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Case Report

Steering through parotid calculi in a 35 year old female: A case report

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A R T I C L E I N F O

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A B S T R A C T

Sialolithiasis, the predominant affliction affecting salivary glands, constitutes approximately half of all major salivary gland pathologies. The vast majority of salivary gland calculi, roughly 80-90%, are located in the submandibular gland, with 5-10% in the parotid gland, and an estimated 0-5% in the sublingual and other minor salivary glands. Typically, sialolithiasis manifests in adults between the ages of 30 and 60, with a greater occurrence in males. This malady is predominantly observed in adults and is infrequent in children. Herein, we present a case study of calculus retrieval in a 35-year-old woman, characterized by recurrent swelling and pain in the left parotid gland, exacerbated during meals due to ductal obstruction impeding saliva flow. This hindrance impedes gland drainage, resulting in inflamed ducts. Sialadenitis subsided following calculus removal.

1. Introduction

Sialolithiasis, also referred to as salivary gland stone or salivary calculi, is characterized by the obstruction of salivary glands or their ducts due to calcified deposits known as sialoliths. Symptoms may include swelling, pain, and potential infection, though pain is absent in approximately 17% of cases.[1] In acute obstructive instances, the gland appears enlarged, and excretory ducts proximal to calculi may be visibly dilated. In chronic cases, glandular fatty atrophy can develop, making the condition asymptomatic unless a secondary infection arises.[2] It accounts for about 30% of salivary gland disorders, with the submandibular gland being the most frequently affected (80% to 95%), followed by the parotid gland (5% to 20%). Involvement of the sublingual gland and minor salivary glands is uncommon (1% to 2%).[3] This condition predominantly affects individuals aged 30 to 60 years, with a low occurrence in children, comprising only 3% of pediatric cases. It is more prevalent in males, particularly when the parotid gland is involved.[4] Sialoliths vary in size from less than 1 mm to several centimeters in diameter. While most calculi are smaller than 15 mm, giant salivary gland calculi larger than 10 mm are rare and mainly documented in isolated case reports.[5] Salivary gland calculi have several proposed causes, including mechanical, inflammatory, chemical, neurogenic, infectious, and foreign body-related factors.

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Typically, it is a combination of these factors that leads to the formation of amorphous tricalcium phosphate, which subsequently crystallizes into hydroxyapatite, initiating the development of calculi.[6] Hydroxyapatite then serves as a catalyst for the accumulation of additional deposits of various substances. Salivary gland calculi are relatively uncommon consequences of salivary gland inflammation, forming due to the mineralization of debris, although the exact cause remains uncertain.[7] Burnstein et al.'s study utilized x-ray diffraction and chemical analyses to identify apatite, magnesium-substituted whitlockite, and octacalcium phosphate as the main crystalline components of six parotid gland calculi.

Although parotid calculi can occur at any age, they are rare in children, and the reasons for this are not fully understood.[8] Two hypotheses have been proposed regarding sialolithiasis in children: one suggests that children have lower salivary calcium concentrations compared to adults, while the other suggests that children have a higher rate of salivary flow than adults. The two most common bacterial pathogens associated with sialadenitis are Staphylococcus aureus and Streptococcus viridans.[9] Less frequently, sialadenitis is associated with a group of gram-negative bacilli collectively known as HACEK, which includes Haemophilus parainfluenzae, Haemophilus aphrophilus, Haemophilus paraphrophilus, Actinobacillus actinomycetemcomitans, Cardiobacterium hominis, Eikenella corrodens, and Kingella kingae. These bacteria together account for approximately 5 to 10% of all cases of infective endocarditis.[10] Parotid duct sialolithiasis has been documented in pediatric medical literature on several occasions.[11] Seifert et al. reviewed 1,200 cases of major salivary gland calculi in 1986 and found that the parotid was involved in fewer than 10% of cases, with a ratio of parenchymal to ductal occurrences of 1:35.[12] Fesharaki et al. published the first case report of a pediatric patient with intraparenchymal parotid calculi in 1979.[13] Parotid fistulas are exceedingly rare, particularly in the pediatric age group, and may be congenital or result from various causes such as trauma, surgical complications, infection, malignancy, and calculi.[14] There are no effective pharmacological options available to dissolve salivary calculi. Dilation of salivary ducts is typically employed for small and distally located calculi, while sialoadenectomy or surgical extirpation of the gland is performed for larger or more proximally located calculi. These procedures carry inherent risks, including bleeding, infection, Frey's syndrome, facial nerve injury, postoperative neurologic damage, sialoceles, salivary fistulae, and facial scarring; although the risks are minimal.[15]

