

Case Report

Giant Submandibular gland stone largest in India: Management and review of literature: A rare case report

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Abstract:

Sialoliths are calcereous concretions seen in the salivary gland parenchyma or its duct, accounting more than 50% of salivary gland diseases. Giant sialoliths measuring 35 mm are rare, with only 19 cases published in literature. Ninety-five percent of the giant sialoliths reported were in the submandibular gland, all occurring in male patients. Preoperative imaging help in defining the size, location of lesion and helps in planning approach. We are reporting a rare case of giant submandibular gland sialolith in a 52 year male patient measuring 40 mm (probably largest in India), which was removed through transcervical approach under general anesthesia.

KEYWORDS: Sialoliths, calcereous concretions , salivary gland, Giant, largest

INTRODUCTION

Sialoliths are calcereous concretions seen in the salivary gland parenchyma or its duct, accounting more than 50% of salivary gland diseases. Incidence of sialolithiasis is about 12 in 1000 persons in the adult population every year, thus making it one of the most common cause of acute and chronic infections of submandibular gland with male predominance.[1] Nearly 80% of reported cases of sialolithiasis are from submandibular gland or its duct, 6% in the parotid gland, and 2% in the sublingual gland or minor salivary glands.[2] Clinically, they are round to ovoid, rough or smooth, and of a yellowish color. They develop because of the deposition of mineral salts around a nidus of bacteria, desquamated cells, or mucus. Sialoliths are composed of organic and inorganic substances, mainly calcium phosphate, and smaller amounts of carbonates in the form of hydroxyapatite. Interestingly composition of submandibular gland stones is mainly inorganic(82%) compared to that of parotid gland stone(inorganic 51%. Organic 49%).[1] Most of Sialolith measure between 5–10 mm in size, and stones over 10 mm are unusual. Giant sialoliths measuring 35 mm are rare, with only 19 cases published in literature [Table 1].[3-18] Ninety-five percent of the giant sialoliths reported were in the submandibular gland, all occurring in male patients. We are reporting a case of a giant sialolith measuring approximately 40mm(probably largest in india), in an middle age male patient along with a review of literature.

Case report:

A 52-year-old male patient reported to the Department of Otolaryngology in a tertiary care center with a chief complaint of swelling on the right side of the floor of the mouth for more than 20 year duration. His main complaints were recurrent attacks of pain and swelling in right submandibular region, on and off pus discharge from mouth. Physical examination revealed a diffuse swelling in the right submandibular region with no other significant findings. Intraoral examination revealed a large, firm to hard, non-tender swelling on right floor of the mouth. The mucosa over the swelling was normal.

On the basis of history and clinical examination, a provisional diagnosis of sialolithiasis of the right submandibular gland was considered with a differential diagnosis of chronic submandibular sialadenitis, foreign body and lymph node calcification. CECT oral cavity with neck revealed a well-defined, large radiopaque mass medial to the right body of the mandible along the course of Wharton's duct [Figures 1]. CECT also revealed extension of stone in parapharyngeal region.[Figure 2]. A diagnosis of right submandibular duct sialolith was confirmed. Patient was prescribed oral antibiotics along with anti-inflammatory drugs and was posted for transcervical excision. Intraoperatively duct was incised to remove the stone from submandibular duct. [Figure 3]. Sialolith measured approximately 40 mm and was of yellow-to-white in color [Figure 4].

Figure 1:

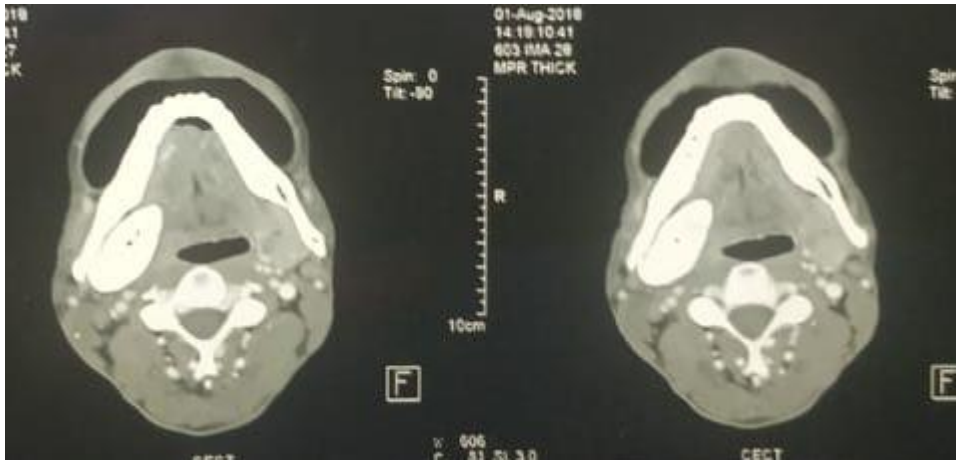


Figure 1: CECT oral cavity with neck: showing 20*40*26 mm hyperdense lesion in right submandibular duct

