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Submandibular Gland Sialolithiasis - A Case Report

KEYWORDS

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ABSTRACT *Sialolithiasis is one of the most common diseases of the salivary glands. It is a condition characterized by an obstructive phenomenon in a salivary gland or in its excretory duct due to a calculus. The clinical presentation is usually characterized by local swelling, pain, infection of the affected area, and dilation of the salivary duct. Sialolithiasis usually affects adults between the third and fourth decades of life, with a frequency of 12:1000. Although the frequency of sialolithiasis is relatively high, the occurrence of giant sialoliths, larger than 1.5 cm in any diameter, is rare. For this reason few studies are found in the pertinent medical literature.*

This report describes a case of giant sialolith in a 26-years-old male, addressing the clinical features, the diagnosis and the surgical procedure performed to restore salivary flow.

Salivary gland lithiasis (Sialolithiasis) is the most common diseases of the major salivary glands after mumps. It accounts for approximately more than 50 % of all salivary gland diseases.[1] The estimated rate of occurrence is 12 of 1000 persons in adult population every year, with the incidence being higher in males (ratio of 2:1) with the most probable age group being 30 to 60 years.[1] Most salivary calculi (80% - 95%) are found in the submandibular gland, whereas 5% - 20% of the cases are encountered in the Parotid gland.[2] The duct is more commonly affected than the parenchyma (ratio being 7:3).[3]

The presence of multiple calculi in submandibular gland is a rare event, as is simultaneous stone formation in multiple salivary system.[4] Radiopacity is not a feature in 40% of Parotid and 20% of submandibular stones; therefore different imaging techniques may be needed to localize them.[5] Microliths may be expelled or continue to grow until the duct is obstructed. Clinically, they are round or ovoid, rough or smooth and of a yellowish colour. They mainly contain Calcium phosphate with smaller amounts of carbonates in the form of hydroxyapatite, with smaller amounts of Magnesium, Potassium and Ammonia.[6] Submandibular stones are composed of 82% inorganic and 18% organic material, whereas Parotid stones are composed of 49% inorganic and 51% organic material.[4]

Sialoliths commonly measure between 5 and 10 mm in size, and all stones over 10 mm can be reported as sialoliths of unusual size.[7]

We report a case of sialolith measuring 19 mm in length, highlighting the management aspects with minimal glandular dysfunction.

A CASE REPORT

A 26 year old man reported complaints of severe pain and swelling in the right submandibular region, while having food, since last five years. There was an episode of fever and associated swelling in the same region a week before he reported to the hospital. Swelling was controlled after a course of antibiotics for seven days. Presently, his pain was aggravated on eating. There was no weight loss, anorexia or burning sensation in the oral cavity.

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On extraoral examination, no gross facial asymmetry was noted. Intraoral examination revealed no swelling of the overlying mucosa in the affected region. On bimanual palpation, a stony hard obstruction was felt in the right submandibular duct region.

FIGURE 1. Preoperative frontal view of the patient

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Radiographic examination with a mandibular occlusal radiograph along with a panoramic radiograph was carried out. It revealed an unusually large sialolith, approximately 2 cm in its greatest length in the right submandibular region. A Cone Beam Computed Tomography (CBCT) was done to exactly localize the sialolith and rule out any radiolucent stones. A diagnosis of Sialadenitis secondary to unusually large sialolithiasis of the right submandibular duct was made.

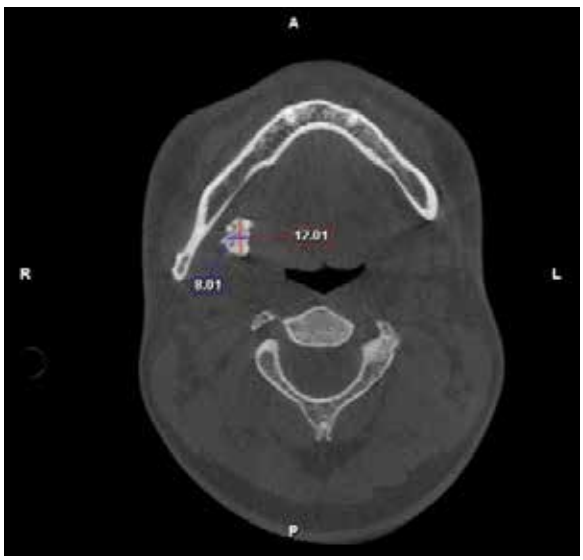

FIGURE 2.Preoperative Cone Beam Computed Tomography (CBCT)

