Heliyon 9 (2023) e16243

Contents lists available at ScienceDirect

Heliyon

journal homepage: www.cell.com/heliyon

Research article

Ameloblastoma of the jaws in adult: A retrospective review of local recurrent lesions based on the resection margin in the adjacent apparent healthy tissues

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ARTICLE INFO

Keywords:
Ameloblastoma
Jaws
Resection
Margin
Recurrence

ABSTRACT

Background: The surgical treatment of ameloblastoma of the jaws remains contentious due to the variable recurrence rate amongst its variants, the tumor's local invasive behavior, and the lack of consensus among surgeons concerning the extent of resection in the contiguous healthy tissues.
Objective: To determine the recurrence rate of ameloblastoma and its association with the resection margins.
Materials and methods: This is a retrospective cohort study of the medical records of patients who underwent surgical resection of the jaws as the primary modality of treatment for ameloblastoma. Clinical data over the 26 years were analyzed for age, gender, site of the lesion, size, radiographic appearance, histopathological sub-type, and the incidence of recurrence post-treatment. Descriptive and bivariate statistics were computed.
Results: A retrospective audit of 234 cases was included in the study that was typical (solid/multicystic) ameloblastoma. The age of patients ranged from 20 to 66 years with a mean age of 33.4 ± 9.6 years, and a male-to-female ratio of 1.2: 1 ($P = 0.52$). The follicular and plexiform types accounted for the majority of histopathological variants (89.8%; $P = 0.000$). Overall, 6.8% of cases relapsed after the initial primary surgery. The rate of recurrence was high with a resection margin of 1.0 or 1.5 cm than 2.0 cm ($P = 0.001$). No case of recurrence was seen with a resection margin of 2.5 cm margin.
Conclusion: A low recurrence rate of 6.8% was noted in our series of cases. A wide 2.5 cm resection margin is recommended in the adjacent healthy tissues.

1. Introduction

Ameloblastoma is an odontogenic epithelial tumor that can originate from an embryonic enamel organ, remnants of the dental lamina, epithelium of dentigerous cysts, or the basal cells of the epithelium of the oral mucosa [1,2]. It remains innocuous at its early stage [1], until it reaches a large size to cause expansion and perforation of the adjacent tissues with a significant facial deformity [3,4]. Numerous risk factors can contribute to the genesis of ameloblastoma, such as chronic inflammation, chemicals, human papilloma

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https://doi.org/10.1016/j.heliyon.2023.e16243
Received 17 October 2022; Received in revised form 26 April 2023; Accepted 10 May 2023
Available online 13 May 2023

virus infections, nutritional deficiencies, poor oral hygiene, and distinct genetic polymorphisms [5].

The World Health Organization (WHO) 2017 classification included conventional ameloblastoma, unicystic ameloblastoma, extraosseous/peripheral ameloblastoma, and metastasizing (malignant) ameloblastoma. The classification amalgamates the solid/polycystic and desmoplastic types into conventional ameloblastoma, while the unicystic ameloblastomas comprise intraluminal, luminal, and mural variants [6]. In 2022, the WHO restructured the classification of ameloblastoma and included adenoid ameloblastoma, with novel histological features comprising of ameloblastoma-like constituents, tubular, cribriform patterns, and helical cell clusters, with or without dentin-like structures [7]. Since it has no biological significance and may be confused with unicystic ameloblastoma, the term "solid/multi-cystic" for typical ameloblastoma was removed from the classification [6]. Desmoplastic ameloblastoma is reclassified as a histopathological variant rather than a clinicopathologic variant. Central ameloblastoma is regarded as a group of intra-osseous ameloblastomas. Follicular, plexiform, acanthomatous, desmoplastic, granular cell, and basal cell patterns are the histopathological variants of ameloblastoma. All of these histological types can be seen as a single lesion, combinations of two or more histopathological variants, or hybrid lesions with other odontogenic neoplasms. Metastasis of histopathological benign ameloblastoma is regarded as "metastasizing ameloblastoma."

