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Anterolateral Sialolithiasis in Wharton's Canal

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Case Report
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Abstract

Salivary glands are exocrine glands whose functions include the production of saliva to moisten the mouth and thus help and facilitate the digestive process. Sialolithiasis is an entity that can commonly present in these glands and is mainly due to the presence of intraglandular stones or within the salivary duct system. These sialoliths can grow abruptly and cause major obstructions and associated secondary complications. This study presents the case of a 48-year-old female patient who was referred to the Maxillofacial Surgery Department of Kitui Hospital in Kenya due to increased volume in the left anterolateral region of the floor of the mouth, which was associated with pain when consuming acidic, salty, or even very sweet foods, with a reported evolution time of 1 year. She also reported that sometimes the pain was so severe that she could observe a slight increase in volume in the submandibular region on that side, which gradually disappeared as the pain subsided. An occlusal X-ray was indicated, which confirmed the presence of a sialolith in Wharton's duct. Some laboratory studies were performed, and then the excision was carried out through an intraoral canalicular sialolectomy. The presentation of our case is part of a paradigm shift documented in recent literature, where the management of sialolithiasis has evolved from glandulectomy as the standard toward minimally invasive and organ-preserving strategies. This transformation, initiated in the 1990s, has reduced the rate of glandular resections from 40-50% to less than 5% in specialized centers.

Keywords: Salivary Glands, Sialolithiasis, Sialolectomy

Introduction

Salivary glands are part of the digestive glandular system. Among their basic functions is the synthesis of saliva deposited in the oral cavity, helping to maintain its moisture and facilitating all processes carried out there. They are generally classified into Major Salivary Glands (parotid, submandibular, and sublingual glands) and Minor Salivary Glands (distributed throughout the oropharyngeal mucosa, for example, in the labial, buccal, lingual, palatal, floor of the mouth, and oropharyngeal mucosa) [1].

Due to these characteristics, the presence of calculi or sialoliths becomes frequent. Sialolithiasis is characterized by the presence of calcified structures in intraglandular tissue or in their ducts that drain into the oral cavity. These calculi are formed by two parts: an inorganic body where minerals such as calcium phosphate, carbonate, magnesium, and ammonium can be found, and an organic matrix of amino acids and carbohydrates. Although several studies reveal that the exact mechanisms of calculus formation remain unknown [2,3].

It is a multifactorial entity, allowing the identification of several factors that may increase the risk of their formation, such as morpho-anatomical elements including saliva composition, crystallization, salivary duct stenosis, diverticula in salivary ducts, supersaturation with calcium and phosphorus among other elements. External factors

such as direct trauma to the ducts, infections, among others, are also involved [3]. Diagnosis is primarily made through anamnesis and physical examination. Sialolithiasis generally causes pain and swelling of the involved glandular tissue due to salivary flow obstruction, which if completely occluded could cause constant pain, swelling of the area, and even the patient could present signs and symptoms of a systemic infection. In cases where the sialolith is located within the gland, it may be asymptomatic in some patients [4].

Different imaging modalities can be used to evaluate clinically obstructed salivary glands, including plain radiography, computed tomography, conventional or digital sialography, computed sialography, magnetic resonance sialography, and ultrasound [5-8]. Sometimes it is necessary to differentiate these entities from others that can affect the oral cavity, such as those associated with soft tissues like dental or non-dental infectious processes, mucoceles, ranulas, among others [9]. Regarding the treatment of sialolithiasis, options are varied and depend on the condition of the gland, the associated infectious process, and the location of the calculus. Therefore, removal of the sialolith would be a priority, whether directly through dilatations of the excretory duct using inflatable probes for spontaneous expulsion, intraoral canalicular sialolectomies, extracorporeal lithotripsy to fragment calculi smaller than 2 mm in diameter, endoscopic removal, and even in cases associated with acute or chronic infections and intraglandular locations, gland removal is a viable option [10-12].

The aim of this case is to demonstrate the importance of gradual oral cavity examination and detailed physical examination by healthcare personnel for each patient, in order to detect any abnormality or developing process early and improve our patients' quality of life.

