ISSN 2307-8960 (online)

World Journal of *Clinical Cases*

World J Clin Cases 2023 August 6; 11(22): 5193-5415

Published by Baishideng Publishing Group Inc

World Journal of Clinical Cases

Contents

Thrice Monthly Volume 11 Number 22 August 6, 2023

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NAME OF JOURNAL
World Journal of Clinical Cases

ISSN
ISSN 2307-8960 (online)

LAUNCH DATE
April 16, 2013

FREQUENCY
Thrice Monthly

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PUBLICATION DATE
August 6, 2023

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World Journal of Clinical Cases

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World J Clin Cases 2023 August 6; 11(22): 5382-5390

DOI: 10.12998/wjcc.v11.i22.5382

ISSN 2307-8960 (online)

CASE REPORT

Long-term rare giant sialolithiasis for 30 years: A case report and review of literature

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Specialty type: Medicine, research and experimental

Provenance and peer review: Unsolicited article; Externally peer reviewed.

Peer-review model: Single blind

Peer-review report's scientific quality classification

Grade A (Excellent): 0
Grade B (Very good): B
Grade C (Good): C
Grade D (Fair): 0
Grade E (Poor): 0

P-Reviewer: Corvino A, Italy; Ozcan C, Turkey

Received: May 4, 2023
Peer-review started: May 4, 2023
First decision: May 25, 2023
Revised: June 1, 2023
Accepted: July 6, 2023
Article in press: July 6, 2023
Published online: August 6, 2023

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Abstract

BACKGROUND

Sialolithiasis is one of the most common salivary gland disorders, most commonly affecting the submandibular gland. Submandibular sialolithiasis can be treated using non-invasive conservative measures and invasive treatments. Treatment selection was based on the ductal system anatomy and the size and location of the stones. This study aimed to review the updates on sialolithiasis treatment and compare the different management strategies of the variables.

CASE SUMMARY

This report presents a case of a long-term, rare, and giant sialolithiasis within the submandibular gland parenchyma for 30 years in an older adult. Our patient presented with painless right submandibular swelling. Computed tomography revealed a calcified mass measuring 35 mm × 20 mm within the right submandibular gland. In this case, the infection and fibrosis of the affected gland and size of the stone did not provide us with other alternatives except for the excision of the involved gland. Thus, right submandibular sialoadenectomy was performed *via* the transcervical approach. After the surgery, the patient recovered without any complaints, side effects, or complications.

CONCLUSION

Tailored management is important for preserving gland function, maintaining low risk, and reducing patient discomfort.

Key Words: Huge sialolithiasis; Submandibular gland; Treatment; Complications; Case report

Core Tip: Giant sialoliths are uncommon, and their long duration is rarely documented. This report presents a case of a long-term, rare, and giant sialolithiasis within the submandibular gland parenchyma for 30 years in an older adult. We reviewed 44 cases of giant sialoliths that were reported between 1981 and 2022. Based on the comparative table, our case is the second largest with the longest duration. This case highlights the importance of proper diagnosis and treatment.

Citation: Mao JS, Lee YC, Chi JCY, Yi WL, Tsou YA, Lin CD, Tai CJ, Shih LC. Long-term rare giant sialolithiasis for 30 years: A case report and review of literature. *World J Clin Cases* 2023; 11(22): 5382-5390
URL: https://www.wjgnet.com/2307-8960/full/v11/i22/5382.htm
DOI: https://dx.doi.org/10.12998/wjcc.v11.i22.5382

INTRODUCTION

Sialoliths are calcified salivary stones that can form within the secretory system of the salivary glands. Although various hypotheses have been proposed, the exact etiology of these sialoliths remains unclear. Most patients with sialolithiasis present with symptoms such as local pain, focal swelling, trismus, xerostomia, and lumps in the throat. Inflammation with oozing pus may be noted in some cases due to secondary infection.

