Bilateral Mucous Retention Cyst of the Submandibular Salivary Gland: A Rare Unusual Presentation and Review of the Literature

***Adewale Francis ADEJOBI, *Olawunmi Adedoyin FATUSI, **Azuka Raphael NJOKANMA, **Ayodele Gbenga AKOMOLAFE,**Abiodun Saheed OLATUNJI**

[Department of Oral and Maxillofacial Surgery, Faculty of Dentistry, Obafemi Awolowo University Ile-Ife,** Department of Oral and Maxillofacial Surgery, Obafemi Awolowo University Teaching Hospital Complex (OAUTHC), Ile-Ife]*

Correspondence
Dr. Adewale Francis Adejobi

Department of Oral and Maxillofacial Surgery, Faculty of Dentistry, Obafemi Awolowo University, Ile-Ife.
Email: walerosy2@gmail.com

ABSTRACT

Background: Mucous Retention Cyst (MRC) are non-neoplastic, developmental salivary gland lesion. Although more common in the minor salivary gland, few cases have been reported in the major salivary gland.

Objective: To draw the attention of clinicians to a rare occurrence of MRC in the submandibular gland bilaterally.

Case Report: The index case presents as a slowly progressive painless lesion of 10 years duration associated with aesthetics concerns. The lesion appeared as a double jaw with extension into the neck. Though lobulated and fluctuant, aspirate yielded yellow coloured, non-viscous and non-foul smelling fluid. Ultrasonography revealed a multilobulated mass with mobile heterogeneous fluid-like content in the submental and submandibular regions bilaterally. The massive size of the lesion limited the ultrasonic visualization of the submandibular salivary glands. The patient could not afford advanced imaging. During surgery, the lesion had involved the submandibular gland bilaterally with attachment to the hyoid bone and in close relation to the recurrent laryngeal nerve. Although the nerve was preserved, the patient experienced temporary hoarseness, which resolved with medication. Histopathologic examination of the excised lesion revealed the lesion as Mucous Retention Cyst.

Conclusion: The massive size of the lesion, delayed presentation and financial constraints in obtaining advanced imaging could have contributed to the diagnostic dilemma. This rare case will help the clinicians include MRC as a differential diagnosis of lesions involving submandibular salivary gland bilaterally. It also draws the attention of the maxillofacial surgeons to the unexpected injury to the recurrent laryngeal nerve and its management in a low-income setting environment.

Keywords: Non-neoplastic, Mucous Retention Cyst, bilateral submandibular salivary gland.

Adewale F. Adejobi
https://orcid.org/0000000256285805
Olawunmi A. Fatusi
https://orcid.org/0000000208191476
Azuka R. Njokanma
https://orcid.org/0000000170938748
Ayodele G. Akomolafe
https://orcid.org/000000028868101X
Abiodun S. Olatunji
https://orcid.org/000000028526259X

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INTRODUCTION

Mucous Retention Cysts (MRC) are non-neoplastic, aneurysm-like dilatation of the salivary ducts containing mucous lined with epithelium. MRC is usually seen in the 3-6 decades of life without gender predilection. The lesion accounts for 0.9% of all salivary gland lesion[1] and can arise within either major or minor salivary glands[2]. However, when major salivary gland is affected, parotid involvement accounts for 90% of MRC of all major salivary gland, with a mean age of 45 years[3] while only 7.8% is seen in the submandibular gland[1]. This report presents a huge MRC with involvement of bilateral submandibular salivary gland seen at our clinic.

The aetiology of Mucous Retention Cyst remains unclear. However, the pathogenesis of lesions has been linked to obstruction of the salivary duct.[4, 5] It may involve the major salivary or minor salivary glands. The parotid gland is the commonly involved major salivary gland while intraorally one or multiple minor salivary glands may be involved commonly in the buccal mucosa, floor of the mouth or lips. Ductal obstruction of the minor salivary gland may occur as a result of mucus plugs or stones causing ductal swellings, thus producing ductal ectasia commonly confused with mucous retention cysts.[6]

