


## Clinicopathological study of sialolithiasis: a retrospective analysis of 11 cases

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### Abstract:

Sialolithiasis is one of the most common benign conditions that affect the salivary glands. The disease is characterized by obstruction of the gland parenchyma or ductal system that reduces the salivary flow rate. Most sialoliths occur in the major salivary glands and usually cause acute and chronic infections. Since an inaccurate clinical diagnosis of sialolithiasis is often reported, the aim of this study was to describe the clinical and histopathological features of 11 cases of sialolithiasis over a period of 13 years retrieved from the Oral Pathology Service of the Federal University of Bahia, Brazil. Most sialolithiasis cases involved the major salivary glands and were identified between the 4<sup>th</sup> and 5<sup>th</sup> decade of life. There was a slight female predominance. The lesions were usually symptomatic and appeared as a single nodule of firm consistency. The most common location was the submandibular gland, followed by the minor salivary glands (oral mucosa). Surgical excision was performed in all cases. Histopathology revealed a lamellar pattern of calcification of the sialoliths, chronic inflammation, and ductal squamous metaplasia.

**Keywords:** Salivary gland calculi, Submandibular gland, Pathology, Diagnosis

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Article received on December 10, 2018.  
Article accepted on February 14, 2019.

DOI: 10.5935/2525-5711.20190006



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## INTRODUCTION

Sialolithiasis is one of the most common benign conditions that affect the salivary glands. It is caused by the formation of sialolith/calculus within their parenchyma or ductal system. Sialoliths are composed of several ratios of organic and inorganic substances being their mechanism of formation incompletely understood<sup>1,2</sup>. Some authors have showed that sialolithiasis is more frequently located in major salivary glands, particularly in the submandibular and parotid glands<sup>1,2</sup>. This seems to be due to the long and tortuous path of the major duct and also the nature and consistency of the submandibular gland saliva, which is thicker in consistency, rich in phosphorous and has a high pH that is conducive of sialolith formation<sup>3</sup>. There is a higher predominance of sialolithiasis in middle-aged patients being male population slightly more affected on their minor salivary glands<sup>2,4</sup> whilst female subjects demonstrated higher incidence on major salivary glands<sup>5,6</sup>.

Although lesions are usually asymptomatic, patients with sialolithiasis typically may experience post-prandial salivary pain and swelling caused by obstruction of the physiological salivary flow<sup>7</sup>. Histologically, the sialolith usually has a lamellar structure which displays alternating eosinophilic and basophilic layers disposed either concentrically or in a globular shape calcified zone<sup>8</sup>. Depending on the calcification degree of the sialolith, conventional radiographs can show small radiopacity which might support a more accurate diagnosis along with clinical and histopathological features of sialolithiasis<sup>8,9</sup>. This condition may also result in a painful chronic inflammation of the salivary ducts (sialodochitis) and/or parenchyma (sialodentitis)<sup>10</sup> which may requires, if not spontaneously resolved, surgical approachment either by direct incision (sialodochotomy) or endoscopic removal of the sialolith (sialoendoscopy) or lastly by excision of the salivary gland being its recurrence not expected<sup>2,10</sup>.

Thus, the aim of this study was to describe a serie of 11 cases of sialolithiasis in major and minor salivary glands with an emphasis on their clinicopathological features.

## MATERIALS AND METHODS

After approval of the study by the Ethics Committee of the Federal University of Bahia, 11 histopathological records comprising the period from 2002 to 2015 and containing the term sialolithiasis were retrieved from the archives of the Oral Pathology Service, School of

Dentistry, Federal University of Bahia) and analyzed retrospectively. The data were checked for clinical information of the lesions (location, signs, symptoms, size, and clinical diagnosis). According to the clinical records, the lesions were surgically removed by excision. All tissue samples were fixed in formalin, routinely processed and embedded in paraffin, and the blocks were cut into 4- $\mu$ m thick sections and stained with hematoxylin and eosin. The slides were revised by one experienced pathologist who recorded the histopathological features of each lesion (structure and number of sialoliths, presence of sialolith inside the salivary duct or parenchyma, ductal metaplasia, and presence of chronic inflammation).

