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Sialolithiasis in the sublingual gland

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Abstract
Sialolithiasis is a major cause of salivary gland dysfunction. The submandibular gland is the most common site followed by the parotid gland. The sublingual gland and minor glands are very rare sites for stone formation. This paper describes a case of multiple sialoliths arising in the sublingual gland. They presented on the right floor of the mouth. The sublingual gland and sialoliths were completely removed with careful preservation of the lingual nerve and Wharton's duct. This was an uncommon sialolithiasis of the sublingual gland in a 14-year-old female.

Key words: Child; Salivary Gland Calculius; Sublingual Gland

Introduction

Sialolithiasis is a common finding and occurs mainly in the submandibular gland (80–90 per cent), and to a lesser degree in the parotid gland (five to 20 per cent). The sublingual gland and the minor salivary glands are very rarely affected.[1] The incidence of sublingual stones has been reported as zero to 6.4 per cent since 1950s.[1–7] The higher incidence (6.4 per cent) of sublingual stones in some reports[2,3] may be related to confusion with stones situated in the anterior portion of Wharton's duct. Minor salivary glands, when involved, are usually in the buccal mucosa or upper lip, forming a firm nodule that may mimic a tumour.[8]

The high incidence of submandibular sialolithiasis may be explained by the pH, mucin content and the high calcium concentration in this gland. The length and route around the mylohyoid muscle and secretion against gravity may also contribute to this predilection.[9] The likely mechanism of stone formation in the sublingual gland and minor glands is mechanical trauma with mucus extravasation, which serves as a nidus for stone formation. Many theories have been put forward to explain salivary calculi formation, such as calcification around foreign bodies.

Demographically, sialolithiasis shows a male predominance in most studies.[2–7] Sialolithiasis in children is rare,[10] although all ages may be afflicted and patients in their third to sixth decade represent the majority of cases.[1] Sialoliths in patients as young as 16 months have been noted.[10] Children presenting with sialoliths have an average age of around 10-years-old and a strong male preponderance for all salivary glands.[11]

This is a report of a case of sublingual sialolithiasis in a 14-year-old with a brief review of the literature.

Case report

A 14-year-old female presented at our hospital with a one year history of intermittent painful swelling of the right floor of mouth upon eating and aggravation of swelling of one month duration. She did not complain of any other symptoms such as dyspnoea or dysphagia. The right sublingual area showed an infected swelling and a hard mass with tenderness was noted. The submandibular gland was not enlarged on bimanual palpation. No other lymphadenopathy was found on physical examination.

A contrast-enhanced computed tomography CT scan showed diffuse swelling and three sialoliths of the right sublingual area (Figure 1). The sialoliths were located

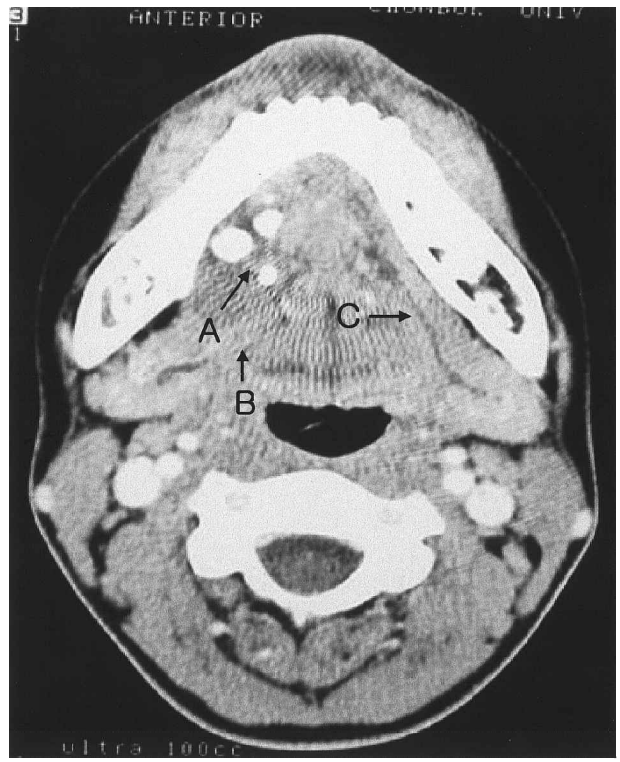

Fig. 1

Axial contrast-enhanced CT scan showing multiple sialoliths (A), partial obliteration of right Wharton's duct (B) and patent left Wharton's duct. Right sublingual space shows oedematous swelling.

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FIG. 2
Intra-operative findings showing multiple stones in the sublingual gland.

anterior to the submandibular gland and lateral to the Wharton's duct. The right submandibular duct was well identified, medially displaced and partially obliterated, and the hilum was well preserved. The left submandibular duct and hilum were also well preserved. These findings were compatible to a sialolithiasis arising in the sublingual gland. No evidence of lymphadenopathy was found on computerized tomography.

The removal of the sialoliths and sublingual gland was performed through a transoral approach. The sublingual gland and sialoliths were dissected as carefully as possible to preserve the lingual nerve and Wharton's duct. At operation, multiple round sialoliths, were found in the sublingual gland with capsule formation (Figure 2). The diameter of the largest one was 0.9 cm. It was a white surfaced, solid mass (Figure 3). The Wharton's duct and lingual nerve were well preserved (Figure 4).

Following surgery the patient remained well and was discharged on the fifth post-operative day. A follow-up examination showed no recurrence after 17 months.

