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A Case Report of Submandibular Gland Sailolithiasis with Recent Modalities of Treatment

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ABSTRACT

Sialolothiasis formation of calcific concretions within the parenchyma or ductal system of the major or minor salivary glands of which submandibular gland has the highest predilection for the sialolithiasis .For small and accessible stones conservative therapies are indicated and for large and inaccessible stones surgical management should be considered .Treatment for sialolithiasis has undergone significant changes since the 1990s. Following the development of new minimally invasive and gland-preserving treatment modalities, a 40 -50% rate of gland resection was reduced to less than 5%.We are presenting a case report with sialolithiasis in the submandibular gland along with the review of literature. The purpose of this paper is to add a case to the literature and review the recent advances in the treatment procedures.

Keywords:Submandibular gland ,Sialolithiasis, Sialolith, Sialolithotomy , ESWL, TDS, ISWL.

I. INTRODUCTION

Sialolithiasis is considered to be the most prevalent salivary gland disorder. In that, Submandibular gland has the highest predilection for sialolithiasis with 80% occurrence rate, followed by the parotid (19%) and the sublingual (1%) glands.Sialolithiasis is commonly seen between the agegroup of 30 and 60 years. Males are affected two times as much as females of which only 3% is reported in pediatric cases.[1]The clinical symptoms are pain and swelling which may be recurrent and most intense during meals.Size of theCalculus may vary from less than an mm to a few centimeters. While patients havingmild sialoliths can undergo classic treatment, those with standard-size or larger sialoliths needs sialolithotomy.Another method is using sialagogues with gland massage to promote salivation and flush the small sialoliths through duct orifice.Extracorporeal shock-wave lithotripsy (ESWL),(2-6) refinement and extension of methods of transoral duct surgery (TDS),(7-9) and in particular diagnostic and interventional sialendoscopy (intSE)(10-11) are major parts of the new treatment plan. It has observed that combining the different treatment modalities further increases the effectiveness of therapy, as has been especially evident with the combined endoscopic–transcutaneous approach. Among these combined approaches, sialendoscopy-guided intraductal shock-wave lithotripsy (ISWL) emerged as an alternative treatment modality within last 5–10 years.(12) All of these minimally invasive techniques became established as part of the state of the art for treating sialolithiasis and were regarded as forming a substantial part of the published minimally invasive treatment plans.(9,13,14,15)

II. CASE REPORT

A 55year old male patient presented our institute with chief complaint of pain, swelling and pus discharge while having food on the right side of the floor of the mouth since 10 days.For the last 6-7 years he was under medication for the same.

On facial examination a diffused swelling over the right submandibular region was found. On bimanual palpation the lesion was firm and tender on the right side of the anterior floor of the mouth. Intraorally rightsubmandibular duct opening was inflamed and erythematous along with discharge of

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pus.Ultrasound scanning and mandibular occlusal radiograph reveals two calcified lesions in the anterior region of right Wharton's duct.

Figure 1:Intraoral view of patient showing Erythematous diffused swelling on the right side of the floor of mouth.

The mandibular occlusal radiograph revealed two radio opacities, one solid radioopacity extending from 44 to 45 tooth region and another cluster of small radioopacities situated opposite to the 46region.

Figure2: Mandibular occlusal radiograph of patient showing sialoliths on right side of floor of mouth

On ultrasound scanning, submandibular duct was dilated and measured upto 7mm. Ultrasound revealed multiple calculi over right Wharton's duct ; one solidmass of size 6mm in the distal part of the duct and another cluster of multiple calculi 2 cm from the ductal orifice.On the basis of clinical and radiographic findings, we arrived at the diagnosis ofright submandibular sialolithiasis.Due to the position of the calculi (close proximity of cluster of calculi towards the hilum)and the patient preference ,surgical removal was thetreatment of choice in this scenario.

