A Case of Sialolithiasis in a Child

Tomomichi Oka / Jouji Nomura / Yoshihiko Matsumura / Shigeaki Yanase / Takayuki Nagata / Shiori Uno / Toshiro Tagawa

Sialolithiasis is a comparatively rare disease in children. Here, we report the case of a female aged 5 years and 7 months old with sialolithiasis of the submandibular duct, and we examine the causal factors, diagnostic techniques and treatment methods for the disease based on a review of the literature.
J Clin Pediatr Dent 31(2):139-141, 2006

Downloaded from http://meridian.allenpress.com/jcpd/article-pdf/31/2/139/1748149/jcpd_31_2_k846l578232g26tr.pdf by Bharati Vidyapeeth Dental College & Hospital user on 25 June 2022

INTRODUCTION

Sialolithiasis is often observed in the oral region, and is caused by development of a calculus in the salivary gland or duct. This disease is mostly seen in adults or young adults, and seldom develops in children.[1-3] Here, we report a case of a 5-year-old female with sialolithiasis of the right submandibular duct, and we provide a literature review of childhood sialolithiasis.

CASE REPORT

A Japanese female child aged 5 years and 7 months, visited the Department of Oral and Maxillofacial Surgery at Mie University Hospital for a swelling of the right sublingual region. Approximately two months before the first visit, the patient visited a nearby dentist after her mother found painless swelling in the sublingual region. Antibiotics were administered by the dentist, but no improvement was observed. The patient subsequently developed a sense of discomfort during swallowing and therefore it was recommended that she should visit our department. A symmetrical facial configuration was confirmed, and no abnormalities in regional lymph nodes were observed. Intraorally, a diffuse swelling was found mainly in the right sublingual papilla, and a rice-grain sized yellowish white stone-like mass was palpable directly below the mucosa and located approximately 5 mm posterior to the sublingual papilla. There was no tenderness, and the patient had good salivary flow from the sublingual papilla (Fig. 1). No shadow was apparent on panoramic and occlusal radiographs, suggesting the absence of a salivary calculus (Fig. 2). However, based on clinical diagnosis of sialolithiasis of the right submandibular duct, sialolithotomy was performed intraorally under local anesthesia and a calculus was located in the duct approximately 5 mm posterior to the sublingual papilla. Removal of the mass as one clot was possible due to lack of adhesion, and the postoperative outcome was good with no abnormalities. The patient is currently under observation as an outpatient.

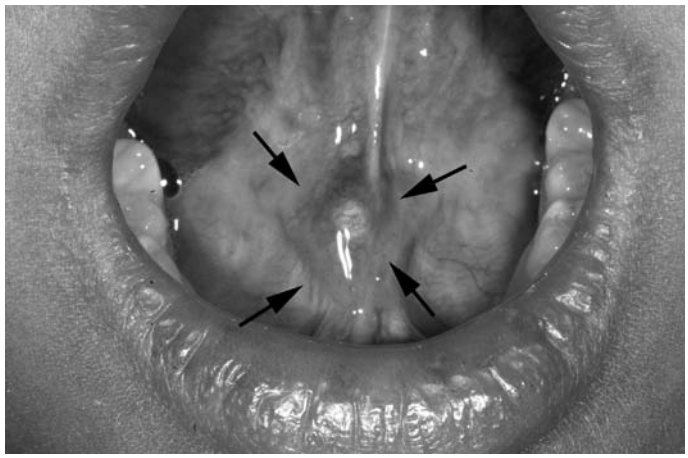

Figure 1: Intraoral findings. A rice grain-sized yellowish-white stone-like mass was revealed.

Figure 2: Dental X-ray findings. No calculus could be seen

The removed mass was a yellowish-white calculus of 2 x 1 mm in size. The mass was comparatively soft and was easily cut with a sur-

From the Department of Maxilofacial Surgery, Faculty of medicine, Mie University Hospital

Send all correspondence to: Tomomichi Oka, Department of Maxilofacial Surgery, Faculty of medicine, Mie University 2-174, Edobashi, Tsu, Mie 514-8507, Japan

Tel: +81-59-232-1111

Fax +81-59-231-5207

E-mail: okatomo@clin.medic.mie-u.

