

ORIGINAL ARTICLE

Submandibular gland excision: a 16 year clinicopathological review of cases in a Nigerian Teaching Hospital

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Abstract

Aim: This study presents a review of the clinicopathological features of excised submandibular glands over a 16 year period of time at the Lagos University Teaching Hospital, Nigeria.

Materials and methods: A retrospective review of excised submandibular glands over a period of 16 years (January 1990 to December 2005) was conducted. Parameters studied included: age and gender of patients, symptoms and duration of symptoms and histologic diagnosis.

Results: A total of 47 patients had their submandibular gland excised during the period of the study. There were 28 men and 19 women (men : women ratio of 1.5:1). Mean age [standard deviation (SD)] of patients at presentation was 40.5 ± 18.1 years (range, 13–84 years). There were 34 (72%) histologically diagnosed neoplasms and 13 (28%) non-neoplastic pathologies. Patients with neoplastic pathologies were significantly older than those with non-neoplastic pathologies ($P = 0.004$). The most common neoplasm was pleomorphic adenoma (35%), followed by adenocystic carcinoma (12%) and anaplastic carcinoma (12%). Chronic sialadenitis with or without sialolithiasis (85%) was the most common non-neoplastic pathology.

Conclusion: The most common submandibular salivary gland pathology that necessitated surgical excision, in our institution, was neoplasia, predominately pleomorphic adenomas. The most common non-neoplastic pathology was chronic sialadenitis with or without sialolithiasis.

Introduction

The submandibular gland is the second largest salivary gland in the human body¹. Each submandibular gland weighs approximately 10–15 g and is anatomically divided into superficial and deep parts by the posterior edge of the mylohyoid muscle¹.

Pathologies affecting the submandibular gland can be divided into two broad groups: non-neoplastic and neoplastic pathologies². Patients with submandibular salivary gland pathologies usually present with two main symptoms: pain and/or swelling. It is, however,

the element of pain which makes patients seek medical treatment early¹. It is for this reason that patients with neoplastic submandibular gland pathologies tend to delay seeking medical treatment because of the paucity of symptoms compared with patients with inflammatory submandibular diseases^{1,3}.

Although, submandibular gland excision is undisputedly the treatment of choice for patients with neoplastic salivary gland pathologies, excision of the submandibular gland may not be regarded as the standard operation for patients with non-neoplastic salivary pathologies^{1,4}. However, in patients who undergo

submandibular excision for non-neoplastic pathologies, the procedure has been proven to be effective in eliminating pre-operative symptoms with minimal complications⁴.

There is paucity of reports in the literature regarding general indications for submandibular gland excision^{1,3,5,6}. A search through the literature did not reveal any such report from Africa.

Therefore, this study presents the clinicopathological review of cases of submandibular gland pathologies that warranted surgical excision at the Lagos University Teaching Hospital, Nigeria over a period of 16 years.

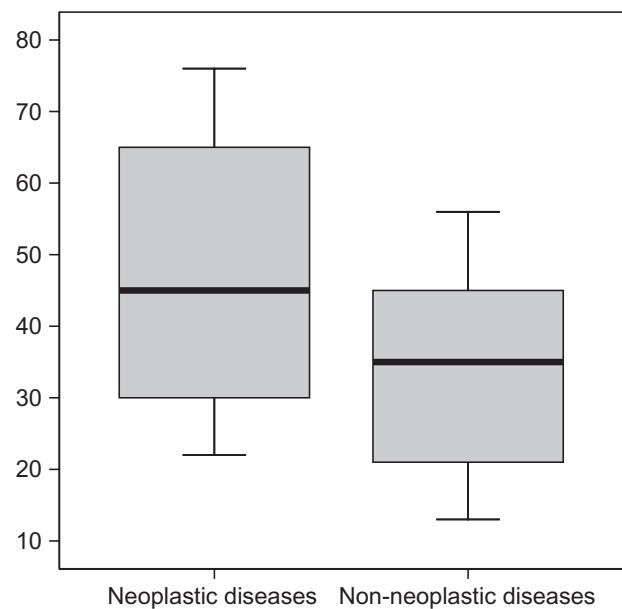
Materials and methods

A retrospective review of clinicopathological features of patients who had submandibular salivary gland surgical excision at the Lagos University Teaching Hospital over a period of 16 years (January 1990 to December 2005) was conducted. Patient records were retrieved from the records file of the Departments of Oral and Maxillofacial Surgery, Oral Pathology and Biology and morbid anatomy of the same institution. A standardised pro-forma was used to collect the following data: age and gender of patients, symptoms and duration of symptoms and histologic diagnosis. Two of the authors re-evaluated the haematoxylin and eosin-stained histologic sections.

