SURGICAL MANAGEMENT OF A LARGE RARE SUBMANDIBULAR SIALOLITHIASIS - A CASE REPORT AND REVIEW OF LITERATURE

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Abstract

The most prevalent pathology affecting the salivary glands is sialolithiasis. Because of the submandibular gland's long, tortuous, tiny orifices, increased mineral content, alkaline pH, high saliva viscosity and gravity-defying drainage, sialolithiasis of the gland is most frequent.The preferred course of treatment for minor, accessible stones is conservative care, such as draining the tube while receiving palliative care.

Surgical therapy should be chosen for sialoliths that are big and numerous. In this report, we describe a patient who had three different-sized sialoliths in the right submandibular duct. The patient was treated with an intraoral surgical approach involving lingual frenum drainage under local anaesthesia. Follow-up was done on a regular basis and no surgical complications occurred before, during, or after the procedure

Categories: Dentistry, Pain Management

Keywords: radiopaque lesion, rare submandibular duct sialolithiasis, intra oral approach, sialolithiasis, submandibular gland

Introduction

Sialolithiasis is the most common pathology of the salivary glands [1] which affects more than 1% of the global population [9]. Sialolithiasis is defined as tartar in the salivary gland or its excretory system, causing an obstructive phenomenon [4-5].

It occurs most often in Wharton's duct followed by Stensen's duct, and rarely in the sublingual glands [6]. Sialolithiasis of the submandibular gland is most common due to the alkaline pH and high viscosity of saliva, higher mineral content, long, tortuous and narrower orifices of its ductal system, and drainage of the submandibular gland against gravity [2]. Sialolithiasis usually presents as swelling followed by pain of the involved gland which worsens on having food [7,10]. There is more swelling and pain when the stone is stuck in the duct than when the stone is in the gland itself [11]. The size of salivary stones can vary from a few millimetres to several centimetres [8]. The

largest reported stone in the submandibular canal was 72 mm long [11]. For small and easily accessible stones the treatment of choice is conservative treatment such as draining the tube with palliative care. For large and multiple sialoliths, surgical treatment should be chosen [3].

In this report, we present the case of a patient with three sialoliths of different sizes in the submandibular canal. We treated this case with an intraoral surgical approach, where lingual frenum drainage was performed under local anaesthesia.

The purpose of this article is to describe the diagnosis and surgical treatment of sialolithiasis of the submandibular duct using an intraoral approach.

Case Presentation

A 35-year-old woman presented to the Department of Oral and Maxillofacial Surgery, Sathyabama Dental College and Hospital with a chief complaint of swelling on the floor of the mouth associated with pain. Patient was not aware of the exact timeline of onset of her symptoms. No relevant findings were found during the external investigation. Oral examination revealed a 1 x 2 cm swelling on the right side of the floor of the mouth that extended from the lingual frenum to the first buccal area along the course of the submandibular canal (Fig 1). The upper mucosa was found to be normal. On palpation, the swelling was firm to hard in consistency, non-tender and not attached to the underlying structures. It extended approximately 5 cm along the course of the duct, and salivary flow was found to be normal. Occlusal radiographic examination revealed 2 radiopaque masses measuring 5 x 1.5 cm extending anteriorly and medially from the mandibular right incisor to the first buccalregion in the floor of the mouth, suggestive of sialolith. Axial section of CT scan revealed radiopaque material in the lingual part of the mandible on the right side. Two distinct radioactive masses suspected to be sialoliths were found on the right side of the lingual area of the mandible (Fig 2). The coronal and sagittal sections were also directed to the sublingual radioactive organs in the right mandibular region. Evaluation of substance P in saliva by polymerized chain reaction test was done before and after the procedure. After administration of local anaesthesia with adrenaline, a retraction suture was placed around the canal distal to the stone which was then withdrawn anteriorly. Yellow colour coded 24G IV cannula measuring OD 0.7 mm and length 19 mm with flow rate 20 ml/min was used to maintain the patency of the right submandibular Wharton duct. We secured the cannula with 3-0 black braided silk with suture (Fig 3). A mucosal incision was made in the visible area of the sialolith to expose the Wharton duct, and then a linear incision was made along the course of the Wharton duct. A skin hook was placed on both sides of the incisions to retract it, and then a blunt dissection was done with a curved haemostat to reveal the visibility of the anterior sialolith (Fig 4). Blunt dissection was performed around the sialolith to expose each calculus and the first sialolith was removed which was approximately 2 x 1 cm in size (Fig 6). Then, two-handed milking of the canal was done to remove the second calculus which was less than 0.5 cm in size (Fig 7). The last calculus measuring 1.25 x 0.5 cm was removed by double milking of the canal to expose the calculus which was removed using an explorer (Fig 8). Figure 9 shows complete removal of all 3 calculi. Saline irrigation and

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milking of the gland was performed to remove small stones and mucus plugs in the duct. The wound was approximated with some 3-0 black braided silk thread. The IV cannula was also secured around the opening of the cannula with 3-0 silk thread after the IV cannula needle was removed (Fig 10). Patient received postoperative instructions along with antibiotics and analgesics.