## RESULTS

The sample consisted of 11 cases. Women were more frequently affected ( $n = 7$ ) than men ( $n = 4$ ). The age ranged from 11 to 80 years, with a mean age of 41.3 years. Most cases were identified between the 4<sup>th</sup> and 5<sup>th</sup> decade of life. One of the 11 patients was 11 years old and three were  $\geq 60$  years. The duration of sialolithiasis was available in three cases and ranged from 2 months to 8 years. The diameter of the sialoliths ranged from 0.03 to 4 cm (mean 0.83 cm). The most common location was the submandibular gland ( $n = 8$ ), followed by the oral mucosa ( $n = 2$ ) and upper lip ( $n = 1$ ). Information about the shape of the lesion was available in five cases and consistency was described in seven cases. The lesions appeared as a nodule in three cases and had an oval shape in two. A firm consistency of the lesions was reported in seven cases and fibrous in one case. The clinical findings are summarized in Table 1.

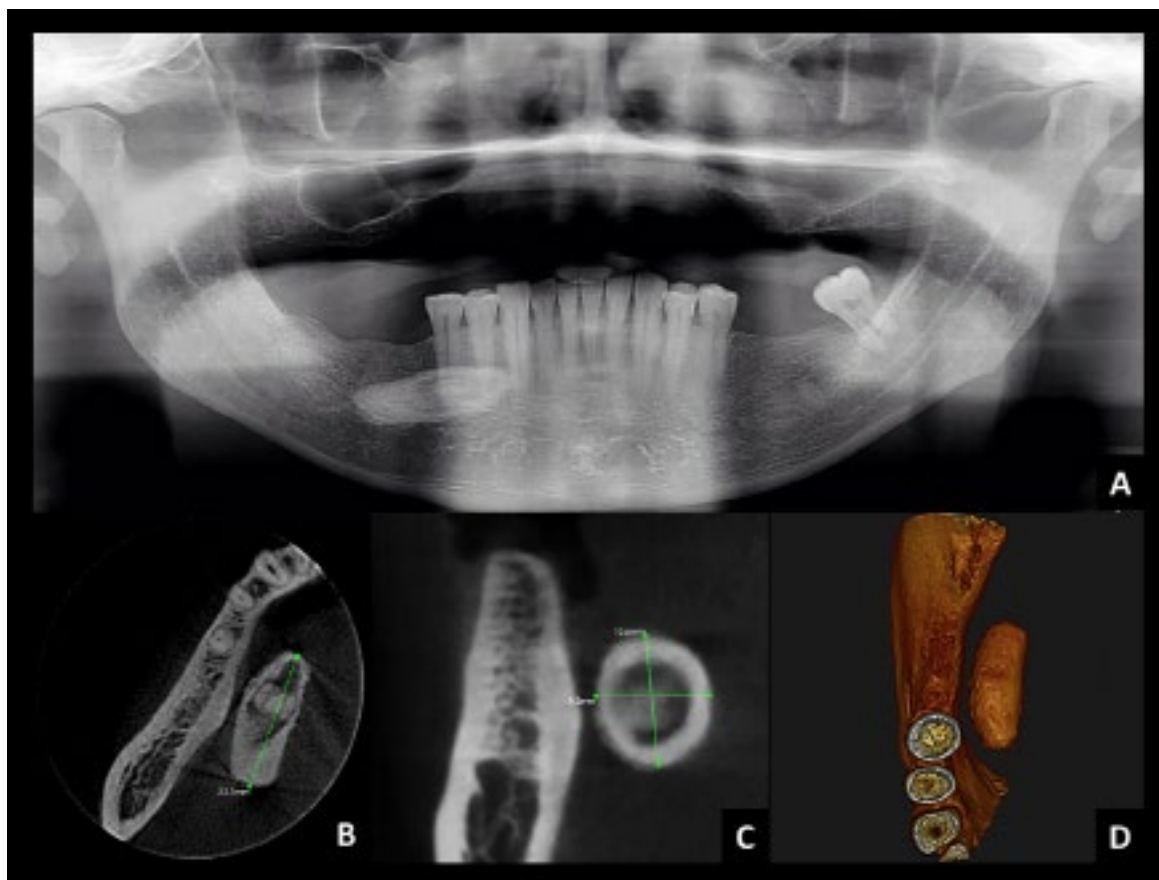
With respect to symptoms, two clinical charts documented painful lesions. Purulent discharge was reported in two cases. The most frequent clinical diagnoses were sialolithiasis (11/11; 100%) and chronic sialadenitis (1/11; 9%) (Table 1).

