Case Report

Bilateral hilar sialoliths in a child: A rare occurrence

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Abstract

Salivary sialolithiasis is a well-known cause for obstructive disease of the submandibular and parotid glands. However, the condition frequently occurs unilaterally, and it is uncommon to find a patient reporting with stones in both submandibular or parotid glands. Children below the age of 16 years rarely suffer from salivary stones. Thus, bilateral sialoliths in a child are extremely rare, with only four previous cases been reported in the literature. This is an additional case report of bilateral submandibular sialolithiasis occurring in the hilar area in a 13-year-old boy.

Keywords: Bilateral salivary stones, pediatric, sialolithiasis, transoral sialolithotomy

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INTRODUCTION

The incidence of sialolithiasis peaks in the third to sixth decades of life. Approximately 80%–90% of sialoliths occur in the submandibular gland, 5%–15% occur in the parotid gland and the remainder occurs in the sublingual and minor salivary glands.[1] Bilateral sialolithiasis, however, is a rare condition accounting for <3% of all cases according to several reports.[2]

In pediatric populations, sialolithiasis remains a rare cause of salivary dysfunction accounting for just 3% of all cases.[1] Consequently, in children, bilateral sialolithiasis is an extremely uncommon occurrence. Till date, only four cases have been reported in the English language literature. We report the fifth case of pediatric bilateral sialoliths present in the hilar region of both submandibular glands in a child.

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CASE REPORT

A 13-year-old male patient reported to the Department of Oral and Maxillofacial Surgery with the chief complaint of recurrent swelling of the right submandibular gland for 2 months [Figure 1]. The swelling would become noticeable during meal times. It was not associated with any significant pain, fever or pus discharge. Past medical history revealed palatoplasty for cleft palate 5 years ago. On examination, the right submandibular gland was noticeably swollen causing facial asymmetry. It was firm and tender to palpation. A small but discrete calculus was felt in the floor of the mouth in the region of the hilum of the right gland. There was no flow of saliva from the orifice of the Wharton's duct on the right side, whereas on the left it was normal.

The presence of the sialolith was confirmed on ultrasound examination which was performed only on the right side.

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A subsequent noncontrast computed tomography scan revealed two sialoliths, one in each submandibular gland. The one on the right was 5.5 mm × 3.5 mm, whereas the one on the left was 2 mm × 2 mm in size [Figure 2].

Since the right sialolith in the hilum of the gland was causing obstructive symptoms, it was decided to remove it transorally. Under general anesthesia, the right submandibular duct was exposed in the floor of the mouth and followed proximally to reveal the stone lying in the hilum of the gland. An incision over the stone, readily allowed it to be shelled out [Figure 3]. After thorough irrigation, only the anterior part of the mucosal incision was sutured loosely. No attempt was made to perform a sialodochoplasty. The stone in the left gland was managed conservatively as it was small and asymptomatic. As and when it causes obstructive symptoms, decision will be taken to remove it.

On follow-up at 7 days, the submandibular swelling had completely subsided and the intraoral wound had healed satisfactorily. The patient has been asked to stimulate saliva flow by sucking Vitamin C tablets or lemon slices and massage both glands gently to encourage salivary flow.

DISCUSSION

Sialoliths occur more commonly in the submandibular duct than in the parotid duct because the former is longer and located below the excretion port. These factors cause the saliva to move against gravity and undergo reversal at a right angle to the posterior border of the mylohyoid muscle, facilitating retention of saliva in the submandibular duct. In addition, high concentrations of calcium and phosphate in the saliva in the submandibular gland allow the formation of apatite. The saliva secreted from the submandibular gland is more alkaline and mucinous than that secreted from the parotid gland, and the rate of saliva movement is slow; thus, sialolithiasis occurs frequently in the submandibular gland.[1]

The prevalence of sialolithiasis is low in children because the condition requires a considerable amount of time to develop and the sublingual papillae and cross-sections of the salivary glands are very small, making invasion by foreign substances difficult.[3] In addition, concentrations of calcium and phosphorus in the saliva increase with age, facilitating sialolith formation in adults. In children, saliva flow is rapid; thus, most sialoliths are located distally. Due to the smaller size of these sialoliths, obstruction is short-lived and quickly results in the patient visiting the hospital.[4,5] Most calculi are relatively small (<1 cm, 93.1%) and located in the distal duct (62%).[5]

A review of all reports with fifty patients or more on sialolithiasis 1913–1989, reported bilateral sialolithiasis in 0.5%–2.2% of all salivary stones in all patients.[2] In children specifically, there are only four previously reported cases of bilateral sialolithiasis in the literature [Table 1].[6-9] The ages at which these were detected

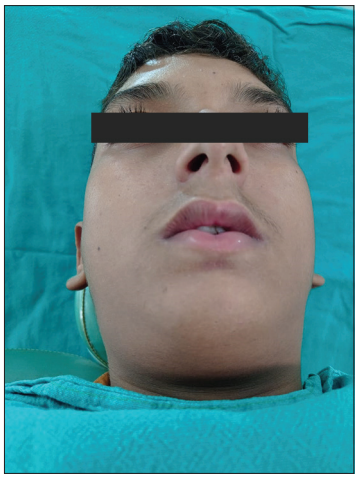

Figure 1: Preoperative view of patient showing the diffuse swelling in the right submandibular region