Follow-up and removal of sutures was done after seven days. Recovery was found to be normal and the patient was asymptomatic (Fig 11).

FIGURE 1: Preoperative Intraoral picture

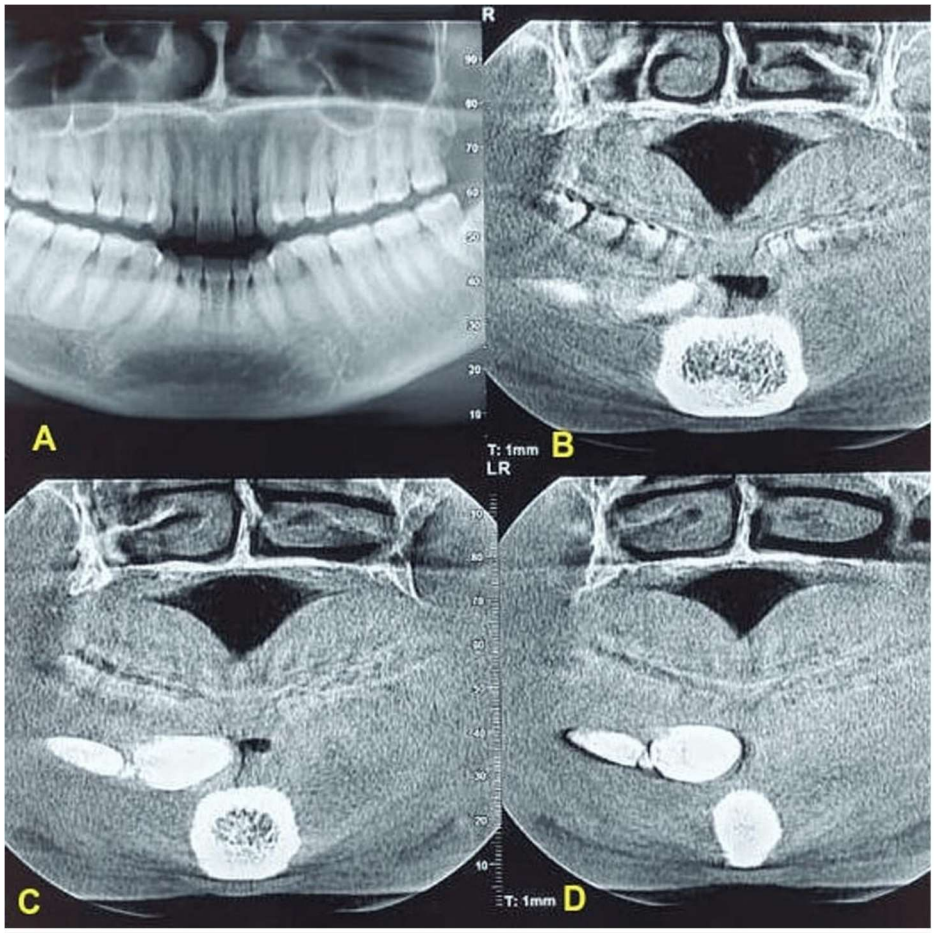

FIGURE 2: A, Pre operative OPG image. B,C,D Shows 3 sialolith located on right side of the floor of the mouth

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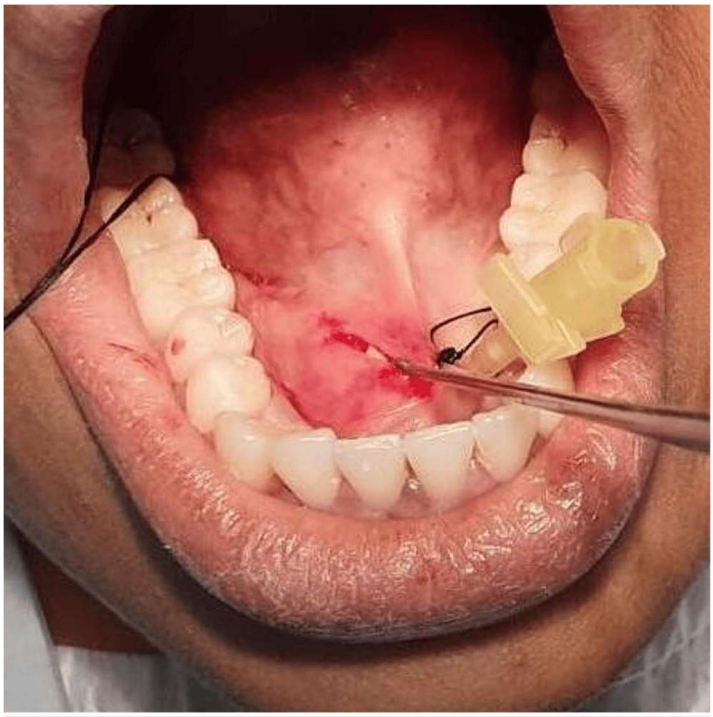

FIGURE 3: 24G Yellow cannula placed inside the right Wharton's duct to maintain the patency with intraoral mucosal incision

