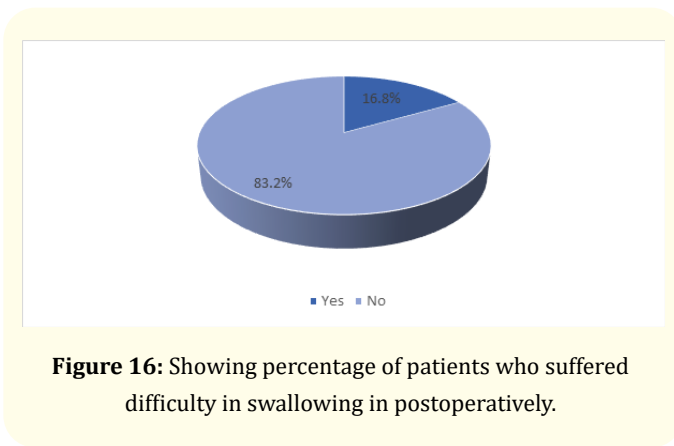
**Table 2:** Summarized causes of failure of follow ups.



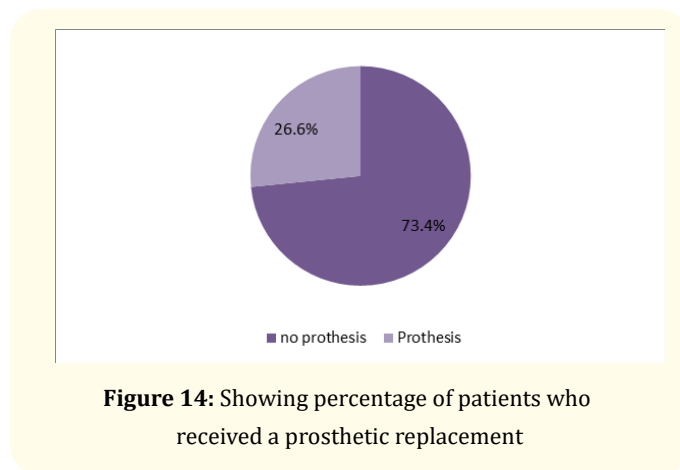
**Figure 15:** Showing percentage of patients who developed speech problems.



**Figure 13:** Showing percentage of recurrence of ameloblastoma.



**Figure 16:** Showing percentage of patients who suffered difficulty in swallowing in postoperatively.

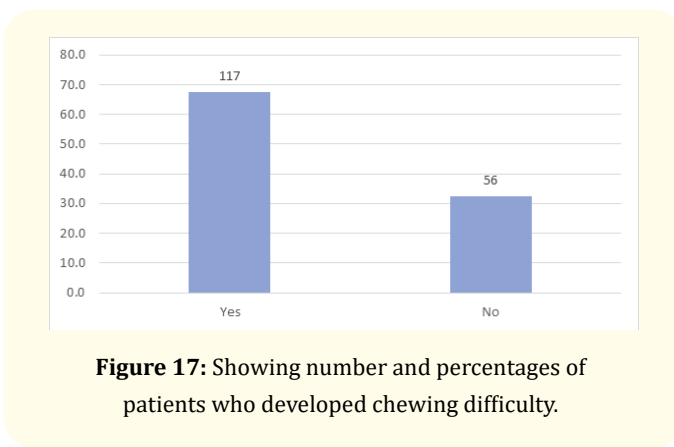


**Figure 14:** Showing percentage of patients who received a prosthetic replacement

Speech was affected in 65 (37.6%) patients while 108 (62.4%) patients didn't complain of speech changes (Figure 15).

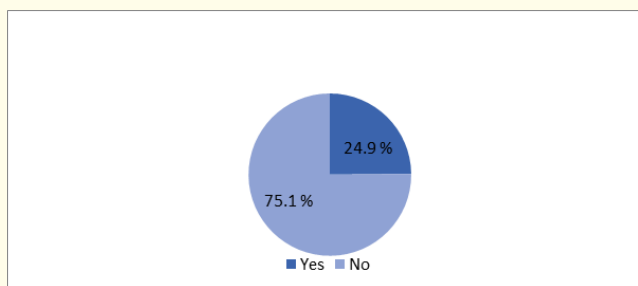
Out of 173 patients, 29 (16.8%) patients developed swallowing difficulty (Figure 16).