Imaging exams were available for five patients and the characteristics of the sialolith were evaluated. Panoramic radiography generally revealed a radiopaque oval or elongated nodule, mainly located on the inner side of the mandible (Figure 1A). Additionally, in a few cases, 3D reconstruction of cone beam computed tomography (CBCT) showed a sialolith of heterogeneous, almost totally hyperattenuating appearance, but with a hypoattenuating central zone in the mandible (Figure 1B-D).

Histopathological specimens were available in all 11 cases. The number of salivary calculi was as follows: five cases (1 calculus), two cases (2 calculi), one case (3

**Table 1.** Summary of the clinical findings for sialolithiasis

Cases	Gender	Age	Clinical form	Location	Duration	Size (cm)	Clinical Consistency	Pain	Purulent drainage	Clinical diagnosis
1	F	39	n/a	Upper lip	Unknown	1.1	n/a	n/a	yes	Sialolithiasis
2	F	63	Nodular	Submandibular gland	Unknown	4.0	Firm	yes	n/a	Sialolithiasis
3	M	63	n/a	Buccal mucosa	4 months	0.03	Firm	n/a	n/a	Sialolithiasis
4	F	11	Nodular	Submandibular gland	2 months	0.6	Firm	no	n/a	Sialolithiasis
5	F	24	n/a	Submandibular gland	8 years	2.5	Firm	yes	n/a	Sialolithiasis
6	F	n/a	n/a	Submandibular gland	Unknown	1.2	n/a	n/a	n/a	Sialolithiasis
7	F	80	Oval	Buccal mucosa	Unknown	0.7	Fibrous	n/a	n/a	Sialolithiasis
8	M	34	n/a	Submandibular gland	Unknown	0.07	Firm	n/a	n/a	Sialolithiasis
9	M	42	Nodule	Submandibular gland	Unknown	0.2	n/a	n/a	n/a	Sialolithiasis
10	F	37	n/a	Submandibular gland	Unknown	0.15	Firm	n/a	n/a	Sialolithiasis
11	M	41	Oval	Submandibular gland	Unknown	0.23	Firm	n/a	n/a	Sialolithiasis Chronic sialo-denitis



**Figure 1.** Elongated sialolith superimposed on the premolars and projecting backward (a). Axial, coronal and 3D CBCT images showing a hyper-/hypoattenuating nodule located in the floor of the mouth, measuring approximately 2.3 cm in length and 1.0 cm in diameter (b, c), with the same orientation as the submandibular gland duct (d).

calculi), and one case (4 or more calculi) (Figure 2A). In 10 cases, the salivary calculus displayed laminated layers of organic and inorganic substances (lamellar and concentric patterns) (Figure 2B). No laminated appearance was observed in one case. The ducts of the salivary glands were present in four cases. The ductal epithelial cells exhibited alterations compatible with squamous metaplasia in four cases (Figure 2C). A discrete and moderate chronic inflammatory infiltrate was found in two and three cases, respectively (Figure 2C). In one case, a sialolith was observed in chronic sclerosing sialadenitis (Küttner's tumor) located in a submandibular gland.