Data were analysed using the SPSS for Windows (Version 12.0; SPSS Inc, Chicago, IL, USA) statistical software package, and presented in descriptive and tabular forms. Comparison of means was done with Student's *t*-test. *P* value was set at ≤ 0.05 .

Results

A total of 47 patients had their submandibular glands excised during the period of the study. There were 28 men and 19 women (men : women ratio of 1.5:1). Mean age [standard deviation (SD)] of patients at presentation was 40.5 ± 18.1 years (range, 13–84 years). There were 34 (72%) histologically diagnosed neoplastic pathologies and 13 (28%) non-neoplastic pathologies. Patients with neoplastic pathologies were significantly older than those with non-neoplastic ones ($P = 0.004$) (Fig. 1). Pleomorphic adenoma (26%) was the most common lesion, followed by chronic sialadenitis with or without sialolithiasis (salivary calculi/stones) (21%).



$P = 0.004$

Figure 1 Box plot comparative analysis of mean age (years) of patients with neoplastic and non-neoplastic diseases of submandibular gland. Medians and quartiles are displayed in the box plot and the horizontal lines outside the box represent the extreme values (y-axis = age).

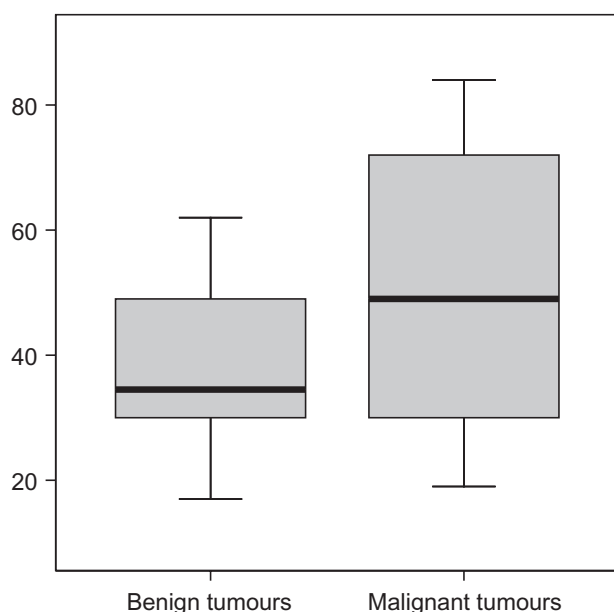
Neoplastic submandibular salivary gland pathologies

Surgical excision of the submandibular gland was carried out on 34 patients because of neoplastic pathologies. There were 22 men and 12 women with men : women ratio of 1.8:1. The mean age (SD) of patients in this group was 43.2 ± 9.0 years (age range, 17–84 years). In this group of patients with neoplastic pathology, malignant tumours were diagnosed in 18 (53%) patients and benign tumours in 16 (47%) patients (Table 1). Patients with benign tumours were significantly younger than those with malignant tumours ($P = 0.02$) (Fig. 2). The most common neoplastic pathology was pleomorphic adenoma (35%), followed by adenocystic carcinoma (12%), anaplastic carcinoma (12%) and malignant lymphoma (9%) (Table 1).

The primary clinical symptom in most patients (27 patients) was painless swelling, followed by painful swelling (four patients). Painful swelling with ulceration of the skin was the primary symptom in three patients with malignant neoplasm. Duration of symptoms before presentation was between 1 month and 12 years.

