CASE REPORT

SIALOLITHIASIS IN REMNANT OF WHARTON'S DUCT- A CASE REPORT WITH RADIOLOGICAL REVIEW

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ABSTRACT

Sialolithiasis in remnant of Wharton's duct after removal of submandibular salivary gland is a rare condition. Stones may become symptomatic in remnant of Wharton's duct. It may be due to remnant from original surgery or via de novo formation. Patients undergoing submandibular gland excision for sialolithiasis should be made aware of this condition. Even the surgeon operating for sialolithiasis should be aware of this condition and should take necessary precautions while performing the surgery. We present a case of 30 years old male patient presenting with sialolithiasis in a remnant of Wharton's duct, two years after the removal of submandibular salivary gland.

Introduction

Commonest disease of salivary gland is sialolithiasis which accounts for 50% of disease of large salivary glands.[1] It affects 12 in 1000 of adult population.[2] Males are twice affected as females. Though children are rarely affected, review of literature reveals 100 cases of submandibular calculi in children of age 3 weeks to 15 years.[3] 80-90% of sialoliths develop in submandibular gland, 5-10% develop in parotid gland and the reminder in the sublingual and minor salivary glands.[2]

Multiple calculi in submandibular salivary gland and simultaneous sialolithiasis in more than one salivary gland are rare. They are usually unilateral and do not cause dry mouth. 40% of parotid and 20% of submandibular calculi are not radio-opaque.[4]

Case Report

A 30 years old male patient presented with mild swelling and pain in right submandibular region for two months. He gave history of removal of right submandibular salivary gland two years ago for a large calculus in right submandibular salivary gland. Ultrasound done with high resolution linear probe on frequency 8-11 mega Hertz showed absence of right submandibular salivary gland (history of prior removal of submandibular gland for sialolithiasis). A linear anechoic tubular structure of calibre 3 mm was noticed in floor of the mouth on right side extending over length of 2 cm. It showed an echo-reflective lesion of size 7 mm causing posterior acoustic shadowing (Fig. 1 and 2). A diagnosis of sialolith within remnant of Wharton's duct was made.

Discussion

Majority of calculi in submandibular gland are located in Wharton's duct, with the ratio of 7:3 compared to intra-glandular stones.[1] Sialolithiasis are usually found in distal portion of the duct or at the hilum of submandi-

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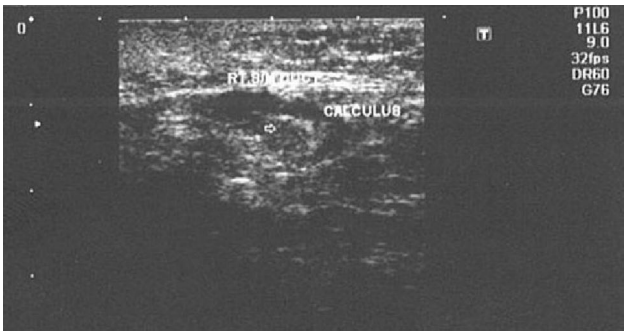

Figure 1: Ultrasound in longitudinal (LS) axis of right submandibular region showing dilated Wharton's duct with echoreflective calculus (marked by arrow) in medial portion of remains of Wharton's duct. (Previous history of removal of right submandibular salivary gland).

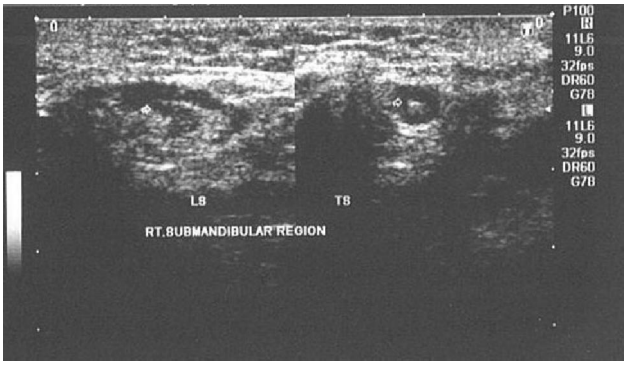

Figure 2: Ultrasound in longitudinal (LS) and transverse (TS) axis of right submandibular region showing dilated Wharton's duct with echoreflective calculus (marked by arrow) in medial portion of remains of Wharton's duct. (Previous history of removal of right submandibular salivary gland).

bular gland with few in parenchyma.[2] Sialolith usually measures from 1 mm to less than 1 cm, rarely it measures more than 1.5 cm. Giant sialoliths are rare.[5] Though it occurs in any age, frequently it is seen in 3rd to 6th decades of life.[2] Usually it presents with pain in submandibular region with associated swelling which may increase during eating.

