Uncommon Localization of a Sialolith in a Child's Frenum

Patricia Fernanda Dias, DDS, MSD Erika Calvano Kuchler, DDS, MSD Maria Elisa Janini, DDS, MSD, PhD Lucianne Cople Maia, DDS, MSD, PhD

ABSTRACT

Sialoliths or salivary calculi appear as hard nodules of unknown etiology and are usually located in the anterior portion of Wharton's duct (submandibular gland ducts), affecting mainly adult men. The purpose of this report was to describe an uncommon case of submandibular sialolith located in a child's lingual frenum and address the semiologic and semiotechnical aspects related to the case's diagnosis. (J Dent Child 2009;76:161-4)

Received January 29 2008; Last Revision April 14, 2008; Revision Accepted April 17, 2008.

Keywords: Salivary gland calculi, submandibular gland, oral diagnosis, child

S ialolith, or salivary calculus, is a calcified structure that develops in salivary glands or their ducts,^{1,2} appearing as a hard nodule located under a yellowish or white mucosa.² An increase in the salivary gland's size or discomfort—usually associated with meal times may accompany the sialolith.¹⁻⁴

The exact etiology is still unknown, but it is thought that sialoliths result from the deposition of calcium salts around organic debris accumulated in the duct lumen.^{2,5} Symptoms and clinical characteristics can be sufficient for establishing a diagnosis.⁶ Panoramic, periapical, and occlusal radiographs, however, are also crucial in this diagnostic process. Sialoliths are usually seen as radiopaque masses of variable sizes and equivalent to the quantity of calcified material.^{3,5-8} Among the treatment options, there are conservative approaches in which salivary glands are milked or moist heat is applied, the patient is instructed to ingest fluids, and the use of saliva-stimulating medications is prescribed.^{2,3,9-11} Furthermore, endoscopic laser lithotripsy and extracorporeal shock wave lithotripsy are commonly used.^{9,10,12,13} Intensive treatment consists of surgical incision in the salivary duct to remove the calculus.^{2,3,10,12,14-16}

There is a slight predominance of sialoliths in men,^{1,8,18} with it being relatively common in adults and rarely seen in children.^{1,2,8,13,15,16} The submandibular gland is involved in most cases,^{1,2,8,9,16} mainly in the anterior two thirds of Wharton's duct.^{1,3,5} Anatomically, this duct is long and has 2 sharp curves and a small ostium located in the buccal floor's anterior region.¹⁶ Anatomical alterations in both duct and ostium, however, were not addressed in the dental literature consulted.

The purpose of this paper was to report the uncommon case of a submandibular sialolith located in a child's lingual frenum and to indicate the diagnostic strategies.

Drs. Dias and Kuchler are masters in Pediatric Dentistry, Dr. Janini is associate professor, Department of Oral Pathology, and Dr. Maia is associate professor, Department of Paediatric Dentistry and Orthodontics, all in the School of Dentistry, Federal University of Rio de Janeiro, Rio de Janeiro, Brazil. Correspond with Dr. Maia at <u>rorefa@terra.com.br</u>

CASE REPORT

An 8-year-old boy was brought by his caregiver to the Department of Pediatric Dentistry of the School of Dentistry of Federal University of Rio de Janeiro, Rio de Janeiro, Brazil, for treatment of a "small ball" in the child's lingual frenum. During anamnesis, the child stated the lesion had appeared some months ago, with no history of trauma in the region or any other associated symptom. In addition, no relevant findings were found in the medical history.

During the clinical examination, a yellowish solid nodular lesion measuring approximately 4 x 3 mm was found on the left side of the lingual frenum, which was hypertrophied (Figure 1). The nodule shifted transversally along the frenum, depending on the tongue's movements, even "disappearing" (Figure 2a-c). Both frenum and nodule were restricted to the buccal floor when the patient kept his tongue at rest (Figure 2d). After milking the submandibular glands, it was observed that the ostia of Wharton's ducts were anatomically located in the middle portion of the lingual frenum, with reduced salivary flow on the left side.

During radiographic examination, a lower occlusal view was not helpful (Figure 3a). Therefore, a parallel



Figure 1. Frontal view of lingual frenum showing lesion on the left side.

periapical radiograph of the frenum was taken, revealing a well-defined radiopaque image (Figure 3b) that led to the diagnosis of a submandibular sialolith.

Because the calculus was small and had been present in the duct for some time without being spontaneously expelled, the treatment option was to remove it surgically under local anesthesia. The sialolith was removed (Figure 4) and submitted to histological analysis (Figure 5a and b).

The patient has been followed up every 3 months since then and no sign of recurrence was noted after 1 year (Figure 6).

DISCUSSION

According to the literature, the presence of sialoliths in children is rare. In the reported case, the patient had a submandibular gland sialolith. Calculi are more likely to form in this gland because of the high calcium and phosphate concentration and the high viscosity and alkalinity of the submandibular gland saliva. In addition, Wharton's duct is anatomically longer and sinuous, with a smaller ostium. All these factors contribute to a slow salivary flow^{1,2,5,14,16} against the force of gravity, which causes stagnation and potential calculus formation.¹⁶

The atypical anatomy of Wharton's duct seen in the present case—with its ostium located in a higher position than normal, namely, in the middle of the lingual frenum instead of on the buccal floor—contributed to the sialolith formation in this pediatric patient.

