

Self-Extrusion of Unusual Size Submandibular Sialolith: A Case Report

Olağandışı Boyutlu Submandibular Sialolitini Kendi Kendine Ekstrüzyonu: Bir Olgu Sunumu

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ABSTRACT

Sialolithiasis is most commonly affects submandibular gland and duct due to its saliva content properties and anatomical location. The common size of sialolith is between 5mm and 10mm and it is called unusual in size when bigger than 10mm. The exact aetiology of its formation is still unknown. Typical presenting symptom is long history of fluctuating submandibular swelling which is associated with meal. Diagnosis is usually based on the history, clinical examination and supplemented by radiographic finding. Treatment is depending on size, location, and the number of stone. We present a case of unusual size of submandibular stone, 15mm x 5mm which is self-extruded by conservative management.

Keywords: Sialolithiasis, sialolith, submandibular gland

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ÖZET

Sialolithiasis, tükürük içeriği özellikleri ve anatomik konumu nedeniyle en sık submandibular bez ve kanalı etkiler. Sialolitini yaygın boyutu 5 mm ile 10 mm arasındadır ve 10 mm'den büyük olduğunda olağandışı olarak adlandırılır. Oluşumunun kesin etiyolojisi hala bilinmemektedir. Tipik başvuru semptomu, yemekle ilişkili uzun süre dalgalı submandibular şişlik öyküsüdür. Tanı genellikle öyküye, klinik muayeneye dayanır ve radyografik bulgularla desteklenir. Tedavi taşın büyüklüğüne, konumuna ve sayısına bağlıdır. Biz konservatif tedavi ile kendiliğinden ekstrüde edilen 15mm x 5mm boyutlarında olağandışı bir submandibular taş vakası sunuyoruz.

Anahtar Sözcükler: Sialolithiasis, sialolith, submandibular bez

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INTRODUCTION

Sialolithiasis present in 1.2% of the general population in a post-mortem study and it is the most common disorder of major salivary glands (1). Submandibular gland and duct are the most common affected sites followed by parotid, sublingual, and minor salivary glands (2-4). This is because the saliva from submandibular gland is more alkaline, has higher concentration of calcium and phosphate and contains more mucous content compare to other salivary glands. Furthermore, this saliva is drain against gravity and submandibular duct is comparatively longer than other major salivary glands. In majority of cases, sialolith are form within Wharton's duct with proximal part more frequently affected. The sialolith is commonly unilateral and single in number. The common sizes of sialoliths are between 5mm and 10mm. Sialoliths of unusual size are referring to all stones with size more than 10mm (5). It is called giant sialolith when the size larger than 15mm (6). Largest reported Wharton's duct stone was 83mm x 12mm (7). It occurs predominately at the middle age with no gender predilection in more recent study (2). The exact aetiology of sialolith formation is still unknown. It is believed to start with formation of nidus consisting of salivary mucin, bacteria, or desquamated epithelial cells which later become deposited by mineral salts (5). Risk factors like dehydration, oral or dental infection or trauma to salivary duct or gland may predispose to calculus formation. Patients usually presented with history of chronic submandibular swelling and pain during food intake. The diagnosis is based on the patient's history and clinical examination and supplemented by radiographic findings. Examination finding is depending on the site of calculus formation. Sometime gritty sensation can be felt on bimanual palpation at the floor of mouth along the course of Wharton's duct especially if stone situated at the distal duct. Most of submandibular calculi are radiopaque and can be detected by conventional dental radiograph especially occlusal and panoramic view (7). Treatment is depending on size, location, and the number of stone (8).