## DISCUSSION

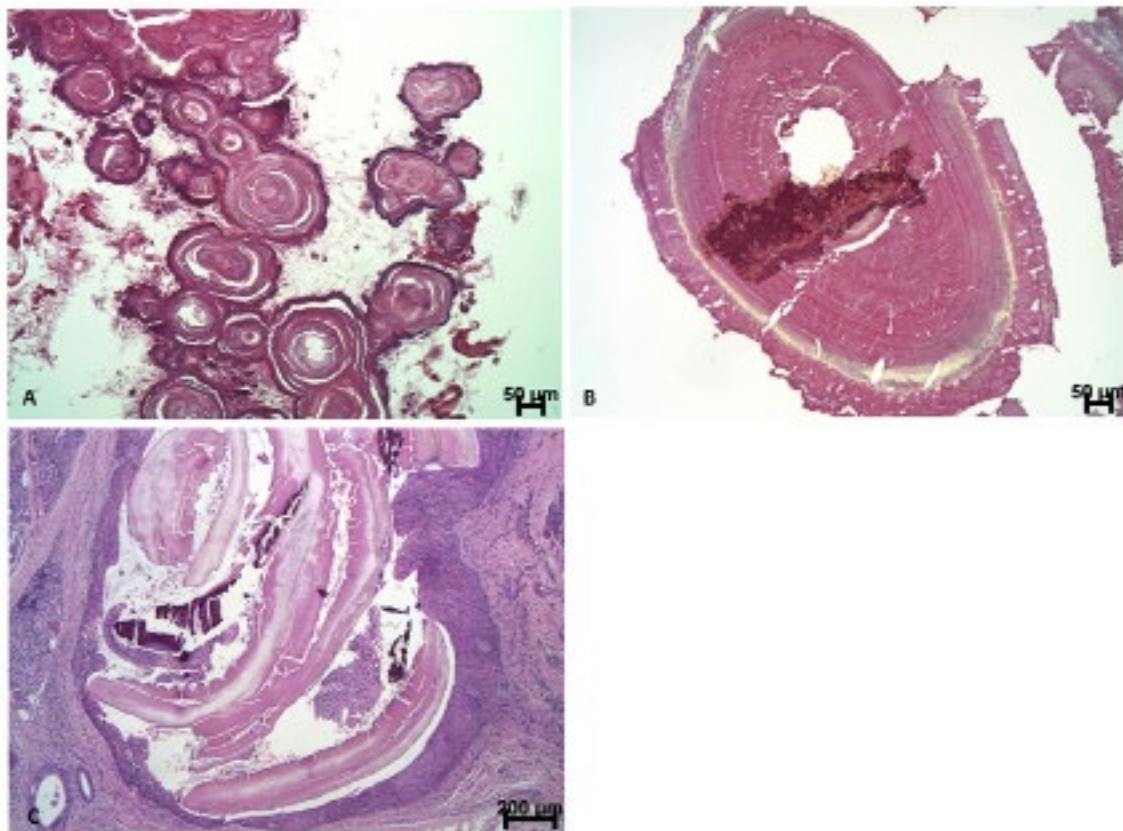
The current study investigated the clinico-pathological aspects of sialolithiasis of salivary glands diagnosed over a period of 13 years. The small number of cases reviewed might suggest that it is a rare disease.

Overall, in the current study, a slight female predominance was observed. However, previous studies have reported almost equal gender distribution<sup>11</sup>, a slight male

predominance<sup>4</sup> or female predilection<sup>8,12</sup> for sialolithiasis in minor salivary glands. For major salivary glands, female subjects demonstrated higher frequency<sup>5,6</sup>. The majority of patients were middle-aged with a mean age of 41.3 years which corroborates previous reports<sup>4,8,11</sup>. Sialolithiasis is rarely observed in infants, adolescents<sup>13</sup> and in our study those individuals were not frequent. The duration of the lesions ranged from 2 months to 8 years which is in accordance with the literature<sup>8</sup>.

A variation of the size of the sialoliths were also observed. In this study, their diameter ranged from 0.03 – 4cm (mean 0.98cm). According to the literature, the average sizes may vary from 0.33 cm to 1.79cm<sup>11,14</sup>. Some studies have demonstrated that the submandibular and parotid glands are more frequently affected by sialolithiasis when compared with minor salivary glands<sup>1,2</sup>. In our study, sialoliths in the submandibular gland was observed in eight cases while the minor salivary glands were less affected. This was consistent with the fact that it commonly occurs in the submandibular gland as previously reported<sup>1,2</sup>.

Considering only the minor salivary glands, the



**Figure 2.** Multiples microsialoliths exhibiting concentric feature (a). An extensive unique displaying laminated layers of organic and inorganic substances in a concentric pattern (b). Ectasic glandular duct exhibiting squamous metaplasia and fragments of concentric lamellas in the lumen. In addition, a chronic inflammatory infiltrate is observed (c).

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most common sites related to sialoliths was the buccal mucosa followed by the upper lip. Some authors have reported upper lip, buccal mucosa, lower lip as the most affected sites<sup>4,8,14</sup>. It is important to mention that sialoliths have been associated particularly to sites where trauma has been caused by the dentition, such as the lips and the buccal mucosa close to occlusal lines, which may indicate a possible etiological factor<sup>8,14</sup>. The resultant inflammation from trauma modifies the composition and properties of the salivary gland or duct, and may lead to sialolith formation<sup>25</sup>. However, no history of trauma prior to the development of the lesions was observed in the clinical charts assessed for this study. Clinically, lesions of sialolithiasis have been described rounded, oval or cylindrical and mobile or firm nodules<sup>8,11</sup> which is in accordance with our clinical findings.