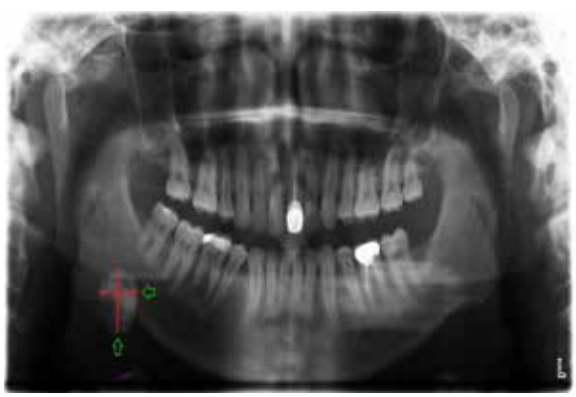

FIGURE 3.Preoperative Orthopantogram (OPG)

The diagnosis was explained to the patient. After induction of local anesthesia, Sialolithotomy was performed via intraoral approach. An linear incision was placed on the right submandibular region on floor of the mouth, directly over the sialolith. Blunt dissection was carried out and the Submandibular gland duct (Wharton's duct) was identified. The duct was separated and the sialolith was delivered out of the opening that was made.

After successful removal was accomplished, the area was thoroughly irrigated with saline and Povidone-Iodine solution. The wound was then close using No. 4 Vicryl interrupted sutures. The Sialolith recovered measured approximately 19 mm in length.

The scientific importance of the case was explained to the patient and his consent was taken for publication in scientific literature.

FIGURE 4. Transoral Sialolithotomy performed by placing incision directly over the stone

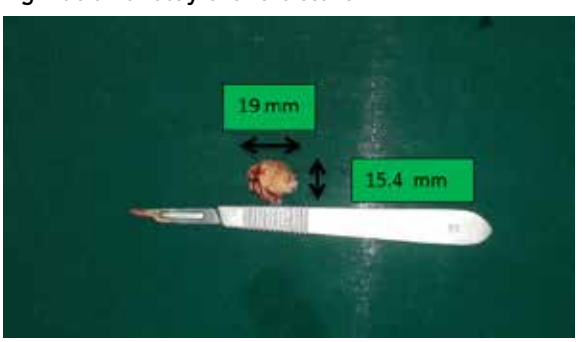

FIGURE 5. The Sialolith after it was delievered out

DISCUSSION

Although large sialoliths have been reported in the body of salivary glands in the literature, they have rarely been reported in the salivary duct. Brusati and Fiamminghi removed a sialolith from the left submandibular duct of a 55 year old man measuring 27 x 31 mm.[8] Leung et al removed a sialolith of 14 x 9 mm from right submandibular duct.[1] The sialolith removed in this case was comparable to these. Interestingly, the patient age in the current study was just 26 years old, as compared to the various studies that have been reported in the literature.

ETIOPATHOGENESIS

Salivary duct lithogenesis mostly proceeds due to relative stagnation of calcium rich saliva. They are thought to occur as a result of deposition of calcium salts around an initial organic nidus consisting of altered salivary mucins, bacteria and desquamated epithelial cells.[5] For stone formation, it is probable that the intermittent stasis of saliva causes a shift of the mucoid element into a gel form, which in turn provides the framework for stone formation.[6] Traditional theories propagate the idea of a bi-phasic sialolith formation: a central core and a layered periphery.[3] The central core forms due to salt precipitation, while the second phase incorporates both the organic and inorganic forms.[9]