A slow progressive buccolingual jaw expansion with an egg-crackling sensation and tooth mobility are some of its hallmarks [5,8,9]. Ameloblastoma accounts for about 1.3% of all jaw tumors and cysts and is the second most common odontogenic tumor accounting for 11%–18% of all odontogenic neoplasms [3,4,10,11].

Clinical, radiologic, and histopathological findings are used to formulate a diagnosis of ameloblastoma [11–14]. It presents with diverse radiological appearances including unilocular radiolucency, radiopacity, and mixed radiopacity/radiolucency, but a multilocular radiolucency showing a "soap bubble" appearance associated with the resorption of tooth roots is a characteristic hallmark [14,15]. Treatment of these tumors remains contentious due to their local invasive behavior, high rate of recurrence, and lack of consensus amongst the surgeons regarding the extent of resection in the adjacent apparent healthy tissues [12,13,16]. The therapeutic challenge is how to address the therapeutic failure with minimal morbidity and complications. Towards achieving this goal, the surgeon is required to assess the patient's medical history; location, size, macroscopic and histopathological subtypes of the ameloblastoma, and patient's compliance towards a regular follow-up [11,17]. Nonetheless, other treatment modalities including adjunct therapies have been reported to be effective, the preferred treatment is jaw resection [17–19]. Majority of studies have categorized surgical procedures as conservative or radical, none of the studies have explored the link between the recurrence rates to the resection margin in the adjacent apparent healthy tissues. The objective of this study was to determine the recurrence rate of ameloblastoma and evaluate its association with the resection margins in the adjacent apparent healthy tissues. The Null hypothesis is that the wider the resection margin in the adjacent apparent healthy tissues, the less the likelihood of recurrence.

2. Materials and Methods

A retrospective audit of medical records of patients diagnosed with ameloblastoma who underwent jaw resection as the primary modality of treatment. The records were accessed from University of Calabar Teaching Hospital Calabar, Nigeria, Department of Oral and maxillofacial surgery, from January 1995 to December 2020.

The objective was to determine the recurrence rate of ameloblastoma and to evaluate its association with the resection margin in the contiguous healthy tissues. This study aims to evaluate the status of resection margins with the risk of recurrence. The study was provided a waiver from ethical clearance by the Regional Ethics Committee of the institution because of its retrospective design and on the condition that the research data are not shared. Written informed consent was obtained from each patient before treatment.

According to the WHO, an adult is a person older than 19 years of age unless national law delimits an earlier age [20]. Clinical, radiologic, and histopathological findings were used to make a diagnosis of ameloblastoma of the jaws. All the ameloblastomas that were presented were also diagnosed and classified according to WHO criteria [6,7]. Cases included in the study were patients 20 years of age and above, who had jaw resection as the treatment of ameloblastoma during the study period, completed medical records, and kept a minimum of 5 years of follow-up appointments after the initial surgery. The cases excluded, were patients below 20 years of age who had jaw resections; those within complete medical records, and failed to attend postoperative follow-up visits for a minimum period of 5 years after the initial surgery.

For the mandibular ameloblastomas, the site of occurrence was categorized into anterior (incisal–canine), body (premolar–molar region), posterior (distal to the third molar), and bilateral (across midline) regions [2]. The posterior area also included the angle, ramus, coronoid and condylar processes. For maxillary tumors, the site was subdivided into anterior (incisal– canine) and posterior (distal to canine) regions [2]. Any tumor affecting two or more sites was assigned to the region approximating the center of the lesion [2].

Conventional radiographs were used to diagnose ameloblastoma and its recurrence was assessed using posteroanterior (PA) of the skull, two (right and left) oblique laterals of the mandible, Submentovertex, lateral skull, and occipitomental views. The radiographs were certified as standardized by the Association of Radiologists in Nigeria (Calabar branch). The images were evaluated by three examiners: an oral and maxillofacial surgeon who regularly deals with orofacial oncology, a senior resident in oral and maxillofacial surgery oncology, and a radiologist with expertise in oncology. Although computed tomography (CT) and magnetic resonance imaging (MRI) are the gold standards in the radiological evaluation of ameloblastoma, particularly in the maxillary region, non-availability and unaffordability for the patient precluded their routine use for the diagnosis of ameloblastoma in the study institution during the period studied.