The submandibular gland is the most commonly affected site for sialoliths, accounting for approximately 80%-90% of cases[1]. The rate of giant sialolith formation within the submandibular gland is high, and several contributing factors have been proposed, including a longer Wharton's duct, larger duct caliber, and tortuous course of the Wharton's duct accompanied by a slower salivary flow rate within the submandibular gland compared with that in other salivary ducts. The slower salivary flow rate is caused by saliva in the submandibular glands flowing against gravity, and the dependent position of the gland makes it more prone to stasis. Additionally, owing to the higher calcium and phosphate levels, the saliva within the submandibular glands tends to be more alkaline and may induce sialolithiasis[2].

Sialoliths usually range from 1 mm to 10 mm in size, and those measuring 15 mm in one dimension are classified as giant sialoliths[3]. In this report, we present a case of giant right submandibular sialolithiasis that was treated by surgical ablation for approximately 30 years and compare various giant sialolith cases and their different treatments with respect to variables such as size, duration, and symptom presentation.

CASE PRESENTATION

Chief complaints

A 75-year-old woman presented to our institution with painless right submandibular swelling with a persistence of 30 years.

History of present illness

The clinical presentation was mostly asymptomatic, with only slight facial asymmetry and occasional right-sided aural fullness. However, the indurated mass began to manifest focal swelling 1 mo prior, with intermittent severe pain that radiated to the ipsilateral neck and contralateral face. The additional onset of dysphagia, odynophagia, and a subsequent decrease in appetite were also noted but without associated postprandial swelling or xerostomia.

History of past illness

The patient had no known medical history.

Personal and family history

The patient denied alcohol or cigarette use and confirmed a family history of sialolithiasis.

Physical examination

Bimanual palpation revealed a raised right erythematous submandibular mass measuring 40 mm × 30 mm overlying the skin. The swelling was indurated, tender on palpation, and firm in consistency. No other obvious cervical lymphadenopathies could be palpated. Intraorally, no stones or purulent discharge was secreted from the Wharton's duct.

Laboratory examinations

White blood cells count levels were elevated (13800/μl).

Imaging examinations

Computed tomography (CT) revealed a calcified mass measuring 35 mm × 20 mm within the right submandibular gland (Figure 1).

FINAL DIAGNOSIS

Combined with the patient's medical history, the final diagnosis was submandibular sialolithiasis.

TREATMENT

Excision of the right submandibular gland and sialoliths was performed using a standard extraoral approach. Intraoperative findings revealed that the right submandibular gland was indurated and filled with giant sialoliths. Neighboring structures, such as the hypoglossal, lingual, and marginal mandibular nerves, were preserved (Figure 2).

OUTCOME AND FOLLOW-UP

The postoperative was uneventful, with no complications. Histopathological examination confirmed the diagnoses of sialolithiasis and chronic sialadenitis. A giant, yellow, hard sialolith measuring 25 mm × 17 mm × 17 mm was noted. Microscopically, the submandibular gland showed varied dilatations of the salivary ducts (ductal ectasia). Ulcerative changes in the dilated ductal epithelium with areas of squamous metaplasia, periductal fibrosis, and moderately mixed acute and chronic inflammatory infiltrates with lymphocytic aggregate foci were also observed.

DISCUSSION

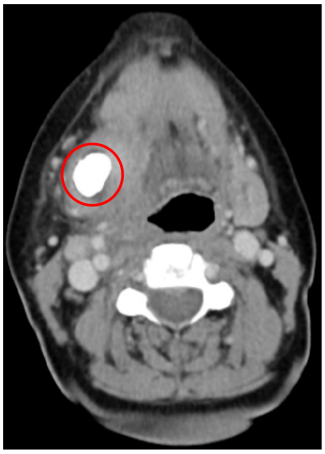
Several treatment modalities for submandibular sialolithiasis exist; however, the primary goal is to preserve gland function, maintain low risk, and reduce patient discomfort[4]. Non-invasive conservative measures for submandibular sialolithiasis include glandular focal massage and trans-oral removal. Glandular focal massage is important during or after the administration of sialagogues, and irrigation is primarily indicated in cases of small intraductal stones. Transoral removal can be considered if stones are palpated bimanually and/or localized within the perihilar region of the gland using ultrasonography[4,5]. If a superimposed infection is suspected, antibiotics should be prescribed and, when warranted, combined with simple sialolithotomy[5,6].

