Unusually large sialolith of submandibular gland

Haci Taner Bulut

ABSTRACT

Introduction: Pathologically sialolithiasis is a disease which results in the obstruction of a salivary gland by a sialolith. Sialolith is generally seen in small size and their sizes range from 1 mm to 1 cm. Large salivary gland calculi are infrequent and defined as the size of 1.5 cm or larger. Only a few cases of large sialolith of the submandibular glands have been reported in literature. Imaging methods have an important role in making a diagnosis and in planning further management, operative or otherwise.

Case Report: A 52-year-old male patient with multiple stones in the submandibular gland admitted to our hospital with swelling at submandibular region. Plain films show large stones at the region of submandibular gland. The maximum stone size was 1.8x0.8 mm. Submandibular gland size increased due to the sialoadenitis caused by stones. After surgery, the patient had a nearly normal function of the glands for three months.

Conclusion: The swelling which is seen in the submandibular region most commonly originates from sialolithiasis of submandibular gland, so it should be carefully evaluated by clinicians. Diagnostic imaging methods may complement each other in examining glands with sialolithiasis and may offer a promising diagnostic strategy for treatment and follow-up studies in sialolithiasis.

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CASE REPORT

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Keywords: Sialolithiasis, Large sialolith, Submandibular gland, Imaging

How to cite this article

Bulut HT. Unusually large sialolith of submandibular gland. Int J Case Rep Images 2014;5(9):625–628.

doi:10.5348/ijcri-2014110-CR-10421

INTRODUCTION

Sialolithiasis accounts for the most common cause of diseases of salivary glands [1–7]. Pathologically, sialolithiasis is a disease results in the obstruction of a salivary gland by a sialolith [8, 9]. The clinical symptoms of sialolithiasis are pain and swelling due to enlargement of involved gland [1–3, 7, 9]. Those symptoms help clinicians to diagnose easily. Nonetheless, pain is not seen in all the cases [2, 10]. About 80% of all reported cases of sialolith occur in the submandibular gland [1–3, 6, 9]. The sublingual and minor salivary glands are seldom involved [1–5, 7, 9]. Sialolithiasis is generally found between 30 and 60 years of age and it has a frequent prevalence in male patients [9, 11]. Sialoliths are generally seen in small size and their sizes range from 1 mm to 1 cm. Large salivary gland sialolith which is large 15 mm are considered rare [7, 9]. Only a few cases of large sialolith of the submandibular or parotid glands have been reported in literature. Imaging methods have an important role in making a diagnosis and in planning management. Plain radiography and sialography, magnetic resonance imaging (MRI) scan, computed tomography (CT) scan, and ultrasound all have a role [12]. The aim of this study is to evaluate the clinical and radiographical findings of the patient with large sialolith of submandibular gland.

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Received: 05 June 2014

Accepted: 27 June 2014

Published: 01 September 2014

CASE REPORT

A 52-year-old male patient admitted to our hospital with painless swelling at submandibular region. Extra-oral examination revealed swelling and palpable mass. In intraoral examination, bimanual palpation revealed a hard elongated mass and multiple stones in a large size. Plain film, axial CT, and MRI scans were obtained for radiological examination. Stone location, shape, and size were estimated on plain film, MRI and CT scans. Posterior-anterior and sagittal plain films show large calculi at the region of submandibular gland (Figure 1). Non-enhanced axial CT-scan showed large hyperdense masses (sialoliths), localized within the left Wharton duct and enlargement of the affected submandibular gland. The maximum stone size was 1.8x0.8 mm, as measured directly on the axial CT scan (Figure 2). Submandibular gland size increased due to the sialadenitis caused by stones. Due to sialadenitis, coronal Short tau inversion recovery (STIR) and enhanced axial T1-weighted magnetic resonance images of affected gland showed higher signal intensity compared with normal gland on right side (Figure 3). Sialoliths removed with surgery. After surgery, the patient had a nearly normal function of the glands for three months.

Figure 1: A 52-year-old male with left submandibular sialolithiasis. Posterior-anterior (A) Sagittal, (B) Plain films showing large stones at the region of submandibular gland.

Figure 2: (A, B) Non-enhanced axial computed tomography scan showing sialoliths and enlargement of the affected submandibular gland.

Figure 3: Coronal short tau inversion recovery (STIR) (A) Enhanced T1-weighted, (B) Magnetic resonance images of affected gland showing higher signal intensity compared with normal gland on right side.

