

Original article

**SALIVARY GLAND NEOPLASMS IN UNUSUAL SITES IN THE HEAD AND NECK REGION:
A REVIEW OF 57 CASES IN IBADAN**

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ABSTRACT

BACKGROUND: The seromucinous glands of the nasal cavity, larynx, bronchi and ectopic sites share similar histology and repertoire of neoplasms with major and minor salivary glands. These rare neoplasms can be termed salivary gland neoplasms in unusual sites (SGNUS).

OBJECTIVE: To determine the pattern of presentation of SGNUS in the head and neck region.

METHODS: The records of the Departments of Oral Pathology and Pathology, University College Hospital Ibadan were examined and all histopathologically diagnosed salivary gland tumours (SGTs) occurring outside commonly known sites were included in the study. Demographic data such as patients' age, gender, tumour topography and diagnosis were documented and analysed.

RESULTS: SGNUS made up 13.8% of SGTs seen with malignant tumours constituting 91.2%. The most common histological sub-type was adenoid cystic carcinoma (ACC) with 31.6% cases, while the mean age of occurrence of SGNUS was 47.9 ± 16.2 years. The tumours occurred more in males with M: F ratio of 1.5:1. The site with the most common affectation was the nasal cavity with 35.1%. **CONCLUSION:** SGTs in unusual sites in the head and neck region have a tendency to be malignancies. They occur more frequently in males and are prevalent in the nasal cavity as shown by this study.

Keywords: Ibadan; Neoplasms; Salivary-gland; Topography; Unusual-site

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INTRODUCTION

The salivary gland system is composed of three paired major salivary glands (parotid, submandibular and sublingual) and numerous minor glands found mainly in the submucosa of the oral cavity. However, there are other seromucinous glands, mainly in the nasal cavity, larynx, bronchi¹ and ectopic sites, which do not produce saliva but share similar histology with major and minor glands¹ and have similar repertoire of neoplasms.^{1,2} These neoplasms can

be termed salivary gland neoplasms in unusual sites (SGNUS) and are rare tumours that typically occur in the head and neck region but occurrences in distant sites like the large bowel and the rectum have been reported in literature.^{3,4}

The common topography of SGNUS (including ectopic sites) is the lateral part of the neck, pharynx, middle ear, parotid lymph nodes or extranodal lymphoid tissue within the parotid. They may occasionally be sited in the gingivae, mandible, brain, parathyroid, thyroid and

cerebello-pontine angle.⁵⁻⁹ The occurrence of ectopic tumours has been attributed to abnormal persistence of vestigial structures or misplacement of salivary gland rests along embryologic pathways of migration during development and differentiation of primitive embryologic structures.²

Many studies have described the pattern of presentation of salivary gland tumours (SGTs) in the major and minor intraoral glands^{10, 11} but literature on SGNUS is limited to case reports and case series.^{5, 6, 8, 9} This study therefore aims to determine the pattern of presentation of SGNUS in the head and neck region using archival records.

MATERIALS AND METHODS

The archival data of the Department of Oral Pathology and the Department of Pathology, University College Hospital Ibadan spanning 20 years were examined and all entries considered to be SGNUS of the head and neck region were retrieved. This included all histopathologically diagnosed SGTs occurring outside the major, minor intraoral and accessory salivary glands from January 1994 to December 2013. Tumours arising from ectopic sites such as the mandible, neck and tonsils were also included while cases with incomplete records were excluded.

Demographic data such as patients' age, gender, tumour topography and diagnosis were documented and analysed using SPSS for Windows (version 20.0; SPSS Inc. Chicago, IL). Ethical approval for the study was obtained from the Oyo State Research Ethical Review Committee (AD 13/479/435).

RESULTS

A total of 413 biopsies and surgical specimens diagnosed as salivary gland neoplasms (SGNs) were reviewed over the study period, of which 57 were histopathologically diagnosed in sites earlier defined in this study as SGNUS. This constituted 13.8% of the entire salivary gland tumours seen within this period. Benign tumours were 5 (8.8%) while malignant tumours were 52 (91.2%). Adenoid cystic carcinoma (ACC) was the most common histological sub-type with 18 (31.6%) cases [Figure 1], followed by

adenocarcinoma not otherwise classified (Adeno Ca NOS) with 15 (26.3%) cases, while 10 (17.5%) cases were muco-epidermoid carcinoma (MEC) [Table 1, Figure 2]. The most common benign tumour seen was pleomorphic adenoma (PA) with three cases (5.3%) [Figure 3].

