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Case Report

The submandibular sialolithiasis in an eight-year-old child: a case report

Rajkumari P. Khatri*

Dr. Rajkumari'S ENT and Head and Neck Oncology Centre, Indore, Madhya Pradesh, India

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***Correspondence:**
Dr. Rajkumari P. Khatri,
E-mail: raj.sachdeva@rediffmail.com

ABSTRACT

Sialolithiasis is a commonly encountered disease of the salivary glands and represents up to 30-50% of all salivary gland diseases. However, the condition is rarely encountered in the paediatric population. The formation of a salivary stone is believed to be secondary to the deposition of calcium salts around a nidus, which is commonly associated with desquamated epithelial tissue or sloughing from a recent bacterial infection. Patients with submandibular sialolithiasis usually present with acute swelling over the neck associated with pain, fever, and purulent intraoral discharge. The calculi were located on the right in 100.0% of paediatric patients. Moreover, 20.0% of paediatric patients exhibited sialoliths in the glands. In this paper, we present the case of an 8-year-old male child who presented with a history of swelling over the right submandibular region since four months and had not responded to the conservative treatment given for the last four months. On examination, a small, rounded salivary calculus was found at the Wharton duct orifice near the papilla. A minimally invasive surgical procedure of intraoral excision of the calculus was done, and the patient was prevented from undergoing surgical excision of the gland. A large number of relatively small and distal sialoliths in paediatric patients can be removed using intraoral retrieval under local or general anesthesia. The clinical evaluation of the entire salivary tract with a high index of suspicion helps in early diagnosis and avoids the development of an abscess and other complications that may require surgical excision of the gland.

Keywords: Sialolithiasis, Salivary gland calculi, Paediatric sialolithiasis, Stone in the salivary duct, Submandibular gland, Paediatric sialoadenitis

INTRODUCTION

Sialolithiasis is commonly seen in the adult population, representing up to 30% of all salivary gland diseases.[1-3] However, the prevalence of paediatric sialolithiasis is rare, accounting for approximately 3-13.8 % of all sialolithiasis cases.[2-6] Some of the most important factors contributing to stone formation are salivary stasis, ductal inflammation, stenosis, and injury.[4,7] A sialolith is a calcareous concretion that may occur in the ducts of the major or minor salivary glands or within the glands themselves. This condition can lead to inflammation, bacterial infection, and an abscess.[8] The duration of symptoms in paediatric patients was shorter than that in adult patients (1.0 months verses 11.5 months). The most common chief complaints in both groups were swelling, followed by pain. A single calculus was more common in paediatric patients (85.7%) than in adult patients (65.1%). The calculi were located in the submandibular gland in 20.0% of paediatric patients and 35.5% of adult patients. The calculi were 5 mm in diameter in 100% of the paediatric patients. The calculi were retrieved intraorally under LA in 80% of cases and by spontaneous expulsion from the duct in 20% of cases.[9] Children with sialolithiasis are generally healthy without systemic illness except for symptoms of acute inflammation; most of their chief complaints consist of

intermittent unilateral pain and swelling in the submandibular region, usually associated with meals.[5]

Sialolithiasis can occur equally on the right or left side at any age, but it is more common in middle-aged adults, with a peak incidence between the fourth and sixth decades, and rare in children.[6,10] The calculi were located on the right in 100.0% of paediatric patients. It has been shown that the long upward path of the submandibular duct (Wharton's duct) is the most affected by calcifications.[10,11]

The unilateral swelling in the submandibular region should raise the question of obstructive pathologies such as sialolithiasis and stenosis. The diagnosis of salivary stones in children is difficult because the lower limit of radiologic detection of salivary stones is 2 mm in diameter, a size sufficient to cause a ductal obstruction.[6]

In sialolithiasis, the calcareous deposits within a salivary gland or its excretory duct cause obstruction to normal salivary flow. Consequently, the mechanical obstruction causes stasis of salivary flow and the formation of sialectasis, which predisposes to retrograde infection from the oral cavity in the gland.[12] The formation of a salivary calculus is believed to be secondary to the deposition of calcium salts around a nidus, which can be caused by desquamated epithelial or sloughing from a recent bacterial infection.[13]

