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TRIPLE TROUBLE – AN UNCOMMON CASE OF MULTIPLE SIALOLITHS OF DIFFERENT SIZED (LOCATED AT DIFFERENT PLACES) IN THE DUCT OF RT SUBMANDIBULAR SALIVARY GLAND, RETRIEVED SURGICALLY IN AN EMERGENCY ATTEMPT UNDER LOCAL ANESTHESIA.

Dental Science

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ABSTRACT

Saliva is an important viscus or serous secretory fluid of major and minor salivary glands of oral and perioral region, plays very important role in digestion of foods, lubrication of oral mucosa and plays a key role in making the teeth caries resistant. There are many disease or conditions, where the salivary secretions not only reduced but also gland functions are grossly jeopardized. The most common disease after mumps, in which the gland or glands function get defunct, is sialolith, which may be single/multiple in numbers or small to giant in size. Here we are presenting an uncommon case of multiple sialoliths which were removed surgically from Right Wharton's duct by trans-oral sialolithotomy, under local anesthesia, in an emergency surgical intervention, not only to give the patient much needed relief from excruciating pain, but also to save the entire gland & duct from surgical removal under GA.

KEYWORDS

Sialolith, Salivary gland, Wharton's duct.

INTRODUCTION

Saliva is a very important viscous/serous secretory fluid of three pairs of major and en-numbers of minor salivary glands, as situated peri/intra orally, helps in lubrication of mouth, swallowing of foods, making the teeth resistant to caries by flushing action and aid in digestion.

The major secretory glands are the parotid, sub-mandibular, and sublingual glands. The minor salivary glands are those smaller glands and groups of glands in the palate, buccal/labial mucosa, and floor of the mouth that secrete primarily mucus.

There are many diseases, which may involve the salivary gland, of which salivary sialolithiasis is probably one of the most common diseases of the major glands after mumps (2).The majority of the sialolith occurs in the submandibular gland or it's duct, which probably may be, as a consequence of mucinous saliva, tortuous and near 90 degree bend of duct, gravity, and the small orifice of the duct.(6) Sialolithias usually occur in third to fifth decade of life, and slightly more in males (4:3) (3,5) than in female counterparts, and is rare in children. The right and left glands and ducts are equally affected, although bilateral occurrence is rare as well as the, multiple sialoliths in the same duct or gland are also not very common.

Here we are going to report a very interesting case of multiple sialoliths, removed from right submandibular gland duct of an elderly female under local anesthesia, by trans-oal sialolythotomy .Three stones were of different sizes, measuring from 18mm to 4.8mm and located at different places of the duct (two from canine premolar area and the third one medial to ramus, posterior to lower second molar) were surgically retrieved in an emergency attempt to give the patient relief from excruciating pain, as she presented to our OPD after routine OPD hour.

CASE REPORT

A 61 year old female patient presented with the chief complaint of moderate swelling under surface of the tongue (right side) along with excruciating pain and pus discharge for past three days. She had given the history of some treatment/self-medication, which did not give relief her from pain and swelling. She also complaints of few episodes of occasional swelling and pain while taking the foods for last nine months.

Extra-oral examination revealed, tender and diffuse swelling in right submandibular region without any notable erythema, raise in local temperature, regional lymphadenopathy and trismus. Patient's vital signs were recorded and found within normal limit except mild tachycardia and fall in systemic BP. Anemia, jaundice, cyanosis, clubbing were also ruled out subsequently. Intra-oral examination revealed a diffuse swelling involving right canine–premolar area over the floor of the mouth. On bi-digital palpation, there was tendered, firm swelling involving right floor of the mouth (due to obstruction of the duct, causing stagnation of saliva) and purulent discharge from the opening of the orifice of Wharton's duct (indicating secondary infection subsequent to salivary stagnation),both of these contributed to severe pain which the patient had.

Standard occlusal X-Ray and NCCT of mandible revealed presence of elongated, differently sized radio-opacity in canaine-premolar area and medial to ramous of mandible posterior to right lower third molar. On the basis of detailed history, clinical & radiological findings, the case was diagnosed as chronic suppurative submandibular sialadenitis, secondary to sialodocholithiasis in the right Wharton's duct. Surgical removal of sialolith was planned under local anesthesia on an emergency basis. Patient was advised for routine blood and urine examination along with immune-surveillance for HIV & HBV/HCV. The patient was admitted to surgical ward for the preparation of trans-oral sialolithotomy, after ascertaining the laboratory findings and negative result of immune-surveillance.

