

CASE REPORT

Salivary Fistula with a Calculus!!

¹Vidya Bhushan Rangappa, ²MS Soumya, ³V Sreenivas

ABSTRACT

Orocutaneous fistula is a pathologic communication between the oral cavity and cutaneous surface. Orocutaneous fistula can occur because of dental infections, salivary gland lesions, neoplasms, or branchial fistula. Overwhelming majority comprises of parotid fistula, submandibular fistula is indeed a rare finding. We report a very unusual case of a large calculus in submandibular gland with a communicating neck fistula.

Keywords: Submandibular gland, Fistula, Neck, Calculus.

How to cite this article: Rangappa VB, Soumya MS, Sreenivas V. Salivary Fistula with a Calculus!! Int J Head Neck Surg 2014;5(2):96-98.

Source of support: Nil

Conflict of interest: None

INTRODUCTION

Orocutaneous fistula is a pathologic communication between the oral cavity and cutaneous surface. Orocutaneous fistula can occur because of dental infections, salivary gland lesions, neoplasms, or branchial fistula.¹ Sialolithiasis can occur at any age but patients in third to sixth decade represents most cases and it is considered rare in children. Literature search reveals scarcity of cases of salivary fistula secondary to submandibular gland calculus.

DISCUSSION

Sialolithiasis accounts for more than 50% of diseases of the large salivary glands and is thus the most common cause of acute and chronic infections.² More than 80% occur in the submandibular gland or its duct, 6% in the parotid gland and 2% in the sublingual gland or minor salivary glands.³ Anatomically, the submandibular duct is longer than the parotid, traversing upward and forward from the gland to the oral floor.⁴ This antigravity direction of flow may also predispose for increases incidence of calculus in submandibular gland. Sialolithiasis can occur at any age but patients in third to sixth decade represents most cases and it is considered rare in children.⁵ We were not able to ascertain the exact incidence

of oro-cutaneous fistula secondary to submandibular gland calculus probably due to its rarity of presentation. On the basis of a review of the literature, most of the sialoliths are usually of 5 mm in maximum diameter and all the stones over 10 mm should be reported as a sialolith of unusual size.⁶ Salivary fistulas are not very common entity in day-to-day practice. Overwhelming majority comprises of parotid fistula, submandibular fistula is indeed a rare finding. Most of the submandibular fistulas are congenital in origin and may be associated with abnormalities of the branchial apparatus.⁷ Salivary fistula can arise either from the gland parenchyma, which is generally more common in parotid gland or from the main salivary duct. Acquired cases can rarely occur and is generally traumatic in origin.⁷

Salivary calculi are usually unilateral in occurrence and round to oblong, have an irregular (majority) or smooth surface, vary in size from a small grain to the size of a peach pit, and are usually yellow. The stones may occur in the duct or gland, with multiple stones not uncommon.⁸

Stones apparently develop as a result of an initial organic nidus followed by the deposition of inorganic material, both of which are derived from the salivary fluid. The filamentous stroma or nidus is not bacterial in nature but rather precipitated mucoids and possibly salivary proteins.⁸

Experimental studies have identified the increased salivary magnesium concentration as a key factor in determining the sialolithiasis.⁹ When the stone reaches a size to obstruct the duct, the secretion in the gland is hampered. This condition facilitates destruction of the gland.¹⁰

Salivary gland calculus obstructing the drainage pathway causes salivary duct swelling, without any obvious reason or at meal times. The symptoms, referred by the patients during the meal times, are due to the higher stimulation of the salivary secretion and to the duct's obstruction that prevents its smooth flow. When affected by a salivary colic, the patient refers an acute pain, sense of swelling and pressure in the floor of the mouth.¹¹

The fistulas arising from the major salivary duct leaks profusely even at the thought of food,⁸ which was happening with our patient.