Pre-operative investigations were under normal limits and the patient was under antibiotic coverage .Lignocaine 2% with adrenaline given on right side of floor of the mouth.Mucosal incision was given parallel to Wharton's duct from right side canine to the second molar.The duct was opened and a calculi of size approximately 6x3x2mm was removed from the distal part of the duct and sialolith withvaried sizes of 4x2x1mm,3x2x1mm,2x1x1mm,2x2x1mm, 1x1x1mm and 0.5x0.5x1mm from the posterior part of the duct.Irrigation was done with betadine andsaline solution. Incision in the duct is not sutured and mucosal suturing was done with 3-0 solus.

III. DISCUSSION

Sialolithiasis is the formation of calcific concentrations within the parenchyma or ductal system of the major or minor salivary glands.Incidence of Sialolithiasis in Submandibular gland is 80%,Parotid gland is 19% and Sublingual gland is 1% Sialolithiasis is usually seen between the age group of 30 and 60 year.It is uncommon in children as only 3% of all sialolithiasis cases has been reported in the pediatric populations.Males are affected twice as much as females.(1)The cause for larger incidence of the calculus in the submandibular gland is the mixed and viscid nature of the secretions, more inorganic contents, uphill and tortuous course of the duct. The calculi commonly composed of inorganic calcium and sodium phosphate salts. It arise from deposition of these salts around the nidus of debris within the duct lumen. (16)

Usually Weight of the calculi varies from 1mg to almost 6gand Size ranges from 2.1 to 10mm. Most calculi are smaller than 10mm in size ;gaint ones of size larger than 15mm occurring rarely. Location from where the stone originates will determine the shape of a salivary stone. Stones originating from the ductal system are elongated whereas stones originating from the hilus are round or oval in most cases .(36)

According to Clinical symptoms includes pain and swelling.The pain has classical relation with the meals. The pain is aggravated during the meals. If the blockage of the duct is complete,

symptoms will be severe. The obstruction invariably leads to retrograde infection in the gland and the clinical signs of chronic sialadenitis like, purulent discharge, diminished flow of saliva are present.

On palpation calcified, hard calculus may be palpable. In the submandibular gland bimanual palpation of the gland and duct may show presence of calculus. The calculus in the submandibular duct is best seen on the mandibular occlusal radiographs and the calculus in the hilum is seen on lateral oblique view of mandible.

Sialography plays an important role in identifying the location of the calculus.

There is filling defect in the main duct. Ductal dilatation in the proximal to the calculus. The emptying phase usually shows contrast medium retained behind the stone.

Management of sialolithiasis can be conservative or radical depending upon the size, location, and mobility of the stone (23).Conservative management of sialolithiasis includesantibiotic therapy, anti-inflammatory drugs (NSAIDs), massaging the salivary gland, and sialagogues in which antibiotic therapyis the prime treatment option for Signs of infection including cervical lymphadenopathy, purulent discharge from the salivary ducts, or erythema surrounding the salivary ducts.Definitive surgical treatment can be instituted once the acute stage subsides.(23)

Treatment for sialolithiasis has undergone major changes Since 1990s,as the new minimally invasive and gland-preserving treatment modalities, was developed and a 40–50% rate of gland resection was reduced to less than 5%.(11,13)

The traditional management of obstructive salivary disorders suggests sialadenectomy in case of proximal,hylar or intra parenchymal sialoliths,duct dilatation, incision and dissection in case of distal stones, and sialodochoplasty(20).These techniques are supposed to have some risks such as permanent mandibular nerve palsy(1-8%),lingual nerve palsy(1-5%),so it has been replaced by minimally-invasive gland-preserving techniques,such as shock-wave lithotripsy, refinement and extension of methods of transoral duct surgery (TDS), particular diagnostic and interventional sialendoscopy (intSE)(6)and robot assisted sialolithotomy with sialendoscopy(35).This may be used individually or combined, which reduces the need for gland removal and its unpleasant complications and loss of function, and assure complete clinical recovery in more than 80% of patients.(19)

