



Case Report

JMR 2015; 1(1): 05-07
January- February
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A huge salivary calculi of the submandibular gland: a case report with the review of literature

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Abstract

Sialolithiasis is the most common salivary gland disease. Submandibular sialolithiasis is the most common site. Sialoliths greater than 15 mm in any one dimension is considered as the larger one. Salivary stones with size greater than 30 mm are rare. We report a case of a huge sialolith (approx. 38 mm*30 mm) of the submandibular gland along with the review of literature. Sialendoscopy is newer diagnostic as well as therapeutic modality recently gaining fame. The aim of presenting this case report is to understand etio-pathogenesis, clinical presentation and management of submandibular sialolithiasis.

Keywords: Huge submandibular gland stones, Submandibular sialolithiasis, Sialolithiasis.

Introduction

Sialolithiasis is the most common salivary gland disease. Sialolithiasis affects the Submandibular gland (83%) followed by the Parotid (10%), Sublingual and Minor salivary glands (7%).^[1] Sialolithiasis occurs in 12 per 1000 adult population each year. Sialolithiasis affect males twice as common than females.^[1-3] Sialolithiasis affects both sides equally. Bilateral stones are less common.^[2] 88% of salivary calculi are smaller than 10 mm in size.^[4] Sialoliths with size greater than 15 mm in any one dimension are considered as the giant one, occurrence of which is rare.^[5] Person of any age may be affected by the salivary gland stones. But it is mainly seen in third and fourth decades of life.^[3] Children are rarely affected.^{[4],[6]} Sialolithiasis patient presents with pain and swelling due to obstruction of the salivary ducts classically at the time of meal.^[1] So, it is called "Mealtime syndrome" also.

Case Report

Permission from the institute ethical committee has been taken. A 46-year-old male patient presented in the E.N.T. outpatient department, P. D. U. Medical College, Rajkot, with the history of the right submandibular swelling for 7 years associated with pain in the right floor of mouth and the submandibular area for 10 days. The swelling was small in size initially then increased progressively up to present level of about Orange. The size of the swelling increased after the meal. He has a history of tobacco chewing for last 15 years with poor oral hygiene. The medical history of the patient was not significant. On examination, there was a mass in the right submandibular region measuring 10 cm x 8 cm, tender with increased temperature without attachment to skin or underlying structures. Bimanual palpation revealed a firm mass of 10 cm x 8 cm with hardness within.

Ultrasound of the neck showed approximately 4 cm*3 cm sized hyper echoic area within the enlarged submandibular gland. Patient was put on antibiotics and anti-inflammatory drugs and planned for surgery five days after completion of therapy. We had excised the submandibular gland with stone by making an incision along Resting Skin Tension Line 2 cm below the mandibular border. Stone was 38 mm*30 mm, light yellowish in color (Figure 1 and 2). The post operative period remained uneventful.

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Figure 1: Intra-operative image of submandibular gland with calculi within the gland

