Case Report

Giant sialolith of submandibular gland: A case report

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ABSTRACT

Sialolithiasis is the most common disease of the salivary glands after mumps. The main site of salivary gland stones formation is the submandibular gland, followed by parotid and sublingual gland. Here, we report the case of a 70-year-old male with giant submandibular gland calculus of size $3.3 \text{ cm} \times 2.1 \text{ cm} \times 1.5 \text{ cm}$ with an inflamed gland. We reviewed the literature and found that submandibular gland sialolith measuring 10 mm in greatest dimension is very common, but large sialolith like the one seen in this case is relatively rare and only 18 cases have been reported in the literature with calculus size >3.0 cm till date. The largest reported sialolith was 6 cm in length and had a weight of 50 g. Non-enhanced computed tomography is recommended when salivary stones are suspected.

Key words: Giant submandibular gland sialolith, Non-enhanced computed tomography, Radiograph, Sialolithiasis

Sialolithiasis (also termed salivary calculi or salivary stones) is a condition where a calcified mass or sialolith forms within a salivary gland. Sialolithiasis is considered to be the most common salivary gland disorder, occurs in middleaged adults, and accounts for about 1.2% of unilateral major salivary gland swelling [1]. >80% of the sialoliths occur in the submandibular gland or its duct, 6% in the parotid gland, and 2% in the sublingual gland or minor salivary glands [1-3].

The salivary stones are typically composed of calcium phosphate or calcium carbonate. Some factors which favor the stone formation and are inherent to the submandibular gland is longer and larger caliber duct, flow against gravity, slower flow rates, and higher alkalinity along with higher mucin and calcium content of the saliva [4]. Here, we present the case of giant submandibular gland calculus with inflammation of gland.

CASE REPORT

A 70-year-old male was referred to the radiology department with a chief complaint of pain and swelling in the left submandibular region for 10 months. On per oral examination, a 4 cm tender and firm mass were palpated bimanually in the floor of the mouth near the left submandibular gland. The ductal opening was not dilated.

Radiograph of the neck in the anteroposterior and lateral projection shows a large calcification in the left submandibular region (Fig. 1). Non-enhanced computed tomography (NECT) of the face and neck region showed a calculus of size 2.1 cm \times 1.5 cm \times 3.3 cm (Anteroposterior, transverse, and craniocaudal dimensions, respectively) within the left submandibular gland (Fig. 2). The submandibular gland was also enlarged in size. Mass effect noted in the form of medial displacement of the left palatine tonsil, effacement of the left

oropharynx, pharyngeal mucosal space, and retropharyngeal mucosal space.

After establishing the diagnosis radiologically, the submandibular gland was excised through an incision on the skin crease 3 cm below the lower border of the mandible and directly over the palpable left submandibular gland. The gland with the calculus was dissected. The calculus was taken out from the submandibular gland and the free duct of the submandibular gland was ligated to the mucosa. After which, the wound was closed in layers after an insertion of a drain. The specimen was sent for histopathology which revealed a bulky submandibular gland, measuring 4.0 cm in its largest dimension with the calculus measuring $3.2 \text{ cm} \times 2.2 \text{ cm} \times 1.5 \text{ cm}$. The calculus was oblong, yellowish in color and had a granular surface (Fig. 3). The patient was recalled after 3 weeks, there was no recurrence and the patient was asymptomatic.

DISCUSSION

Sialolithiasis is the second most common disease of the salivary glands after mumps [1]. More than 80% of the sialoliths occur in the submandibular gland or its duct, 6% in the parotid gland, and 2% in the sublingual gland or minor salivary glands [1-3]. The exact location of the sialolith is the parenchyma of the left submandibular gland.

Some factors inherent to the submandibular gland tend to favor stone formation there such as longer and larger caliber duct, flow against gravity, slower flow rates, and higher alkalinity along with higher mucin and calcium content of the saliva [4]. The submandibular gland hosts the largest stones with the largest reported one being 6 cm in length [5].

Sialolith larger than 1.5 cm has been deemed "giant sialoliths" and only 13 cases of sialoliths >55 mm are reported [6].

Submandibular gland sialolith measuring up to 10 mm in greatest dimension is very common, but large sialoliths like the one seen in this case are rare [7]. Sialolithiasis of the submandibular gland can be completely asymptomatic [8]. Common symptoms vary from a painless swelling, moderate discomfort to severe pain with large glandular swelling accompanied by trismus, and usually associated with eating [9].

NECT and ultrasound (USG) can demonstrate sialoliths with high accuracy and can correctly localize them anatomically, but USG is less accurate than NECT in distinguishing multiple clusters of stones from single large stones [10,11]. NECT scan provides a precise location of calculus in the duct system. As USG has limited sensitivity and limited negative predictive value, it does not allow a reliable exclusion of small salivary gland calculi. Therefore, further, diagnostic investigations such as radiograph



Figure 1: Radiograph of cervical spine lateral and anteroposterior projection showed radio-opacity in the left submandibular region



Figure 2: Non-enhanced computed tomography of the face and neck region axial section shows hyperdense mass in the left submandibular region





and NECT are recommended to detect calculi in patients with normal sonographic findings and suspected lithiasis [12,13].

Submandibular gland calculi have been reported to be radiopaque in 80–94.7% of cases and can be seen on plain films [3,13,14]. However, if an abscess or an inflammatory process is suspected, contrast may be administered after identifying the stone on unenhanced scans [10]. Excision of the submandibular gland carries a risk of permanent or temporary marginal mandibular nerve palsy [15].

CONCLUSION

Occlusal radiographs are extremely useful in showing radiopaque stones. Major salivary glands are affected by a variety of diseases, few of which show characteristic features on imaging. NECT is the mainstay of imaging in sialolithiasis to confirm the diagnosis and evaluate the gland and adjacent changes. Newer techniques such as scintigraphy, NECT, and sialoendoscopy have revolutionized the diagnostic aspect of sialolithiasis and should be used for the accurate prediction of the entity.

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