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Case Report

Giant submandibular sialolith of left side with unique branching pattern: A case report

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ABSTRACT

Background: Salivary gland calculi account for the most common disease of the salivary glands. The majority of sialoliths occur in the submandibular gland or its duct and are a common cause of acute and chronic infections. Sialoliths attaining a size of more than 1.5 centimetres are rare.
Materials and Methods: This case report presents a middle-aged male patient with enlarged tender swelling with left submandibular gland. Mandibular occlusal radiography followed by computed tomography was done. Surgical removal of stone was done under local anaesthesia.
Results: Conventional mandibular cross-sectional occlusal radiograph was taken and it revealed a dense radio-opacity on left side with branching with the posterior extent not seen and other round shaped radio-opacity superior to giant stone which is superimposed and visible on careful examination. Computed tomography revealed a single hyperdense mass in left submandibular region with obvious branching seen. Medical management followed by left submandibular intraoral stone extraction along with second round shaped sialolith was performed and it revealed one large stone measuring 2.5 centimetres with branching & other one with 0.7 centimetres. The patient had smooth recovery with no complications on follow-up.
Conclusion: In the present case our patient presented with typical clinical and radiographic findings of a rare giant intraductal submandibular sialolith with lateral branching. Calcification in the accessory salivary gland was unique in our case. The present case illustrates the need for proper diagnosis and treatment of choice in the case of salivary gland diseases.

1. Introduction

Sialolith is one of the most common diseases of salivary glands. It is estimated to have a frequency of 0.15% in the adult population with slight male predilection.[1–3] Sialoliths are always found in the distal portion of the duct or at the hilum of the submandibular gland with a few in parenchyma.[4] Sialolith is a calcified organic matter that is formed within the secretory system of the salivary glands. A nidus of the salivary organic material becomes calcified and forms a sialolith.[5] Higher rate of giant sialolith formation in the gland is due to the tortuous course of the Wharton's duct, high calcium and phosphate levels, and dependent position of the gland which makes it prone to stasis.[6] Clinical and radiographic examination are required for the correct diagnosis and treatment plan. Here we report an interesting case of a large sized sialolith in the parenchyma of the left submandibular gland.

2. Case Report

A 37-year-old male patient reported to Department of Oral Medicine and Radiology with the chief complaint of mild pain, excessive salivation and bad breath from his mouth for past 1 year. Patient was relatively alright 1 year back, then he started experiencing a small swelling in the floor of mouth on left side. Initially the size was small and gradually it increased to the present size. He experienced recurrent swelling & mild pain in the same region with excess salivary flow with some salty discharge from swelling and bad breath

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due to it.

On intraoral examination, Single, ovoid, dome-shaped intraoral swelling of size approximately 1×3 cms seen in floor of mouth extending from midline to lingual frenum towards left side along the floor of mouth upto the lingual vestibule (Figure 1). Overlying surface appeared stretched, pale pink & translucent with no pulsations or varicosities. Borders were diffuse which blended along normal adjacent mucosa. Swelling was afebrile, tender and was without any discharge on palpation. It was non-compressible, fluctuant & mobile, not attached to any underlying structures. By bimanual palpation of submandibular salivary gland was tender on palpation. Opening of Wharton's duct was raised and inflamed on palpation.

Fig. 1: Intra-oral dome shaped swelling of left submandibular region

A provisional diagnosis of submandibular sialolith of left side was made. Conventional mandibular cross-sectional occlusal radiograph was taken and it revealed a dense radio-opacity on left side of mid-line. It was cylindrical in shape with pointed end anteriorly, branching seen laterally and running posteriorly with posterior extent not visible on radiograph suggestive of salivary gland calculi of submandibular gland (Figure 2). On careful examination of occlusal radiograph, we found out another round shaped radio-opacity superior to giant stone which was less dense.

Computed tomography was advised as posterior extent was not visible on conventional radiograph. Axial section of CT scan showed cylindrical shaped dense radio-opacity with lateral branching on left side of mandibular arch along the medial border of mandible of size 1×2.5 cms suggestive of giant submandibular gland duct calculi. Coronal section of CT showed well defined radiopacity on left side of submandibular region (Figure 3). Patient was referred to oral surgeon for removal of giant submandibular sialolith. A submucosal incision was given on floor of mouth on

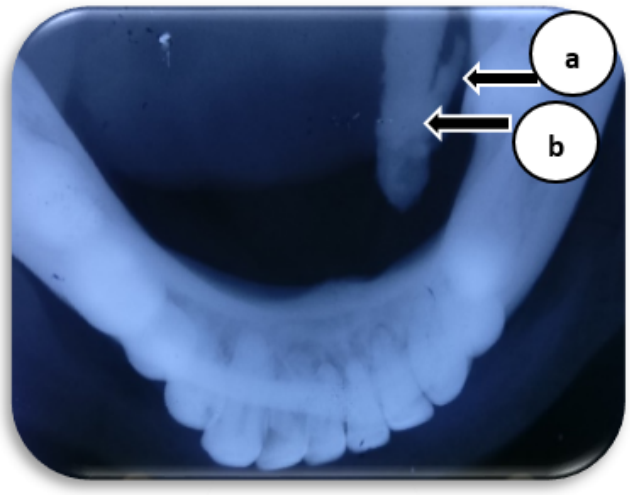

Fig. 2: Mandibular occlusal radiograph showing **(a)** Submandibular sialolith of left side with lateral branching **(b)** Round shaped radio-opacity superior to giant stone