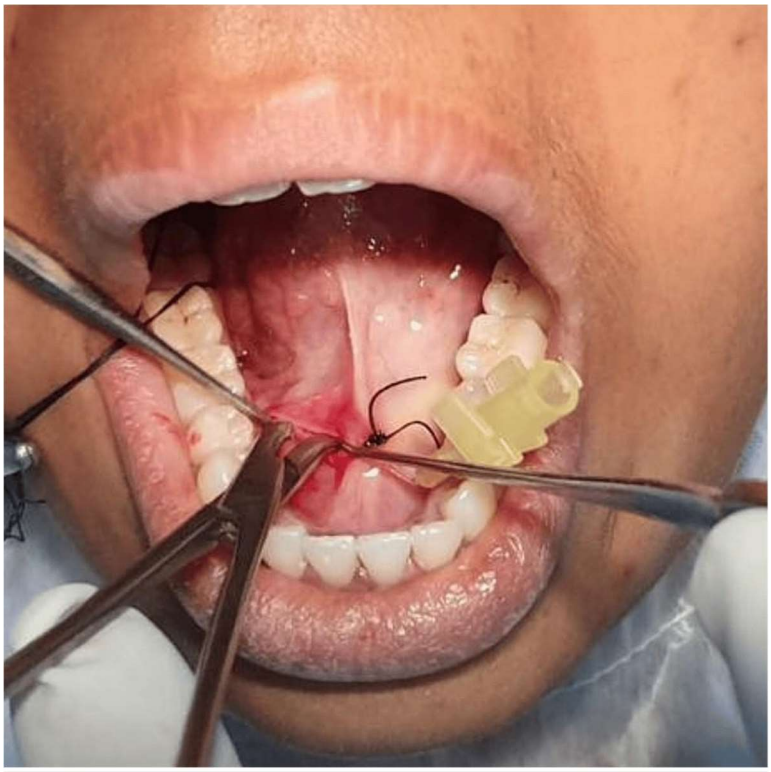

FIGURE 4: Blunt dissection done with scissors and incision edges are retracted with skin hook

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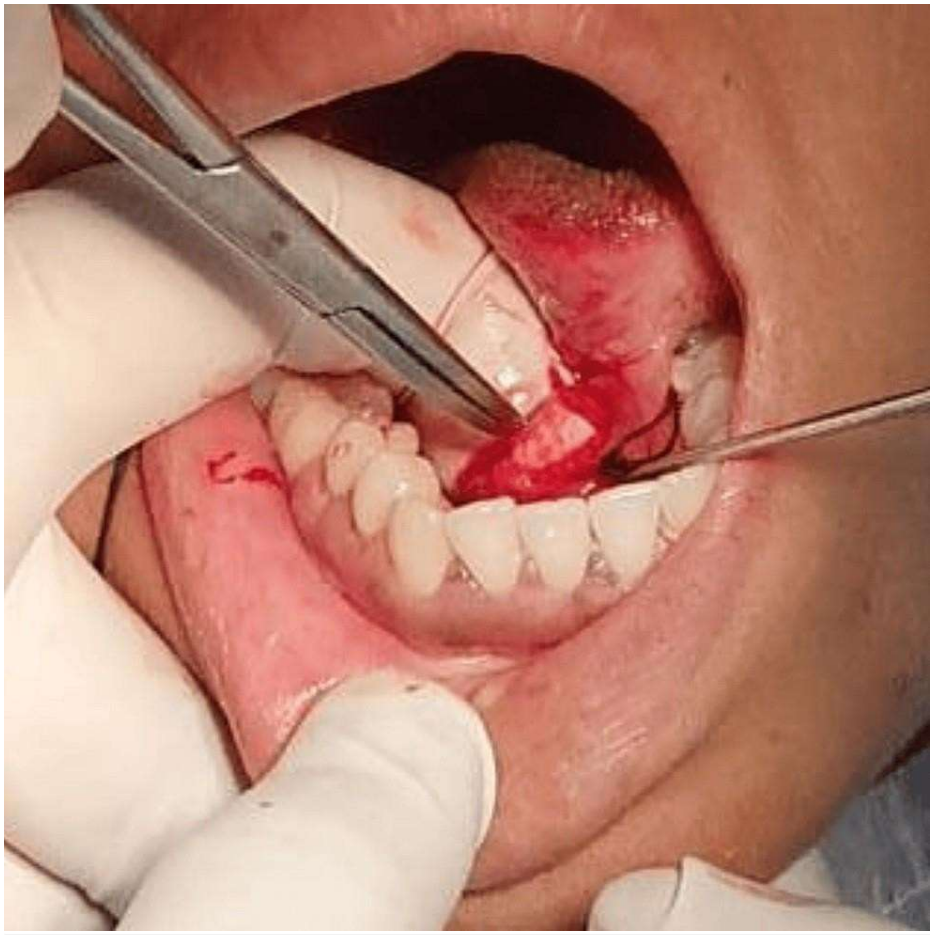

