

Giant Submandibular Sialolith: A Case Report and Review of Literature

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ABSTRACT

Sialolithiasis is the most common disease of salivary glands. Commonly, sialoliths measure from 5 to 10 mm in size and stones over 10 mm can be reported as sialoliths of unusual size. Giant sialoliths measuring more than 35 mm are extremely rare, with only around 17 cases published in the literature. We report an asymptomatic case of a sialolith in the submandibular duct measuring 62 × 20 mm and weighing 40 gm, highlighting the management aspects and also review the literature.

Keywords: Sialolithiasis, Sialolithotomy, Sialodochoplasty.

INTRODUCTION

Sialolithiasis is the most common (50%) disease of salivary glands. Its estimated frequency is 1.2% (12 in 1000) in the adult population every year, with a slight male predominance (2:1).¹ Most salivary calculi occur in the submandibular gland (80-95%), whereas 5 to 20% occur in the parotid gland.¹⁻³ The sublingual gland and minor salivary glands are rarely (1-2%) affected.¹ Salivary calculi are usually unilateral, clinically they are round or ovoid, rough or smooth with yellow in color.¹ Calculi generally consist of a mixture of different calcium phosphates (mainly hydroxyl-apatite and carbonate-apatite) together with an organic

matrix.⁴ Submandibular stones are 72% inorganic and 18% organic material in nature, whereas parotid stones are composed of 49% inorganic and 51% organic material.¹ About 40% of parotid and 20% of submandibular stones are not radio-opaque and sialography or other imaging techniques (computed tomography scan, ultrasound) may be required to locate them.⁴

Commonly, sialoliths measure from 5 to 10 mm in size and stones over 10 mm can be reported as sialoliths of unusual size. They rarely measure more than 15 mm. Giant sialoliths measuring more than 35 mm are rare, with only around 17 cases published in the literature (Table 1).⁵⁻²²

Table 1: Giant sialoliths (larger than 35 mm) reported in the literature

Sl no	Study	Gender	Age (Years)	Gland	Location	Size (mm)	Weight (gram)
1.	Meyers, 1942 ⁵	Male	50	SM	Duct	50	NR
2.	Mustard, 1945 ⁶	Male	42	SM	Duct	56	NR
3.	Allen, 1956 ⁷	Male	49	SM	Duct	35	NR
4.	Cavina and Santoli, 1965 ⁸	Male	59	SM	Duct	70*	18
5.	Cavina and Santilo, 1965 ⁸	Male	53	SM	Both	60	33
6.	Hoggins, 1968 ⁹	Male	52	SM	Paren	50	NR
7.	Rust and Messerly, 1969 ¹⁰	Male	66	PAR	Duct	51	NR
8.	Rust and Messerly, 1969 ¹⁰	Male	58	NR	Paren	35	NR
9.	Raksin et al, 1975 ¹¹	Male	52	SM	Duct	55	9.5
10.	Isacsson and Nils-Erik, 1982 ¹²	Male	48	SM	Duct	36	NR
11.	Tinsley, 1989 ¹³	Male	48	SM	Paren	50	23.5
12.	Hubar et al, 1990 ¹⁴	Male	65	SM	Duct	52	17.5
13.	Akin and Esmer, 1991 ¹⁵	Male	45	SM	Paren	45	NR
14.	Paul and Chauhan, 1995 ¹⁶	Male	45	SM	Duct	45	4.2
15.	Bodner, 2002 ²⁰	Male	50	SM	Duct	50	NR
16.	CL Montes et al, 2007 ²¹	Male	34	SM	Duct	36	12
17.	Manjunath Rai, 2009 ²²	Male	60	SM	Duct	72	45.8
18.	Present case	Male	50	SM	Duct	62	40

Abbreviations: SM: Submandibular gland; PAR: Parotid gland; Paren: Parenchymal; NR: Not reported.

*this case was described as having "hen's egg" size

Ninety-five percent of the giant sialoliths reported were in the submandibular gland. We report a case of one of the largest submandibular sialoliths in the literature measuring 62 mm and weighing 40 gm, highlighting the management aspects and also communicate the results of a search of the literature on giant sialolith larger than 35 mm.

CASE REPORT

A 50-year-old man was seen in the outpatient clinic of the division of oral and maxillofacial surgery at Government Dental College, Bangalore, India. His main complaint was a swelling in the left side of the floor of the mouth for more than 2 years duration. Clinical history revealed that the patient was in good health; with no other signs, symptoms or abnormalities. Extraoral examination revealed swelling in the left submandibular region with no other significant findings. On intraoral examination, a large, firm, nontender swelling in the left floor of mouth in the region of the submandibular duct was noted with no color changes of the surrounding mucosa (Fig. 1). There was no history of pain and or swelling aggravating during meals. Mandibular occlusal radiograph showed a large J-shaped radiopaque mass extending back beyond the lower left 1st permanent molar located within the Wharton's duct (Fig. 2). A diagnosis of left submandibular duct sialolith was made.