Ultrasound-guided piezoelectric extracorporeal shockwave lithotripsy has been utilized for evaluating and treating salivary gland calculi. Educating patients about the underlying pathology and management strategies, particularly emphasizing the importance of staying hydrated and maintaining optimal oral hygiene, is crucial. These measures can help alleviate symptoms and reduce the risk of dental complications.[16]

2. Case Report

A 35 year old female patient came to the Department of Oral Medicine, Diagnosis and Radiology, PDM Dental College & Research Centre, Bahadurgarh, Haryana Pradesh with chief complaint of pain and swelling in the left cheek region since 3 months. Pain was dull and intermittent in nature with history of recurrent swelling during meals and subsided on its own after meals. Extra-oral examination was non-contributory at the time of examination. Intra-oral examination revealed a firm and tender swelling in left buccal mucosa at level of occlusal plane of maxillary first molar. When the gland was palpated, saliva could be seen at the duct orifice and the gland was tender. Intra oral periapical radiographic film was placed against left stenson's duct opening and examination revealed faint solitary ovoid radio-opaque fleck. A cone beam computed tomography scan was taken, which confirmed the presence of calculi (Figure 1). Based on these findings the patient was diagnosed with a sialolith in left stenson's duct. Surgical removal of the sialolith was planned and performed under local anesthesia (Figures 2 and 3). With antibiotic coverage under local anesthesia, incision was placed longitudinally along the margins and was placed at the ductal orifice and calculi was exposed and retrieved. The calculus was removed in total and was followed by a satisfactory healing process. The sialolith was of size.8 X.8cm (Figure 4).

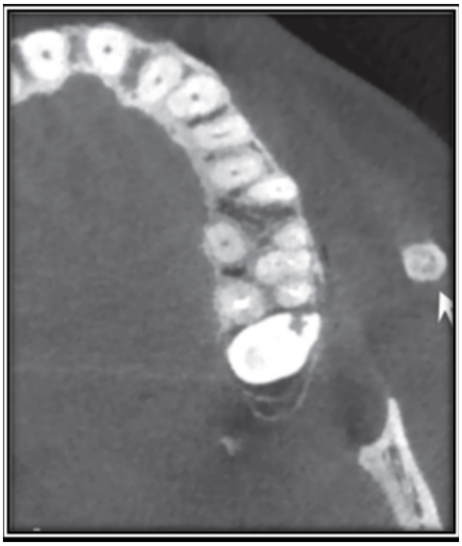

Figure 1: Cone beam computed tomography (Axial scan) depicting radiopaquesialolith in left parotid gland