The surgeries were done by an oral and maxillofacial surgeon who regularly performs ablative procedures in the orofacial region. During the surgical procedures, the length between the limit of the tumor was determined clinically as well as radiologically and the

resection margins in the adjacent apparent healthy tissues were recorded in centimeters (cm) using a metallic ruler and tape: 1.0, 1.5, 2.0, and 2.5. The affected soft tissue margins were also excised with a margin of safety similar to that of the affected bony tissues or where there is a margin of doubt, a frozen section was also done to confirm the margin status. The data were collected using a pro-forma questionnaire prepared for the study. Clinical records of the patients over the 26-year study period were analyzed for age, gender, site of the lesion, size, radiographic appearance, histopathological sub-type, and recurrence after treatment. A retrospective chart review was then utilized for the compilation of data of interest.

The primary outcome variable was the rate of recurrence of the tumor after jaw resection during follow-up appointments. The primary predictor of the recurrence of lesion after treatment was the presence of ameloblastoma of the jaws and the length of the resection margin. Other independent predictor variables were age, gender, site of the lesion, size, radiographic appearance, and histopathological subtypes.

Follow-up was done by clinical and radiographic examinations. A successful outcome of treatment was determined if the patient is free of symptoms and signs of the disease locally at the surgical and/or distant sites. Recurrent ameloblastoma was defined as relapse after the initial surgery with symptoms and signs of the disease locally at the surgical and/or distant sites. Recurrences of tumor were diagnosed based on the patient's complaints and clinical and radiological evaluation during follow-up reviews.

The data obtained were analyzed with EPI Info 7, 2012 software (US Centers for Disease Control and Prevention, Atlanta, GA, USA). The Chi-square and Fisher's Exact tests were used to determine the differences between two groups and the Kruskal-Wallis test was for differences among three or more groups. *P*-values <0.05 were considered significant.

3. Results

The Null hypothesis was investigated with Fisher's exact test using the initial data provided for the study and was found to be true (P = 0.02, significant). In this study, 234 patients with ameloblastoma of the mandible and maxilla were evaluated and each subject presented with only one type of histopathological variant. Clinicopathological, all the cases that presented were typical (solid/multicystic) ameloblastoma whereas unicystic ameloblastoma, peripheral and malignant forms were not recorded.

Table 1 shows the age and gender distribution of subjects. The age of patients ranged from 20 to 66 years with mean age, of 33.4 ± 9.6 years. A majority (n = 221, 94.4%) of the patients were in the age range of 20–49 years (Table 1, P = 0.000). The males outnumbered females in all age categories with a male-to-female ratio of 1.2: 1 (Table 1, P = 0.52). The distribution of clinical sizes of ameloblastoma according to the histopathological types is shown in Table 2. A majority (n = 141, 60.2%) of the tumors measured 5.1–9.0 cm (P = 0.03). As shown in Tables 2 and 3, the follicular and plexiform histopathological variants accounted for the majority (n = 210, 89.8%; P = 0.000). Table 3 also shows a majority of the cases were located in the mandible than in the maxilla (P = 0.000). In addition, the majority of the lesions were located in the posterior aspect of the mandible (body, ramus) and posterior maxilla (P = 0.000, Table 3).

The distribution of radiological appearances of the tumor is shown in Table 4. This was statistically significant (P = 0.001) in favor of multilocular radiolucency which formed the majority. The mixed type comprised both radiolucency/radiopacity radiological appearances. The resections done in the mandible are segmental (n = 163, 69.6%), marginal (n = 24, 10.3%), hemi-(n = 11, 4.7%) and sub-total (n = 11, 4.7%) mandibulectomies. In the case of maxillary lesions: partial (n = 15, 6.4%), hemi-(n = 7, 3.0%) and sub-total (n = 3, 1.3%) maxillectomies. The distribution of the histopathological types according to the resection margins is shown in Table 5. In the majority of the 234 patients (n = 162, 69.3%), resection margins of 1.5 cm and 2.0 cm were predominant (P = 0.02).