Table 1 Histologic distribution of neoplastic diseases of submandibular salivary gland

Histologic types	Frequency (%)
Benign	16 (47)
Pleomorphic adenoma	12
Oncocytic adenoma	1
Papillary cystadenoma	1
Fibrolipoma	1
Angiomatous fibrous histiocytoma	1
Malignant	18 (53)
Anaplastic carcinoma	4
Adenocystic carcinoma	4
Malignant lymphoma	3
Mucoepidermoid carcinoma	2
Adenocarcinoma	2
Squamous cell carcinoma	2
Carcinoma ex-pleomorphic adenoma	1
Total	34 (100)



$P = 0.02$

Figure 2 Box plot comparative analysis of mean age (years) of patients with benign and malignant neoplastic diseases of submandibular gland. Medians and quartiles are displayed in the box plot, and the horizontal lines outside the box represent the extreme values (y-axis = age).

Non-neoplastic submandibular salivary gland pathologies

Non-neoplastic pathologies were responsible for the surgical resection of submandibular gland in 13 patients. The mean age (SD) of patients in this group was 33.3 ± 14.0 years (range, 13–56 years). There

Table 2 Histologic distribution of non-neoplastic diseases of submandibular gland

Histologic types	Frequency (%)
Chronic sialadenitis	8 (62)
Chronic sialadenitis + sialolithiasis	2 (15)
Sialolithiasis	1 (8)
Mucocele	2 (15)
Total	13 (100)

were six men and seven women with men : women ratio of 1:1.2. Chronic sialadenitis, with or without sialolithiasis, was the most common non-neoplastic pathology and was histologically diagnosed in 10 (77%) patients (Table 2). Two cases of mucocele of submandibular gland origin were also recorded.

The record of primary clinical symptoms was available in seven (54%) patients with chronic sialadenitis (with or without sialolithiasis) of which four patients reported painful swelling and three reported intermittent painless swelling. Duration of symptoms in patients with sialadenitis ranged between 5 and 48 months. Submandibular swelling was the primary symptom of the two patients with mucocele with duration of 6–7 months before presentation.

Discussion

Clinical studies of pathology of the submandibular gland comprising large number of patients are infrequent in the literature, although, this gland is most frequently affected by the non-neoplastic pathology of the major salivary glands⁴. The submandibular gland has often been overlooked in relation to the parotid gland for two main reasons. First, parotid gland lesions are more commonly encountered than those of the submandibular glands^{1,7,8–13}. Second, the challenging anatomy of the parotid gland, made possible by the interposing facial nerve, has consistently generated more interest and study than the anatomy of the submandibular gland¹.

The present study agrees with previous studies^{1,3,4} in the literature regarding the age of onset and sex distribution of patients with submandibular salivary gland pathologies. In general, patients with neoplastic pathologies were significantly older than those with non-neoplastic pathologies, and those with benign neoplastic pathologies were significantly younger than those with malignant neoplastic ones. These findings suggest that neoplastic pathologies of submandibular salivary gland, especially the malignant ones, affect patients of older age group.

Neoplastic pathologies were more common than non-neoplastic pathologies in the present series in contrast to previous reports in the literature^{1,3}. Non-neoplastic pathologies were responsible for 61% of submandibular gland excision in a series reported by Gott and Sethl¹. While chronic sialadenitis with or without sialolithiasis has been widely reported as the most common lesion of the submandibular gland^{1,3-7}, and pleomorphic adenoma was the most predominant lesion in the present series. This discrepancy may be due to the fact that only surgically excised submandibular glands were included in the present study. Perhaps, neoplastic pathologies of submandibular gland are more common than non-neoplastic ones in our environment or it might be that many cases of chronic sialadenitis in our environment are amenable to antibiotic therapy.

Previous studies^{7,14} have reported more malignant neoplastic pathologies than benign ones. Although there were more malignant neoplastic pathologies than benign ones in the present study, the difference was, however, negligible. Other authors^{1,3} have, however, reported more benign neoplastic lesions than malignant ones. Pleomorphic adenoma was the most common neoplastic pathology of submandibular gland in the present series, constituting 26% of all pathologies of submandibular gland and 35% of all neoplastic pathologies. This is consistent with previous studies in the literature.^{1,3} Rallis *et al.*³ in a study on 55 submandibular gland excisions in their institution reported pleomorphic adenoma as the most common neoplastic pathology, constituting about 6% of all the submandibular gland lesions. Gott and Sethl¹, in another series on 93 submandibular gland excisions, reported that 33% of all cases seen were due to pleomorphic adenomas.

