CASE REPORT

Submandibular gland sialolithiasis with chronic sialadenitis: A case report

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Abstract

The most common non-neoplastic salivary gland disease is obstructive sialadenitis. Sialoliths are the main cause of obstructive submandibular sialadenitis. Sialolithiasis leads to recurrent painful swelling of the involved gland, which increases in size during meals. Sialoliths occurs commonly in the submandibular gland in 80% of the cases. Pathogenesis of sialolithiasis seems to be based on anatomical position of the salivary duct and gland. This article reports a case of submandibular gland sialolithiasis with chronic sialadenitis of the right side in 55-year-old male patient with recurrent symptoms of pain and swelling during mealtime. Clinical and radiographic findings are important in determining the precise location and size of the sialolith. It helps in establishing the right treatment for the individual patient. Diagnosis of sialolithiasis is based on careful consideration of patient history and thorough clinical examination which demonstrates its typical symptoms.

Keywords: Sialadenitis, sialolith, sialolithiasis, submandibular gland

Introduction

Sialolithiasis also called as mealtime syndrome is the second most common disease of major salivary gland. It occurs commonly in middle-aged adults with the incidence of 12 in 1000 of the adult population. It is common in males than females in the ratio of 2:1.[1]

It most commonly involves the major salivary gland, especially submandibular gland or its duct. Sialoliths occur as a result of deposition of calcium salts around an organic material such as inspissated mucous, ductal epithelial cells, salivary proteins and foreign bodies. Stasis of saliva may lead to acute and chronic infections.[2]

Intraductal stones are comparatively common than intraglandular stones. Hilar stones tend to become very large and round in shape before becoming symptomatic unlike, ductal stones which show elongated form. Diagnosis of sialolithiasis is easy due to typical and obvious clinical features, but in order to establish the right treatment, various imaging studies are mandatory.[3]

We report a case of right submandibular gland sialolithiasis with chronic infection in 55-years-old male with its diagnostic imaging and treatment plan.

Case Report

The 55-year-old male patient reported to the Department of Oral Medicine and Radiology with the chief complaint of swelling and pain in the lower right side of the face since 1 year. Pain was sudden in onset. It was moderate to severe in nature and continuous. Pain aggravated with intake of food and relieved on medication. There was a history of swelling 1 year back. Swelling was initially smaller in size. There was a history of recurrent increase in size of swelling during meals that decreased in size after food. After few months, swelling size became persistent.

Patient also gave the history of epileptic attack 2½ years back and infarct in his temporo-parietal lobe of the brain. He was on antiepileptic and anticoagulant drugs since 2 years. His family and allergy history were non-contributory. He also gave a history of smoking 3-4 cigarettes/day since 10 years but quit the habit 2 years back.

On extra oral examination, facial asymmetry of the right side of the face was present. Diffuse swelling on right submandibular region measuring 3 cm × 2.5 cm in size, roughly oval in shape. Surface over the swelling was normal. Swelling was firm to hard in consistency and tender on palpation [Figure 1].
On intraoral examination, there was no obvious finding. Wharton’s duct on the right side was tender on palpation. On milking the gland, salivary secretion was decreased from the orifice. Other findings showed generalized gingival recession, missing 14 31 34 35 36 41 45 and grossly destructed 16 24 25 46.

Considering the history and examination, a provisional diagnosis of a submandibular sialolithiasis was given. Mandibular true occlusal radiograph was advised, which showed no significant finding. Panoramic view was advised, and it showed a focal radiopaque area of approximately 1 cm in size present anterior to angle of mandible of the right side in the line of the inferior border of mandible [Figure 2]. Computed tomography (CT) sialography was advised which showed dilatation of the duct in the proximal portion with focal collection of dye near the hilum of the gland [Figure 3].

Based on position of the sialolith, sialoadenectomy of right submandibular gland was planned. Submandibular gland and sialolith specimen were obtained and sent for histopathological examination [Figure 4]. Histopathological findings revealed stone with lesional tissue devoid of epithelium, made up of glandular structure having the lobular arrangement of acini separated by fibrous septae. Numerous intralobular ducts along with focal areas of ductal hyperplasia were seen. Focal areas showed acinar degeneration. Dense chronic inflammatory infiltrate predominantly of lymphocytes were seen suggestive of submandibular gland sialolithiasis with chronic sialadenitis. Patient was put on antibiotics and recalled after 1 week for follow-up and swelling had subsided [Figure 5].