Case Presentation

A 48-year-old female patient, of black race, from Kitui, Kenya, with a history of hypertension, presented to our service accompanied by her relatives reporting discomfort and pain in the floor of the mouth, associated with meal times and more specifically with acidic, salty, bitter, and very sweet foods. She reported that upon contact with these foods, severe pain first appeared in that area, but after a few minutes it gradually decreased, sometimes even experiencing a sensation of increased salivary secretion. Upon questioning, the patient denied the presence of toxic habits, allergies, oral cavity trauma, or any other relevant data, stating that the symptoms and signs began approximately 1 year ago.

The extraoral physical examination provided no relevant findings. The intraoral physical examination revealed a significant increase in volume in the left anterolateral region of the floor of the mouth, specifically along the course of Wharton's duct of the left submandibular gland, with a hard and rough consistency on palpation (Figure 1).

Figure 1: Intraoral volume increase in the left anterolateral region of the floor of the mouth, affecting Wharton's duct.

Complementary blood studies were performed, with results within normal values. A neck ultrasound was ordered, more specifically of the salivary glands, to rule out any irregularities regarding size, echogenicity, and other elements, which provided no relevant findings. Regarding radiographic studies, the lateral occlusal X-ray showed a radiopaque image, calcified and tubular in appearance, large, tortuous, located in the affected duct (Figure 2).

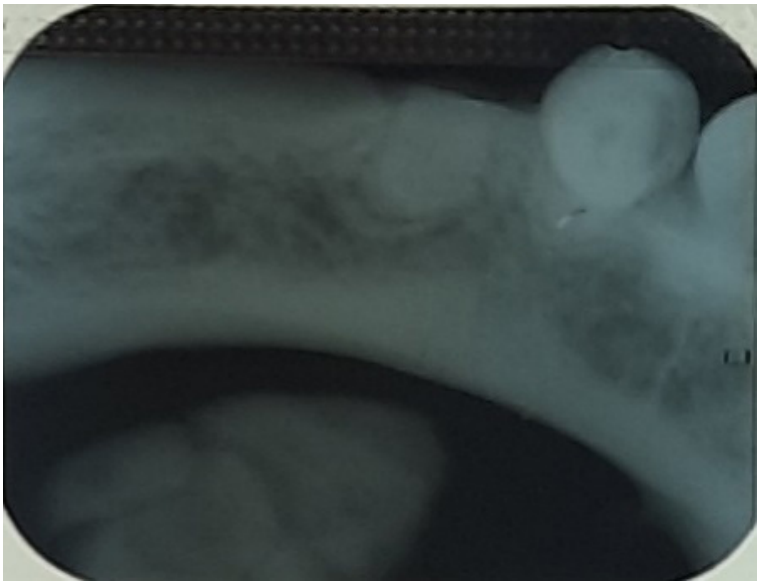

Figure 2: Radiopaque image, tubular in appearance, in the floor of the mouth, lateral to the mandibular body, located in Wharton's duct.

As surgical treatment, an intraoral canalicular sialolectomy was performed to expose the calculus and facilitate its excision. A large calculus with the previously described characteristics was observed (Figure 3).