Shock-wave lithotripsy was one of the initially introduced method, and is probably the most standardised of the newer techniques. since the late 1980s , salivary stones was treated with **Extra-corporeal shock-wave lithotripsy**.(ESWL) has been successfully used to treat in a way that is similar to that used in urology and gastroenterology. salivary stones were fragmentized into smaller pieces using the shock-waves generated by the change in impedance at the stone/water interface, which induces a compressive wave that spreads through the stone and an expansive wave that pits and cavitates it, (24)thus making it more easily cleared from the salivary duct system spontaneously after sialogogue-induced salivation, or during endoscopic procedures. with Shock-wave lithotripsy. The shock waves may be produced extra-corporeally by piezoelectric(25) and electromagnetic techniques,(26-28) or intra-corporeally by using electro-hydraulic,(29) pneumatic,(30,31) or laser sources during interventional sialendoscopy.(32,33)Advantages of ESWL are,easy to perform, repeatable, safe, generally well-tolerated, and can be carried out on an outpatient basis without anaesthesia, and main limitations are the stones often cannot be completely cleared by salivary flow, and small residual fragments remaining inside the ductal system may cause recurrences. In order to remove all of the remnants, It is better if any ESWL treatment is followed by sialendoscopy .Other disadvantages are that it is time-consuming (it generally requires repeated 30-minute sessions at intervals of a few weeks) and is not currently approved by the US Food and Drug Administration. For these reasons, together with the cost of the equipment and its maintenance, and the rapid development of advanced interventional sialendoscopic procedures using optical miniaturisation and micro-instruments, fewer centres are now offering ESWL, which is being gradually replaced by endoscopically assisted intra-corporeal techniques for stones which are endoscopically accessible.(13)

According to Johannes Zenk et al **Transoral removal of the stones** within the distal part of the Whartons duct in the floor of the mouth is not a major surgical problem,whereas sialolithectomy within the duct posterior to the first molar or even more proximally in the so-called comma area(where the ducts turns inferiorly at the posterior border of the mylohyoid muscle)is difficult and may be hazardous to lingual nerve.Nevertheless , in the literature expanded

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incision of the duct has been continually favoured as a gland preserving therapy.(20)

In **intra-corporeal lithotripsy**, lithotripsy probe was placed inside the salivary duct system and under endoscopic guidance they produces the shock-waves toward the stones.The energy needed to fracture the stone is usually provided by means of a laser beam, pneumatic devices, or electro-hydraulic probes(22). The primary indication for ISWL is with stones in which gland-preserving treatment is not possible using the well-established techniques . ISWL has also been used for when there is difficult anatomy in the ductal system (narrow duct, duct variations),when the location is difficult (with poor accessibility)or in the presence of multiple stones as a single-mode treatment, and also in combination with other techniques.(22)

Gundlach et al. reported that in 1990s ,the first successful application of **endoscopically guided intra-corporeal lithotripsy for salivary stones using a laser beam** was introduced and it achieves 92% of stone clearance. It was done using Holmium YAG (yttrium-aluminum-garnet) or pulsed dye lasers(33)the former is associated with a high risk of soft tissue damage, and their difficulty of use is attributable both to their thermal effects and absorption by the surrounding tissues ; the latter are harmless, but extremely expensive.(22)

Raif et al. recently proposed the development of an erbium (Er) fibre delivery system for endoscopic lithotripsy of salivary stones: hollow metal wave guides optimised for an Er: YAG laser were end-sealed with a polished sapphire rod of 0.63 mm, designed to adapt to the laser and the sialoendoscope(22)

Sialoendoscopy was first described in 1991 by Katz .(34)He used a 0.7 mm flexible endoscope to remove salivary stones with Dormia baskets.It can be used as diagnostic or interventional tool for inflammatory and obstructive pathology of the ductal system ,thus providing an alternative method to open surgery and its related complications.Sialendoscopy is a conservative minimally invasive treatment by using 0.8 to 1.6mm width micro-endoscope which is outfitted with a light ,camera and tools.By using grasping tools like forceps or basket retrieval of the stones is possible. One of the absolute contraindication to this procedure is complete distal obliteration of the duct that is impenetrable by the endoscope. The most frequent side-effect of sialoendosopy is a transient glandular swelling due to the irrigation with physiological solution in 80-100% of cases , but ductal strictures (2-4%) or lacerations (1-8%), basket block (6%), infections (2-3%), temporary lingual nerve paresthesia (0.4-0.6%), ranula formation (0.6-0.9%) and bleeding (0.5%) have also been described(22).To reduce these side-effects a proper training with a high level of dedicated experience is needed ,especially for doing balloon dilatation or laser lithotripsy.

