

Cervical fistula caused by submandibular sialolithiasis

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Abstract

In oto-rhino-laryngology, cases of submandibular sialolithiasis are common. Submandibular sialoadentis with sialolith may cause severe complications such as deep neck abscess and sepsis. We introduce a rare case of a cervical fistula with abscess caused by submandibular sialolith. The patient had diabetes. We performed drainage of the left submandibular gland that included a Wharton duct stone and abscess by an external skin incision approach. Submandibular sialoadentis due to sialolith would likely progress to neck abscess and the formation of a neck skin fistula; moreover, the condition can be worsened by the coexistence of diabetes. This neck abscess with skin fistula could have caused potentially fatal complications such as carotid artery rupture or sepsis. In such cases the infected source should be carefully removed as soon as possible.

Introduction

Sialolithiasis within the salivary gland ducts or parenchyma is sometimes encountered in oto-rhino-laryngology. The area surrounding a submandibular sialoadentis due to sialolithiasis can often show an inflammatory reaction. However, cases of cervical fistula with abscess caused by submandibular sialoliths are likely very rare. To our best knowledge, this report is the only two.^{1,2} We experienced a case of cervical fistula with abscess caused by submandibular multiple sialolith. This case report presents the clinical findings, imaging examination, treatment, postoperative progress and some considerations based on various studies.

Consent for publication

We confirm the patient's anonymity and obtained consent for publication of the clinical findings, image examination and pathological examination.

A copy of this document is available for review by the editor of this journal.

Case Report

A 72-year-old Japanese man complained of left submandibular pain and swelling for three months. His past history included diabetes. He consulted another hospital and was diagnosed with sialolithiasis of the left submandibular gland. A month before, the left submandibular pain and swelling became worse with a neck skin ulcer. Therefore, he was introduced to our hospital. At first examination, his left neck showed poor granuloma in a skin defect with swelling and redness (Figure 1). Computed tomography (CT) scan revealed some stones 2-20 mm in diameter in the left submandibular gland and Wharton duct. Moreover, a fistula existed from the left submandibular gland to the skin defect (Figure 2). The body temperature was 36.0°C. Vital signs were stable. In the blood examinations the white blood cells count was 8600/ μ L (normal range: 4000-8000 μ L), C-reactive protein was 0.3 mg/dL (normal range: 0-0.3 mg/dL), and HbA1c 8.9% (normal range: 4.6-6.2%). Biopsy and bacteria culture from the granuloma in the neck defect ruled out tumor diseases and tuberculosis.

We removed the left submandibular gland together with the Wharton duct stones and poor granulomas by an external skin incision approach (Figure 3). In the operative findings, the submandibular gland hardly adhered to its surrounding tissues. However, the main vessels (such as internal jugular vein, internal, external and common carotid artery) and nerves (lingual nerve, hypoglossal nerve and vagus nerve) were not damaged. The postoperative wound was left open rather than sutured to allow drainage of the pus from the wound. The histopathologic findings of the submandibular gland were nonspecific chronic inflammation.

The postoperative course was good. The open wound could close naturally by the administration of antibiotics, local treatment of the wound and diabetic therapy for one month.

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Discussion and Conclusions

Sialolithiasis within the salivary gland ducts or parenchyma, is somewhat common in the adult population,³ with 12 per 100,000 people affected, and is characterized by symptoms of recent glandular swelling pain, and trismus. Traditional medical management consists of treatment with antibiotics and anti-inflammatory drugs, with the hope of spontaneous expression of the stone through the normal papilla. Surgical management is reserved for stones located in the proximal duct, and glandular resection is reserved for deeply embedded stones.⁴ Submandibular sialoadentis with sialolith may cause severe complications such as deep neck abscess or sepsis. We could find only two relevant papers in the literature concerning cervical fistula caused by submandibular sialolith. One case report described a neck fistula but there was no large defect in the neck skin as in our case.^{1,2} Our case suggested the possibility that submandibular sialoadentis due to sialolithiasis could progress to a neck abscess and the formation of a neck skin fistula; moreover, the condition would likely be worsened by the co-existence of diabetes. Our patient showed poor granuloma



Figure 1. At first examination, the left neck showed a skin defect with redness, swelling and poor granuloma.

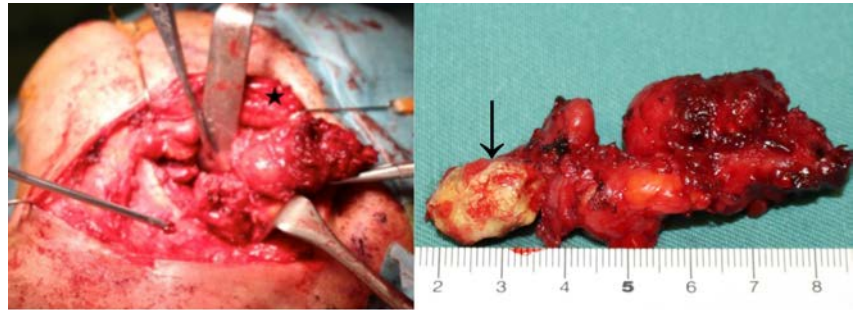


Figure 3. On the left there is the left submandibular gland (asterisk) including a Wharton duct stone. The specimen (right) shows the submandibular gland with the main stone (arrow).

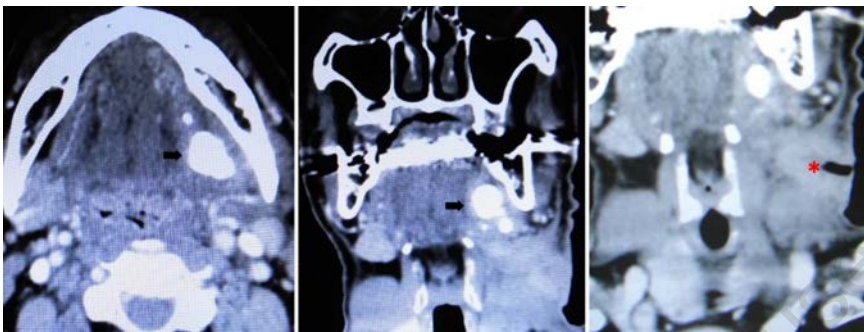


Figure 2. In the preoperative computed tomography (CT) (left, middle), some stones (arrow) 2-20 mm in diameter could be found from the submandibular gland to the Wharton duct. Right CT showed a fistula (asterisk) between the left submandibular gland and the skin defect.



Figure 4. A post-operative finding could show complete closure of the fistula.

in the skin defect on the left neck, and was therefore at risk of potentially fatal complications such as carotid artery rupture or sepsis. We could confirm neither tumor diseases nor tuberculosis by biopsy and bacteria culture. In such cases the infected sources should be carefully removed as soon as possible.

Recently, some hospitals have presented minimally invasive surgery such as by using sialoendoscopy through an oral approach for submandibular sialolithiasis.^{4,5}

Kleiszak⁴ attempted the extirpation by the above method in a case of submandibular sialolithiasis with Wharton duct fistula in the oral cavity, but there was no cervical fistula as in our case. In our case, for complete removal of the infected focus and drainage, we selected an external skin incision

approach and removed the left submandibular gland with Wharton duct stones and poor granulomas. The postoperative wound was left open and was not sutured in order to allow drainage of the pus from the wound. The postoperative course was good. The open wound could close naturally by the administration of antibiotics, local treatment of the wound and diabetic therapy for one month.

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