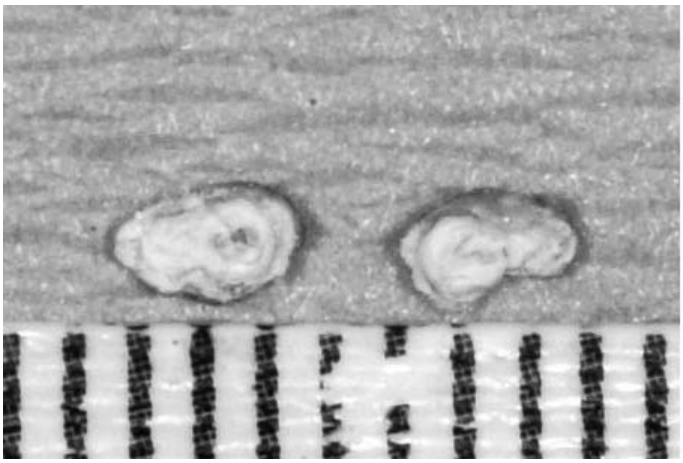

Figure 3: Removed calculus. A calculus of 2 x 1 mm in size was found, with a cross-sectional surface showing a layered structure.

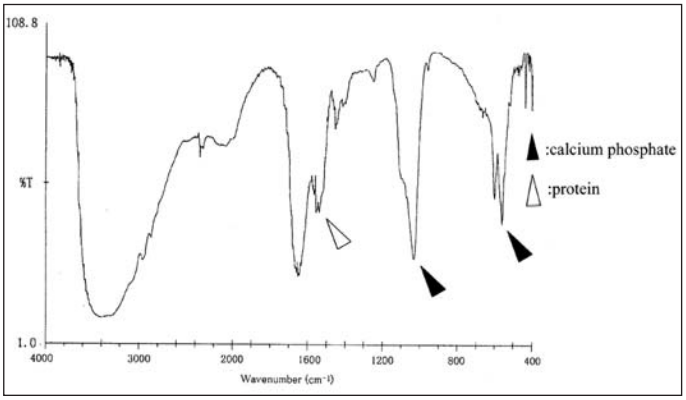

Figure 4: Analysis of the calculus. It contained 12% calcium phosphate and 88% protein by infrared absorption spectrophotome.

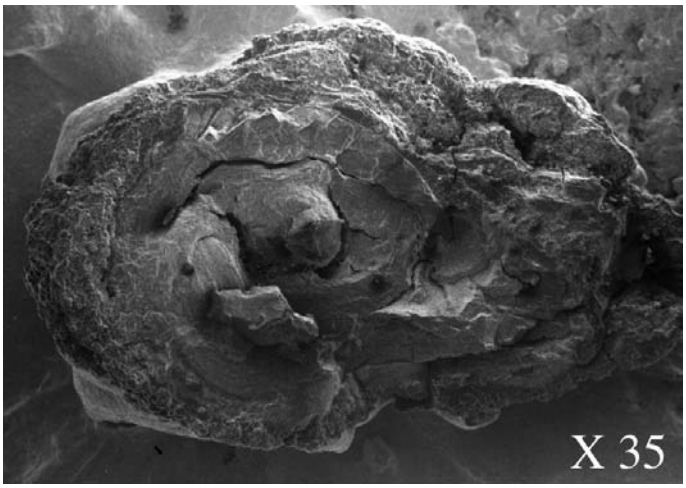

Figure 5: Scanning electron microscopy. No nucleus-like material in the central part of the calculus is showed.

gical knife, revealing a cross-sectional surface with a layered structure (Fig. 3). Infrared absorption spectrophotometry showed the mass to contain 12% calcium phosphate and 88% protein, and calcium and phosphorous were detected in elemental analysis of the calculus surface (Fig. 4). Scanning electron microscopy indicated a cobblestone appearance in the central part of the cross-sectional surface of the calculus, in addition to vorticose cracking, but nucleoids were not observed in the central part of the calculus (Fig. 5).