One hundred seventeen (67.6%) patients complained of difficulty in chewing while 56 (32.4%) patients did not complain of chewing difficulty (Figure 17).



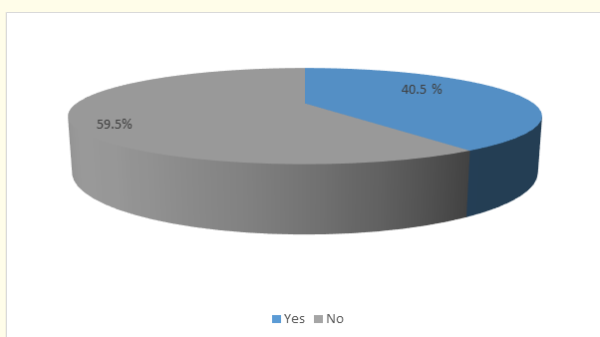
**Figure 17:** Showing number and percentages of patients who developed chewing difficulty.

Limitation of mouth opening was the complaint of 43 (24.9%) patients while 130 (75.1%) patients showed no change in the mouth opening (Figure 18).



**Figure 18:** Showing percentage of patients who developed limitation of mouth opening.

Out of 173 patients, 70 (40.5%) developed changes in their appearance, while 103 (59.5%) showed no changes in their appearance (Figure 19).



**Figure 19:** Showing reported changes in appearance.

Out of 70 patients who showed change in appearance, 43 (61.4%) patients their lesions were located posteriorly in the jaws (Table 3-6).

Gender	Unsatisfied	Satisfied	Total
Male	59	40	99
	59.6%	40.4%	
Female	55	16	71
	77.5 %	22.5 %	

**Table 3:** Cross tabulation between gender and satisfaction.

P-value = 0.014.

Age group	Unsatisfied	Satisfied	Total
Group1 (0 - 20)	0	2	2
	0.0%	100.0%	
Group2 (21- 30)	20	13	33
	60.6%	39.4%	
Group3 (31 - 40)	39	10	49
	79.6%	20.4%	
Group4 (41 - 50)	34	7	41
	82.9%	17.1%	
Group5 (51 - 60)	13	7	20
	65.0%	35.0%	
Group6 (61 - 70)	7	10	17
	41.2%	58.8%	
Group7 (71 - 80)	1	6	7
	14.3%	85.7%	
Group8 (81 - 90)	0	1	1
	0.0%	100.0%	

**Table 4:** Cross tabulation between age groups and satisfaction.

Change in Appearance	Anterior	Posterior	Both	Total
Yes	5	43	22	70
	7.1%	61.4%	31.4%	

**Table 5:** Cross tabulation between Change of Appearance and defect location.

Depending on patients and co-patient’s expressions twenty-nine (16.8%) of the patients developed changes in psychological status from a total of 173 patients (Figure 20).

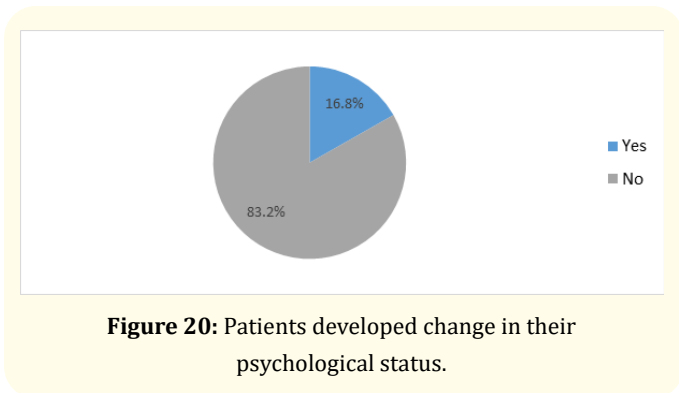
Forty-five (26%) patients reported a change in their daily activity towards the low side postoperatively while 128 (74%) patients reported no change in daily activity (Figure 21).

