

Giant Sialolith of the Submandibular Salivary Gland

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We report the case of a 48-year-old man who had an unusually large submandibular gland sialolith (2.6 cm in greatest dimension), which led to sialadenitis and subsequent abscess formation. We describe the management of this patient and review the literature with emphasis on the various modalities available for diagnostic imaging.

Introduction

Sialolithiasis is the second most common disease of the salivary glands after mumps [1]. It is also the most common disease of submandibular glands in middle-aged adults [2]. Sialolithiasis is estimated to affect 12 in 1000 of the adult population with males being affected almost twice as much as females [3, 4]. Children are very rarely affected [5]. More than 80% of the sialoliths occur in the submandibular gland or its duct, 6% in the parotid gland and 2% in the sublingual gland or minor salivary glands [1, 6, 7]. Simultaneous lithiasis in more than one salivary gland is rare, occurring in fewer than 3% of cases. Also, 70 to 80% of cases feature solitary stones; only about 5% of patients have three or more

stones [6]. There is no left or right predominance [8]. The stones themselves are typically composed of calcium phosphate or calcium carbonate in association with other salts and organic material such as glycoproteins, desquamated cellular residue, and mucopolysaccharides. Bacterial elements have not been identified at the core of a sialolith [9]. Some factors inherent to the submandibular gland tend to favor stone formation there like longer and larger caliber duct, flow against gravity, slower flow rates and higher alkalinity along with higher mucin and calcium content of the saliva [10]. The submandibular gland hosts the largest stones with the largest reported one being 6cm in length [11]. Most submandibular stones are found in the salivary duct (75 to 85% of cases) [12]. Hilar stones tend to become very large before becoming symptomatic. Ductal stones are elongated in shape whereas hilar stones tend to be oval [13].

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Abbreviations: CT, computed tomography

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Case Report

A 48-year old man presented to the Department of Otolaryngology with a persistent draining wound over the right submandibular region. He had started experiencing symptoms like odynophagia and dysphagia two

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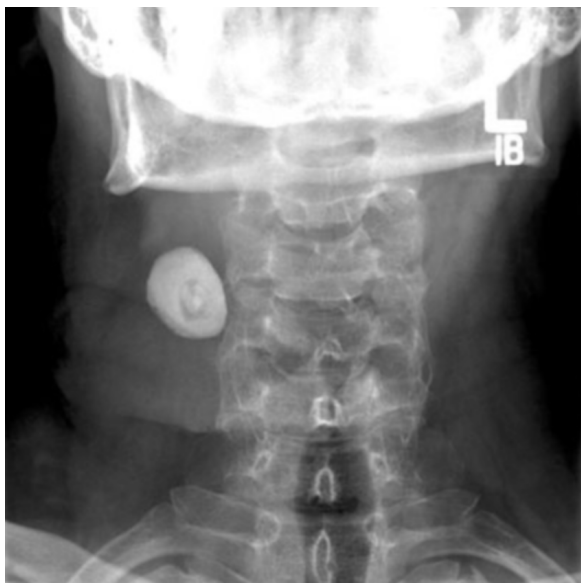


Figure 1A. Radiograph of the neck in the antero-posterior projection shows a large calcified mass in the submandibular region.



Figure 1B. The calcified mass as seen on the left lateral projection of the neck.

years ago and was diagnosed as suffering from chronic right submandibular sialolithiasis at an outside hospital. He subsequently developed an abscess over the area and underwent two incision and drainage procedures. Consequently he developed a constantly draining wound over the incision site. On clinical examination, a 2-cm erythematous incision was noted over a 2-cm indurated mass in the right submandibular region. The area was tender and purulence could be expressed by palpation. Per-oral examination revealed a 2-cm tender and firm mass in the right floor of the mouth near the submandibular gland. The ductal opening was normal.

