CASE REPORT

Australian Dental Journal 2009; 54: 61–65

doi: 10.1111/j.1834-7819.2008.01091.x

Multiple, large sialoliths of the submandibular gland duct: a case report

TC Huang,* JB Dalton,* FN Monsour,† NW Savage‡

**Dento-Maxillofacial Radiology, School of Dentistry, The University of Queensland.*
†Maxillofacial Unit, Royal Brisbane and Women's Hospital, Queensland.
‡Oral Medicine, School of Dentistry, The University of Queensland.

ABSTRACT

This paper reviews the major clinical and radiographic features of sialoliths and illustrates these with an unusual case of multiple sialoliths within the submandibular gland duct. The differential diagnosis of other calcific structures both within and outside the salivary gland that may mimic a sialolith is also presented.

Key words: Calcification, case reports, radiopaque lesion, sialolith, salivary gland.

(Accepted for publication 4 June 2008.)

INTRODUCTION

Salivary gland/duct stones or sialoliths are calcifications that accumulate within the salivary gland parenchyma and associated ductal systems.[1] 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.[2] 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.[2–4] 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.[3] Bilateral stones occur in around 2.2 per cent of cases.[3]

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.[2,3] 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.[1,3] Sialoliths can often be detected on palpation, especially when they are located above the mylohyoid muscle or in the buccal mucosa and lip.[1,5] Sialoliths in the submandibular gland duct are usually diagnosed after longer asymptomatic periods than those in the parotid gland duct.[6] This is due to greater ductal volume between the hilus and submandibular papilla and the ability to accommodate the obstruction while still allowing saliva to flow past the obstruction.[6] The severity of pain and swelling is pressure-associated and dependent upon the degree of obstruction and residual duct patency.[1] Recurrent partial obstructions are the usual clinical diagnoses and correlate with mild symptoms that self-correct within a short period following stimulation, usually meal related.[4] Complete obstruction however, presents as an emergency situation with severe symptoms including a tense swollen gland with marked sensitivity, ductal swelling and on occasion suppuration which may collect as a discrete abscess or drain from the duct orifice.[1,4] This may be accompanied by localized cellulitis, malaise and fever.[4] The signs and symptoms are listed in Table 1.

Salivary calculi larger than 1 cm are rare. A review of literature by Lustmann found that of 302 sialoliths studied, 79.8 per cent were 1 cm or less and only 7.6 per cent greater than 1.5 cm.[2] This present article reviews the significant clinical features of sialoliths and reports a case of multiple calculi in the submandibular duct. Patients will often present to the general dental practitioner for diagnosis and it is important that the matter can be dealt with competently and referral to surgical management arranged.

CASE REPORT

A 57-year-old female presented with pain and swelling focal to the left submandibular triangle with a duration

Table 1. Signs and symptoms

Swelling
Anatomical asymmetry
Size fluctuation, usually rapid onset and partial resolution over one to several hours
Residual glandular swelling
Decreased stimulated salivary flow compared to the contralateral gland
Pain (intensifies during mealtimes or when salivary flow is stimulated)
Stones commonly visible in submandibular duct
Swelling and erythema of submandibular papilla for distal stones
Suppuration (uncommon)
Localized cellulitis (uncommon)

of around three months. Intra-orally she was aware of a firm mass in the floor of the mouth. Her medical history was unremarkable and she had not had any previous similar episodes involving either site.

Clinically, there was a visible swelling of the left submandibular triangle which, on careful examination, was localized to a tense and sensitive submandibular salivary gland. The overlying skin did not show erythema or a temperature differential compared with the contralateral side and although the adjacent lymph nodes were difficult to palpate due to generalized sensitivity, none were obviously enlarged. The cervical nodes remained uninvolved. Intra-orally the left floor of the mouth was oedematous with a multi-nodular enlargement of the submandibular duct with two presumed calcifications palpable. It was not possible to examine the lingual fossa due to swelling and sensitivity. The calcifications were fixed and did not move antero-posteriorly with gentle manipulation. Stimulated flow from the submandibular gland was difficult to judge due to sensitivity but there was no discharge of either saliva or suppuration from the submandibular papilla.

The panoramic radiograph showed an elongated radiopaque structure superimposed over the roots of the 31 to 37 and with obscuration of the root anatomy (Fig 1). An occlusal radiograph revealed at least three sialoliths occupying most of the length of the left submandibular duct extending from the distal extent proximally to the molar area and lingual fossa (Fig 2). Lamination can be seen within the sialolith.