CASE REPORT

A 17-year-old man presented with acute left submandibular swelling for 3 days duration. The swelling was increasing in size and associated with pain especially during meal. He could not tolerate any solid foods due to the severe pain. He had multiple episodes of similar symptoms for the past 3 years, however the symptoms were minimal and resolved by gentle massage. He also had fever for the past 2 days, however denied any obstructive symptoms like shortness of breath or dysphagia. There was no history of foreign body ingestion. Upon examination, he looked dehydrated and presence of left submandibular swelling measuring 2.0 cm x 2.0 cm, which was tender and firm on palpation (Figure 1). Bimanual palpation was positive, thus confirmed submandibular gland in origin. Oral cavity examination revealed floor of mouth was oedematous and presence of pus mixed with blood discharge from the left Wharton's duct opening upon pressing on the left submandibular swelling (Figure 2). There was no gritty sensation felt at the floor of mouth and dental hygiene was good. Other ear, nose and throat examinations including oropharynx and flexible nasopharyngolaryngoscopy revealed no medialisation or other significant findings. Lateral neck x-ray was done by emergency department team to rule out retropharyngeal abscess, before the case was referred to otorhinolaryngology, head, and neck surgery team. There was no widening of prevertebral soft tissue to suggest retropharyngeal abscess, however, a radiopaque calculus seen at the left submandibular region, anterolateral to anterior border of hyoid bone (Figure 3). Patient was referred to dental clinic for occlusal view x-ray to confirm the present of sialolith but unfortunately x-ray machine was non-functioning at that time. Subsequently, the patient was admitted to ward with impression of left submandibular gland abscess secondary to left submandibular sialolithiasis. He was started on intravenous antibiotics, namely amoxicillin clavulanate and metronidazole. Analgesia and intravenous drip were given for pain control and rehydration respectively. He was advised for frequent massage over the swelling site to drain pus through the Wharton's duct. On day two of admission, patient reported sialolith was self-extruded through the duct after massage over left submandibular region. The stone measured 15mm x 5mm (Figure 4). He was pain free after that with gradually resolving left submandibular swelling. He was discharged home on day five of admission and sialography was done as outpatient showed no residual sialolith.



Figure 1. 2.0 x 2.0 cm swelling at the left submandibular region with no overlying skin colour changes.

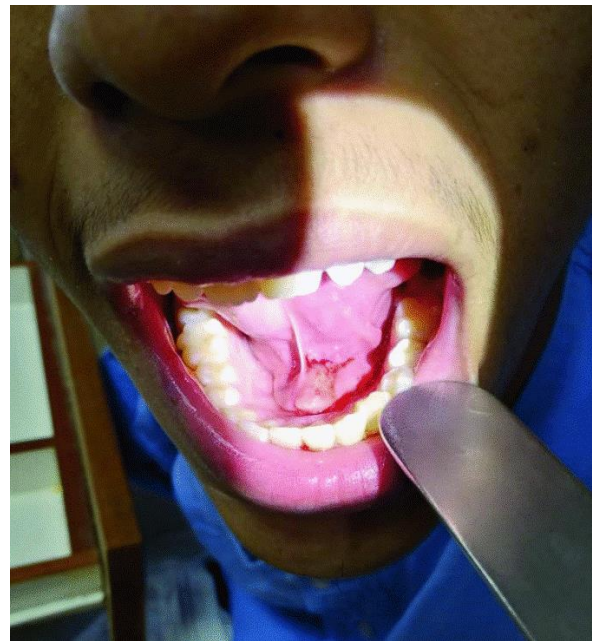


Figure 2. Oral cavity examination shows oedematous floor of mouth and presence of pus mixed with blood discharge from the left Wharton's duct opening.

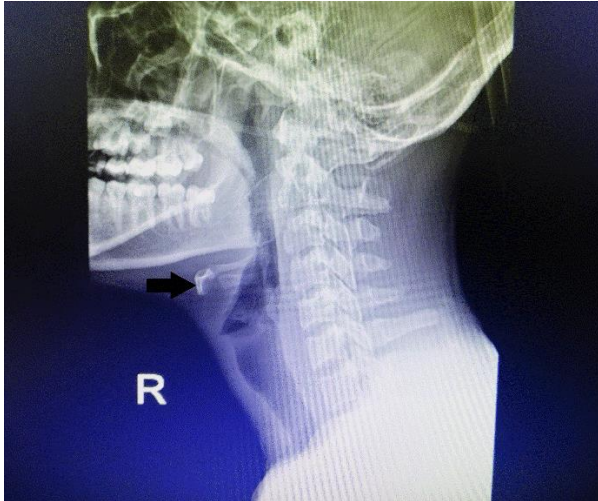


Figure 3. Lateral neck soft tissue X-ray shows radiopaque calculi at the left submandibular region, anterolateral to anterior border of hyoid bone (arrow).



