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Giant Submandibular Sialolith of Remarkable Size in the Comma Area of Wharton's Duct: A Case Report

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Sialolithiasis accounts for more than 50% of the salivary gland diseases, with an estimated 12 of 1,000 persons in the adult population being affected every year.[1] Most salivary calculi (80%-95%) occur in the submandibular gland, whereas 5% to 20% are found in the parotid gland.[2] The sublingual gland and minor salivary glands are rarely (1%-2%) affected.[2] Male patients are affected twice as much as female patients.[3]

Multiple calculi in the submandibular gland are rare, as is simultaneous lithiasis in more than 1 salivary system.[4] Radiopacity is not a feature in 40% of parotid and 20% of submandibular stones; therefore sialography or other imaging techniques (computed tomography scan, ultrasound) may be required to locate them.[3] Clinically, the stones are round or ovoid, rough or smooth, and of a yellowish color. They consist of mainly calcium phosphate with small amounts of carbonates in the form of hydroxyapatite, as well as smaller amounts of magnesium, potassium, and ammonia.[5] 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.[6] Giant sialoliths measuring more than 35 mm are rare, with only around 16 cases published in the literature. Ninety-four percent of the giant sialoliths reported were in the submandibular gland.[7]

We report a case of a sialolith in the submandibular duct measuring 72 mm in length and weighing 45.8 g, highlighting the management aspects of this giant sialolith with minimal glandular dysfunction.

Report of a Case

A 60-year-old man reported complaints of severe pain and swelling in the left lower submandibular region for a duration of 1 month. There were episodes of pain in the same region for last 2 years but of a moderate variety that the patient could tolerate. Presently, his pain was intermittent, of the pricking type and sharp in nature, radiating to the tongue. The pain became aggravated during eating and was relieved by rest. Swelling was gradual in onset, progressing to the present size. There were occasions of mild swelling during meals for the last 6 months, which the patient had been ignoring. There was no associated history of fever, malaise, weight loss, anorexia, or burning sensation in the oral cavity.

On extraoral examination, the patient showed diffuse swelling over the left submandibular region measuring 8 × 6 cm, with normal overlying skin (Fig 1). There were no signs of sinus, fistula, or ulceration in the affected region. The swelling was warm and tender on palpation with a firm consistency. No nodular or matting characteristics were noted. Intraoral examination showed inflammation and induration of the left floor of the mouth with absent salivary flow from the left Wharton's duct orifice. However, no pus discharge was detected from the duct orifice. The left submandibular gland was tender on bimanual palpation.

Radiographic examination with a panoramic radiograph showed a giant sialolith, cylindrical in shape and approximately 6.5 cm in length, in the left submandibular region extending to the floor of the mouth (Fig 2). A diagnosis of giant sialolithiasis of the left submandibular duct in the comma area with chronic sialadenitis of the left submandibular gland was made.

After induction of local anesthesia, sialolithotomy with sialodochoplasty was performed via an intraoral approach. Upward and medial pressure was applied to the submandibular gland, and an incision was placed directly over the sialolith to expose it. After sufficiently mobilizing the sialolith, we attempted to deliver it out through the opening. The larger portion of the sialolith was delivered out first with the sinus forceps; however, the fragmented portion

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FIGURE 1. Preoperative front view showing swelling in left submandibular region.

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remaining thereafter could not be visualized clearly. Thorough exploration and continuous massaging of the submandibular gland with upward and medial pressure were required to clearly visualize and mobilize the distal portions of the stone with the sinus forceps. We appreciated that the stone was not a cylindrical structure but, rather, had multiple fingerlike projections in the distal half, which occurred like undercuts when we attempted to remove the stone. Complete removal of the distal half of the sialolith was finally accomplished with a curved sinus forceps (Fig 3).

After successful removal of the sialolith as confirmed by a panoramic radiograph, a 1-cm-wide opening remained, which was marsupialized intraorally with No. 4 silk sutures

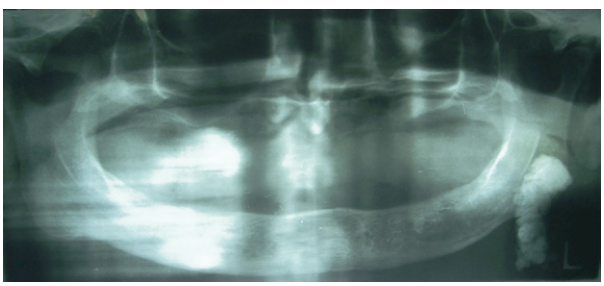

FIGURE 2. Preoperative panoramic radiograph showing giant sialolith in left submandibular region.

