

# An Unusual Case of Submandibular Sialolithiasis in Early Childhood: A Case Report

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## Abstract

Sialolithiasis is the most common condition of the salivary glands and it is characterized by the development of a calcified structure within a salivary duct. This condition can lead to inflammation, bacterial infection, and abscess. The different forms of management range from the direct massage of the duct to the complete removal of the gland. This report describes an unusual case of submandibular sialolithiasis in a four-year-old girl. The main complaints were pain when eating and swelling in the floor of the mouth. The diagnosis was sialolithiasis in the duct of the submandibular gland. Since the salivary stone was palpable and located in the anterior portion of the duct, a minimally invasive procedure was planned. An excisional biopsy was performed and a unilateral salivary stone was removed. The histopathological analysis confirmed the diagnosis. Although rare in children, understanding the diagnostic process and the different treatment modalities leads to a favorable prognosis.

## Keywords

Pediatric Dentistry, Sialolithiasis, Biopsy, Salivary Gland Calculi

## 1. Introduction

Sialolithiasis is a benign condition characterized by the development of a calcified structure within the duct of a major salivary gland [1] [2]. This condition can result in the obstruction of the gland, which can lead to inflammation due to harmful salivary enzymes, bacterial infection (sialadenitis), or, in rare cases, ab-

sciss. The incidence is estimated to be one in 10,000 to one in 30,000 individuals [3].

It has been shown that the long upward path of the submandibular duct (Wharton's duct) is the most affected by calcifications [1] [4]. Sialolithiasis can occur equally on the right or left side at any age but it is more common in middle-aged adults and rare in children [4]. There are some divergences regarding whether sialolithiasis affects one of the sexes more than the other [5].

The factors associated with the formation of a salivary stone are not well understood. In general, the etiologic factors are divided into two major groups: anatomical factors affecting the formation or flow of saliva (*i.e.*, duct stenosis or inflammation) and composition factors (*i.e.*, increased calcium content or altered enzyme function). The main risk factors are dehydration, smoking, increased local levels of calcium, diuretics, and drugs that reduce salivary flow; however, the cause is idiopathic in many cases [6].

Patients with sialolithiasis have a variety of symptoms, the most common of which is unilateral swelling of the salivary gland, decreased saliva production, and the acute onset of pain that worsens when eating [2] [7]. Modern diagnostic techniques involve ultrasound, computed tomography (CT), magnetic resonance imaging (MRI), and sialoendoscopy [8] [9].

The condition is usually managed by removing the stone. However, a variety of treatment options exist for sialolithiasis, such as medications (sialogogues), direct massage of distal stones out of the duct, and a variety of surgical procedures. Only a few cases of sialolith have been reported in children [2] [4] [5]. The present report describes an unusual case of sialolithiasis in a child and discusses the management of the condition with a minimally invasive surgical intervention.

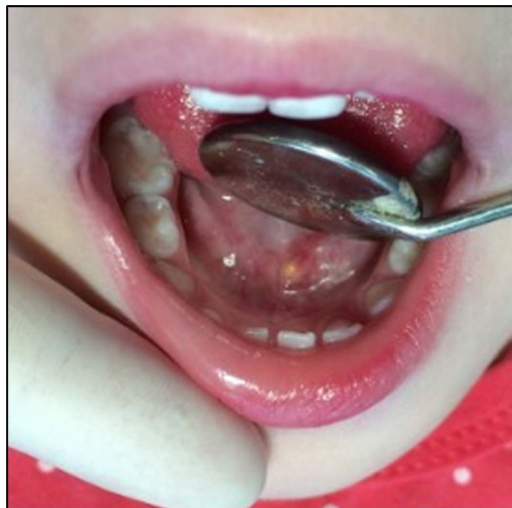
## 2. Clinical Case

A four-year-old girl visited the Department of Pediatric Dentistry at School of Health and Life Sciences, *Pontifícia Universidade Católica* in the state of *Rio Grande do Sul*, Brazil, with the complaint of pain in the floor of the mouth, especially during meals. The mother reported that the child occasionally had slight swelling in the left submandibular region. The intraoral examination revealed a palpable firm mass near the orifice of the submandibular duct. Bimanual palpation of the left submandibular salivary gland further indicated that the mass was mobile and tender (**Figure 1**).

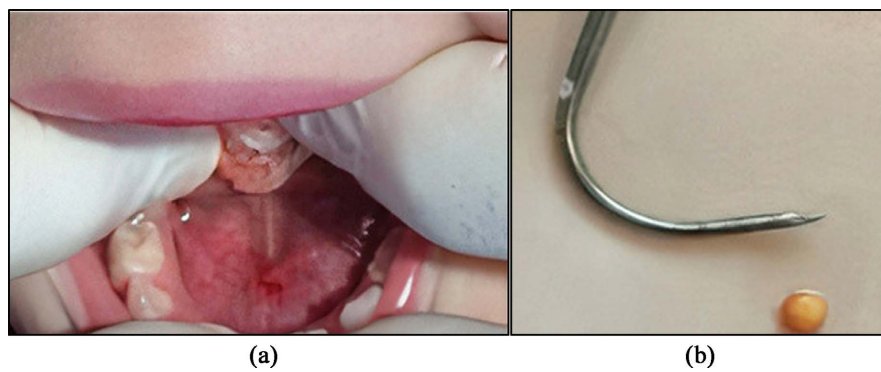
The calcified lesion was yellowish and translucent and moved within the duct when pressed. When the gland was "milked", the yellowish mass shifted to the lumen of the duct but was not expelled. The preliminary clinical diagnosis was of a sialolith in the duct of the left submandibular gland, requiring histopathological confirmation. Due to the urgency and the presence of the sialolith in the most superficial portion of the duct, excisional biopsy was proposed. Children are more sensitive to the effects of ionizing radiation. Complementary imaging