DISCUSSION

The most widespread illnesses of the salivary gland are sialoliths [1, 3, 9, 13]. Sialoliths are generally seen in small size and their sizes range from 1 mm to 1 cm [1–3, 5, 9]. The mean size of sialoliths is reported as 6 to 9 mm. They infrequently measure more than 1.5 cm. Large salivary gland calculi are infrequent and defined as the size of 1.5 cm or larger [2, 9–11]. Most of the studies have conducted that the common symptoms of sialoliths are recurrent pain and swelling of the associated gland, because sialoliths generally does not block the flow of saliva fully [1–3]. Nonetheless, large sialoliths have been frequently reported in the body of salivary glands, they have infrequently been described in the salivary ducts, particularly without any complaints from the patients [2, 4, 14]. In this study, clinical and radiological features of one case which have large sialoliths in the size of 1.8 cm were presented. The sialoliths were located into Wharton ducts and the patient complained painless swelling. Some uncommon large salivary stones may be noticed unless the patient has a long history, due to the fact that lesions are usually asymptomatic. It is conducted that the stones may expand in the proportion of about 1 to 1.5 mm each year [2]. Hence, it is possible to presume that sialoliths of our case began to develop many years ago.

In the diagnosis of sialoliths, history and careful examination come to the fore. Pain and swelling of involved gland at the time of meal are of great importance. Bimanual examination in the floor of the mouth may show a palpable stone in a great number of cases of submandibular sialoliths. Bimanual palpation of the gland is very useful because a uniformly solid and hard gland indicates a hypo-functional or non-functional gland [1, 2, 8]. A case in the this study has a history of painless swelling in the floor of the mouth at mealtimes. Extra-oral examination revealed swelling and palpable mass. In intraoral examination, bimanual palpation revealed a hard elongated mass and multiple stones in a large size.

In the diagnoses of sialolithiasis, imaging methods are very useful. Plain radiographs are useful in showing radiopaque stones. It is very uncommon for patients to have a combination of radiopaque and radiolucent stones and 40% of parotid stones may be radiolucent [15]. Sialography is thus useful in patients showing signs of sialadenitis related to radiolucent stones or deep submandibular stones. Sialography is, however, contraindicated in acute infection or in significant patient contrast allergy. Nowadays, magnetic resonance sialography (MR sialography) imaging is recommended in diagnosis of sialoliths, but this method is not appropriate to see the inner duct of the salivary glands. Developed in the 1990's as an endoscopic method, sialoendoscopy technique enables clinicians to examine the ductal system completely and it can be used not only for diagnosis but also for treatment [16]. The CT scan is useful in identifying small calculi within the salivary gland or duct. It can also show localization and number of stones in the gland and measure size of stones. Contrast-enhanced CT scan has a potential to show enlargement of gland due to sialadenitis. Features of the submandibular glands affected by sialolithiasis can well evaluate with MRI scan [12]. Hence, it can possible to differentiate, acute or chronic stage of sialadenitis with MRI scan. It can also show the location of stones and shapes of ducts. In this context, MRI scan using T1-weighted and STIR sequences, can provide effective information about the pathologic status of the gland parenchyma affected by sialolithiasis [12]. Moreover, the extent, acute and chronic nature of this obstruction may reflect by MRI findings of the gland parenchyma [12]. In this study, the sialoliths were observed clearly in plain radiographs, but estimation of size and location of stones is limited. The MRI scan and CT scan were suitable in precise preoperative estimation of stone's size and location. These results suggest that MRI features may reflect acute obstruction, and a combination of CT and MRI scans in examining glands with sialolithiasis may offer a promising diagnostic strategy for treatment and follow-up studies in sialolithiasis.

Different treatment options may be selected according to the size and location of the sialolith. The treatment of choice of small sialolith should be medical instead of surgical. However, if the stone is too large or located in the proximal of duct, piezoelectric extracorporeal shock wave lithotripsy or surgical removal of the stone or gland may be required [1–3]. Sialoendoscopy is a new way and minimally invasive technique for treating obstructions of the ductal system and can be used with operation in large salivary stones [17]. Recurrent or continuous obstruction of the salivary duct may lead to acute or chronic sialadenitis or even to the perforation of the oral mucosa [18]. In this case, the sialoliths were removed by surgical excision. The clinicians should evaluate carefully the painful or painless swellings in submandibular area. This condition seems to be the most common disease in submandibular gland and Wharton duct due to the

presence of gland lithias. Large submandibular sialoliths should be treated by appropriate approach to avoid possible severe postoperative complications.

CONCLUSION

The swelling which is seen in the submandibular region most commonly originates from sialolithiasis of submandibular gland, so it should be carefully evaluated by clinicians. Diagnostic imaging methods may complement each other in examining glands with sialolithiasis and may offer a promising diagnostic strategy for treatment and follow-up studies in sialolithiasis.

Author Contributions

Haci Taner Bulut – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

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Article citation: Bulut HT. Unusually large sialolith of submandibular gland. *Int J Case Rep Images* 2014;5(9):625–628.

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