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Case Report Article

Giant sialolith of submandibular gland: case report

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Abstract

Introduction: The sialolith, also known as saliva stone or sialolithiasis is a calcified structure which develops inside the salivary ductal system or on the salivary gland parenchyma; it grows gradual, asymptomatic, and slowly. Most of the stones are sized less than 10 mm and only 7% of them are larger than 15 millimeters; those are considered giant salivary gland stones. **Objective:** This study aimed to report a case of two sialoliths that have merged, forming a giant sialolith, located in the Wharton duct in the left submandibular gland. **Methods and Results:** Clinical diagnosis was confirmed by occlusal and panoramic radiographs. A surgical removal was performed with intraoral incision under local anesthesia, through sialolith anchorage by suture thread and removal of two sialoliths, sized about 13 mm and 16 mm. Marsupialization suture was performed at the local where the incision was made, forming a new gland duct. **Conclusion:** The sialolithiasis treatment methods are very different and should be taken into account the affected gland, size and location of sialolith, always opting for the more conservative methods.

Introduction

The sialolith, also known as salivary stone or sialolithiasis is a calcified structure developing inside the salivary duct system or parenchyma with gradual, slowly, and asymptomatic growth [3, 7]. Sialoliths are common in patients at 30 and 40 years occurring more in males than females, but not in children [2, 4]. Sometimes, sialolith are asymptomatic generally when the obstruction is incomplete, so that the saliva surpasses the stone and is eliminated [2]. On the other hand, the complete obstruction is associated with swallowing, fever, pain, gland infection and drainage of purulent exudate via the duct orifice, characterizing the sialodenitis. The gland volume may increase during meals due to great salivary secretion [3, 4, 7]. Long-time obstructions result in infections leading to gland atrophy with alterations in salivary secretion function and gland fibrosis [7].

The etiology of salivary stone is still unknown. One hypothesis is the anatomical irregularities of the gland conduct associated with a change in salivary secretion and composition resulting in increasing the saliva viscosity and obstruction of the terminal ducts of the glands. This obstruction is caused by the deposition of calcium salt around an organic matrix constituted of foreign bodies, bacteria, and epithelial cells [1, 3, 7]. Not only the increase of the saliva alkalinity, but also the salivary stagnation, the presence of infection/inflammation, physical trauma on the gland duct are predisposing factors to the stone formation [4].

The salivary stone size may vary from below 1 mm to few centimeters of diameter. Most of the stones measure less than 10 mm, and only 7% are greater than 15 mm, which are classified as giant salivary stones, little reported in the literature [1, 4, 7]; sialoliths grow by deposition, at an estimated rate from 1 to 1.5 mm per year [4].

The correct diagnosis is important involving clinical examination, inspection, palpation, gland manipulation (to verify the amount of saliva secreted), imaging examination as occlusal/panoramic radiographs, lateral cephalograms, lateral oblique mandible radiograph. Radiographically, salivary stones are radiopaque images in the gland and conduct areas. Other valuable imaging examinations are computed tomography, ultrasound, magnetic resonance imaging, sialography, and duct endoscopy. Alternative methods such as lithotripsy, xeroradiography, and endoscopy have been suggested and used for selected cases [4, 6]. Each one of the methods has an indication depending on the affected gland and stone size [2]. The sialography is indicated in cases of signs of sialodenitis, radiolucent stones, submandibular stones, deep parotid stones, and contraindicated in cases of infection, patients allergic to the radiograph contrast solution. In the endoscopy, the location of the gland alteration is important because when isolated, it suggests chronic inflammatory disorder; when involving multiple glands, it suggests Sjögren syndrome, endocrine disorders, metabolic alterations, and nutritional deficiency. When the sialolith are palpable in the gland parenchyma at preauricular or submandibular area, it is necessary to differ from regional lymph nodes presenting granulomatosis or hematopoietic alterations [4, 7].

The sialolithiasis account for 30% of the salivary gland pathologies, affecting more commonly the submandibular gland (83 to 94%), followed by parotid (4 to 10%) and sublingual (1 to 7%). The high incidence of submandibular stone is explained by the pH, the amount of mucin and high concentration of calcium in saliva associated with the long, tortuous, and ascending path and length of the Wharton duct of the submandibular gland and thicker mucoid secretion against gravity [1, 3, 7]. The sialolith may also form inside small salivary glands more frequently in upper lip or mucosa jugal [2, 3].

Clinically, these stones may be fusiform, round, or ovoid, soft or hard, yellowish, depending on the constitution; generally single. Generally, antibiotics, analgesics, and antipyretics are prescribed. Multiple or bilateral sialolith are uncommon [3].