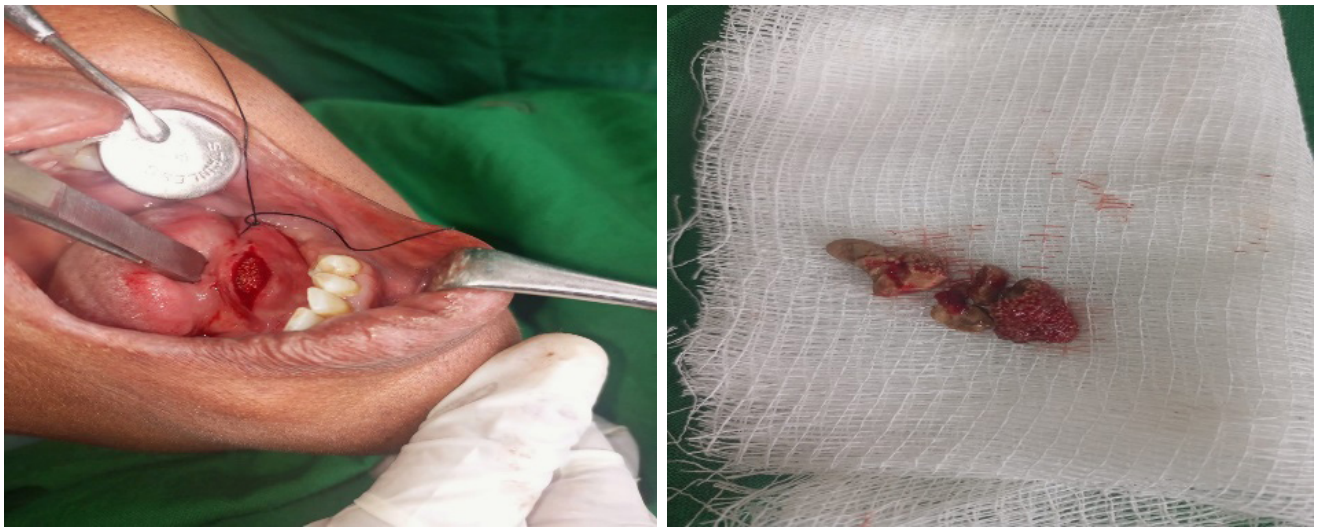

Figure 3: Intraoral canalicular sialolectomy. Extraoral image of the sialolith.

The patient had a favorable evolution, with scheduled follow-up appointments, voluntarily giving her consent for the publication of her case for medical and scientific purposes. This is an interesting case from a medical standpoint due to the size and shape of the calculus, in addition to the tortuous duct of the involved gland, which made spontaneous expulsion almost impossible.

Discussion

Sialolithiasis is one of the most common diseases of the salivary glands, with the submandibular gland being the most frequently involved for various reasons, including that its saliva is more seromucous, due to its alkaline nature and richness in mucin, its drainage occurs in an ascending

direction, against gravity, and Wharton's duct is longer and more tortuous, with its main portion being wider than its orifice [4,10,21,23].

Sialoliths are generally formed by two types of components: an organic group and an inorganic group. Among the organic constituents we can find saliva products (glycoproteins and mucopolysaccharides), cellular detritus, and lipids. The main inorganic component is carbonate-apatite and not hydroxyapatite, accompanied by other calcium salts, in addition to other different elements such as phosphate, magnesium, iron, copper, and zinc. They can have different shapes and sizes, with a smooth or irregular surface, and regarding color, they can be yellow or brownish [9,13,16].

Regarding their etiopathogenesis, it can be included that changes in pH, dehydration, reduced salivary flow rate, ductal abnormalities, and retrograde migration of food, bacteria, or foreign bodies from the oral cavity can favor calculus formation. Other authors suggest that it may also be due to trauma, which possibly hinders salivary flow and causes stasis, bacterial, viral, fungal infections, or local irritations. It is estimated to generally affect the adult population, with a dominant prevalence in men [3,4].

Among the most frequent clinical manifestations are swelling and colicky pain [1-3]. This is also known as "meal-time syndrome," which is associated with increased salivation, since the dilated obstructed duct generates intraglandular pressure and pain episodes occur that can be transient, mild, or more severe. In other cases, the calculus lodges in the duct lumen but does not completely obstruct it, allowing some salivary secretion to pass, so it may be asymptomatic [5]. Long-standing obstruction can cause fibrosis and atrophy of the involved gland, with subsequent loss of its secretory function [4,5].

In the presented case, immediate pain followed by gradual relief as saliva flowed through the duct affected by the calculus were evident signs and symptoms. The diagnosis of sialolithiasis is made through anamnesis and a thorough clinical examination that includes palpation of the affected area. In the case of the submandibular gland, if it is located in the duct, it can be felt when examining the floor of the mouth, and if it is intraglandular, it may be indurated and firm, which would guide us to think of an infectious or chronic process of the gland [2,4,5].