The patient was made to seat on the dental chair with 45 degree back angulation and anterior stones were first located accurately by radiographs and palpation after achieving the bilateral pterigomandibular block. A suture was passed through the floor of the mouth, below the duct and behind the stones (anterior), and was tied to prevent the stones from sliding (1) backward. A towel clamp was placed through the tip and, and the side of the tongue to obtain retraction and control of the assistant. The gland was then palpated extra-orally and pushed upward, toward the floor of the mouth to fix the intraoral tissues under tension and make the stones easier to locate & palpate. When the incision was placed, consideration was given to two vital structures, the lingual nerve and the sublingual gland. Thus an incision of 2cm was designed for the anterior stones and medial to the plica. Blunt dissection was carried out in the direction of the sialolith. A small incision was placed over the duct overlying the first sialolith. Blunt dissection was continued through the loose tissues of the space until the duct was encountered. When the duct was isolated, a longitudinal slit was made directly over the stone. The stones were

carefully removed with a small artery forcep. After the anterior stones were removed, a small aspirating cannula was passed to remove any pus, mucous plugs, and satellite stone that remained posteriorly. A probe was then passed from the caruncle to the surgical opening to ensure patency of the anterior end of the duct. No effort was made to close the duct properly. The wound edges were sutured at the level of the mucosa only after retrieval of three stones.

The patient was prescribed with Inj. Cefotaxime 1gm, 12hrly, Inj Metronidazole 500 mg, 8hrly, Inj Amikacin 500mg, 12hrly, Inj Ranitidine 12hrly and Inj Diclofenac sodium 75mg (IM) SOS. Postoperatively patient was reviewed after 24hr, 48hr and seven days consecutively and was discharged from the hospital after removal of the suture on seventh post-operative day.

DISCUSSION

Exact cause of formation of sialolith (6) is not very clear, but there are some theories that support the process of formation of stones in the glands like, may be related to dehydration, which increases the viscosity of the saliva; reduced food intake, which decreases the demand for saliva; or certain medications that lower the production of saliva, such as anti-histaminics, anti-hypertensives and antipsychotics, and antidepressants. The lack of saliva can also be results of several circumstances, which includes aging, Sjogren's syndrome, and radiotherapy of the head and neck region etc. Some factors like alkaline PH, increased calcium concentration by submandibular gland, and the tortuous course of the duct, narrow and opening of duct at an acute angulation against the gravity, contributes as predisposing factors for submandibular gland. A study conducted by Sheman and McGurk (07) indicated that there is no link between water hardness and sialolithiasis or sialadenitis, also suggested that increased calcium intake might not lead to sialolithiasis.

Chemically, sialoliths are composed (8) of organic substances like glycoproteins, mucopolysaccharides, and cellular debris and inorganic substances mainly calcium carbonates and phosphates invariable ratios. Stones in the salivary ducts or glands are formed by deposition of calcium salt around a central nidus, which may consist of aggregation of desquamated epithelial cells, bacteria, foreign bodies or products of bacterial remnants. Once the stone achieved the size bigger than the duct, causes obstruction, which prevents the free flow of saliva, and results in stagnation or accumulation of saliva under pressure, clinically represents by pain and swelling. Careful bidigital palpation of the floor of the mouth, along Wharton's duct on a posterior to anterior direction may reveal the presence of a sialolith.

The incidence of sialolith in Wharton's duct is not uncommon but what is not common that is presence of multiple numbers of stone, specially one is very large(>15mm). Several times it has been observed that small sized stones in duct comes out with a normal salivary flow with no or little inflammatory features. But incase of the sialoliths, which are much bigger than the size of the duct, then it will cause mechanical obstruction and thereby produces various signs and symptoms, which demands surgical intervention.

