

ORIGINAL ARTICLE

Clinico-Pathologic Spectrum of Salivary Gland Tumours and Its Management in a Suburban Tertiary Centre

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ABSTRACTS

Salivary gland tumors are heterogeneous group of lesions with complex clinico-pathological characteristics, distinct biological behaviors and considerable epidemiological differences. To evaluate the clinico-pathologic distribution of salivary gland tumours and compare the findings with the previous reports. A retrospective study of patients with histopathological diagnoses of epithelial salivary gland tumours over an eight year period. A total of 28 patients with M: F of 1.3: 1. Over 60% of the patients were within the 3rd – 5th decade of life. The mean ages of patients with malignant and benign tumours were 57.3 ± 15.4 and 34.7 ± 13.9 years respectively. Over 71% of the tumours were benign and greater proportions (57.1%) of the malignant as well as benign tumours occurred in males. Majority 78.6% were parotid tumours of which 72.7% were benign and pleomorphic salivary adenoma was the most common (71.4%) benign tumor. Adenoid cystic carcinoma is the most common malignant tumour (50% of the malignant tumours and 14.3% of all the tumour types). The treatment modality for the patients included surgical excision with or without chemo-radiation. Some variations exist in the clinico-pathological distribution of SGTs in this study from those previously reported among other native Africans. Further studies are recommended to clarify these findings and to possibly unravel the factors responsible for racial, gender and geographic variations in salivary gland tumours.

Key words: Salivary gland tumours, clinico-pathological characteristics, pleomorphic adenoma, Adenoid cystic carcinoma, surgical excision, chemo- radiation

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INTRODUCTION

Salivary gland tumours are heterogeneous tumours which are diffusely distributed around the face and in the upper aerodigestive tract. Clinical and epidemiological surveys reported salivary gland tumours account for less than 2% of all human tumours [1], and between 2.8 and 10.0% of all head and neck tumours [2] and they can thus be regarded as being relatively rare. Like many other tumours in human beings, the cause of salivary gland tumours remain largely unknown, however reported predisposing factors include genetics, social factors like, smoking, diet, chronic sialadenitis, as well as previous ionizing irradiation. Salivary glands are generally classified as major or minor. Most of the tumours occur in the major salivary glands especially the parotid (65 to 85%) and submandibular (10%) glands, while the remaining occur in the minor salivary glands which spread across the upper aerodigestive tract [2, 3, 4]. Sublingual salivary gland tumours are said to be rare in occurrence 5–8. While benign tumours especially pleomorphic salivary adenoma are generally regarded to be more common than malignant ones, acinic cell carcinoma and muco-epidermoid carcinoma are the common malignancies reported in many studies among pure negroid populations [4, 7].

Despite being relatively uncommon, neoplasms of salivary gland are of clinical importance, because of the deformation of the facial structures and the associated morbidities caused by them and also their proximity to important head and neck structures that pose considerable clinical management challenges [7].

Management of salivary gland tumours depends on the location, extent, type and the biologic behaviours of the tumours. While most of the benign lesions can be managed with surgical excision with wide tumour margins, the malignant ones will require adjuvant chemo or radiotherapy, and sometimes immunotherapy in addition to surgery. Management of the salivary gland tumours can be particularly challenging not only due to the variability of salivary glands, but also the diversity of the tumour types. Furthermore, the outcome of management will also depend on the tumour biology and behavior. For instance, tumours that have perivascular or perineural spread metastasize easily to distant sites and are more difficult to control. In fact, salivary gland tumors are heterogeneous group of lesions with complex clinico-pathological characteristics and distinct biological behaviors [1]. Considerable epidemiological differences in the distribution of salivary gland tumors have been reported between continents, and also in different parts of the same continent [5, 6]. Thus the incidence and distribution of salivary gland tumors is influenced by geographical and racial factors [3]. Ladoke Akintola University of Technology (LAUTECH) Teaching Hospital is a tertiary health institution that is located in Osogbo, South Western Nigeria. The hospital caters for patients from about four contiguous states in south-western Nigeria. Osogbo is in the rain forest zone in Nigeria, population is about 290,000 (2006 Nigerian National population figure), majority of whom are farmers.

Studies that had been done in different parts of Nigeria on salivary gland tumours had reported inconsistent findings, and this study aims to document the clinico-pathologic distribution and the treatment modalities employed in managing salivary gland tumours seen in a relatively young tertiary health facility in a sub-urban town in Nigeria. This could assist in guiding our decisions concerning screening, pattern of presentation and best management options available, in order to ensure a favourable outcome.