FIGURE 5: Bimanual palpation of Rt Submandibular Gland

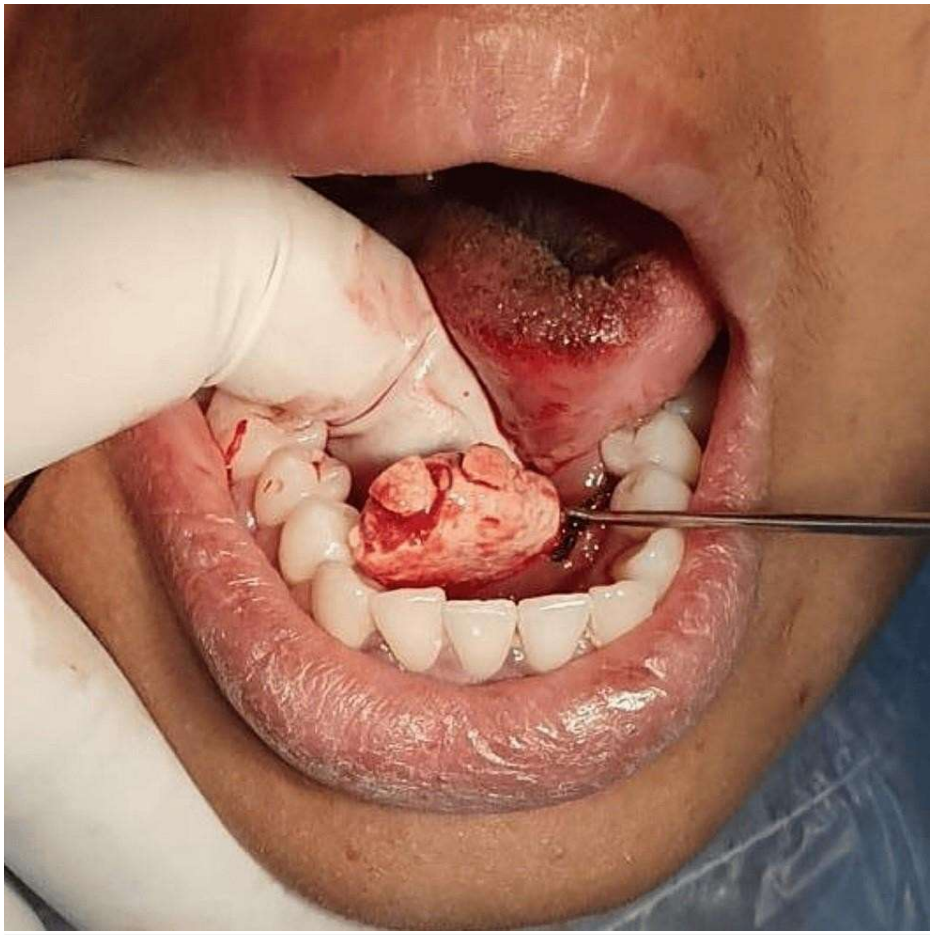

FIGURE 6: Removal of 1st Large calculi by Bimanual palpation of Rt Submandibular gland

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FIGURE 7: Removal of 2nd calculi removed by Adson's non toothed forceps