Thirteen (12.4%) patients lost or stopped their jobs after surgery out of a total of 105 employed patients (Figure 22).

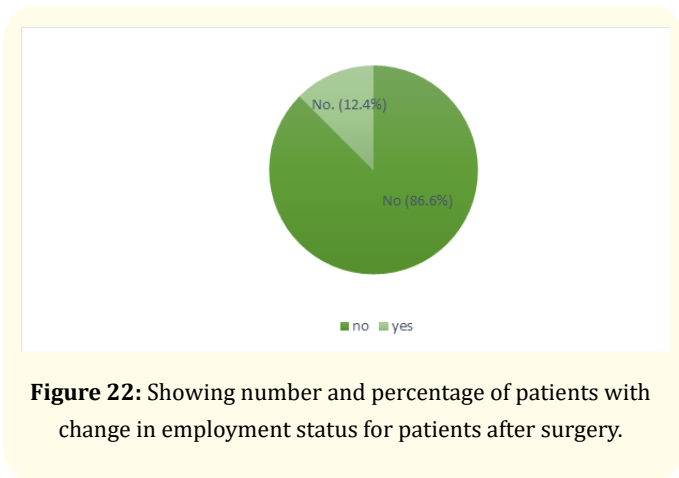
Type of treatment	Anterior	Posterior	Both	Total
Enucleation	6	33	4	43
	13.95%	76.74%	9.30%	
Marginal resection	3	17	7	27
	11.11%	62.96%	25.93%	
Segmental resection	4	32	28	64
	6.25%	50.00%	43.75%	
Hemimandibulectomy	1	35	4	40
	2.50%	87.50%	10.00%	
Hemimandibulectomy with disarticulation	0	22	4	26
	0.0%	84.62%	15.38%	
Partial maxillectomy	2	1	0	3
	66.67%	33.33%	0.0%	
Total maxillectomy	0	2	4	6
	0.0%	33.33%	66.67%	

**Table 6:** Cross tabulation between type of treatment done and lesion location.

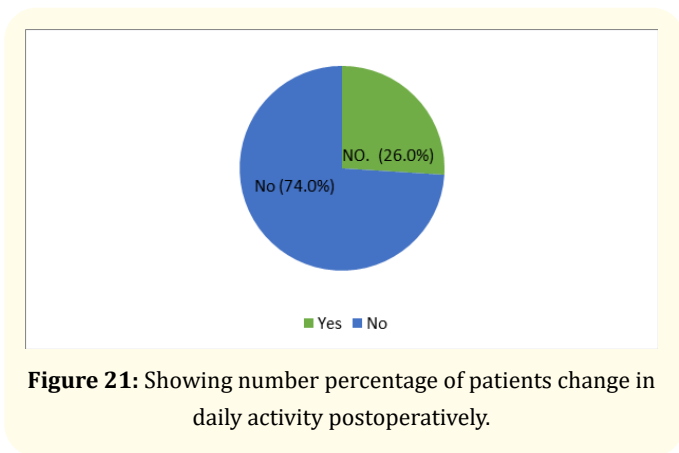
P-value = 0.00.



**Figure 20:** Patients developed change in their psychological status.



**Figure 22:** Showing number and percentage of patients with change in employment status for patients after surgery.



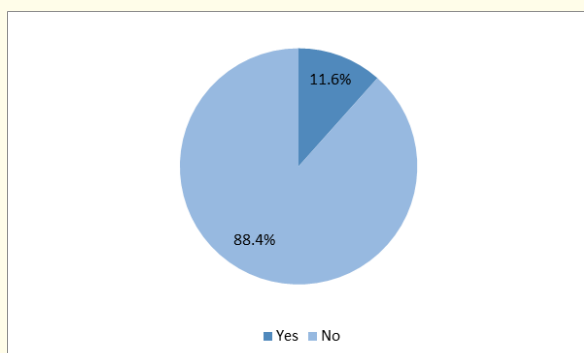
**Figure 21:** Showing number percentage of patients change in daily activity postoperatively.

