

CASE REPORT

Pleomorphic Adenoma of the Lower Lip Extending to the Oral Commissure: First Case Report from Nigeria and Literature Review

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ABSTRACT

Background: Pleomorphic adenoma is the most common benign salivary gland tumour, predominantly affecting the parotid and minor salivary glands of the palate and upper lip. Involvement of the lower lip is exceptionally rare and poorly documented, with no prior cases from Nigeria or sub-Saharan Africa involving the oral commissure.

Methods: We report a rare case of pleomorphic adenoma in a 62-year-old Nigerian woman who presented with a two-year history of a painless, slowly enlarging nodular swelling of the lower lip extending to the left commissure. Clinical evaluation revealed a well-circumscribed, firm mass

measuring 1.5 cm in its widest dimension. The lesion was excised via an intraoral approach under local anaesthesia, and histopathological examination was conducted to confirm the diagnosis. Postoperative follow-up was done over a three-year period.

Results: The lesion was successfully excised with clear margins. Histopathology confirmed the diagnosis of pleomorphic adenoma, showing characteristic features such as epithelial and myoepithelial components in a myxoid stroma. The patient experienced psychological relief post-excision, and no recurrence was observed during three years of follow-up.

Conclusion: Pleomorphic adenoma of the lower lip is extremely rare, particularly with commissure involvement. This case, the first documented in Nigeria and sub-Saharan Africa, underscores the

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need to consider pleomorphic adenoma in the differential diagnosis of lower lip swellings. Accurate clinical evaluation and histopathological confirmation remain crucial for effective diagnosis and management.

INTRODUCTION

Salivary gland tumours (SGT) constitute less than 3% of all head and neck neoplasms, with pleomorphic adenoma (PA) being the most common type¹. Also known as benign mixed tumour, pleomorphic adenoma arises from both epithelial and mesenchymal components. The term was first introduced by Willis to describe its distinct histological pleomorphism seen under light microscopy². Historically, it has also been referred to as mixed tumour, enclavoma, branchioma, endothelioma, and endochroma³.

These tumours primarily affect the major salivary glands, most commonly the parotid, followed by the submandibular gland and minor salivary glands within the oral cavity⁴. Minor salivary glands are presents in the mucosal of oral cavity and oropharynx, secreting saliva directly into the oral cavity. Lesion involving the minor salivary gland are mostly malignant but benign lesion do occur. Benign lesion of the minor salivary occur at the different sites in the oral cavity, the junction of the hard and soft palate being the most frequent site, followed by the upper lip and buccal mucosa (5.5%). Less common sites include the pharynx (2.5%), retromolar area (0.7%), floor of the mouth, and alveolar mucosa⁵. These tumours most often occur in individuals between the fourth and sixth decades of life and show a slight female predominance⁶. Histologically, they are composed of spindle-shaped myoepithelial and polygonal epithelial cells within a variable stromal background, including myxoid, chondroid, and mucoid elements.

Pleomorphic Adenoma of the lip is rare, with that of the lower lip occurring in every six lesions seen⁷. There is no documented case of lower lip pleomorphic adenoma in Nigeria as at the time of presenting this case. Globally, few cases of lower lip

pleomorphic adenoma were reported⁷⁻¹⁰. A scientific literature search in English for cases of pleomorphic adenoma published in across major database such as PubMed, Web of Science, ScienceDirect, EMBASE, SpringerLink, Scopus, JSTOR, Jaypee Digital, Google Scholar, MEDLINE and Cochrane between 2000 and 2025 identified twelve articles reporting lower lip involvement. Four of these were excluded due to insufficient clinical or histopathological details, leaving eight relevant cases for review and are presented in table 1 below. Most of the reported cases were reported in Asia followed by America, No African cases reported to date; this is the first from sub-Saharan Africa.

This case report presents a rare instance of pleomorphic adenoma arising in the lower lip in a 62-year-old female from Yoruba ethic group in Southwest Nigeria. It highlights the clinical features, histological findings, and surgical treatment of this unusual presentation. (see Table 1).