tests were ruled out due to the superficial location of the lesion near the orifice of the submandibular duct. Thus, according to ALARA principles (“as low as reasonably achievable”), imaging tests were not necessary, respecting good radiological practices to minimize or eliminate unnecessary radiation in the diagnosis. Anesthesia was initially performed with 10% lidocaine spray (100 mg/ml) (Xylestesin®, Cristália, São Paulo, Brazil) over the surface of the duct outlet and epithelial tissue surrounding the lesion. The tongue was raised and held with gauze by an assistant. The mass was secured with a hemostat. The surgeon made an incision with a #11 scalpel blade and completely removed the mass from the floor of the mouth. Bleeding was minimal and hemostasis was achieved quickly after the incision. No suture was performed. The patient did not complain of immediate intraoperative or postoperative pain and normal conditions were found during clinical evaluation immediately after the procedure (**Figure 2(a)** & **Figure 2(b)**).

The lesion was sent for histopathological analysis along with the clinical data obtained on the day of the biopsy. The stone was a yellowish tissue fragment with a smooth surface, round shape, and hard consistency, measuring 2 mm in diameter. The histopathological analysis revealed a rounded mass with calcified



**Figure 1.** Sialolith in duct outlet of submandibular gland.



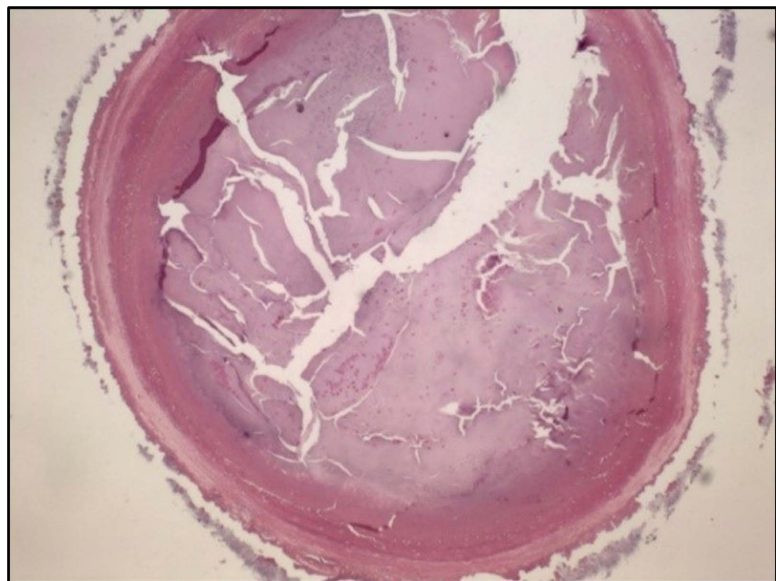
**Figure 2.** Immediate post-surgical excisional biopsy (a) and sialolith after its removal (b).

lamellar and globular rings, confirming the diagnosis of a sialolith (**Figure 3**).

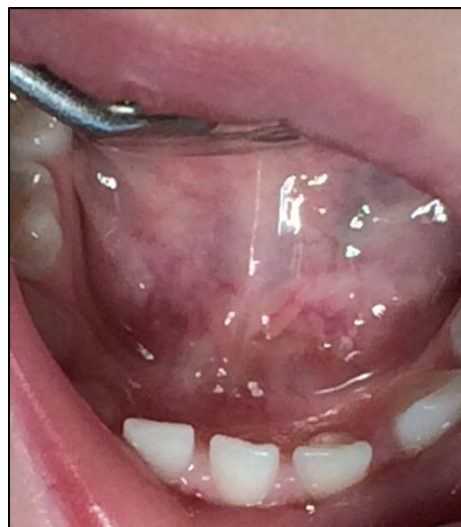
The postoperative evaluation performed seven days after the surgical procedure revealed good recovery and the patient reported no episodes of pain or bleeding. The region had no local scar and appeared within normal limits during the examination of the floor of the mouth. The outlet of the gland duct appeared normal, with no change in volume and no rigidity (**Figure 4**). Ethical approval was obtained from the university (certificate number: 3.377.556) and the patient's guardian granted written informed consent for publication of the case.

### 3. Discussion

Sialolithiasis is the most common condition of the salivary glands. However, less than 5% of cases occur in the pediatric population and the current literature offers



**Figure 3.** Histopathological image showing round calcified mass (HE 4X).



**Figure 4.** Postoperative clinical outcome.

few clinical case reports [5]. This report presents an unusual case of sialolithiasis managed with a minimally invasive surgical intervention in a young child.