CT of the head and neck region showed a 2.2 cm by 1.8 cm stone within the medial aspect of the right submandibular gland with enlargement of the gland itself. A 1-cm in diameter ring-enhancing hypodensity was also noted superiorly to the gland, which most likely represented a small abscess.

The gland and the calculus were excised via an incision in the skin crease 2 cm below the lower border of the mandible and directly over the palpable submandibular gland. The skin was ellipsed around the area of the draining sinus and granulation tissue. The gland with

the calculus was dissected free and the wound was closed in layers, with insertion of a vacuum drain. Pathology revealed a submandibular gland, which measured 5.5 cm in its largest dimension with the stone measuring 2.6 x 2.1 x 1.5 cm. The calculus was round, tan in color and had a granular surface. The calculus was surrounded by white fibrous tissue. Adjacent to the calculus, there was unremarkable lobulated yellow salivary gland tissue.

Discussion

Sialolithiasis of the submandibular gland can be completely asymptomatic [9]. Common symptoms vary from a painless swelling, moderate discomfort to severe pain with large glandular swelling accompanied by trismus and usually associated with eating [13]. Sialoliths are commonly 1-10 mm in size, but giant sialoliths (greater than 3.5 cm) have been reported occasionally [14]. Sialo-cutaneous fistula (as seen in this case) is also a possibility with megaliths because of the obstruction of the duct resulting in stasis, infection and subsequent rupture through the skin [11]. Long-term obstruction in

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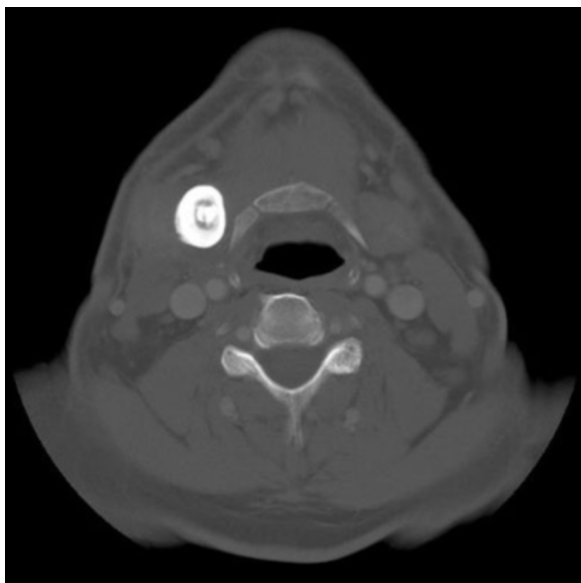


Figure 2A. CT scan of the neck with contrast demonstrating a large sialolith of the submandibular gland on the right side. Both soft tissue and bone windows are on display here. The stone lies on the medial aspect of the gland and is seen to compress the normal glandular tissue.

the absence of infection can lead to atrophy of the gland with resultant lack of secretory function and ultimately fibrosis [15]. Large sialoliths like the one seen in this case are relatively rare. The largest reported sialolith was 6-cm in length and had a dry weight of 50 grams [11]. Submandibular gland calculi have been reported to be radiopaque in 80 to 94.7% of cases and can be seen on plain films [7, 16, 17].

CT and ultrasound can demonstrate sialoliths with high accuracy and can correctly localize them anatomically. US is less accurate than CT in distinguishing multiple clusters of stones from single large stones [18, 19]. Generally, CT in this setting is best performed without administration of contrast material, since small opacified blood vessels may simulate small sialoliths. However, if an abscess or an inflammatory process is suspected, contrast may be administered after identifying the stone on unenhanced scans [18]. T2-weighted MR imaging with thin sections can be used noninvasively to evaluate the ductal architecture of the salivary glands and to visualize