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Another theory proposed that there is a sudden intermittent increase in the bicarbonate content of saliva, leading to alteration in calcium phosphate solubility and henceforth, their precipitation.[6,10] A retrograde theory has also been put forward where substances or bacteria in the oral cavity might migrate into salivary ducts and become nidus for further calcification.[3]

Submandibular sialolithiasis is more common as its saliva is (1) more alkaline, (2) has an increased concentration of calcium and phosphate, (3) and has a higher content of mucus as compared to saliva of other glands. In addition, the Wharton's duct is longer along with an antigravity flow.[1]

Stone formation is not associated with systemic calcium or phosphate imbalance.[6] Electrolytes and parathyroid hormone studies in patients with sialolithiasis have not shown abnormalities.[11] Gout is the only systemic illness that predisposes a patient to sialolithiasis[11], although in gout the stones are predominantly made of Uric acid.[6]

CLINICAL FINDINGS

Sialolithiasis typically causes pain and swelling of the involved salivary gland by obstructing the food related surge of salivary secretion. Calculi may cause stasis of saliva, leading to bacterial ascent into the gland parenchyma[6], and therefore infection, pain and swelling of the gland. Some may be asymptomatic until the stone passes forward and is visible in the duct orifice. Long term obstruction may lead to atrophy of the gland with resultant lack of secretory function and ultimately fibrosis.[6]

DIAGNOSIS

Careful history and examination are essential tools in diagnosis of Sialoliths. Pain and swelling of the concerned gland during mealtimes are important. Complete obstruction causes nagging persistent pain and pus may be seen draining from the duct, associated with systemic infection.[12]

Bimanual palpation of floor of the mouth, in posterior to anterior direction, reveals a stone in large number of cases of submandibular calculi. For Parotid stones, careful intraoral palpation around Stenson's duct orifice may reveal a stone, while deeper stones are often not palpable.[6]

Imaging studies are very useful in diagnosing Sialoliths. Occlusal radiographs are useful in showing radiopaque stones. Sialography is useful in patients showing signs of sialadenitis related to radiolucent stones or deep submandibular/parotid stones. Other diagnostic methods include the use of Computed Tomography, Cone Beam Computed Tomography, MRI or Sialadenoscopy.

TREATMENT

Various treatment modalities have been in practice for Sialolithiasis. Patients may be benefitted from a trial of conservative management, especially if the stone is small.[6] The patient must be well hydrated and the clinician must apply moist warm heat and gland massage, while sialagogues are used to promote saliva production and flush the stone out.

Almost half of the submandibular sialoliths lie in the distal third of the duct. They are amenable to simple surgical release through an incision in the floor of the mouth, which is relatively simple to perform and not usually associated with complications.[13] If the stone is sufficiently forward, it can be milked and manipulated through the duct orifice. This can be done with the aid of lacrimal probes and dilators to open the duct.

The duct may need opening to retrieve the stone. This involves a transoral approach where an incision is made directly onto the stone. In this way more posterior stones, 1-2 cm from the punctum, can be removed by cutting in the longitudinal axis of the duct. Care is taken as the lingual nerve lies deep, but in close association with the submandibular duct posteriorly.[14]

If the gland is damaged by recurrent infection and fibrosis, or calculi have formed within the gland, it may require removal.

Alternative methods of treatment have emerged such as the use of Extracorporeal Shock Wave Lithotripsy (ESWL) and more recently the use of Endoscopic Intracorporeal Shockwave Lithotripsy (EISWL), in which shockwaves are delivered directly to the surface of the stone lodged within the duct without damaging adjacent tissue (piezoelectric principle).

CONCLUSION

Unusually large sialoliths pose a diagnostic and therapeutic challenge for the clinician. The choice of surgical approach to access the sialolith requires careful evaluation and localization. Newer treatment modalities such as ESWL and EISWL are effective alternatives to the conventional surgical excision for smaller sialoliths. However, Transoral Sialolithotomy still remains the mainstay of management.

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