Konigsberger et al. in 1993 introduced **endoscopically controlled intra-corporeal electro-hydraulic lithotripsy** by placing a flexible fibroscope with an additional probe inside the ductal system. The tip of the probe electrode placed 1 mm in front of the stone and the shock wave was produced. The clinical trial led to complete stone fragmentation in 20/29 patients with sub-mandibular sialolithiasis . Iro et al. criticised this procedure on the basis of the results of in vitro and experimental animal studies, as having a high risk of ductal iatrogenic injuries and being scarcely efficacious at low voltage, and it has now been abandoned on account of possible tissue damage.(22)

Arzoz et al. in 1996 introduced a **Endoscopically controlled intra-corporeal pneumatic lithotripsy** bya rigid 2.1 mm urethroscope with a 1 mm working channel in order to perform intraductal stone fracture using both a pneumoblastic lithotriptor and a laser device under endoscopic control in 12 patients. Pneumatic lithotriptors work by means of ballistic energy and can be likened to a biological “pneumatic hammer”.its major complication wastheir high risk of ductal perforation.

Several groups reported transoral robot-assisted extraction of deeply located and large submandibular sialoliths , also in combination with sialendoscopic assistance. But 10–20% of stones are located intraparenchymally, and they were not primarily accessible using TDS, any modification of intSE, or combined surgery, so that ESWL often remained as the only available treatment method. In addition to this, a further 5–15% of stones could not be successfully treated by any method.(12)

Sequential or simultaneous combination of ESWL and ISWL has been reported in a few studies, in which ISWL contributed to the improvement of treatment results and supplemented the combined treatment options. In an earlier study, Katz et al. reported that a combination with sequential use of the two techniques led to an improvement in the results of around 30% .(6)Koch et al. reported that patients presenting with difficult sialolithiasis could be treated with ISWL after the application of ESWL with a success rate of > 90%. In that study, stones that were not adequately visible and/or accessible for intSE could be mobilized using ESWL and were ultimately successfully treated with ISWL in

95% of cases in both major salivary glands. (21)A combination of ISWL and TDS has also been reported in cases of difficult sialolithiasis in the SMG. Stones that are not removable with TDS (persistent sialolithiasis), residual stones in patients with multiple sialolithiasis, and recurrent stones after TDS in posthilar to intraparenchymal locations are important new indications for ISWL, as has been shown in recent publications. (12)

According to Christopher Razavi ,they demonstrates the safety and efficacy of RASS (robot assisted sialolithotomy with sialendoscopy)in the management of large submandibular salivary gland sialoliths. We attribute this preliminary success of RASS to the enhanced visualization, magnification, and technical advantages of the procedure.(35)

IV. CONCLUSIONS

In conclusion, recent advances in sialendoscopy-guided treatment for salivary stones have been made and have been reported in published studies during the last 5–10 years. The literature results point to the growing importance of ISWL, for which very good results have been reported. This has led to an expansion of the scope for less invasive, gland preserving treatment. It is an effective, but not always a faster form of treatment. ESWL is still of value, particularly in combination treatment, which further improves the results. ESWL can also make stones amenable to ISWL treatment, and the two methods can supplement each other. As combination treatment is more promising in cases of difficult sialolithiasis, there may be greater effort involved in such cases. This must be discussed with the patient, and adequate counselling is needed. As all of the procedures can be performed with local anesthesia, general anesthesia can be avoided in the great majority of cases, representing a major advantage for patients. In the light of this, gland-preserving treatment should always be considered as the first choice.

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