

ORIGINAL RESEARCH

Recurrence of orofacial tumours and tumour-like lesions after ablative surgery: A 15-year retrospective review of 27 cases

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Abstract

Background

With recurrence of orofacial tumours and tumour-like lesions after surgical treatment, there is increased facial deformity, and the accompanying secondary surgery leads to further facial mutilation and impaired patients' well-being. This study aimed to determine the recurrent orofacial lesions after their surgical ablation, in addition to the demographics of patients, histopathological diagnosis, site of the lesion, duration between initial surgery and recurrence, and the corrective surgery.

Methods

A retrospective analysis of the medical records and histopathological reports of patients, who presented in the Oral and Maxillofacial Unit of the institution over 15 years, was undertaken. Information on demographics, histopathological diagnosis, site of the lesion, duration between initial surgery and recurrence, and the corrective surgery were obtained.

Results

During this period, 368 patients that were treated had complete clinical data, while 27/368 (7.3%) presented with recurrent lesions. They were aged 6 to 59 years, with male-to-female distribution ratio of 1:1.3 ($P=0.3$). The benign lesions accounted for 24 (88.9%, $P=0.000$) cases, while odontogenic tumours recurred ($P=0.001$) more than the other types of lesion. The lesions were seen more often in the mandible ($P=0.001$), and the frequency of patients was highest in the 21-30 years age group ($P=0.02$). Most of the recurrent cases were seen within 2 years after the initial surgery (85.2%, $P=0.001$). Majority of the corrective surgeries were segmental mandibular resection (37.1%), hemi-mandibulectomy (25.9%) and hemi-maxillectomy (18.5%).

Conclusions

This study showed that 7.3% of the lesions recurred after initial ablative surgery. Benign lesions particularly ameloblastoma and fibromyxoma which are locally invasive constitute the majority of the cases that relapsed. A future prospective study will address the limitations of this study.

Keywords: lesions, orofacial, biopsy, recurrence

Introduction

The orofacial complex as documented by Regezi,¹ Fernandes et al.,² and Servator et al.,³ can be affected by a wide spectrum of lesions which may be benign and circumscribed, locally infiltrative or malignant. Other authors such as Adekeye et al.,⁴ Avelar et al.,⁵ and Kamulegeya and Kalyanyama⁶ stated that these lesions can cause swelling, pain, tooth displacement, resorption or loss; poor periodontal health, infection, foetor oris, paraesthesia/anaesthesia, malocclusion, facial deformity and other serious clinical consequences. There are several reports⁷⁻¹¹ from around the globe on

the recurrence of these orofacial tumours and tumour-like lesions after treatment, including studies from Nigeria and other African countries, but most of these studies did not focus on the recurrence of the lesions after initial treatment as the study's primary objective.

In our centre, during pre-operative evaluation of patients afflicted by the lesions, we rely on clinical judgment, biopsy report and plain radiographic views to determine the extent of surgery. We do not use computed tomography (CT) scan and magnetic resonance imaging (MRI) where they might be indicated to assess patients radiologically during the

pre-operative evaluation, and neither do we use intra-operative frozen section when appropriate because of unavailability of these services in our centre. These radiological and histopathological investigations aid the surgeon to determine the extent of surgery and prevent recurrence of the lesion after the ablative surgery. As recorded by Young et al.,¹² and Malik,¹³ in centers where these facilities are available and utilized, it has contributed significantly towards reduction of recurrence of the lesion after initial ablative procedure. These imaging techniques offer advantages over plain radiographs during patients' radiological evaluation before treatment as noted by Malik,¹³ and Kovisto et al.¹⁴ On the other hand, intra-operative frozen section may be required in the treatment of infiltrative and malignant neoplasm which assists the surgeon to determine their extent of spread.¹³

Kovisto et al.,¹⁴ and Anyanechi et al.,¹⁵ observed that with the recurrence of lesions after surgical treatment, apart from the resultant further facial deformity, more extensive surgery will be required for its eradication which will inadvertently adversely affect facial esthetics, function of orofacial complex, and the patients' overall well-being. Consequently with further facial mutilation due to the accompanying secondary surgeries, some authors^{16,17} observed that the economic burden on the family and society is worsened, in addition to the psychological trauma and distress undergone by the patients.

