



RESEARCH ARTICLE

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CAN MEGALITH OF WHARTON'S DUCT BE TREATED IN OUTPATIENT DEPARTMENT? A RARE CASE

*¹Dr. Nilam U. Sathe, ²Dr. Avinash Malekar, ²Dr. Anup Shrinivas and ²Dr. Muniram Pawara

¹Associate Professor, Dept of ENT and Head – Neck Surgery, Seth G. S. Medical College and KEM Hospital, Parel, Mumbai - 400 012

²Senior Resident, Dept of ENT and Head – Neck Surgery, Seth G. S. Medical College and KEM Hospital, Parel, Mumbai - 400 012

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*Corresponding author: Dr. Nilam U. Sathe,

ABSTRACT

Introduction: Sialolithiasis is a common salivary gland disease most commonly occurring in submandibular gland. Calculi more than 15 mm in size are considered giant or megalith. This case report describes a megalith of Wharton's duct in 70 years old male, its pathophysiology and subsequent patient management. To our knowledge there are very few cases of megalith of such size has been described and sialolith presented in our study is perhaps the largest ever reported to date so far in the literature. **Case Report:** 70 years old male patient presented to our ENT outpatient department with complaints of intermittent pain and swelling in right side of floor of mouth since last 6 months. On Intra oral examination, a yellowish, hard mass seen protruding out from Wharton's duct ostium on right side in floor of mouth. A provisional diagnosis of right submandibular sialolith was made. Sialolith was removed in toto in OPD using Tilley's forceps and gentle manipulation under local anesthesia. It was measured to be 8x2.5cm which was unusually giant sialolith with smooth surface present in submandibular gland duct. **Conclusion:** Megalith or giant sialolith are encountered rarely and may be asymptomatic for long time. The diameter value of Wharton duct ranges between 0.2 mm and 2.2 mm. Also this case report highlights the ability of Wharton's duct to dilate up to large extent to accommodate such giant sialolith up to 8cm. In our patient the sialolith was really huge probably the largest one till reported in literature. Such huge sialolith can be removed with simple manoeuvring and instruments in out patient department under local anesthesia which can avoid major surgical procedure in operation theatre and it is a day care procedure. The Wharton duct has a natural capacity to regain its normal size after removal of such megalolith.

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INTRODUCTION

Submandibular gland, owing to its position, duct course and characteristic of salivary content is common site for sialolithiasis. Commonly sialoliths measure from 5mm-10mm. Sialoliths more than 10mm has been termed unusual in size. Stones larger than 15 mm in length are called "giant stones" or "megaliths" and are relatively rare in occurrence (Wallace, 2010). Several authors have reported that sialolithiasis occurs in 80% to 95% in the submandibular gland, 5% to 20% in the parotid gland, and 1% to 2% in the sublingual and minor salivary glands (Bodner, 2002) Submandibular sialolith may be asymptomatic for long period time and patient may present

with pain and swelling due to obstruction of duct classically during meal. Diagnosis can be made by history, physical examination and using Ultrasonography, Occlusal radiographs, Sialography or CT scan. The location and size of sialoliths are important factors in deciding which surgical procedure will be performed, especially in giant sialoliths (Wallace, 2010). Treatment methods include intraoral removal of sialolith, sialodochotomy, shock wave lithotripsy, sialoendoscopic removal, intraductal laser fragmentation and submandibular gland excision.

CASE REPORT

70 years old male patient presented to ENT outpatient department of KEM hospital, Mumbai, with complaints of

intermittent pain and swelling in right side of floor of mouth since last 6 months. Initially patient ignored the problem and does not seek any medical attention. Later in course part of swelling was extruded spontaneously and later patient presented to hospital with yellowish hard mass in right side floor of mouth.



Figure 1. Showing Part of megalith visible out of duct in floor of mouth on right side

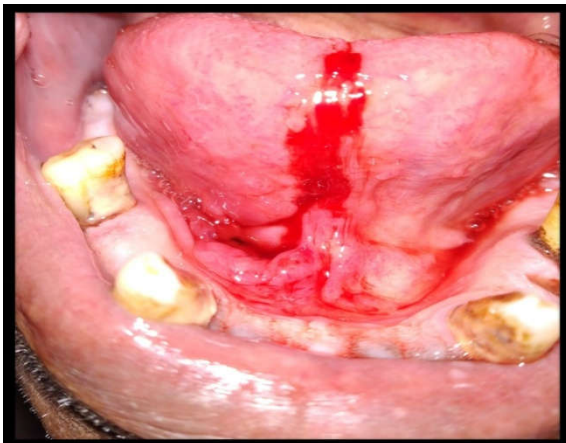


Figure 2. Showing Dilated submandibular duct and its ostium after sialolith removal



Figure 3. Showing Removed megalith from Wharton's duct measuring approx. 8x2.5cm

On Intraoral examination, a yellowish, hard mass seen protruding out from Wharton's duct ostium on right side in floor of mouth (Figure 1). Overlying mucosa was normal without any signs of inflammation. Bimanual palpation revealed the presence of a palpable hard mass along Wharton's duct. A provisional diagnosis of right submandibular sialolith was made. Sialolith was removed in toto in OPD using Tilley's forceps and gentle manipulation under local anaesthesia. It was measured to be 8x2.5cm (Figure 3). Patient thereafter treated with antibiotics, analgesics and antiseptic mouthwash for 7 days. Post procedure USG neck does not show any residual calculi and bilateral submandibular glands were normal. Postoperative healing was uneventful (Figure 4).