Sialolithiasis is an important diagnosis to exclude in children with recurrent salivary gland infections, as it is a treatable condition. Obstructions of the salivary ducts may lead to inflammation from destructive salivary enzymes and a superimposed bacterial infection, or in rare cases, abscess formation. Early diagnosis and prompt treatment are vital to reducing the morbidity of such patients, especially in the paediatric age group. We presented a rare case of paediatric sialolithiasis in an 8-year-old boy. We will discuss the clinical presentation and, particularly, the surgical management after the failure of medical treatment prevented the surgical excision of the gland, emphasising the need for a complete clinical evaluation of the salivary tract in cases of salivary gland swelling.[1]

CASE REPORT

An 8-year-old boy presented with chief complaints of recurrent pain and swelling in the right submandibular region, which have occurred over a period of 4 months (Figure 1).

The swelling increased in size with the meals, and there was no history of pain or fever. He has not responded to the conservative treatment with antibiotics, analgesics, and anti-inflammatory agents given by different family physicians for the last 4 months. The past history was unremarkable. The general and systemic examinations were normal, and all vitals were stable. The right submandibular region had localised swelling that was approximately 43 cm in size, and the right submandibular gland was bimanually palpable with mild tenderness. Intraoral examination revealed a small greyish-white stone at the orifice of the duct at the sublingual papilla (Figure 2). No salivary flow was produced by gentle manipulation of the gland.

On investigations, the T and D count revealed total count 8900/cumm and differential count P-52, L-42 , E-3, M -3, rest all blood tests were normal. The ultasonography of the right submandibular region revealed an enlarged gland only, no calculi was seen. Under the coverage of antibiotics and analgesic and anti-inflammatory drugs, intraoral removal of salivary duct stones was done under GA by giving a small incision over the calculi. After the stone was removed, the salivary flow was restored to normal.

The patient had an uncomplicated recovery, and the swelling in the right submandibular region reduced in size within a week. Gradually, over a period of 3 months, he was relieved of the swelling completely, and there were no signs or symptoms of recurrence.

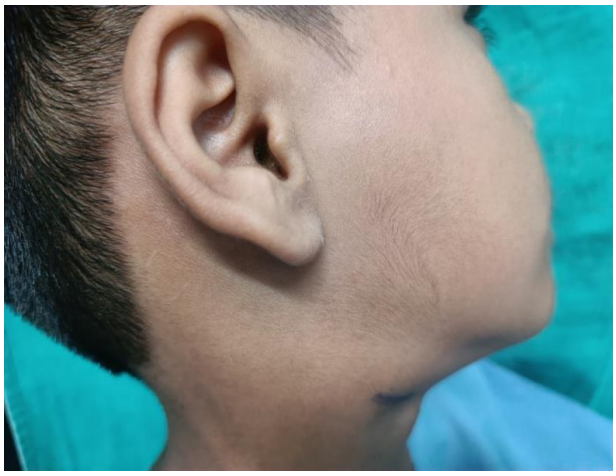

Figure 1: Revealing swelling in the submandibular region due to enlarged submandibular gland.

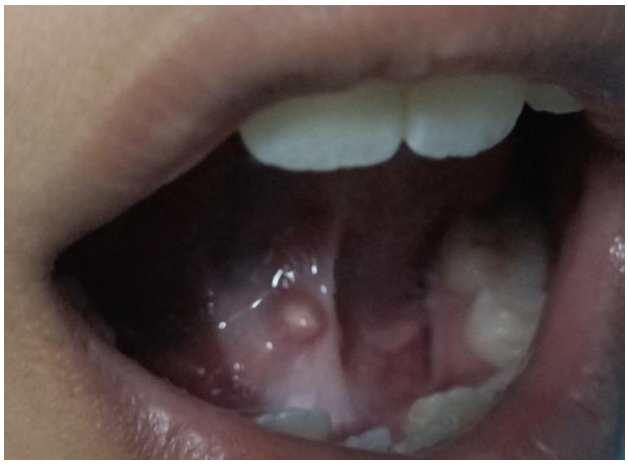

Figure 2: Revealing the calculi at the orifice of the Wharton's duct.

