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Case Report

An interesting case of asymptomatic giant submandibular sialolithiasis: a case report

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ABSTRACT

Sialolithiasis is the most common disease of salivary glands. On the other hand, giant submandibular sialoliths or megaliths are quite rare. Our case report is of an unusually large submandibular sialolith called a giant sialolith as dimensions exceed 15 mm in size. Patient was asymptomatic on presentation and had preserved gland function. A firm mass was detected on the left floor of mouth and on transoral sialolithotomy a single large calculus of 40 mm, in greatest dimension, was removed in toto. Though submandibular gland sialoliths are generally asymptomatic in nature, giant calculi usually present with symptoms including pain and swelling of the involved gland caused by the accumulation of saliva, due to blockage of the lumen of Wharton's duct by the calculus. This is a rare case report of an asymptomatic giant submandibular sialoithiasis, its diagnosis and management.

Keywords: Sialolithiasis, Submandibular sialolith, Giant sialolith, Giant submandibular sialolith, Megalith

INTRODUCTION

One of the most common disorders of the salivary glands is sialolithiasis.[1] More than 80% occur in the submandibular gland or its duct, probably because of its more viscous saliva, longer duct, and higher mineral content in the saliva. Only 4-10% have been reported in the parotid gland and 1-7% in the sublingual gland or minor salivary glands.[2] Sialolithiasis is defined as formation of stones in the salivary glands or their duct and the term sialolith indicates a calcified mass within the gland or its duct. Sialolithiasis is usually seen between the age of 30 and 60 years.[1] Twelve per 1000 adult population are reported to suffer from the condition each year, with males affected more than females.[3] Though sialoliths are one if the most common non neoplastic major salivary gland pathology, giant sialoliths or Megaliths are not common and are defined as formation of a salivary gland stone larger than 15 mm in its greatest dimension.[4] Megaliths are harbored in submandibular gland due to anatomical and physiological characters that favors the formation of these stones. The rarity of the condition, its diverse clinical presentation makes it an important topic for reporting and discussing.

CASE REPORT

A 68-year-old male patient, diabetic, was referred to the outpatient department of our hospital for opinion and management of a firm mass on the left side of floor of mouth. A high-resolution ultrasound neck showed features of chronic left submandibular sialadenitis with significantly dilated intraglandular ductules and submandibular duct with impacted large calculi proximal to the opening of the duct, near to floor of mouth. There was significant edema of floor of mouth and pus extruding from submandibular duct. Patient did not have any signs of systemic infection. Under antibiotic coverage left submandibular sialolith excision was done. A single large calculus with dimension of 4 cm×2 cm was removed in toto. Good salivary flow was noted immediately after sialolith was removed. Sialoflo stent

was placed in the duct and secured in place with sutures. Patient was stable post procedure and was discharged on oral antibiotics.

Figure 1: Submandibular sialolith showing larger dimension of 40 mm.

Figure 2: Submandibular sialolith showing smaller dimension of 20 mm.

DISCUSSION

Most of the sialolith are usually of 5 mm in maximum diameter and all the stones over 10 mm should be reported as a sialolith of unusual size. Furthermore, they are classified as giant in case any dimension exceeds 15 mm. One of the largest sialoliths of 72 mm size was reported by Rai et al.[5]

Aetiology

Though definite aetiology is still ambiguous, sialoliths are thought to occur as a result of deposition of mineral salts around an initial nidus consisting of salivary mucin, bacteria or desquamated epithelial cells. They form as a result of mineralization of debris that has accumulated in the lumen of the duct. This debris includes bacterial colonies, exfoliated ductal epithelial cells, mucus plugs, foreign bodies or other cellular debris. Factors like stagnation of salivary flow, dehydration, and change in salivary pH associated with oropharyngeal sepsis, impaired crystalloid solubility, high alkalinity, and increased calcium content, and physical trauma to salivary duct or gland may predispose to calculus formation.[1]

Two stages of sialolith formation can be found in the literature, central core formation and layered periphery formation.[6] In the first phase, mineral salts bound by certain organic substances precipitate to form the central core. Then, in the second phase, some organic and inorganic materials deposit around the central core in layers. Parotid and submandibular stones are thought to frequently form around a nidus of inflammatory cells or foreign body and a nidus of mucous respectively. Boynton et al reported an unusual case in which a facial hair of the patient got entrapped in the Wharton's duct and acted as a nidus for the formation of a sialolith.[7] A retrograde theory suggested that any substance or bacteria of the oral cavity, that had migrated into the salivary ducts, can act as a nidus for further calcification.[6]