Overall, 16 (6.8%) cases relapsed after initial surgery, and these were 12 (5.1%) in the mandible and 4 (1.7%) in the maxilla. For the mandibular lesions, one (0.4%) was located in the anterior region, 2 (0.8) in the body, 3 (1.3) bilateral, and 6 (2.6) posterior, Fig. [1 (A-D)]. In the maxilla, 3 (1.3) cases were located posteriorly and one (0.4%) anteriorly. These recurrences occurred between 3.4 and 7.8 years (mean 3.9 ± 0.47 years) after the initial surgery. Also, 3 (1.3%) cases occurred in females while males accounted for the rest, 13 (5.6%), and their ages ranged from 28 to 47 years (mean 39.4 ± 1.6 years).

Table 6 shows that after initial surgery, desmoplastic, basal cell, and acanthomatous variants are more likely to recur than follicular, plexiform, and granular cell ameloblastoma (P = 0.03). The distribution of the recurrent lesions is shown in Table 7. The wider the resection margin, the lower the frequency of recurrence, and recurrent tumors occurred more frequently if the resection

Table 1
Distribution of age and gender of the 234 patients that presented with ameloblastoma.

Age groups (years)	Gender		Total
	Male	Female	
	n (%)	n (%)	n (%)
20–29	36 (15.4)	30 (12.8)	66 (28.2)
30–39	48 (20.5)	45 (19.2)	93 (39.7)
40–49	35 (15.0)	27 (11.5)	62 (26.5)
50–59	5 (2.2)	4 (1.7)	9 (3.9)
60–69	3 (1.3)	1 (0.4)	4 (1.7)
Total	127 (54.4)	107 (45.6)	234 (100.0)

Age: $\chi^2 = 116.44$, df = 8, P = 0.000.
Gender: $\chi^2 = 116.44$, df = 8, P = 0.52.

Table 2
Distribution of sizes of ameloblastoma in the 234 patients according to the histopathological types.

Types of lesion	Sizes of lesion in centimeter (cm)						Total (%)	
	3.1–5	5.1–7	7.1–9	9.1–11	11.1–13	13.1–15		
	n	n	n	n	n	n		P
Follicular	14	2	37	12	10	7	109 (46.6)	0.03
Plexiform	11	31	33	16	7	3	101 (43.2)	
Desmoplastic	0	1	5	0	1	1	8 (3.4)	
Acanthomatous	0	2	0	3	0	2	7 (3.0)	
Basal cell	1	1	2	0	1	0	5 (2.1)	
Granular cell	0	0	0	3	1	0	4 (1.7)	
Total	26 (11.1)	64 (27.3)	77 (32.9)	34 (14.5)	20 (8.6)	13 (5.6)	234 (100.0)	

Table 3
Distribution of the 234 histopathological variants according to the site of occurrence.

Site	Histopathological variants						Total	
	Follicular	Plexiform	Desmoplastic	Acanthomatous	Basal	Granular		
	n	n	n	n	n	n	n (%)	P
Mandible							**209(89.3)**	0.000
Posterior	46	44	0	1	1	0	92 (39.3)	
Body	39	36	2	2	2	2	83 (35.5)	
Bilateral	13	9	0	0	2	2	26 (11.1)	
Anterior	3	1	3	1	0	0	8 (3.4)	
Maxilla							25 (10.7)	
Posterior	7	9	1	2	0	0	19 (8.1)	
Anterior	1	2	2	1	0	0	6 (2.6)	
Total	109	101	8	7	5	4	234 (100.0)	

Table 4
The distribution of radiological appearances of ameloblastoma according to the histopathological variants.

Type of lesion	Radiological appearances				
	Radiopacity	Mixed	Multilocular	Total	
	n	n	n	n (%)	P
Follicular	1	11	97	109 (46.6)	0.001
Plexiform	1	12	88	101 (43.2)	
Desmoplastic	3	3	2	8 (3.4)	
Acanthomatous	1	2	4	7 (3.0)	
Basal cell	0	1	4	5 (2.1)	
Granular cell	0	1	3	4 (1.7)	
Total	6 (2.6)	30 (12.8)	198 (84.6)	234 (100.0)	

Table 5
Distribution of the histopathological types according to the length of resection margin.