The calculi were surgically exposed and removed (Fig 3). The stones were individually dissected free due to extensive fibrosis and adhesion to the wall of the left submandibular salivary duct. Following removal of all stones, the duct was cannulated and expanded and became patent with transmission of normal secretory elements. Proximal repositioning of the duct orifice to the mid-left floor was undertaken with a circular suture back technique to prevent subsequent fibrosis and duct stricture.

Four calculi were identified at operation and removed with a combined length of 4 cm in their largest dimension (Fig 4). They were irregular to oval in shape with rough, multi-nodular and irregular surfaces. There was also evidence of fibrotic tissue covering the calculi.

Fig 1. Panoramic radiograph showing the submandibular salivary stone superimposed over the roots of the left anterior and posterior teeth.

Fig 2. Lower occlusal radiograph showing two definite submandibular gland duct stones and possibly a third.

DISCUSSION

Submandibular sialoliths measuring less than 1 cm in greatest dimension are quite common but larger sialoliths are considerably less so.[2,7,8] Their presence can cause salivary gland dysfunction and obstruction of salivary flow resulting in chronic or acute bacterial infections. Varying degrees of atrophy of the glandular

Fig 3. Removal of the distal and largest sialolith from the submandibular gland duct.

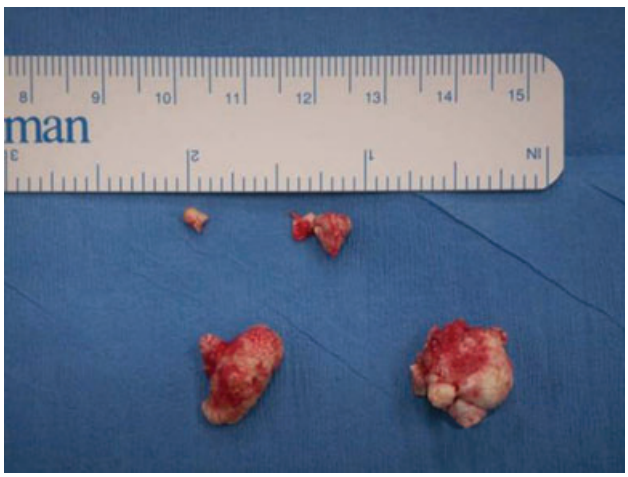

Fig 4. Four discrete calculi were removed from the duct and are shown to scale.

parenchyma with ductal ectasia/expansion and fibrosis of the interstitium may also occur. This depends upon the position of the stone within the ductal system, hilum or gland parenchyma and the duration and degree of obstruction and resultant retrograde pressure. Intraductal stones may lie free within the duct and able to move antero-posteriorly depending upon pressure build-up from saliva trapped within the proximal sector or intra-oral manipulation. Stones present for an extended period and particularly those with an irregular external surface may provoke a focal inflammatory reaction within the duct wall with resultant scarring and enhanced obstruction.

There are several factors that may contribute to the increased incidence of sialoliths in the submandibular gland. These include the more viscous mucus content of the saliva and the high concentration of calcium phosphate. This creates a more alkaline pH which not only favours the solid-liquid phase exchange of calcium phosphate species in the mouth and maintenance of the dentition but causes precipitation of the more reactive species dibasic calcium phosphate dihydrate/brushite. This process is helped by the ascending course and narrow orifice of Wharton's duct compared to the calibre of the duct itself, both of which encourage stagnation of saliva.[6,9]

Table 2. Radiographic features

Radiopaque (homogeneous or with a laminated structure)
Some may be radiolucent
Cylindric or irregularly-shaped
Anatomical position important
Imaging must include full duct length and gland
Stone orientated antero-posteriorly within duct
Fixed stones tend to be more rounded

The initial radiographic examination of sialoliths is usually undertaken with plain films (Table 2). Lustmann found that sialoliths were detected in 94.7 per cent of cases using intra-oral radiographs alone.[2] Large and well mineralized calculi are visible on plain radiographs but small or partially mineralized calculi may remain undetected.[6] Significantly, Blatt found that around 20 per cent of sialoliths remain unseen on plain film examination due to a low mineral content.[4] Sialoliths developing in the hilus of the submandibular gland tend to be oval and may grow larger before becoming symptomatic reflecting the dynamics of fluid flow around the developing stone and the ductal structure in this part of the arborization system.[5,10] Given the postero-inferior position of the gland, a mandibular oblique lateral radiograph may be useful for visualization.[10] Sialoliths located within the duct distal to the hilum tend to be elongated, again due to fluid flow characteristics and a more defined luminal architecture, and are better visualized with an occlusal radiograph displaying the floor of the mouth without overlap from other anatomy (Fig 2).[10] Sialoliths are well visualized on panoramic and periapical radiographs but can be obscured with superimposition over the roots of the premolar and molar teeth and muscle attachment ridges on the cortices of the mandible. Intraductal, large stones will usually show as an anteriorly inclined stone due to the ascending course of the duct from the flexure in the lingual fossa to the anterior floor of the mouth. Both small and sometimes large sialoliths can be asymptomatic and can appear as coincidental findings on radiographs.[11]