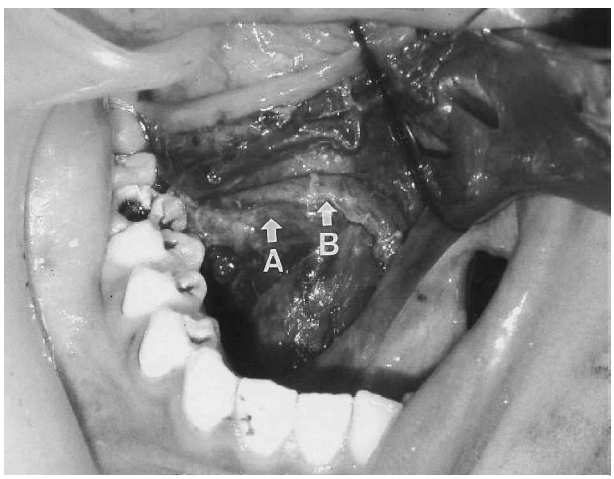

FIG. 3
After removal of sublingual gland and stones, the lingual nerve (A) and Wharton's duct (B) are well preserved.

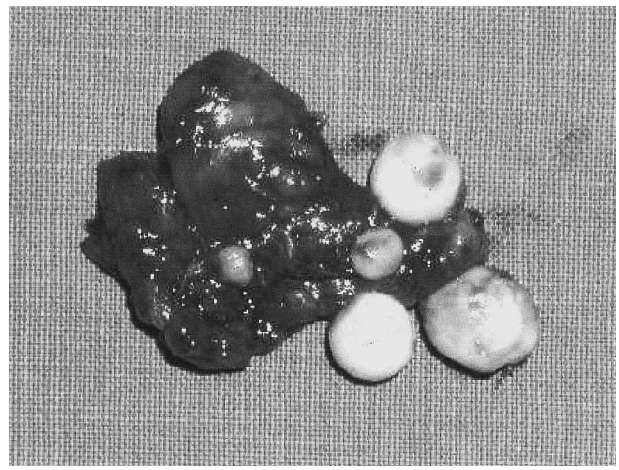

FIG. 4
Resected specimen of the sublingual gland and multiple sialoliths.

- **This is a case report of a patient presenting with calculi in the sublingual gland**
- **These were removed using a trans-oral route and the paper describes this method of removal**

Discussion

Salivary glands contain small concretions called microliths in the intraglandular ducts. These microliths induce microobstruction and result in chronic sialadenitis. The intermittent stasis of saliva changes the mucoid element and results in stone formation. The natural history of sialolithiasis is to cause obstruction of the salivary flow and to lead to bacterial ascent into the parenchyma of the gland. Long-standing obstruction by a sialolith can lead to damage to the acini of the gland and atrophy of the gland with a resultant loss of secretory function and ultimately fibrosis.[12]

The submandibular gland system, when compared with parotid and sublingual glands, is more susceptible to sialolith formation because of its unique physiological characteristics, its contents are more alkaline and have a higher concentration of calcium and phosphate. However, the cause of sialolith formation is still largely unexplained and individual susceptibility to sialolith formation is still a mystery. Calcium and phosphate concentrations in saliva are very important, but not the most critical factors in determining individual susceptibility to the formation of a sialolith. The elevation of protein and urea content in saliva are also important. Some children may exhibit an unusually high salivary calcium concentration, mostly in the submandibular gland and uncommon in the parotid and sublingual gland. This case report might be related to an unusually high salivary calcium concentration in the sublingual gland.

Most sialolithiasis of the sublingual gland presents with a painful swelling or in some cases with a painless swelling. Patients may be afflicted with a recurrent salivary colic and spasmodic pains upon eating. However, not all patients with stones will be symptomatic. This depends on the relative obstruction of salivary outflow. The patient may have repeated infections as well as abscess formation. Salivary stones also may be discovered incidentally on dental radiographs or during routine examination.

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The size of the sialolith varies from a small grain to giant sialoliths. A stone of 55 mm in length is the largest reported in the submandibular gland.[13] Single salivary stones are found in 70 to 80 per cent of cases and multiple in the remaining cases, with approximately five per cent of patients having three or more stones.[1] Sialoliths in the sublingual gland are usually round or oval shaped. However, stones in Wharton's duct may be elongated. Parotid stones are usually smaller and more often multiple. With regard to stone location, parotid stones are found to be in the hilum or parenchyma of the gland approximately 50 per cent of the time. Submandibular stones most commonly are found in the duct (75 to 85 per cent).[14] Submandibular stones close to the hilum of the gland tend to be larger and may become symptomatic. Patients with multiple stones may have stones located in different positions along the duct and gland. Their surfaces may be smooth or irregular.

Sublingual stones are treated surgically by complete sialadenectomy through a transoral approach. However, in the submandibular gland the most appropriate mode of treatment is primarily dependent on the location of the stone and may be, transoral siallithotomy or extraoral sialadenectomy. Radiograph or sialogram information can be helpful in planning treatment for these patients. Transoral removal of the sublingual gland usually requires a general anaesthetic. The floor of the mouth is incised from the orifice of Wharton's duct to the lingual aspect of the third molar. The submandibular duct is dissected out, and the lingual nerve is identified and protected. The duct was isolated along with the lingual nerve. The lingual nerve can be seen through the distal part of the incision passing forwards and across medially under the submandibular duct.[15]

In summary, this was an uncommon case of sialolithiasis of the sublingual gland presented in a 14-year-old female. The sialoliths in the sublingual gland were multiple, round and smooth surfaced. The sublingual gland and sialoliths were dissected and removed totally with isolation of Wharton's duct and preservation of lingual nerve.

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