DISCUSSION

Sialolithiasis accounts for 50% of the major salivary gland diseases localised to the head and neck region. Although sialolithiasis is a relatively common disease in adults, it is rare in paediatric cases, with <150 cases reported in the literature.[5,9] Males are more frequently affected than female in paediatric patients.[14,15] This study also showed the involvement of the right side (100%), these differences in the sex and side distribution are unclear.

Sialoliths are clinically round or ovoid, ranging from 1 mm to 1 cm, can be rough or smooth, and are usually unilateral and yellowish in colour.[16] The majority of the calculi in the paediatric populations were <5 mm in diameter, Ikuri et al reported a case of formation of a calculus >10 mm in diameter that was located in the wharton's duct on the right side. The sialolith generally contains calcium phosphate, proteins, and calcium carbonate. In adult cases, the ratio of calcium phosphate is higher than in paediatric cases.[5]

The exact pathogenesis of sialolithiasis is unknown, and various hypotheses have been proposed. One hypothesis states that an initial organic nidus may occur that progressively grows because of the deposition of organic and inorganic 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 duct may cause sialolithiasis.[5,17]

Im et al 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, forming the outer lamellae (layers).[18] The exact ratios of inorganic material found within a sialolith depend on the chemical environment in which it forms.[18]

Approximately 85% of sialoliths occur in the submandibular gland, which is located on the floor of the mouth near the lingual frenulum close to the orifice or in the anterior 2/3 of the wharton's duct.[4,14,17,19]

This occurrence may be related to (1) the longer and larger calibre duct and slower salivary flow rate; (2) the saliva flows against gravity; (3) the presence of more alkaline saliva, which facilitates the precipitation of inorganic salts; (4) the high mucin content of the saliva, which is more viscous, results in a more stagnant flow of secretions in the submandibular gland; and (5) the high calcium and phosphorous concentration of the submandibular saliva.

Lower rates have been reported in the parotid 5%-10%, sublingual 5%, and minor salivary glands <1%.[2,5,14,20] Moreover, the Wharton duct is larger than other sublingual ducts. In general, the etiologic factors associated with the formation of a salivary stone 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.[21]

Marchal et al proposed a retrograde theory for sialolithiasis formation that involves the retrograde migration of food particles, foreign bodies, or pathogens ascending from the oral cavity into the salivary ductal system, which subsequently acts as a nidus for further calcification resulting in salivary stone formation.[17]

However, the reason for the low incidence of these diseases in children may be due to the relatively long time required for the formation of a salivary calculus, faster salivary flow rate, low concentration of calcium and phosphate in saliva, and because sublingual papillae and salivary glands are very small in children, rendering it difficult for foreign matter to enter.[5,14,19] According to McCullom et al, salivary calculi form more easily in adults because calcium and phosphate concentrations increase with age.[22]

The duration of symptoms was significantly shorter in paediatric patients (0-3 months, mean 1 month). This could be attributed to a lower tolerance for uncomfortable symptoms among the patients or the parents. The high salivary flow in paediatric patients may result in more severe symptoms among them.[3] Patients with sialolithiasis have a variety of symptoms, the most common of which are unilateral swelling of the salivary gland, decreased saliva production, and the acute onset of pain that worsens when eating.[19] In the present case, the child presented with unilateral swelling in the neck region associated with mild pain, there was no fever as he had been under the cover of antibiotics and analgesics off and on for 4 months.

There are considerable clinical differences in sialolithiasis between children and adults. The duration of symptoms seems to be shorter in paediatric 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 paediatric sialolithiasis mirrors that found in adults.[23,24] Moreover, salivary gland infections related to sialoliths do not appear to recur in paediatric patients.[23]

It is also important to perform a careful evaluation of the functioning and anatomy of the gland and duct involved. A palpable stone is found during bimanual examination in most cases. As a complement to the physical examination, there are a variety of radiographic tests for the diagnosis of sialolithiasis, including ultrasound, computed tomography (CT), magnetic resonance imaging (MRI), and sialoendoscopy.[21,25]

In the present case, the physical examination was sufficient to establish the treatment strategy. The

management of sialolithiasis should begin with conservative measures, such as massaging of the salivary gland, stimulation of the gland by eating acidic foods to stimulate the salivary flow, non-steroidal analgesic anti-inflammatory drugs, and sialogogues like lemon or orange candy. Good hydration, frequent hot compresses, and antibiotics. As a consequence, the stone is often spontaneously expelled, especially when it is superficial and mobile.[13,22,23] The smaller stones with a dimension less than 2 mm may pass out spontaneously in adults. However, spontaneous passing may be difficult in children due to the smaller ductal opening.[13]

