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Surgical management of submandibular gland sialolithiasis in a children: A case report and literature review

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Abstract
Sialolithiasis is a relatively common condition in adults but is rarely found in children (3%–13.8%). The majority of sialoliths are formed (92%) in the Wharton duct.
Clinical manifestations of this special entity differ from those of adult sialolithiasis in some aspects, that is, shorter duration of obstructive symptoms, smaller and distally located calculi.
We report a case of a 9-year-old boy with a sialolith in the left anterior Wharton's duct. Radiographic and clinical examination confirmed this diagnosis. The calculus was removed transorally under local anaesthesia. Follow-up examinations showed recovery of the function of the affected gland.

Keywords: Sialolith, sialolithiasis, submandibular gland, wharton's duct

Introduction

Sialolithiasis is the most common disorder of the salivary glands and may range from tiny particles to stones that are several centimeters in length. According to Levy *et al*., the prevalence of submandibular gland sialolithiasis is 80%, 19% in the parotid and 1% in the sublingual glands [1]. It is rarely found in children (3%–13.8% of all sialolithiasis cases) [2-3].
The submandibular gland is most frequently involved because of its anatomic location, long, tortuous duct with a narrow orifice compared to the main portion of duct. Along with this salivary stasis and ductal inflammation and injury are important factors contributing to stone formation. It is believed that intermittent salivary stasis results in an alteration of the mucoid elements of saliva forming an organic gel. This gel becomes the framework for the deposition of salts, which leads to the development of calculi. In the sub- mandibular gland, development of a sialolith might be the primary event that results in stagnation of saliva and inflammation, encouraging bacterial migration and resulting in sialadenitis [4].

Case report

A 9-year-old healthy male was referred to the pedodontic and prevention department of the consultation and dental treatment center of Rabat. He gave a history of increase in the size; since 1 month; of a hard and asymptomatic mass in the left Wharton's duct.
The extra oral examination was without particularities. On palpation, the presence of small lymphadenopathy was detected under the left mandibular angle, it was painless and not very mobile compared to the deep plane.
Intra-oral examination (Fig. 1) showed an inflamed Wharton's ductal orifice and a calcified stone was visible, palpable and with well defined limites in the anterior part of the left submandibular duct area, precisely at the sublingual caruncle. A deposit of tartar was present on the vestibular and lingual surfaces of the 41 and 31. (Fig. 2)
On occlusal radiograph of mandible, a radiopaque mass was evident with a diameter of 7 mm in the anterior portion of the duct of left submandibular gland. (Figs. 3)
Oral prophylaxis was performed and the patient was prepared for surgery. Left-side inferior alveolar nerve block and lingual nerve block local anaesthesia was administrated (Figs. 4)
Silk suture was tied behind the sialolith around the duct, so as to isolate the stone and to prevent movement of the stone to the inner part of the duct or hilum of the gland (Figs. 5 and 6). An incision of 6-7 mm made on mucosa of the left submandibular duct above the sialolith

which measuring about 7 mm × 3 mm was gently removed without injuring the lingual nerve and without postoperative stenosis (Figs. 7 and 8), then sutures were placed. (Fig. 9)

Post-operative instructions were given. Antibiotic (Amoxicilline 500 mg, 2 times per day for 7 days) and analgesic (Paracetamol 500 mg, 3 times per day for 3 days) were prescribed, and the patient was recalled after 10 days (Fig. 10). The patient was completely symptom free; in particular the lymphadenopathy has disappeared; and the wound had healed. (Fig. 11).

Fig 1: inflammation of Wharton's ductal orifice and calcified stone at the sublingual caruncle.

Fig 2: Deposit of tartar on the vestibular and lingual surfaces of the 41 and 31.

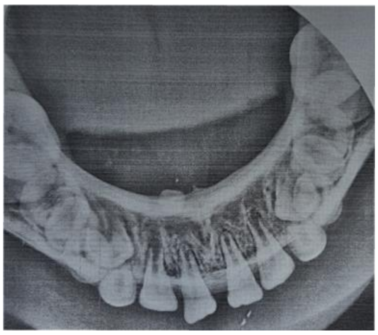

Fig 3: Occlusal radiograph of mandible showing a radiopaque mass, in the anterior portion of the duct of left submandibular gland.

Fig 4: administration of local anesthesia.

Fig 5, 6: Isolation of the stone by silk suture to prevent his movement to the inner part of the duct or hilum of the gland.

Fig 7, 8: Incision on mucosa above the sialolith which is removed.

Fig 9: Placement of sutures.

Fig 10: Follow-up after 10 days.

Fig 11: Wound had healed.

Discussion

Salivary gland plays a vivacious role in sustaining homeostasis in oral microenvironment by secreting saliva, which in turn encompasses various components comprehensive of antimicrobial modules [5].

Prevalence

Sialolithiasis is a common salivary gland disorder characterised by the obstruction of the salivary secretion because of calculi. They accounting for approximately one third of salivary gland disorders [6].

