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Clinical Record

Sialolithiasis in a residual Wharton's duct after excision of a submandibular salivary gland

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Abstract

Treatment of salivary stones includes both surgical and non-surgical techniques. Surgical approaches range from excision of the sialolith, for those near the duct orifice, to removal of the affected salivary gland and its associated duct, for stones near the hilum of the gland. We present a case of two sialoliths triggering an acute infection in a residual Wharton's duct, 12 years after the removal of the associated submandibular gland. Excision of the sialoliths and treatment of the infected duct via sialodochoplasty was successfully performed in this patient. If the Wharton's duct is not removed with the associated submandibular gland, the potential for infection and continuous growth of dormant calcifications exists. We also address the aetiology, pathogenesis, and management of patients with sialolithiasis in the absence of a major salivary gland.

Key words: Submandibular Gland; Salivary Ducts; Calculus

Introduction

Eighty to ninety per cent of sialolithiasis cases occur in the submandibular glands and Wharton's duct. Sialoliths are more common in adults during the third to sixth decades of life but can also be found in children.[1,2] The treatment of salivary stones includes both surgical and non-surgical techniques.[1,3–9] Excision of the sialolith is one surgical approach; another may also include the removal of the affected salivary gland and its associated duct. If the Wharton's duct is not removed during sialadenectomy, the potential for infection and continuous growth of dormant calcifications exists.

We present a case of two sialoliths triggering an acute infection in a residual Wharton's duct, 12 years after removal of the associated submandibular gland. Excision of the sialoliths and treatment of the infected duct via sialodochoplasty was successfully performed in this patient.

Case report

A 36-year-old white woman presented with the complaint of a hard and painful swelling in the right floor of her mouth. The patient reported that the swelling had been present for approximately one month and had initially been accompanied by a sharp pain that eventually abated and presented only while eating. The patient's medical history revealed repeated episodes of sialolithiasis in the right Wharton's duct, which had culminated in the surgical excision of the right submandibular gland in 1992. The patient's other medical conditions included a history of surgical fusion of vertebrae L3–L5 and arthroscopy of her left knee in 1999. In addition, mild asthma, migraines, and limited mouth opening due to myofascial pain were reported. The patient was taking hydrocodone and cyclobenzaprine hydrochloride for back pain, albuterol for asthma, and Depo-Provera® for oral contraception. The patient reported no history of smoking, alcohol or recreational drug use.

Clinical examination revealed normal extra-oral and intra-oral anatomical structures, except for evidence of a 2 cm, indurated, erythematous swelling in the right anterior floor of the mouth. The swelling was not tender to palpation. A panoramic radiograph showed the presence of two ovoid radiopacities below the inferior border of the mandible. A computed tomography (CT) study of the floor of the mouth and the neck was ordered to further assess the radiopaque lesions.

Two weeks later, the patient was seen again to review the CT study, which was remarkable only for the presence of two large, radiopaque masses within the right Wharton's duct (Figure 1). The posterior mass closest to the location of the previously excised right submandibular gland measured 11 × 4 mm, and the anterior mass closest to the right Wharton's duct orifice measured 4 mm in diameter. There was no radiographic evidence of residual right submandibular gland tissue.

At the time of this consultation, the patient reported increased pain from the area of the swelling and a daily purulent discharge that had begun a few days after her initial visit. Clinical examination revealed a purulent discharge upon palpation of the right floor of the mouth. At this point, it was elected to perform exploratory surgery, sialodochoplasty and excision of both sialoliths under local anaesthesia.

