



Case report

Unusual giant calculus of the submandibular duct: Case report and literature review

Zephania Saitabau Abraham^{a,*}, Mary Mathias^b, Aveline Aloyce Kahinga^c^a Department of Surgery, University of Dodoma, School of Medicine and Dentistry, Dodoma, Tanzania^b Department of Otorhinolaryngology, Temeke Municipal Hospital, Dar es Salaam, Tanzania^c Department of Otorhinolaryngology, Muhimbili University of Health and Allied Sciences, Dar es Salaam, Tanzania

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ABSTRACT

Introduction and importance: Majority of salivary gland stones (sialoliths) occur in the submandibular gland (Wharton's duct and parenchyma) accounting for 80% of cases. A Giant calculus of more than 3 cm is a rare encounter.

Case presentation: We present a 45-year old male patient who undergone surgical removal of a giant submandibular gland calculus which was reported by the patient as a result of a hard mass beneath the tongue with occasional pain being experienced during intake of meals.

Clinical discussion: Clinical assessment revealed a painless palpable hard mass beneath the tongue though with some dull pain being experienced during intake of meals. Local examination showed a hard mass at the sublingual region but not adhered to surrounding structures. The overlying intraoral mucosa appeared normal and not inflamed and with neither enlarged ipsilateral submandibular gland nor cervical lymph nodes. The patient was then prepared for surgical removal of the calculus under general anesthesia where a single giant calculus (measuring 4 cm) was extracted by marsupialization of Wharton's duct.

Conclusion: A giant calculus of more than 3 cm is a rare encounter and they remain to be one of the causes of submandibular gland dysfunction.

1. Introduction

Most of the salivary gland sialoliths involve the submandibular gland (Wharton's duct and parenchyma) accounting for 80% of all the calculi. It involves commonly the submandibular duct (Wharton's duct) [1–9]. Formation of calculi within the Wharton's duct may be due to several reasons such as flow of saliva against gravity, high calcium and mucin content and its more alkaline though the exact aetiopathogenesis remains unknown [10]. The work has been reported in line with the SCARE 2020 criteria [11].

2. Case presentation

A 45-year old male patient presented to a health facility complaining of a hard mass beneath the tongue with occasional dull pain being marked during intake of meals.

The patient denies a prior history of recurrent pain or swelling at the right submandibular gland region.

Local examination showed a hard mass at the sublingual region but not adhered to surrounding structures. The overlying intraoral mucosa appeared normal and not inflamed and with neither enlarged ipsilateral submandibular gland nor cervical lymph nodes. Computerized tomography scan of the head/x-rays and sonography were not done due to financial constraints by the patient.

Laboratory investigations were found to be normal; Hemoglobin was 13 g/dl, serum sodium, potassium and calcium had normal values. The patient was then prepared for surgical removal of the calculus under general anesthesia where a single giant calculus (measuring 4 cm) was extracted by marsupialization of Wharton's duct.

Postoperatively; He was then kept on intravenous ceftriaxone for 24 h and then maintained on tablets cephalexin 500 mg 12-hourly for 7 days and oral corticosteroids (tabs prednisolone 10 mg once daily for 7 days and then 5 mg once daily for the next 7 days). Postoperatively the patient made an uneventful recovery with no any flare upon 6-months of follow up (Table 1).

* Corresponding author at: Department of Surgery, University of Dodoma, School of Medicine and Dentistry, Tanzania.

E-mail address: zsaitabau@yahoo.com (Z.S. Abraham).

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3. Discussion

Unlike the rarely encountered calculi within the parotid gland; flow of saliva against gravity, high calcium and mucin content and its more alkaline pH can explain preferential calculi formation in the submandibular gland [10].

Aetiopathogenesis of salivary gland calculi may be due to several factors such as; Wharton's duct being wider in diameter, the flow of salivary secretions in the submandibular gland being against gravity and such secretions being more alkaline compared with those of the parotid gland, the submandibular saliva appears to be more mucinous compared to those of the parotid gland and similarly, submandibular saliva has a higher concentration of calcium and phosphate [9,10].

Moreover, precipitation of mineral salts has been proposed to be implicated in the pathogenesis of salivary gland calculi. Also, several local, chemical, and mechanical factors in the precipitation of the mineral salts are involved.

Presence of infection, inflammation, stasis of saliva, physical trauma, introduction of foreign bodies, and the presence of desquamated epithelial cells appears to be the epicenter for precipitation of mineral salts that consequently plays role in pathogenesis of salivary gland calculi. [3,9] In the late 1950s, Harril et al. proposed that gels formed by coalescence of mucins plays a role in pathogenesis of calculi through mineralization [12]. Similarly, Sherman and McGurk demonstrated the presence of water hardness is not strongly associated with the occurrence of salivary calculi [13].

Epidemiologically, while submandibular gland calculi rarely affect children, it appears to have slight male predominance and usually appears to affect those in their third to sixth decades of life [2,3,5,8,9,11,14–18].