Figure 4. Showing Operative site after 1 month follow up

DISCUSSION

Sialolithiasis is a common disease of the salivary glands and a major cause of salivary gland dysfunction. There are three requirements for sialolith formation: a nest, saliva stagnation, and minerals precipitation. Physical trauma, infection, and inflammation of the gland predispose the appearance of the sialolith by promoting the precipitation of salts (Bodner, 1995). Sialolith primarily consists of calcium phosphate, carbonate hydroxyapatite in combination with an organic matrix of glycoproteins, and mucopolysaccharides. Salivary gland stones occur most commonly between the fourth to sixth decades of life. Males tend to develop calculi more frequently than females. Stones larger than 15 mm in length are called "giant stones" or "megaliths" and are relatively rare in occurrence (Wallace, 2010). Stones have been shown to grow at a rate of 1 mm to 1.5 mm per year. Consequently, it could be hypothesized that it could take at least 10 years to obtain a stone classified as a megalith (Wallace, 2010). The ability of a calculus to grow and become a giant sialolith depends mainly on the reaction of the affected duct. If the duct adjacent to the sialolith is able to dilate, allowing nearly normal secretion of saliva around the stone, it might be asymptomatic for a long period and eventually a giant calculus will be created (Paul, 1995 and Rai, 2009). Submandibular sialolithiasis is more common as its saliva is (i) More alkaline, (ii) has an increased concentration of calcium and phosphate, and (iii) has a higher mucous content than saliva of the parotid and sublingual glands. In addition, the submandibular duct is longer and the gland has an antigravity flow (Ledesma-Montes, 2007). He predisposition to calculi, and ability to tolerate expansion, lead

to a higher incidence of giant calculi associated with this gland (Rai, 2009). A few case reports on submandibular calculi have resulted in perforation of the Floor of the mouth or cutaneous fistula (Karengera, 1998 and Sutay, 2003). To our knowledge, the sialolith presented and removed in our study is perhaps the largest ever reported to date, as compared with published data.

The diameter value of Wharton duct ranges between 0.2 mm and 2.2 mm. Narrowest duct diameter observed at Wharton's duct ostium (Zenk, 1998). Our study also highlights the capacity of Wharton's duct to dilate almost 10 times to its mean diameter as size of removed megalith in our case was 8x2.5cm. Patient may be asymptomatic for longer period due to this ability of duct to dilate, allowing normal salivary flow around the stone and eventually megalith will be formed. Megalith can completely obstruct the salivary flow and can lead to inflammation of gland. It can also cause fibrosis and atrophy of the affected gland. To prevent these complications calculi should be completely removed as early as possible. Treatment objective for sialolith is to remove the stone and restore normal salivary flow. Numerous treatment methods has been described depending upon the size & position of stones and ranges from application of moist warm heat with gland massage, use of sialogogue, and transoral removal to complete gland removal (sialoadenectomy) (Rai, 2009). Sialodochotomy is a well-reported technique for the intra-oral removal of ductal stones, including giant calculi (Fowell, 2012). Sialendoscopy is now an established intervention for stone removal, and has been described for use in giant salivary calculi (Wallac, 2010). Large intraglandular calculi inaccessible via intraoral approach are treated with submandibular gland excision. Other treatment methods includes extracorporeal shock wave lithotripsy, endoscopic intracorporeal shock wave lithotripsy (ISWL), intraductal laser fragmentation and combined approach technique. In our patient the sialolith was really huge probably the largest one till reported in literature. Usually surgical procedures are done to remove the sialolith in operation theatre. We were successful in removing such a huge salivary duct calculi in Out patient department with surface anaesthesia (10% lignocaine spray) in the floor of mouth and with the basic instrument Tilley's nasal forceps and massage on the calculi with the finer tips in submandibular area without any complications. The patient recovered with regaining near normal size of submandibular duct after the removal of stone in one month postoperatively.

Summary

- Megalith or giant sialolith are encountered rarely and may be asymptomatic for long time.

- Early diagnosis and treatment in cases of megalith is necessary to restore normal salivary flow and to prevent long term salivary obstruction which may result in fibrosis and atrophy of the affected gland.
- The diameter value of Wharton's duct ranges between 0.2 mm and 2.2 mm. Also this case report highlights the ability of Wharton's duct to dilate up to large extent to accommodate such giant sialolith up to 8cm.
- Such huge sialolith can be removed with simple maneuvering and instruments in outpatient department under local anesthesia which can avoid major surgical procedure in operation theatre and it is a day care procedure.
- The Wharton duct has a natural capacity to regain its normal size after removal of such megalolith.

Conflict of interest: There is no conflict of interest.

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