Invasive management of submandibular sialolithiasis consists of open surgery (which can be performed with transoral duct surgery[7-9] or a transcervical approach to the submandibular space[10], extracorporeal shock wave lithotripsy[11-15], interventional sialendoscopy, and intraductal shock-wave lithotripsy[16-19]. These invasive methods are recommended for stones > 7 mm with symptoms[20]. Treatment selection is mostly based on ductal system anatomy and the location of the stones[7].

Transoral duct surgery may be considered for stones of all sizes in the papilla, owing to the narrow ostium of the papilla and stones in the distal and middle ducts. Small mobile stones (not exceeding 3-5 mm) in the papilla distal and middle ducts can also be extracted using interventional sialendoscopy. The proximal duct, however, is curved and narrow; thus, interventional sialendoscopy is difficult to perform. In cases where the stones are immobile, impacted, palpable, or large, transoral duct surgery is recommended, although mechanical fragmentation or intraductal shockwave lithotripsy followed by fragment extraction is also an option. Intraparenchymal stones are rare; however, if small, mobile stones (not exceeding 3–5 mm) can be visualized during endoscopy, interventional sialendoscopy, including mechanical fragmentation or intraductal shock wave lithotripsy, is recommended. Transoral duct surgery with submandibulectomy is indicated for large palpable stones (10 mm) located posthilar to the intraparenchymal region. Finally, salivary gland massage combined with a sour diet and sialogogue use is recommended after the removal of salivary stones[5].

In summary, transoral duct surgery offers better accessibility to the duct system and remains the preferred treatment modality; however, intraductal shock wave lithotripsy has gradually replaced transoral duct surgery in cases of smaller salivary stones. Compared with an earlier algorithm for the management of submandibular stones published in 2009, the indications for extracorporeal shockwave lithotripsy have since been adjusted, and the newly developed intraductal shockwave lithotripsy is also available. Although extracorporeal shockwave lithotripsy is considered less important, the procedure remains an option for intraparenchymal stones that are not visible or accessible. Finally, botulinum toxin injections may be considered in situations where the patient is deemed inoperable[7].

Among all the invasive treatment methods, adenectomy has a higher success rate than other techniques. Open surgery *via* the transcervical approach has a lower risk of infection because the oral space does not communicate with the neck space, whereas the intraoral approach avoids facial scarring. However, open surgical approaches are associated with



DOI: 10.12998/wjcc.v11.i22.5382

Figure 1 Axial view of the head and neck computed tomography with contrast showing an irregular opacity of 35 mm 20 mm in the parenchyma of the right submandibular gland.

several complications. Examples include postoperative neurological damage, particularly hypoglossal, lingual, and marginal mandibular nerve injuries, hemorrhage, hematoma, and disfiguring scars. Temporary marginal mandibular nerve damage is the most common complication of transcervical submandibular gland surgery. This complication was reported to be 36% by Smith *et al*[21], and permanent damage was reported to be as high as 12% by De *et al*[22]. in the literature. Beahm *et al*[23] reported rates between 7.7% and 36% in a previous literature revie. To reduce the risk of marginal mandibular nerve injury in the transcervical surgical approach to the submandibular gland, three surgical maneuvers are recommended: (1) The gland was identified to be lower than the hyoid bone, and the marginal mandibular nerve was not examined; (2) The marginal mandibular nerve is located at the level at which it leaves, tracks, and protects the parotid tail; and (3) The facial veins are located at a lower level and ligated, and the nerve is suspended by elevation; thus, the marginal mandibular nerve is removed from the surgical area.

However, Smith *et al*[21] reported that when a lower approach was used, no permanent marginal mandibular nerve palsy occurred, although 36% of the nerves were temporarily dysfunctional. This may have been due to stretching during the lower surgical approach to the glands. The hypoglossal nerve is partially sheltered by the posterior belly of the digastric muscle, which is located in the inferomedial portion of the lower one-third of the submandibular glands. This may explain why hypoglossal nerve damage has been reported very rarely (0%-1.4%)[24] compared with marginal mandibular nerve injuries. In this case report, no permanent or temporary complications occurred during the postoperative follow-up. Although the submandibular glands contribute 69% of the salivary secretion volume in the resting state[25], xerostomia was not reported in our case. This could be explained by the fact that other salivary glands were sufficiently functional to compensate for the resected glands.