There are various and latest diagnostics aids are available for the accurate diagnosis of sialoliths. The conventional methods for detecting obstructions in the ducts of submandibular/sublingual salivary gland are standard occlusal radiographs whereas lateral oblique X ray of mandible or panoramic radiographs are still being primarily used for the stone in the hilum or substance of the submandibular gland. Other than conventional radiography, advanced imaging modalities are also used like, sialography, xeroradiography (specially for radiolucent sialoliths), ultrasonography, scintigraphy and computerized tomography and magnetic resonance imaging. The NCCT helps in planning and performing the surgery pre/per operatively by revealing accurate size, shape, numbers and the locations, specially in multiple stones.

In our case, presence of multiple no and different sized of the sialolith, and the variable position of the stones (specially the posterior most one, which was far posterior to lower second molar), made surgical removal difficult. Moreover the inaccessibility for the tongue and relation of lingual nerve and the duct attributed the difficulty in few more folds. Trans-oral intervention with single 2cm single incision medial to plica, was thought to be most appropriate to save the gland and other vital structures while removing the three stones. Sialoliths in Wharton's duct and gland can be divided in to three (10) groups; (a) those in the anterior part of the duct, (b) stones in the posterior part of the duct, and (c) the third one when the stones are found at the hylum of the gland or within the gland. Seward described the principles of surgical management very comprehensively. Sialoliths present at the orifice or in the distal half of the duct, are removed trans-orally, because of easy accessibility.

There are various documented complications (10) of trans-oral sialolythotomy like, lingual nerve paresthesia, anesthesia, formation of ranula, as a result of disruption of the sublingual salivary duct etc. Extra-oral approach also carries a risk of permanent/temporary damage of branch of facial (marginal mandibular) nerve (10) in 8% cases. Sialo-cutaneous fistula is also a possible unwanted ending of mega liths because of the severe obstruction of the ducts, resulting in stasis, infection and subsequent rupture through the skin.

CONCLUSION

Multiple sialolithiasis of submandibular gland is uncommon disorder of the ducts and parenchyma of salivary glands. In this case report, retrival of three sialoliths by trans-oral sialolithectomy from the anterior as well as posterior part of Wharton's duct was carried out successfully under local anesthesia without any immediate or latent post-operative complications, while preserving wetness of the mouth by preserving the whole anatomy of the gland. Commonly surgical excision of the gland is undertaken, if there are multiple sialoliths in the duct; or if the sialolithis in the substance of the gland, or in cases of recurrent sialadenitis secondary to sialolithiasis. There are various modalities of treatment of sialadinitis are there like, conservative & noninvasive management (local heat application, massage and sialogogues) (11) for small single non occluding type may be preferred. Sialoliths in the anterior part of the duct are managed by sialolithectomy through a ductotomy, performed intra-orally, under local anesthesia in a co-operative patients. The sialoliths in the posterior part of the duct, are treated by sialolithectomy through a ductotomy performed trans-orally, preferably under general anesthesia. Multiplesialoliths in the duct, or sialolyths below the posterior edge of mylohyoid muscle, or at the hilum of the gland or within the gland, or where the gland has been damaged by recurrent infection and followed by fibrosis, may require surgical removal of the gland and the duct along with the stones. Excision of the whole gland along with the duct through an extra-oral approach under general anesthesia, is preferred when a patient does not respond to any conservative therapy/ductotomy by trans-orally. The future holds great promises due to the development of couple of non-invasive procedures, such as extra corporeal shockwave lithotripsy and endoscopic lithotripsy and so on.

LIST OF PHOTOGRAPHS

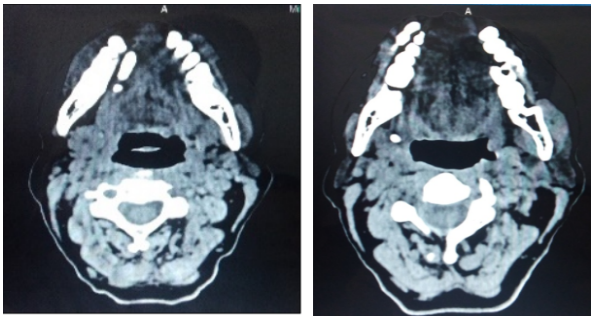


Fig no 1&2: shows sialolith in Rt Wharton's duct right canine–premolar area & medial to ramous of mandible posterior to right lower third molar



Fig no: 3 shows largest sialolith in canine- premolar area (18 mm) as measured in RVG



Fig no: 4 shows placement of an incision of 2cm medial to plica, as designed for the anterior stones & retrieval of stone



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