MATERIALS AND METHODS

The study was a retrospective study of all patients diagnosed with salivary gland tumour who were managed in the department of Otorhinolaryngology, Ladoke Akintola University of Technology Teaching Hospital, Osogbo, Osun State, Nigeria, over an eight year period between January 2005 and December 2012. All the patients with histopathological diagnoses of epithelial salivary gland tumours were included in the study. Excluded were patients with salivary glands whose case records could not be located, those with incomplete information and others with salivary gland swellings histologically confirmed to be non-epithelial tumors.

The clinical and pathological information retrieved from patients' hospital records included; patient's socio-demographics, clinical features and its duration, anatomical location of the tumor, Histopathologic classification, management and outcome. Histological classification of tumors was according to the 1991 WHO Classification of salivary gland tumors.

The information obtained was fed into a spreadsheet and the data generated was analysed using SPSS version 14 (Illinois, USA). Descriptive analysis was done as proportions and the results presented in tabular forms and charts.

RESULTS

Data of 28 patients were included in the study consisting of 16 (57.1%) males with (M: F) of 1.3: 1. The age of the patients ranged from 14 to 73 years (mean 40.4 ± 17.2 years). There were 327 head and neck tumours during the period thus; the salivary gland tumours represented 8.6% of all the head and neck tumours in this study. Over 60% of the patients were within the 3rd – 5th decade of life, with a quarter in the 21-30 years age group. Figure 1 shows the age and sex distribution of the patients. Most males were ≤ 40 years while most females were ≥ 41 years. There is no patient below 10 years or above 75 years. The mean age of patients with malignant tumours was 57.3 ± 15.4 years while mean age for those with benign tumours was 34.7 ± 13.9 years. Twenty (71.4%) of the tumours were histologically confirmed as benign while 28.6% were malignant tumours. Greater proportions (57.1%) of the malignant as well as benign tumours occurred in males.

Site distribution analysis shows that majority 22 (78.6%) were parotid tumours of which 16/22 (72.7%) were benign. Figure 2 shows distributions of the salivary gland tumours by site. None of the tumours was found in the sublingual salivary gland.

Histologically, pleomorphic salivary adenoma was the most common (71.4%) benign tumor and it represented 53.6% for the total tumour types in the study; this was followed by adeno-lymphoma (20%) of the benign tumours and 14.3% of the total tumour types. The parotid gland was the most frequent site for benign tumours (75%), followed by submandibular gland (15%). For the malignant varieties, adenoid cystic carcinoma is the most common, representing (50%) of the malignant tumours and (14.3%) of all

the tumour types. Mucoepidermoid carcinoma was the second most common histological malignant type of tumours (37.5%) of the malignant varieties and (10.7%) of the total tumour types. While adenoid cystic carcinoma was most commonly found in the parotid gland, the distribution of the other histological types is shown in Table 1. The treatment modality for the patients included surgical excision with or without chemo- radiation. Superficial parotidectomy was the most common surgical intervention performed in 64.3% of the patients. One patient had ulceration of the parotid gland secondary to hot formentation, he had wedge biopsy for histological diagnosis and subsequently had chemo-radiation. The various surgical treatment modalities are shown in Figure 3. All the seven (31.8%) patients with malignant tumour except one with acinic cell carcinoma had chemo-radiation, two patients with giant tumours who had large wound defects that could not be closed primarily at surgery , had delto-pectoral flaps raised to cover the defects.