Moreover, regarding the oncological risk to which patients with chronic inflammation and lithiasis are exposed[26], we reviewed some studies on salivary gland tumors. Most salivary gland tumors arise in the parotid gland (70%), followed by the minor salivary glands (22%) and submandibular glands (8%)[27]. Differential diagnoses for benign submandibular tumors include pleomorphic adenomas, Warthin's tumors, and oncocytomas. Histopathological characteristics can be used to distinguish between them. Pleomorphic adenomas are characterized by thick irregularly marginated capsules[28,29]. Oncocytomas have thin capsules and monomorphic oncocytes without mitosis or necrosis[29,30]. The lymphatic population is the most common cytology and histology of Warthin's tumor[28,29]. Histopathological examination of our patient did not reveal the above characteristics, and the characteristics of malignant transformation include local invasion into the muscular, perineural, and lymphatic structures, as well as microscopic features, including nuclear atypia, cellular polymorphism, mitoses, and focal necrosis, were absent too. Thus, salivary gland tumors were excluded.

Owing to the similarities in the clinical presentation of benign salivary tumors and submandibular sialolithiasis, radiological imaging is essential to distinguish between the two entities. Ultrasound is recommended for the initial assessment of salivary gland abnormalities; however, ultrasound is insufficient because information on the surrounding structures cannot be provided. Its accuracy in the identification of benign salivary lesions can be increased using elastography or contrast-enhanced ultrasound; however, the European Federation of Societies for Ultrasound in Medicine and Biology do not recommend contrast-enhanced ultrasound for the characterization of salivary gland lesions in clinical practice. Thus, further studies are required to investigate the diagnostic roles of contrast-enhanced ultrasound and elastography in the evaluation of salivary gland lesions. Corvino *et al*[26] described the utility of CT and magnetic resonance imaging in the initial staging, histologic grading of salivary gland malignancies, and preoperative planning. Positron emission tomography with F-18 fluorodeoxyglucose is useful for the evaluation and clinical management of head and neck lesions[26].

Giant sialoliths are rare and classified as those measuring 15 mm in one dimension[3]. We compared cases of giant sialoliths in the submandibular gland from 1981 to 2022 and their different treatments according to variables such as size, duration, and symptom presentation. The cases and their details are listed in Table 1.

Table 1 Comparative table of various cases of giant submandibular sialolithiasis with different treatments