In order to ascertain the burden of non-usage of these facilities in our center among other confounding variables that might influence treatment outcome like the surgeons' competence, this study determines the recurrent orofacial lesions after their surgical ablation over a period of 15-years. It is hoped that the outcome of this study will stimulate improvement on the quality of surgical services rendered to patients that attend this tertiary health institution.

Methods

A retrospective analysis of the medical records and histopathological reports of patients who presented in the Oral and Maxillofacial Unit of the Department of Dental and Maxillofacial Surgery of the study institution from January 2002 to December 2016, a period of 15 years, was undertaken. Patients with complete data were included in the study whereas those with incomplete data were excluded. The Health Research and Ethics Committee of the institution exempted the study from ethical clearance because of its retrospective nature.

Information on demographics, histopathological diagnosis, site of the lesion, duration between initial surgery and recurrence, and the corrective surgery were obtained. The WHO 2005 criteria as documented by Barnes et al.,¹⁸ were used for the classification of odontogenic tumours. Data were recorded and analyzed using the statistical package for the social sciences for windows 17.0 (SPSS Inc, Chicago, III, USA). Descriptive statistics and test of significance were also used. A p-value of less than 0.05 was considered significant.

Results

During the period of the study, 371 patients who had jaw lesions were operated upon while 368 of them had complete clinical data for analysis, whereas 27/368 (7.3%) patients reported back with recurrent lesions. The 27 patients were aged 6 to 59 years (mean 28.7 ± 7.4), with male-to-female distribution ratio of 1:1.3, ($P=0.3$). The lesions that recurred after initial treatment were categorized into three types in Table 1. The benign lesions accounted for 24, 88.9% ($\chi^2=74.531$, $df=8$, $p=0.000$) while the rest were malignant. Odontogenic tumours occurred significantly ($\chi^2=74.531$, $df=8$, $p=0.001$; Table 1) higher than the other types of lesion. The lesions presented more often in the mandible ($P=0.001$, Table 1) than maxilla in the ratio 2:1. The frequency of patients was more in the 21-30 years age group than the other age categories ($P=0.02$, Table 1). Under non-neoplastic lesions, central giant cell granuloma (CGCG) presented more frequently ($P=0.03$). The patient who had recurrent diffuse lipomatosis of the left cheek and mandible secondary to congenital left hemifacial hypertrophy, also presented with left extra-capsular temporomandibular joint (TMJ) ankylosis after the initial surgery. The muco-epidermoid carcinoma was histologically of the high grade.

The initial treatment done for the patients are shown in table 2. The recurrent lesions occurred between 6.3 months to 5.9 years (mean 2.8 ± 1.3 years) after the initial surgery. Most of the recurrent cases presented within 2 years after treatment ($n=23$, 85.2%, $P=0.001$, Table 2). The corrective surgeries done for the patients are also shown in table 2. The patient, who had recurrent diffuse lipomatosis complicated by extra-capsular TMJ ankylosis, had the ankylosis released after the excision of the lesion and fibrous band. Those who had malignant lesions also received radiotherapy treatment post-surgery when they were referred to other centres.

The follow-up of the patients after the corrective surgery ranged from 3.5-11.6 years (mean 5.6 ± 3.8). The patient who had high grade muco-epidermoid carcinoma was lost to follow-up after 4.2 years.