Ultrasonography represents an excellent first-level diagnostic technique, because it reveals ductal and highly mineralized stones with a diameter of at least 1.5 mm with a accuracy of 99%.¹² Forty percent of parotid and 20% of submandibular stones are not radiopaque and sialograph

¹Postgraduate, ²Assistant Professor, ³Associate Professor

¹⁻³Department of ENT and Head and Neck Surgery, St John's Medical College and Hospital, Bengaluru, Karnataka, India

Corresponding Author: Vidya Bhushan Rangappa, Post-graduate, Department of ENT and Head and Neck Surgery St John's Medical College and Hospital, Bengaluru, Karnataka India, Phone: 22065735, e-mail: drbhushanhns@gmail.com



may be required to locate them.¹² The occlusal radiograph is the most reliable method of viewing the submandibular sialolith. Lateral plate radiographs are effective only with stones in the submandibular gland that project below the inferior border of the mandible.⁸

Sialography is an adequate technique to detect salivary gland calculi that allows the visualization of the whole duct system. However, sialography is not indicated in the case of acute infections or patients sensible to contrast medium.¹¹ Our patient too developed reaction to the test dose of the contrast medium and henceforth this investigation was deferred.

While many stones may remain asymptomatic, recurrent obstruction and sialadenitis are potential hazards of a conservative approach. Migration of a salivary stone from the duct into the tissues is a rare complication of conservative management but should be considered when a decision is made to leave a calculus in place.¹² If the patient is having a symptomatic calculus, the treatment should be removal of the gland or the calculus. There are various modalities currently available like extracorporeal and intracorporeal salivary lithotripsy and radiologically guided stone removal by Dormia basket,¹² simple surgical release through an incision in the floor of the mouth.³ If the calculus size is big and if the patient develops recurrent episodes of sialadenitis, complete excision of the submandibular gland is indicated. Our patient had no history of trauma or previous surgical procedure. Most probably she has developed fistula secondary to the calculus obstructing the salivary flow. The dependant drainage of the saliva is more easier than the antigravity flow through the submandibular gland duct. Our case also highlights the fact that a clinical suspicion of calculus should always be kept in mind, when the patient presents with a discharging fistula in the neck without any swelling in the submandibular gland. The surgeon can be easily misled by diagnosis of branchial fistula, if the submandibular gland is not palpated or proceed with surgery without imaging. This may lead to erroneous diagnosis and improper excision of the tract and high chance of recurrence. Complete excision of the tract in continuity with the gland avoids recurrence.

CASE REPORT

A 55-year-old homemaker, presented to us with history of swelling in the left submandibular region since 6 months. Swelling was insidious onset, increased during chewing food. She noticed watery discharge from the side of the left neck since 2 months. Watery discharge and pain increased at the thought of food and during food intake. She had not undergone any surgical procedure till date. There was no history of trauma. On examination, left submandibular gland

was enlarged, 3 × 3 cm. The gland was nontender. It was firm in consistency and was bimanually palpable. Intraoral examination revealed hard mass palpable in the proximal part of the whartons duct, probably calculus. She had a fistula opening on the left side of the neck along the anterior border of sternocleidomastoid muscle (Fig. 1). The fistulous opening was present in the mid 1/3rd of the sternocleidomastoid muscle. Watery discharge with pus could be expressed on pressure over the submandibular gland. Ultrasound examination of the neck revealed radiopaque calculus in the proximal whartons duct measuring 1 × 1 cm and a tract was noted to extend from this point to the neck. Lateral view X-ray of the neck too revealed radiopaque calculus approximately 1 cm size just inferior to the lower border of angle of mandible (Fig. 2). Sialography could not be done as she was hypersensitive to contrast medium. She was counselled and was treated conservatively for 6 weeks. Due to nonimprovement of symptoms, she was planned for left submandibular gland excision and fistulectomy. Intraoperatively, the gland was enlarged and firm in consistency. The calculus was present at the junction of the submandibular duct and the gland.



