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Long-term Rare Huge Sialolithiasis for 30 Years: A case report and literature review

Sialolithiasis Submandibular gland

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Abstract

BACKGROUND

Sialolithiasis is one of the most common salivary gland disorders, with the submandibular gland being the most affected. Submandibular sialolithiasis can be treated with noninvasive conservative measures and invasive treatments. Treatment selection was based on the ductal system anatomy and size and location of the stones. The purpose of this paper is to review the updates on sialolithiasis treatment, along with a comparative table of their different management of variables.

CASE SUMMARY

This report presents a case of long-term, rare, and huge sialolithiasis within the submandibular gland parenchyma for 30 years in an older adult. Our patient presented with painless right submandibular swelling. Computed tomography (CT) 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 the size of the stone left us with no other options except for 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.

CONCLUSION

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

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

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Core Tip:

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 cases of sialolithiasis present with symptoms such as local pain, focal swelling, trismus, xerostomia, and lumps in the throat. Inflammation with pus oozing 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 contributory factors have been proposed, including a longer Wharton's duct, larger duct caliber, and the tortuous course of the Wharton's duct accompanied by a slower salivary flow rate within the submandibular gland compared to other salivary ducts. This is because saliva in the submandibular glands flows against gravity, and the dependent position of the gland makes it more prone to stasis. Additionally, owing to higher calcium and phosphate levels, saliva within the submandibular glands tends to be more alkaline and may induce sialolithiasis in the submandibular gland.[2]

Sialoliths usually range from 1 mm to less than 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 for approximately 30 years that was treated by surgical ablation and compare various giant sialolith cases and their different managements with respect to variables such as size, duration, and symptom presentation.

CASE PRESENTATION

Chief complaints

A 75-year-old female presented to our institution with painless right submandibular swelling for 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 one month prior, with intermittent severe pain that radiated to the ipsilateral neck and contralateral face. 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

WBC count levels were elevated (13800/ul).

Imaging examinations

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

FINAL DIAGNOSIS

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

TREATMENT

Excision of the right submandibular gland and sialolithiasis was performed using a standard extraoral approach. The 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. [Fig. 2].

OUTCOME AND FOLLOW-UP

The postoperative course 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 dilatation 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

There are several treatment modalities for submandibular sialolithiasis; however, the primary goal should always be to preserve gland function, maintain low risk, and reduce patient discomfort[4]. Noninvasive 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 and is primarily indicated in cases of intraductal stones that are small. Transoral removal can be considered if the stones can be palpated bimanually and/or localized within the perihilar region of the gland using ultrasound[4,6]. 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[8,9] or a transcervical approach to the submandibular space[10]), extracorporeal shock-wave lithotripsy[11,12,13,14,15], interventional sialendoscopy and intraductal shock-wave lithotripsy[16,17,18,19]. These invasive methods are recommended for stones > 7 mm with symptoms[20]. Treatment selection is mostly based on the ductal system anatomy and location of the stones[7].

Transoral duct surgery may be considered for stones of all sizes at the papilla, and owing to the narrow ostium of the papilla, for stones at the distal and middle ducts. Small mobile stones (not exceeding 3-5 mm) in the papilla and distal and middle ducts can also be extracted using interventional sialendoscopy. The proximal duct, on the other hand, is curved and narrow, so interventional sialendoscopy is hard 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, and interventional sialendoscopy, including mechanical fragmentation or intraductal shock wave lithotripsy, is recommended. Transoral duct surgery with submandibulectomy is indicated for palpable, large stones (> 10 mm) in the 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[6].

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, it is still 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 can avoid facial scars. However, an open surgical approach is associated with complications. For example, postoperative neurological damage, especially hypoglossal, lingual, and marginal mandibular nerve injuries; hemorrhage; hematoma; and disfiguring scars. Temporary marginal mandibular nerve damage is the most common complication after transcervical submandibular gland surgery. This complication was reported to be 36% by Smith *et al*, and permanent damage was as high as 12% by De M *et al* in the literature[21,22]. Beahm *et al* reported rates between 7.7% and 36% in a previous literature review[23]. To reduce the risk of marginal mandibular nerve injury in the transcervical surgical approach to the submandibular gland, three surgical maneuvers are recommended.

The gland is identified as being lower than the hyoid bone, and the marginal mandibular nerve is not sought.