At the time of surgery, a scant purulence oozed from the orifice of the right Wharton's duct. A lacrimal probe was inserted into the orifice to cannulate the right Wharton's duct for approximately 15 mm until resistance was met

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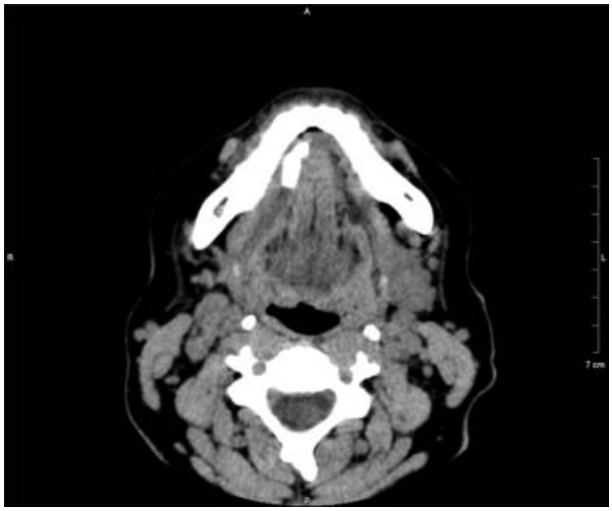

FIG. 1
Computed tomography scan revealing anterior and posterior sialoliths within the right Wharton's duct.

(Figure 2a). A Bovie electrocautery needle tip on a 20 W cutting setting was used to incise along the tissue above the lacrimal probe to include the duct orifice. Two retraction sutures were placed for stabilization of the opened

FIG. 2
(a) Identification and cannulation of the right Wharton's duct with a lacrimal probe. (b) Retraction sutures placed at the orifice of the right Wharton's duct. The anterior sialolith is displayed. (c) Posterior sialolith with surrounding bloody purulence representative of infection.

FIG. 2 Continued.

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duct. At this time, the 4 mm anterior salivary stone was identified and removed from the duct (Figure 2b). The lacrimal probe was then further inserted posteriorly until resistance was once again noted. Blunt dissection was performed with a mosquito haemostat and copious purulence was expressed from around the sialolith which necessitated incision and drainage from the surrounding soft tissues. The duct was further opened with the Bovie. The posterior sialolith was then identified and removed from the duct (Figure 2c). More purulence was expressed and a blind pouch was identified that continued posteriorly into the right submandibular triangle below the mylohyoid muscle attachment. Following irrigation, the duct was sutured open to the surrounding floor of the mouth mucosa via dochoplasty in order to promote healing by secondary intention and to substantially shorten the length of the residual Wharton's duct. The patient was prescribed ciprofloxacin 500 mg twice daily, chlorhexidine rinse twice daily and ibuprofen 800 mg thrice daily.

The excised specimens were fixed in 10 per cent buffered formalin and decalcified. Microscopic examination of 5 μm paraffin-embedded sections revealed two fragments of calcified material with typical concentric laminations displaying basophilic layers (Figure 3). Amidst the calcified material, bacterial colonies, acute inflammatory cells and surgical haemorrhage were observed.

The patient was seen for follow-up examination two weeks after surgery; there was no evidence of wound dehiscence, infection or purulent discharge.

Discussion

Our case is unique and underscores the potential complications arising from leaving a residual excretory salivary duct after the excision of a submandibular gland. It is conceivable that our patient's sialoliths had been present in the Wharton's duct since the time of submandibular gland removal and had remained quiescent for 12 years until a superimposed infection unveiled them. It is also possible that the stones had formed following excision of the submandibular gland.

The exact mechanism of sialolith formation is unclear. It is thought that desquamated epithelial cells, microorganisms, and salivary duct derived lysosomes and mitochondria-like structures may be the matrix that precipitates calcium present in the duct.[10] Although some authors have suggested that a diet high in calcium may play a role in salivary stone formation, the calcium found in sialoliths appears to originate from saliva.[2,9] A possible explanation for the predominance of salivary stones in Wharton's duct may be the alkaline pH, high mucin and Ca^{+} content of saliva. In addition, the anatomical characteristics of Wharton's duct – a long ascending trajectory that circumvents the posterior border of the mylohyoid muscle, may contribute to salivary gravitational flow stasis and, perhaps, to calcium precipitation.[1]