Ref.	Year	Size (mm)	Symptoms/signs	Duration	Treatment	Complication
Zakaria[35]	1981	33	Swelling in the right submandibular region, increasing pain during meals	20 yr	The right submandibular gland was excised	None
Naraynsingh[36]	1985	60	Painless hard left submandibular swelling	3 yr	Incision of left submandibular gland *via* a transcervical approach	None
Frame and Smith [37]	1986	30	Recurrent pain and swelling of the right side of the face	N/A	Excise the gland and calculus	None
		25	Slight discomfort in the area	N/A	Sialo-adenectomy *via* a standard incision	None
Kaltman and Eichner[38]	1987	45	Pain and swelling over the left submandibular area	15 yr	Standard extraoral approach	None
Raveenthiran and Hayavadana Rao [39]	2004	35	Painless swelling in the floor of her mouth	N/A	Transoral sialolithotomy	None
Yildirim[40]	2004	30	Huge, firm mass below the right angle of the mandible	N/A	Excised *via* extraoral approach	None
Chan and Patel[41]	2006	35	Painful mass in the left floor of the mouth	N/A	Transoral sialodochoplasty and removal calculus	None
Graziani *et al*[42]	2006	22	Asymptomatic	N/A	Transorally	None
Ledesma-Montes *et al*[43]	2007	36	Painful mass located on the right side of the floor of the mouth	12 yr	Surgical excision of the sialolith	None
Biddle and Arora [44]	2008	26	Persistent draining wound over the right submandibular region, odynophagia, and dysphagia were noted	2 yr	Excised *via* standard incision	None
Rai and Burman[45]	2009	72	Episodes of pain for last 2 yr, but severe pain and swelling in the left lower submandibular region	1 mo	Sialolithotomy with sialodocho-plasty *via* an intraoral approach	None
Soares *et al*[46]	2009	25	Hard elongated mass along the right Wharton's duct and a reduced salivary flow	4 mo	Dissected with the patient under local anesthesia	None
Rivera-Serrano and Schaitkin[47]	2011	23	Chronic unrelenting purulent discharge from bilateral submandibular ducts	N/A	Transoral sialolithotomy approach	None
Krishnan *et al*[48]	2009	34	Recurrent pain and swelling	8 yr	Transoral approach	None
Emir *et al*[49]	2010	35	Mass on the right side of the floor of the mouth and fistulization of the Wharton duct into the oral cavity	N/A	Removed transorally from the opening of the fistula at the floor of the mouth	None
Shetty and Sharma [50]	2010	27	Firm painful swelling in the left submandibular region	1 mo	Gentle extraction	None
Boffano and Gallesio[51]	2010	22	Swelling and pain in the submandibular right region	N/A	Removal of the stone and sialodochoplasty were done *via* an intraoral approach	None
Arunkumar *et al*[52]	2015	20	Recurrent right submandibular swelling with pain during meals and gradual regression	8 mo	Removed intraorally	None
Pandarakalam *et al* [53]	2013	40	Swelling, gradually increased in size and became painful recently	4 yr	Excision of the right submandibular salivary gland	None
Iqbal *et al*[54]	2012	35	Hard swelling in the anterior oral floor of mouth	N/A	Intraoral incision	None
Ben-Shoshan and Lacroix[31]	2014	17	Mild pain in the submandibular area	1 yr	Fell out without any intervention	None
Gupta *et al*[3]	2013	28	Intermittent, dull aching pain, and swelling in left submandibular area	3 mo	Intraoral approach	None
Bhullar *et al*[55]	2015	31	Pain in the left submandibular region associated with meals	6 mo	Transoral sialolithotomy	None
Akinyamoju and Adisa[32]	2015	44	Hard, painless, left floor of mouth swelling, moderate pain at mealtimes	1 yr	Removed non-surgically	None

Shahoon *et al*[56]	2015	55	Difficulty breathing and a foul-tasting mouth since 21 days previously with history of right submandibular swelling episodes	21 d	Surgical removal	None
Arslan *et al*[57]	2015	35	Recurrent pain, swelling, and redness in the left submandibular area	6 mo	Submandibular gland resection	None
Oliveira *et al*[58]	2016	30	Pain over the right floor of mouth and submandibular region	N/A	Excision of the right submandibular gland and stone *via* standard extraoral approach	None
Weinberg and Albers[59]	2016	38	Swelling at the submandibular region, pus from Wharton's duct was noted	N/A	Excision	None
Gadve *et al*[60]	2016	25	Pain and swelling in the floor of mouth on the left side	1 mo	Remove the sialolith surgically	None
Goh *et al*[61]	2016	35	Left submandibular swelling, increasing pain, fever and discharge was noted	5 yr	Submandibulectomy with transcervical approach	None
Omezli *et al*[62]	2016	37	Painful and persistent purulent discharge from unilateral submandibular ducts	N/A	Intraoral approach	None
Lim *et al*[63]	2017	50	Right submandibular swelling, increased in size rapidly causing discomfort and pain	10 yr	Right submandibulectomy	None
Iwai *et al*[64]	2017	41	Right submandibular swelling several times	40 yr	Submandibular sialoadenectomy	None
Sakthivel *et al*[65]	2017	50	Recurrent pain and swelling in the right submandibular region	6 mo	Intraoral removal of stone	None
Rodrigues *et al*[33]	2017	45	Painless mass in the neck	3 yr	Removed without anesthesia or surgical excision	None
Singh *et al*[66]	2020	42	Swelling over right side of floor of mouth and pain during meals	2 yr	Sialolithotomy and marsupialization of Wharton's duct	None
Abraham *et al*[67]	2021	40	Hard mass beneath the tongue with occasional dull pain	N/A	Transoral surgical removal of the right submandibular calculus	None
Thong *et al*[68]	2021	25	Right submandibular swelling	2 wk	Excision of right submandibular gland	None
Kumar *et al*[69]	2021	28	Firm mass below the left angle of the mandible	4 yr	Excision of the left submandibular gland and stone *via* trans-cervical approach	None
Brooks *et al*[34]	2021	16&19	Firm, ovoid swelling in the floor of the mouth approximate to the left sublingual caruncle with thin, slightly pale overlying mucosa	N/A	Patient declined any additional diagnostic measures	None
Ungari *et al*[70]	2022	23	Painful swelling in the left side of the mouth floor	20 yr	Excision of the left submandibular gland and stone *via* trans-cervical approach	None
Mohsin *et al*[71]	2022	28	Moveable, firm, extensive non-tender swelling in the posterior floor of the mouth	15 yr	Incision of the right submandibular duct's longitudinal axis and removed the stones	None