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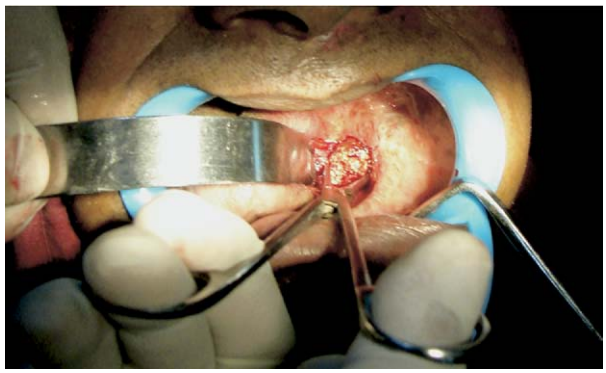

FIGURE 3. Transoral sialolithotomy performed by placing incision directly over stone with subsequent delivery of sialolith in fragments by sinus forceps.

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placed along its margins. Flow of submandibular gland fluid was visualized from the opening created. The sialolith recovered measured approximately 72 mm in length (on realignment) and weighed 45.8 g (Fig 4). Postoperatively, the intraoral opening was regularly irrigated, and after 2 months, the opening was surgically reduced to 3 mm (Fig 5). After a 2-year follow-up, the patient was asymptomatic with satisfactory glandular function. Flow of saliva occurred from the new opening created during stone retrieval.

We explained the scientific importance of the patient's disease to him, and he provided consent for publication of his case in the scientific literature.

Discussion

Giant sialoliths are rare findings in clinical oral pathology, with sizes ranging from 35 to 70 mm and all of them occurring in male patients.[7-11] Although giant sialoliths have been reported in the salivary glands, they have rarely been reported in the salivary ducts.[1] The largest sialolith reported in the literature was 70 mm in length in Wharton's duct and was

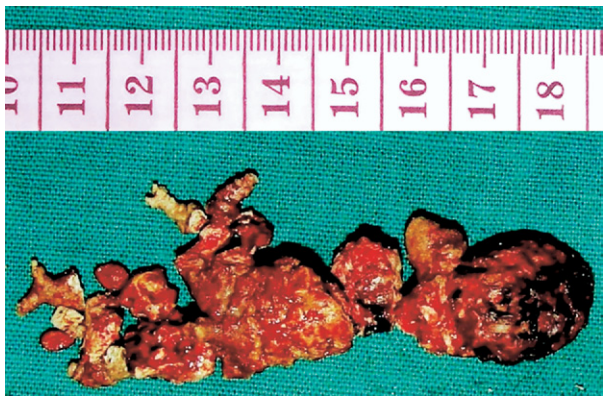

FIGURE 4. Giant sialolith realigned and measured to be approximately 72 mm in length and 45.8 g in weight.

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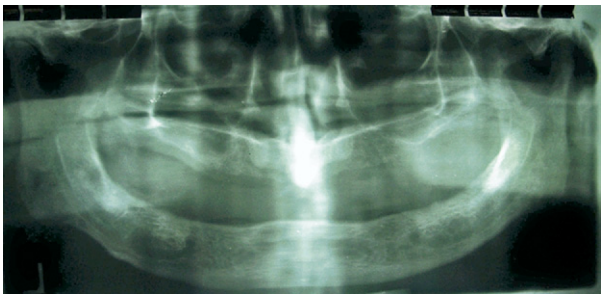

FIGURE 5. Postoperative panoramic radiograph showing complete removal of sialolith after transoral sialolithotomy.

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described as having a "hen's egg" size.[7] The weight of giant sialoliths can vary from very light specimens of approximately 12 g to heavy specimens of approximately 93 g.[7,9,11] To our knowledge, the sialolith presented in this study is perhaps the largest ever reported to date, as compared with published data (Table 1).