CASE REPORT

A 62-year-old female patient of Yoruba ethnicity presented to the outpatient clinic of the Department of Oral and Maxillofacial Surgery, OAUTHC, with a two-year history of a gradually enlarging, painless swelling on the inner surface of the lower lip extending to the corner of her mouth as shown in figure 1. She first noticed the swelling incidentally, following a discomfort sensation when her tongue contacted the mucosa lining the lower lip towards the left commissure area. The patient claimed it has “mentally disturbed her”, this could be due to the visible swelling. She reported no difficulty in mouth opening or functional impairment. However, she observed a slight increase in size over the past six months. There was no history of similar swelling elsewhere in the body, no history of repeated mucosal bite trauma from tooth edges, repeated ulceration, or fibrotic healing. There was no relevant medical or dental history. Additionally, no prior trauma to the affected site was reported.

Extraoral examination revealed no significant abnormalities. Intraoral inspection identified a

Table 1: Reported Cases of pleomorphic adenoma of the lower lip between 2000 and 2025 in both males (M) and females (F)

No	First Author's name	Country	Year	Age (years)	Sex	Size	Trauma	Provisional Diagnosis	Imaging	Treatment	Follow-up	Recurrence
1	To et al	Hong Kong	2002	25	M	1cm	No	Benign minor salivary tumour	Nil	Excision	24 months	No
2	Nourwal et al	Saudi Arabia	2019	26	M	2.5 x 2.0cm	No	Fibroma	OPG, USS	Excision	18 months	No
3	Costa et al	Brazil	2015	37	F	NS	Yes	Fibroma	NS	Excision	24 months	No
4	Sood et al	USA	2014	46	M	1.5 x 1.2 cm	No	Lipoma Sebaceous cyst	Nil	Excision	No	*NS
5	Umemori et al	Japan	2022	69	F	1.5x 2.0 cm	No	Benign minor salivary tumour	USS MRI	Excision	2 weeks	No
6	Gotoh et al	Japan	2022	58	M	1.1x1.7	Yes	Dermoid cyst	USS MRI	Excision	24 months	No
7	Sengul et al	Turkey	2011	49	M	1.5x0.7	*NS	*NS	Nil	Excision	40 months	No
8	Akram et al	Pakistan	2023	30	F	1.0x1.0cm	No	*NS	CT	Excision	3 months	No

*NS – Not Stated

distinct, oval-shaped mass with a smooth mucosal surface, located on the lower lip towards the left commissure. The swelling measured 1.5x0.5 cm, with a non-tender rubbery firm consistency. There was no other mass noticed in the oral cavity except the lower lip swelling as shown in Figure 1.

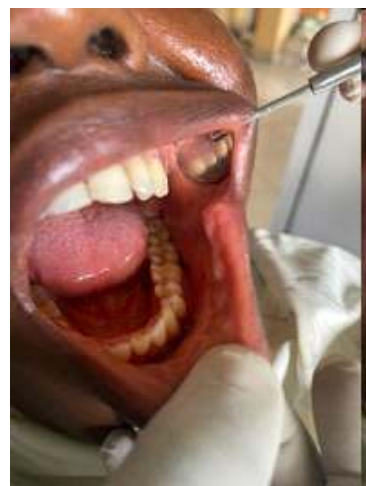


Figure 1: Discrete mass of the lower lip extending to the left commissure

Given the presence of a slowly progressing, painless mass at the junction of the upper and lower lips in a middle-aged woman, a benign salivary gland tumour was considered the provisional diagnosis. Differential diagnoses included fibroma, fibrosed mucocele, polymorphous low-grade adenocarcinoma, and fibrolipoma. No imaging was done due to its small size and the classic benign features of the lesion. Patient have not had prior treatment for the lesion before presentation to our facility.

The patient was scheduled for wide surgical excision of the mass under local anaesthesia via an intraoral approach following informed consent. The mass was well-encapsulated, a characteristic often seen in pleomorphic adenoma of the minor salivary glands (as opposed to that of major salivary glands) which typically present as well-delineated and mobile lesions.

Careful dissection between the fibres of the orbicularis oris muscle was carried out and the lesion was removed completely with a clean muscular bed as shown in Figure 2 and 3.



Figure 2: The nodular mass present in the muscular bed (orbicularis oris) of the lower lip and left commissure



Figure 3: Mass carefully dissected and delivered with the anchor suture.