After induction of local anesthesia, giant sialolith was removed in a minimally invasive manner via transoral sialolithotomy and sialodochoplasty. Following isolation of the duct, longitudinal incision was placed into the duct over the stone, sharp dissection was done and sialolith was exposed. Direct cut-down was avoided because of the risk of ductal stenosis. The larger (Fig. 3) and subsequently remaining portion of the sialolith was delivered out with sinus forceps taking care to protect the lingual nerve. It was measured to be 62 × 20 mm; weighed around 40 gm, off white to yellow in color and cross-section showed central nidus surrounded by concentric layers (Fig. 4).

After a 3-year follow-up, the patient was asymptomatic with satisfactory glandular function and normal, undisturbed salivary flow.

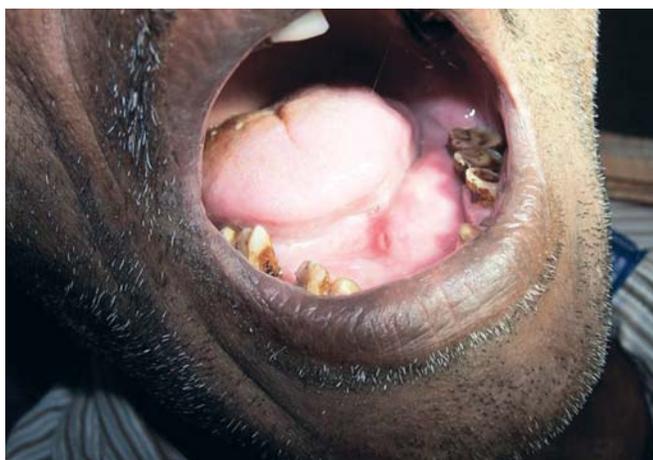


Fig. 1: Intraoral examination showing a large, firm, nontender swelling in the left floor of mouth



Fig. 2: Mandibular occlusal radiograph revealing a large radiopaque mineralized body extending back beyond the lower left 1st permanent molar



Fig. 3: Large stone after removal measuring around 62 × 20 mm (including the bent portion)



Fig. 4: Sialolith off white to yellow in color and cross-section showed central nidus surrounded by concentric layers

DISCUSSION

Submandibular sialoliths measuring less than 10 mm in greatest dimension are very common but larger sialoliths

are considerably less.²³ To our best knowledge, the sialolith presented in this case is one of the largest reported to date, as compared with the published data (Table 1). No report has mentioned about the width which in this case was almost an inch. Other cases of large calculi cited by Zakaria¹⁷ and Brusati and Fiamminghi¹⁸ are known by their weight only which in present case is 40 gm.

Demographic Variables

Giant sialoliths are rare findings in clinical oral pathology with sizes ranging from 35 to 72 mm and all of them occurring in male patients (Table 1)⁵⁻²² and so is the present case. All the patients were older than 42 years except the case presented by Ledesma Montes C et al²¹ where it was 34 years. In present case patient is 50 years old. Sialolithiasis in children is considered rare.¹⁹

Etiology and Pathogenesis

The exact etiology and pathogenesis of salivary calculi is largely unknown. They are thought to occur as a result of deposition of calcium salts around an initial organic nidus consisting of altered salivary mucins, bacteria and desquamated epithelial cells.¹ Traditional theories suggest that formation of calculus occurs in two phases; a central core and a layered periphery. The central core formed by precipitation of salts which are bound by certain organic substances. The second phase consists of layered deposition of organic and nonorganic material. Another theory has proposed that an unknown metabolic phenomenon can increase the salivary bicarbonate content, which alters calcium phosphate solubility and leads to precipitation of calcium and phosphate ions.¹ A retrograde theory has proposed that aliments, substances or bacteria within the oral cavity might migrate into the salivary ducts and become the nidus for further calcification.^{1,2} Marchal et al²⁴ further suggested that variation in sphincter-like mechanism in the first 3 cm of Wharton's duct can be responsible for easier retrograde migration of oral materials. Reduced salivary flow due to stagnation, dehydration and change in salivary pH associated with oropharyngeal sepsis, introduction of foreign bodies, impaired crystalloid solubility² and physical trauma to salivary duct or gland may predispose to calculus formation.^{1,7,8} Recently, Sherman and McGurk²⁵ showed that water hardness is not significantly associated with the incidence of salivary calculi.

