CASE REPORT

https://doi.org/10.5125/jkaoms.2017.43.3.178
pISSN 2234-7550 · eISSN 2234-5930

Peripheral odontogenic myxoma in a 12-year-old girl: a rare entity

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Abstract (J Korean Assoc Oral Maxillofac Surg 2017;43:178-181)

Peripheral odontogenic myxoma is a rare odontogenic tumor representing an extra osseous counterpart of central odontogenic myxoma. It is commonly seen in gingiva between the 3rd and 4th decades of life and appears predominantly in females. Compared to central odontogenic myxoma, it is a less aggressive, slow-growing lesion with a low recurrence rate. However, close postoperative follow-up is required because of the unlimited growth potential of incompletely removed lesions. It shares many features with other soft tissue myxoid proliferations occurring in the oral cavity and hence needs to be differentiated from them. Very few cases of peripheral odontogenic myxomas have been reported and, to the best of our knowledge, no case has been reported in a pediatric patient. We present an unusual case of peripheral odontogenic myxoma occurring in a 12-year-old girl located in the anterior mandibular gingiva, with an emphasis on differential diagnosis.

Key words: Myxoma, Odontogenic tumours, Gingiva, Mandible, Mast cells

[paper submitted 2016. 5. 4 / revised 2016. 7. 25 / accepted 2016. 8. 3]

I. Introduction

Odontogenic myxomas are relatively rare benign odontogenic tumors that arise from the ectomesenchyme of the tooth-forming apparatus and are composed of spindle shaped/rounded/angular cells embedded in abundant mucoid stroma. Odontogenic myxomas can be categorized into central and peripheral variants[1-4]. Very few case reports of peripheral odontogenic myxomas (POMs) are available in the literature. Clinically and histologically, POMs resemble many other soft tissue lesions. Hence, recognizing and diagnosing POMs is necessary for the careful planning of conservative treatment and follow-up to rule out intraosseous extension[5]. This article presents a rare case of POM in a pediatric patient with a special emphasis on differential diagnosis.

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II. Case Report

A 12-year-old girl presented with a growth on the mandibular gingiva between tooth #31 and #32 (Fig. 1. A) that appeared three months prior. It was 1×1.5 cm in size, firm in consistency, and adherent to the mandibular gingiva but not fixed. The overlying mucosa was normal in color and texture.(Fig. 1. A) Radiologically, the intraoral periapical view showed drifting of tooth #31 and #32 without any erosion of alveolar bone.(Fig. 1. B) Based upon the clinical and radiographic findings, the lesion was provisionally diagnosed as a pyogenic granuloma. The lesion was completely excised and curetted under local anesthesia.

Gross examination of the excised tissue revealed a soft-to-firm grayish white pedunculated mass. Microscopically, an H&E stained section showed well-circumscribed lesional tissue separated from the overlying stratified squamous parakeratinized epithelium by fibrous tissue. The lesional tissue consisted of relatively acellular loose myxoid stroma with scattered spindle-to-stellate-shaped cells and many delicate proliferating capillaries. A minimal amount of collagen fibers was seen.(Fig. 2. A) The presence of numerous mast cells (MCs) was confirmed by toluidine blue staining.(Fig. 2. B) The lesional tissue was strongly positive for reticulin staining and showed alcinophilia.(Fig. 2. C, 2. D, respectively)