Between 1981 and 2022, 44 giant sialoliths were noted. The mean giant sialoliths size is 34.70 mm (two cases were within the range of 15-19 mm; 14 cases within 20-29 mm; 15 cases within 30-39 mm; seven cases within 40-49 mm; three cases within 50-59 mm; one case within 60-69 mm and one case within the range 70-79 mm). The median duration of giant sialoliths was 2 years (11 cases were less than a year; 9 cases were within the range of 1-5 years; two cases were within 6-10 years; three cases were within the range of 11-15 years; two cases were within the range of 16-20 years; one case was within the range of 36-40 years; and 15 cases did not discuss the duration). Of the 43 tumors, 23 and 21 were located in the right and left submandibular glands, respectively. Of the giant sialoliths, 90.70% were surgically removed (incision or excision), either intra-orally or extra-orally (transcervical), whereas 9.30% were removed using other methods. The patients with giant sialoliths reported by Ben-Shoshan and Lacroixl[31] did not undergo any intervention. Akinyamoju and Adisa[32] and Rodrigues *et al*[33] removed the stones non-surgically. Brooks *et al*[34] reported that a patient declined any additional diagnostic measures and was lost at follow-up. No postoperative complications were observed in these cases.

CONCLUSION

Giant sialoliths are rare, and their longer durations have rarely been documented. This report presents a case of long-term, rare, and giant sialolithiasis within the submandibular gland parenchyma for 30 years in an older adult. Our patient

DOI: 10.12998/wjcc.v11.i22.5382

Figure 2 Excised sialolith and right submandibular gland.

presented with typical clinical and radiographic findings. In this case, the infection and fibrosis of the affected gland and size of the stone provided us with no other alternatives except for the excision of the involved gland. Thus, right submandibular sialoadenectomy was performed *via* the transcervical approach. After the surgery, the patient recovered smoothly without any complaints, side effects, or complications. Based on the comparative table, this is the second largest case with a long duration that we would like to share. This case highlights the importance of proper diagnosis and treatment. Clinicians are advised to tailor management according to the patient and to keep in mind that the primary goal should always be to preserve gland function, maintain low risk, and reduce patient discomfort.

FOOTNOTES

Author contributions: Mao JS and Lee YC conceived the study and contributed to protocol development, data collection, data analysis, interpretation of results, and drafting of the article; Mao JS and Lee YC collected the data; Chi JCY, Yi WL, Tsou YA, Lin CD, Tai CJ, and Shih LC provided critical feedback, direction, and article revisions; Mao JC, Lee YC, and Shih LC contributed to the protocol development, interpreted the results, and finalized the manuscript; all authors have approved the final version of the manuscript submitted for publication; Mao JS and Lee YC contributed equally to this study.

Supported by The China Medical University Hospital, No. DMR-110-242 and No. DMR-110-057.

Informed consent statement: All study participants or their legal guardians provided written informed consent prior to study enrollment.

Conflict-of-interest statement: The authors declare no conflict of interest.

CARE Checklist (2016) statement: The authors have read the CARE Checklist (2016), and the manuscript was prepared and revised according to the CARE Checklist (2016).

Country/Territory of origin: Taiwan

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S-Editor: Lin C
L-Editor: A
P-Editor: Zhao S

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