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CASE REPORT

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Management of a giant pleomorphic adenoma of the soft palate: A case report

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Key Clinical Message

Benign mixed salivary gland tumor is comprised of epithelial and myoepithelial cells and represents up to 80% of tumors of the parotid gland. It is relatively rare in the soft palate and in other minor salivary glands. Surgery is the standard care.

Abstract

Salivary gland tumors are relatively rare and morphologically diverse group of lesions. Pleomorphic adenoma (PA) is the most common salivary gland tumor, accounting for approximately 80% of all major and minor salivary gland tumors. PA usually affects the parotid gland. Huge PA occurring in soft palate is extremely rare. Patients with these tumors are usually between at the age of 40 and 60 years. The tumors exhibit pleomorphic nature microscopically that may pose diagnostic challenges to pathologists as may confuse PA histopathologically with other salivary gland tumors. Surgery is the standard treatment. The purpose of writing this case study is to describe unusual case of a giant PA of the soft palate found in a 44-year-old male successfully managed at our facility.

KEYWORDS

giant, mixed tumor, pleomorphic adenoma, soft palate, surgical excision

1 | INTRODUCTION

Salivary gland tumors represent a significant group of neoplasms categorized by a great range of types and morphological variations.[1] The majority of these tumors are benign, of which about 80% are pleomorphic adenomas (PA).[2] PA derives its name from the heterogeneous morphological architectural pattern seen under light microscopy.[3] It is also known as "mixed tumor, salivary gland type", which describes its pleomorphic appearance as opposed to its dual origin from epithelial and myoepithelial elements.[3] PA tumors mostly involve the parotid gland; however, if it occurs in the minor salivary glands, the palate would be the most common site.[4,5] Other sites in the oral cavity include the floor of the mouth, lip, buccal mucosa, tongue, pharynx, and retromolar area.[4] PA may occur at any age, but mainly affect patients from 40 to 60 years of age and has a predilection for women.[6,7]

Case studies describing huge PA occurring on soft palate are relatively rare.[2] Clinically, these tumors appear to be well circumscribed and nodular; with a smooth surface that generally present without pain, having a firm consistency, slow growth and do not attach themselves to the adjacent tissue. The tumors usually are covered by normal

overlying mucosa.[6] They can be mobile except when occurring in the hard palate.[8] The treatment of choice is surgical removal. Given correct surgical removal, the prognosis is excellent.[8] Herein, we report a case of a giant PA of the soft palate in a 44-year-old male patient; the tumor was treated successfully by surgical excision. A brief review of the literature is also provided.

2 | CASE PRESENTATION

A 44-year-old male patient reported to our institution's department of dental surgery because of slow growing intraoral swelling for 10 years duration. The new growth had gradual onset, and was increasing in severity over time. It was associated with difficult in swallowing due to mechanical obstruction. Snoring while sleeping, difficult in speaking, and weight loss were also reported. The patient denied history of throat pain, difficulty in breathing, fever, and night sweats. In addition, there was no previous history of trauma or any injuries. His past medical history was essentially unremarkable.

On examination, a huge mass measuring 9.4 × 7.9 × 8.3 cm on the soft palate was seen (Figure 1A). The mass was firm, non-tender, circumscribed, and faintly lobulated with smooth surface extending to uvula and bilateral anterior pillars of the tonsil. There was no regional lymphadenopathy, and his general and systemic examination were essentially normal. Full blood picture and vital signs were within normal limits. Computed tomography (CT) scan of the head and neck highlighted a well circumscribed and encapsulated soft palate solid mass with prominent blood vessels; its capsule took the anterior surface of soft palate (Figure 1B). The CT scan did not show any bony involvement by the lesion. Fine needle aspiration cytology (FNAC) results were suggestive of PA. Thus, the working diagnosis of PA with differential diagnoses of other salivary gland neoplasms including carcinoma ex PA (malignant mixed tumor), polymorphous adenocarcinoma, and epithelial myoepithelial carcinoma were considered.

