



SUBMANDIBULAR SIALOLITHIASIS IN DIFFERENT PRESENTATIONS: A CASE SERIES

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ABSTRACT

Sialolithiasis occurs in approximately 1% of the population. Around 70-90% of salivary stones occur in submandibular salivary gland. Sialolithiasis with sialadenitis is commonly occurring salivary gland disorders. Sialadenitis is an inflammatory condition of the salivary gland. It usually occurs insidiously causing pain and it may also lead to development of fibrous mass within the salivary gland.

Here we discuss 3 cases of submandibular sialolithiasis with different presentations and how each case was managed with different surgical approach.

INTRODUCTION

Sialolithiasis is a common salivary gland disease, it is commonly seen in the Wharton's duct. As the Wharton's duct is located in the floor of the oral cavity so it has high tendency for secondary infection by oral bacteria which affects the gland in a retrograde manner.

Sialolithiasis along with sialadenitis is a commonly occurring salivary gland disorder. Sialolithiasis accounts for around 50% of all disease that occur in the major salivary glands, and for approx. 66% of all obstructive salivary gland diseases. Salivary gland stones are more common in males than in females. The most common age group in which they occur is between 30-60, and they are quite uncommon in children.

Sialadenitis is an inflammatory disease of the salivary gland. It usually occur insidiously causing pain to the patient and may also lead to development of fibrous mass within the salivary gland. The cause of sialadenitis is mostly due to the salivary duct calculi(sialolith or salivary stones) which cause stasis of saliva within the ducts. This condition is usually accompanied by tenderness and pain of the involved site

CASE 1

A 45yr old male presented to ENT OPD with chief complaint of left side neck swelling from last one month, which was gradual in onset and progressively increasing in size, associated with pain and decreased appetite with no history of fever, difficulty swallowing. On examination swelling noted on left side neck in submandibular region ,approx. 3*4cm in size, globular, with no local rise of temperature, tender, firm in consistency, freely mobile, not moving with deglutition. IMAGE 1 shows the preoperative picture of the patient.

Oral cavity examination revealed normal findings.

USG NECK was done and left submandibular gland showed evidence of echogenic foci with posterior acoustic shadowing at the distal end of the submandibular duct measuring approx. 5.0mm. The gland shows enlargement, heterogenous and hypo-echogenesity with dilated intra-glandular ducts and hyper-vascularity on CDFI.

FNAC from swelling of left submandibular gland shows CATEGORY 2-non neoplastic lesion (According to Milan System). Features suggestive of Acute Sialadenitis.

After proper conservative management to reduce inflammation left submandibular gland excision under GA was done and tissue was sent for histopathology. IMAGE 2 shows the intraoperative picture. IMAGE 3 and IMAGE 4 shows the excised specimen and histology finding in the patient.

Features were suggestive of **SIALOLITHIASIS WITH SIALADENITIS**.



IMAGE 1 -PREOPERATIVE PICTURE



IMAGE 2 – INTRAOPERATIVE PICTURE



IMAGE 3 – EXCISED SPECIMEN SHOWING STONE AND SUBMANDIBULAR GLAND.

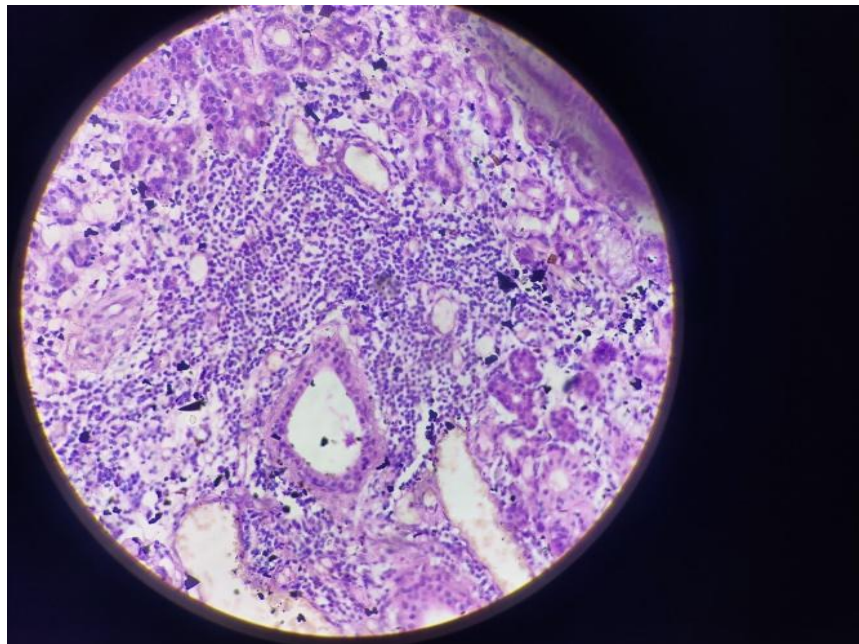


IMAGE 4 - Seromucinous glands and dilated ducts showing dense chronic inflammatory infiltrate.