Mucous Retention Cyst of the major salivary gland is common in the parotid gland usually as slowly progressive painless swelling.[7, 4, 5]. Submandibular gland mucous retention cyst is extremely rare. However, it should be considered in the differential diagnosis of swelling at the submandibular triangle. Possible differential diagnoses for swellings in the submandibular triangle include neoplastic and non-neoplastic lesion. Neoplastic lesions include tumors of the submandibular salivary gland, the tail of the parotid gland, the Hodgkin's disease and non-Hodgkin's lymphomas. Non-neoplastic causes include branchial cleft cyst, thyroglossal duct cyst, hemangioma, laryngocele, teratoma, dermoid cyst, thymic cyst, mononucleosis, cat scratch disease, mumps, benign lymphoepithelial cyst, sialadenitis, Sjögren syndrome, submandibular cellulitis or abscess and submandibular mucoceles.[1,2,5] Mucoceles can be extravasation cysts or retention cysts. Mucous extravasation cyst is found commonly in children and young adults, while retention cyst is found in the elderly[8]

The index case presented a diagnostic dilemma due to its clinical features and constraints in diagnosis. The objective of this study was to review the literature on major salivary gland MRC with the report of an index case at our center of MRC involving bilateral submandibular gland.

CASE PRESENTATION.

A 24-year-old male undergraduate student presented in the Oral and Maxillofacial Surgery clinic of the Obafemi Awolowo University Teaching Hospital Complex, Ile-Ife, with a slowly progressive painless bilateral upper neck swelling, noticed 10 years ago. No reduction had been noticed since the onset. The patient could not recall any traumatic event related to the onset of the lesion. Swelling is not associated with dysphagia, respiratory disturbance, hoarseness, mastication or discharge. He had withdrawn from school due to the psychological effect of the massive swelling. Cigarette smoking and alcohol consumption were the positive social habit. The patient had presented 3 years earlier but absconded due to financial constraints.

Examination revealed localized oval-shape multinodular swelling extending superiorly from the inferior border of the mandible to 4cm above the manubrium, inferiorly and bounded laterally by the anterior border of the sternocleidomastoid muscle bilaterally. The swelling was fluctuant, the overlying skin was intact (without pigmentation, erythema and ulceration) and there was no paresis of the lower lip. The submandibular lymph nodes were palpable, non-tender and freely mobile. Other cervical lymph nodes (Levels Ia, II, III, IV, V and VI) were not palpable. Aspirate from the lesion yielded yellow coloured, non-foul smelling aspirate without granules. Intra-orally, there was no swelling in the floor of the mouth. The Warthin's duct was patent, expressing clear saliva. A provisional diagnosis of benign lymphoepithelial cyst was made. Ultrasonography revealed multi-lobulated mass with mobile heterogeneous fluid like content in the submental and submandibular region bilaterally. The submandibular glands could not be visualized. Advanced imaging could not be done because patient could not afford the cost of Magnetic Resonance Imaging.

Patient was scheduled for lesion excision under general anaesthesia using the Risdon incision. At surgery, there was bilateral involvement of the submandibular gland with attachment of the lesion to the hyoid bone and the glands were excised along with the lesion. The lesion also wrapped round the recurrent laryngeal nerve, thus the nerve was teased out of the lesion. An active drain was placed for 4 days and discontinued after 24 hours of non-discharge. Post-operatively, patient experienced mild hoarseness due to traction of the recurrent laryngeal nerve but this resolved with

oral Neurobion, administered at one tablet 8-hourly daily for 3 weeks. The surgical specimen was diagnosed histopathologically as Mucous Retention Cyst. Histopathology result revealed a densely collagenised connective tissue stroma lined by stratified squamous epithelium. Within the connective tissue stroma were mucous and serous salivary glands, ducts and muscles. Also seen were granulation tissue, endothelial lined vascular channel and areas of heamorrhage.

The patient developed a hypertrophic scar at the site of surgical incision and was treated with intra-lesional triamcinolone. No recurrence was seen at 6 month post-surgical review.