The adequate treatment will depend on the affected gland, stone size and location. Generally, conservative techniques are recommended for small sialoliths, such as patient's hydration, application of internal and external heat, milking, massage of the gland with acid fruit (lemon), and removal of small stones close to the duct orifice through dilating with the aid of catheter. More invasive techniques, such as surgical removal via intraoral access are indicated for greater salivary stones and/or those located in the gland parenchyma, mainly when the stone is at the distal third of the gland, and it can be easily touched. The affected duct is exposed through an incision on the mouth's floor which is easily performed and little associated with complications. Thus, the stone is seen and removed. The duct is sutured to the oral mucosa, and left open for proper drainage [2, 7]. Also, tissue transfixation or suture thread anchorage involving the duct to promote the obstruction prevents the sialolith displacement towards the gland [1].

This study aimed to report a clinical case of giant sialolith located in the Wharton duct of the submandibular gland treated through surgical removal.

Clinical case

Patient, male, melanoderm, aged 50 years was referred to the Center of Dental Specialties in the city of Cruzeiro do Oeste, Paraná (Brazil), with chief complaint of confection a total denture. During anamnesis, the patient reported no systemic alteration. At the intraoral examination, the presence of a foreign body was noted on the left side of the mouth's floor, in the area of the submandibular gland duct with decreasing of saliva during milking, but without the presence of purulent secretion. During palpation, a hard, mobile submucosal nodule in the area of the left Wharton duct of the submandibular gland was noted. The radiograph examination (occlusal and panoramic radiographs) showed two radiopaque long masses in the area of the submandibular duct resembling the canine tooth shape (figure 1). When asking about the nodule, the patient reported that he already noted the nodule, as the years went by. However, he did not know to specify exactly how many years and the nodule never disturbed him. He also did not report any discomfort, history of infection, purulent drainage, and fever. Based on the history and radiographic examinations, the diagnosis was sialolithiasis.

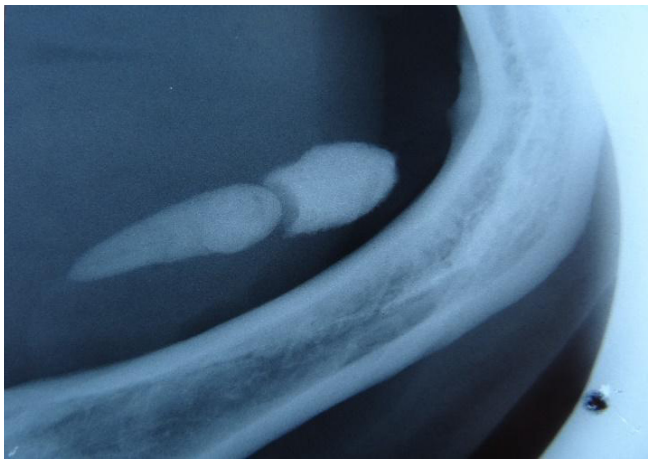

Figure 1 – Occlusal radiograph evidencing the radiopaque mass, resembling the canine tooth shape

The proposed treatment was the surgical removal through intraoral access under local anesthesia. The sialolith anchorage was accomplished through suture thread aiming to favor the incision and prevent its displacement towards the gland. A small incision followed by the divulsion of the tissues was performed by preserving the sublingual caruncle area (figure 2). The duct was located and dissected up to remove the giant stone completely (figures 3 and 4). Because of the sialolith size and duct destruction, the duct epithelium was sutured to the mouth's floor epithelium (marsupialization), aiming to guide the formation of a new site for salivary drainage. We prescribed amoxicillin 500 mg and analgesics for pain control (dipyrone 500 mg/ml) postoperatively.

