Large sialolith in the submandibular gland of a child

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Abstract
Sialolithiasis is a disorder encountered by oral surgeons that is rarely seen in children, although it is rather common in adults. Most sialolith found in children are smaller than 5 mm in diameter, and the majority of reported cases have been treated by surgically. We report a 9-year-old boy with a sialolith that measured $12 \times 3.5 \times 3$ mm, which had developed in Wharton’s duct and was then spontaneously passed.

Introduction
A sialolith is a calcareous concretion that may occur in the ducts of the major or minor salivary glands, or within the glands themselves. The submandibular gland is most commonly affected, in most cases, the calculus is found close to the orifice or in the anterior two-thirds of Wharton’s duct. Formation of a sialolith is considered to be the result of calcium salt around a central nidus, which may consist of desquamated epithelial cells, bacterial or micro-organismal decomposition products, or foreign bodies. The high incidence of submandibular calculi in this gland has been explained by the high concentration of calcium salts, pH, and mucin content located there1. Although sialolithiasis is not uncommon in adults, only about, 3% of cases have been reported in children. This low frequency of sialolithiasis in young patients is due to the long period required for sialolith formation, faster salivary flow rate, lower concentrations of calcium and phosphate in saliva, and smaller orifice size for foreign body entrance. In adults, average calculus size is $6.3$ mm (range, 2–30 mm), but most are less than 5 mm in children2.3).

Children who present with sialolithiasis are generally healthy and without systemic illness, except for symptoms of acute inflammation. Most of their chief complaints regard intermittent and unilateral pain and swelling in the submandibular region, usually associated with eating.

We describe here a case of submandibular gland sialolithiasis in a 9-year-old child, which resulted in the formation of a calculus greater than 10 mm in diameter that was located in Wharton’s duct on the right side.

Case report
A 9-year-old boy was referred to our hospital with the chief complaints of tenderness and swelling of the floor of the mouth with spontaneous pain, which had occurred over a period of 3 days. The patient was first seen by a general practitioner, who made a diagnosis of acute sialodochitis, and prescribed a 2-day regimen of an antibiotic (cefotiam dihydrochloride). The past medical history of the patient was unremarkable. Physical examination revealed no acute distress, and body temperature, pulse, and blood pressure findings were within normal ranges. Clinical examination revealed swelling in
the right submandibular region, and diffuse swelling of the right submandibular gland was palpable with associated tenderness (Fig. 1). In addition, the right submandibular nodes were palpable and about the size of the little finger, and showed mobility and significant pain. Trismus was not present. Intraoral examination revealed that the left-side Wharton’s duct was normal, with clear salivary flow produced by gentle manipulation of the gland. In contrast, Wharton’s duct on the right side exhibited a prominent edema, with swelling, discharge of pus, and erythema of the orifice (Fig. 2). Radiographic examination revealed a radiopaque mass in the anterior one-third of Wharton’s duct, close to the orifice (Fig. 3). A diagnosis of submandibular acute sialodochitis caused by a sialolith was made. One day after beginning the prescribed antibiotics, a sialolith spontaneously migrated from the gland (Fig. 4). The calculus was an ash gray-colored oval, and measured approximately 12 mm long and 3.5 mm in diameter.

Physicochemical chemical analysis revealed the
presence of 67% calcium phosphate, 28% protein and 5% calcium carbonate. The patient had an uncomplicated recovery and was released 1 day after the migration of the calculus. He was asymptomatic, and a subsequent follow-up examination 1 month later revealed the disappearance of submandibular swelling. Four months later, there were no signs or symptoms of recurrence.

**Discussion**

Although sialolithiasis accounts for 50% of the major salivary gland diseases localized to the head and neck region, individual reports of management of pediatric patients with sialoadenitis or sialolithiasis are limited. In children, 80% to 90% of cases are found in the submandibular gland, compared with 5% to 10% in the parotid gland, and approximately 5% in the sublingual and other minor salivary glands. The submandibular gland salivary outflow includes large amounts of calcium and phosphorus, compared with that of the parotid gland, and includes mucin, which has a high viscosity. Further, the submandibular duct has an opening on the floor of the month that can easily retain saliva, and Wharton’s duct is longer than other sublingual ducts. These characteristics may account for the preponderance of reports of occurrence in this gland.

