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Giant Parotid Sialolithiasis: Report of a case with review of literature

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Introduction:

Sialoliths are calcified structures that develop within the salivary ductal system [(1)]. Sialolithiasis is the most common disease of salivary glands [(2)]. Sialolithiasis accounts for 30% of salivary diseases [(1)]. It is estimated that it affects 12 in 1000 adult population. Males are twice as much affected as females [(2)], especially in cases of parotid gland lithiasis [(1)]. Sialolithiasis usually occurs between the ages of 30-60 years, though cases of teenagers have also been reported [(3)]. Children are rarely affected [(1)]. Sialolithiasis mainly occurs with reference to sub-mandibular gland (80%-90%), to a lesser degree in parotid gland (5%-20%) and the sublingual and minor salivary glands (1% to 2%).

Bilateral or multiple-gland sialolithiasis is occurring in fewer than 3% of cases [(3)]. In patients with multiple stones, calculi may be located in different positions along the salivary duct and gland. Sialolithiasis occurs equally on the right and left sides. 40% of parotid and 20% of submandibular stones are not radiopaque and sialography may be required to locate them [(4)]. The sialolith should be at-least 30% mineralized to be detected by radiographic study. Salivary calculi are usually unilateral and are not a cause of dry mouth [(4)]. Clinically they are round or ovoid, rough or smooth and of a yellowish color. They consist of mainly calcium phosphate with smaller amounts of carbonates in the form of hydroxyapatite, with smaller amounts of magnesium, potassium and ammonia. This mix is distributed evenly throughout [(5)].

Submandibular stones are 82% inorganic and 18% organic material whereas parotid stones are composed of 49% inorganic and 51% organic material [(6)].The organic material is composed of various carbohydrates and amino acids [(6)]. Bacterial elements have not been identified at the core of a sialolith [(6)]. The difference in chemical properties of the saliva secreted by both the glands explain the difference between the organic and inorganic constituents of parotid and submandibular gland.

Commonly, Sialoliths measure from 1 mm to less than 1 cm. Giant salivary gland stones (GSGS) are those stones measuring over 1.5 cm and have been rarely reported in the medical literature.[(6,7)] GSGS measuring over 3 cm are extremely rare, with only scant reported cases.[(8)]

This case presents the clinical and radiographic features of a rare giant sialolith of Parotid duct measuring 1.5 cm ×1.3 cm in size with a reflection of review of literature, demographic variables and possible aetio-pathogenesis.

Case report:

A 56 year old female reported to the department of Oral Medicine, Government Dental College and Hospital, Burdwan, West Bengal, with chief complaint of swelling and pain in the right side of the face since 5-7 days. History revealed that the onset of symptoms was 5-7 days prior to consultation in the Dental College but at the same time the pain was of recurrent in nature for the past 5-6 months. She had a dull aching pain in the area of complain, but suddenly developed agonizing pain in the right side of the upper back tooth region followed by swelling. The pain was localized, severe and continuous in nature but aggravated during meals. Swelling was of sudden onset and progressed rapidly. The past medical and dental history including personal and family history was unremarkable.

Figure 1: Showing swelling over the Parotid region

General examination revealed no obvious abnormalities. Extra-oral examination revealed facial asymmetry due to swelling on the right side of the face. The swelling on inspection was diffuse and within the anatomic location of the Parotid gland. The skin over the swelling was smooth with no secondary changes. On palpation, the swelling was warm, firm in consistently and tender. On clinical examination, the mouth opening was normal. Maxillary teeth on right side showed mild attrition and extrinsic staining with no evidence of pulp involvement of mobility in any of the teeth. An ill defined swelling was present in the right buccal mucosa localized near the anatomic orifice of right Stensen's duct with no obvious color change of the surrounding mucosa. The opening of the Stensen's duct was inflamed. Intra-oral palpation revealed swelling to be hard and tender. Bidigital palpation and extra oral digital pressure over Parotid gland revealed pus discharge from the opening of Stensen's duct. It also revealed a hard mass along the course of the duct.
Correlating the history and otolaryngologic findings, the condition was diagnosed as acute suppurative Parotitis due to Sialolithiasis of the right Parotid duct with differential diagnosis of Phlebolithiasis due to Sub-masseteric hematoma.