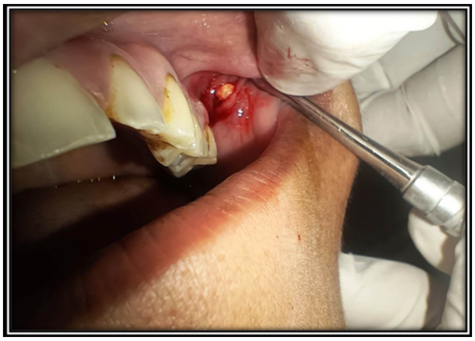

Figure 2: Surgical removal of calculi from left parotid gland

Figure 3: Excised specimen

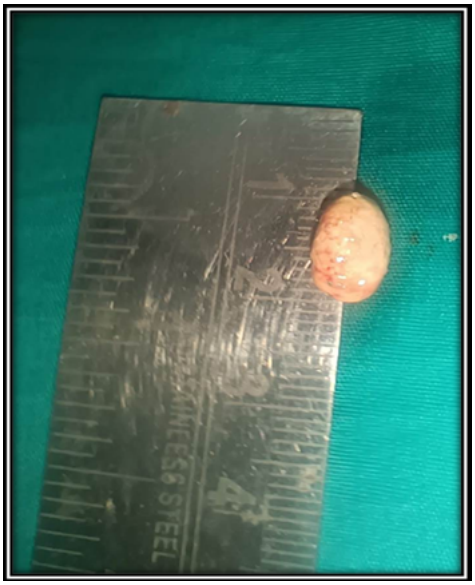

Figure 4: Ovoid shaped sialolith measuring .8X.8 cm

3. Discussion

The parotid gland, located superior to the angle of the mandible, lies superficially to the masseter muscle and drains through Stenson's duct opposite the maxillary second molar.[17] Sialolithiasis, predominantly observed in adults and rarely in children, is associated with various factors such as blockage of salivary flow, decreased saliva production, alterations in salivary pH due to sepsis in the oropharynx, and reduced solubility of crystalloids. Additionally, a retrograde theory suggests that the migration of food, bacteria, or foreign objects from the oral cavity into the duct system may contribute to calculi formation through subsequent calcification.[18] According to conventional theories, sialoliths develop in two stages: initially, a central core composed of salts bound with organic substances forms, followed by the accumulation of organic and inorganic material to form the outer layer.[19] Clinically, sialolithiasis can present acutely with symptoms such as pain, swelling, redness, and discharge from the duct, or as a cheek abscess accompanied by gland enlargement. Symptoms may worsen during eating. Calculi often lead to recurrent painful swellings or unresolved infections of the parotid gland, typically decreasing in size over time.[20] Interventional radiology procedures, commonly employed, utilize fluoroscopic techniques to visualize within the duct, facilitating direct extraction. A variety of investigative modalities are available for diagnosing parotid calculi, utilizing multimodal imaging to evaluate theses.[21] However, not all are radiopaque. Plain radiography can detect approximately 80-90% of submandibular (typically situated within the duct) and around 60% of parotid duct s (more commonly located within the gland itself), possibly due to variations in glandular secretion composition.[22] Oblique views are often necessary to visualize s distinct from surrounding bone and teeth. Computed tomography is excellent at visualizing calculi within the duct and the gland; however, its spatial resolution may not detect very small calculi.[23] Additionally, while computed tomography can assess the gland, it is not as effective as Magnetic Resonance Imaging.

In acute obstructive cases, the gland may appear enlarged, hyper dense, and show stranding and enhancement following contrast administration.In chronic cases, fatty atrophy will be evident, with reduced parenchymal volume replaced by fat. MRI can visualize larger calculi, map ductal anatomy, distinguish acute from chronic obstruction, and identify glands with incomplete obstruction. Calculi appear as low signal regions outlined by high signal saliva on T2-weighted images.[24] Computed Tomography scans are more sensitive than plain X-ray films in detecting calculi and can effectively visualize abscesses and ductal anatomy. However, the specificity of Computed Tomography scans alone in diagnosing focal inflammatory disease, with or without calculi, is approximately 75%.[25]

This specificity increases to 90% when combined with clinical and laboratory findings. Obtaining both contrast and non-contrast films is crucial to prevent misidentifying artefacts as calculi. While sialography effectively delineates ductal anatomy, it poses challenges in children and is contraindicated in acute settings. Magnetic Resonance Imaging, offering reduced radiation exposure, enables visualization of larger calculi, mapping of ductal anatomy, and assessment of gland condition.[26] It can differentiate between acute and chronic obstruction and identify glands with incomplete obstruction due to its superior tissue discrimination. Sialo-Magnetic Resonance Imaging, a recently introduced non-invasive diagnostic method, shows promise in evaluating salivary gland disease. It is highly effective in precisely outlining the size and position of calculi within salivary gland ducts.[27] Sialography is considered the gold standard investigation for salivary gland disorders, involving the injection of radio-opaque dye into the duct and obtaining serial radiographs. It can identify calculi and strictures, with smaller calculi sometimes flushed out during the procedure. It excels at delineating the exact size and location of s within salivary gland ducts and is particularly useful for delineating ductal anatomy.