Type of lesion	Length of margin of safety (cm)					
	1.0	1.5	2.0	2.5	Total	
	n (%)	n (%)	n (%)	n (%)	n (%)	P
Follicular	23 (9.8)	39 (16.7)	36 (15.4)	11 (4.7)	109 (46.6)	0.02
Plexiform	17 (7.3)	41 (17.5)	27 (11.5)	16 (6.8)	101 (43.2)	
Desmoplastic	0 (0)	3 (1.3)	3 (1.3)	2 (0.9)	8 (3.4)	
Acanthomatous	0 (0)	2 (0.9)	4 (1.7)	1 (0.4)	7 (3.0)	
Basal cell	1 (0.4)	1 (0.4)	3 (1.3)	0 (0)	5 (2.1)	
Granular cell	0 (0)	1 (0.4)	2 (0.9)	1 (0.4)	4 (1.7)	
Total	41 (17.5)	87 (37.2)	75 (32.1)	31 (13.2)	234 (100.0)	

margin was either 1.0 or 1.5 cm than 2.0 cm (Table 7, P = 0.001). Furthermore, no tumor treated with a 2.5 cm resection margin relapsed including the entire granular cell variant (Tables 6 and 7).

Radiologically, all the cases that recurred showed multilocular radiolucency. These recurrent tumors were successfully managed by

