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UNUSAUAL PRESENTATION OF SUBMANDIBULAR DUCT AND GLAND CALCUI: CASE REPORT

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Abstract

Sialolithiasis is one of the most common pathologies of the submandibular gland; sialoliths account for about 80 percent of all salivary duct calculi.The majority of sialolith occurs in the submandibular gland or its duct and is common cause of acute and chronic infection. Salivary stones larger than 15 mm are classified as giant sialoliths. They are uncommon in the practice of otolaryngology, and their management has always been a therapeutic challenge. This report presents the two unusual and rare cases of large sialolith of the submandibular duct as well as gland measuring 70x11mm and 54x25mm respectively.

Introduction

Salivary gland / duct stones or sialoliths are calcifications that accumulate within the salivary gland parenchyma and associated ductal systems. They develop from a mineralization nucleus of debris including bacterial colonies, shed ductal epithelial cells and cell remnants, mucus plugs and foreign bodies[1].Eighty to 90 per cent of sialoliths develop in the submandibular gland system, and 10–20 per cent in the parotid gland. Only 1 per cent of calcifications occur in the sublingual gland which may be due to a dominant mucoid secretion and very short ductal tree[2]. Most patients present with a single stone but multiple stones occur in 32 per cent of cases in the parotid gland and 22 per cent in the submandibular gland. Bilateral stones occur in around 2.2 per cent of cases. Sialoliths are typically more common in middle-aged males but some studies suggest a male to female ratio of 1:1 and with ages ranging from 12 to 93 years[4]. The most frequent clinical presentation is swelling and pain in the area of the affected gland with a prodromal awareness varying from less than six months to 30 years. Sialoliths can often be detected on palpation, especially when they are located above the mylohyoid muscle or in the buccal mucosa and lip[2- 4]. Sialoliths in the submandibular gland duct are usually diagnosed after longer asymptomatic periods than those in the parotid gland duct[2-4].

Case report:

Case 1:

A 26-year-old male presented at the department of ENT, Rural Medical College, Loni with complaints of 1) severe pain and swelling in the right lower submandibular region for a duration of 7 days. There were episodes of pain in the same region for last 2 years but of a moderate intensity 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. 2) Firm mass in the anterior part of the right side of the floor of the mouth.

On neck examination, the patient showed diffuse swelling over the left submandibular region measuring 7× 6× 5cm, with normal overlying skin. 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 hard, inflammation, induration, swelling of the right Wharton's duct (fig. 1). The left submandibular gland was tender on bimanual palpation.

Radiologically patient was evaluated which includes, lower occlusal radiograph and X- ray neck AP and Lateral view which showed the mass to be radiopaque and extending back beyond the lower right first permanent molar (fig. 2&3). A diagnosis of right submandibular duct calculus was made and sialolithotomy was planned under local anaesthesia, after giving local anaesthesia upward and medial pressure was applied to the submandibular gland, and an incision was placed directly over the sialolith to expose it, multiple stone were removed measuring to be 70mm long when kept together to greatest length (fig. 4). The larger portion of the sialolith which was of 20mm, delivered out first with the sinus forceps then thorough exploration and continuous massaging of the submandibular gland with upward and medial pressure was applied to mobilize the distal portions of the stone with the sinus forceps. Wharton's duct stoma kept open to facilitate drainage of left fragments of stone. Postoperatively patient was relieved of pain and swelling regressed. The patient was reviewed one weeks post operatively to check salivary function of the gland. On review the right submandibular gland was palpable but clear saliva could be expressed from the Wharton's duct stoma on massage.

Case 2:

A 44-year-old man presented at the department of ENT, Rural Medical College, Loni with the compliant of swelling, in his left submandibular region that had been present for 6 months. On examination it reveals that swelling was hard, non-tender, local temperature not raised and bimanually palpable. Neck radiograms (fig. 5). and ultrasonography revealed a sialolith of 54 mm in length and 25 mm in diameter at its widest portion. Blood pressure and pulse rate were within normal limits. Chest radiograms, electrocardiography, total blood count, urine sediment, liver and kidney function test were also normal. Under general anaesthesia, a surgical resection of the left submandibular gland was performed (fig. 6&7). Post-operatory course was good and the patient was discharged after two days. No injury to lingual or hypoglossal nerve occurred.