Other imaging techniques that may be used to diagnose sialoliths include sialography, ultrasound, computed tomography and magnetic resonance sialography. Sialography is rarely indicated and should be restricted to those cases with a suspected ductal stricture or other obstruction but without a calcification visible on routine imaging. Ultrasound will locate a sialolith but, with the subsequent requirement for

conventional imaging, it is of limited clinical usefulness and in most cases introduces an unnecessary step in the diagnostic sequence. Until proven otherwise it is prudent to consider and exclude the presence of multiple sialoliths in any patient presenting with a sialolith. These may be located in either or both the duct and the gland. Computed tomography is useful in any situation where there are multiple stones or when the stone is situated in a site not readily examined intraorally, for example, the lingual fossa and proximally including intraglandular stones and similarly for the parotid gland.

In most cases of sialolithiasis, treatment is advised either for management of symptoms or, in quiescent lesions, to prevent periductal inflammation and fibrosis and the development of an obstructive situation. When the stone is small, conservative management such as moist heat, increased intake of fluids, sialagogues and gentle massage of the gland towards the gland duct opening may be all that is required to allow spontaneous release of the stone.[1,5] A small sialolith near the orifice of the duct may also be removed following widening of the orifice with a lacrimal probe.[2] If this is not successful or a large stone is present, surgical removal is necessary. Sialoliths in the gland duct can often be removed without damage to the gland but intraglandular sialoliths generally require removal of the gland.[1,8,12] Stone removal in the posterior part of the duct or removal of the gland may lead to complications such as damage to the lingual and hypoglossal nerves or bleeding into the floor of the mouth. Haemorrhage in the floor of the mouth can lead to major complications and can even be life threatening. Therefore, postoperative observation is vital. This procedure is usually performed under general anaesthesia to better control any bleeding and to dissect and protect the lingual and hypoglossal nerves. Other treatments used successfully in the management of sialoliths include interventional sialendoscopy with wire-basket extraction for small sialoliths (< 4 mm) and fiberoptic laser lithotripsy with basket retrieval for larger sialoliths (> 4 mm).[13] In any retrieval procedure within the ductal system, care must be exercised firstly to ensure the stone does not track proximally and be lost to the extraction process or that both the duct and adjacent anatomical structures are not damaged to the extent of causing significant scarring or other anatomical deficit on healing.

It is important to distinguish sialoliths from other calcific structures outside the salivary gland and considerations in the differential diagnosis are listed in Table 3.[14,15] A mandibular torus or osteoma can occur in a similar position but both remain in a constant relationship with the mandible on films with different angulations. Their surface contour both clinically and radiographically also reflect the nature of these two lesions and are almost always diagnostic. Due to the large size of this patient's sialolith on the panoramic radiograph, the presentation does mimic an osteoma but pain is usually not a symptom.

Table 3. Differential diagnosis

Mandibular torus
Osteoma
Calcified lymph nodes
Phleboliths and other vascular calcifications
Tuberculosis of lymph nodes or of the salivary gland itself
Calcified atherosclerotic plaques in major blood vessels
Myositis ossificans
Metastasis from distinct calcifying neoplasms

Calcified lymph nodes are usually both radiopaque and radiolucent with mottled and irregular borders.[9,10] A calcified submandibular lymph node may be difficult to differentiate from a submandibular sialolith due to its position near the submandibular gland and a similar projection on a radiograph. Differential diagnosis of a calcified mass in the submandibular area would favour a sialolith due to its relative higher incidence. In this instance, an ultrasound may be of assistance in differentiating the typical echopattern of a lymph node.

Phleboliths are calcified thrombi occurring in venules, veins or vascular malformations/haemangiomas. Phleboliths can occur in the floor of the mouth and tend to have a dense ring with a radiolucent centre giving a bull's eye or target appearance on an occlusal radiograph.[9,14] A radiopacity in the floor of the mouth in the absence of sialadenitis and the presence of a clinically discernible varicosity would favour a diagnosis of phlebolith. However, there is a reported case of cavernous hemangioma with numerous phleboliths in the submandiblar gland which clinically and radiographically (on plain film and computed tomography imaging) resembled sialolithiasis.[16]

Calcification in a short section of the facial artery near the submandibular area could simulate a sialolith. If a longer area of artery is involved, the serpentine, calcified image is usually diagnostic.[17] A calcification in the common carotid usually lies next to the C3–C4 and more laterally on a panoramic view than a sialolith.[18]

A rare differential diagnosis may include myositis ossificans. Although most common in the masseter, other muscles of mastication such as the temporalis, medial pterygoid and lateral pterygoid can also be involved.[19] Myositis ossificans in the medial pterygoid can mimic a submandibular sialolith.[20] However, trismus should alert the clinician to the possibility of myositis ossificans.