Discussion

This study showed that 7.3% of the lesions recurred after the initial surgical treatment, and these were classified as odontogenic, non-neoplastic, non-odontogenic and malignant occurring significantly more often in the mandible and in the 21-30 years age groups. As most of the lesions that recurred were locally infiltrative, it may be that the surgeons' could not get beyond the microscopic limit of the lesions to carry out adequate treatment because they were deceitfully guided by the macroscopic extent, and the appearance of the lesions on plain radiographs. However, it is difficult to compare this recurrent rate with those of earlier studies because of dissimilarities in the study designs like the type of lesions considered, age of the patients, study duration, treatment modalities used, competence of the surgeons, and period of follow-up among other denominators that might have been used to calculate the previous rates. Furthermore, as noted by Arotiba et al.,¹⁹ the rate of recurrence in this study may be

Table 1. Distribution of the age and gender of patients according to the type and site of lesions.

Type of lesion	Total, n (%)	Site n		Sex n		Age group, years n					
		Mandible	Maxilla	Male	Female	0-10	11-20	21-30	31-40	41-50	51-60
Odontogenic	16 (59.3)										
Follicular ameloblastoma	7	6	1	2	5	0	1	2	2	2	0
Odontogenic fibromyxoma	6	4	2	4	2	0	0	3	1	1	1
Plexiform ameloblastoma	2	1	1	1	1	0	0	0	0	1	1
KCOT	1	1	0	0	1	0	0	0	0	1	0
Non-neoplastic	6 (22.2)										
CGCG	4	3	1	3	1	0	0	2	0	0	2
Fibrous dysplasia	2	0	2	0	2	1	0	1	0	0	0
Non-odontogenic	2 (7.4)										
Ossifying fibroma	1	0	1	0	1	0	1	0	0	0	0
Diffuse lipomatosis	1	1	0	1	0	1	0	0	0	0	0
Malignant tumors	3 (11.1)										
MEC	1	1	0	1	0	0	0	0	1	0	0
Fibrous histiocytoma	1	1	0	0	1	0	0	0	1	0	0
Osteogenic sarcoma	1	0	1	0	1	0	0	1	0	0	0
Total	27 (100.0)	18 (66.7)	9 (33.3)	12 (44.4)	15 (55.6)	2 (7.4)	2 (7.4)	9 (33.3)	5 (18.5)	5 (18.5)	4 (14.8)

KCOT, keratocystic odontogenic tumour; CGCG, central giant cell granuloma; MEC, mucoepidermoid carcinoma

Benign lesions: $X^2=74.531$, $df=8$, $p=0.000$; odontogenic tumours: $X^2=74.531$, $df=8$, $p=0.001$

underestimated as the follow-up period was short and recurrences have occurred 30 years after initial surgical treatment of some of the lesions identified in this study.

The recurrent odontogenic tumours were significantly more common than the other lesions that presented, and this may be attributed to their being commoner in occurrence and do exhibit locally invasive behavior as reported by Kamulegeya and Kalyanyama⁶ and Parkins et al.⁷ Similar higher prevalence has been reported by other researchers.^{7, 20, 21} Ameloblastoma was the most common lesion in this group. The rate of recurrence of ameloblastoma which usually range from 0.6 to 46.4% depending on the study depends on the size of the lesion and site, histological type and the extent of surgical resection carried out during treatment as shown by several authors.^{13, 22, 23} Malik,¹³ and Anyanechi and Saheb,²⁴ showed that ameloblastoma in most clinical situations is diagnosed in the advanced stage of the disease due to absence of symptoms initially and the very slow tumour progression. This study confirmed earlier reports^{13, 23, 24} that follicular ameloblastoma exhibits a significantly higher recurrence rate than the plexiform type. Also, as earlier documented by other researchers,^{13, 22, 24} when conservative treatment like enu-

cleation and curettage is done rather than surgical resection with extension into normal healthy tissues, the more likely there will be recurrence of the lesion. Resection was done for all the ameloblastoma that recurred ($n=9$, 33.3%) in this study, and this approach may have been responsible for the lower frequency rate obtained when compared with earlier reports of Pinto et al.,¹¹ and Goteti.²²