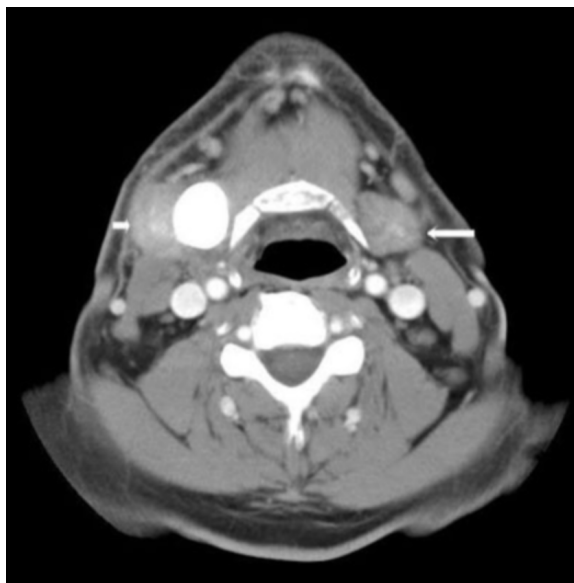


Figure 2B. Soft Tissue Window The short arrow shows the compressed submandibular gland adjacent to the sialolith and the long arrow shows the normal gland for comparison.

the effect the sialolith has on the ductal system [20-22]. MR is mostly not needed but sometimes it may be used to see strictures of the salivary duct, when another concurrent process is suspected and in complicated cases [23]. Sialography is contraindicated in the acute setting of sialadenitis because of the possibility of exacerbating the symptoms associated with the infection. Also the act of instrumentation may lead to post traumatic edema and stricture formation. The need to perform sialography should be restricted to a very few number of cases when clinical assessment, serology (especially for the autoimmune causes), conventional radiography (the stones may be radiolucent), and/or CT cannot facilitate diagnosis of the cause of the chronic sialadenitis. MR sialography can replace conventional sialography [23].

If the stone is small, conservative management may be attempted with local heat, massage and sialogogues [9]. Infection should be treated with antibiotics and these cases should be combined with simple sialolithotomy when required [13, 24]. If the stone lies in the distal one-third of the duct, a simple surgical release can be performed by an incision in the floor of the mouth.

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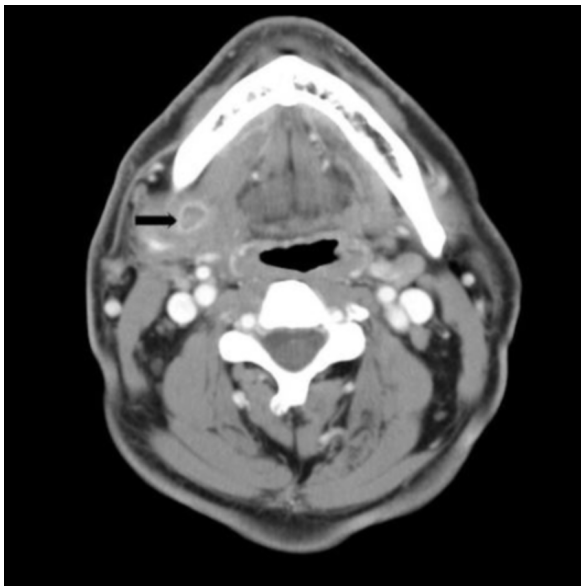


Figure 3. CT scan of the neck with contrast showing a ring enhancing lesion on the right side in the area of the submandibular gland compatible with a diagnoses of abscess. This abscess lies just superiorly to the sialolith of previous figures (black arrow).

Incising the duct transorally can retrieve more posterior stones. Care is taken as the lingual nerve lies deep to the submandibular duct posteriorly and the two structures are closely associated. If the gland has been damaged by recurrent infection and fibrosis, or calculi have formed within the gland, it may require removal. Excision of the submandibular gland carries a risk of permanent or temporary marginal mandibular nerve palsy [25]. In the case described in this report, no permanent or temporary nerve palsy occurred. Alternative methods of treatment have emerged such as the use of extracorporeal shock wave lithotripsy and more recently the use of endoscopic intracorporeal shockwave lithotripsy [26].

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