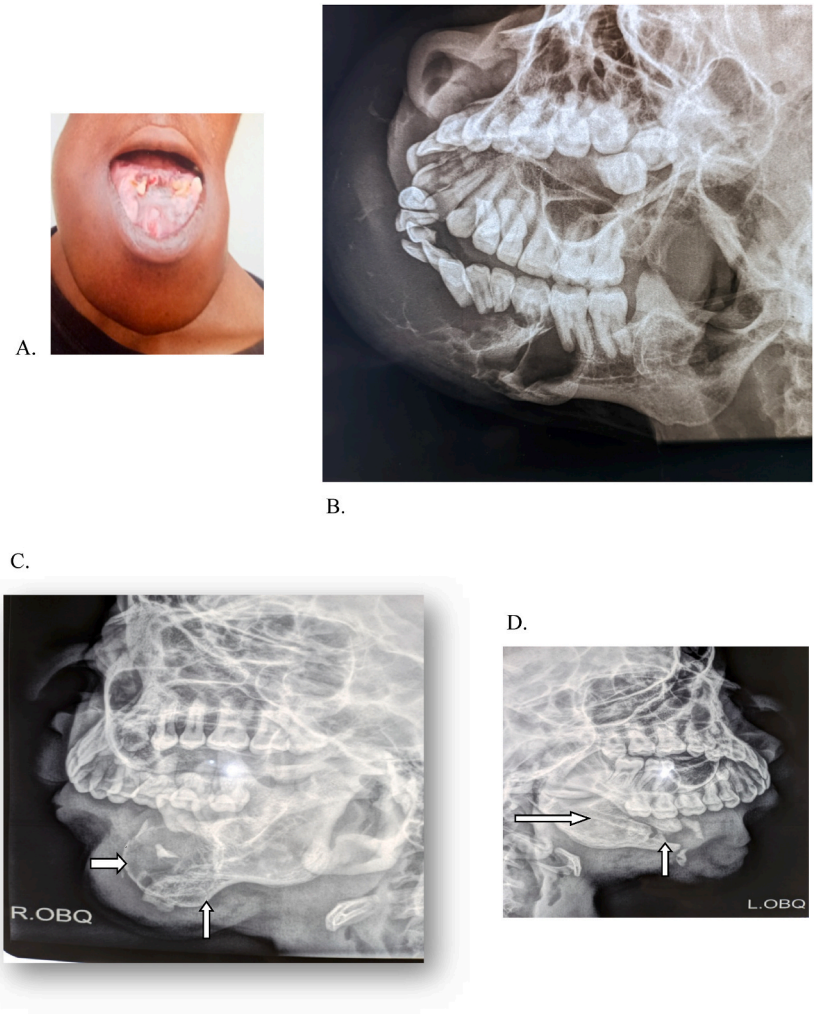

Fig. 1. A. shows a 25-year old male who presented with follicular ameloblastoma of the mandible noticed 3.2 years prior to presentation. **B.** Lateral oblique view of the mandible of the patient shows the lesion as a multilocular radiolucency with a "soap bubble" appearance associated with the resorption of roots of the affected dentitions. **C.** Lateral oblique mandibular view of the same patient showed the lesion recurred on the right side of the remnant body of the mandible 3.4 years after subtotal mandibulectomy with an initial resection margin of 1.5 cm. **D.** 6.5 years after secondary surgery to excise the remnant mandible with resection margin of 2.5 cm, there has not been recurrence of the lesion rather bone regeneration was observed on the left side.

Table 6
Distribution of the recurrent rates among the various histopathological types.

Type of lesion	No recurrence	Recurred	Total	
	n (%)	n (%)	n (%)	P
Follicular	102 (93.6)	7 (6.4)	109 (100.0)	0.03
Plexiform	96 (95.0)	5 (5.0)	101 (100.0)	
Desmoplasti	6 (75.0)	2 (25.0)	8 (100.0)	
Acanthomatous	6 (85.7)	1 (14.3)	7 (100.0)	
Basal cell	4 (80.0)	1 (20.0)	5 (100.0)	
Granular cell	4 (100.0)	0 (0)	4 (100.0)	
Total	218 (93.2)	16 (6.8)	234 (100.0)	

extending the resection margins to 2.5 cm during the second surgery. No recurrence has been recorded after the second surgery. Overall, the follow-up of patients ranged from 5.1 to 18.3 years (mean 11.5 ± 3.8 years) after the initial surgery. In the recurrent cases, follow-up ranged from 3.7 to 8.5 years (mean 6.1 ± 0.4 years) after the second surgery. During these periods, there was no evidence or record of any other recurrence except those reported.

Table 7
Distribution of recurrent lesions according to the length of resection margin and histopathological variants.

Type of lesion	Length (cm) of resection margin during initial surgery					
	1.0	1.5	2.0	2.5	Total	
	n (%)	n (%)	n (%)	n (%)	n (%)	P
Follicular	4 (25.0)	2 (12.5)	1 (6.25)	0 (0)	7 (43.75)	0.001
Plexiform	2 (12.5)	2 (12.5)	1 (6.25)	0 (0)	5 (31.25)	
Desmoplastic	1 (6.25)	1 (6.25)	0 (0)	0 (0)	2 (12.5)	
Acanthomatous	1 (6.25)	0 (0)	0 (0)	0 (0)	1 (6.25)	
Basal cell	1 (6.25)	0 (0)	0 (0)	0 (0)	1 (6.25)	
Total	9 (56.25)	5 (31.25)	2 (12.5)	0 (0)	16 (100.0)	

4. Discussion

Ameloblastoma is a locally aggressive, benign, and odontogenic tumour. The World Health Organization has recategorized the types of ameloblastomas based on the effect of the tumour on treatment and the recent progression in molecular genetics.

Radical approaches for ameloblastoma may offset the recurrence rate; however, the associated facial deformity has a prodigious influence on the physiology of the orofacial apparatus and the psychology of patients, exclusively in younger patients. In contrast, conservative treatments such as enucleation have a high recurrence rate with an added risk of malignant transformation and distant metastasis following repeated tumor manipulation.

The current study reveals a low recurrence rate of 6.8% by deploying radical approaches with wide resection margins. The risk of recurrence was more common if the resection margin in the adjacent apparent healthy tissues was either 1.0 or 1.5 cm than a wide 2.0 cm margin. In addition, there was no recurrence seen in patients who had a 2.5 cm resection margin. The recurrence rate varies from study to study depending particularly on the study design. The rate in this study is higher than the 4.5% reported by Hong et al. [21], but lower than several others [2,4,22,23]. It is possible that the recurrence rate might increase in the future as the optimum follow-up period in this study was 18.3 years. Some researchers have reported recurrences several decades after the initial treatment which reaffirms the insidious biological behavior of this tumor and reemphasizes the need for long-term follow-up [1,8,11,24].