The high recurrence rate of odontogenic fibromyxoma after surgical treatment is well documented in the literature,^{13, 25, 26} and can be as high as 25.0% especially if enucleation and curettage is the treatment modality. The recurrence is directly related to the local infiltrative nature of the lesion and the surgical procedure administered to the subject with conservative approach to treatment resulting in a higher rate of recurrence than resection as reported by Malik,¹³ and Singaraju et al.²⁷ In this series, radical treatment were used and that may have accounted for the lower complication rate (22.2%), although the rate in this study was in relation to all the lesions that presented and not restricted to only odontogenic fibromyxoma.

Odontogenic keratocystic tumour's (OKCT) wide variation in recurrence rate is thought by some researchers^{5, 7,}

Table 2. Distribution of lesions according to initial surgery, duration between initial surgery and recurrence, and the corrective surgery

Type of lesion	Total, n (%)	Initial operation n				Duration, years n			Corrective operation n					
		EC	Excision	MJR	SMR	<1	1-2	>2	SMR	Excision	Hman	Hmax	Pdb	PM
Odontogenic	16 (59.3)													
Follicular ameloblastoma	7	0	0	4	3	1	5	1	3	0	3	1	0	0
Odontogenic fibromyxoma	6	0	0	3	3	2	3	2	2	0	2	2	0	0
Plexiform ameloblastoma	2	0	0	1	1	0	1	1	0	0	1	1	0	0
KCOT	1	0	0	0	1	0	0	1	1	0	0	0	0	0
Non-neoplastic	6 (22.2)													
CGCG	4	2	0	2	0	0	4	3	3	0	0	0	0	1
Fibrous dysplasia	2	2	0	0	0	1	1	0	0	0	0	0	1	1
Non-odontogenic	2 (7.4)													
Ossifying fibroma	1	1	0	0	0	1	0	0	0	0	0	0	0	1
Diffuse lipomatosis	1	0	1	0	0	1	0	0	0	0	1	0	0	0
Malignant tumors	3 (11.1)													
MEC	1	0	1	0	0	1	0	0	0	0	1	0	0	0
Fibrous histiocytoma	1	0	1	0	0	1	0	0	1	0	0	0	0	0
Osteogenic sarcoma	1	0	1	0	0	1	0	0	0	0	0	1	0	0
Total	27 (100.0)	5 (18.5)	4 (14.8)	10 (37.1)	8 (29.6)	9 (33.3)	14 (51.9)	4 (14.8)	10	1	7	5	1	3

EC, enucleation and curettage; MJR, marginal jaw resection; SMR, segmental marginal resection; Hman, hemi-mandibulolectomy; Hmax, hemi-maxillectomy; Pdb, paring down bone; PM, partial maxillectomy

¹³ to be due to its thin and fragile wall and the plethora of treatment approaches, ranging from simple curettage with or without adjunct therapy to resection of the affected jaw. It was observed by earlier investigators ^{24, 28, 29} that OKCT recurrence rate after enucleation is 5-62% whereas enucleation with adjunct therapy or resection is associated with lower recurrence rates which are in agreement with the present study (3.7%). However, some authors, Raghav et al., ³⁰ and Cunha et al., ³¹ stated that the clinical characteristics, radiographic and histopathologic parameters may affect relapse of OKCT and should individually guide the choice of treatment.

The non-neoplastic lesions were fibro-osseous, while the fibrous dysplasia was of the monostotic type. The levels of the transcription factor C-fos are raised in fibrous dysplasia, leading to gene over-expression and the resultant dysplastic change in bone; but the pathogenesis is still poorly understood as noted by Kashima et al. ³² However, genetics (non-inherited), trauma, endocrine disturbances and molecular basis have been mentioned by Regezi et al., ³³ as being involved in the pathogenesis and relapse of the lesion after treatment. The recurrence of central giant cell granuloma confirmed the assertion of Malik, ¹³ that enucleation and

curettage may not be completely adequate to eradicate this condition, and therefore recommended resection where appropriate.