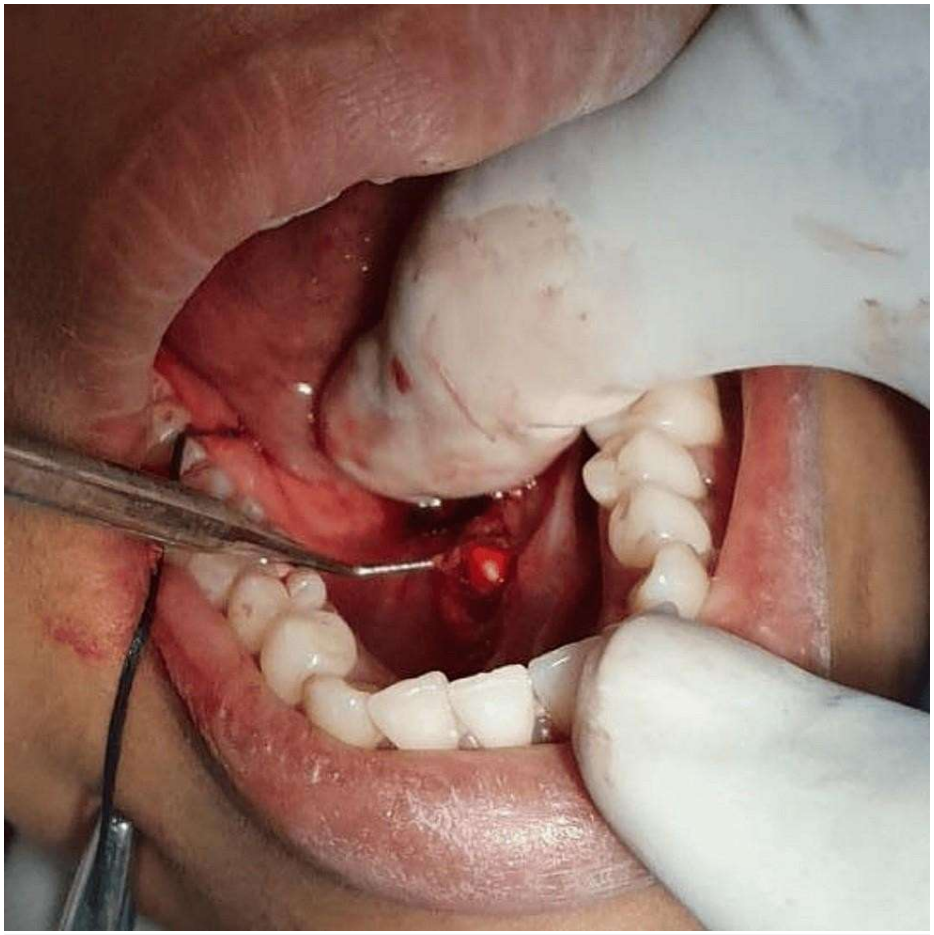

FIGURE 8: Removal of 3rd calculi by probe

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FIGURE 9: Excised Sialolith in order