While jaw resection has been established as a preferred treatment for ameloblastoma, the dispute of optimum resection margins has always been questioned by different investigators. Some researchers support the resection margin to be at least 1–2 cm beyond the radiological limit of the tumor to ensure that all micro-lesions i.e. microscopic pseudo pods are completely excised [8,11,21,23]. Like the present study, recurrent ameloblastoma have been reported when the resection margin is 1–2 cm [2,4,5,25]. On the contrary, Carlson et al. [26], recommended 1–1.5 cm resection margin, and noted that researchers who recorded failure of conservative surgeries for the tumor pointed to the lack of uniform success of radical surgeries in salvaging these failures as the basis for the controversy in the literature regarding the appropriate length of the resection margin during treatment. According to the WHO, typical (solid/-multicystic variant) ameloblastoma has a greater tendency to recurrence after treatment than the other variants and emphasizes the need for wide resection margins during their treatment [6,7]. This is in agreement with the findings of this study. This study, therefore, recommends a 2.5 cm resection margin since no recurrence was recorded in the present group of patients. Recurrence is believed to be the result of several risk factors, such as tumor subtype, initial therapeutic approach, and tumor behavior [1,4,17,23,25,27].

Some researchers may argue that the 2.5 cm resection margin may cause unnecessary facial mutilation and functional impairment. It is better to put in perspective the consequences of the recurrence of the tumor and second surgery that might lead to more severe facial deformity, functional disability, and financial woes for the patient. Radical jaw resection is the only effective treatment for this type of ameloblastoma as there is a link between the resection margin and relapse of the tumor [1,8,11]. While the main objective remains to achieve a complete resection, the focus should be on how to achieve this without performing a disproportionate surgery. The treatment of typical (solid/multicystic) ameloblastoma is therefore complex, as it must be as minimally destructive as possible due to the benign nature of the lesion, but also be adequately extensive to prevent recurrence as the tumor is locally invasive and sometimes metastasize to distant tissues [21,28,29]. Much of the anatomy of the affected part of the jaws can be conserved following surgery if the patients present early for management, before the lower border, angle, condyle and coronoid processes of the mandible are eroded; the buccal and lingual/palatal cortices are perforated reducing the ensuing morbidity [1,5,30,31]. Patients' age, sex, location, and extent of the tumor should be taken into account while determining the margin of resection, and try to avoid unnecessary radical resection, which will cause less morbidity, facial deformity, and functional impairment [4,27]. Conservative treatments like enucleation, enucleation with curettage, and marsupialization including dredging are not recommended for typical (solid/multicystic) and recurrent ameloblastoma because of its local invasive nature and sometimes metastatic ability [1,3,4,28]. The prognosis of recurrence after treatment appears to be more related to the surgical planning before the evaluation of the various histological subtypes [17,32].

The histopathological patterns of typical (solid/multicystic) ameloblastoma reported in this study have been documented by other researchers [11–13,19]. The follicular type was the most commonly found in this study. This finding is consistent with previous studies [1,4,10], but contrasts the reports of Siar et al. [2], and Fregnani et al. [13], which found that the majority of patients exhibited a plexiform pattern followed by the follicular variant. As in the present study, desmoplastic, acanthomatous, basal, and granular cell types are less frequently reported [11,12,25,28]. Granular cell variant presented no relapse after treatment. This corroborates the findings of some earlier researchers [11,28], but differs from those of Reichart et al. [1], and Hong et al. [20], who recorded recurrences in their cases. In this study, desmoplastic, basal cell, and acanthomatous variants are more likely to recur after surgery than

follicular, plexiform, and granular cell ameloblastoma. Some researchers have a different view and noted that tumors with a follicular, granular, or acanthomatous growth pattern have a relatively higher likelihood of recurrence potential than desmoplastic, plexiform, and unicystic subtypes [1,2,13,19,25].