From a total of 173 patients, 20 (11.6%) patients developed negative changes in their socioeconomic status (Figure 23).

**Discussion**

Ameloblastoma is the most common clinically significant odontogenic tumor. Its relative frequency equals the combined frequency of all other odontogenic tumors, excluding odontomas [2]. In Sudan of all odontogenic tumors ameloblastoma was the most frequent (64.7%) [26]. Ameloblastomas are tumors of odontogenic





**Figure 23:** Changes in Socioeconomic status of patients after surgery for ameloblastoma.

epithelial origin. Are slow-growing, locally invasive tumors that run a benign course in most of cases. They occur in three different clinicoradiographic situations, which deserve separate consideration because of differing therapeutic entities and prognosis: Conventional solid/Multicystic (about 86% of all cases). Unicystic (about 13% of all cases) and Peripheral (extraosseous) (about 1% of all cases). Ameloblastoma is chiefly a lesion of adults. It occurs predominantly in the fourth and fifth decades of life, and the age range is very broad, extending from childhood to late adulthood the rare lesions occurring in children are usually cystic and appear clinically as odontogenic cysts. There appears to be no gender predilection for this tumor. Ameloblastomas may occur anywhere in the mandible or maxilla, although the mandibular molar-ramus area is the most common site. Lesions usually are asymptomatic and are discovered during routine radiographic examination or because of asymptomatic jaw expansion. Occasionally, tooth movement or malocclusion may be the initial presenting sign. Despite the increasing literature about odontogenic tumors, specifically ameloblastoma, little is known about the demographic data and treatment outcomes of these tumors in Sudan. The importance of such study in this filed reflect from the fact that there is considerable presentation of patients with ameloblastoma increased need to know the demographic data and treatment outcomes for such patients. The demographic data of the presently reported patients are more or less in accordance with those obtained from the literature. Regarding the gender predilection in the present study, males were most commonly affected with males to female's ratio 1.4:1, in

consistence with Akinosi JO., *et al*, kameyama YS., *et al*, Siar CH., *et al*, Fomete B., *et al*, Adekeye EO, Arotiba JT., *et al*, Shoor H., *et al*. and Ali M., *et al*. [6,8,10,13,16,18,20,26]. Third and fourth decades being the most common (29.7% and 22.5%) occurrence in this study with a mean age of 49 years which is in agreement with Akinosi JO., *et al*, Kameyama YS., *et al*, Ledesma C., *et al*, Adekeye EO, Shoor H., *et al*, Arotiba JT., *et al*. and Ali M., *et al*. [6,8,9,16,18,20,26]. In Maxillo-mandibular distribution of ameloblastoma, the mandible was the mostly involved (94.8%) in consistence with Small LA, Akinosi JO., *et al*, Kameyama YS., *et al*, Ledesma C., *et al*, Shoor H., *et al*, Adekeye., *et al*, Arotiba JT., *et al*. and Ali M., *et al*. [5,6,8,9,16,18,20,26]. In the present study there is no significant difference in lesion affection between the left and right side of the jaws in agreement with Singh T., *et al*. [29] and in contrast to Kameyama YS., *et al*. and Olitan AA., *et al*. who found that the left side was most affected [8,19]. Prediction of ameloblastoma was mainly to the, molar - premolar area in the present study which is similar to the findings of Akinosi J., *et al*, Kameyama Y., *et al*, Ali M., *et al*. and Singh T., *et al*. [6,8,26,29]. In the present study the right side of the maxilla was mostly affected in contrast to the mandible where the left side is most commonly involved. In this study multilocular radiolucency was the most common radiographic presentation (72.7%) in accordance with Small LA, Siar H., *et al*, Butt FMA., *et al*, Adekeye EO, Arotiba JT., *et al*. and Singh T., *et al*. [4,10,12,18,20,29] and in contrast with Shoor H., *et al*. and Olitan AA., *et al*. studies [16,19]. In the present study (86%) of lesions associated with root resorption while (14%) associated with unerupted teeth compared to (11%) in Butt FMA., *et al*. report [12] No single standard type of therapy can be advocated for patients with ameloblastoma. Rather, each case should be judged on its own merits. Prime considerations are whether the lesion is solid, cystic, extraosseous, or malignant, and its location. Although the optimal surgical approach remains controversial, it remains the favored treatment modality. Conservative surgical options in this study, were utilized in about (20.5%) of cases. Various forms of resection were performed for the rest. Segmental resection was the most common treatment performed (30%). Treatment form was selected based on the extent of the lesion, histopathological subtype, patient factors and available facilities which is nearly equivalent to study done by Olitan AA., *et al*. [19] the ultimate goal of the treatment of ameloblastoma is restoration of function and aesthetics. Most patients who were managed by surgical resection also un-