Figure 4. Self-extruded of submandibular sialolith from the left Wharton's duct, measuring 15mm x 5mm.

DISCUSSION

Patient with submandibular sialolithiasis usually has long history of fluctuating submandibular swelling and pain which typically associated with meal. The present case also presented with these symptoms; however, patient did not seek any medical attention initially as his symptoms were minimal and the pain was bearable. As time passed, his symptoms became worsening, that could be explained by gradual increasing size of sialolith and finally obstruct the Wharton's duct. Presence of oedematous floor of mouth and pus discharge from the Wharton's duct orifice were other points that support the diagnosis.

Important differential diagnosis that needs to consider in this case is Ludwig's angina because the condition can progress very rapidly and lead to upper airway obstruction. Gout is the only systemic disorder known to predispose to sialolith (4). There was no known risk factor identified in the present case. Radiology investigation is important tool to support the diagnosis. Majority of submandibular sialolith are radiopaque and can be detected on a radiograph. Intraoral radiograph or occlusal view was found to be more diagnostic than standard extraoral radiograph (8). This is due to the stone usually masked by the bony structures and teeth in extraoral radiograph especially when the stone is small. In the present case, due to its location, size and radio-opacity, the stone can be visualised even on lateral neck x-ray. Sialography and ultrasound are more useful imaging tools in cases of radiolucent stone. Other advantages of sialography include able to detect salivary duct stricture or stenosis, presence of accessory salivary duct or gland and visualise small sialolith. However, this method is contraindicated in acute infection and contrast-allergy patient. Ultrasound is a non-invasive procedure with high sensitivity and specificity to detect submandibular sialolith. Sialography was preferred in present case to assess any residual stone because most of the time ultrasound cannot detect small calculi less than 2mm (8). Sialendoscopy is another helpful diagnostic procedure when there is no stone visualized on imaging, but patients having persistent symptoms suggestive of sialolithiasis. Furthermore, small stone can be removed using stone retrieval basket during the procedure (9). Newer techniques like computerized tomography (CT) scan and magnetic resonance imaging (MRI) are helpful when other techniques provide ambiguous results. CT scan can detect any associate abscess or ranula and able to differentiate between calculi and vascular structures. MRI is more useful in identification of smaller stones and to differentiate acute from chronic obstruction. Imperatively, the size, site and number of stone are determinants for the treatment modalities. Small stone usually treated conservatively with administration of sialogogues and regular gland massage. Antibiotic is warranted if patient presents with signs of infection and hydration should be maintained by intravenous drip in poor oral intake and dehydrated patient. Sialendoscopy is indicated for small stone with diameter of 4mm to 5mm (10). For stone size 4mm to 8mm in diameter, it is generally need combination of sialendoscopy and extra-corporeal lithotripsy. Failure of the mentioned procedures denote for open surgery like transoral, external, or combination of the approaches. For the present case, the stone was self-extruded after regular massage on the submandibular gland, even the size was relatively big (15mm x 5mm). Theoretically, unusual and giant sizes submandibular sialolith are very difficult to pass through the Wharton's duct because the mean diameter of the duct is between 0.5 mm and 1.5 mm with the narrowest diameter at the ostium (11).

CONCLUSION

Thorough history taking is important in diagnosing submandibular sialolithiasis because most of the patients usually presented with typical symptoms. They can present with infection or abscess secondary to retrograde infection as the result of salivary flow impairment by the presence of the stone. Appropriate imaging techniques are required to confirm the diagnosis as well as define the location, size, and number of the stone. These informations are very crucial in determining the types of management.

Conflict of interest

No conflict of interest was declared by the authors.

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