However, it is difficult to perform in children and contraindicated in acute settings due to the risk of exacerbating infection.[28] Ultrasonography, on the other hand, can accurately identify ductal and highly mineralized calculi with a diameter of at least 1.5 mm, boasting a high accuracy rate of 99%. It can detect up to 90% of salivary duct calculi, presenting them as strongly hyperechoic lines or points with distal acoustic shadowing. However, it's worth noting that small calculi (< 1.5 mm) may not cast a shadow, and some radiolucent calculi can be visualized using ultrasound. It also capable of visualizing calculi that are radiolucent, which may not be detectable on X-ray films, as approximately 20-40% of is not radio-opaque.[29] Generally, there is minimal confusion because the clinical presentation is relatively specific. The differential diagnosis depends on the imaging modality used. For plain radiography and computed tomography scans, potential differentials include other calcific foci such as hemangiomas/phleboliths and atherosclerotic calcifications. This underscores the need for careful interpretation to distinguish between these entities and salivary gland calculi. Both contrast and non-contrast films should be obtained to avoid misidentifying artefacts as calculi. In the case of sialography, filling defects observed may have various explanations, including an injected bubble of air, tumor, or blood clot.[30] Conservative treatment options for salivary gland calculi typically involve non-invasive techniques before considering more invasive procedures. Hydration helps promote saliva flow, which can aid in flushing out smaller calculi. Hot compresses can help relax the duct and facilitate the movement of calculi. Sialagogues, substances that stimulate saliva production, can also help dislodge the calculi. Anti-inflammatories help reduce inflammation and pain associated with the condition. Antibiotics are necessary if there is an accompanying infection.[31] For small calculi, methods like applying moist warm heat, using sialogogues, and gland massage are often effective in dislodging calculi from the duct. Additionally, smaller calculi can sometimes be removed through the duct orifice using bimanual palpation, a technique where pressure is applied externally while manipulating the calculi internally. Flushing with normal saline under minimal pressure can push calculi out, which can be useful for distal duct calculi that have migrated proximally in a dilated duct.[32] Calculi less than 2 mm may pass out on their own in adults but rarely do so in children due to the smaller size of the ductal opening. Sialoendoscopy is typically reserved for calculi smaller than 5 mm, enabling direct visualization and extraction using endoscopic imaging and tools. It is a minimally invasive technique, initially introduced by Katz in 1991 and further developed by Marchal et al. and Nahlieli et al.[33] Numerous reports have discussed the use of sialendoscopic surgery for parotid gland sialolithiasis, focusing on sialolith removal.[34] However, in cases where conservative measures fail or if there are complications such as infection, simple sialolithotomy may be necessary. This procedure involves making a small incision in the duct to remove the calculi. It's crucial to monitor for complete resolution, as obstruction of the ductal opening can impede saliva flow and lead to further complications. According to Klein and Ardekian, sialoliths up to 4-5 mm in diameter are considered suitable for sialendoscopic removal, while dealing with sialoliths located deep within the hilum poses a challenge for surgeons.[35] Increasingly, non-surgical options exist to treat symptomatic, including extracorporeal sialolithotripsy, endoscopic removal, and endoluminal balloon dilatation and calculi extraction. Persistent debate surrounds the endoscopic approach, as there is no agreement on the largest diameter of s that can be extracted without fragmentation or whether impacted or hard can be effectively treated solely by endoscopy. Extracorporeal shockwave lithotripsy serves as an alternative for calculi in the proximal duct when transoral retrieval is not possible.

The typical use of this method involves the combination of sialendoscopy with extracorporeal lithotripsy utilizing a Thulium laser to fragment s sized between 4 and 8 mm. Nonetheless, interventional sialendoscopy, particularly in cases involving large or narrowed ducts, has faced setbacks even among experienced surgeons, with a failure rate of approximately 20%.[36] While alternative procedures such as extracorporeal shock wave lithotripsy and endoscopic calculi removal have been outlined, their effectiveness in pediatric cases is emphasized as requiring further investigation. Ktilkens et al. conducted extracorporeal shockwave lithotripsy on 42 patients with parotid calculi, noting that 5 out of 10 patients with intraparenchymal parotid calculi were free after treatment. The average follow-up duration for all patients in their study was 63 months.[37] Calculi smaller than 1.0 cm in diameter tend to exhibit improved responsiveness to extracorporeal Shock Wave Lithotripsy. Invasive modalities entail a transoral approach for distal duct calculi, involving exposure of the ductal opening or direct incision over the calculi for removal. The ductal opening may also be stitched to the surrounding mucosa to enhance gland drainage. Surgical interventions might be required for more intricate cases, such as a transoral approach for distal calculi or a transcervical approach for proximal calculi, particularly if the transoral method proves ineffective. General anesthesia is highlighted as preferable for certain procedures, especially those involving difficult intra-oral approaches or calculi located near critical structures like the hilum of the parotid duct. Alternatively, parotidectomy is considered for larger, proximal calculi associated with chronic inflammatory changes in the gland. Patients experiencing recurrent or persistent symptoms should be directed to specialized treatment. Excision of the salivary gland may be considered for patients with persistent symptoms unresponsive to conservative or medical management, or for those with large intraglandular calculi.[38]

4. Conclusion

The diagnosis and treatment of salivary gland calculi, emphasizing the importance of considering both non-invasive and invasive approaches based on the size and location of the calculi. It explores various surgical methods, such as transoral duct incision and external approaches, and underscores the role of endoscopy-assisted removal for larger or impacted calculi. While minimally invasive techniques have advanced, gland excision may be necessary in cases of symptomatic or recurrent sialadenitis due to multiple or large calculi, or when less invasive methods prove ineffective.

5. Source of Funding

None.

6. Conflict of Interest

None.

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