It is important to remove a pleomorphic salivary adenoma because of its potential for malignant change. Malignant transformations in large recurrent pleomorphic adenomas have been reported^{15,16}. Malignancy can arise from epithelial (carcinoma ex-pleomorphic adenoma) or mesenchymal (true malignant mixed tumour)¹⁶. The diagnosis of malignant mixed tumour requires the presence of a recognisable-mixed tumour (primary or recurrent) in association with a carcinoma and/or sarcoma¹⁶.

The most common malignant lesion in our study was adenocystic carcinoma, followed by anaplastic carcinoma and malignant lymphoma. This contrasts the findings of lymphoma and acinus cell carcinoma by Gott and Sethl¹, and squamous cell carcinoma by Rallis *et al.*³. Other authors^{14,17} have also reported adenocystic

carcinoma as the most frequent malignant tumour of submandibular salivary gland.

Sialadenitis with or without sialolithiasis has been widely reported as the most common non-neoplastic pathologies of the submandibular salivary gland^{1,3,4}. This is also confirmed in the present series. In the present series, two out of the 10 cases of sialadenitis were associated with sialolithiasis, and one patient had submandibular gland excision caused by sialolithiasis (Table 2). In a series of 233 patients with non-neoplastic pathologies of the submandibular gland, Ellies *et al.*⁴ reported that 79.4% and 11.6% cases were due to sialadenitis with or without sialolithiasis and sialolithiasis respectively. Defined as an acute, subacute or chronic salivary gland inflammation, sialadenitis may result from a variety of causes which may be obstructive (ductal obstruction from calculi formation) or non-obstructive (systemic diseases, trauma and specific infective agents) in nature¹. The causative factor is, however, frequently unknown¹.

Three (24%) cases of non-neoplastic pathologies in our study were associated with sialolithiasis. Compared with other salivary glands, submandibular gland has an unusual propensity to sialolith (calculi) formation¹. More than 80% of salivary calculi occur in the submandibular gland or its ducts^{1,18}. The reasons are speculated to be the following: (1) the submandibular gland has a long duct with slow flow rates; (2) the flow of saliva is against gravity; and (3) saliva from the submandibular gland contains more alkaline, mucin and calcium than that from the other glands^{1,19,20}. Although the aetiology is unknown, it has been suggested that the biological process of calculi formation in salivary glands is characterised by reduced secretory activity, alterations in electrolyte concentrations and impairment of glycoprotein synthesis in the salivary glands, all of which could result from structural deterioration of the cell membranes during aging²¹. Mimura *et al.*¹⁹ in a recent biophysical analysis of submandibular salivary calculi showed that mitochondria and lysosomal bodies from the degenerative ductal system of the submandibular gland are an aetiological source for calcification in the salivary gland.

Although surgical excision of the salivary gland is a well-established treatment modality for sialolithiasis, new endoscopic/non-invasive techniques which allow the conservative removal of salivary calculi have been developed²²⁻²⁴. These include lithotripsy, basket retrieval technique and balloon dilatation. With these techniques, over 70% of stones can now be retrieved leaving a functioning gland²⁴.

The majority of patients with neoplastic submandibular pathologies in our series presented with a

painless swelling, and patients with malignant lesions experienced painful swelling with ulceration. Painful swelling experienced by small proportion of patients may be due to the rapid stretching of the submandibular gland capsule¹. Painful swelling with ulceration is a feature of malignant salivary gland pathology²⁵. In this study, the primary clinical symptom in patients with sialadenitis with or without sialolithiasis was a painful swelling or intermittent painless swelling. This is in consonance with features associated with sialadenitis^{4,25}.

Conclusion

The most common submandibular salivary gland pathology that necessitated surgical excision, in our institution, was neoplasia, predominately pleomorphic adenomas. The most common non-neoplastic pathology was chronic sialadenitis with or without sialolithiasis. Patients with neoplastic pathologies were significantly older than those with non-neoplastic pathologies, and those with benign neoplastic lesions were significantly younger than those with malignant neoplastic ones.

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