Pathology

Microscopic evaluation of the gland revealed a chronic sialadenitis with infiltration of lymphocytes in the stroma and destruction of the acini and of the main duct[5].

Discussion

Although large sialoliths have been reported in the body of salivary glands, they have been rarely been reported in the salivary ducts. Messerly removed a 51 mm long calculus that occupied the entire length of Stenson's duct in 66-year-old man. Brusati and Fiamminghi removed a sialolith from the left submandibular duct of a 55- year-old man measuring 27x31 mm. More recently Leung *et al.* removed a sialolith 14x9 mm from the right submandibular duct[10]. The sialolith removed in our first and second case were far bigger measuring 70x11mm and 54x25mm.

Aetiology

The exact aetiology and pathogenesis of salivary calculi is largely unknown. Genesis of calculi lies in the 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. For stone formation it is likely that intermittent stasis produces a change in the mucoid element of saliva, which forms a gel[9]. This gel produces the framework for deposition of salts and organic substances creating a stone. 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. Submandibular stones are thought to form around a nidus of mucous, whereas parotid stones are thought to form most often around a nidus of inflammatory cells or a foreign body[7,9]. Another theory has proposed that an unknown metabolic phenomenon can increase the saliva bicarbonate content, which alters calcium phosphate solubility and leads to precipitation of calcium and phosphate ions. A retrograde theory for sialolithiasis has also been proposed. Aliments, substances or bacteria within the oral cavity might migrate into the salivary ducts and become the nidus for further calcification. A case in which a stone formation around a vegetal nidus was histologically proven has been reported. 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 Submandibular sialolithiasis is more common as its saliva is

(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. In addition, the submandibular duct is longer and the gland has an antigravity flow[9]. Stone formation is not associated with systemic abnormalities of calcium metabolism. Electrolytes and parathyroid hormone studies in patients with sialolithiasis have not shown abnormalities. Gout is the only systemic illness known to predispose to salivary stone formation, although in gout the stones are made predominantly of uric acid. The proposed association between hard water areas and salivary calculi has been shown to be incorrect. The lack of association holds equally for both sexes. One study has suggested a link between sialolithiasis and nephrolithiasis, reporting an association in up to 10% of patients. Sialolithiasis typically causes pain and swelling of the involved salivary gland by obstructing the food related surge of salivary secretion[10,11]. 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. Some may be asymptomatic until the stone passes forward and can be palpated in the duct or seen at the duct orifice. It may be possible that obstruction caused by large calculi is sometimes asymptomatic as obstruction is not complete and some saliva manages to seep through or around the calculus. Long term obstruction in the absence of infection can lead to atrophy of the gland with resultant lack of secretory function and ultimately fibrosis[12].

Diagnosis

Careful history and examination are important in the diagnosis of sialolithiasis. Pain and swelling of the concerned gland at mealtimes and in response to other salivary stimuli are especially important. Complete obstruction causes constant pain and swelling, pus may be seen draining from the duct and signs of systemic infection may be present[6].

Bimanual palpation of the floor of the mouth, in a posterior to anterior direction, reveals a palpable stone in a large number of cases of submandibular calculi formation. Bimanual palpation of the gland itself can be useful, as a uniformly firm and hard gland suggests a hypo-functional or nonfunctional gland. For parotid stones, careful intraoral palpation around Stenson's duct orifice may reveal a stone[6].Deeper parotid stones are often not palpable. When minor salivary glands are involved they are usually in the buccal mucosa or upper lip, forming a firm nodule that may mimic tumour. Imaging studies are very useful for diagnosing sialolithiasis. Occlusal radiographs are useful in showing radiopaque stones. It is very uncommon for patients to

have a combination of radiopaque and radiolucent stones; 40% of parotid stones may be radiolucent[9]. 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.