derwent a reconstructive procedure either immediately (90%) or delayed (10%). Three maxillary defects were reconstructed with soft tissue flaps (temporalis muscle flap). For mandibular defects, 90 cases were reconstructed with titanium reconstructive plates only without an osseous component. 40 cases reconstructed with hard tissue component, (the iliac crest) was the most common source of bone graft used and in 35 cases. Fibula graft was used in 3 cases and costochondral graft which was the least bone graft used in 2 cases, all were free grafts. These results are nearly similar to the findings of the study done by Singh T, *et al.* [29] In the present study follow up periods of ameloblastoma patients divided in a regular annual manner. Nineteen (11%) patients developed recurrence after surgery, 14 of these cases underwent conservative surgery (10 cases unicystic and 4 cases solid), 2 cases of unicystic ameloblastoma operated by marginal resection and 3 cases of solid ameloblastoma operated by marginal resection. Such findings support the opinion of radical surgery as treatment modality versus conservative one (Gardener DG and Vohra FA, *et al.* [7,24]). However, the rate of recurrence is considered low if compared with Kim SG, *et al.* (21.1%) Nakamura N, *et al.* (19%), Hasegawa T, *et al.* (48.7%) and Singh T, *et al.* (14.7%) [22,23,25,29]. In this study (73.4%) patients developed teeth-related problems, as it was not possible to construct a replacement. Speech was affected in (37.6%) patients, (16.8%) patients developed swallowing difficulty and (67.6%) patients complained of difficulty in chewing, Limitation of mouth opening was the complaint of (24.9%) patients. Seventy of our patients showed change in appearance, 43 of them having their lesions located posteriorly in in the jaws however they were satisfied even with large lesions that left large defects. In the present study (26%) of patients reported a change in their daily activity towards the low side postoperatively, while (12.4%) of patients lost or stopped their jobs after surgery and (11.6%) of patients developed negative changes in their socioeconomic status. Regarding family relations, eleven patients (5.8%) got divorced due to the disease, 6 were females and 5 were males.

### Conclusion

This study was done in Sudanese population to raise the knowledge about ameloblastoma demographic data and treatment outcomes. The study revealed that the ameloblastoma most frequently occurred in third and fourth decades of life a slight male's predilection than females. Study revealed that the posterior mandible was the most frequently affected with left side prediction. In the maxilla

affection more at right side opposite to the mandible. In this study whatever the type of ameloblastoma radical surgeries showed less recurrence rate rather than conservative one despite the disfigurement that may result. This study showed the importance of long term post-operative follow up (clinical and radiographic) as a crucial step to ensure prognosis and prevent recurrence. Decreasing ability strength, ignorance and distance prevented patients from coming for further reconstruction and prosthetics replacement. Study showed that there was direct impact of ameloblastoma disease and treatment provided on patient's quality of life. Limitations of this study were manifested because of incomplete patient's files and records, lack of some patient's cooperation and failure to contact directly with those who live distance.

### Conflict of Interest

The authors declare that there is no conflict of interest.

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