The mean age of occurrence of SGNUS was 47.9 ± 16.2 years while the age range was 16 to 79 years with peak age incidence in the seventh decade of life (Table 2). The mean age of males was 45.7 ± 15.5 , while that for females was 51.8 ± 16.2 but there was no statistically significant difference in these mean ages. ($t = -1.49$, $df = 55$, $p = 0.070$).

SGNUS occurred more in males (59.6%) compared to females, giving a M: F ratio of 1.5:1. Table 3 shows the topography of the types of SGNUS in the head and neck region. Nasal tumours were the most common with 20 (35.1%) cases followed by 17 (29.8%) maxillary sinus tumours, 9 (15.8%) intraosseous mandibular tumours, 4 (7%) nasopharyngeal tumours and two (3.5%) neck tumours. Other sites such as sphenoidal sinus, larynx, tonsil, infra-orbital rim and temporal regions recorded one (1.8%) case each.

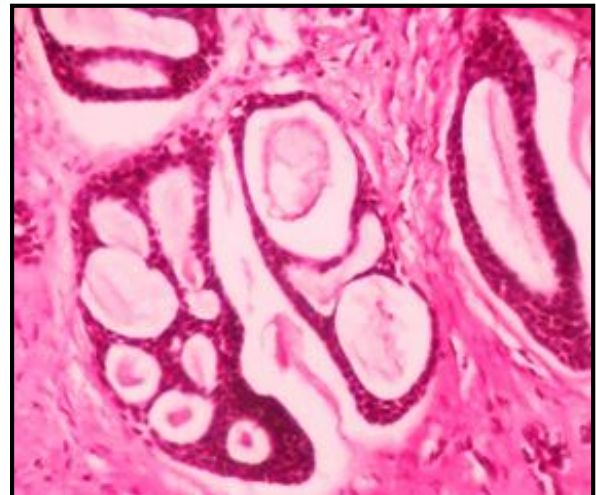


Figure 1: Adenoid Cystic Carcinoma - Section shows tubular and cribriform patterns within fibrous connective tissue (H&E, X100)

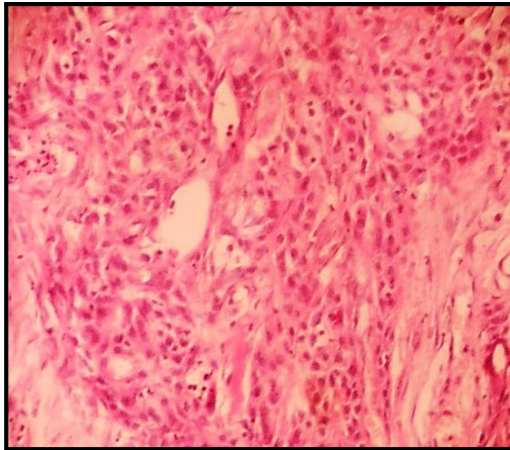


Figure 2: Muco-epidermoid Carcinoma - Section shows sheets of epidermoid cells and mucous cells in a loose fibrous connective tissue stroma (H&E, X100)

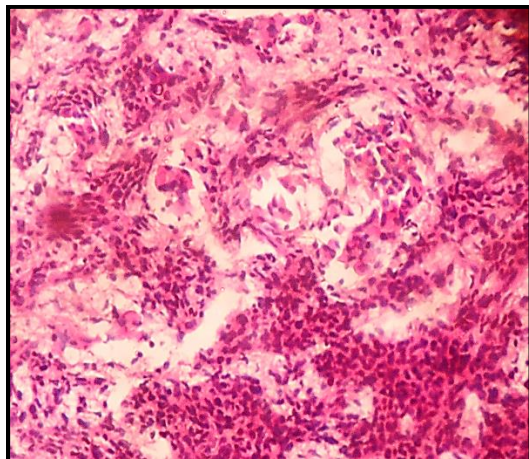


Figure 3: Pleomorphic Adenoma - Section shows irregular nests of myoepithelial cells within a myxoid connective tissue stroma (H&E, X100)