Figure 1: A picture of a 24-year-old male with bilateral Mucous Retention Cyst of the Submandibular Salivary Gland

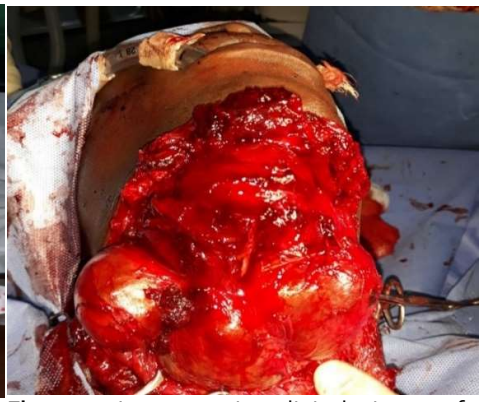


Figure 2: Intraoperative clinical picture of the lesion exposed

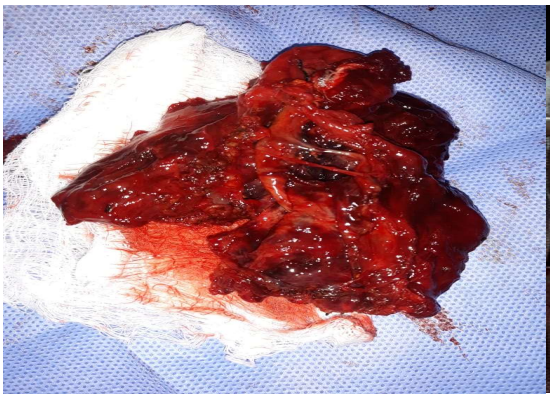


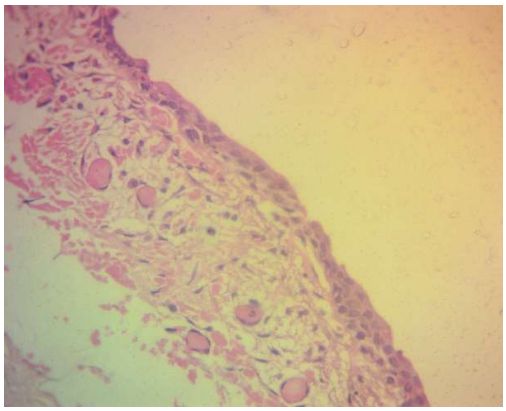
Figure 3: Excised surgical lesions

Figure 4: Intraoperative picture of the surgical bed with hyoid bone exposed

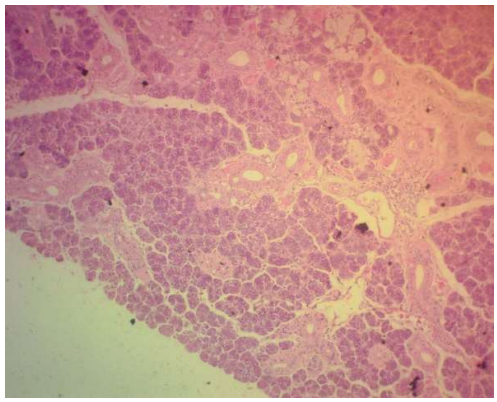
Figure 5: Surgical access sutured with an active drain in-situ

Photomicrograph

Figure 6a and 6b: Photomicrograph show cystic cavity lined by stratified squamous epithelium. There is a surrounding fibrous connective tissue with small blood vessels (A) and lobules of mixed mucus and serous salivary gland acini (B) [H&E x 100]



A



B

DISCUSSION

A mucous retention cyst is a true developmental cyst that is lined by epithelium derived from salivary gland tissue. This cyst are usually separate from the normal salivary duct although there is some controversy surrounding cyst-like dilatation of salivary ductal epithelium as a result of obstruction which represents more of a ductal ectasia rather than a true cyst.[5]

Mucous retention cysts usually produce asymmetric swelling of the submandibular triangle. We however, presented a rare index case in Nigeria of an adult male who presented with synchronous mucous retention cysts involving the submandibular salivary glands bilaterally. Mucocele sometimes has a slight female prevalence of about 1.3:1,[9] however, our case was a male. The prevalence of mucoceles is 2.4 cases per 1000 persons, commonly occurring in individuals aged 3-20 years old[10]. Mohan et al[2], in their 15- year retrospective review of non-neoplastic salivary gland lesions, found 3 of the 5 MRC seen in major salivary gland, to be located in the sublingual gland. Choi et al.[9] also reported a case of bilateral submandibular mucoceles in a 16-month-old child, however the reported case was that of a mucous extravasation cyst.