Figure 1: Age and gender distribution of the patients

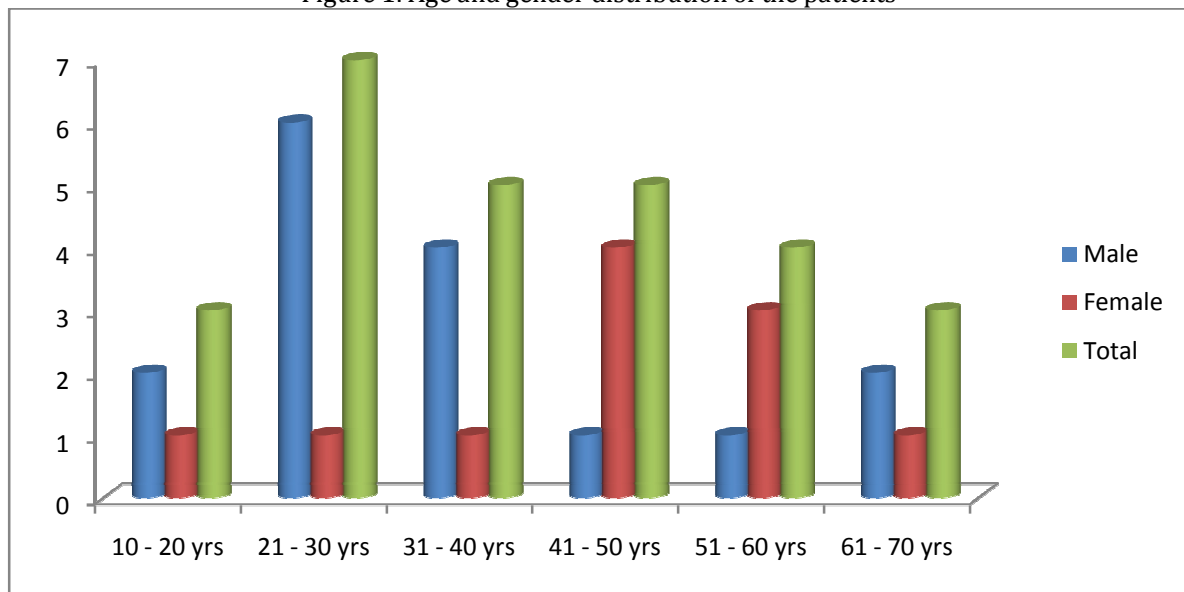


Figure 2: Site distribution of salivary gland tumours

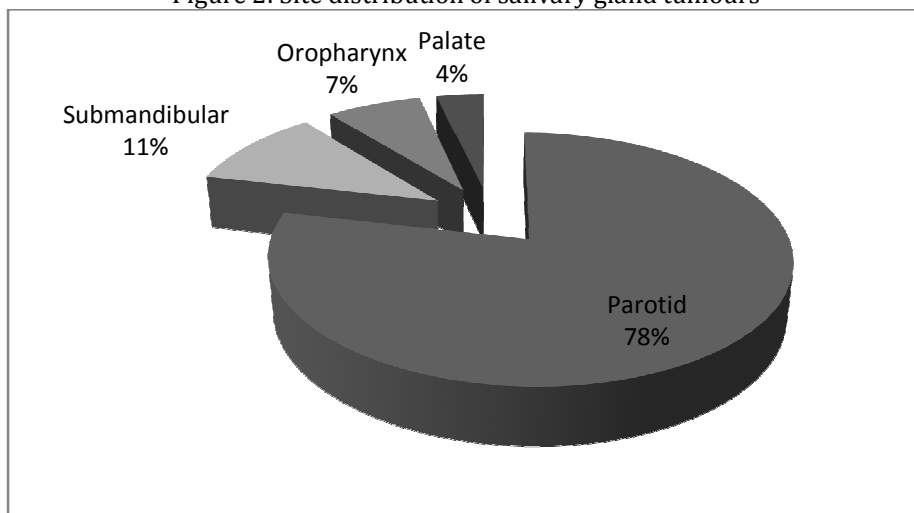


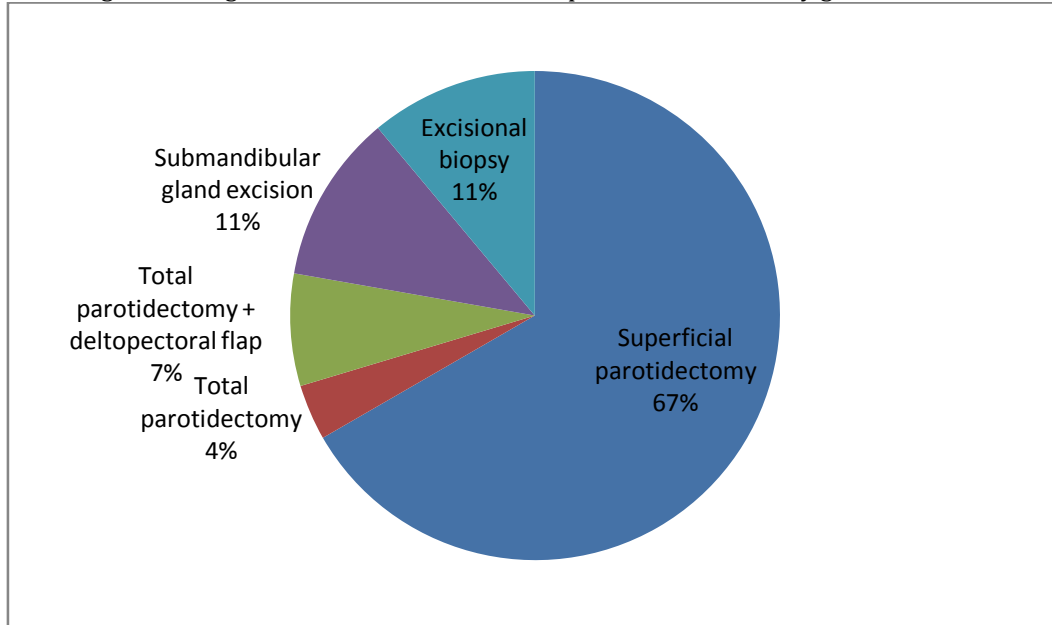
Table 1: Various histological types and distributions of salivary gland tumours