Figure 2: Intra- oral examination showing a large firm non-tender nodular swelling over the orifice of Stensen's duct

PA view of the face with puffed check revealed an oblong radio-opaque mass overlapping the shadow of the right ramus of the mandible on the right side. This confirmed the provisional diagnosis of Sialolith in the right Parotid duct. Findings of blood and serum biochemistry were within normal limits.

Surgical removal of the Sialolith was done in the Department of Oral and Maxillofacial Surgery under local anesthesia. Conventional surgical procedure was slightly modified by placing the incision behind the papilla to prevent posterior slipping of the stone as because the stone was present close to papilla. After the stone was accessed it was delivered using an ocular probe and the mucosa was sutured with horizontal mattress suture. The Sialolith was 1.5 cm in its maximum length and 1.3 cm in diameter in its maximum width.

Post operative 2 months follow up showed that the patient was asymptomatic with satisfactory glandular function and normal undisturbed clear salivary flow.

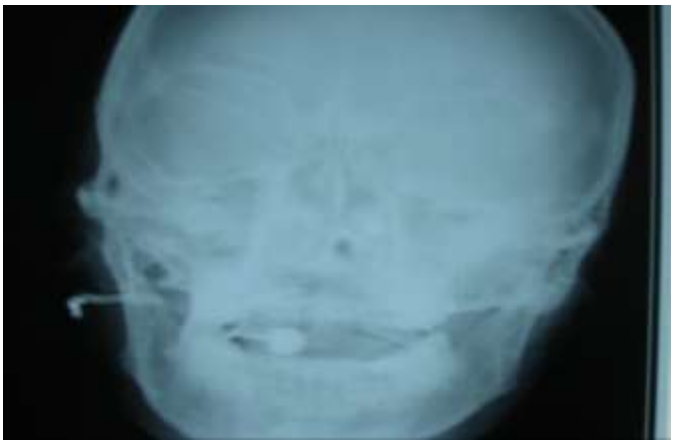

Figure 3: PA View of the skull.

Figure 4: PA View of the skull with higher contrast.

Figure 5: Lateral Cephalogram View.

Figure 6: The Sialolih shown to scale after surgical removal.

Discussion:

Although large Sialoliths have been reported in the body of salivary glands, [5,6,7,8] they have been rarely been reported in the salivary ducts.[9,10,11,12] Messerly removed a 51 mm long calculus that occupied the entire length of Stenson's duct in a 66-year-old man.[12] Brusati and Fiamminghi removed a sialolith from the left submandibular duct of a 55- year-old man measuring 27x 31 mm.[12] More recently Leung *et al*. removed a sialolith 14x9 mm from the right submandibular duct.[13] The sialolith removed in our case was comparable to these.

A review of the literature by Ledesma-Montes et. al found only 16 reported cases of stones having a size or 3.5 cm or greater.[8] Commonly, Sialoliths measure from 5 to 10 mm in size and stones over 10 mm can be reported as Sialoliths of unusual size. They rarely measure more than 15 mm. Giant Sialoliths measuring more than 35 mm are rare, with only around 17 cases published in the literature (Table 1).[(17)]