Figure 2 – Sialoliths anchorage with suture thread and incision

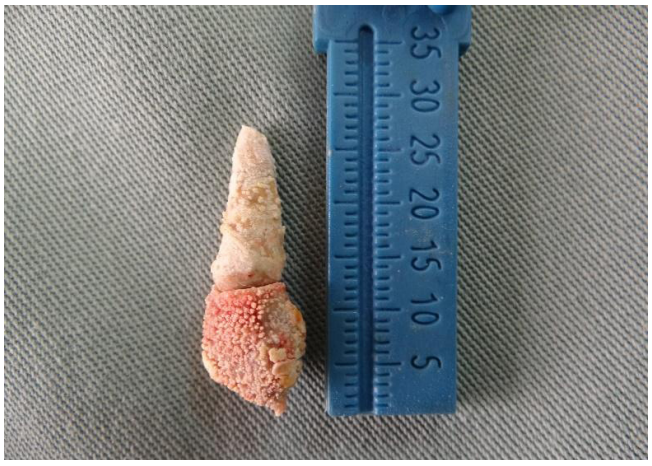

Figure 3 – Sialolith removed and fitted one to the other

Figure 4 – Sialoliths separated

Discussion

The sialolithiasis affects approximately 1.2% of the population, mostly in males, but regardless of breed. It mainly affects mid-age individuals, although young and the elderly are affected, but rarely children. The sialoliths are normally unilateral and they can be single or multiple. A single sialolith occurs in 75.3% of the cases; two sialoliths occur in 15.6%, three sialoliths occur in 2.9%; and four to eight sialoliths occur in 6.2% of the cases [5]. This case report described the presence of two embedded sialoliths in a male patient aged 50 years, without history of trauma, infection, or inflammation.

The sialolithiasis diagnosis is the result of a comprehensive history of the patient, typical symptoms, and findings of the clinical and imaging examinations. The radiograph is of paramount importance in diagnosing by achieving a success rate of 90% of the cases [1]. Considering the radiographic incidences, periapical and panoramic radiographs are contraindicated in these cases due to bone superimposition resulting in false diagnosis of intraosseous lesion, while a simple occlusal radiograph can show the stone located in the mouth's floor. The panoramic, cranium posterior-anterior, mandible lateral oblique radiographs are indicated in cases of parotid glands together with tangential radiograph for soft tissues [9, 10].

The giant stones are extremely rare. Ledesma-Montes *et al*. [8] reviewed 16 cases with stones measuring between 3.5 and 7 cm, and found 15 cases that affected the submandibular gland and one affecting the parotid gland. The cases affecting the submandibular gland were most frequently located in the duct.

The sialolith size may determine the symptomatology. Sometimes, the sialolith is asymptomatic, generally when the obstruction is incomplete and the saliva can surpass the stone and be eliminated. When the sialoliths are small, they enable the normal salivary flow without signs and symptoms. Greater sialoliths can cause the sudden increase of the glands, mainly during meals, followed by tension and pain that progressively reduce with salivary flow [7, 9]. In this case, despite of the sialolith size, the duct was not obstructed, and according to the patient, without pain symptomatology.

The salivary stones grow by deposition, at an estimated rate of 1 a 1.5 mm per year [4]. In this present case, the sialoliths were fitted to each other, one measuring 13 mm and the other measuring 16 mm, resulting in 27 mm.

The proper treatment will depend on the affected gland, size and site of the stone. Generally, conservative techniques are recommended for small sialoliths, while the surgical removal is indicated for greater ones [2, 3, 7]. Corroborating with the recommended treatment for this sialolith type, surgical removal was performed via intraoral access because more conservative treatments would be ineffective for this type of giant sialolith.

The tissue transfixation or sialolith anchorage with the suture thread involving the duct can be performed to promote the obstruction, preventing the sialolith displacement towards the gland [1]. In this present case, we opted by performing the anchorage with the suture thread on the sialolith, but not on the duct, aiming to prevent its displacement and provide a more accurate incision on mucosa searching the minimum dilaceration of the duct epithelium in the case of giant.

After removing the stone, the duct should be sutured to oral mucosa, leaving open for proper drainage [7]. Aiming to guide the formation of a new salivary drainage and avoid that the duct collabates, the duct epithelium was sutured to the epithelium of the mouth's floor (marsupialization) [1, 7].

If the gland is infected, antibiotics should be prescribed together with the treatment. In some situations with purulent drainage, swallowing and systemic signs of infection, first the infection should be proper treated and then the gland should be removed to eliminate the stone [7]. Aiming to avoid infection and allow proper postoperative period and healing, considering the patient's age, after the surgical procedure, we prescribed antibiotics (Amoxicillin 500 mg) and analgesics (Dipyrone 500 mg).

Conclusion

- Many imaging methods exist to diagnosis the sialolithiasis and the occlusal and panoramic radiographs are the most accessible ones;
- The occlusal examination is the most suitable for sialoliths located in the submandibular gland because of the incidence;
- The dentist should know the methods for the early diagnosis that enables the simplified treatment and the most favorable prognosis;
- The available methods for treatment sialolithiasis depend on the affected gland, size/site of the stones. However, whenever possible, one should opt for the most conservative method or that most suitable to the specific situation of the patient;
- The previous anchorage with suture thread enables a defined and minimum incision favoring the minimally invasive removal and preventing the sialoliths displacement towards the gland.

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