Liu et al., ¹⁶ reported 15% recurrence of ossifying fibroma in their series which is higher than the result in the present study while MacDonald-Jankowski ³⁴ reported 0-28% recurrence rate after initial treatment. Some earlier researchers, ^{34, 35} reported that the recurrence rates of the aggressive forms of ossifying fibroma can be as high as 30-38% after surgery. The lesion is usually well delineated from the surrounding normal structures and recurrence is reported ³⁶ to be rare if completely excised. The multi-focal developmental nature of this lesion was suggested by Pui-chee et al., ³⁷ but they noted that radical surgery does not appear to be justified after recurrence. The recurrence of lipomatosis after excision complicated by extra-capsular TMJ ankylosis is very rare in the orofacial region, ^{13, 38} and will require further investigation in a future prospective study.

The recurrence rate of the malignant neoplasm in this study is lower than in most other series. ^{4, 6, 7} This rate may not be necessarily correct in this environment, and is probably due to the limited number of such patients that were

treated initially in this centre because of the unavailability of the necessary facilities like radiotherapy required for their treatment as was documented in an earlier study.³⁹ Most of the patients were referred to other centres for treatment. The malignant lesions that were surgically treated initially were suspected not to be malignant. The relapses that occurred after treatment are due to the infiltrative and metastatic nature of the conditions.^{4,6}

The age, gender and site distribution of subjects in the present study is consistent with earlier reports.^{1,6} However, unlike the present study, Adekeye et al.,⁴ who studied only intra-oral malignant neoplasm recorded the maxilla as the most common site of recurrence which may be attributed to the complex anatomy of this region and the infiltrative and metastatic characteristics of the conditions they treated.

From the available literature, the time of recurrence of these lesions vary from study to study, but according to earlier researchers,^{1,4-7} remain unpredictable. The period of relapse recorded in the present study is comparable with those of Liu et al.¹⁶ although their study was restricted to patients who had only ossifying fibroma of the jaws. The methods of treatment used to eradicate these conditions after relapse though more radical than the initial surgical procedures, were effective, and these have been reported earlier by other investigators.^{1,4,6,7,13}

Some patients (n=3/371, 0.8%) had incomplete clinical data and were excluded from data analysis. However, this exclusion may not have affected the outcome of this study as the percentage is quite negligible. Follow-up of patients in the study environment is poor.³⁹ Probably not all the patients that had recurrence of lesion reported to the hospital for clinical evaluation and further treatment. The non-availability of CT scan, MRI, intra-operative frozen section and radiotherapy services precluded their utilization in cases where they might have been indicated, and these may have reduced the frequency of patients treated and increased the recurrence rate of the lesions managed in this series.

The intervention of agencies of government and non-governmental organizations is required to assist this institution in the provision of the medical services that are not rendered in this tertiary health facility. It is hoped that if made available, these facilities will reduce to the barest minimum the recurrence rate of the lesions identified in this study, thereby facilitating the achievement of the core healthcare system values in the study environment. A future prospective study might also address the limitations of this retrospective analysis by controlling the variables that influence treatment outcome. However, life time follow-up is advocated for the odontogenic and malignant tumours identified in this study.

Conclusions

This study showed that 3.7% of the lesions recurred after initial ablative procedures. Benign lesions particularly ameloblastoma and fibromyxoma that are locally invasive constitute the majority of the cases that relapsed which significantly occurred more often in the mandible and in the

21-30 years age groups. The intervention of agencies of government and non-governmental organizations is required to assist in the provision of the medical services such as CT scan, MRI, radiotherapy and intra-operative frozen section that are not rendered in this tertiary health institution which are necessary in the management of these patients. A future prospective study will address the limitations of this study by controlling the variables that influence treatment outcome.

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