The mass was delivered using an anchor suture and placed in a preservation medium containing 10% formalin before being transported to the histopathological laboratory for examination. The lower lip defect arising from the excision was closed primarily in layers (muscle and mucosa layer) with vicryl 3/0 suture. This is presented in figure 4 below.



Figure 4: The surgical site after suture placement.

The final histopathologic diagnosis confirmed pleomorphic adenoma, with no clinical evidence of recurrence after a three-year follow-up period as presented in figure 5.



Figure 5: Clinical picture of the surgical site at 3years post operative follow

The review timelines and clinical findings at each review are presented in table 2 below. The patient reported significant relief following excision and expressed satisfaction with both the aesthetic and functional outcome.

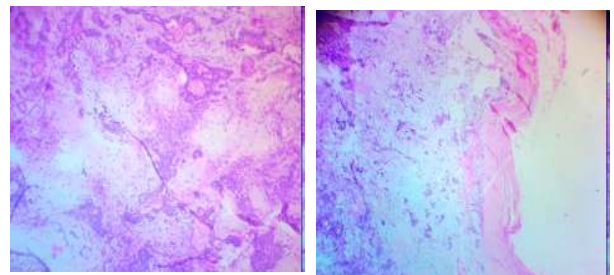
Table 2: Review Timelines and clinical findings

Review Timeline	Procedure	Clinical Findings
Day 0	Presentation and excision of swelling	Asymptomatic, discrete swelling
1 week	Review of surgical site, and suture removal	Apposed wound edges and satisfactory healing
3 month	Review of surgical site, and discuss histopathological report with patient	Surgical site has healed satisfactorily.
6 month	Review of surgical site	No recurrence observed

1 year	Review of surgical site	No recurrence observed
2 year postoperative	Review of surgical site	No recurrence observed
3 years postoperative	Review of surgical site	No recurrence observed

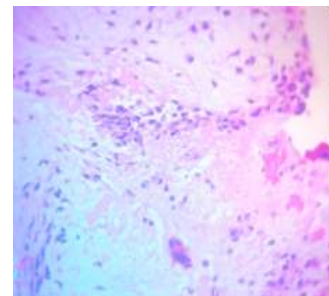
Microscopic Features

Histopathological examination revealed Sheets of epithelioid and clear myoepithelial cells in mucoid stroma. The epithelial elements are arranged in duct-like structures, sheets, clumps and consists of polygonal, spindle or stellate shaped cells.



A

B



C

Figure 6: A This shows epithelial cells in a myxomatous stroma. Some of the epithelial cells have formed duct-like structures. B. Histology shows an encapsulated glandular tissue that is composed of islands and nests of epithelial cells in a myxomatous stroma while C. Nests of plasmacytoid cells are seen within myxomatous stroma

Ethical Approval

Ethical approval was obtained from OAUTHC Ethics and Research Committee with protocol number: ERC/2025/07/09. The study registration number are – International: IRB/IEC/Z0004553 and National: NHREC/17/03/2021

Informed Consent

Written informed consent was obtained from the patient for the publication of this case report.

DISCUSSION

The lower lip is a functionally and aesthetically important component of the orofacial region. It is composed of skin, mucosa, adipose tissue, the orbicularis oris muscle, minor salivary glands, blood vessels, and nerves¹¹. Although minor salivary glands are distributed throughout the oral cavity, neoplastic transformation in the lower lip is rare⁷. Among SGTs, PA is the most common benign variant, predominantly affecting the parotid gland and the minor salivary glands of the palate. Its occurrence in the lower lip is uncommon and less frequently reported compared to the upper lip^{7,12}. To the best of our knowledge, the index case represents the first documented case of lower lip pleomorphic adenoma involving the oral commissure in Nigeria and, more broadly, in Africa

Several studies have confirmed a predilection for benign SGTs to involve the upper lip^{7,13,14}, while malignant variants are more commonly seen in the lower lip^{12,15}. In a large review by Alsanie *et al.*¹⁵ involving 5,736 SGTs, pleomorphic adenoma (PA) was identified as the most common benign tumour across all participating centres. The incidence of PA varied geographically, ranging from 64% in Europe to 88% in Africa, with Asia and South America reporting incidences of 80% and 87%, respectively. Notably, only a small proportion of these tumours were in the lips. When present, benign lesions such as PA demonstrated a predilection for the upper lip. Umemori *et al.*⁷ reviewed six cases of lip PA and reported only one case arising from the lower lip. The present case represents a rare occurrence of PA

in the lower lip, extending to the oral commissure, a site with even fewer reports in the literature.