Salivary calculi or sialoliths are calcified structures or concretions located in the parenchyma or ductal system of the salivary glands. Although this special entity is a relatively common in adults, it is rare in pediatric cases, with <150 cases reported in the literature [7].

The low incidence of these diseases in children may be due to the relatively long time required for the formation of a salivary calculus and because sublingual papillae and salivary glands are very small in children, rendering it difficult for foreign matter to enter. Furthermore, a calculus is more easily formed in adults than in children because the concentration of calcium and phosphate ions in the resting saliva increases with age [7]. Man Ki Chung *et al*. suggests that a male predominance is not characteristic of pediatric cases [8].

Symptoms

On the basis of the review of the literature, 3% of the patients with a submandibular stone have no symptoms or minimal discomfort [9-6].

Indeed, the pain and swelling are caused by the obstruction of the salivary flow in the affected gland, resulting in accumulation of saliva and a subsequent increase in intra-glandular pressure. Larger stones are most often incriminated in this symptomatology. In incomplete obstruction of the duct, saliva can seep through or around the sialolith. In these cases, a salivary stone can be symptomless and these stones may be an incidental finding on a dental panoramic radiograph [6-10].

Compared with sialolithiasis in adult patients, pediatric sialolithiasis has different clinical characteristics such as

A short duration of symptoms (0–3 months, mean 1.0 month) than in adult patients (0–120 months, mean 11.5 months). This manifestation can be explained by the facts that the pediatric population is less exposed to various infections and has intact salivary gland function. In addition, the production of saliva in affected glands appears to be less compromised in pediatric patients. Moreover, a lower tolerance to uncomfortable symptoms among the patients or the parents [7, 8], more distal location due to a higher salivary flow, and relatively small size of stones [8].

Biochemical composition [6]

Sialoliths consist of both organic and inorganic material, but there is a great variation in the relative contribution.

Submandibular stones contain less organic material (9–12% of the dry weight). - The organic part consists of collagen, neutral and acid glycoproteins, other proteins (probably bind to calcium ions in sialoliths) [11], lipids and carbohydrates such as glucose and mannose.

Slomiany and co-workers [12] suggested that the lipids and phospholipids are important for the initiation of the mineralisation of a salivary stone.

Submandibular stones contain between 70–80% of inorganic matrix. The mineral component is proportional to the size of the sialolith, suggesting that mineralisation of the organic matrix increases with time. Hydroxyapatite ($Ca5(PO4)3OH$) is present in all submandibular stones, frequently together with whitlockite ($Ca3(PO4)2$). Stones retrieved from an infected gland may also contain ammonium and magnesium [13, 14].

Pathogenesis

Salivary duct stones may occur in all of the salivary glands, but they are mainly found in the submandibular glands (80-90%), especially the submandibular duct (Wharton duct) because on the one hand of its long, narrow, winding transection. On the other hand of the more-alkaline pH, mucin content, high $Ca2+$ concentration of submandibular saliva and salivary flow against gravity, [13-15].

Nevertheless, the exact mechanism of the formation of calculi is still unclear and various hypotheses have been proposed.

One hypothesis states that an initial organic nidus may occur that progressively grows because of the deposition of inorganic and organic substances, whereas another hypothesis states that intracellular microcalculi are excreted in the canal and function as a nidus in the ductal system. Furthermore, debris, bacteria, or substances migrating in the salivary ducts may cause sialolithiasis [7, 8].

Seong Jun Won and col suggested that pediatric sialolithiasis may result from the intrinsic factors of the salivary gland itself. Such as the anatomy or function of the affected gland not from oral or oropharyngeal infections [16].

Furthermore, it is generally accepted that bacterial infection initiates the process of calculi formation due to increasing salivary pH and organic matters. Infection can also disturb the status inside the salivary duct by changing morphoanatomic factors (salivary duct stenosis, inflammation or injury) and saliva composition factors (high supersaturation or crystallization inhibitors deficit) [8].

Thus, it can be concluded that the aetiologic factors implied in the sialolith formation can be classified in two large groups: a) saliva retention due to morphoanatomic factors (salivary duct stenosis, salivary duct diverticuli, etc.) and b) saliva composition factors (high supersaturation, crystallisation inhibitors deficit, etc.). The existence of a bacterial infection can also favour the development of sialoliths [9-13].

Studies have shown that sialolithiasis can be related to primary hyperparathyroidism, as the latter can lead to altered electrolytic imbalance which in turn can lead to the formation of salivary calculi. In a retrospective analysis done in US, it has been shown that approximately 8100 salivary stone cases could have coexisting hyperparathyroidism, in which 2700 cases, sialolith might be the only presenting symptom [17]. In a study done by Rakesh *et al*. Comparison between sialolith and nephrolith was done in an ultrastructural level, which demonstrated that nephrolith is related to electrolytic imbalance in urine whereas in sialolith it is related to electrolytic imbalance in saliva. Sialolith is predominantly composed of hydroxyapatite crystals whereas in nephrolith, it is calcium oxalate and phosphate. Approximately, 10% of the patients showed a possible link between sialolith and nephrolith [18, 19].