DISCUSSION

Sialolithiasis is a disease accompanied with development of a calculus in the salivary gland duct, and is often observed in patients aged 30–50 years old. Many cases of this disease have been observed in the submandibular gland due to the properties of saliva (calcium concentration and pH) and the long-curved duct.[1-3] However, there are only a few reports of this disease in children; we found only 29 cases (including our case) in patients aged 14 years old or younger in the English literature. Doku *et al.*[4] reported 11 cases of patients aged 14 years old or younger with sialolithiasis of the submandibular gland over a 50-year period, and Reuther *et al.*[5] reported 15 confirmed cases of the disease from 1898 to 1973. An additional report suggests that about 3% of cases of sialolithiasis develop in children.[3] The low onset rate in children may be due to the relatively long time required for formation of a calculus, since the sublingual papilla of the duct is extremely small, making it difficult for foreign material to enter. Furthermore, a calculus is formed more easily in adults than in children because the concentrations of calcium and phosphate ions in resting saliva increase with age[3,5]; however, the mechanistic details of calculus formation have not been determined.

Table 1: Age and Gender

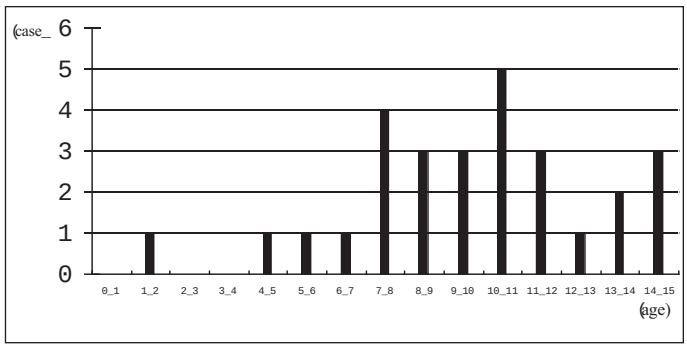

Table 2: 2 size and site

size	< 5mm	10(67.6%)
	≧ 5mm	5(33.3%)
site	submandibular duct	25(86.2%)
	submandibular gland	2(6.8%)
	parotic gland	2(6.8%)

In the literature cases of childhood sialolithiasis, the youngest patient was 13 months old, the oldest was 15 years old. The number of patients aged 10 years old was largest (5 cases, 17%), followed by 4 patients aged 7 years old (13%), (Table 1). Other reports show a similar age frame.[1,3,5]

In our study, 17 male children (58%) and 12 female children (41%) were included, showing a male-to-female ratio of 1.25:1 (Table 1), a larger number of male children has also been reported (3.5:11,5 and 3:13). The calculus size after removal was described in 15 of the 29 cases: 10 cases had a calculus smaller than 5 mm (67.7%), and 5 cases had a calculus of 5 mm or larger (33.3%)

Downloaded from http://meridian.allenpress.com/jcpd/article-pdf/31/2/139/1748149/jcpd_31_2_k846l578232g26tr.pdf by Bharati Vidyapeeth Dental College & Hospital user on 25 June 2022

(Table 2) which are similar to previous reports.[1,6-8] This may be because the time period required for children to develop clinical symptoms is shorter than adult patients.

Of the 29 literature cases, 25 (86.2%) had a calculus in the submandibular duct, 2 (6.8%) had a calculus in the parotid gland, and 2 (6.8%) had a calculus in the submandibular gland (Table 2). Other reports found 10 of 12 cases (83%) with a calculus in the submandibular duct, and 2 cases (16%) with a calculus in the parotid gland. Most cases of calculus development were observed near the sublingual papilla or in the front two-thirds of the submandibular duct, which may be due to a small calculus being able to move toward the sublingual papilla through the duct.