left side at the site of pathology and submucosal flap was raised (Figure 4), tissues cleared to visualise the stone. After removal of stones, submucosal resorbable sutures were placed to close the site (Figure 5). The sialoliths was measuring 2.5cms and 1cm and were yellow to white in colour with rough granular surface texture (Figure 6). Vicryl 3-0 sutures were given for approximation of wound and wound closure (Figure 7). Following the post-operative instructions patient was recalled after seven days for follow-up. The healing was found to be satisfactory and salivary flow was found to be normal and patient was relieved of the symptom.

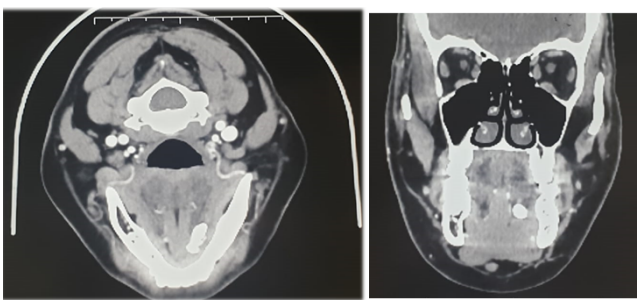

Fig. 3: Axial & coronal section of computed tomography showing radio-opacity in submandibular region of left side

3. Discussion

Sialolithiasis is a rare disease with an estimated frequency of 1.2% in the adult population with male predilection.[1] In year 2017, Thimmarasa et al reported a case of giant submandibular sialolith measuring 35mm or more in an adult female which the first case reported in a literature.[7]

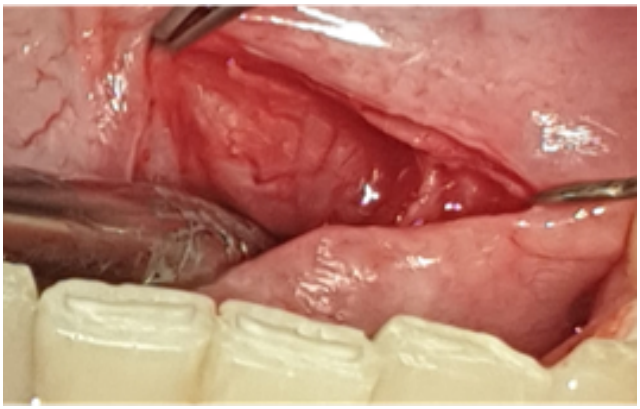

Fig. 4: Submucosal flap reflected

Fig. 7: Final closure with intermittent suturing

Fig. 5: Intra oral picture after stone removal

Fig. 6: Giant stone measuring 2.5cms and other one measuring 1cms

The disease can occur at any age, but it appears more frequently in the third to sixth decades of life.[1] The majority of the sialoliths are formed from phosphate and oxalate salts. They are also composed of varying ratios of organic and inorganic substances. The organic substances are glycoproteins, mucopolysaccharide's and cellular debris, inorganic substances are mainly calcium carbonate and calcium phosphate.[8]

Gaint sialoliths are a rare finding and their size varies from approximately from 1.5 cm-7 cm. A review of literature in 2007[9] found 16 reported cases of salivary stones having a size up to 35 mm with the largest stone reported measured 55 mm. In our case we report two sialoliths one over other which is very unique about the case and is not been seen in any literature so far. In year 2016, Omimah reported that there are only few cases documented in the literature so far.[10] The clinical and radiographic methods for diagnosis include conventional radiographs like occlusal and panoramic radiographs, sialography, ultrasound and computed tomography.[5] Mandibular occlusal radiograph is best conventional method to visualize radiopaque stones.

Ultrasonography is widely reported to be useful in detecting salivary stones. As many as 90% of all stones larger than 2 mm can be detected ultrasonographically.[11] Sialendoscopy is an accepted intervention, for the elimination of giant salivary sialolith. The characteristic ultrasound reveals an echo reflection with marked dorsal sound shadowing. Computed tomography is also highly diagnostic.[12] There are different treatment options available regarding removal of submandibular stones. If the stone is smaller in size, it can be milked out through ductal orifice of submandibular gland by bimanual palpation. If it is larger in size, surgical excision is the only preferred treatment of choice. Complications from sialolith include acute sialadenitis, ductal stricture, ductal dilatation and recurrent infections.

4. Conclusion

In the present case our patient presented with typical clinical and radiographic findings of a rare giant intraductal submandibular sialolith with lateral branching and round shaped. Calcification in the accessory salivary gland was unique in our case. The present case illustrates the need for proper diagnosis and treatment of choice in the case of salivary gland diseases. Post-operative follow up is essential to ensure that the patient is symptom free and stone free long term. To conclude detailed history taking, clinical and radiographic examination are important in determining the precise location and size of the sialolith in order to indicate the right treatment for the patient.

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