

Submandibular Sialolithiasis Perforating the Floor of Mouth: A Case Report

Case Report

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Abstract

Calculus formation in salivary glands (sialolithiasis) is one of the most common diseases of the salivary glands and is most commonly seen in the submandibular gland. Not only can the stones be small and inside the duct, they may also get larger and reside inside the gland. We can easily see submandibular sialolithiasis perforating the floor of the mouth in cases having sialolithiasis for decades and not having any treatment and its transoral removal as we look in the medical literature. A 52-year-old lady who had rejected surgical treatment for submandibular sialolithiasis for 25 years is presented as a

case in this study. Treatment of the case was performed effectively by taking out the calculus transorally with a simple intervention whose examination revealed that the calculus was seen to perforate the floor of the mouth. It is aimed to stress with this case that transoral removal of submandibular sialolithiasis that perforates the floor of the mouth without performing external approaches is a method that has to be thought in the first step.

Keywords: Floor of mouth, submandibular, sialolithiasis, calculi

Introduction

Calculus formation in salivary glands (sialolithiasis) is one of the most common diseases of salivary glands. It frequently occurs after 25 years of age. It is two times more common among men than among women. The incidence in adult population is 12 in 1000. Although sialolithiasis is most commonly seen in the submandibular gland (approximately 85%), calculus formation may also be seen in the parotis, sublingual, and other salivary glands. Factors such as stasis of the salivary flow, increased mucous secretion, epithelial inflammation, calcium deterioration, and electrolyte metabolism play a role in the pathogenesis of sialolithiasis (1). The sialoliths can not only be small and be present inside the duct but also get larger and reside inside the gland. Sialolithiasis may cause infected sialadenitis and clinically presents itself with painful swelling under the jaw. In a patient complaining of pain and swelling under the jaw during or after a meal, detection of stones in the Wharton's duct during the bimanual palpation of the mouth floor is diagnostic. Combination of ultrasonography (USG) and neck tomography can be useful in the case of non-palpation or uncertain diagnosis. For therapy, the localization of the sialolith is of great importance. Although a simple surgery without an incision is sufficient to remove the sialoliths that perforate the mouth of the Wharton's duct, intraoral or external surgery from the neck may be needed for the stones that reside in the duct or the gland. It has been reported in the literature that very large stones also perforate the floor of the mouth (2). In

our study, we present a case of a patient with submandibular sialolithiasis with spontaneous perforation of the floor of the mouth.

Case Report

A 52-year-old female patient who presented with pain under the tongue and swelling to the Dokuz Eylül University, Faculty of Medicine Emergency Department was admitted for swelling under the jaw and oral intake restriction; she was referred to our clinic. Patient's history revealed that she had submandibular sialolithiasis for 25 years and had rejected surgical treatment despite other doctors' recommendations. The patient who could not describe recurrent episodes of sialadenitis in the previous years developed swelling under the chin, and the swelling got bigger in the last couple of days. In the patient's examination, an image compatible with off-white-yellow colored sialolith perforating the mouth of the right Wharton's duct was observed (Figure 1). The sialolith was removed from the mouth of the duct without using any anesthetic agents with the aid of forceps. The size of the stone that was removed was 2.5×0.8×0.6 cm (Figure 2). The patient was prescribed 150-mg clindamycin capsules 4×1 and analgesic-anti-inflammatory for sialadenitis, and a soft food diet for a few days was recommended. Because the mouth of the duct was quite enlarged because of the stone and the integrity of the duct was not impaired, an additional surgery was not needed. A check-up after a week showed that the patient's saliva drainage from the mouth of the duct was normal. The first



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month check-up also did not reveal any problems. The patient's informed consent for this case report and the use of images for this purpose was obtained.

Discussion

Although submandibular sialolithiasis is usually seen among adult population, it can rarely be seen in the pediatric population (3, 4). This may be because of the submandibular duct being longer, its flow being horizontal and in the opposite direction of gravity, the mouth of the duct being narrow, and the submandibular saliva being more alkaline and containing more mucous secretions. Eighty percent of the submandibular sialolithiasis is in the Wharton's duct, and 20% of it is in the gland. Its size may differ from a few millimeter to a few centimeter (5). The salivary gland stones are thought to grow approximately up to 1–1.5 mm in a year (6). Submandibular sialolithiasis generally tends to be asymptomatic. However, when the stone reaches the appropriate size to obstruct the duct lumen, it can cause swelling and pain in the gland. In this situation, patients complain of recurrent pain and swelling developed with eating. In case of secondary infection, a clinical picture is characterized with high fever and occurrence of purulent discharge into the mouth. As a complication of sialolithiasis, acute or chronic sialadenitis may develop.