The marginal mandibular nerve is located at the level at which it leaves the parotid tail and tracks and protects it.

The facial veins are located at a lower level and ligated, and the nerve is suspended by elevation; thus, the marginal mandibular nerve is kept out of the surgical area.

However, Smith *et al* reported that when a low 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 gland. The hypoglossal nerve is partially sheltered by the posterior belly of the digastric muscle, which is located in the inferomedial portion of the lower 1/3 of the submandibular glands. This may be the reason why hypoglossal nerve damage has been reported very rarely (0–1.4%)[24] compared to marginal mandibular nerve injuries. In this case report, no permanent or temporary complications occurred during the postoperative follow-up. Even though, 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 the other salivary glands were sufficiently functional to compensate for the resected glands.

Moreover, regarding to the oncological risk to which patients with chronic inflammation and lithiasis are expose[26], we also reviewed some papers about salivary gland tumors. Most salivary gland tumors arise in the parotid gland (70%), followed by minor salivary glands (22%) and submandibular glands (8%)[27]. The differential diagnosis for benign submandibular tumors includes pleomorphic adenoma, Warthin's tumor and oncocytomas. Their histopathological characteristics are the point that can distinguish them. Pleomorphic adenomas are characterized by the thick and irregularly marginated capsules[28,29]. Unlike oncocytomas, which have thin capsules and have monomorphic oncocytes without mitoses and necrosis[29,30]. While lymphatic population is the common cytology and histology of Warthin's tumor[28,29]. Histopathological examination of our patient does not present the above characteristics, and the characteristics of malignant transformation include local invasion into 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 tumor was ruling out.

Due to the similarities in clinical presentation of benign salivary tumors and submandibular sialolithiasis, radiologic imaging is essential in distinguishing between the two entities. Ultrasound is recommended for initial assessment of salivary gland abnormalities, but it is insufficient as the information of surrounding structures cannot be provided. Its accuracy in the identification of salivary benign lesions can be increased by using elastography or contrast-enhanced ultrasound, but the EFSUMB do not recommend contrast-enhanced ultrasound for the characterization of salivary gland lesions in clinical practice. Thus, further studies are needed to investigate the diagnostic role of contrast-enhanced ultrasound and elastography in salivary gland lesion evaluation. Corvino A and colleagues described the utility of computed tomography (CT) and magnetic resonance imaging (MRI) in the initial staging, histologic grading of salivary gland malignancies and preoperative planning. Besides, positron emission tomography with F-18 fluorodeoxyglucose (PET-FDG) is useful in the evaluation and clinical management of head and neck lesions[26].

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

There were a total of 44 cases of giant sialoliths from 1981 to 2022. 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 two 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). Out of 43 cases, 23 and 21 were located in the right and left submandibular gland respectively. Of the giant sialoliths, 90.70% cases were surgically removed (incision or excision), either intraorally or extraorally (transcervical) and 9.30% were removed by other methods. The giant sialoliths reported by Ben *et al* fell out without any intervention[51]. Akinyamoju *et al* and Gustavo *et al* removed the stones nonsurgically [65,54]. In the case of John *et al*,[70] the patient declined any additional diagnostic measures and was lost to follow-up. No postoperative complications were observed in these cases.

CONCLUSION

Giant sialoliths are rare; however, their longer duration is rarely documented. This report presents a case of long-term, rare, and huge sialolithiasis within the submandibular gland parenchyma for 30 years in an older adult. Our patient presented with the typical clinical and radiographic findings. In this case, the infection and fibrosis of the affected gland and the size of the stone left us with no other options except for 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 such 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.

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Jit-Swen Mao and *Yu-Chien Lee* were involved in the conception of the study and contributed to protocol development, data collection, data analysis, interpretation of results, and drafting of the article. *Jit-Swen Mao* and *Yu-Chien Lee* collected the data. *Jessie Chao Yun*, *Chi,Wan-Ling Yi*, *Yung-An Tsou*, *Chia-Der Lin*, *Chih-Jaan Tai*, and *Liang-Chun*

Shih provided critical feedback and direction as well as article revision. *Jit-Swen Mao*, *Yu-Chien Lee,* and *Liang-Chun Shih* contributed to protocol development, interpreted the results, and finalized the manuscript. All authors have approved the final version of the manuscript submitted for publication.

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