Usually, sialoliths may cause stasis of saliva, leading to bacterial ascent into the affected gland. As a result, it might cause damage of the parenchyma and yield a suppurative process associated with fistula and purulent drainage via mucosa or skin<sup>16,17</sup>. In the current report, two cases of purulent discharge was referred in the retrieved data.

The most common clinical hypotheses for sialolithiasis have been reported such as: mucocele, sialodentitis, fibroma and salivary gland tumor<sup>4,8,11</sup>. In our case series, the most frequent clinical diagnosis reported was sialolithiasis. Likewise the occurrence of unaccurate clinical diagnosis for sialolithiasis is often reported on the literature<sup>4,8,18</sup> with studies ranging from 0% to 20%<sup>4,11,18</sup> of compatibility between clinical and histopathological diagnoses.

Our study identified a predominance of single sialoliths which has been often described for other authors<sup>8,11,14</sup>. However, lesions with multiple sialoliths on minor and major salivary glands also have been found<sup>19,20,21</sup>. According to a previous report, multiple sialoliths might indicate the existence of multiple areas of sialolithogenesis<sup>4</sup>.

When associated with clinical information, the diagnosis of sialolithiasis by plain images is not a difficult task<sup>8,9</sup>. However, when the sialolith reaches large proportions other clinical hypothesis might be considered, especially if it is located in the mandibular angle. Thus, computerized tomography notably multidetector computed tomography (MDCT) provides a better resolution for soft tissues enabling a more accurate

diagnosis for sialolithiasis as previously demonstrated in one case of our study<sup>21</sup>. In addition, sialoliths identified by orthopantomography is a common diagnostic exam as demonstrated in this study and also in the literature.

Histopathologically, our cases fulfilled the diagnostic criteria of sialolithiasis which is assumed to be formed by initial central core of organic material which grows progressively by a layered deposition of organic and inorganic materials creating a lamellar pattern or a globular shape calcified zone<sup>4,8,11</sup> being also hypothesized that mitochondria and lysosomes bodies originated from the ductal system play a role in the etiology of salivary glands stones<sup>22</sup>. Likewise, in this study, most of the lesions were formed by calcified nodular structures in a lamellar arrangement. Recently, it has been observed that lipids are the main components of the initial central core of the sialolith. Also, whitlockite crystallites are involved in the initial mineralization process and progressive transformation into more stable hydroxyapatite crystallite<sup>23</sup>.

Morphological alterations involving the duct or glands are common findings in sialolithiasis. Salivary duct cells can display features of squamous or mucous metaplasia which results from a persistent ductal obstruction associated with chronic inflammatory process on periductal zone<sup>4,8,19</sup>. We observed ductal squamous metaplasia in some of our cases associated with a mild to moderate chronic inflammatory infiltrate.

In one case, the histological diagnosis was chronic sclerosing sialadenitis located in a submandibular gland. It has been reported that sialoliths are also considered one of the main etiological factors for chronic sclerosing sialadenitis<sup>21,24,25</sup>. Sialoliths can cause obstruction of salivary flow or stasis of secretions leading to an outcome of acinar cell atrophy, periductal sclerosis, ductal dilation and retention of salivary secretions accompanying edema and inflammatory cell infiltration<sup>25</sup>. In our case, we found a sialolith associated to the mentioned lesion.

As concerns the treatment, a sialolith must always be removed, since a long term obstruction of the salivary gland duct can lead to inflammation and secondary infection<sup>1,8,10</sup>. Non-invasive conservative treatments, including the use of sialogogues and irrigation<sup>18</sup> as well as surgical treatments such as sialodochotomy, sialoendoscopy, extracorporeal shockwave lithotripsy and excision of the gland have been reported<sup>2,8,10</sup>. In general, recurrence is not a common finding and may indicate undiagnosed

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multiple sialoliths<sup>8,11,15</sup>. In the current case series report, surgical excision of the lesions was performed in all cases and recurrence was not observed.

Finally, the sialolithiasis seems to be a nonfrequent lesion in our Oral Pathology Service as between 2.631 biopsies this lesion represented 0.41%. Nonetheless, the sialolithiasis should be considered in the differential diagnosis of lesions that particularly affects the submandibular glands.

### CONFLICT OF INTEREST

We have no conflicts of interest.

### ETHICS STATEMENT/CONFIRMATION OF PATIENTS' PERMISSION

This study was approved by the Institutional Review Board of our hospital, and we did not use any private information about the patients.

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