Uncommon cases like the one reported here require careful differential diagnosis. Because this lesion was located in the buccal floor's middle region when the tongue was at rest, the hypothesis of lymphoepithelial cyst, epidermoid cyst, and dermoid cyst^{18,19} had been raised, but subsequently ruled out because the nodule was solid. In the diagnostic semiology, Wharton's ducts were found to be atypically positioned in the lingual frenum's middle portion. Additionally, the reduced salivary flow on the side exhibiting the lesion raised the suspicion of an obstruction in keeping with a sialolith, which justified a radiographic examination.



Figure 2. (A) Lateral view of the lingual frenum, (B) Note the lesion dislocating transversally, C) until disappearing, (D) View of the tongue at rest. Note nodule on the buccal floor.

It should be emphasized, however, that the radiographic examination was adjusted because of the lesion's location. A periapical film was positioned parallel to the lingual frenum, which allowed the sialolith to be clearly visualized.



Figure 3. Lower occlusal radiograph suggesting normality (A) and parallel radiograph of the lingual frenum showing radiopaque mass (B).



Figure 4. Macroscopic image of sialolith removed with surgery.

Conservative measure was used to stimulate spontaneous elimination of the calculus because, although it was small, it had not been expelled for several months. As most submandibular sialoliths are located in duct portions that facilitate their removal through surgical procedures,¹⁰ this was the treatment choice. This technique, performed under local anesthesia by making a small incision to remove the calculus, is easy and requires only one appointment.^{2,10,12,14-16}



Figure 5. Microscopic images of sialolith. Note remaining organic debris surrounded by concentric layers of calcified material. Magnification of 10x (A) and 40x (B).



Figure 6. Frontal view of lingual frenum after 1-year follow-up with no recurrence.

CONCLUSION

Because sialoliths are not common in children, special attention should be paid to their characteristic signs and symptoms to achieve the correct diagnosis. It should be remembered that small anatomical variations in the location of the salivary ducts may cause sialolith formation in unusual sites. It should also be emphasized that early removal of the sialolith allows complete recovery and normal functioning of the affected gland.

REFERENCES

- 1. Di Felice R, Lombardi T. Submandibular sialolithiasis with concurrent sialoadenitis in a child. J Clin Pediatr Dent 1995;20:57-9.
- Neville BW, Damm DD, Allen CM, Bouquot JE. Oral and Maxillofacial Pathology. 2nd ed. Philadelphia, Pa: WB Saunders; 2002.
- 3. Karengera D, Yousefpour A, Sadeghi HM, Reychler H. Sialolithiasis in children as a diagnostic dilemma. Eur Arch Otorhinolaryngol 2000;257:161-3.
- 4. Leung AK, Choi MC, Wagner GA. Multiple sialoliths and a sialolith of unusual size in the submandibular duct: A case report. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 1999;87:331-3.
- 5. Soares LP, Gaião de Melo L, Pozza DH, Pinheiro ALB, Gerhardt de Oliveira, M. Submandibular gland sialolith in a renal transplant recipient: A case report. J Contemp Dent Pract 2005;6:127-33.
- 6. Shinohara Y, Hiromatsu T, Nagata Y, Uchida A, Nakashima T, Kikuta T. Sialolithiasis in children: Report of four cases. <u>Dentomaxillofac Radiol 1996</u>; 25:48-50.
- 7. Horie N, Shimoyama T, Tanebayashi Y, Ide F. Parotid sialolithiasis in a child. J <u>Clin Pediatr Dent</u> 1995;20:61-2.
- 8. Nahlieli O, Eliav E, Hasson O, Zagury A, Baruchin AM. Pediatric sialolithiasis. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2000;90:709-12.

- 9. Blasco JL SN, Alcaina MB, Perez SH, Padilla VS, Garcia-Algar O. Expulsion espontanea de un calculo salival submandibular. An Pediatr (Barc) 2003;59: 393-5.
- 10. Escudier MP, Drage NA. The management of sialolithiasis in 2 children through use of extracorporeal shock wave lithotripsy. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 1999;88:44-9.
- 11. Waseem Z, Forte V. An unusual case of bilateral submandibular sialolithiasis in a young female patient. Int J Pediatr Otorhinolaryngol 2005;69: 691-4.
- 12. McGurk M, Escudier MP, Brown JE. Modern management of salivary calculi. Br J Surg 2005;92: 107-12.
- Steiner M, Gould AR, Kushner GM, Weber R, Pesto A. Sialolithiasis of the submandibular gland in an 8-year-old child. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 1997;83:188.
- 14. Tosios K, Rallis G, Vallianatou D, Vlachodimitropoulos D. Yellow-white tumor on the floor of the mouth. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2006;101:701-4.
- 15. Escudier MP, McGurk M. Symptomatic sialoadenitis and sialolithiasis in the English population: An estimate of the cost of hospital treatment. <u>Br Dent J</u> 1999;186:463-6.
- Peterson LE, Hupp JR, Tucker MR. Contemporary Oral and Maxillofacial Surgery. 3rd ed. St. Louis, MO: Mosby; 1998.
- 17. Bodner L, Fliss DM. Parotid and submandibular calculi in children. Int J <u>Pediatr Otorhinolaryng</u>ol 1995;31:35-42.
- 18. Ho MW, Crean CS. Simultaneous occurrence of sublingual dermoid cyst and oral alimentary tract cyst in an infant: A case report and review of the literature. Int J Paediatr Dent 2003;13:441-6.
- 19. Mathews J, Lancaster J, O'Sullivan G. True lateral dermoid cyst of the floor of the mouth. J Laryngol Otol 2001;115:333-5.

Copyright of Journal of Dentistry for Children is the property of American Academy of Pediatric Dentistry and its content may not be copied or emailed to multiple sites or posted to a listserv without the copyright holder's express written permission. However, users may print, download, or email articles for individual use.