Sublingual Gland Sialolithiasis: A Case Series and Literature Review

Introduction

With an incidence of 2-6% in the general population, obstructive sialadenitis, with or without sialoliths, represents the most common inflammatory disorder of the major salivary glands. Sialoliths most commonly occur in the submandibular gland, outnumbering the parotid gland by approximately 3:1, while they occur in the sublingual gland in just 0.6-6.4% of cases.4,10 Because the sublingual gland is a rare site for sialolithiasis, it may be underdiagnosed and may not be examined as a Wharton’s duct stone.

Sialolithiasis most commonly produces regularly recurring swelling and pain of the involved gland, particularly during meals. In some cases, sialolithiasis may be an incidental finding on imaging. Traditional treatment for sialolithiasis includes surgery, often either intracutaneous or external incision of the entire gland and stones. Sialendoscopy, first performed with a flexible endoscope by Katz in 1993, and then with a rigid sialendoscope by Nakhle et al in 1994, has increased in popularity, as it permits diagnostic examination and allows stone removal with gland preservation.11 However, not all stones are amenable to retrieval via sialendoscopy, and recent studies highlight determining factors such as size, site, type and number of stones, along with functional status of the gland.12

Combined-endoscopic and transoral approaches have been described to avoid sialoadenectomy for stones considered too large, for stones situated within the submandibular gland or at the hilum, and for stenosis that is too tight to be dilated.11,12 We describe 3 patients with sialoliths located at the takeoff branch to the sublingual gland, a rare location that presents challenges for diagnosis and endoscopic removal. There are no reports describing sialolithiasis in this location, and there are no reports of sialendoscopy as a treatment method for sublingual gland sialolithiasis. Each patient in our series was managed with a combined approach, using sialendoscopy and transoral excision of the sublingual gland.

Case Reports

Case 1. A 20-year-old female presented after a 6mm sialolith was incidentally found on a CT scan of her neck. On retrospective review, she had one episode of mild submandibular gland swelling, the underling sialendoscopy, which identified the calculi filling a branching point midway down Wharton’s duct (See figure 1). Attempts to retrieve the stone with a basket, as well as with fragmentation, were unsuccessful. Guided palpation of the stone and by location of the scope with visualization of the stone, transoral cut down was performed with incisions along the mid floor of the mouth. The sublingual gland was encountered and dissected to allow access to Wharton’s duct (See figure 2). Both the stone and sublingual gland were removed, with preservation of the duct and the lingual nerve. Sialendoscopy was repeated, and several remaining small fragments of stone were identified and retrieved with a basket. Sialendoscopy repeated again confirmed that there were no residual stones and that Wharton’s duct was patent. Follow-up at 12 days showed the incision to be well healed.

Case 2. A 74-year-old female presented with only one previous episode of submandibular gland swelling. On presentation, she had a stone palpable on the posterior floor of mouth that was large, but separate, from her sublingual gland, while CT showed a sublingual, an anterior one at mid floor and a posterior one within the sublingual gland, distinct and separate from the submandibular gland. She underwent diagnostic sialendoscopy which demonstrated the anterior stone located within mid part of duct. It was too large for endoscopic removal and sialolithotomy was performed. Repeated sialendoscopy did not reveal the second stone, so transoral cut down was performed in similar fashion to case 1. It was not until resolution of the sublingual gland that the stone was identified embedded within the gland. Wharton’s duct was excised, allowing visualization of stone off the side of Wharton’s duct and showing the duct to be widely patent throughout its length. At 6 months follow-up, she was doing well with no recurrent symptoms.

Case 3. A 17-year-old female presented, like the others, with a single episode of submandibular gland swelling that had occurred 3 months prior. CT scan revealed a 6 mm calcified anteriorly on the floor of the mouth. In exam, the stone was palpable more posteriorly than would be predicted by the scan. Sialendoscopy revealed the stone floating, remained in the main duct, but off of a side channel toward the sublingual gland. Due to size and location, the stone and sublingual gland were resected with a mid floor of mouth cut down approach as described in case 1. Repeat sialendoscopy showed a widely patent duct with the sialolithotomy site off to the side. At 12 days postoperatively, she had a well healed incision with good salivary flow from Wharton’s duct.

Discussion

The incidence of sublingual gland sialolithiasis varies in the literature from 0-6.4%.3,4,5 Older studies from the 1950s and 1960s report a higher incidence of 6.4%, which may be falsely elevated due to confusion with stones in the anterior portion of Wharton’s duct.1 A more recent series by Zeich et al demonstrated that out of 163 patients who had sialolithiasis originating in the sublingual gland,9 the relationship between the submandibular gland and sublingual gland remains unclear, but there is frequently a direct connection between the two ductal systems, as evident from our sialendoscopy findings. Furthermore, the part of the submandibular gland superior to the mylohyoid muscle is frequently non-communicating with the posterior part of the sublingual gland and is indistinguishable except histologically. In our review of over 230 sublingual gland cases from 2000 to present at our tertiary care academic center, only three cases of sublingual gland sialolithiasis were identified.

We report 3 cases of sialolith located within the duct to the sublingual gland treated with a combined endoscopic and transoral cut down approach to remove the sialolith and the sublingual gland. The average size of excised stone was 0.5±0.1 cm. At follow-up, all patients were doing well with no recurrent symptoms.

Sialolithiasis most commonly produces regularly recurring swelling and pain of the involved gland, particularly during meals. Lustmann et al report that 96% of patients experience symptoms for 1 year or longer before treatment. However, symptoms associated with sublingual gland sialolithiasis appear to be less frequent and less severe. In one case report, the finding was incidentally discovered on imaging performed for a different TMJ dysfunction dysfunction syndrome.13 In each of our cases, the patient presented with mild symptoms of sublingual gland swelling of short duration. Despite symptomatic relief to the submandibular gland, pathology revealed chronic sialadenitis affecting the sublingual gland. These stones may have originated in Wharton’s duct and subsequently became embedded in the branch point to the sublingual gland. In this location, they no longer obstructed Wharton’s duct and symptoms did not persist.

References