CASE 2

A 52 yr male patient presented to OPD with chief complaint of swelling on right side of neck, which was progressively increasing in size and associated with pain which increased after intake of food and relieved on taking oral medications for pain.

On palpation swelling was palpable on right side neck, firm, non-tender, mobile, not moving with deglutition.

USG NECK was suggestive of right side submandibular sialolithiasis.

Surgical management was done which involved right submandibular gland excision under GA with removal of stone. IMAGE 5 shows the intraoperative picture.

The specimen was then sent for histopathological examination which confirmed the case to be **SIALOLITHIASIS** right submandibular gland. IMAGE 6 and IMAGE 7 shows the Excised specimen and histopathology findings.

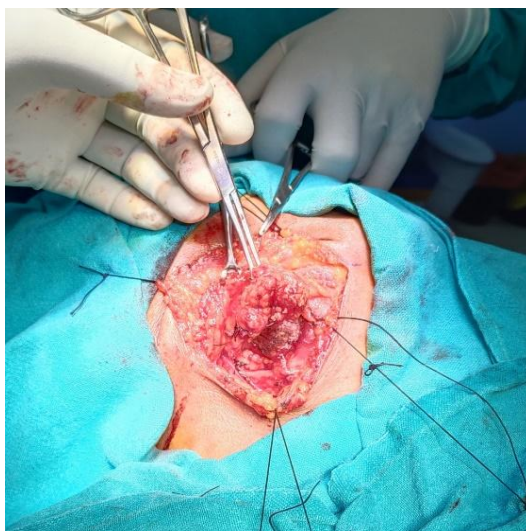


IMAGE 5 - INTRAOPERATIVE PICTURE



IMAGE 6 – EXCISED SPECIMEN SHOWING SUBMANDIBULAR GLAND WITH STONE.

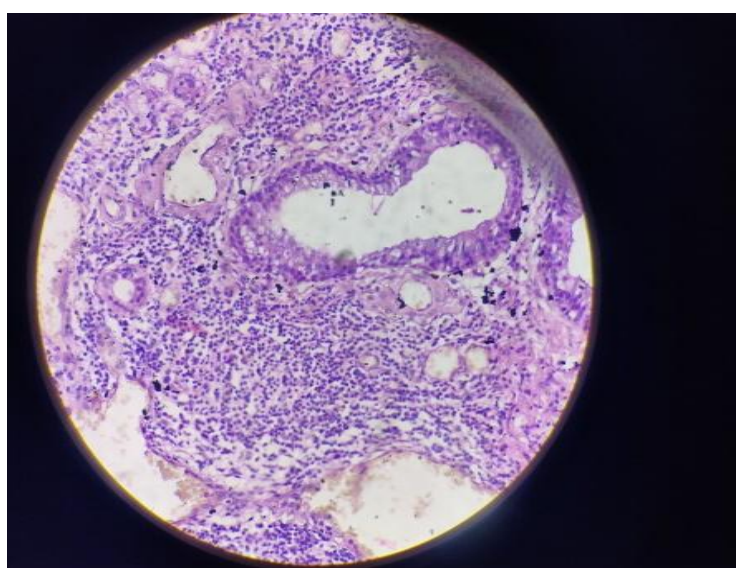


IMAGE 7 - HIGH POWER VIEW SHOWING DUCTAL MUCINOUS METAPLASIA AND SURROUNDING CHRONIC INFLAMMATORY INFILTRATE.

CASE 3

A 45yr old male patient presented to ENT OPD with swelling in floor of mouth from last 10 days which was progressively increasing in size , associated with pain which was localised and increased on chewing food and relieved by medication.

There was no history of fever, discharge from swelling, decreased mouth opening .On examination the swelling was bimanually palpable, tender,firm

USG NECK was suggestive of submandibular sialolithiasis

Management involved surgical removal of stone under GA by giving transoral incision followed by antibiotics and anti -inflammatory medication. IMAGE 8 and IMAGE 9 shows the transoral excision of the stone .



IMAGE 8 - INTRAOPERATIVE PICTURE



IMAGE 9 - TRANSORAL EXCISION (REMOVAL OF STONE)

DISCUSSION

Salivary gland calculi are far more common in submandibular gland than parotid gland because of various factors such as high calcium and mucin content of saliva produced in the submandibular gland, the direction of salivary flow and a tortuous structure of Wharton duct¹. The size of salivary gland calculi may vary from formation of small particles to large concretions. The average size of the salivary calculi is usually between 3 mm-17 mm². They are usually found in the duct of the gland and the hilus, whereas in around 9-17% of cases sialolith occurs in the gland parenchyma³. It is the most common cause of acute and chronic infection in the major salivary glands⁴.

Many theories suggest that formation of calculus occurs in two phases

Stones have a central core and a layered periphery. The central core is formed by precipitation of salts which are binded by certain organic substances⁵. In the second phase there is layered deposition of organic and nonorganic material⁶.