They usually consist of calcium phosphate with smaller amounts of carbonates in form of hydroxyapatite, with smaller amounts of magnesium, potassium and ammonium.[6] Submandibular stones are 82% inorganic and 18% organic material. Organic material is composed of various carbohydrates and amino acids. Bacterial element is not identified in case of sialolith.[6] Formation of sialolith is a multifactorial event in which disturbances in secretion, microliths and bacteria play a major role. Nuclei of sialoliths are mostly made of inorganic materials like calcium carbonate and calcium phosphate, few studies have shown that the nidus can be bacterial matter.[1] The nucleus is surrounded by laminar layers of both organic and inorganic substances whose contents varies in each sialolith. The outer shell is coated by organic material like glycopreoteins, mucopolysaccharides, lipids and cell detritus.[1] The stone usually grows by 1-1.5 mm each year.[1] There is higher incidence of sialolithiasis in submandibular gland as compared to parotid. The factors involved are:-

a) Longer and wider diameter of Wharton's duct than Stenson's duct.
b) Salivary flow in submandibular gland is against gravity.
c) Secretion of submandibular gland is more alkaline as compared to parotid saliva.
d) Saliva from submandibular gland contains higher quantity of mucin proteins while parotid saliva is entirely serous.
e) Submandibular saliva contains more of calcium and phosphate as compared to in other salivary glands.[2]

Pathogenesis of calculus in remnants of Wharton's duct is difficult to assess. One possibility is that a smaller sized calculus would have been present at time of initial surgery for removal of submandibular salivary gland which later on can grow. The patient must have retrograde flow of saliva from mouth into Wharton's duct to allow increase in size of the calculus. 90% of salivary ducts in distal duct have a sphincter like mechanism which can allow retrograde migration of oral material.[7] Another possibility is stone can develop in the Wharton's duct de novo after excision of submandibular gland. The nucleus may be present before or can develop secondary to subclinical infection in the duct with subsequent layering of nucleus added by retrograde flow of saliva into the duct. Further research and studies are needed to understand pathogenesis of formation of sialolith in remnant of Wharton's duct after removal of submandibular salivary gland.[1]

Sialolithiasis can be diagnosed radiographically, sialography, ultrasonography (US),computed tomography (CT), magnetic resonance imaging (MRI) and MRI sialography. Sialolith can be diagnosed radio-

graphically as a radiopaque structure which may be homogenous or with a laminated structure. Few calculi may be radiolucent. On USG, calculus is seen as well-defined echoreflective round or oval structure causing posterior acoustic shadowing. It is diagnostic method for detecting non-opaque calculi.US can show dilatation of proximal salivary duct. Calculi smaller than 2 mm may not produce acoustic shadowing. Small calculi in intraparenchymal ducts with no duct distension may be missed on US. Hyperechogenic air bubbles with saliva may simulate stones. Unenhanced CT is the best method for detection of radiopaque sialolithiasis within gland or along course of duct. It is useful in differentiating a cluster of fine stones from a large calculus. It cannot detect radiolucent calculi. It has disadvantage of poor visualization of salivary ducts with exposure of patient to ionizing radiation and high cost of examination. Magnetic Resonance Sialography is a non-invasive method with no exposure of patients to ionizing radiation or iodine contrast administration. It can be carried out in acute inflammation of the salivary gland. It is an alternative of digital subtraction sialography, especially in cases of acute sialoadenitis or failure of salivary opening cannulation. It allows precise evaluation of salivary ducts and its branches upto tertiary level. It is performed before and after stimulation with lemon juice. It allows visualization of intraglandular duct dilatation, co-existing stenosis, visualization of tiny calculi not found with USG.[8]

Conclusion

This case emphasizes that even after removal of submandibular salivary gland, sialolith can develop in remnants of Wharton's duct and cause symptoms even after initial surgery.

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