Tumour types	Sites			Total
	Parotid	Submandibular	MSG	
Benign				
Pleomorphic adenoma	14 (50%)	1(3.6%)	0(0%)	15(53.6%)
Adenolymphoma	1(3.6%)	2(7.1%)	1(3.6%)	4(14.3%)
Basal cell adenoma	1(3.6%)	0(0%)	0(0%)	1(3.6%)

Malignant				
Acinic cell carcinoma	1(3.6%)	0(0%)	0(0%)	1(3.6%)
Mucoepidermoid carcinoma	2(7.1%)	0(0%)	1(3.6%)	3(10.7%)
Adenoidcystic carcinoma	3(10.7%)	0(0%)	1(3.6%)	4(14.3%)
Total	22(78.6%)	3(10.7%)	3(10.7%)	28(100%)

NB: MSG = minor salivary glands

Figure 3: Surgical treatment modalities for patients with salivary gland tumours.



DISCUSSION

There is a wide variation in the global incidence and clinico-pathological characteristics of salivary gland tumours (SGTs). Most published studies from the African continent on salivary gland tumours also revealed considerable epidemiological differences in different parts of the continent. Poverty and ignorance might have contributed to under-reporting of some of these tumours in the African sub-region as many patients with seemingly innocuous, low-morbidity conditions such as benign salivary tumours may not bother to seek orthodox medical care. Thus hospital prevalence of SGTs in this continent may not adequately reflect the picture of the condition in the general population. The present prevalence of 8.2% of all H&N tumours in this study is however similar to the previous published studies in the sub-region [2].

While Adeyemi et al. [9] in Ibadan reported no sex predilection for intraoral salivary gland tumours, most other studies [4, 6, 7, 11] from the African continent reported female preponderance in their reports. The finding of male predominance in this study was however similar to the findings of Silas et al. [10] in Jos Nigeria and Oti et al. [5] in Ghana. While Silas et al. [10] had attributed the preponderance to cultural factors that do not allow females attend clinic alone without permission from their male counterparts in the Northern part of Nigeria, this reasoning may not be tenable in our own region. Though not full-prove, our small sample size might be responsible for this in our study. Some reports from the western world also reported a male preponderance [7, 12].

The findings from this study of over 60% of the patients being in their 3rd – 5th decade of life and the age group 21 – 30 years being most affected agreed with the previous report [7]. Ochicha et al. [6] in Kano, Nigeria reported prevalence of SGTs among 13 – 60 years and that over 50% occurred in 2nd – 4th decade of life. The mean ages for benign and malignant tumours in the present study of 34.7 ± 13.9 years and 57.3 ± 15.4 years respectively agrees with the findings from most African studies [4, 5, 7, 8]. Yaor et al. [8] opined that though salivary gland neoplasms represent a significant health problem in Africa, patients are generally younger at presentation. In general the age profile of the Africans was much lesser than the reported ages in the Western literature where benign lesions appeared in the fifth to seventh decade and malignancies a decade or two later among the elderly [6, 7]. This substantiate the earlier reports that there are a number of clinico-pathological differences between the salivary gland tumors in black population and those from white populations of Europe and United States [4, 7].

There is no consistency from most reports about the proportion of benign to malignant varieties in Africa. While report from eastern African countries like Tanzania, Kenya and Uganda showed almost equal proportions of benign and malignant varieties in their studies [4], some published studies from Zimbabwe, Nigeria and Mali had reported greater proportion of benign varieties [4, 7, 10]. Vuhahula *et al.*[7] in Uganda reported a significant proportion of malignant tumors in their series. The findings of 71.4% benign and 28.6% malignant varieties in the present study is similar to the findings of Silas *et al* in Jos, Nigeria [10], Oti *et al.* [5] in Ghana and de- Oliveira *et al*1 in Brazil while Ochicha *et al*6 in Kano, Nigeria reported a slightly lower proportion (56.4%) of benign varieties.