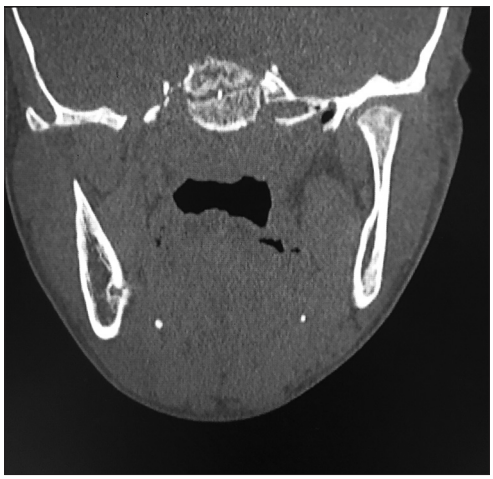

Figure 2: Coronal section showing calcifications in the submandibular gland hilar region bilaterally

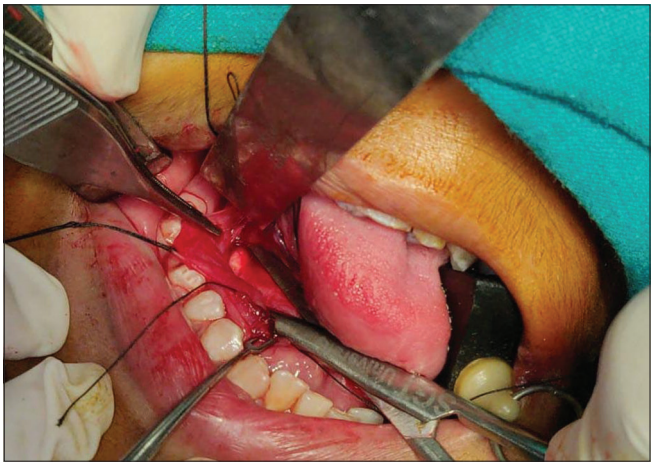

Figure 3: Intraoperative view showing the stone within the hilum

Table 1: Cases of pediatric bilateral sialoliths reported till date

Cases	Publication	Age	Sex	Symptom	Location	Treatment
1	Bodner and Fliss (1995)	16	Male	Pain	Right - Proximal duct Left - One in proximal and one in distal duct	Transoral sialolithotomy
2	Waseem and Forte (2005)	11	Female	Pain and swelling	Right - Proximal duct Left - Proximal duct	Conservative
3	Kim, Park, Son and Woo (2012)	15	Male	Postprandial bilateral swelling	Right - Hilum Left - Hilum	Transoral simultaneous bilateral sialolithotomy
4	Yoshii, Iwai, Sugiyama *et al*. (2017)	7	Male	Swelling L gland	Right - Hilum migrated to papilla Left - Duct-papilla	Transoral bilateral consecutive papillotomy
5	Present case (2020)	13	Male	Swelling R gland	Right - Hilum Left - Hilum	Transoral R. sialolithotomy

ranged from 7 years to 16 years and only one among them was a female child. Of the nine stones detected in these children, three occurred in the distal duct while six were located in the proximal part of the Wharton's duct. While three of these children underwent sialolithotomy, one was managed conservatively. None of them required removal of the submandibular salivary gland.

As noted above, children usually present early with symptoms of salivary obstruction. The fact that treatment is initiated promptly in the course of the disease as well as the inherent tendency toward repair and regeneration of tissues in children, it is reasonable to expect the gland to completely recover functionally after sialolithotomy. Even when the stone is located in the proximal duct, nowadays every attempt is made to remove it rather than subject the child to sialadenectomy as professed in the earlier days. Advanced imaging techniques and improved surgical understanding have made transoral removal of the stone safe and predictable. When small; stimulating saliva outflow and massage often cause the stone to either expel spontaneously or to travel from the proximal to the distal duct. Often it may lodge in the punctum, from where it can be retrieved easily.

When facilities are available, removal of submandibular stones through a transoral approach, in combination with the use of ultrasound and sialendoscopy will relieve symptoms in a majority of cases. In patients with symptomatic deep intraparenchymal submandibular stones, extracorporeal or intraductal lithotripsy and ultimately submandibulectomy are the options of choice.[10]

In our patient, the right stone was large enough to cause symptoms. Although lodged in the hilum of the gland, it was palpable intraorally, thus a transoral sialolithotomy was planned. This was performed successfully through an intraoral incision under G. A. The left stone, also located in the hilum being small and asymptomatic is being managed conservatively. The patient has been kept under long-term follow-up for any recurrences.

CONCLUSION

Bilateral sialolithiasis, itself an uncommon entity, is extremely rare in children. Along with our case, only five cases of pediatric bilateral salivary stones have been reported so far. Our patient was a 13-year-old boy with stones in the hilum of the submandibular gland bilaterally. The stone was removed from the right side through a transoral approach while the left one is being managed conservatively.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient (s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initial s will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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