Table 1: Giant Sialoliths (Larger than 35 mm) reported in literature

Sl.No.	Study	Gender	Age (years)	Gland	Location	Size (mm)	Weight (Gram)
1.	Meyers, 1942	Male	50	SM	Duct	50	NR
2.	Mustard, 1945	Male	42	SM	Duct	56	NR
3.	Allen, 1956	Male	49	SM	Duct	35	NR
4.	Cavina and Santoli, 1965	Male	59	SM	Duct	70	18
5.	Cavina and Santoli, 1965	Male	53	SM	Both	60	33
6.	Hoggins, 1968	Male	52	SM	Paren	50	NR
7.	Rust and Messerly, 1969	Male	66	SM	Duct	51	NR
8.	Rust and Messerly, 1969	Male	58	PAR	Paren	35	NR
9.	Raskin et.al, 1975	Male	52	NR	Duct	55	9.5
10.	Isacsson and Nils-Erik, 1982	Male	48	SM	Duct	36	NR
11.	Tinsley, 1989	Male	48	SM	Paren	50	23.5
12.	Hubar et.al, 1990	Male	65	SM	Duct	52	17.5
13.	Akin and Esner, 1991	Male	45	SM	Paren	42	NR
14.	Paul and Chauhan, 1995	Male	45	SM	Duct	45	42
15.	Bodner, 2002	Male	50	SM	Duct	50	NR
16.	CL Montes et.al, 2007	Male	34	SM	Duct	36	12
17.	Manjunath Rai, 2009	Male	60	SM	Duct	72	45.8
18.	Present case	Female	56	PAR	Duct	15	22

Abbreviations: PAR- Parotid Gland, SM- Sub-mandibular Gland, Paren- Parenchymal, NR- Not reported.

(A) Demographic variables:

Giant Sialoliths are rare findings in clinical oral pathology with sizes ranging from 35 to 72 mm and all of them occurring in male patients (Table 1) [17]. The present case unusually is a female. All the patients were older than 42 years except the case presented by Ledesma Montes C et al [17] where it was 34 years. In present case patient is 56 years old. Sialolithiasis in children is considered rare. [1]

(B) **Etiology and Pathogenesis**: [18.19, 20]

The exact etiology and pathogenesis that probably leads to the genesis of salivary calculi has not yet been unearthed, but proposed multi-factorial concept of varied investigators for sialolith development can be discussed:

1. Calcium salt deposition due to stagnation of calcium rich saliva around an initial organic nidus consisting of altered salivary mucins, bacteria and desquamated epithelial cells.
2. Change of mucoid content of saliva due to intermittent stasis produces a gel like salivary mass over which deposition of inorganic and organic substances occurs leading to sialolith genesis
3. Associated causes of salivary calculi formation may be reduced salivary flow due to stagnation, dehydration, change in salivary Ph, oro-pharyngeal sepsis, introduction of foreign bodies, impaired crystalloid solubility and physical trauma to salivary duct
4. Traditional theories suggest that the formation occurs in two phases: a central core and a layered periphery. The central core is formed by the precipitation of salts, which are bound by certain organic substances. The second phase consists of the layered deposition of organic and non organic material.
5. Unknown metabolic phenomenon can increase the saliva bicarbonate content, which alters calcium phosphate solubility and leads to precipitation of calcium and phosphate ions.
6. A retrograde theory for sialolithiasis has also been proposed.
7. Aliments, substances or bacteria within the oral cavity might migrate into the salivary ducts and become the nidus for further calcification.
8. A case in which a stone formation around a vegetal nidus was histologically proven has been reported
9. Salivary stagnation, increased alkalinity of saliva, infection or inflammation of the salivary duct or gland, and physical trauma to salivary duct or gland may predispose to calculus formation.
10. Submandibular sialolithiasis is more common as its saliva is [7-14] (i) more alkaline, (ii) has an increased concentration of calcium and phosphate, and (iii) has a higher mucous content than saliva of the parotid and sublingual glands. (iv) in addition, the submandibular duct is longer than Stensen's duct, wider in diameter, narrow orifice and the gland has an antigravity flow, all of which encourage stagnation of saliva. (v) Marchal et al further suggested that sphincter like mechanism in first 3cm of the Wharton's duct may be responsible for retrograde migration of oral materials
11. Stone formation is not associated with systemic abnormalities of calcium metabolism. [3] Electrolytes and parathyroid hormone studies in patients with sialolithiasis have not shown abnormalities.[23] Gout is the only systemic illness known to predispose to salivary stone formation,[23] although in gout the stones are made predominantly of uric acid
12. The proposed association between hard water areas and salivary calculi has been shown to be incorrect as proved by Sherman and McGurk recently.[23] The lack of association holds equally for both sexes.[23] One study has suggested a link between sialolithiasis and nephro-lithiasis, reporting an association in up to 10% of patients.[3,5]

(C) Diagnosis:

The diagnosis of salivary lithiasis is based on a detailed patient's case history supplemented by a thorough clinical evaluation and roengenographic findings.