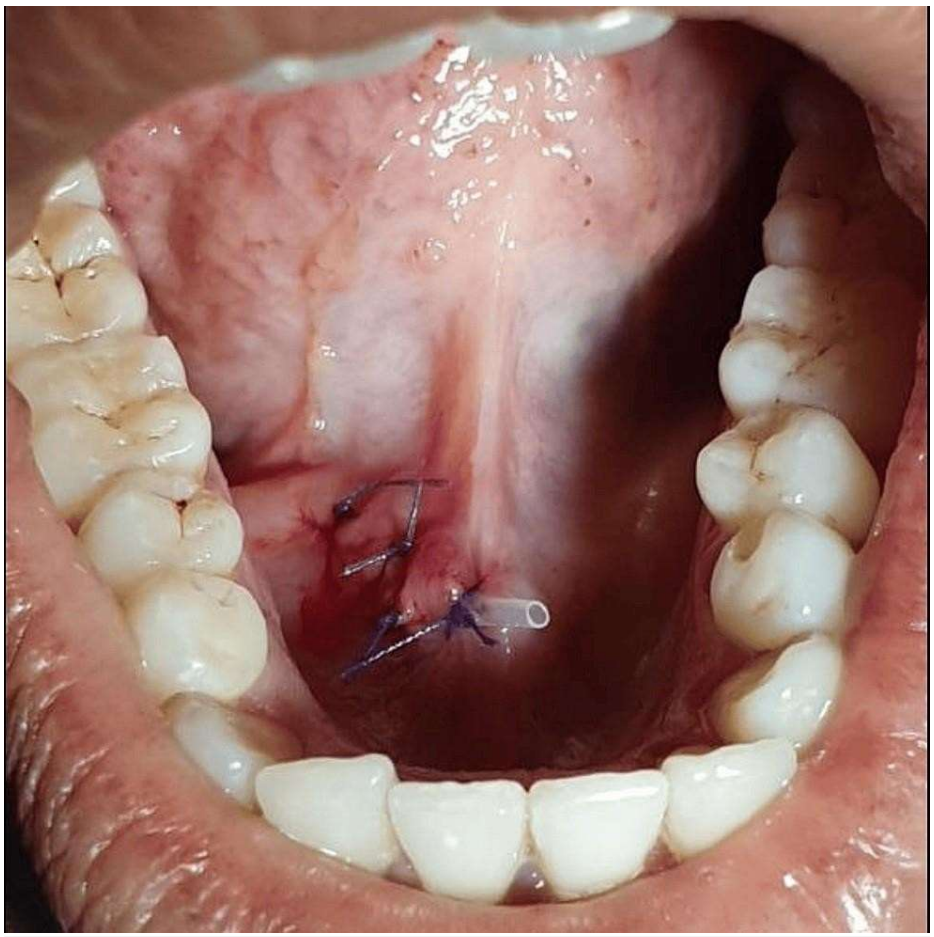

FIGURE 10: Wound closure with stent in place

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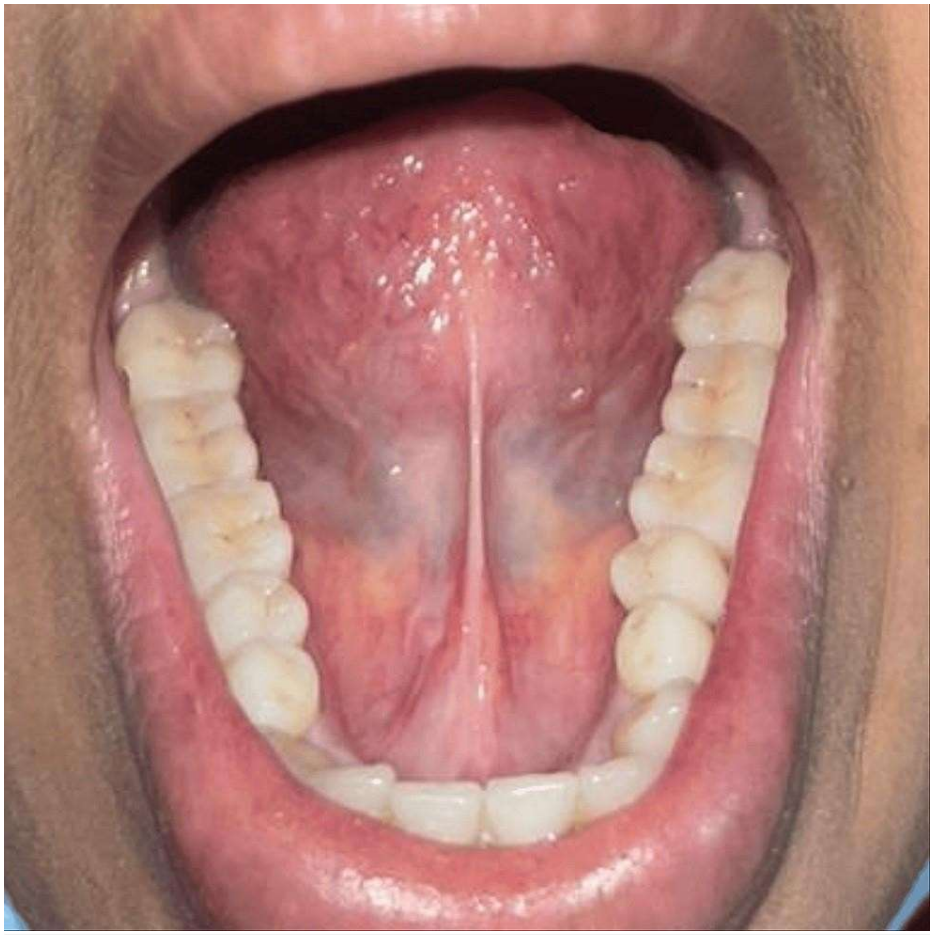

FIGURE 11: Postoperative healing after 3 month with patent Rt Wharton's duct

Discussion

REVIEW OF LITERATURE ON DIAGNOSIS OF SUBMANDIBULAR DUCT SIALOLITHIASIS

Sialolithiasis of the submandibular ducts refers to the formation of stones (sialoliths) in the duct system of the submandibular gland [11]. This condition is characterized by recurring pain and swelling that often worsens during meals [5]. Diagnosing sialolithiasis of the submandibular duct can be difficult, but various imaging methods can help [12]. The purpose of this review is to summarize the current literature on diagnostic methods for submandibular ductal sialolithiasis and to discuss their advantages and limitations.

A plain x-ray

Plain radiography (intraoral or panoramic radiography) is often used to detect salivary gland stones due to its accessibility and low cost. However, their sensitivity is limited because small or uncalcified stones may not be detected [13].

Ultrasound

Ultrasound is a non-invasive, inexpensive and widely available imaging technique for the diagnosis of sialolithiasis [15]. It shows high sensitivity and specificity in stone detection in most cases [4]. Ultrasound can also assess salivary gland inflammation or fluid accumulation around the stone [15]. However, its accuracy depends on the skill and experience of the user.

Computed Tomography (CT)

A CT scan can provide detailed cross-sectional images of the salivary glands and their associated anatomy. This technique has excellent sensitivity and specificity for the detection of stones and complications of sialolithiasis, such as abscesses [5]. However, CT scans expose patients to ionizing radiation and are more expensive than other imaging methods [16].

Magnetic resonance imaging (MRI)

Magnetic resonance imaging (MRI) offers the advantage of imaging soft tissues without ionizing radiation [17]. MRI can detect salivary duct obstruction and glandular inflammation associated with sialolithiasis [18]. However, small calcifications may not show up well with MRI and the procedure is time-consuming and more expensive than other methods.

Sialography

Sialography involves the injection of a radioactive contrast agent into the salivary duct system and imaging with x-ray equipment. Although invasive, it provides detailed information on canal anatomy and function. Because of its invasiveness, exposure to radiation, and potential complications (such as allergic reactions), sialography is usually reserved for cases where other imaging methods are inconclusive [4].

Conclusion

Diagnosis of submandibular ductal sialolithiasis requires a combination of clinical findings and imaging studies. Ultrasound is recommended as the primary imaging modality due to its non-invasiveness, low cost and high sensitivity. If the ultrasound results are inconclusive or additional anatomical details are needed, computed tomography can be used. Magnetic Resonance Imaging can provide valuable information about soft tissues but may be limited in detecting small calcifications. Sialography has a role in difficult cases where other methods have been inconclusive.

VARIOUS APPROACHES AND THEIR EFFECTS:

There are various intraoral approaches to treat submandibular ductal sialolithiasis, such as sialendoscopy, marsupialization (Sialodochopasty), and transoral incision. Each technique has its own advantages and complications. The intraoral approach shares advantages such as being minimally invasive and better cosmetic results because it avoids external incisions and scars, preserves salivary gland function and faster recovery due to less postoperative discomfort.