Although the aetiology of MRC remains controversial, the pathogenesis of the lesion have been linked to obstruction of the salivary duct.[4, 5] The failure of the patient to recall related traumatic event and non-involvement of the sublingual gland may also support this assumption. The increased pressure within the salivary duct causes dilatation, leading to proliferation of the ductal epithelium

Patients with mucocele present with asymptomatic, painless swelling in their mouths or may present as a neck swelling especially in cases diagnosed as plunging ranula. Most presentations are also unilateral. However, our patient presented with a massive swelling involving the submandibular triangle bilaterally and sub-mental region. The swelling was also of ten years duration (10 years) at the time of presentation for treatment. The patient cited financial constraints as a reason for late presentation. This is supported by the conclusion in the study conducted by Stierman et al[11] on assessing effective health coverage in the low and middle income countries. They identified affordability as a barrier to effective health coverage.

Patient's aesthetics is usually compromised due to the resultant neck swelling in submandibular mucocele. The swelling may also interfere with speech and swallowing and may cause respiratory compromise if the swelling displaces the tongue upward and backward.[12, 13] Our patient cited aesthetic compromise as his chief complaint as his look had been severely compromised due to the resultant neck swelling.

Clinical findings of a submandibular mucocele usually reveal a dome-shaped, non-tender, fluctuant, swelling in the submandibular region and may range in size from small lesions to lesions of grotesque proportions in size. This was similar to presentation in the index case. The non-tender, fluctuant and massive swelling in our patient resulted in a double jaw appearance. (Figure 1)

Non neoplastic lesion of the salivary gland presents clinically as tumours with similar pathologic features, making diagnosis difficult and errors

serious.[14] Diagnosis of mucoceles is usually clinical and ultrasonography, computed tomography (CT) /magnetic resonance imaging (MRI), are investigative methods that have been used to evaluate the lesions, for anatomical extent and association with surrounding vital structures[15]. However, the use of ultrasonography remains the most affordable and least invasive method of investigating these lesions[16] especially in low to middle income economies like Nigeria. It however has some limitations as other lesions such lymphangioma, lipomas and hemangioma may show similar sonomorphologic characteristics. The expertise of an experienced radiologist is thus invaluable in evaluating these lesions. Advanced imaging such as CT & MRI are rarely required except the mucocele involves the neck/cervical area and are usually indicated in submandibular mucocele[17]. However, our patient could not afford advanced imaging as he claimed financial constraints. Histopatholgical analysis is also important to differentiate between benign and malignant disease. A fine needle aspiration cytology can be employed for this purpose. The provisional diagnosis of the index case was benign Lymphoepithelial cyst. Although the benign nature could be confirmed by FNAC, the histopathologic investigation was able to describe the tissue of origin of the lesion with a definitive diagnosis of MRC of the submandibular glands.

Treatment of submandibular mucoceles is surgical excision of the mucocele along with excision of the involved submandibular salivary gland to prevent recurrence.[4, 5] Bilateral excision of the submandibular mucocele and submandibular salivary gland was done for our patient under general anaesthesia through a cervical neck incision. The lesion in this patient extended to the hyoid bone and meticulous dissection was done to detach the lesion in its entirety while preserving the recurrent laryngeal nerve. Patient's aesthetics was restored after surgery with no recurrence at 6 month postoperatively.

CONCLUSION

This case report presents a rare occurrence of MRC in submandibular glands bilaterally and should be included in the differential diagnosis of a long standing lesions involving submandibular gland bilaterally. The huge size of the lesion, delayed presentation and financial constrains in obtaining advanced imaging could have contributed to the diagnostic dilemma. It also draws the attention of the maxillofacial surgeons to the possibility of injury to the recurrent laryngeal nerve and its management in a low income setting environment.

Source of support
Nil
Conflict of interest
None declared

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