If conservative management is unsuccessful, several different treatment methods have been proposed for sialolithiasis based on the size, number, and location of the sialolith. Approximately 40% of all submandibular stones are found in the distal portion of the Wharton duct or at the orifice, can be removed by a simple, minimally invasive, intraoral procedure. 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.[26] In the present case report, as the swelling of the gland was further increased in size following conservative management and there was no spontaneous expulsion of the stone, the decision was taken to remove the stone intraorally from the duct orifice.[5] Sialodenectomy is rarely necessary for the treatment of extensive proximal stone and in cases where there is no improvement after years of obstruction and recurrent inflammation but carrries the risk of damage to the lingual nerve of 1% substantial reduction of the non-stimulated salivary flow, which may affect oral hygiene, risk of the development of dental caries, xerostomia, and the risk of scar formation, alteration in the skin sensation and injury to the gustatory nerves, and transient functional disturbance of the marginal mandibular branch of the facial nerve, which are most often encountered in up to 12.5% of cases, while permanent lesions have been reported in 7% of these cases.[6,8,28]

Most sialoliths were removed using a surgical procedure, but in a few cases, they spontaneously migrated out.[5] Sialoendoscopy is another successful treatment for submandibular ductal stones because it reduces the risk of missed stones, nerve injury, and the requirement for sialoadenectomy.[27] Although we did not use this method, because of non-availability of instruments, high cost, future studies should focus on this. Extracorporeal shock wave lithotripsy of salivary calculi entails a risk of damaging the salivary gland, the ear, dental amalgams, or the central nervous system.[28]

Recently, several minimally invasive procedures have been introduced for the management of sialolithiasis. Such as extracorporeal sonography and intracorporeal endoscopically controlled lithotripsy, which have completely changed the therapeutic methods utilized.

Various techniques of sialodochotomy have been described in the literature, but there is an associated risk of injury to the lingual nerve, which passes in close proximity to the Wharton duct.[26]

CONCLUSION

Although sialolithiasis is the most common benign cause of salivary gland swelling, it is uncommon in children and should not be overlooked in the differential diagnosis of paediatric patients with submandibular swelling and pain. A high index of suspicion should be maintained, as a delayed diagnosis may cause significant discomfort and morbidity to the patient. This article presented the management of the obstruction of a submandibular duct in an 8-year-old male child using a simple, effective, minimally invasive surgical intervention. In paediatric patients with a large number of relatively small and distal calculi, intraoral removal of sialoliths is recommended as the treatment of choice. If the sialolith is located in the anterior 1/3rd of the Wharton's duct, initial treatment with antibiotics and sialogogues should be given to treat the acute symptoms and possibly to stimulate a spontaneous migration out of the duct; the decision to excise the gland should be balanced against the risk of excision.

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REFERENCES

1. Thong HK, Mohamad MH, Sabir HAPP, Tengku KTMI. Recurrent Submandibular Sialolithiasis in a Child. Cureus. 2020;12(12):e12163.
2. Chung MK, Jeong HS, Ko MH, Cho HJ, Ryu NG, Cho DY, et al. Pediatric sialolithiasis: what is different from adult sialolithiasis? Int J Pediatr Otorhinolaryngol. 2007;71(5):787-91.
3. Lustmann J, Regev E, Melamed Y. Sialolithiasis. A survey on 245 patients and a review of the literature. Int J Oral Maxillofac Surg. 1990;19(3):135-8.
4. Bhavesh D, Trivedi BD. Surgical removal of submandibular gland sialolithiasis in a 9-year-old girl: a case report. Pediat Dent J. 2014;24:111-4.
5. Konishi I, Fukumoti F, Yamada A, Nonaka K, Fujiwara T. Large sialolith in the submandibular gland of a child. Pediatr Dental J. 2005;15(1):143-6.
6. Faure F, Querin S, Dulguerov P, Froehlich P, Disant F, Marchal F. Pediatric salivary gland obstructive swelling: sialendoscopic approach. Laryngoscope. 2007;117(8):1364-7.