Location

With the exception of one case, all the giant sialoliths were located in the submandibular gland (94.44%) and only an isolated case was found within the Stensen's duct of the parotid salivary gland. Several factors that may contribute to the increased incidence of sialoliths in the submandibular gland include as follows:

1. The submandibular excretory duct is wider in diameter and longer than the Stenson's duct,⁷ has an antigravity

flow^{1,21} and the narrow orifice all of which encourage stagnation of saliva.²³

2. The submandibular saliva contains a higher quantity of mucin proteins and increased content of calcium and phosphate.^{1,21,23}
3. Submandibular salivary secretion is more alkaline compared with pH of the parotid saliva.^{1,21,23}

Diagnosis

The diagnosis of salivary calculi is based on patient's history and clinical examination which are supplemented by radiologic findings.²⁶ Patient may present with pain and swelling of the concerned gland at meal times and in response to other salivary stimuli, pus may be seen draining from the duct and signs of systemic infection.¹ Patients with small and sometimes large sialoliths can be asymptomatic¹ as noted in the current case. Bimanual palpation of the floor of the mouth in a posterior to anterior direction reveals palpable stone in a large number of cases in submandibular sialolith.¹ For parotid stones, careful intraoral palpation around Stenson's duct orifice may reveal a stone. Deeper stones are often not palpable.¹

Sialoliths are well visualized on panoramic and periapical radiographs but can be obscured with super imposition over the roots of premolar and molar teeth and hence, better visualized by an occlusal radiograph without overlap from other anatomy.^{4,23} Given the posterior-inferior position of the gland, a mandibular lateral oblique radiograph may be useful for visualization.²³ Other imaging techniques that may be used to diagnose sialoliths include sialography, ultrasound, computed tomography and magnetic resonance sialography.²³ Sialography is useful in patients with sialadenitis, radiolucent stones or deep submandibular/parotid stones. It is, however, contraindicated in acute infections or in significant patient contrast allergy.¹ Ultrasound will locate a sialolith within the gland with the subsequent requirement for conventional imaging.^{14,23}

Computed tomography is a noninvasive technique¹¹ useful in any situation where there are multiple stones or when the stone is in a sight not readily examined intraorally,²³ but scans do not provide the precise localization of a sialolith within the duct system.² Scintigraphy though not indicated for the diagnosis of sialolithiasis but may be useful as a complimentary, exploratory technique where sialography is not indicated.¹¹

Recent advances in optical technology have led to the development of sialoendoscopy, a new diagnostic means of directly visualizing intraductal stones; allow an almost complete exploration of the duct system² and to determine the exact location of the obstruction.²⁷

Management

The treatment options for salivary calculus are generally dictated by the position of the stone.²⁶ When the stone is small

it may be removed by application of moist warm heat and gland massage, while sialogogues can be used to promote saliva production and flush the stone out of the duct.¹ Salivary calculi which lie in the distal third of the duct are amenable to simple surgical release through an incision in the floor of the mouth where incision is made directly onto the stone in the longitudinal axis of the duct. If the stone is sufficiently forward it can be milked and manipulated through the duct orifice. Sialolithotomy within the duct posterior to the first molar or even more proximally in the so-called comma area is difficult and may be hazardous to the lingual nerve. Nevertheless, expanded incision of the duct has been continually favored as a gland preserving therapy.³ If the gland has been damaged by recurrent infection and fibrosis or calculi have formed within the gland, then the gland may require removal.^{1,20} Sialoadenectomy is the mainstay of the surgical management for the majority of intraglandular stones.^{1,22}

Alternative methods of treatment have emerged such as shock wave lithotripsy, sialoendoscopy, interventional radiology, endoscopically video-assisted transoral retrieval of stones² and intraductal laser fragmentation.²⁸

In sialolithotripsy, the calculus is broken and the fragments are subsequently washed down the duct⁵ without damaging the adjacent tissues.¹ Extracorporeal lithotripsy has limitations; stones in the middle third of the submandibular ducts are closely related to mandible and thus can not be targeted. Intracorporeal lithotripsy has a drawback of ductal perforation.²⁶ Sialoendoscopy initially used for diagnostic purposes, is now scheduled for intervention in the obstructive salivary gland diseases, the only absolute contraindication to the procedure is complete distal obliteration of the duct that is impenetrable by the endoscope.²

CONCLUSION

Asymptomatic giant sialolith of remarkable size may pose both diagnostic and therapeutic challenge for the clinician. They are usually seen in males during 4th to 5th decade of life predominantly involving submandibular gland. Transoral sialolithotomy with sialodochoplasty or sialoadenectomy remains the mainstay of management. Newer treatment modalities are effective alternatives to conventional surgical treatment for smaller but probably not giant sialoliths.