The management of the behavior of children in the first years of life is one of the major challenges in the clinical practice of pediatric dentistry. At the same time, parents have high expectations regarding the behavior of their children and the performance of the dentist. Behavioral problems during dental care are generally related to the fear, anxiety and pain. This is a barrier that leads to postponing appointments, which impedes a timely diagnosis and treatment, leading to an increase in the incidence and severity of oral problems. In the present case report, anamnesis with those responsible and a careful physical examination of the child were fundamental for the child's behavioral adaptation and for the guarantee of the parents' trust.

While there is no evidence proving the true origin of this type of calcium deposit, theories have been put forth to explain sialolithiasis, such as calcification around a foreign body, desquamated epithelial cells, and presence of microorganisms in the duct [2] [9] [10] [11] [12] [13]. Alkaline pH, the mucin content of saliva, and a high calcium concentration may also explain the pathogenesis of sialolithiasis [14]. According to McCullom *et al.* [11] salivary calculi form more easily in adults because calcium and phosphate concentrations increase with age.

Sialoliths are clinically round or ovoid, ranging from 1 mm to 1 cm, they can be rough or smooth, and usually unilateral and yellowish in color [10]. Im *et al.* [15] studied the mineralization mechanism and growth of a submandibular gland sialolith and found that an organic core grew as inorganic materials were deposited and calcified in alternating layers. The inner core is considered the initial sialolith, which subsequently enlarges due to the deposition of additional organic and inorganic material, forming the outer lamellae (layers). The mineralization process may include an initial whitlockite development and successive transformation into more stable hydroxyapatite. The exact ratios of inorganic material found within a sialolith depend on the chemical environment in which it forms [15].

Approximately 85% of sialoliths occur in the submandibular gland, which located on the floor of the mouth near the lingual frenulum [2]. This occurrence may be related to 1) the longer and larger caliber duct and slower salivary flow rate, 2) the fact that saliva flows against gravity, 3) the presence of more alkaline saliva, which facilitates the precipitation of inorganic salts, and 4) the high mucin content of the saliva, which is more viscous, resulting in a more stagnant flow of secretions in the submandibular gland. Lower rates have been reported in the parotid (5% to 10%), sublingual, and minor (<1%) salivary glands [16].

There are considerable clinical differences in sialolithiasis between children and adults. The duration of symptoms seems to be shorter in pediatric patients compared to adults. Other differences are the more distal location, relatively smaller sialoliths, and lower recurrence. On the other hand, the dominant location of pediatric sialolithiasis mirrors that found in adults [12] [17]. The low

prevalence of sialolithiasis in children may be related to the generally long time required to form a salivary stone, which would explain the higher occurrence in older patients [1] [2]. Moreover, salivary gland infections related to sialoliths do not appear to recur in pediatric patients [12].

Pain and swelling of salivary glands during meals are generally associated with sialolithiasis. According to those responsible, pain and swelling in the submandibular region were common, especially during meals. It is also important to perform a careful evaluation of the functioning and anatomy of the gland involved. A uniformly solid, hard gland during bimanual palpation indicates a hypo-functional or non-functional gland. A palpable stone is found during bimanual examination in most cases [14]. As a complement to the physical examination, there is a variety of radiographic tests for the diagnosis of sialolithiasis. In the present case, the physical examination was sufficient to establish the treatment strategy, later confirmed by the histopathological examination. While conventional radiography is useful, indication of imaging techniques using ultrasound, computed tomography, or sialography can be performed, as they provide better sensitivity for diagnosis, in addition to determining the precise location [4] [6] [18].

The management of sialolithiasis should begin with conservative measures, such as massaging of the salivary gland, stimulation of the gland by eating acidic foods, nonsteroidal anti-inflammatory drugs, and sialogogues. As a consequence, the stone is often spontaneously expelled, especially when it is superficial and mobile [11] [12]. If conservative management is unsuccessful, treatment is dictated based on the size, number, and location of the sialolith. In the present case report, conservative surgery was chosen, since most salivary calculi in children are obviously palpable and located close to the duct orifice, which allows for minimally invasive procedures or surgeries related to approaches that can be performed through minimal entry ports in the body, which are capable of performing the procedure fully, with minimal inflammatory response, less postoperative pain and minimal bleeding [5]. The procedure involves a small incision that exposes the salivary stone trapped in the duct of the involved gland, followed by its complete removal by excision. Sialodenectomy is rarely necessary for the treatment of sialolithiasis. The minimally invasive procedures discussed above have excellent success rates with minimal morbidity in comparison to traditional surgical techniques [2] [19].

#### **4. Conclusion**

Sialolithiasis is the most common benign cause of salivary gland swelling but it is relatively rare in childhood. This article presented the management of the obstruction of a submandibular duct in a young patient using a simple, effective, minimally invasive surgical intervention. Understanding the diagnostic process and different treatment modalities leads to a favorable prognosis. A disclosure approach can aid in ensuring that patients receive appropriate initial manage-

ment, resulting in the quicker resolution of symptoms.

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## Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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