7. Flint PW, Haughey BH, Robbins KT, Lund VJ, Niparko JK, Richardson MA, et al. Cummings otolaryngology—head and neck surgery. 5th ed. Philadelphia: Elsevier Health Sciences; 2005: 1151-61.
8. Alves MA, Veiga LC, Coelho EMRB, Kramer PF, Rodrigues PH, Joo BB. Weber An Unusual Case of Submandibular Sialolithiasis in Early Childhood: A Case Rep Open J Stomatol. 2022;12:347-54.
9. Inui A, Itou R, Oyama T, Tamura Y, Kubota K, Kobayashi W. Comparison of sialolithiasis in paediatric and adult patients. Oral Sci Int. 2017;14:37-9.
10. Shinohara Y, Hiromatsu T, Nagata Y, Uchida A, Nakashima T, Kikuta T. Sialolithiasis in children: report of four cases. Dentomaxillofac Radiol. 1996;25(1):48-50.
11. Wilson KF, Meier JD, Ward PD. Salivary gland disorders. Am Fam Physician. 2014;89(11):882-8.
12. Lustmann J, Regev E, Melamed Y. Sialolithiasis. A survey on 245 patients and a review of the literature. Int J Oral Maxillofac Surg. 1990;19(3):135-8.
13. Konishi I, Fukumoto S, Yamada A, Nonaka K, Fujiwara T. Large sialolith in the submandibular gland of a child. Pediatr Dent J. 2005;15:143-6.
14. Bodner L, Fliss DM. Parotid and submandibular calculi in children. Int J Pediatr Otorhinolaryngol. 1995;31(1):35-42.
15. Nahlieli O, Eliav E, Hasson O, Zagury A, Baruchin AM. Pediatric sialolithiasis. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 2000;90(6):709-12.
16. Marwaha M, Nanda KD. Sialolithiasis in a 10 year old child. Indian J Dent Res. 2012;23(4):546-9.
17. Marchal F, Kurt AM, Dulguerov P, Lehmann W. Retrograde theory in sialolithiasis formation. Arch Otolaryngol Head Neck Surg. 2001;127(1):66-8.
18. Im YG, Kook MS, Kim BG, Kim JH, Park YJ, Song HJ. Characterization of a submandibular gland sialolith: micromorphology, crystalline structure, and chemical compositions. Oral Surg Oral Med Oral Pathol Oral Radiol. 2017;124(1):13-20.
19. Francis CL, Larsen CG. Pediatric sialadenitis. Otolaryngol Clin North Am. 2014;47(5):763-78.
20. Bodner L. Salivary gland calculi: diagnostic imaging and surgical management. Compendium. 1993;14(5):572.
21. Binar M, Gokgoz MC, Aydin U, Yavan I, Karahatay S. Chronic sialadenitis due to the stone inside the accessory duct of submandibular gland. Surg Radiol Anat. 2017;39(10):1165-8.
22. McCullom C, Lee CY, Blaustein DI. Sialolithiasis in an 8-year-old child: case report. Pediatr Dent. 1991;13(4):231-3.
23. Won SJ, Lee E, Kim HJ, Oh HK, Jeong HS. Pediatric sialolithiasis is not related to oral or oropharyngeal infection: A population-based case control study using the Korean National Health Insurance Database. Int J Pediatr Otorhinolaryngol. 2017;97:150-3.
24. Epker BN. Obstructive and inflammatory diseases of the major salivary glands. Oral Surg Oral Med Oral Pathol. 1972;33(1):2-27.
25. Hammett JT, Walker C. Sialolithiasis. Treasure Island: StatPearls Publishing; 2023.
26. Zenk J, Constantinidis J, Al-Kadah B, Iro H. Transoral removal of submandibular stones. Arch Otolaryngol Head Neck Surg. 2001;127(4):432-6.
27. Chu DW, Chow TL, Lim BH, Kwok SP. Endoscopic management of submandibular sialolithiasis. Surg Endosc. 2003;17(6):876-9.
28. Iro H, Schneider HT, Födra C, Waitz G, Nitsche N, Heinritz HH, Benninger J, Ell C. Shockwave lithotripsy of salivary duct stones. Lancet. 1992;339(8805):1333-6.

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