Most sialoliths previously reported were removed using a surgical procedure, while in a few cases they spontaneously migrated out. In pediatric patients with sialolithiasis, it is easy for the sialolith to be passed from the duct, since most are located near the orifice of Wharton’s duct, salivary flow is faster than that from other salivary glands, maturation of the sialolith is generally poor, the tissues surrounding the ducts are soft, the size of the sialolith is usually less than 5 mm in size, and swelling and pain are milder than in adults. For these reasons, the first choice of treatment for pediatric sialolithiasis in the submandibular gland may be to wait for spontaneous migration. In fact, a sialolith with a diameter greater than 10 mm in size passed from the duct 1 day after starting antibiotic treatment observed in the present case, to our knowledge, is the largest. The sialolith is generally containing calcium phosphate, protein, and calcium carbonate. In adult cases, the ratio of calcium phosphate is high compared with pediatric cases. In present case, the composition of sialolith is approximately same as the previous report in child. Additionally, the concentration of calcium carbonate may provide evidence that this sialolith was made of saliva, but not other hard tissue or foreign material. Approximately 40% of all submandibular stones are found in the distal portion of Wharton’s duct or at the orifice, and can be removed by simple intraoral procedures performed under local anesthesia. For calculi that lie in the proximal duct or gland, the treatment of choice has been sialoadenectomy, which is effective in eradicating symptoms, but carries the risk of nerve injury. Recently, several new minimally invasive techniques have been introduced for the treatment of sialolithiasis, such as extracorporeal sonography and intracorporeal endoscopically controlled lithotripsy, which have completely changed therapeutic methods utilized. In the case of parotid duct stones, the long-term outcome with extracorporeal lithotripsy has been quite satisfactory, with 50% of all patients reported to be free of stones and 80% free of symptoms. In comparison, fewer than 30% of patients who suffer from sialolithiasis of the submandibular gland and receive lithotripsy treatment are reported to be free of stones. Therefore, the benefits of a minimally invasive technique, compared to those of other moderately invasive or other invasive surgical and gland-preserving techniques, must be considered for these patients.

Various techniques of sialodochotomy have been described in the literature, with the major point of concern being the risk of injury to the lingual nerve, which passes in close proximity to Wharton’s duct. Surgical excision of the gland is recommended in cases with extreme proximal stone localization, due to the anatomical circumstances and the assumption that the submandibular salivary gland will not tend to exhibit improvement after years of obstruction and recurrent inflammation.

Diagnosis is very important for correct selection of treatment of sialolithiasis in children. As in most cases with spontaneous passage of a sialolith, in the present case the stone was localized in anterior portion of Wharton’s duct, and its passage did not depend on its size. Preservation of gland function in conjunction with low-level risk and minimal discomfort for the patient should be the primary objectives of treatment of sialolithiasis. Apart from problems such as scar formation, disturbances of skin sensation, and injury to the gustatory nerves, transient functional disturbances of the marginal branch of the facial nerve are most often encountered in up to 12.5% of open gland excisions. Further,
permanent lesions have been reported in 7% of these cases\textsuperscript{20}\textsuperscript{21}. In addition, unilateral excision of the submandibular gland also leads to substantial reduction of nonstimulated flow of saliva, which may significantly affect oral hygiene, risk of caries, and the development of xerostomia\textsuperscript{21}. Conservative treatment should therefore be selected for sialolithiasis in children.

A few reports have noted submandibular calculi that passed out of the duct spontaneously, 1–2 days following stimulation. These were successful results of initial management protocols that included instructing the child to suck on a sour lemon or orange candy to stimulate salivary flow. In adults, the submandibular gland also leads to substantial results of initial management protocols that included instructing the child to suck on a sour lemon or orange candy to stimulate salivary flow. In adults, the submandibular gland also leads to substantial.

No spontaneous passage of calculi has been reported from Stenson’s duct. This may be because occurrence is less frequent, however, because Stenson’s duct perforates the buccinator muscle, it may be anatomically difficult for stones to pass spontaneously. During treatment planning, the fact that submandibular sialoliths in children have been reported to pass spontaneously following stimulation of salivary secretion should be taken into consideration, especially in cases in which the surgical alternative is a sialadenectomy.

Finally, if the sialolith is located in the anterior third of Wharton’s duct, antibiotic treatment should be given first to treat the acute symptoms and possibly to stimulate a spontaneous migration out through the duct.

References