One theory suggests that an unknown metabolic phenomenon can increase the salivary bicarbonate content, which in turn alters the calcium phosphate solubility and leads to precipitation of phosphate and calcium ions⁷.

Sequelae of long-standing calculi include sialadenitis, profound cystic

dilatation, duct dilation and gland atrophy⁸. Other causes of infection in salivary glands are a variety of microbial agents including bacteria, fungi, parasites, mycobacteria, viruses and protozoa⁹. The diagnosis is based on patient's history and clinical examination which are further supplemented by radiologic findings.

Bimanual palpation of the floor of the mouth reveals palpable stone in a large number of cases in submandibular sialolithiasis

A prompt management is very important. The primary objective of the treatment is to restore normal salivary secretion. The treatment of choice may vary according to the location, size and number of stones. When stones are small, moist warm heat application with the administration of sialogogues and gland massage help to flush out the stones from the duct¹⁰. Small sialoliths can also be removed through the duct orifice by bimanual palpation. The treatment options for submandibular sialoliths are intraoral removal of stone, resection of the gland and interventional sialendoscopy.

There are two main surgical approaches for removing salivary gland stones, depending on the gland involved and location of the stone. The transoral approach involves making an incision inside the mouth, directly over the affected salivary duct. Transoral removal of stone should be the treatment of choice in patients having submandibular stones which can be bimanually palpated and localized by ultrasound within the perihilar region of the gland¹¹.

The extraoral approach involves making an incision on the skin below the jawline just below the mandible for submandibular gland stones¹². This approach helps in case of deeper stones or when the transoral approach is not feasible.

CONCLUSION

Sialolithiasis alone or with sialadenitis is a commonly occurring salivary gland disorder. Sialadenitis is an inflammatory disease of the salivary glands. The presence of a salivary gland calculi must be diagnosed beforehand so as to decrease the risk of a subsequent salivary gland infection like sialadenitis which may have detrimental effects.

Surgical management should be considered when the stone is large in size or is inaccessible or conservative therapies turned out to be unsatisfactory. Transoral removal should be the treatment of choice in patients with submandibular stones that can be bimanually palpated and are localized on ultrasound within the perihilar region of the gland.

REFERENCES

1. Lucarelli A, Perandini S, Borsato A, Strazimiri E, Montemezzi S. Iodinated contrast-induced sialadenitis: a review of the literature and sonographic findings in a clinical case. *J Ultrason* 2018;18 (75):359–364
2. Gurwale SG, Gore CR, Gulati I, Dey I. Immunoglobulin G4-related chronic sclerosing sialadenitis: an emerging entity. *J Oral Maxillofac Pathol* 2020;24(Suppl 1):S135–S138
3. Koch M, Zenk J, Iro H. Algorithms for treatment of salivary gland obstructions. *Otolaryngol Clin North Am* 2009;42(06): 1173–1192
4. Capaccio P, Torretta S, Pignataro L. The role of adenectomy for salivary gland obstructions in the era of sialendoscopy and lithotripsy. *Otolaryngol Clin North Am* 2009;42(06):1161–1171
5. Abdel Razek AAK, Mukherji S. Imaging of sialadenitis. *Neuroradiol J* 2017;30(03):205–215
6. Zhang YY, Hong X, Wang Z, et al. Diagnostic utility of submandibular and labial salivary gland biopsy in IgG4-related sialadenitis. *Clin Rheumatol* 2020;39(12):3715–3721
7. Lustmann J, Regev E, Melamed Y. Sialolithiasis. A survey on 245 patients and a review of the literature. *Int J Oral Maxillofac Surg* 1990;19(03):135–138
8. Yiu AJ, Kalejaiye A, Amdur RL, Todd Hesham HN, Bandyopadhyay BC. Association of serum electrolytes and smoking with salivary gland stone formation. *Int J Oral Maxillofac Implants* 2016;45 (06):764–768
9. Gurwale SG, Gore CR, Gulati I, Dey I. Immunoglobulin G4-related chronic sclerosing sialadenitis: An emerging entity. *J Oral Maxillofac Pathol*. 2020 Feb;24(Suppl 1):S135-S138.
10. Steiger R, Storck C. [CME ORL 5. Bilateral pre-auricular swelling. Sialadenitis of all salivary glands in bulimia]. *Praxis (Bern 1994)*. 2012 May 23;101(11):748-9.
11. Mauz PS, Mörike K, Kaiserling E, Brosch S. Valproic acid-associated sialadenosis of the parotid and submandibular glands: diagnostic and therapeutic aspects. *Acta Otolaryngol*. 2005 Apr;125(4):386-91.
12. Urits I, Orhurhu V, Chesteen G, Yazdi C, Viswanath O. Acute Sialadenitis After Intubation. *Turk J Anaesthesiol Reanim*. 2020 Jun;48(3):263.