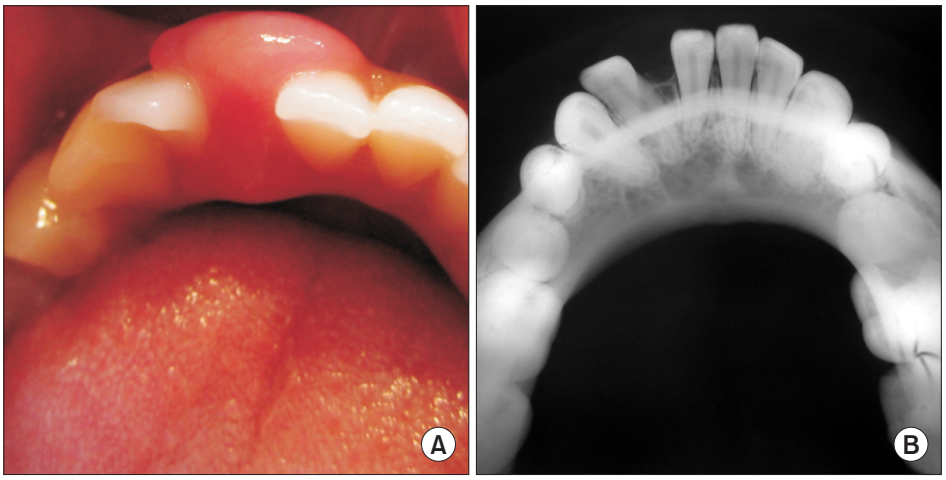

Fig. 1. A. Clinical photograph showing the gingival mass extending buccolingually between teeth #31 and #32. B. Occlusal radiograph showing drifting of #31 and #32 without bone involvement.
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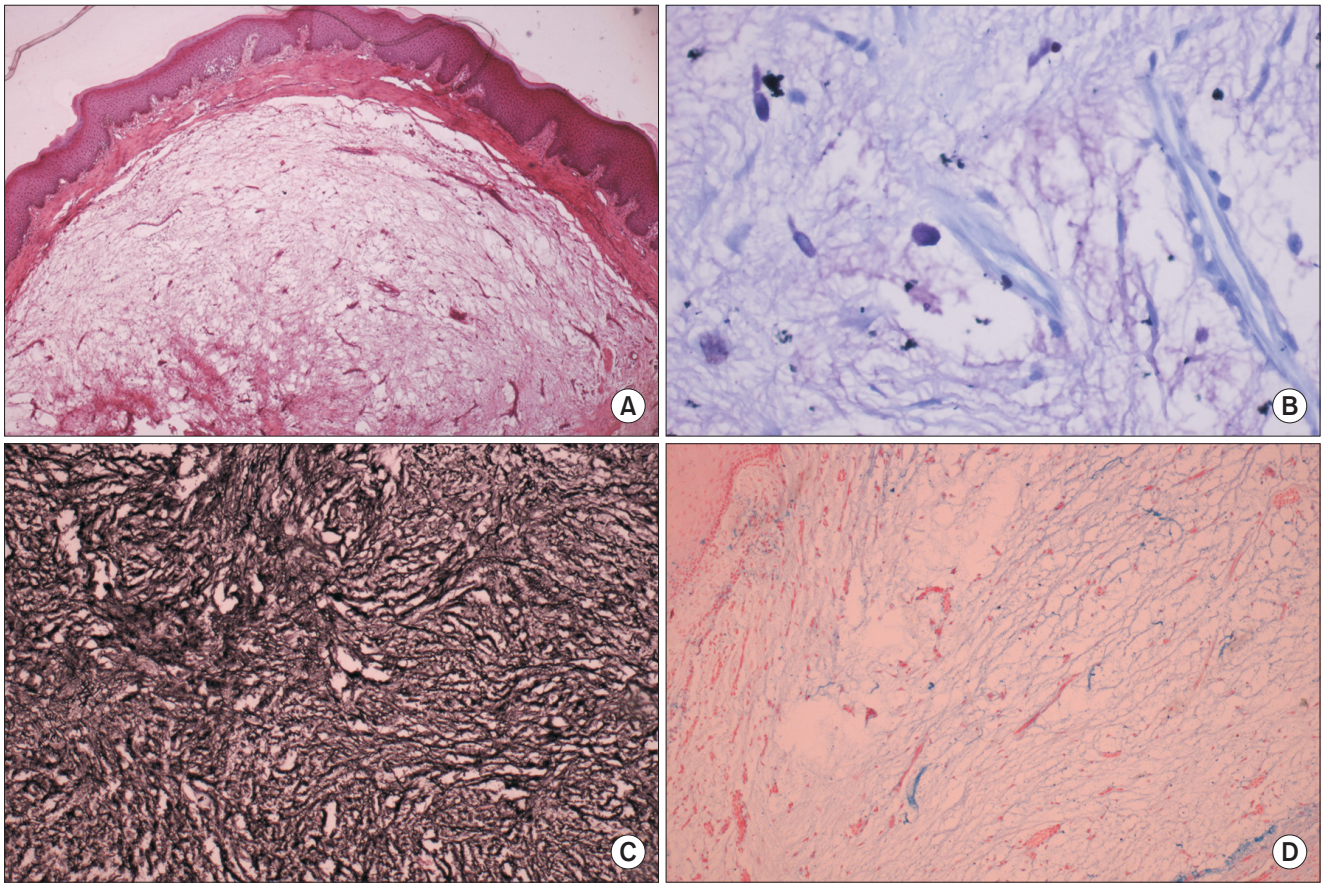