Treatment

Patients presenting with sialolithiasis may benefit from a trial of conservative management, especially if the stone is small.6The 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[9]. 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. Almost half of the submandibular calculi lie in the distal third of the duct and 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. If the stone is sufficiently forward it can be milked and manipulated through the duct orifice[9]. This can be done with the aid of lacrimal probes and dilators to open the duct. 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. The duct may need opening to retrieve the stone[9]. 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 directly onto the stone 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. Subsequently, the stone can be grasped and removed. No closure is done leaving the duct open for drainage[9]. If the gland has been damaged by recurrent infection and fibrosis, or calculi have formed within the gland, it may require removal. Parotid stone management is more problematic as only a small segment of Stenson's duct is approachable through an intraoral incision. In addition, opening Stenson's duct can be complicated by subsequent stenosis of the duct whereas this is rare in the submandibular gland. As a result, parotidectomy is the mainstay of surgical management for the majority of intraglandular stones[9]. This is reserved for patients whose symptoms do not respond to conservative therapy and suffer from recurrent pain and swelling. 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). Both extra and intracorporeal lithotripsy are gaining increasing importance in the treatment of salivary stone disease[9-11]. In extracorporeal piezoelectric lithotripsy, the average size of fragments produced is about 0.7 mm. 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[11]. 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 anaesthetic, facial nerve damage, surgical scar, Frey's syndrome, and causes little discomfort to the patient whilst preserving the gland[12].

Conclusion

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[9]. Newer treatment modalities such as

extracorporeal short-wave lithotripsy and sialoendoscopy are effective alternatives to conventional surgical excision for smaller sialoliths[9]. However, for giant sialoliths, transoral sialolithotomy with sialodochoplasty or sialadenectomy remains the mainstay of management.

References

1) Liao G, Su Y, et al. Sialoendoscopic secondary intervention after failure of open sialolithectomy. J Oral Maxillofac Surg. 2010; 68(2):313-318

2) Cawson R, Odell E. Cawson's neoplastic and non-neoplastic diseases of the salivary glands. Essentials of Oral Path and Oral Med. Philadelphia (PA): Churchill Livingstone; 1998. p. 239-240.

3) Gehani E, Krishnan B, et al. Submandibular giant sialoliths: report of two cases and review of the literature. Ear Nose Throat J. 2010;89(6):E1-4.

4) Cottrell D, Courtney M, Bhatia I, Gallagher G, Sundaarajan D**,** Intraoral Removal of a Giant Submandibular Sialolith Obstructing Wharton's Duct: A Case Report, Journal of the Massachusetts Dental Society 2011;60:14-16

5) Ellis GL, Auclair PL, Gnepp DR. Surgical pathology of the salivary glands. Philadelphia: W.B. Saunders Company, 1991.

6) Williams MF. Sialolithiasis. Otolaryngol Clin North Am 1999 ;32:819–834.

7) Huang TC, Dalton JB, Monsour FN, Savage NW, Multiple, large sialoliths of the submandibular gland duct: a case report Australian Dental Journal 2009; 54: 61–65

8) Rai M, Burman R, Giant Submandibular Sialolith of Remarkable Size in the Comma Area of Wharton's Duct: A Case Report, J Oral Maxillofac Surg 67:1329-1332, 2009

9) Siddiqui S. J. Sialolithiasis: an unusually large submandibular salivary stone, BDJ 2002;193: 89- 91

10) Akin I, Esmer N. A submandibular sialolith of unusual size: a case report. *J Otolaryngol* 1991; 20: 123-125.

11) Zenk J, Werner G, Hosemann M D, Iro H. Diameters of the main excretory ducts of the adult human submandibular and parotid gland – a histological study. *Oral Surg, Oral Med, Oral Pathol, Oral Radiol, Endod* 1998; 85: 576-580.

12) Iro H, Schneider H Th, Fodra C *et al*. Shockwave lithotripsy of salivary duct stones. *Lancet* 1992; 339:1333-1336