The ability of a calculus to grow and become a giant sialolith depends mainly on the reaction of the affected duct. If the duct adjacent to the sialolith is able to dilate, allowing nearly normal secretion of saliva around the stone, it might be asymptomatic for a long period and eventually a giant calculus will be created.[12] A sialo-oral fistula develops most likely when bacteria set up an acute exacerbation in the stagnating and retained saliva located behind the stone. The inflammatory debris obstructs the residual narrowed duct lumen, further exacerbating the inflammation. The resulting inflammatory process around a large stone may lead to tissue breakdown and spontaneous stone extrusion with intraoral fistula formation.

In contrast to the small-sized calculi, 20% to 30% of which are radiolucent, giant sialoliths are mostly radiopaque and are easily depicted on panoramic radiographs, probably because their lithogenesis is long enough for calcification to be completed.[2]

Calcification, however, can also be visualized very early by use of a computed tomography scan, which is sensitive even to stones that are radiolucent on standard radiographs.[13] Although the standard occlusal radiograph is the most reliable method of viewing the submandibular sialolith, the region visualized is limited posteriorly to the second molar, making it unsuitable for giant sialoliths, which occur frequently in the posterior portions of Wharton's duct. The posterior fourth of the duct, which includes the comma area to the hilum and body of the gland, can be visualized only by placing the x-ray cone posterior to the gland and directing it in an upward, anterior, and slightly medial direction.[14] In our report the standard occlusal view did not show any sialolith because the stone was located in the comma area of Wharton's duct; therefore a panoramic radiograph was taken to show the sialolith of this remarkable size.

The treatment objective for giant sialoliths, as for the standard-sized stones, is restoration of normal salivary secretion. The giant sialolith should be removed in a minimally invasive manner, via a transoral sialolithotomy, to avoid the morbidity associated with sialadenectomy.[2] Whenever the stone can be palpated intraorally, it is best to remove it through an intraoral approach.[1] The cardinal rule when performing stone removal from Wharton's duct is to first isolate the duct and then provide a longitudinal incision into the duct over the stone to retrieve it.[14] By direct cut down of the stone, the initial incision is taken directly to the depth of the stone without primary isolation of the duct. Direct cut down is not advised because of the risk of ductal stenosis, except when the sialoliths are at the orifice of the duct or when there is a large stone in the submandibular gland pushing the gland upward and anteriorly.[14] More posterior stones, 1 to 2 cm from the punctum, can be removed by cutting directly into the stone in the longitudinal axis of the duct while carefully protecting the lingual nerve.

Table 1. GIANT SIALOLITHS MEASURING MORE THAN 55 mm OR WEIGHING MORE THAN 20 g REPORTED IN LITERATURE

Study	Gender	Age (yr)	Gland	Location	Size (mm)	Weight (g)
Mustard,[8] 1945	M	42	SBM	Duct	56	NR
Cavina and Santoli,[9] 1965	M	59	SBM	Duct	70	18
Cavina and Santoli,[9] 1965	M	53	SBM	Duct and parenchyma	60	33
Raskin et al,[10] 1975	M	52	SBM	Duct	55	9.5
Tinsley,[11] 1989	M	48	SBM	Parenchyma	50	23.5
Current study	M	60	SBM	Duct	72	45.8

Abbreviations: M, male; SBM, submandibular gland; NR, not reported.

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Giant sialoliths are accompanied by long-standing salivary gland sialadenitis resulting in a grossly fibrotic and poorly functioning gland. However, after elimination of the obstruction, the apparent resiliency of the submandibular gland results in no adverse symptoms. Submandibular gland removal is indicated only when there is a stone of substantial mass within the gland itself that is not surgically accessible intraorally and when there are small stones present in the vertical portion of Wharton's duct from the comma area to the hilum.[15]

Giant sialoliths of a remarkable size pose a diagnostic and therapeutic challenge for the clinician. The choice of surgical approach to access the sialolith and the consideration for preserving the submandibular gland require careful evaluation when dealing with giant sialoliths. Newer treatment modalities such as extracorporeal short-wave lithotripsy and sialoendoscopy are effective alternatives to conventional surgical excision for smaller sialoliths. However, for giant sialoliths, transoral sialolithotomy with sialodochoplasty or sialadenectomy remains the mainstay of management.

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