Fig. 2. A. H&E stained section (×10) showing loose myxomatous lesional tissue separated from the overlying stratified epithelium by a fibrous capsule. B. Toluidine blue-stained section (×40) showing mast cells in myxoid stroma. C. Reticulin-stained section (×10) showing strong positivity. D. Lesional tissue showing reactivity to Alcian blue staining (×10).
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Lesional tissue showed vimentin positivity and S-100 negativity. Based on these findings, a final diagnosis of POM was established. After excision of the lesion, the migrated teeth reverted to their normal position. The two-year follow-up period was uneventful.

III. Discussion

Odontogenic myxoma is a mesenchymal lesion of uncertain histogenesis that microscopically mimics dental pulp or follicular connective tissue[1,2]. Odontogenic myxomas are

classified as central/intraosseous and peripheral/extra osseous variants[2,3].

POM is a very rare lesion with a reported incidence less than that of other peripheral odontogenic tumors[4]; data on POM clinicopathologic features remain scarce[4].

Relevant literature suggests that peripheral myxomas of the intraoral tissues should be named POMs because soft tissue myxomas are usually seen extrafacially in skeletal muscles, dermal and subcutaneous tissues and do not occur in the oral cavity[5].

Several theories have been put forth regarding the pathogenesis of POM. One hypothesis states that altered primitive fibroblast/myofibroblasts produce excess mucopolysaccharides. And most of these cells are incapable of forming mature collagen. Other authors have suggested an origin derived from mesenchymal cells, such as dental papilla, dental follicle, or periodontal ligament[5,6].

POMs most commonly present clinically as pedunculated or sessile, painless, exophytic masses located in the gingiva[4,5]. Most of the reported cases of POMs occur in 4th to 6th decade of life[4,5,7-10]. In contrast, our case was found in a 12-year-old girl. POMs show a predilection for females and most reported cases have occurred in the maxilla[2,4,5,7-9], with only a few cases including the present case reported in the mandible.

The size of the lesions ranges from one centimeter to several centimeters, with two reported cases being very large[4].

Radiologically, some of the reported cases of POMs showed displacement of the associated teeth without root resorption. Localized erosion of alveolar bone was also observed in some cases[4,5,8]. In our case, the lesion caused tooth displacement without any bony erosion.

Clinically, POMs may mimic similar lesions like peripheral odontogenic lesions, peripheral giant cell granuloma, fibroma, lipoma, pyogenic granuloma, giant cell fibroma, traumatic fibroma, neurofibroma, focal oral mucinosis and other malignant and metastatic connective tissue tumors[2,4,5,7,10,11]. Histological examination is necessary to differentially diagnose these lesions.

POMs are poorly circumscribed myxoid proliferations outside the bone. They show little encapsulation and their rapid growth may be due to an accumulation of mucoid ground substance mimicking an aggressive neoplasm. The neoplasm is composed of haphazardly arranged stellate, spindle shaped and round cells in a loose myxoid stroma. Typically, a delicate vascular network and stellate fibroblasts are diagnostic of POM[4]. Odontogenic epithelial rests may not be obvious in most lesions and are not necessary for establishing a final diagnosis[5].

Interestingly, in our case of POM, we found a scattered distribution of MCs. MCs may also play an important role in the growth and expansion of odontogenic tumors and their presence is associated with poor prognosis[12,13].

It has been suggested that the MCs are associated with remodeling of the extracellular matrix in neoplastic alterations as they produce and release proteolytic enzymes favoring the migration of both endothelial and tumor cells as well as the release of angiogenic factors stored within the stromal tissue, leading to a higher degree of aggressiveness of odontogenic myxoma[12,14]. However, the presence of MCs has not been reported previously in POMs.