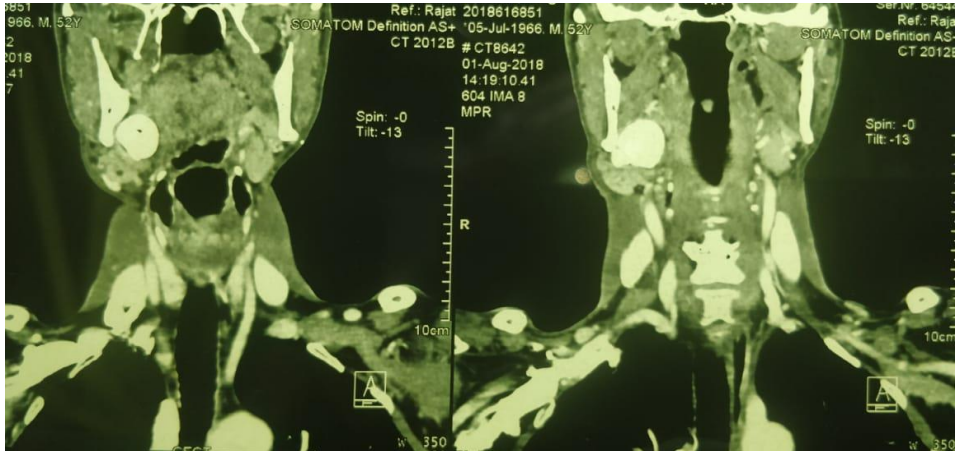


Figure 2: parapharyngeal extension of stone

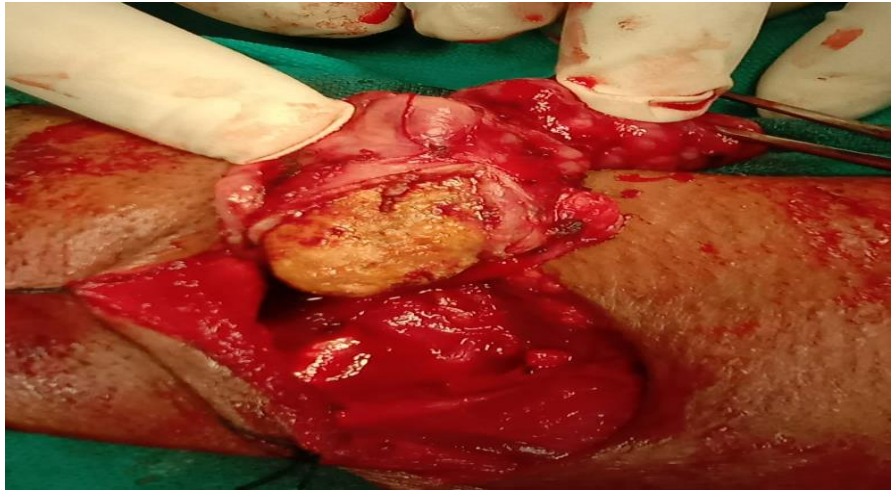


Figure 3: intraoperative image of stone after incising submandibular duct

Figure 4: postoperative image showing submandibular stone measuring around 20*40*26 mm



Discussion:

Sialoliths commonly involve major salivary glands. Frequency around 1.2% in the adult population, with male predominance.[1,14,16]. Mainly reported in the submandibular salivary gland with few incidences in the parotid and sublingual glands. Submandibular salivary gland is commonly affected because of the longer and wider duct, salivary flow against gravity, alkaline pH and higher content of mucin proteins, calcium, and phosphate.[17] Sialolith formation occur in two phases, namely, central core and periphery phase. Central core phase is mainly by the precipitation of salts, which are bound by certain organic substances such as various carbohydrates and amino acids. The second layered periphery phase consists of layered deposition of organic and inorganic material. Parotid stones are formed around a nidus of inflammatory cells or a foreign body whereas submandibular stones are thought to form around a nidus of mucous.[18] Peak incidence occurs between the ages of 20 and 60 years, and is uncommon in children.[14,19]. Giant sialoliths are a rare entity with sizes varying from 3.5 to 7 cm, mostly in male patients [Table 1]. Bhoji TV et al gave a detailed study of various giant sialolith reported till date in literature.[23]. Large calculi are also known by their weights.[20,21] Weight of the giant sialoliths was not communicated in all these previously reported cases, however, according to the reported data, they can be very light specimens (4.2 g) or can attain a heavy weight (33 g), as can be seen in our case with a reported weight of 18 g. Several factors contribute to the development of sialolith in the submandibular gland. The saliva from the submandibular gland has a high content of mucin and flows uphill in a wider and longer duct as compared to that in the parotid gland. Stenson's duct (parotid gland) is narrow and the serous saliva from the parotid gland flows downhill. In addition, the saliva secreted by the submandibular gland is alkaline and has a high content of calcium and phosphate, which promotes stone formation.[1,16,18,20].

Table 1:

Study	gender	age	gland	location	size(mm)	weight(gm)
<i>Meyers, 1942[2]</i>	Male	50	SM	Duct	50	NR
<i>Mustard, 1945[3]</i>	Male	42	SM	Duct	56	NR
<i>Allen, 1956[4]</i>	Male	49	SM	Duct	35	NR
<i>Cavina and Santoli, 1965[5]</i>	Male	59	SM	Duct	70	18
<i>Cavina and Santoli, 1965[5]</i>	Male	53	SM	Both	60	33
<i>Hoggins, 1968[6]</i>	Male	52	SM	Paren	50	NR
<i>Rust and Messerly, 1969[7]</i>	Male	66	PAR	Duct	51	NR
<i>Rust and Messerly, 1969[7]</i>	Male	58	NR	Paren	35	NR
<i>Raksin et al., 1975[8]</i>	Male	52	SM	Duct	55	9.5

<i>Isacson and Nils -Erik, 1982[9]</i>	Male	48	SM	Duct	48	NR
<i>Tinsley, 1989[10]</i>	Male	48	SM	Paren	50	23.5
<i>Hubar et al., 1990[11]</i>	Male	65	SM	Duct	52	17.5
<i>Akin and Esmer, 1991[12]</i>	Male	45	SM	Paren	45	NR
<i>Paul and Chauhan, 1995[13]</i>	Male	45	SM	Duct	45	4.2
<i>Bodner, 2002[14]</i>	Male	50	SM	Duct	50	NR
<i>Montes et al., 2007[16]</i>	Male	34	SM	Duct	36	12
<i>Manjunath Rai[17]</i>	Male	60	SM	Duct	72	45.8
<i>Babu, 2011[18]</i>	Male	50	SM	Duct	62	40
<i>Bhovi et al 2017[23]</i>	Female	60	SM	Duct	35	13
<i>This case</i>	Male	52	SM	Duct	40	18

Table 1: various studies showing giant sialolith with there distribution
SM-submandibular gland, PAR- parotid gland,Paren- parenchyma, NR- not reported

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. 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. Inflammatory process around a large stone may lead to tissue breakdown and spontaneous stone extrusion with intraoral fistula formation. Imaging studies are very useful for diagnosing sialoliths. To visualize the radiopaque stones in the submandibular duct, the best view is a standard mandibular occlusal radiograph. Radiopacity is not a consistent feature in most of the sub mandibular stones, and hence, sialography, ultrasound, computed tomography, and magnetic resonance sialography may be required for locating the stone.[15,21] Different treatment options may be selected according to the size and location of the sialolith. Small stones can be milked out through the ductal orifices using bimanual palpation. If a stone is large, piezoelectrical extracorporeal shock wave lithotripsy or surgical removal of the stone or gland may be required.[22].

Conclusion:

Sialoliths can obstruct the secretory duct, resulting in a chronic retrograde infection because of a decrease in salivary flow. Giant sialolith poses both diagnostic and therapeutic challenges for the clinician. Preoperative imaging help in defining the size,location of lesion and helps in planning approach. We are reporting a rare case of giant submandibular gland sialolith in a male patient(probably largest in india), which was removed through transcervical approach under general anesthesia.

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