Patient of a salivary calculi usually complain of pain and swelling of the involved salivary gland which gets aggravated during meals and with salivary stimulants. The pain is mainly due to obstruction of salivary flow as noted in the present case. The pain usually disappears during the daytimes and is of acute lacerating in

nature on onset at meals. [19,20] Since the glands are encapsulated, space for expansion is hardly present which causes pain due to salivary pooling within the obstructed duct. [5]

Calculi may cause stasis of saliva, leading to bacterial ascent into the parenchyma of the gland resulting in Sialadenitis with or without associated systemic infections. Pus may be seen draining out from the gland on palpation of the gland. [16, 17, 18] Some Sialoliths may be asymptomatic. [7, 20] Long-term obstruction, in the absence of infection can lead to atrophy of the gland with resultant lack of secretory function and ultimately fibrosis. Sinus, fistula or even ulceration of the involved gland is seen in chronic cases. [21, 22]

Bimanual palpation of the floor of the mouth, in a posterior to anterior direction, may reveal a palpable stone in majority of the cases of submandibular gland calculi. [20] For parotid gland stones, careful intraoral palpation around Stenson's duct orifice may reveal a stone. [20] Deeper parotid stones are often difficult to palpate or not palpable. Minor salivary gland involvement usually in the buccal mucosa or upper lip, forms a tumor like firm nodular mass. [5, 20]

Giant salivary gland stones (GSGS) are those stones measuring over 1.5 cm and have been rarely reported in the medical literature.[25] GSGS measuring over 3 cm are extremely rare, with only scant reported cases with varied weight. 94.44% of giant Sialoliths were located in submandibular gland. [19] The sialolith presented in our case is 1.5cm in size with 22 grams in weight.

A wide range of radiographic techniques are known to exist for correlating the clinical diagnosis with roengenographic diagnosis of salivary calculi. [19,20] These diagnostic methods for detecting obstructions in the salivary ductal system include routine (occlusal and panoramic) radiographs, sialography, ultrasound, magnetic resonance sialography and computed tomography . [24, 25]

Sialoliths are well visualized on panoramic and periapical radiographs but can be obscured with super imposition over the roots of premolars and molars teeth and hence, better visualized by an occlusal radiograph without overlap from other anatomy. [19, 20] Given the postero-inferior position of the gland, a mandibular lateral oblique radiograph may be useful for visualization. [19, 20]

Sialography is thus useful in patients showing signs of Sialadenitis related to radiolucent stones or deep submandibular/parotid stones. Sialography is, however, contraindicated in acute infection or in significant patient contrast allergy. [19,20] Additionally, Iodine of the contrast medium may irritate, potentially rupture an already inflamed ductal system and force bacteria into the gland through ductal system resulting in worsening the situation. [19, 20]

Computed tomography is a non-invasive technique useful in situations of multiple calculi and in such anatomic location not easily examined intra-orally, but scans do not provide the exact anatomic location of the sialolith within the duct system. Non- contrast helical CT scans with multi-planar reconstruction has become the gold standard to detect salivary gland calculi. [19, 20]

Scintigraphy may be a complimentary exploratory technique where sialography is not indicated. Of late, sialoendoscopy is the latest optical technology for in-depth visualization of intra ductal anatomy for location of salivary calculi. [19, 20]

(D) Treatment:

Management of cases of salivary calculi is primarily dependant on the exact anatomic location of the stone. [19, 20, 25]