A preponderance of parotid tumours (78.6%) in this study is higher than the values reported in other studies from Africa [4, 6, 7]. Some of the reasons given for lower prominence of parotid glands tumours in Africa included poverty and ignorance fueling patients' non-chalant attitude towards seeking orthodox medical care [4, 7]. The findings from the present study is however similar to those from Brazil, Iran and most of the western countries [1, 13]. Increased awareness emanating from a relatively high literacy level in the south-western part of Nigeria may partly be responsible for the positive health-seeking attitude. Further studies especially at the community level with larger sample size will be required for further clarification of these findings.

Vuhahula *et al.* [7] however reported equal distribution of tumours between the parotid, submandibular and minor salivary glands in Uganda, while Bahra *et al* 4 in Kenya reported a predominant distribution of the tumours in the minor salivary glands. While preponderance of the palatal tumours among those with minor salivary gland tumour has been a consistent finding from previous studies in both western and African continent [6, 7,] our findings were at variance with these. There was no case of sublingual salivary gland tumour in the present study, confirming earlier reports that the sublingual gland is an unusual site for SGTs tumours. Our findings were also not in tandem with the assertion that parotid tumors were more likely to be malignant in the African [4, 7], since majority (72.7%) of parotid tumours in the study were benign in nature. Our findings however resonated with the statements that minor salivary gland tumours are more likely to be malignant and submandibular tumors are less likely to be malignant [7]. Contrary to the report from the western world that the submandibular gland followed by palate 13 were the main sites of malignancies, all the submandibular gland tumours from this study were benign. Thus it is possible that benign tumors were under-represented, and malignancies accounted for a relatively larger proportion [6, 7], moreso since it was previously reported that the parotid gland is a less likely site for salivary gland tumor in African population and that the tumors that occur in the gland have higher likelihood of being malignant compared with those from Western population [7].

Histologically, pleomorphic adenoma and adenoid cystic carcinoma were the most common benign and malignant tumours respectively, found in our study, in tandem with most of the reports from the African continent, [4, 7]. Ochicha *et al.* [6] however reported muco-epidermoid carcinoma as the most common malignant tumour in their study Like most other studies 6 - 8 in the African continent that reported rarity of warthin tumor, we also did not find case of warthin's tumour in this study. The parotid gland had been the site for most benign and malignant tumours as is the case in the present study [5, 6]. Vuhahula *et al* [7] in Uganda and de Oliveira *et al*1 in Brazil however reported minor salivary gland as the common site for malignant varieties.

Though there had not been consistency about gender predisposition for malignant or benign SGTs our finding of preponderant male distribution in both benign and malignant varieties is similar to the findings.[5] in Ghana. Other studies however reported female preponderance in all tumour varieties [4] while others yet reported variation in sex distribution according to nature of the tumours [4, 6] at different geographical locations. These findings suggest that geographic differences do exist in the pattern and pathology of salivary gland neoplasms.

In most cases, management of salivary gland tumours consists of tumour excision (submandibular, sublingual, minor salivary gland and parotidectomy) and adjuvant therapy –especially radiotherapy for malignancies [14]. Surgical treatment for the parotid gland tumours varies from limited excision to parotidectomy with or without facial nerve sacrifice, to extensive surgeries including neck dissections [14]. Factors that influence surgical therapies include facial nerve invasion, tumour extension, histological characteristics and positive limits of the tumor [14] Surgical excision was the main treatment modality we employed for all our patients while 31.8% who had malignant tumours had adjunctive chemo-radiotherapy. It may sometimes be difficult to appose and close large defects created after tumour excision which will necessitate closure with local, regional or distant flaps. Two of our patients had deltopectoral flap cover of their large defects with resultant good aesthetic outcome with minimal and acceptable donor site morbidity. It was reported that delto-pectoral flap remains a useful, reliable, and versatile regional flap that can be used alone or in combination with other flaps in selected circumstances

for major head and neck reconstruction [15]. Other local and free tissue flaps that had exhibited versatility and usefulness in head and neck reconstructive process with successful cosmetic and functional results especially the myocutaneous flaps.

In conclusion, we found that;

Some variations exist in the clinico-pathological distribution of SGTs in this study from those previously reported among other native Africans. Our treatment modalities were however similar to the standard practice elsewhere comprising mainly surgical excision of tumour, with adjuvant chemo-radiotherapy for the malignant ones.

Further studies are recommended to clarify these findings and to possibly unravel the factors responsible for racial, gender and geographic variations in salivary gland tumours.

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Conflict of interest: The authors declare that they have no conflict of interests.

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