Histologically, the differential diagnosis of POM should include myxoid neurofibroma, myxoid chondrosarcoma, and myxoid liposarcoma, chondromyxoid fibroma, myxoid chondrosarcoma, a myxoid change in fibrosarcoma, botryoid type embryonal rhabdomyosarcoma and pleomorphic adenoma[4]. Awareness of the potential diagnostic pitfalls as well as careful evaluation of the clinical, radiological and characteristic histopathologic findings can narrow down the differential diagnosis[7]. Nerve sheath myxoma typically exhibits lobulated mucoid tissue containing stellate and spindle shaped cells, and condensed connective tissue representing perineurium surrounding the lesion. MCs are characteristically present in this lesion[11]. Oral focal mucinosis is clinically indistinguishable from other similar lesions; however, the connective tissue is alcinophilic and lacks reticulin fibres[15]. Our case showed strong positivity for reticulin staining, thus ruling out oral focal mucinosis.

In our case, we confirmed the results of other studies with respect to S-100 negativity and vimentin positivity. The diagnostic value of immunohistochemistry (IHC) in odontogenic myxomas is limited as the neoplastic cells share antigenic characteristics with many non-odontogenic myxoid proliferations and a specific marker for cells of dental ectomesenchymal origin is lacking[4]. However, IHC findings help to differentiate these lesions from other myxoid lesions.

If left untreated, POMs have unlimited growth potential. POMs without bone destruction are treated by simple excision while those with bone destruction require excision and marginal curettage. POM has a much lower recurrence rate (3%-8%) than central odontogenic myxoma (10%-33%). Therefore, a carefully planned conservative enucleation or semi-radical approach is justified[2,4,5,16]. Close follow-up of these lesions is necessary to rule out intraosseous extension and recurrence.

We conclude that special stains and IHC are valuable tools for the differential diagnosis of these lesions. Overtreatment of POMs should be avoided through the use of definitive diagnosis, especially in pediatric patients, as it may affect the alignment and eruption of teeth. The role of MCs in POMs needs to be further evaluated, since POMs with MCs have not been reported previously.

Conflict of Interest

No potential conflict of interest relevant to this article was reported.

Acknowledgements

The authors would like to thank Bharati Vidyapeeth Deemed University Dental College and Hospital, Sangli, for the necessary support. The authors are also thankful to Dr. Jaydeep Pol, M.D. Pathology, Mahatma Gandhi Cancer Institute, Miraj, for IHC analysis.

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CASE REPORT

https://doi.org/10.5125/jkaoms.2017.43.3.182
pISSN 2234-7550 · eISSN 2234-5930

Removal of submandibular calculi by surgical method and hydraulic power with curved needle: a case report

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Abstract (J Korean Assoc Oral Maxillofac Surg 2017;43:182-185)

Sialolithiasis, the most common salivary gland pathology, is caused by calculi in the gland itself and its duct. While patients with small sialoliths can undergo conservative treatment, those with standard-size or larger sialoliths require sialolithotomy. In the present case study, we removed two sialoliths located beneath the mucosa in the posterior and anterior regions of Wharton's duct, respectively. For the posterior calculus, we performed sialolithotomy via an intra-oral approach; thereafter, the small anterior calculus near the duct orifice was removed by hydraulic power. This method has not previously been reported. There were no complications either during the operation or postoperatively, and the salivary function of the gland remained normal.

Key words: Salivary gland calculi, Sialolithiasis, Sialoliths, Submandibular gland, Hydraulic power

[paper submitted 2016. 7. 9 / revised 2016. 8. 27 / accepted 2016. 9. 16]

I. Introduction

Sialolithiasis, the most common salivary gland pathology, is caused by calculi in the gland itself and its duct[1]. Gland swelling and pain are the chief complaints of presenting patients[2,3]. Most salivary gland calculi occur in the submandibular gland (83%), followed by the parotid (10%), sublingual and minor salivary glands (7%)[4]. Calculus size varies from less than 1 mm to a few centimeters[5]. Most salivary calculi are smaller than 10 mm; giant ones, larger than 15 mm and occurring only rarely, are known as sialoliths[2,6].