CONCLUSIONS

Sialoliths are always a consideration in submandibular and facial pain particularly when related to mealtimes. Their work-up requires a careful history and the

selection of the correct imaging techniques to both confirm the clinical diagnosis and to define the precise position of the calcification. This paper has reviewed the major features of sialoliths and illustrated these with an unusual case of multiple sialoliths within the submandibular duct. At surgery an additional stone was found reinforcing the need for caution at removal. The procedural work is not discussed in detail as this is usually best undertaken by clinicians skilled in salivary gland and ductal surgery to avoid post-procedural morbidity and functional deficit.

REFERENCES

1. Ellis GL, Auclair PL, Gnepp DR. Surgical pathology of the salivary glands. Philadelphia: W.B. Saunders Company, 1991.
2. Lustmann J, Regev E, Melamed Y. Sialolithiasis: a survey on 245 patients and a review of the literature. Int J Oral Maxillofac Surg 1990;19:135–138.
3. Levy DM, William MD, ReMine H, Devine KD. Salivary gland calculi. Pain, swelling associated with eating. JAMA 1962; 181:1115–1119.
4. Blatt IM. Studies in sialolithiasis: III. Pathogenesis, diagnosis and treatment. South Med J 1964;57:723–729.
5. Williams MF. Sialolithiasis. Otolaryngol Clin North Am 1999; 32:819–834.
6. Som PM, Curtin HD. Head and neck imaging. 4th edn. St. Louis: Mosby, 2003.
7. Zakaria MAK. Giant calculi of the submandibular salivary gland. Br J Oral Surg 1981;19:230–232.
8. Siddiqui SJ. Sialolithiasis: an unusually large submandibular salivary stone. Br Dent J 2002;193:89–91.
9. Rabinov K, Weber AL. Radiology of the salivary glands. Boston: GK Hall, 1985.
10. White SC, Pharoah MJ. Oral radiology: principles and interpretation. 5th edn. St. Louis: Mosby, 2004.
11. Graziani F, Vano M, Cei S, Tartaro GP, Mario G. Unusual asymptomatic giant sialolith of the submandibular gland: a clinical report. J Craniofac Surg 2006;17:549–552.
12. Yildirim A. A case of giant sialolith of the submandibular salivary gland. Ear Nose Throat J 2004;83:360–361.
13. Marchal F, Dulguerov P. Sialolithiasis management: the state of the art. Arch Otolaryngol Head Neck Surg 2003;129: 951–956.
14. Bar T, Zagury A, London D, Shacham R, Nahlieli O. Calcifications simulating sialolithiasis of the major salivary glands. Dentomaxillofac Radiol 2007;36:59–62.
15. Mandel L. Tuberculous cervical node calcifications mimicking sialolithiasis: a case report. J Oral Maxillofac Surg 2006; 64:1439–1442.
16. McMenamin M, Quinn A, Barry H, Sleeman D, Wilson G, Toner M. Cavernous hemangioma in the submandibular gland masquerading as sialadenitis: case report. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 1997;84:146–148.
17. Suarez-Cunqueiro MM, Duker Jg, Liebehenschel N, Schön R, Schmelzeisen R. Calcification of the branches of the external carotid artery detected by panoramic radiography: a case report. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2002;94:636–640.
18. Pornprasertsuk-Damrongsri S, Thanakun S. Carotid artery calcification detected on panoramic radiographs in a group of Thai population. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2006;101:110–115.
19. Steiner M, Gould AR, Kushner GM, Lutchka B, Flint R. Myositis ossificans traumatica of the masseter muscle: review of the literature and report of two additional cases. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 1997;84:703–707.
20. Parkash H, Goyal M. Myositis ossificans of medial pterygoid muscle: a cause for temporomandibular joint ankylosis. Oral Surg Oral Med Oral Pathol 1992;73:27–28.

Address for correspondence:
Dr Neil W Savage
Oral Medicine and Pathology
School of Dentistry
The University of Queensland
200 Turbot Street
Brisbane QLD 4000
Email: n.savage@uq.edu.au