The age, gender, and site of occurrence of typical (solid/multicystic) ameloblastoma in this study are consistent with previous reports [1,5,11,12,25]. As with the cases in this study, approximately 80–90% of ameloblastomas occur in the mandible, usually in the body and posterior regions [4,5,12]. Posterior mandibular or maxillary tumor sites require wider resection margins than tumors of the anterior mandibular or maxillary regions due to the higher risk of local invasion and more difficult secondary surgical procedures in case there is recurrence [9,12,27,28]. Multilocular radiolucencies were seen in most cases on the radiograph. This is consistent with earlier reports [10,25,33]. Radiographically, multicystic ameloblastomas sometimes present as multilocular radiolucent images in "soap bubbles" [27,28,33]. Also, other radiological images reported in this study have been documented by other researchers [12,25, 33].

Typical (solid/multicystic) ameloblastoma of the jaws remains a challenge to the oral and maxillofacial surgeon due to the possibility of its relapse and the need for aggressive resections distorting the facial appearance and oral function. This study determined the recurrent rate of typical ameloblastoma and assesses its bearing on the resection margins in the adjacent apparent healthy tissues. The recurrence is particularly high in this sub-type, and if the tumor is located posteriorly in the jaws and the resection margin is grossly inadequate. In the present study, patients presented for clinical evaluation and treatment, and the outcome of treatment yielded a low recurrence rate. A 2.5 cm resection margin is recommended to eliminate the recurrence of the tumor after the ablative procedure.

It is possible that not all adult patients afflicted with this condition in the study community presented for clinical evaluation and treatment during this period. This would have negatively affected the frequency of patients and recurrence rate. The treatment of the patients was done by different surgeons with differing surgical skills. This could as well have introduced some errors that might have affected the treatment outcome. Some patients who had a recurrence of the tumor after treatment probably failed to report back to the hospital for further management. These patients may have opted for alternative means of treatment or decided to live with the disease. This study is also limited by its retrospective design. A prospective study is recommended in the future to provide more valid data devoid of defects associated with the retrospective study. The study fails to explore the probable genes that have mutated and have contributed significantly to providing locomotion to this invasive tumor. The BRAF protein in the mitogen-activated protein kinase pathway (MAPK) has been frequently found to be mutated and thereby igniting this pathway [34].

Odontogenic tumors have overlapping morphologic features and a precise preoperative diagnosis has a significant role in planning their management strategy. Conventional radiographs form the traditional foundation for the assessment of jaw lesions. The arch-like configuration of the jaws and the superimposition of teeth impede diagnostic precision [35]. These primitive radiographs do not precisely trace the tumor size, margins, internal bony architecture, cortical perforation, and three-dimensional expansion [36]. Also, ameloblastomas demonstrate a blend of enhancing solid components and non-enhancing cystic components that is well illustrated on cross-sectional imaging. Lastly, a yardstick of the optimum resection margin and adjuvant treatment options such as targeted therapies for multi-recurrent ameloblastoma must be explored.

5. Conclusions

Ameloblastoma is a benign yet aggressive odontogenic tumor with an inherent ability to erode the hard and soft tissue and yet devoid of histological hallmarks of malignancy. The aggressive, destructive nature of this odontogenic neoplasm with recurrence despite an en-bloc resection is a surgical predicament. This study shows that the recurrence rate of typical (solid/multicystic) ameloblastoma after treatment was low, which was 6.8% of patients that presented. The recurrence was more frequent if the resection margin in the adjacent apparent healthy tissues was either 1.0 or 1.5 cm than 2.0 cm. There was no relapse in patients who had a 2.5 cm margin which must be recommended.

Author contribution statement

Charles Anyanechi: Conceived the study, designed it, did literature search, data acquisition, data analysis, manuscript preparation, editing and review. Is the guarantor and take full responsibility for the integrity of the work.

Sameep S. Shetty: Did literature search including manuscript preparation, editing and review.

Data availability statement

The authors do not have permission to share data.

Additional information

No additional information is available for this paper.

Declaration of competing interest

The authors have no conflict of interest to declare. This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors. The authors have viewed and agreed to the submission.

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