

## SIALADENITIS OF LEFT SUBMANDIBULAR SALIVARY GLAND ASSOCIATED WITH GIANT SIALOLITH - A CASE REPORT

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### Abstract

Salivary gland diseases are relatively common. The most frequent non-neoplastic salivary disorder is obstructive sialadenitis. Sialoliths are the main cause of obstructive submandibular sialadenitis. Sialoliths commonly measure between 5 and 10 mm in size and stones over 35 mm are rare. We are reporting about the clinical and radiographic features of giant sialolith with obstructive sialadenitis.

### Keywords:

*Sialolith, salivary gland, sialadenitis.*

### Introduction

Sialolithiasis accounts for more than 50% of the salivary gland diseases.<sup>1</sup> They are stones within the major and minor salivary glands or in the ducts of these glands.<sup>2</sup> Males are affected more than females.<sup>1</sup> Sialoliths commonly measure between 5 and 10 mm in size, and all stones over 10 mm can be reported as sialoliths of unusual size.<sup>1,3</sup> 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 parenchyma of the gland, and therefore infection, pain and swelling of the gland.<sup>4</sup> Giant sialoliths measuring more than 35 mm are rare.<sup>1</sup> This report presents the clinical and radiographic features of giant sialolith accompanied by long-standing salivary gland sialadenitis.

### Case report

A 55-Year-old male presented with one month history of discomfort and swelling in the left submandibular region. His symptoms were exacerbated with the food intake. Patient reported of having 2 year history of episodic swelling in the same area. Patient medical history revealed that patient was suffering from multiple systemic diseases. On external examination, diffuse swelling over the left submandibular region was observed (figure 1). The overlying skin was normal and area was tender on palpation. The left submandibular gland was tender on bimanual palpation. The oral mucosa was normal in texture, with mild erythema in orifice of left whartin's duct. Pus discharge from the orifice of left Wharton's duct was observed on performing milking maneuver. Mandibular occlusal radiograph did not reveal any pathology. A panoramic radiograph showed a single radio-opaque mass measuring around 4 centimeters in size with respect to left lower border of the mandible. The anterior part of the radiopaque mass had a concentric arrangement due to the presence of a radiolucent circular area, where as the posterior part was uniformly radiopaque. The borders of the radiopaque mass was well defined, overlapped superiorly by lower border of the mandible. A circular radiopaque area extending from the image of the hyoid bone to the angle of mandible was also observed probably due to inflammatory fluid accumulation in the submandibular gland region (figure2). Based on history, clinical and radiological examination diagnosis of sialadenitis of left submandibular salivary gland with sialolithiasis was made. Medications were given to reduce pain and inflammation. Patient was referred

for surgical removal of sialolith. Due to the multiple systemic disorders of the patient, he was referred to higher specialty centre for surgical treatment.

## Discussion

Sialolithiasis is defined as the formation of calcific concretions within the parenchyma or the ductal system of a major or minor salivary gland.<sup>5</sup> Previous reports on salivary calculi indicate that 92 percent occur in the mandibular gland, 6 percent in the parotid gland, and 2 percent in both the sublingual and minor salivary glands.<sup>5</sup> In our case also sialolith was associated with submandibular gland. The submandibular glandular system is the most susceptible for salivary calculi because of the anatomic arrangement of the principal duct and the physiochemical characteristics of its secretion.<sup>6</sup> Sialoliths are the main cause of obstructive submandibular sialadenitis as in our case. They vary in size, shape, and quality.<sup>6</sup> We noticed a single stone mass measuring around 4 cms in size with respect to left submandibular gland.

A pubmed literature search was carried out regarding reported cases of giant sialolith from the year 2000 to 2013. All the reported cases were evaluated for the number, age of the patient at the time of reporting, sex predilection, site, size of the sialolith, any associated special features and the mode of management (table 1).<sup>7-13</sup> Out of 11 cases reported 10 patients were males.<sup>7-13</sup> In present report also male patient was affected. The age range varied from 33 years to 65 years.<sup>7-13</sup> All the cases were involving submandibular gland. The size of the reported cases were 22mm to 65mm.

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. Submandibular stones are composed of 82% inorganic and 18% organic material, whereas parotid stones are composed of 49% inorganic and 51% organic material.<sup>1</sup>

Symptoms of salivary secretion blockage vary, depending on the size and location of the stone. The most common and diagnostic sign of obstruction is increase in swelling of the gland at mealtime. As the gland produces saliva and as the saliva attempts to exit through the duct, the stone effectively acts as a valve to create back pressure within the gland. The patient then experiences a feeling of fullness and pain. Usually with the cessation of the salivary stimulation, the pain and sensation of fullness tend to decrease. Continued obstruction of the duct may lead to chronic inflammation and acute infection.<sup>5</sup> In the presenting case continued obstruction lead to sialadenitis. Inflammatory swelling occurs when organisms from mouth ascend the duct and reach saliva stagnating in the dilated ducts proximal to the calculus. Inflammation due to infection will cause edema in the duct, and the duct will close around the calculus, resulting in obstructive symptoms. Antibiotics can reduce the symptoms temporarily. When the acute stage subsides, the chronic inflammatory process may continue for a long period and cause parenchymal tissue damage. The chronic process may eventually lead to fibrosis and atrophy of the gland. Acute exacerbation of the chronic long standing process may occur.<sup>5</sup>

The differential diagnosis of masses of lymph nodes or the submandibular salivary gland origin can be considered for such swelling in the submandibular region. A careful bimanual, intraoral, and extraoral palpation is the first step in diagnosing and distinguishing between the masses of the submandibular gland and the nonsubmandibular gland origin.