SIALENDOSCOPY:

Sialendoscopy is a minimally invasive method used for both diagnostic and therapeutic procedures. The procedure is mainly performed under local anaesthesia. A thin, flexible endoscope and a light source and camera are inserted into the submandibular canal, usually through the papilla under the tongue. An assessment is made, such as the size and location of the sialolith. Sialendoscopy uses various techniques to remove stones such as basket removal, laser lithotripsy and irrigation with saline solution or drugs. After treatment patients are prescribed antibiotics and post-treatment instructions with regular monitoring [19]. Signs include smaller stones less than 5 mm in diameter and moving stones. For larger stones, sialendoscopy is considered an additional procedure along with other met hods. Sialendoscopy is contraindicated in acute ductal and glandular inflammation because it increases pain and inflammation.

The advantage is that it is a minimally invasive procedure that does not require an incision, preserves the function of the glands, allows the stone to be directly visualized, the recovery time is relatively shorter, and the success rate is high. The disadvantage is that it is effective only for small stones, smaller pieces can reach behind the channel and it is expandable [20].

MARSUPIALIZATION (SIALODOCHOPLASTY):

Sialodochoplasty is a procedure in which post salivary stones removal, the length of the duct is shortened after a catheter is placed to maintain saliva flow, and the edges are sutured to the oral mucosa. Indications are large and persistent salivary gland stones, recurrent sialolithiasis, inflammation and failure of conservative treatment. The advantage is that it prevents recurrence; this shortens the length of the channel, thus eliminating the narrowing of the point. Potential complications include increased postoperative hypoesthesia, increased need for generalanaesthesia, and increased operative time [21].

TRANSORAL SECTION:

A transoral incision is a minimally invasive surgical technique. Indications include the presence of smaller stones and the presence of a single stone when there is no obstruction. Contraindications for a transoral incision are when access to the canal is difficult, severe inflammation and infection, and lack of experience because the possibility of nerve damage during the incision is greater. Complications include excessive bleeding, the possibility of infection and scarring [22].

DISCUSSION:

In the case of submandibular sialolithiasis, pain can vary in individual cases, which can be intermittent, continuous, dull, sharp and even painless. ANAND GUPTA et al. in 2013, reported two cases of submandibular sialoliths of unusual shape with intermittent dull aching pain[22]. VINISHA S. POUSHYA et al. reported pain was sudden moderate and constant. In this particular case, the patient experiences chronic, dull and gnawing pain [23].

VON EULER AND GADDUM discovered substance P in 1931, an 11 amino acid neuropeptide that plays a critical role in pain perception and is produced by neurons located in the spinal cord and trigeminal ganglion [24]. Substance P is significantly detected in dental pain and inflammation and mediates and maintains harmful stimuli and inflammatory processes [25]. In our case, the level of substance P increases due to inflammation of the submandibular gland duct. An increase in the concentration of substance P is also observed in some other cases, such as pulpitis, granuloma, and orthodontic treatment. In th is case, we evaluated substance P levels by polymerase chain reaction (PCR) both before and after surgery. In an article entitled Postoperative healing after surgical removal of a mandibular third molar: A comparative study between two proteolytic enzymes by THARANI KUMAR et al., 2020, the substance P level was evaluated by PCR [26].

Prescribed medications are Oraways cream (containing triamcinolone acetonide), Rutoheal

tablet (combination of trypsin, bromelain, Ruto side and diclofenac) and Metroplex crea m. Triamcinolone acetonide is a synthetic glucocorticosteroid and has both anti-inflammatory and immunosuppressive properties [27]. Its mechanism of action is by inhibiting the white blood cell function, stabilising the cell membranes, and reducing the synthesis of inflammatory mediators like prostaglandins and leukotrienes. This reduces inflammation, subsequently decreasing substance P levels. THARANI KUMAR et al.in 2020, compared two proteolytic enzymes from different origins (serratiopeptidase, trypsin, bromelain, rutin) to prove which provides faster healing and concluded that trypsin, bromelain and rutin combination is more effective because clinically, the size of the swelling and exudate from the surgical site was found to be significantly reduced post-operatively [26].

The diagnosis of sialolithiasis requires careful evaluation and knowledge of the differential diagnosis, as it can be confused with sialadenitis and tumours. Various imaging techniques are used for diagnosis, including 2D techniques such as traditional occlusal radiographs and 3D techniques such as Computed Tomography (CT), Magnetic Resonance Imaging (MRI), Cone Beam CT (CBCT), sonography, sialography, and sialendoscopy. Each imaging method has its own advantages and limitations. Conventional occlusal radiography was the first evaluation choice. In most cases, stones appear oval on conventional X-rays. The main disadvantage is that only larger stones were detected, and smaller sialoliths were missed because few sialoliths are less radioactive due to lower mineral content[5]. MANJUNATH RAI and others mentioned in 2009 that the shape of a sialolith is cylindrical in panoramic radiographs. The disadvantage is the sialolith image sometimes overlaps with other structures [28]. Computed tomography has often been an important modality in the diagnosis of both intraductal and intraglandular sialit hiasis. In CT, onlya small