The oral commissure, where the upper and lower lips meet, is composed of mucosa, skin, and underlying muscle, and is more commonly affected by inflammatory and infectious conditions such as angular cheilitis, herpes simplex virus infection, and condyloma lata¹⁶. The occurrence of PA in this region is exceedingly rare and can be easily misdiagnosed due to overlap with more common benign lesions. The involvement of the commissure in the index case raises diagnostic challenges, particularly when the lesion is firm and slow growing without signs of ulceration or pain, typical of PA presentations^{4,12}.

The rarity of lower lip involvement has been attributed to the constant exposure of the area to trauma and inflammation, potentially leading to destruction of susceptible glandular tissues before neoplastic transformation occurs¹². Chronic irritation, such as angular cheilitis, has been proposed as a possible predisposing or protective factor, though no definitive correlation has been established. Further studies are necessary to explore this hypothesis.

Reported cases of PA in the lower lip show a wide age range of presentation, typically between the third and seventh decades of life⁷. The present case involves a 62-year-old female, aligning with the observation that PAs can present in older individuals. Although malignant transformation is more common with increasing patient age, the lesion in this case remained benign, consistent with the majority of minor salivary gland PAs¹⁶.

Female predominance is another well-established feature of PA^{3,6,9}, which is reflected in the current case. Clinically, PA presents as a painless, slow-growing mass that may remain unnoticed for a long duration. The lesion's firm consistency in this case led to a differential diagnosis that included fibrolipoma and fibrosed mucocele—entities often linked to trauma or irritation¹⁷. However, no history of trauma or habitual lip biting was reported in this patient.

The size of the lesion in this case was comparable to those reported in other studies. Umemori *et al.*⁷ noted an average lesion size of 1.5 ± 0.7 cm in lip PAs, while Lotufo *et al.*¹⁸ described a similarly sized PA in the upper lip of a paediatric patient. This supports the notion that PAs of the lip typically exhibit limited growth and remain well-circumscribed over time.

Imaging studies were not conducted in this case, as the lesion's small size and benign characteristics rendered them unnecessary. This is consistent with other reports of lower lip PA, where imaging was often omitted^{8,19}. However, modalities such as ultrasound, MRI, or CT may be useful in cases of large, recurrent, or diagnostically ambiguous lesions^{7,9,10,20,21}.

Histopathological examination remains the gold standard for diagnosis. While incisional biopsy is generally discouraged in PA of major salivary glands due to the risk of tumour cell seeding through its pseudocapsule,²² PAs of minor salivary glands, such as those in the lips, are usually well-encapsulated and amenable to complete excisional biopsy, which serves both diagnostic and therapeutic purposes^{9,12,21}.

The mainstay of treatment is wide local excision with clear margins to minimize recurrence risk^{2,4,5,10,12}. In the present case, surgical excision was successfully performed without the need for complex reconstruction, as sufficient surrounding tissue allowed for primary closure. Reconstruction in the lower lip can be particularly challenging due to its role in speech, mastication, and facial expression, emphasizing the importance of early detection and conservative surgical planning. The patient has been followed up for three years without recurrence, consistent with literature reporting a low recurrence rate following adequate excision^{2,5,18,23}. The index case is limited because it is a single centre experience and lacks molecular analysis.

CONCLUSION

Pleomorphic adenoma of the lower lip is a rare presentation of a common benign salivary gland

tumour, with even fewer cases involving the oral commissure. Its clinical diagnosis can be challenging due to its resemblance to other benign lip lesions, especially when located in atypical sites. This case highlights the importance of maintaining a high index of suspicion for salivary gland tumours in slow-growing, asymptomatic lip masses. Awareness of this unusual site of occurrence, coupled with careful clinical evaluation and histopathological confirmation, is essential for accurate diagnosis and effective management.

Competing interests

The authors declare no conflicts of interest related to the publication of this case report.

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Authors' contribution

All authors read and approved of the final manuscript.

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