3 | SURGICAL PROCEDURE

The patient was planned for surgery under general anesthesia after obtaining his informed consent. The patient was intubated through endotracheal tube via right nostril by fiber-optic guidance. Dingmans retractor was applied and tumescent solution infiltrated around the lesion. Mucosa around the lesion was marked and incised (Figure 1C); an extensive dissection was accomplished and the whole encapsulated tumor mass was excised together with the mucoperiosteum and overlying mucosa (Figure 1D). Surgical wound (Figure 2A) was closed with advancement of nearby mucosa in layers in a water tight fashion (Figure 2B). Surgery did not affect the speech and swallowing of the patient postoperatively. The excised mass (Figure 1D) was sent for histopathological examination. Macroscopically, the lesion was solid, well demarcated with a gray-white myxoid cut surface.

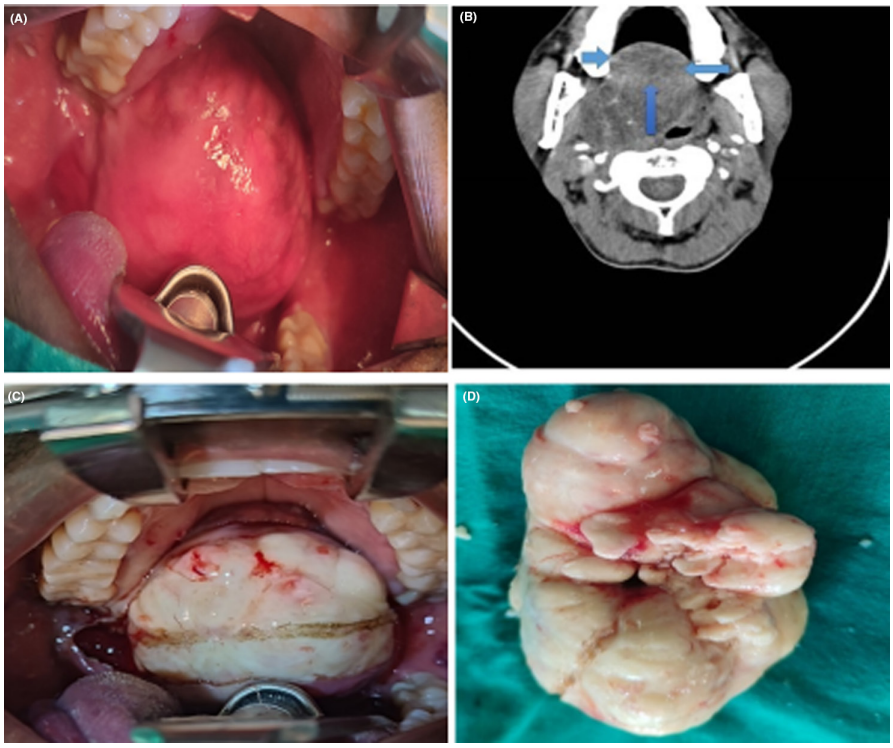

FIGURE 1 Photograph of a patient displaying a huge soft palate mass (A); a computed tomography (CT)-scan image showing a well circumscribed soft palate solid mass (B); appearance of the lesion after the surgical incision of the mucosa over the lesion (C); an excised soft palate mass that was sent for histopathology evaluation (D).