The affected patients normally present with pain either before or during intake of meals and may have a history of recurrent swelling of the involved submandibular gland. (Photograph 1).

Giant calculi are a rare encounter in daily clinical practice and those with dimensions greater than 3 cm are very rare [19]. Such rarity can be depicted from what was documented by Bodner in 2002 who similarly reported that there are only few cases of submandibular gland calculi in the available literatures [1]. Unusually, we extracted a submandibular gland calculus measuring 4 cm from the patient reported in this article (Photograph 2).

It is advisable to perform bimanual palpation and massage of the



Photograph 1. Intraoral appearance of the giant right submandibular duct calculus preoperatively.

affected gland and its duct while observing the nature of saliva such as its flow and whether its clear or not and with such maneuver a calculus can be located anteriorly in the excretory duct [10,18].

Work ups for patients with submandibular gland calculi include head computerized tomography scan where it appears radiopaque in 80–94.7% of cases and also ultrasonography that is widely useful in detecting calculi where about 90% of all sialoliths larger than 2 mm can be detected upon ultrasonography [3,17,20].

In hospital settings, sonography remains to be a standard diagnostic tool and computerized tomography scan is only complementary tool in establishing the diagnosis of salivary calculi though not easily affordable due to its cost but similarly other cost effective work ups like radiographs are also available [15,17].

In circumstances where the calculus is located within the gland then a panoramic radiograph is preferred but when they are small and radiolucent then sialography is recommended [10].

The introduction of minimally invasive surgical procedures has significantly reduced the rate of major salivary gland removal for

Table 1
Comparative table of various cases of operated giant submandibular duct calculi.

Author	Sialolith size	Symptoms/signs	Removal method	Age (years)	Gender
Fowell and McBean	4.1 cm	Pain in the right floor of mouth and submandibular region, exacerbated by swallowing	Excision of the right submandibular gland (SMG) and stone via a standard extra-oral approach	58	Male
Oliveira et al	3.0 cm	Palpation revealed hardness in the right submandibular salivary gland	Intra oral excision of the SMG was done. The sialolith was removed by curettage after direct incision of the duct.	42	Male
Dernegi BB	2.8 cm	Recurrent painful swelling in the left submandibular area. Examination (bimanual palpation) revealed a large firm, mobile, non-tender mass in the left submandibular region	Intra-orally extraction of the SMG calculus was done.	21	Male
Mbalaso et al	4.0 cm	History of a swelling in the right submandibular region and pain associated with purulent discharge. The swelling was found intermittently to wax and wane over some years usually following meals.	Extra-oral excision of the right submandibular gland was done and the calculus extracted following a difficult dissection	45	Male
Fefar et al	3.8 cm	History of the right submandibular swelling associated with pain in the right floor of mouth and the submandibular area. The size of the swelling increased after the meal.	Extra-oral excision of the right submandibular gland was done and a stone of about 3.8 cm was extracted	46	Male
Oteri et al. (Case 1)	2 cm	She had a history of having episodes of left submandibular swelling occurring with meals. These symptoms disappeared within relatively short period, never more	Trans-oral excision of the calculus was done	40	Female
Oteri et al. (Case 2)	1.5 cm	Episodic acute pain in left submandibular area and swelling under the tongue especially at meal times and they remitted within a few hours and exudation was noted in the oral cavity	Intra-oral excision of the calculus was done	51	Female
Abdullah et al	3.6 cm	Complaining of hard swelling in the left submandibular area	Left submandibular intraoral stone extraction with marsuplization of the duct	36	Male



Photograph 2. A giant right-sided submandibular duct calculus post-intraoral surgical removal.

example sialoendoscopy which is the method of choice in small and medium stones and with a high rate of success and gland preservation. Other available techniques include combined transoral and endoscopic approach indicated for large stones in the submandibular gland [1–5,21].

From the available literatures, large calculi have been reported but those measuring more than 3.5 cm are very rare and thus worth to be reported in our report since it measured 4 cm and was found to be encased completely in the submandibular duct.

4. Conclusion

Giant submandibular gland calculi are very rare and they represent a major challenge to oral and maxillofacial surgeons and otorhinolaryngologists especially a desirable surgical approach to minimize the possibility of postoperative complications. Intraoral excision of the calculi should be advocated in routine clinical practice aiming to preserve the gland. This is so far the first reported rare case of giant submandibular gland calculus in an adult Tanzanian patient.

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Ethical approval

Ethical standards were reviewed and approved by the Head of Department of Otorhinolaryngology at Temeke Municipal Hospital.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Registration of research studies

Not applicable.

Guarantor

Dr. Zephania Saitabau Abraham takes full responsibility of the work.

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ZSA-Conceptualization, methodology, writing original draft.

MM-Conceptualization and reviewing the prepared original draft of the manuscript.

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Declaration of competing interest

The authors reports no conflict of interest.

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