Diagnosis

The key step in the diagnosis of sialolithiasis is the elucidation of a thorough history and careful clinical examination. Therefore, bimanual palpation of oral cavity is mandatory to make a diagnosis for patients suspicious of sialolithiasis, because most of calculi are located in the distal duct in pediatric patients [8].

In the same way, the praticien must be attentive and examine the affected side of the floor of the mouth that may be elevated and inflamed.

In addition to an oral examination, the most effective imaging modalities for sialolith are X-rays, sialography, ultrasound and computed tomography. Occlusal and panoramic views are the most common radiographic techniques used to diagnose a sialolith [13].

The detection rates differ between intraoral occlusal radiographs and extra-oral panoramic radiographs. Extra- oral radiographs will detect fewer salivary stones because many calculi will be projected superimposed on bony structures or teeth. This indicates that an occlusal radiograph is the most useful method for detection of a sub- mandibular sialolith [20].

Computer tomography (CT) and cone beam computer tomography (CB-CT) can detect any size of sialolith, but have the disadvantage of a relatively high radiation dose [21].

Ultrasonography allows detection of stones with a diameter of 2 mm or more. This technique has the additional advantage that it also can be used during an acute episode of sialendenitis [22].

Radiolucent sialoliths can be imaged with sialography. A contrast agent is injected into the duct of the affected gland and subsequently radiographs are taken. Sialography is most frequently used for detection of parotid sialoliths. It is contraindicated during acute episodes of sialendenitis and in patients with an allergy to contrast media [23].

Sialendoscopy is a minimally invasive technique to visualise the salivary duct system, usually performed under general anaesthesia. An endoscope with a very small diameter (0.6 mm) is introduced into the duct after the orificium has been dilated with special instruments with increasing diameters from 0.8–1.6 mm. The endoscope has a rinse channel that can be used to flush the duct with saline or an anti-inflammatory rinse. This flushing primarily results in a better image of the salivary ducts but may also have a therapeutic effect [13].

Differential diagnosis for sialolithiasis includes other disease processes that may affect the salivary gland: infections, inflammatory conditions, and neoplastic and non neoplastic masses [24].

Salivary stones should be differentiated from phleboliths, inflammation of maxillary bones, and calcification of lymph nodes [13].

Treatment

The primary objective in the treatment of sialolithiasis should be preservation of gland function in combination with a low level of discomfort for the patient and complications including external scar, loss of facial contour, facial numbness, facial weak- ness, gustatory sweating, and reduced salivary volume [25, 26].

The optimal treatment strategy for pediatric sialolithiasis is sialoendoscopy which is a successful treatment for submandibular ductal stones [16] because it reduces the risk of missed stones, nerve injury, and the requirement for sialoadenectomy [8-16]. This option should be considered as a first intention treatment for all salivary stones in children.

Non-invasive conservative management of sialolithiasis consists of gland massage, in combination with use of sialogogues and irrigation. This treatment has the highest success rate when stones are small and located in the duct [25-27].

Other treatment options are [9]

1. For large sialoliths that are located in the close proximal duct managed by extracorporeal shock wave lithotripsy (ESWL) (ultrasound to break the stone), this method entails a risk of damaging the salivary gland, the ear, dental amalgams, or the central nervous system [28],
2. Endoscopy intracorporeal shock wave lithotripsy (EISWL) is also gaining importance because of less damage to the adjacent tissues during the procedure,
3. The extra oral approach might require some time mainly when dealing with a large stone and a large sialolith requires excision of the gland.

Stone size/location, CT-localization (Computed tomography), and palpability were predictive of calculi that require an incisional approach. If these factors are recognized, the surgeon can consider proceeding directly to incisional sialolithotomy [28, 29].

In conclusion, intraoral removal of sialoliths under local anesthesia is recommended as the treatment of choice in pediatric patients [7-9].

Thus, a functional and anatomical evaluation may be clinically important to treating the sialolithiasis and to preventing the recurrence in pediatric patients. For example, metabolic or chemical analyses could help discriminate the patients at high risk for recurrence [16-30].

Conclusion

Pediatric sialolithiasis presents a special entity. Dental practitioners thus play a pivotal role in timely diagnosis and treatment of sialolithiasis. Treatment modalities should be framed considering location and size of sialolith [5].

Intraoral removal of sialoliths under local anes- thesia is recommended as the treatment of choice in pediatric patients with of the occurrence of a large number of relatively small and distal calculi [7].

Other nonsurgical methods can also be adopted to avoid stricture formation along the course of the duct. Recurrence of sialoliths is uncommon and is estimated to occur in 1%–10% of the patients [7].

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