While patients with small sialoliths can undergo conservative treatment, those with standard-size or larger sialoliths require sialolithotomy[5,7,8]. Most submandibular calculi, located in the distal third of the duct, are removed surgically via direct incision to the stone[5,7,9-11]. This procedure is relatively simple to perform, and does not often result in complications[10]. Sialoliths that are sufficiently small and near the duct orifice can be manipulated and removed. Another minimally invasive method is using sialogogues with gland massage to promote salivation and flush the small sialoliths to the duct orifice[7,8,12]. Carelessness during this treatment, however, can cause rupture of the duct[12].

In the present case study, we removed two sialoliths located beneath the mucosa in the posterior and anterior regions of Wharton's duct, respectively. For the posterior calculus, we performed a sialolithotomy via an intra-oral approach; thereafter, the small anterior calculus near the duct orifice was removed by hydraulic power. Postoperatively, the patient's symptoms disappeared, and there have been no complications since.

II. Case Report

A 21-year-old female patient presented with a chief complaint of a firm mass on the right side of the floor of the mouth and uncomfortable swallowing for the preceding 4 months. She had no pain, fever, chills or lymph node swell-

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This study was supported by a grant of Dong-A University Medical Research Institute.

ing. Intra-oral examination by bimanual palpation revealed a small, firm, and non-tender swelling of the anterior floor of the mouth as well as a large, firm and non-tender swelling in the region opposite #38. Panoramic radiography and cone-beam computed tomography confirmed two calcified lesions in the posterior and anterior regions of the right Wharton's duct, respectively.(Fig. 1) The final diagnosis was sialolithiasis in the right Wharton's duct.

The posterior sialolith was removed intra-orally by sialolithotomy under general anesthesia, incising directly to the stone along the longitudinal axis of the duct and cautiously avoiding the nearby lingual nerve.(Fig. 2) The duct, which was 6 mm in diameter (Fig. 3), was left open, enabling successful, complication-free removal of the anterior sialolith by hydraulic power using a 50-mL syringe with a curved 18-gauge needle.(Fig. 4, 5) After inserting the needle into the open duct, the syringe was used to spout saline water to flush the small sialolith. Based on the saline that came from the duct orifice, we checked that the front of the sialolith was removed. The postoperative radiography was clear, indicating no remaining sialoliths. After one week and then again at two months, the patient was reviewed to assess gland's salivary function, which proved normal.(Fig. 6) To date, after 2 years, the patient has been asymptomatic, showing no recurrence.

III. Discussion

Sialolithiasis is the most common pathology of the salivary glands[1]. The precise pathogenesis is still mostly unknown; however, for treatment of sialolithiasis of the Wharton's duct, there are various methods available, the selection of which depends on the calculi location and size as well as the likeli-