For diagnosis of sialolithiasis, X-ray examination is useful in addition to ocular inspection and manipulation. An X-ray examination of the salivary gland is the most reliable approach, but use of this method may cause a calculus located around the sublingual papilla to move backward. In addition, there is a possibility of onset of acute inflammation caused by the examination[6,8] and since a patient may have pain or discomfort upon injection of radiopaque dye, it may be better to avoid this procedure when possible. Normal X-ray examination is also useful, as shown by the approximately 90% detection rate for sialolithiasis in the submandibular gland.[3] The detection rate with an occlusal X-ray is greater compared to other methods such as P-A and orthopantomography.[2,6,8] However, when a calculus is located in the rear one-third of the duct, or in the gland, the calculus cannot be detected by the occlusal method, and other examinations such as CT, MRI and sonoradiography[11] are required. It should be noted that approximately 20% of sialolithiasis cases in children show X-ray translucency[12], as it occurred in our case. This is thought to be due to the relatively small size of the calculus as well as the insufficient calcification process.[3] In our case, an 88% protein content in the calculus was confirmed, showing an extremely low level of calcification. In such cases a CT, MRI, sonoradiography or X-ray examination of the salivary gland can be necessary. We also consider important to carefully perform manual palpation[8] in the lower intraoral region to enhance the effectiveness of diagnosis, since the calculus is located in the submandibular duct in most cases.

The calculus can be removed intraorally in many patients with sialolithiasis and there are few cases in which the gland itself has to be removed.[4] In addition, since some cases of spontaneous dislodgment have been reported, massage to the submandibular gland may be sufficient if the calculus is located near the sublingual papilla.[7] There are few cases in which removal of the entire gland is necessary, because most calculi are located in the duct; however, removal of the gland should be avoided whenever possible based on esthetics and age-related perspectives. Other means of calculus removal have been implemented such as endoscopic[11] and extracorporeal shock wave lithotripsy, modalities used in the management of renal calculi.[12,13]

REFERENCES

1. Bodner L Aza B. Submandibular Sialolithiasis in Children .J Oral Maxillofac Surg.; 40: 551-4. 1982
2. Felice R Lombardi T. Submandibular sialolithiasis with concurrent sialoadenitis in a child. J Clin Pediatr Dent.; 20: 57–59. 1995
3. Bodner L, Fliss D, Parotid and Submandibular calculi in children. J Pediatric Oto Rhino Laryngology.; 31: 35-42. 1995
4. Doku H, Berkman M. Submaxillary Calculus in Children . Amer J Dis Child; 114: 671-673. 1967
5. Reuther J, Hausamen J. Sialolithiasis der Glandula submandibularis im Kindesalter. Klin Padiat; 188: 285-288. 1976
6. Karengera D, A Yousefpour . Sialolothiasisnin children as a diagnostic dilemma Eur Arch Otorthinolaryngol; 257: 161-163. 2000
7. Walsh S, Robson W, Spontaneous of a submandibular salivary calculus in a child J Laryngologyband Otology; 102: 1052-1053. 1988
8. Zeeshan BA, Waseem Forte V. An unusual case of bilateral submandibular Sialolothiasis in a young female patient J Pediatric Otorhinolaryngolgy; 69: 691-694. 2005
9. Grunebaum M Mankuta J. Submaxillary Sialadenitis with a calculus in an infancy diagnosed by ultrasonography Pediatric Radiology 1985; 15: 191-192.
10. Cornell M Cameron Y David I. Sialolothiasis in an 8-year-old case report Pediatric Dentistry; 13: 231-233. 1991
11. Ottaviani F Capaccio P Campi M Ottavini. A Extracorporeal Electromagnetic Shockwave Lithotripsy for Salivary Gland Stone Laryngoscope; 106: 761-764. 1996
12. Williams M. Sialolithiasis Otolaryngol Clin North Am; 32: 819-834. 1999
13. Iro H Nitche N. Extracorporeal Shockwave Lithotripsy of Salivary Gland Stones Lancet; 2:115. 1989

Downloaded from http://meridian.allenpress.com/jcpd/article-pdf/31/2/139/1748149/jcpd_31_2_k846l578232g26tr.pdf by Bharati Vidyapeeth Dental College & Hospital user on 25 June 2022