part of the sialolith can be seen in each section, making it difficult to define the shape of the sialolith. There are two types of neck CT - contrast-enhanced CT of the neck (CECTN) and non-contrast CTof the neck (NCCTN). In NCCTN, the details of glandular tumours are less visible than in CECTN. Blood vessels can be seen as small sialoliths and cause false positive CECTN results. YM PURCELL et al conducted a study in 2017 to evaluate the accuracy of CECTN in the diagnosis of sialoliths compared to NCCTN in 92 cases and concluded that CECTN is accurate in diagnosis and that monophasic CECTN should be performed insteadof biphasic CT(which includes both CECTN and NCCTN) thus reducing the effective radiation of the patient [29]. MRI can be used to detect both radioactive and soft tissue tumours, and also determine their exact location, size, shape and pathology. The advantage of MRI is that it does not use ionizing radiation. The disadvantages of MRI are it does not recognize obstructive stones in the salivary gland and the technique is expensive.The CBCT method has been found to be very accurate in imaging and shows the exact location and size of the sialolith. The image obtained by CBCT was dimensionally stable and anatomically accurate. In this case, we prefer CBCT to CT because it has lesser radiation than CT and covers the whole part in one section. Melek TASSOKER et al. reported two cases in 2016 where they used CBCT as a diagnostic method and concluded that CBCT is the preferred imaging technique for the diagnosis of salivary stone and its main disadvantage is that it cannot provide information about the anatomy of the duct and the surrounding soft tissue pipeline paper towel [30]. MIGUEL GONCALVES and others in 2017 pointed out in their study that sonography seems to be a very suitable method for diagnosis and treatment. The advantages of sonography are it is economical and the sensitivity is 94.7% and the specificity is 97.4%. Disadvantages of the technique are that it is less sensitive to tumours and stones that are smaller than 2 cm in size [31]. Sialography is the gold standard method to diagnose a sialolith, as it provides a clear image of the stone and ductal morphology. In this method, a radioactive contrast agent is injected into the gland and X- rays are imaged. In our case, we did not use sialography because our patient was allergic to contrast agents and the patient could not afford this technique, which is the main disadvantage of sialography. The technique of sialendoscopy allows direct imaging of the stone and has been used for both diagnostic and therapeutic purposes. It is performed under local anaesthesia. Complications of this technique include sensitivity of the technique and temporary swelling. THOMAS PNIAK et al stated in 2016 that its sensitivityand specificity are significantly higher than ultrasound and sialography [32]. Treatment usually begins conservatively with methods such as sialagogues, hydration, heat, manual manipulation, and non steroidal anti-inflammatory drugs (NSAIDs). If signs of infection or redness appear in the immediate area, antibiotic therapy with first-generation cephalosporins may be necessary. The choice of treatment depends on the size, location and number of stones. Options include interventional endoscopy, transoral ductal excision for larger impacted stones, endoscopy, intraductal shock wave lithotripsy, extracorporeal shock wave lithotripsy and in severe cases submandibulectomy. In this case, we performed an intraoral surgical approach to the sialolith.

MANJUNATH RAI and others on 2010 performed a sialolithotomy with sialendoplasty using an intraoral approach to avoid the morbidity associated with sialadenectomy [28]. IRO et al.

mentioned in 1992 that extracorporeal piezoelectric shock therapy appears to be a safe, convenient, effective, minimally invasive, nonsurgical treatment for salivary stones [33]. ODE NAHLIELI et al. conducted a study in 2011 and concluded that the canal stretching technique is the choice for posterior and ethmoid stones with a diameter of 5 mm to avoid submandibulectomy [34]. YU-TING SUN et al. performed a sialendoscopy with the holmium: YAG laser and reported that it appeared to be effective in fragmenting and removing several larger stones [35]. If all other treatments fail, submandibulectomy should be the last treatment option.

Complications after submandibulectomy may include Frey's syndrome, scarring, xerostomia, and lingual nerve paraesthesia [33]. Therefore, operator skill is critical to minimize these complications. In this particular case, the patient had no postoperative complications during regular follow-up.

Conclusions

Sialolithiasis is one of the most prevalent conditions of the salivary glands which is simple to diagnose based on its clinical characteristics. Computed tomography remains the gold standard for determining the location, size, and quantity of salivary stones. When choosing the treatment modalities one mustconsider the patient's compliance, propensity for micro-invasive procedures, and medical and surgical history.

The primary therapy for sialolithiasis is still surgery with local anaesthesia being preferred wherever feasible. Local anaesthesia may be used during surgery depending on a number of factors such as location, volume required, and number of calculi. Other options such as lithotripsy and other surgical procedures do not seem to be much feasible.

Additional Information

Disclosures

Human subjects: Consent was obtained or waived by all participants in this study. Conflicts of interest: In compliance with the ICMJE uniform disclosure form, all authors declare the following: Payment/services info: All authors have declared that no financial support was received from any organization for the submitted work. Financial relationships: All authors have declared that they have no financial relationships at present or within the previous three years with any organizations that might have an interest in the submitted work. Other relationships: All authors have declared that there are no other relationships or activities that could appear to have influenced the submitted work.

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