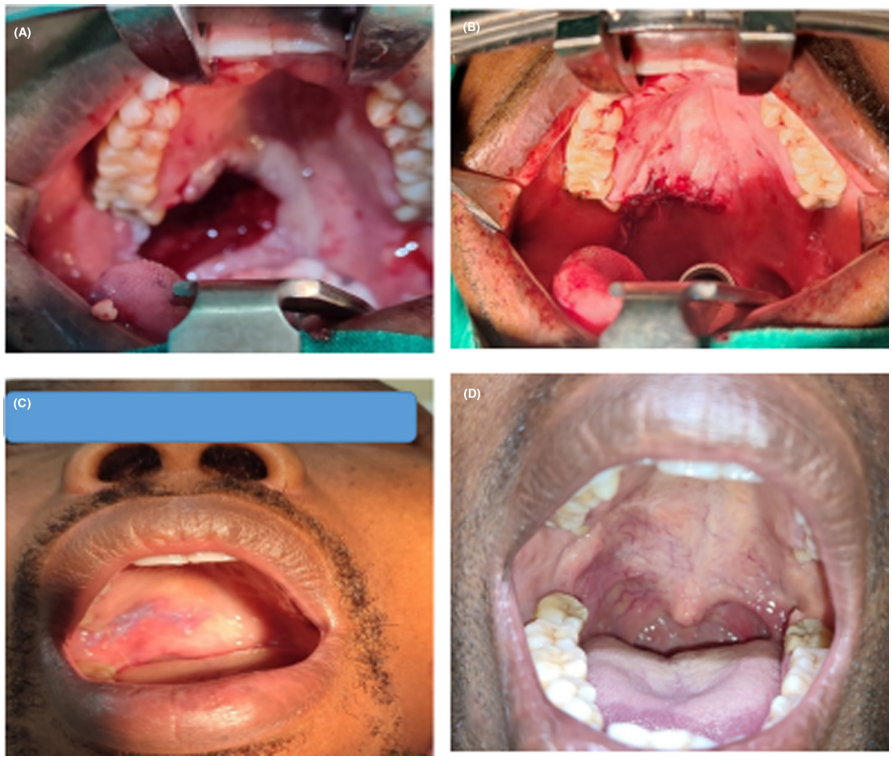

FIGURE 2 Photograph showing the surgical wound site (A); its appearance after being closed in layers in a water tight fashion (B); appearance of the patient at 6 months (C) and 12 months (D) after surgery respectively.

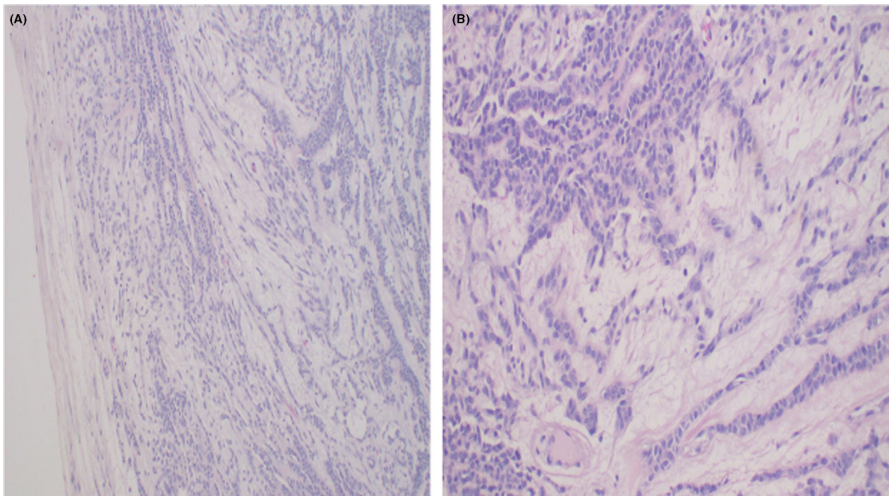

FIGURE 3 Histopathology displaying encapsulated PA lesion composed of epithelial cells with the tendency to form inner layers of ductular structures or cysts and myoepithelial cells forming the outer layer of cysts and tubule structures sparsed within myxoid stroma; H&E staining 10× original magnification (A); photomicroscopy of PA demonstrating myxochondroid stromal component; H&E staining 20× original magnification (B).

Histopathology study of the specimen demonstrated a benign triphasic salivary gland tumor consisting of epithelial (ductal) cells and myoepithelial cells scattered in a myxoid stroma (Figure 3A,B). The findings were consistent with PA. The patient's postoperative course was uneventful. No recurrences were observed in 6 months (Figure 2C) and 12 months (Figure 2D) of follow-up after surgery.

4 | DISCUSSION

PA is the commonest tumor of both major and minor salivary glands with about 70%–80% of it occurring in the parotid gland.[9–11] It is a mixed tumor of the salivary glands derived from epithelial, myoepithelial, mesenchymal, and stromal elements.[5] The most common affected age groups are fourth to sixth decades.[12] This is similar to our patient who was 44-year-old. PA is more common to females than males, with a ratio of about 2:1. Intra-orally, the most common site of this tumor is the palatal area (approximately 73%), followed by upper lip (17%), buccal mucosa, floor of the mouth, tongue tonsil, pharynx, and retro molar area.[4] Palatal tumors mainly occurs on the hard palate because majority of minor salivary glands are located in this area.[4]

Case studies of PA of soft palate are extremely rare. Only few cases have been reported in the English literature (Table 1). In our case the lesion's dimensions were

TABLE 1 Published case studies of soft palate pleomorphic adenoma in the English literature.