Table 1: Histopathological types of SGNUS of the head and neck

Histopathologic type	Frequency	%
Adenoid Cystic Carcinoma	18	31.6
Adenocarcinoma NOS	15	26.3
Muco-epidermoid Carcinoma	10	17.5
Pleomorphic Adenoma	5	8.8
Papillary Adenocarcinoma	3	5.3
Ca-ex pleomorphic adenoma	2	3.5
*Others	4	7.0
Total	57	100.0

*Other histopathological types include: PLGA (Polymorphous low grade adenocarcinoma), Sebaceous Cell Carcinoma, Canaliculi Adenoma and Cystadenoma

Table 2: Age group distribution of SGNUS of the head and neck

Age group	Frequency	%
10-19	4	7.0
20-29	4	7.0
30-39	10	17.5
40-49	11	19.3
50-59	9	15.8
60-69	13	22.9
70-79	6	10.5
Total	57	100.0

Table 3: Topography of SGNUS

Site	Frequency	%
Nose	20	35.0
Antrum	17	29.8
Mandible	9	15.7
Nasopharynx	4	7.0
Neck	2	3.5
*Other sites	5	9.0
Total	57	100.0

*Other sites include Temple, Sphenoid Sinus, Tonsils, Larynx and Infra-orbital region

DISCUSSION

The present classification system of SGTs by the WHO is based on histological typing¹² and does not make provision for topographical characterisation of SGTs; whether they are usual or unusual sites. Some reports have included SGTs arising from minor intraoral glands and other usual sites (e.g. sino-nasal tract, paranasal sinuses) as ectopic sites.^{13, 14} We suggest the use of the terminology 'unusual' site for SGTs occurring outside the major (extraoral) and minor (intraoral) salivary glands as an alternative term for ectopic sites. The paucity of demographic studies on SGNUS made comparison of our study with existing literature challenging, as previous studies are predominantly case reports.

Prior epidemiological studies on SGTs in usual sites reported no particular gender predilection,^{10, 15} however we found a male preponderance in SGNUS with M: F ratio of 1.5: 1 that was similar to findings by Otoh et al.¹¹ The peak age incidence was in the 7th decade and this is similar to peak age of occurrence of malignant SGTs in usual sites from existing studies.^{11, 16} Most studies of benign SGTs in usual sites report a lower peak age,^{17, 18} however, most of the SGNUS in our study were malignant, and had a similar peak age incidence with malignant SGTs in usual sites.^{11, 16}

Indeed a previous account by Ross and Sukis¹³ reported a predominance of malignant SGTs in ectopic sites. This report of a higher frequency of malignant USGNs is consistent with the general rule that, 'the smaller a salivary gland, the more likely an arising tumour is malignant'. This trend is seen in SGTs in usual sites, where malignancies occur more in the smaller minor glands.¹⁰

The most common malignant SGT seen in this study was ACC and this is in concert with studies that report ACC as the predominant minor salivary gland tumours in usual sites.¹⁰ This may suggest similar biologic behaviour and tumour evolution in salivary glands whether they are in usual or unusual sites. Also, the most common benign SGNUS in this study was PA. This is in keeping with literature on SGTs in usual sites where PA is also the most common.¹⁰ A contrasting report however, stated that Warthin's Tumour was the most frequent benign tumour of ectopic tissues' but no case of Warthin's tumour was found in this study.⁹

With regards to the site predilection of SGNUS; this study recorded the nose as the most frequently affected site and this is contrary to the findings of Ferlito et al.,⁹ who in their review of ectopic sites of SGTs found that the neck was the most affected site. The role of racial and geographical disparities in such varied topographical distribution of SGNUS cannot however be excluded based on this study.

In conclusion, SGNUS have a tendency to be malignancies. They occur more frequently in males and are prevalent in the nasal cavity as shown by the findings in the present study. ACC was the most frequently seen malignancy, while PA was the most common benign tumour. Additional research and follow up studies are essential to further characterise these SGTs in unusual sites.

Conflict of Interest: None declared

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