Patients diagnosed with small sized sialolithiasis may benefit from a trial of conservative management.[19,20,25] The patient must be well hydrated and the clinician must apply moist warm heat and gland massage, while sialogogues are used to promote saliva production and flush the stone out of the duct. With gland swelling and sialolithiasis, infection should be assumed and a penicillinase resistant anti - staphylococcal antibiotic prescribed. Most stones will respond to such a regimen, combined with simple sialolithotomy when required. [19, 20, 25]

Submandibular calculi in the distal third of the duct 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 including lingual nerve paresthesia or anesthesia and ranula of the floor of the mouth. [19, 20] If the stone is sufficiently forward, it can be manipulated and delivered through the duct orifice. This can be done with the aid of lacrimal probes and dilators to open the duct. [19, 20] Once open, the stone can be identified, milked forward, grasped and removed. The gland is then milked to remove any other debris in the

more posterior portion of the duct.[19,20] Excision of the submandibular gland by an external approach carries a 0 to 8% risk of permanent or temporary marginal mandibular nerve palsy. [14] The likely reason for this is that the nerve is stretched or cut as it courses next to the superior portion of the gland. [19, 20]

Occasionally, the duct may need opening to retrieve the stone. This involves a trans-oral approach making an incision directly onto the stone. In this way more posterior stones, 1–2 cm from the punctum, can be removed by cutting directly onto the stone in the longitudinal axis of the duct. [19, 20] Care is taken as the lingual nerve lies deep, but in close association with the submandibular duct posteriorly. Subsequently, the stone can be grasped and removed. No closure is done leaving the duct open for drainage. [21, 22]

If the gland has been damaged by recurrent infection and fibrosis, or calculi have formed within the gland, it may require removal. [21, 22]

Parotid stone management is more problematic as only a small segment of Stenson's duct is approachable through an intraoral incision. [20] In addition, surgical opening Stenson's duct can be complicated by subsequent pos surgical stenosis of the duct whereas this is rare in the submandibular gland. [20] As a result, parotidectomy has become mainstay of surgical management for the majority of intra-glandular stones. This is especially mandatory for patients who do not respond to conservative therapy and suffer from recurrent pain and swelling. [20, 21]

Alternative methods of treatment have emerged such as the use of extracorporeal shock wave lithotripsy (ESWL) and more recently the use of endoscopic intra-corporeal 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). [20] Both extra and intra-corporeal lithotripsy is gaining increasing importance in the treatment of salivary stone disease. [20]

In extracorporeal piezoelectric lithotripsy, the average size of fragments produced is about 0.7 mm. [20] Duct diameters are greater than 0.7 mm in general except for at the ostium. Therefore, fragments produced by ESWL would not be prohibited by duct diameters. Findings have also suggested that best results in salivary stone lithotripsy are achieved when the maximum size of stone fragments does not exceed 1.2 mm. [20]

Extracorporeal salivary lithotripsy provides another therapeutic option that carries fewer risks than surgical removal of the affected gland, such as the risks of a general anesthetic, facial nerve damage, surgical scar, Frey's syndrome, and causes little discomfort to the patient whilst preserving the gland.[20]

A retrospective study of patients treated endoscopic ally from 1994 to 1999 showed a success rate of 83% with no severe complications. [20] Endoscopy is a minimally invasive technique for removal of calculi from salivary glands as well as an excellent diagnostic procedure,[20] as miniaturized endoscopes conforming to the physiological widths of the ducts are used to directly view and then deliver shock waves to the stones.[20]

Though sialoendoscopy was initially used for diagnosis of salivary calculi, is now used as an interventional technique in obstructive gland diseases. [21]

Conclusion:

Parotid salivary gland calculi are one of the rare forms of salivary gland obstructive disorder. The epidemiological reviews along with clinical features, etiological causes, diagnosis and possible treatment modalities are discussed and presented in the present case scenario in a bird's eye view perspective so as to enable the clinicians a better approach towards the disorder.

Conflict of interest:

The authors declare no conflict of interests among themselves.

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