S no.	Author name	Article title	Year of publishing
1	Graham et al[16]	Interesting case: Maxillary swing approach for resection of a large pleomorphic adenoma arising in the soft palate	2006
2	Uz et al[4]	Pleomorphic adenoma of the posterior surface of the soft palate causing sleep disturbance: A case report	2017
3	Hmidi et al[19]	Pleomorphic adenoma of the soft palate: major tumor in a minor gland	2015
4	Daryani et al[15]	Pleomorphic adenoma of the soft palate: myoepithelial cell predominant	2011
5	Murthy et al[13]	A large pleomorphic adenoma of soft palate causing sleep apnea syndrome—A case report	2003
6	Yilmaz et al[14]	Giant pleomorphic adenoma of soft palate leading to obstruction of the nasopharyngeal port	2006
7	Kurokawa et al[5]	Extensive necrosis of pleomorphic adenoma in the soft palate: a case report and review of the literature.	2008
8	Oumakhir et al[20]	Pleomorphic adenoma of the soft palate	2008
9	de la Rica y Rodas et al[21]	A case of pleomorphic adenoma of the minor salivary glands (soft palate and parapharynx)	1991
10	Hughes et al[22]	A pediatric soft palate mass. Pleomorphic adenoma	2015
11	Karatzanis et al[17]	Malignant myoepithelioma arising from recurrent pleomorphic adenoma of the soft palate	2005
12	Lazow et al[23]	An unusually large pleomorphic adenoma of the soft palate	1984
13	McCartan et al[24]	Pleomorphic adenoma of the soft palate; an important benign disease in an unusual location	2014
14	Wagner et al[25]	Diagnostic challenge of a deep minor salivary gland neoplasm	2014
15	Sahoo et al[29]	Pleomorphic adenoma palate: Major tumor in a minor gland	2013
16	Osborne et al[26]	Massive pleomorphic adenoma of the soft palate	2004
17	Arampian[27]	Pleomorphic adenoma of the soft palate	1990
18	Glazer et al[28]	Pleomorphic adenoma causing a peritonsillar abscess	2014
19	Assouan et al[30]	An obstructive soft palate tumor	2017
20	Benazzou et al[18]	Mucoepidermoid carcinoma arising from pleomorphic adenoma of the soft palate	2006

9.4 × 7.9 × 8.3 cm, and was located on soft palate which is unusual location. Huge PA lesions of soft palate are unusual and may pose challenge in both diagnosis and treatment. Thus, the unusual size and unusual location of this tumor, both parameters make this case unique. The large tumor size caused compression of uvula and tonsillar pillars in our patient. The huge lesion size in the index case is possibly because the patient neglected it for long time (10 years). Case studies of huge soft palate PA have been previously reported. For instance Forde et al., reported a case of soft palate PA lesion measuring 9.9 cm in a 53-year-old male in the UK.[9] Murthy et al[13] described a case of large PA tumor in soft palate that was causing sleep apnea syndrome, while in their study, Yilmaz et al[14] described a case of a giant PA of soft palate leading to snoring and hypernasal speech.

Most common symptom manifested by large size PA tumors of the soft palate are dyspnea, dysphagia, acute airway obstruction, and obstructive sleep apnea.[2,13,14] This was similar to our case where he presented with difficulty in swallowing, difficulty in speaking and snoring. The symptoms could be due to unusual location and huge size of the lesion. As mirrored in our patient clinically, PA appears as a slow growing, asymptomatic, unilateral firm mass that may become huge if left untreated.[7]

Little is known about the origins of PA neoplasms, except that radiation exposure increases the risk. Equally uncertain is the histogenesis of the various components. A currently popular view is that all neoplastic elements, including those that appear mesenchymal, are of either myoepithelial or ductal reserve cell origin, thus, the designation PA. PA tumors are diagnosed on the basis of thorough history, physical examination and FNAC or histopathology.[10] Grossly as it was mirrored in the index case, PA tumors are solid with gray white cut surface.[11] However, unusual cases describing tumors with extensive necrosis, cystic degeneration, hemorrhage, and tumor-associated infarction of a PA of soft palate have been documented.[5]