Fig. 1: Fistula in left side of the neck



Fig. 2: Calculus inferior to angle of mandible

It was measuring 1 × 1 cm in size (Fig. 3). The tract was noted to extend from the proximal part of the submandibular gland to the skin anterior to the sternocleidomastoid (Fig. 4). An elliptical incision was taken around the fistulous opening and was excised. The specimen was subjected to histopathological examination. It revealed salivary gland



Fig. 3: Calculus

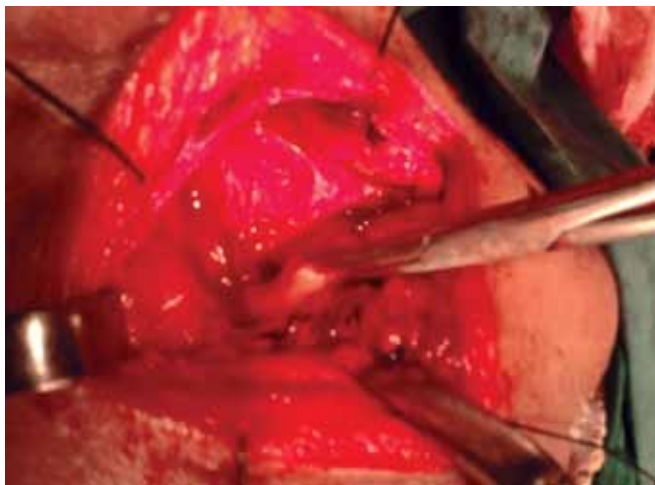


Fig. 4: Fistula tract held using artery forceps

tissue with evidence of atrophy characterized by mildly dilated ducts with marked periductular fibrosis and a mild periductular lymphocytic infiltrate. The fistula tract was lined by granulation tissue with a dense mixed inflammatory infiltrate. Postoperatively patient improved symptomatically. On follow-up for 6 months, she was absolutely asymptomatic.

REFERENCES

1. Saha S, Jha A, Kaur N. Unusual cause of orocutaneous fistula in the neck—case reports in surgery 2012;2012:2.
2. Zenk J, Benzel W, Iro H. New modalities in the management of human sialolithiasis. *Minimally Invasive Therapy* 1994;3(5): 275-284.
3. Sathish R, Chandrashekar L, Ganesan S. Submandibular salivary calculi: a case report. *J Dent Sci Res* 2011;1:16-22.
4. Baumash HD, DDS. Submandibular salivary stones: current management modalities. *J Oral Maxillofac Surg* 2004;62:369-378.
5. Jayachandran S, et al. Giant submandibular sialolith presenting sialocutaneous and sialo-oral fistula: a case report and review of literature. *J Indian Acad Oral Med Radiol* 2011 July-Sep;23(3): 491-494.
6. Batori M, Mariotta G, Chatelou H, Casella G, Casella MC. Diagnostic and surgical management of submandibular gland sialolithiasis: report of a stone of unusual size. *European Review for Medical and Pharmacological Sciences* 2005;9:67-68.
7. Jana AK, et al. Fistula of submandibular gland a rare presentation. *Indian J Otolaryngol Head Neck Surg* 2006 Oct-Dec;58(4): 393-394.
8. Grases F, Santiago C, Simonet BM, Costa-Bauza A. Sialolithiasis: mechanism of calculi formation and etiologic factors. *Clin Chim Acta* August 2003;334(8):131-136.
9. Oteri G, Procopio RM, Cicciù M. Giant salivary gland calculi: report of two cases. *Open Dentis J* 2011;5:90-95.
10. Yoshimura Y, Inoue Y, Odagawa T. Sonographic examination of sialolithiasis. *J Oral Maxillofac Surg* 1989;47:907-912.
11. Cawson RA, Odell EW. *Essentials of oral pathology and oral medicine*. 6th ed. Edinburgh: Churchill Livingstone; 1998 p. 239-240.
12. Drage NA, et al. Migrating salivary stones: report of three cases. *Bri J Oral Maxillofac Surg* 2005;43:180-182.