Discussion

Sialolithiasis is the formation of calcific concretions within the parenchyma or the ductal system of a major or minor salivary gland. Studies indicate that 92% of sialolithiasis occur in the submandibular gland, 6% in the parotid gland, and 2% in both the sublingual and minor salivary glands. In our case, the sialolith

Figure 1: Profile picture of the patient showing extraoral swelling in the right submandibular region

Figure 2: Panoramic view showing focal radiopaque area on the right inferior border of mandible

Figure 3: 3D view of computed tomography-sialography showing focal collection of dye in the proximal portion of duct with dilatation of the duct

Figure 4: Specimen showing sialolith with right submandibular gland
was associated with submandibular gland. Sialoliths are generally seen in small size and their sizes range from 1 mm to 1 cm. Large salivary gland sialoliths, which are larger than 15 mm are considered rare.

Sialolithiasis usually presents with pain and inflammation but in few cases infection of the main gland may present. The exact etiology and pathogenesis of salivary calculi is unknown. Most accepted retrograde theory proposed for sialolithiasis suggested that, retrograde flow of substances or bacteria within the oral cavity into the salivary ducts lead to the formation of organic nidus that further shows calcification. Salivary stasis, increased alkaline nature of saliva, infection or inflammation, and physical trauma to salivary duct or gland may predispose to calculus formation. The submandibular salivary glands are the most commonly related pair of glands in cases of sialolithiasis (around 80% of prevalence). It is explained by a tortuous structure of submandibular salivary duct. The most narrowed path of the referred duct is named "comma area," which is located near to the duct’s outfall. It facilitates the deposition of minerals, such as calcium, creating a proper nidus for salivary stones. In addition, the submandibular salivary gland presents an alkaline environment with high phosphate concentration, which contributes for the formation of hydroxyapatite.

Common symptoms vary from a painless swelling, moderate discomfort to severe pain with large glandular swelling accompanied by trismus and usually associated with swelling while eating. Patient discussed here was having most of the symptoms.

The differential diagnosis of masses of lymph nodes or the submandibular salivary gland origin can be considered for such swelling in the submandibular region. A careful bimanual, intraoral, and extraoral palpation is the first step in diagnosing and distinguishing between the masses of the submandibular gland and the nonsubmandibular gland origin.

Radiographs are a practical and simple way of investigating the ductal system. The traditional diagnostic methods include plain radiographs (occlusal radiograph), sialography, ultrasound, and scintigraphy. In our report the standard occlusal view did not show any sialolith because the stone was located in the posterior region, therefore, a panoramic radiograph was taken to show the sialolith of this remarkable size. Sialography or other imaging techniques may be required to locate them. Sialo-CT and magnetic resonance sialography are more recently introduced diagnostic tools.

The treatment methods for submandibular duct stones are conservative care, operative removal and the minimal invasive surgery such as extracorporeal shock wave lithotripsy, sialendoscopy, etc. The treatment of sialolithiasis is determined by the location and size of the sialolith. Conservative management should be considered in the cases of small stones. Intraglandular sialoliths require submandibular sialoadenectomy. In the present case, submandibular sialoadenectomy was performed as the sialolith was located in the proximal part of the Wharton’s duct.

**Conclusion**

Clinical and radiographic findings are most important factors in determining the precise location and size of the sialolith. In cases of sialoliths associated with sialadenitis, a penicillinase resistant anti-staphylococcal antibiotic will be preferable with follow-up.

**Clinical significance**

For sialoliths located in the distal area of the Wharton’s duct, simple removal of the stone can be performed but sialolithiasis in the proximal portion of the duct or in the parenchyma of the gland, sialoadenectomy has been performed. Few disadvantages noticed during sialoadenectomy are temporary or permanent injury to marginal branch of facial nerve, scar formation, alteration of dermal sensation and functional problems such as reduction in saliva. The patient reported here was showing no significant complications after surgery.

**References**