REFERENCES

- Siddiqui SJ. Sialolithiasis and unusually large submandibular salivary stone. *Br Dent J* 2002;193:89-91.
- Capaccio P, Torretta S, Ottaviani F, Sambataro G, Pignataro L. Modern management of obstructive salivary diseases. *Acta Otorhinolaryngol Ital* 2007;27:161-72.
- Johannes Zenk, Jannis Constantinidis, Basel Al-kadah, Heinrich Iro. Transoral removal of submandibular stones. *Arch Otolaryngol Head Neck Surg* 2001;127:432-36.
- Andretta M, Tregnaghi A, Prosenikliev V, Staffieri A. Current opinions in sialolithiasis diagnosis and treatment. *Acta Otorhinolaryngol Ital* 2005;25:145-49.
- Meyers H. Large calculus in submaxillary gland. *Dent Dig* 1942;48:420.
- Mustard TA. Calculus of unusual size in Whartons duct. *Br Dent J* 1945;79:129.
- Allen NE. A sialolith within the submaxillary duct and gland. Report of a case. *J Oral Surg* 1965;14:65.
- Cavina C, Santoli A. su alcuni casi di calcolosi salivare di particolare interesse. *Minerva Stomatol* 1965;pp 190.
- Hoggins GS. Large calcified mass in the submaxillary gland. *Oral Surg Oral Med Oral Pathol* 1968;25:679.
- Rust TA, Messerly CD. Oddities of salivary calculi. *Oral Surg Oral Med Oral Pathol* 1969;28:862.
- Raskin SZ, Gould SM, William AC. Submandibular gland sialolith of unusual size and shape. *J Oral Surg* 1975;33:142.
- Issacson G, NILS-Erick P. The gigantiform salivary calculus. *Int J Oral Surg* 1982;11:135.
- Tinsley G. An extraordinary large asymptomatic submandibular salivary calculus. *Br Dent J* 1969;166:199.
- Hubar JS, Guggenheimer J, Evan M. Megalith. *Oral Surg Oral Med Oral Pathol* 1990;70:245.
- Akin I, Esmer M. A submandibular sialolith of unusual size. *J Otolaryngol* 1991;20:123.
- Paul D, Chauhan MS. Salivary megalith with a sailo-cutaneous and sailo-oral fistula. A case report. *J Laryngol Otol* 1965;109:767.
- Zakaria MAK. Giant calculi of the submandibular salivary gland. *Br J Oral Surg* 1981;19:230.
- Brusati R, Fiamminghi L. Large calculus of the submandibular gland. Report of case. *J Oral Surg* 1973;31:710.
- Lustman J, Regev E, Melamed Y. Sialolithiasis: A survey on 245 patients and review of literature. *Int J Oral Maxillofac Surg* 1990;19:135.
- Bodner I. Giant salivary gland calculi: Diagnostic imaging and surgical management. *Oral Surg Oral path Oral Med Oral Radiol Endod* 2002;94:320.
- Ledesma-Montes C, Garces-Ortiz M, Salcido-Garcia JF, et al. Giant sialolith: Case report and review of literature. *J Oral Maxillofac Surg* 2007;65:128-30.
- Manjunath Rai, Richi Burman. Giant submandibular sialolith of remarkable size in the comma area of Wharton's duct: A case report. *J Oral Maxillofac Surg* 2009;67:1329-32.
- Huang TC, Dalton JB, Monsour FN, Savage NW. Multiple, large sialoliths of the submandibular gland duct: A case report. *Dent J* 2009;54:1-65.
- Sherman JA, McGurk M. Lack of correlation between water hardness and salivary calculi in England. *Br J Oral Maxillofac Surg* 2000;38:50.
- Marchal F, Kurt AM Dulgerov P, et al. Retrograde theory in sialolithiasis formation. *Arch Otolaryngol Head Neck Surg* 2001;127:66.
- Drage Nicholas A, Brown Jackie E, Escudier Michael P, McGurk Mark. Interventional radiology in the removal of salivary calculi. *Radiology* 2000;214:139-42.
- Oded Nahlieli, Liat Hecht Nakar, Yaron Nazarian, Michael Turner D. Sialoendoscopy: A new approach to salivary gland obstructive pathology. *J Am Dent Assoc* 2006;137:1394-1400.
- Walvekar Rohan R, Bomeli Steven R, Carrau Ricardo L, Schaitkin Barry. Combined approach technique for the management of large salivary stones. *Laryngoscope* 2009;119:1125-29.