To assist diagnosis, USG, computed tomography, or sialography can be used. Nearly 80% of the submandibular stones are radiopaque and allow detection via imaging (7). Due to the fact that up to 40% of the stones in the parotid gland are radiolucent, conventional or sectional sialography is a very useful test for these patients. Although sectional sialography is considered as the "golden standard" technique in the diagnosis of sialolithiasis, the disadvantages are that it requires the use of contrast, and that it cannot be used for patients with acute infections and those who are allergic to contrast (8). For diagnosis, USG, which is an inexpensive and a reliable method, can be used. The stones of sizes larger than 2 mm can be easily seen via USG.

Submandibular stones are classified as anterior and posterior according to their transverse direction relationship with the mandibular first molar tooth. Anterior stones are easy to recognize with occlusal radiographs and usually can be easily excised because they perforate into the mouth (1, 2). Those that do not perforate into the mouth can be removed following the dilation of the duct orifice. Posterior stones sometimes reside in the hilum of the gland and sometimes within the gland. These stones usually may not be palpated with bimanual palpation inside the mouth. An extraoral excision of the stones with the gland may be required (1, 2, 7).

Treatment of sialolithiasis is dependent on the localization and size of the stone as well as on the duration and recurrence frequency of the symptoms. It has a wide spectrum of treatment options from conservative medical treatments such as an-



Figure 1. Sialolithiasis perforating the floor of the mouth



Figure 2. A stone with a size of 2.5×0.8×0.6 cm.

tibiotherapy, sufficient hydration, and hot massage therapy to advanced methods such as surgical excision and extracorporeal shock wave lithotripsy (ESWL) (9). Anteriorly located submandibular stones usually respond well to medical treatment; however, for these patients, it might be necessary to remove the stone through sialolithotomy. This initiative will be carried out in patients for whom it is necessary to be alert to the risk of injury to the lingual nerve. ESWL of stones of smaller size up to 12 mm generally respond well (10). For patients who will undergo this surgery, it is vital to be alert regarding the risk of lingual nerve injury. Generally, stones smaller than 12 mm in size respond well to ESWL (10).

When we refer to the medical literature, we know that patients with submandibular sialolithiasis perforating the floor of the mouth are not rarely seen. Bayindir et al. (1) reported in the case that they presented that the submandibular sialolithiasis perforating the floor of the mouth was 3×2 cm in size. In the case presented by Yaman et al., (2) the stone that was removed by means of Volkmann's curette was 3×3×2 cm in size. In the case presented by Sütay et al., (3) it was reported that the submandibular stone with a size of 37×7 mm was spontaneously extruded from the floor of the mouth without any need for surgery,

and no problem was experienced during the follow-up. Siddiqui et al. (4) reported that the submandibular sialolith with a size of 30 mm was perforating into the mouth and was removed under local anesthesia.

The pathogenesis for the ability of sialolithiasis to grow to such an extent can be summarized as follows. In response to a sialolith growing in the duct, the duct may continue to expand at the same rate. Because the duct expands, these patients tend to remain asymptomatic for many years. However, if the size of the sialolith reaches a point where the duct can no longer expand, a sialo-oral fistula forms, and some part of the sialolith perforates into the oral cavity (11, 12).

In our case, the stone that was anteriorly located and perforated the mouth was removed without the need for any further intervention. Because the mouth of the Wharton's duct dilated owing to the presence of stone and there was a normal amount of saliva formation within, suturing on the mouth of the duct or an additional process was not required. Because the patient also had sialadenitis, she was given 1-week antibiotherapy after the procedure. No problem was detected during the patient's 6-month follow-up. In this case, we want to emphasize on the topic that submandibular sialolithiasis spontaneously protruding from the floor of the mouth is not seen very infrequently and on what kind of method should be applied in these cases taking into consideration similar cases that have been observed in the medical literature. When we assessed our case, it was possible to remove the stone with the help of forceps because the position of the stone was convenient and the duct orifice was sufficiently dilated. Accompanied with these studies, we can see that in similar cases with a simple medical intervention and without any complications, the stone can easily be removed. Although the transoral removal of submandibular sialolithiasis that perforates the floor of the mouth without performing external approaches is a method that has to be considered in the first step, it should be remembered that each case should be considered separately while planning its treatment process.

Conclusion

Transoral removal of submandibular sialolithiasis that perforates the floor of the mouth without performing external approaches is a method that has to be considered in the first step because it is the simplest and the most inexpensive method.

Informed Consent: Written informed consent was obtained from the patient.

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