If the mass is an enlarged lymph node all possible sites in the mouth and face should be carefully examined to detect a source of infection or a primary tumor. A submandibular abscess may form by the direct extension of a preexisting infection or abscess in the mouth. The history and physical findings of an abscess are diagnostic.<sup>14</sup>

Masses within the submandibular gland can be submandibular tumors or Submandibular gland sialadenitis. Submandibular gland sialadenitis commonly occurs. When the duct is obstructed, eating leads to pain and swelling in the gland because the secretions accumulate behind the obstruction. The pain and swelling tend to subside between meals. Similar features were noted in our case.

Radiographs are a practical and simple way of investigating the ductal system. The traditional diagnostic methods include plain radiographs (occlusal film), sialography, ultrasound, and scintigraphy.<sup>6</sup> In our report the standard occlusal view did not show any sialolith because the stone was located in the posterior region; therefore a panoramic radiograph was taken to show the sialolith of this remarkable size. Radiopacity is not observed in 40% of parotid and 20% of submandibular stones; therefore sialography or other imaging techniques may be required to locate them.<sup>1</sup> Sialo- CT and magnetic resonance sialography are more recently introduced diagnostic tools.<sup>15</sup> Magnetic resonance sialography provides two-dimensional or three-dimensional images of the salivary gland

without contrast medium and too much exposure to radiation. Sialoendoscopy is relatively new technique which allows us to explore the ductal system completely. <sup>6</sup>

Sialolith can be distinguished from other soft tissue calcifications because they usually are associated with pain and swelling of the involved salivary gland. Other calcification in this region such as lymph node calcifications do not show features of pain and swelling associated with meals and generally they are asymptomatic. The position and appearance of the calcified mass also helps to differentiate sialolith from tonsilloliths and phleboliths. <sup>16</sup>

The conventional treatment depends on the site of the stone, and can be either by intraoral or external approach. The treatment of choice is surgical excision. Newer treatment modalities such as extracorporeal short-wave lithotripsy and sialoendoscopy are effective alternatives to conventional surgical excision for smaller sialoliths. <sup>17</sup> However, for giant sialoliths, transoral sialolithotomy with sialodochoplasty or sialadenectomy remains the mainstay of management. <sup>1</sup>

In the presenting report clinical and radiographic features of the giant sialolith is highlighted. Early diagnosis and treatment of sialolith is of utmost importance both to reduce patient discomfort and prevent further complications such as chronic sialadenitis.

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**Figures and legends**



*Figure 1 : Patient photograph showing swelling in the left submandibular area*



*Figure 2 : Panoramic radiograph showing sialolith in the left lower border of mandible*

**Table 1: Reported cases of giant sialoliths in the past decade**

SL. No	Author /year	Number of cases/ Age/Sex	Site	Size	Special features	Management
1.	Takeda Y et al 2003 <sup>(7)</sup>	1- 33 Years /Female	Submandibular gland		sialolith with bone formation	
2	Graziani F et al 2006 <sup>(8)</sup>	1- 61 Years /Male	Submandibular duct - right	22mm		Removal by transorally under local anaesthesia
3	Boffano P et al 2006 <sup>(9)</sup>	1 – 48 Years /Male	Submandibular duct- right	22mm		Removal of the giant sialolith and sialodochoplasty were performed via an intraoral approach
4	Rai M, Burman R 2009 <sup>(1)</sup>	60 Years /Male	left submandibular region	65 mm		Sialolithotomy with sialodochoplasty was performed under LA via an intraoral approach
5	Alkurta MT 2009 <sup>(2)</sup>	2-45years/male and 65years/male	1-right submandibular gland. 2-right Wharton's duct	1-28mm 2-31mm		Excised Surgically
6	Shetty BN and Sharma P. 2010 <sup>(10)</sup>	1-50 years /male	Submandibular left	27mm	Protruding calculus from the floor of the mouth.	The calculus was removed by gentle extraction
7	El Gehani R et al 2010 <sup>(11)</sup>	2 cases	Submandibular	1-35mm		Transoral removal of the stone was performed under local anaesthesia
8	Iqbal A 2012 <sup>(12)</sup>	1- 55years / Male	left submandibular duct sialolith	35 mm		Removed the sialolith surgically under local anaesthesia
9	Rauso R 2012 <sup>(13)</sup>	1-56years/Male	left submandibular	56 mm		Removal of the stone and the left submandibular gland was performed via an extraoral incision