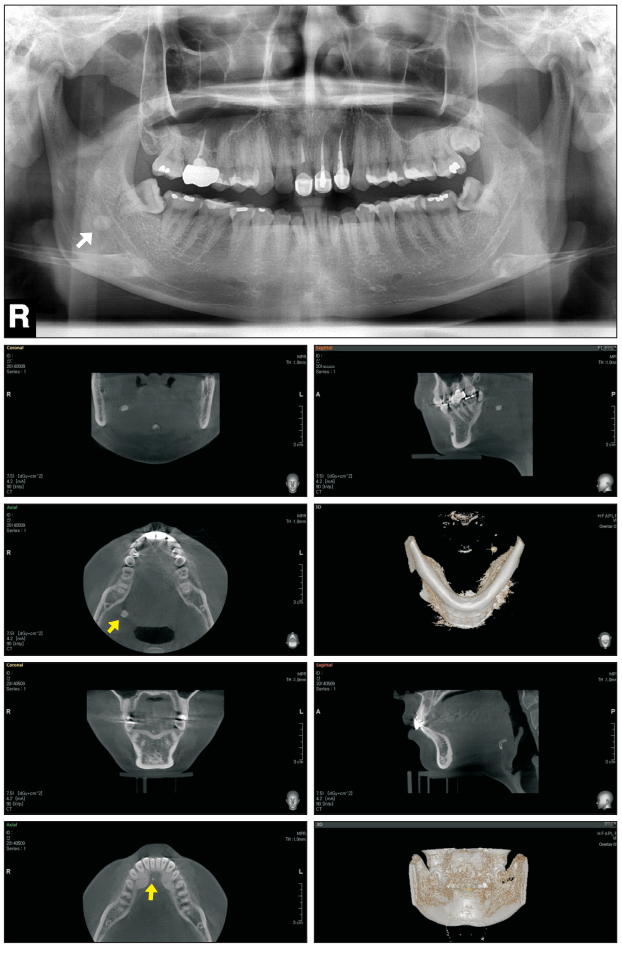

Fig. 1. Preoperative radiographs showing 2 sialoliths. Arrows presenting radiopaque structures.
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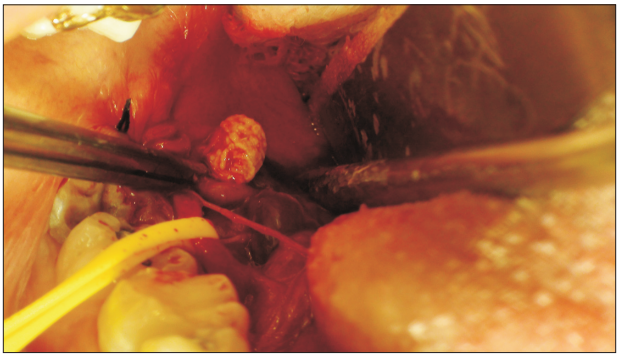

Fig. 2. Intraoperative photograph showing removal of sialolith from Wharton's duct.
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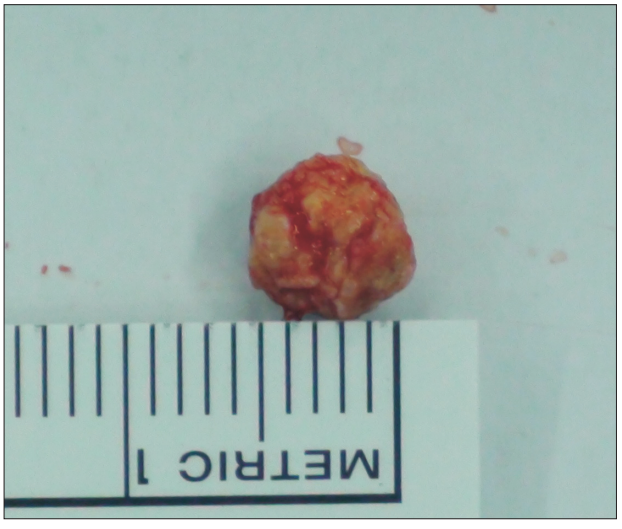

Fig. 3. Excised sialolith.
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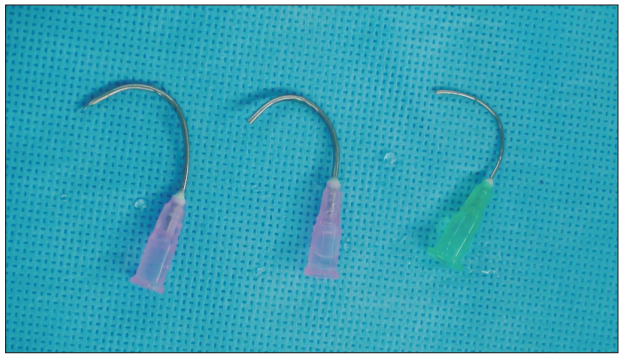

Fig. 4. Curved 18-gauge needles.
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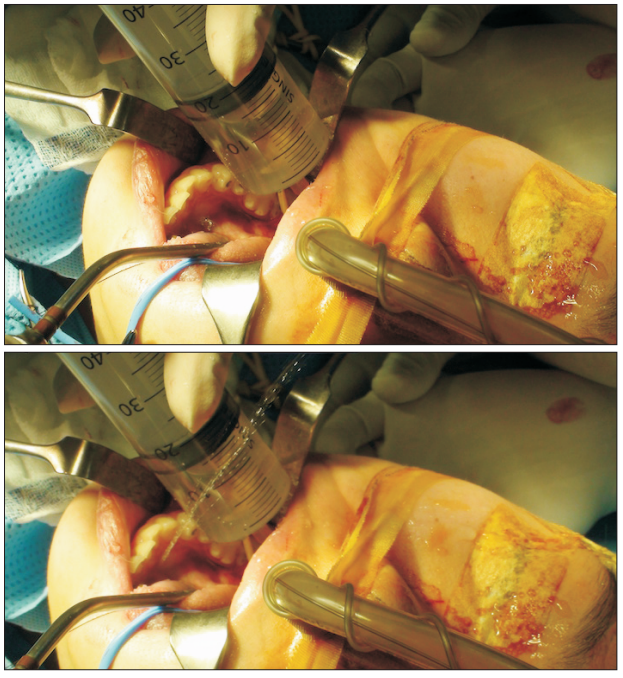