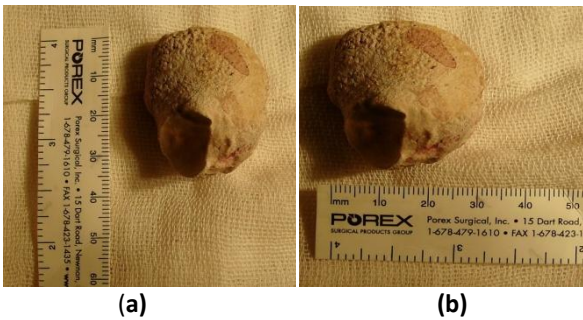


Figure 2: Measurements of huge stone removed from the submandibular gland

Discussion

Exact etiology of formation of the sialoliths is not known, although it is suggested due to defect of migration of autophagosomes through the ductal system or the calcification of mucus plug.^{[1],[3]} They will act as nidus around which layered deposition of both organic and inorganic material in varying proportion occurs. Organic material includes glycoproteins, mucopolysaccharides, bacteria, and desquamated cells while inorganic material includes mainly calcium phosphate and calcium carbonate. Ions like iron, copper, manganese etc are found in lesser amount.^{[2],[6-8]} Presence of streptococcal species is found in almost all calculi in one study.^[9] Sialolithiasis may occur due to altered calcium metabolism, altered pH, reduced water intake, infection, inflammation, tobacco chewing, drugs like antihistamines, antipsychotics etc. Sialolithiasis may be associated with other systemic conditions like gout, nephrolithiasis, chronic liver disease etc. The stones in gout are composed of uric acid crystals.^{[1],[6-8],[10]} Generalized calculi formation involving urinary system, biliary tract and salivary glands were found in 6-10 % of cases.^[3]

Majority stones are less than 10 mm in size. Stones with size greater than 30 mm are rare.^{[4],[11]} A review of literature in 2007 found only 16 reported cases of sialoliths having a size up to 3.5 cm.^[12] Several factors like longer and tortuous course of the submandibular duct, antigravity drainage, high calcium and mucin content, more viscid secretions, alkalinity predispose the submandibular gland to stone disease.^{[6],[7],[13],[16]}

Proper history taking and examination will tell the diagnosis. 80-90% of Submandibular stones are radio-opaque due to their high content of calcium and magnesium carbonates & phosphates leading to their easy diagnosis on x-rays. Other diagnostic methods include Sialography, USG, CT Scan and Sialendoscopy.^[4] Sialography demonstrates anatomy of the ductal system with the presence of stone if any and degree of

glandular damage by chronic disease. However, sialography carries risk of radiation and chances of ductal perforation and retrograde displacement of the stone with injection.^[11] USG is non-invasive method of detecting sialoliths. Stones > 15 mm size particularly with high mineral content are easily detected. USG may detect radio-lucent stones.^[14] CT scan will tell about the exact location of stone of any size. But, it is an expensive tool.^[2] Sialendoscopy is both diagnostic and therapeutic modality. It detects stone even when x-ray and USG fail.^[15]

If stone is small, treatment remains conservative. Maintaining hydration, prescribing sialogogues and massage may be helpful. Antibiotics are added whenever necessary.^[10]

Submandibular stones can be removed surgically through either intraoral or an external approach. For stones located entirely in the duct and close to the papillae, intraoral approach is sufficient. Incision is made longitudinally over the Wharton's duct taking care of lingual nerve which is closely related to it. Stone is then removed and duct left open (Sialodochotomy) or sutured with the surrounding mucosa (Sialodochoplasty). Extra oral approach is indicated for intra glandular stones and stones embedded in the hilum of gland as performed in our case. But, an external approach risks marginal mandibular nerve.^{[7],[16]} Larger stones may be fragmented by using extra shock wave lithotripsy or by endoscopic intracorporeal shock wave lithotripsy or LASER. The later technique has been developed recently.^[10] Extra corporal shock wave lithotripsy breaks stone into fragments of 0.7 mm on an average. The best results in salivary stone lithotripsy are achieved if size of the stone fragments is below 1.2 mm.^[17] Sialendoscopy is performed under local anesthesia. Appropriate size sialendoscope is introduced after dilatation of salivary papilla. Small stones can be retrieved with a wire basket. Larger stones are fragmented with fiber optic laser lithotripsy. Intervention can be performed under direct vision by this technique. Sialendoscopy carries risk of ductal perforation which can be minimized by adequate training.^{[8],[15]}

Conclusion

Occurrence of giant sialolithiasis is rare. More research is required to know the etio-pathogenesis of salivary stone formation. USG is a noninvasive and highly effective investigation. There are various treatment options available depending up on the size and location of sialoliths. The submandibular gland needs to be excised in cases of large stones and stones embedded within the gland. Sialendoscopy is newer diagnostic and therapeutic modality.

Conflict of Interest: No conflict of interest noted. No financial implication.

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