A decreased salivary flow rate is further diminished after sialolith formation and salivary gland infection.[1] Even after excision of the sialolith and clearing of the duct, there is a higher likelihood of recurrent sialolith formation due to the decreased flow rate with subsequent accumulation of intraductal sludge.[11] Therefore, recurrent salivary gland infections and/or recurrent sialolith formation usually necessitate sialadenectomy.[12,13]

Typically, sialadenectomy of the submandibular gland consists of removal of the glandular tissue and its associated duct. Failure to do so can result in complications associated with the presence of intraductal sialoliths, which may lead to recurrent infection or to cutaneous or intraoral fistula formation due to sialolith migration.[4] The most common complaints of patients with this condition are swelling and pain while eating.[1]

Sialodochoplasty of the submandibular gland (a procedure originally introduced to alleviate drooling in mentally disabled patients) has shown success in treating salivary obstruction.[3,5–10,14–17] Application of mitomycin C to the newly created opening of the duct has been shown to prevent premature stenosis.[7]

We used a slightly modified Rontal and Rontal technique to treat our patient.[8] Briefly, the duct was identified with a lacrimal probe and cannulated open with a mosquito haemostat. Retraction sutures were placed as the duct was widened to prevent premature closure. Following the removal of the sialolith(s), the new orifice of the duct was sutured to the surrounding oral mucosa to prevent stenosis.

- **This paper describes a case of two sialoliths triggering an acute infection in a residual Wharton's duct, 12 years after removal of the associated submandibular salivary gland**
- **Excision of the sialoliths and treatment of the infected duct via sialodochoplasty was successfully performed**
- **If the Wharton's duct is not removed with the associated submandibular gland, the potential for infection and continuous growth of dormant calcifications exists**

Removal of small, superficial sialoliths can be accomplished by simple widening of the ductal orifice with a lacrimal probe and subsequent milking of the gland. Larger sialoliths that are less superficial usually require incision along the length of the duct directly over the sialolith, with subsequent dissection around, and removal of, the sialolith. The duct is left open, and the oral mucosa overlying the incised duct is then sutured to the peripheral tissues. No sutures are placed in the duct itself to prevent stenosis. Sialoliths located close to the hilum of the salivary gland are removed along with the gland and its associated excretory duct. Haemorrhage, infection, and possible damage to the cranial nerves V_3 (in particular, the lingual branch of the trigeminal nerve), VII and XII are potential complications of surgical excision. In selected patients,

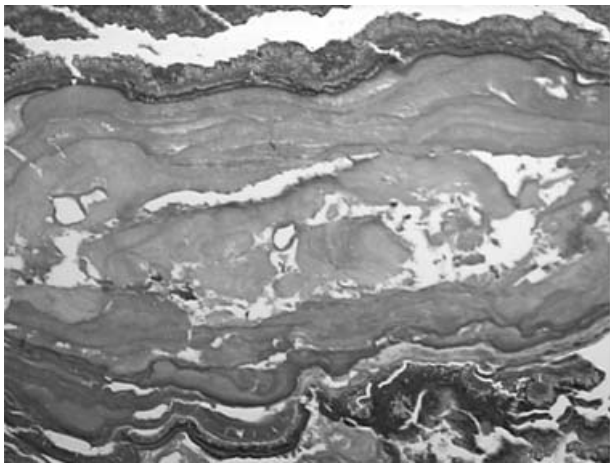

FIG. 3

Low magnification view of the biopsy specimen, demonstrating typical concentric laminations (H&E; ×10 original magnification).

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extracorporeal and intracorporeal lithotripsy represent non-surgical options for the treatment of sialolithiasis.[9]

The treatment of sialolithiasis should be as conservative as possible. In our case, a transoral surgical approach with sialodochoplasty was deemed to be the most appropriate procedure. Maintenance of patency of the treated ductal orifice is crucial to long-term success.

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