Typically, PA tumors exhibit pleomorphic nature microscopically; that is varied histopathologic presentations. The pleomorphic nature may pose diagnostic challenges to both radiologists and pathologists. It may lead to confuse PA histopathologically with other salivary gland tumors. For instance, PA can easily be confounded with myoepithelioma, adenoid cystic carcinoma, mucoepidermoid carcinoma, and basal cell adenoma due to its diverse histopathologic presentations. Similarly, myoepitheliomas might be considered as a subtype of PA, however, these tumors lack the classic characteristic feature of glandulo-ductal differentiation. In addition, they lack of chondromyxoid or chondroid stroma.[11] Daryani et al[15] reported a case of soft palate PA which had predominantly myoepithelial cells with minimal stroma, ductal cells, or tubular elements. Histomorphological features in the index case (Figure 3) showed salient features of PA, these include a well-capsulated cellular tumor of sheets and islands of epithelial cells and rounded myoepithelial cells, along with myxomatous background and ductal architecture, was clearly evident.

The best method for management of palatal PA is an extensive excision with negative margins.[1] Lesions of the gum or palate may occasionally require a safety margin, normally being surgically removed below the periosteum, including the overlying mucosa. Graham et al,[16] documented a modified Weber–Ferguson incision and maxillary swing procedure to resect large PA arising in the soft palate in a 35-year-old man. The removal of bone is not usually required as PA tumors do not invade bone tissue, even though it may cause resorption from pressure. In our case despite of the large size of the tumor which was located on the soft palate; the tumor excision was successfully done and the wound repaired. Swallowing and speech were not impaired after surgery. Uz et al,[4] reported the use of endoscopic assisted trans-oral surgery following preoperative flexible fiberoptic nasopharyngoscopy imaging of the oropharyngeal passage. As evidenced in our patient, PA usually does not recur after satisfactory surgical excision.[4] Reasons for recurrence include partial excision, seeding, cutting through the microscopic extra capsular projections thereby leaving some tumor behind, or rupture of the capsule and unintentional seeding of tumor cells, as is more likely to happen when dissecting close to the capsule.[4] In our patient follow-up was done for 12 months with no signs of recurrence. As it was evidenced in the index case, PA tumors have seldom been observed to undergo malignant transformation. The tumor in the index case was there for a decade. Tumors that transform into malignant typically are likely to be recurrent, huge in size and persistent after resection. In addition, the tumors typically exhibit an abrupt change in growth as well as regional malignancy symptoms such pain, ulceration, spontaneous bleeding, and invasion of nearby tissue.[17,18] Also, transforming tumors into malignant are likely to arise from patients with history of exposure to radiation.

5 | CONCLUSION

Among benign tumors of the minor salivary glands, PA is the commonest, found most often in the oral cavity. The palatine arch is a very rare site for this tumor. Its diverse histological and topographical property make PA tumor unique. Due to its heterogeneous nature and composition, the tumor may pose diagnostic challenges to both radiologists and pathologists. Comprehensive surgical excision

is the treatment of choice. The investigative clinician and treating surgeon must be attentive of its recurrence, longevity, and malignant potential if wrongly diagnosed or treated.

AUTHOR CONTRIBUTIONS

Kanankira A Nnko: Conceptualization; data curation; writing – original draft. **Deogratius S Rwakatema:** Data curation; writing – review and editing. **Jackson M. Mariki:** Data curation; writing – review and editing. **Calvin J Baraka:** Data curation; writing – review and editing. **Raphael T Pima:** Data curation; writing – original draft. **Sosthenes Damas:** Data curation; writing – review and editing. **Alex Mremi:** Conceptualization; investigation; writing – original draft; writing – review and editing.

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CONFLICT OF INTEREST STATEMENT

All authors have declared that no competing interests exist.

DATA AVAILABILITY STATEMENT

There is no data genererated from this study

ETHICS STATEMENT

The patient provided written informed consent to allow for his de-identified medical information to be used in this publication. A waiver for ethical approval was obtained from the authors' institutional review board committee.

CONSENT

Written informed consent for publication of clinical details and images was obtained from the patient.

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