Diagnosis

Careful history and examination are important in the diagnosis of sialolithiasis. Pain and swelling of the concerned gland at mealtimes and in response to other salivary stimuli are especially important. Complete obstruction causes constant pain and swelling, pus may be seen draining from the duct and signs of systemic infection may be present.[8] Bimanual palpation of the floor of the mouth, in a posterior to anterior direction, reveals a palpable stone in many cases of submandibular calculi formation. Bimanual palpation of the gland itself can be useful, as a uniformly firm and hard gland suggests a hypo-functional or nonfunctional gland.[9]

Imaging studies are very useful for diagnosing sialolithiasis. Occlusal radiographs are useful in showing radiopaque stones. Sialography is thus useful in patients showing signs of sialadenitis related to radiolucent stones or deep submandibular/parotid stones. Sialography is, however, contraindicated in acute infection or in significant patient contrast allergy.[9]

Treatment

Patients presenting with sialolithiasis may benefit from a trial of conservative management, especially if the stone is small.[9] With gland swelling and sialolithiasis, infection should be assumed and a penicillinase resistant anti - staphylococcal antibiotic prescribed. Most stones will respond to such a regimen, combined with simple sialolithotomy when required.[10] Almost half of the submandibular calculi lie in the distal third of the duct and are amenable to simple surgical release through an incision in the floor of the mouth, which is relatively simple to perform and not usually associated with complications.[11] The duct may need opening to retrieve

the stone. This involves a transoral approach where an incision is made directly onto the stone. In this way more posterior stones, 1-2 cm from the punctum, can be removed by cutting directly onto the stone in the longitudinal axis of the duct. Care is taken as the lingual nerve lies deep, but in close association with the submandibular duct posteriorly. Subsequently, the stone can be grasped and removed. No closure is done leaving the duct open for drainage.[12]

Alternative methods of treatment have emerged such as the use of extracorporeal shock wave lithotripsy (ESWL) and more recently the use of endoscopic intracorporeal shockwave lithotripsy (EISWL), in which shockwaves are delivered directly to the surface of the stone lodged within the duct without damaging adjacent tissue (piezoelectric principle).[13] Findings have also suggested that best results in salivary stone lithotripsy are achieved when the maximum size of stone fragments does not exceed 1.2 mm.[14] Submandibular gland excision is recommended in cases of substantial intra-glandular caliculi, which are inaccessible via a trans-oral approach. Also, when multiple small stones are present in the vertical and comma portions of Wharton's duct, sialoadenectomy is recommended.[4]

CONCLUSION

Giant salivary calculi are rare, yet most cases present with the classical picture of salivary colic. In our case, patient was asymptomatic and it was found that his gland function was preserved. Intraoral removal of calculi with sialoflo stent placement ensured that post op neo-opening was formed and the salivary flow was not impeded. Intra oral removal of calculi and complete excision ensured preservation of gland function and successful management of giant sialolith without the need of submandibular gland excision.

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REFERENCES

1. Pachisia S, Mandal G, Sahu S, Ghosh S. Submandibular sialolithiasis: A series of three case reports with review of literature. Clin Pract. 2019;9(1):1119.
2. Goaz PW, White SC. Oral Radiology, principles and interpretatio. Soft tissue calcifications. St. Louis: Mosby. 1994: 62-66.
3. Leung AK, Choi MC, Wagner GA. Multiple sialoliths and a sialolith of unusual size in the submandibular duct: a case report. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 1999;87:331.
4. Wallace E, Tauzin M, Hagan I. Management of giant sialoliths: Review of the literature and preliminary experience with interventional sialendoscopy. Laryngoscope. 2010;120:1974-8.
5. Rai M, Burman R. 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.
6. Marchal F, Kurt AM, Dulguerov P, Lehmann W. Retrograde theory in sialolithiasis formation. Arch Otolaryngol Head Neck Surg. 2001;127:66-8.
7. Boynton TT, Lieblich SE. Unusual case of a sialolith: a case report. Oral Surg Oral Med Oral Pathol Oral Radiol 2014;117:9-10.
8. Pollack CV, Severance HW. Sialolithiasis: case studies and review. J Emerg Med. 1990::561-5.
9. Williams MF. Sialolithiasis. Otolaryngologic Clinics of North America. 1999;32:819-34.
10. Pietz DM, Bach DE. Submandibular sialolithiasis. General Dentistry. 1987;35:494-6.
11. McGurk M, Esudier M. Removing salivary stones. Br J Hosp Med. 1995;54:184-5.
12. Siddiqui SJ. Sialolithiasis: an unusually large Submandibular Stone. 2002;193:89-91.
13. Iro H, Schneider H Th, Fodra C. Shockwave lithotripsy of salivary duct stones. Lancet 1992;339:1333-6.
14. Zenk J, Werner G, Hosemann MD, Iro H. Diameters of the main excretory ducts of the adult human submandibular and parotid gland – a histological study. Oral Surg Oral Med, Oral Pathol Oral Radiol, Endod. 1998;85:576-80.

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