Fig. 5. Intraoperative photographs showing successful sialolith removal by hydraulic power.
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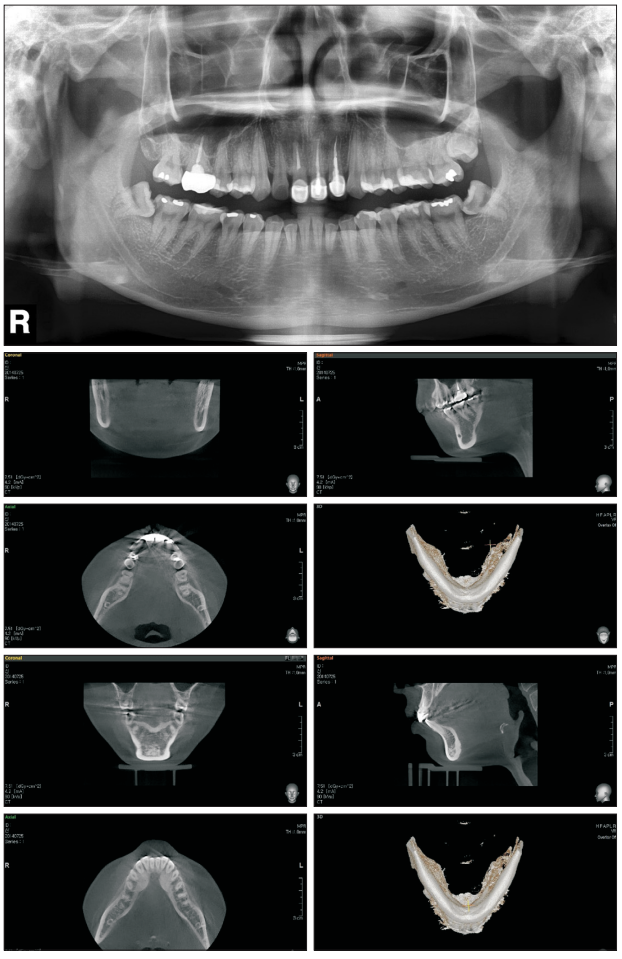

Fig. 6. Postoperative radiographs showing no sialoliths.
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hood of complications based on careful consideration of the patient's specific condition and symptoms[12-14].

The available methods range from conservative management to surgery[5]. The non-invasive options include hydration of the patient, prescription of sialogogues, and gland massage, all of which can be helpful in promoting salivation and consequent flushing of the calculus[7,12]. As for minimally invasive treatment, manipulation using lacrimal probes and dilators to open the duct orifice is useful in order to milk the calculus, manipulate it forward, and then remove it[7]. The invasive, surgical approach entails incision along the longitudinal axis of the Wharton's duct directly onto the stone, most of which are submandibular calculi located in the duct's distal-third region[7,8].

This paper presents a case report of a patient with two sialoliths in the right Wharton's duct. One sialolith, located posteriorly in the distal third of the duct, was surgically excised by sialolithotomy, after which the other, located anteriorly near the orifice, was removed through the duct by hydraulic power. The former surgical method is required to remove large stones in the back of the duct. Then, the least invasive method possible should be used for removal of any stones blocking the duct that are smaller than the diameter of the duct. Careless use of a lacrimal probe is likely to result in rupture of the duct, so we needed a new, easier and safer approach. We chose to try hydraulic power because it is less invasive than a lacrimal probe.

The use of hydraulic power is limited in that it requires another incision and the diameter of the stone should be less

than the diameter of the duct. Removal of only a small stone, located anteriorly near the orifice, can be achieved with conventional methods. Our future work will focus on determining indications for the use of hydraulic power method for less invasive removal of sialoliths.

This method has not been reported previously. There were no complications either during the operation or postoperatively, and the salivary function of the gland remained normal.

Conflict of Interest

No potential conflict of interest relevant to this article was reported.

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