Radiographically, sialoliths appear as homogeneous radiopaque masses with defined borders and radiopacity similar to dental structures, so they can be confused with one of these. For detailed evaluation, different radiological studies can be used, such as partial or total occlusal radiographs as the most indicated, lateral skull radiographs. Among the diagnostic exams, the following stand out: sialography, ultrasound, computed tomography, ultrasonography, magnetic resonance imaging. Currently, among the most used treatments, sialoendoscopy stands out, representing a minimally invasive, outpatient procedure that allows treating and diagnosing pathologies such as sialolithiasis, representing an important alternative [5-8]. Prognoses are usually favorable, as in the presented case, but sometimes they can trigger infectious processes such as recurrent sialadenitis, Ludwig's angina, among others [14,18,19,22,25,36].

There are different treatment options for this entity, ranging from pharmacological to surgical, depending on the individual characteristics of each patient and the severity of the process [2,3]. Among the options are continuous dilatations of the excretory duct using small inflatable balloon probes to try to expel the calculus spontaneously. Other procedures can also be performed such as intraoral canalicular sialolectomy, where an incision is made in the anterior portion of the excretory duct to extract the sialolith, and in other cases of deep or intraglandular lithiasis associated with inflammation and recurrent infection processes of the affected gland, it should be removed via an external approach [10-12].

Many researchers have based their studies on sialendoscopy, such as Lee SH, et al. [20], Strychowsky JE, et al. [7] who conducted a meta-analysis demonstrating that sialendoscopy has a success rate of 80.9% and significantly reduces the need for glandulectomy. Modest MD, et al. [6] documented that with the learning curve, sialendoscopy achieves glandular preservation in >95% of cases. Goodstein L, et al. [8], who describe sialendoscopy as a novel technique even for the sublingual gland, demonstrating its versatility [17,20,27-29,36].

Sialendoscopy has been consolidated as a safe and effective technique. A systematic review and meta-analysis that included 91 studies and 8,218 patients reported an overall success rate of 80.9%, reaching 88.3% specifically in the submandibular gland [15,24,26,32]. Our traditional transoral approach aligns with these figures, although recent studies have identified key predictive factors for the success of the endoscopic procedure.

Takahara, et al. in a series of 144 patients, demonstrated that the success rate of sialendoscopy alone was 40%, establishing that the location, size, and mobility of the calculus are independent predictors. Specifically, it was identified that a major axis <7.5 mm constitutes a measurable standard for successful extraction, while parenchymal calculi are associated with prolonged operative times, greater

complications, and higher risk of fragment retention. This finding is relevant to our case, as it suggests that proper patient selection based on the size and location of the calculus is fundamental for therapeutic success [32-34].

Regarding the combined approach as an emerging standard, the most recent literature emphasizes that sialendoscopy assisted by transoral approaches has become the standard of care for larger submandibular calculi, especially those >8 mm. The therapeutic arsenal has been expanded with complementary techniques. Intracorporeal lithotripsy with Ho: YAG laser or pneumatic lithotripsy has demonstrated success rates exceeding 90% in complex calculi, with no significant differences between both modalities. Likewise, transoral robotic surgery (TORS) emerges as a promising alternative for hilar and intraparenchymal calculi [30,31,33].

Muthunagai R, et al. [21] reported a 30 mm calculus (giant), managed with a transoral approach but emphasizing that >15 mm are infrequent. Raghuvanshi P, et al. [22] described a 42 mm calculus with sialo-oral fistula, requiring open surgery. Sarma M, et al. [23] reported on an 11x8 mm calculus (similar to ours), extracted transorally.

In this context, our case, successfully managed through a conventional transoral approach, constitutes a valid option, although the reviewed evidence suggests that the incorporation of sialendoscopy—whether as a single or combined procedure—offers advantages in terms of precision, glandular preservation, and fewer complications, consolidating itself as the current standard in centers with the availability of equipment and necessary experience.

Conclusions

Sialolithiasis is an entity that must be taken seriously, as it can cause major affections of the glandular system, which would bring a number of organic imbalances that would ultimately affect the digestive process, the oral cavity, and even at a systemic level, since the associated infectious processes could destabilize the patient's general condition.

Health promotion and disease prevention should always be directed at everyone, in order to detect any of these entities early and treat them with the least possible risk and without affecting the health and lifestyle of our patients.

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