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AN UNUSUAL CASE OF SELF EXTRUSION OF A GIANT SUBMANDIBULAR SIALOLITH

Otolaryngology

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ABSTRACT

Sialolithiasis is the commonest benign disease of salivary glands.It usually occurs in the duct,leading to obstruction of salivary outflow and its associated consequences/symptoms.Of the salivary glands,the submandibular gland is the most frequently affected.Sialoliths usually measure around 5mm in maximum diameter.Calculi measuring greater than 15mm are giant sialoliths and have been rarely reported in literature.Diagnosis of calculi is based on history,examination and radiographic investigations.Management,which aims at the elimination of the calculus and restoration of normal salivary flow,may be either medical or surgical,depending on the site and size of the sialolith,and the condition of the gland.Self expulsion of a sialolith is an unusual occurrence.We report a rare case of a giant Wharton's duct sialolith with spontaneous extrusion into the oral cavity.

KEYWORDS

Salivary calculi;Wharton's duct;Giant sialolith

INTRODUCTION

Sialolithiasis is the most common disease of salivary glands.[1]Sialoliths can occur at any age,but appear more frequently in patients in the third to sixth decades of life.[2]Estimate show that it affects 1.2% of the adult population.[1]Males are affected twice as much as females.[3]More than 80% occur in the submandibular gland or its duct,probably due to its secretion being more viscous with a higher mineral content,and a longer duct.4-10% have been reported in the parotid gland and 1-7% in the sublingual gland or minor salivary glands.[4]

0.45% of the population is affected by symptoms caused by salivary calculi,though the estimated frequency including asymptomatic form may exceed 1% in the adult population.[5]Sialolithiasis is clinically characterized by local pain and swelling,reduced salivary flow,restricted mouth opening and purulent discharge.Swelling is usually correlated to food intake,when salivary secretion is enhanced.[6]In many cases,the diagnosis is easy due to obvious clinical features,but for treatment,imaging studies are always necessary.[7]Radiographs presenting a large portion of the maxillofacial region(panoramic radiography,computed tomography)can detect salivary calculi with high accuracy.[5]

Most sialoliths are usually of 5mm in maximum diameter and all stones over 10mm should be reported as a sialolith of unusual size.[7]Giant stones(>15mm) are rare and approximately every tenth or twelfth of stones belong to this category.[8]Large submandibular calculi measuring several centimetres,whether symptomatic or silent,have to be removed promptly for the prevention of possible complications that may occur in the future.Perforation through the floor of the mouth by a submandibular sialolith is rare.[9]We therefore report a case of a giant Wharton's duct sialolith with spontaneous extrusion into the oral cavity.

CASE REPORT

A 36 year old male patient presented to the Department of Otorhinolaryngology and Head and Neck Surgery with the complaint of swelling in the right half of the floor of the mouth since childhood.It had increased in severity for the past 10 days and was associated with pain in the right submandibular region,which was aggravated on food intake.There was no history of any neck swelling.The patient was a non smoker and had no other medical or surgical history.Examination revealed a 2x2cm tender,hard swelling in the right half of the floor of the mouth.The overlying mucosa was erythematous,with a 0.5x0.5cm region of ulceration.The right submandibular gland was enlarged and tender on bidigital palpation.

Ultrasonography of the neck detected right submandibular sialadenitis with dilatation of intraglandular ducts.The patient was started on antibiotic and analgesic.Computerised tomogram(CT) of the neck was performed after 3 days which revealed a single radio-opaque density measuring 16x14x11mm in the terminal right Wharton's duct,suggestive of a calculus and confirming the clinical diagnosis of sialolithiasis (Fig.1).Surgical intervention was planned.However,the following day,during a meal the calculus self extruded (Fig.2) and his symptoms resolved thereafter.He was treated with sialogogues and the site of extrusion was allowed to heal spontaneously.On follow up,there were no complications or evidence of recurrence.

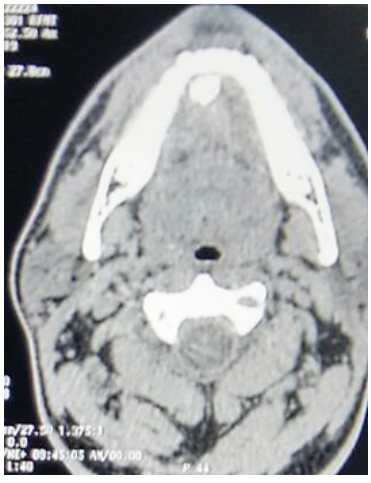

Fig 1-CT showing giant distal right Wharton's duct calculus

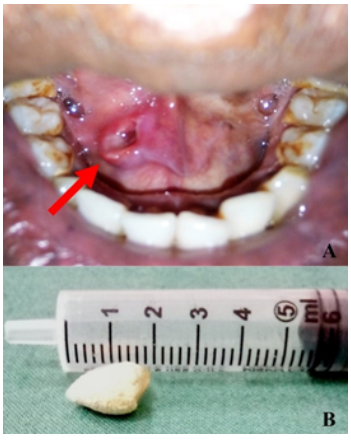

Fig 2A-Floor of mouth following self extrusion of right Wharton's duct calculus; B-Extruded giant calculus

DISCUSSION

Sialolithiasis is considered to be the most common salivary gland disorder.Submandibular gland has the highest predilection for sialolithiasis,with an 80% occurrence rate,followed by 19% in the parotid and 1% in the sublingual glands.[7]The size of sialoliths is usually less than 10mm in diameter.Salivary calculi larger than 15mm(considering the largest diameter) are classified as giant sialoliths and most of them are located in the submandibular gland or in its duct,as seen in our case.[5]About 80-90% of submandibular calculi are located in Wharton's duct i. e.10 % in the parenchyma,57% in the hilum and 34% in the distal duct.[7]In our case,a giant sialolith was identified in the distal duct.

Submandibular sialolithiasis occurs as a consequence of a hampered flow due to inflammatory stenosis of Wharton's duct.Moreover,there are some anatomical factors associated with formation of sialoliths in the submandibular gland,such as:Wharton's duct is the longest among the salivary glands' ducts; the path of Wharton's duct is in an upward direction and the main portion of the duct is wider than the orifice.Along with these anatomical factors,the peculiar composition of submandibular gland saliva(alkaline and rich in mucin)contributes to the initial steps in the sequence of sialolith formation i.e.stasis of saliva and precipitation of salts and organic matrix formation. Dehydration,allergic states and infection of the oral cavity make saliva denser thereby initiating the formation of organic matrix with the accumulation of ductal debris and precipitation of mucoid elements and salts in order to form the organic matrix.When the stone reaches a size to obstruct the duct,secretion in the gland is hampered.This condition facilitates destruction of the gland.[7]

As no obvious precipitating factors were evident in our case,further in-depth review of literature showed that uncertainty about the causes and natural history of sialoliths and other obstructions is being dispelled by clinical and experimental research.Sialoliths are now shown to be secondary to chronic obstructive sialadenitis.Microscopic stones(sialomicroliths)accumulate during secretory inactivity in normal salivary glands and produce atrophic foci by obstruction.Microbes ascend the main salivary duct during secretory inactivity and proliferate in atrophic foci and cause spreading inflammation,leading to inflammatory swelling and fibrosis that can compress large ducts.This leads to stagnation of secretory material rich in calcium that precipitates onto degenerating cellular membranes to form a sialolith.[10]

Sialolithiasis is easy to diagnose on the basis of its clinical features.[11]Swelling is the commonest symptom in a submandibular sialolith(>80%),followed by pain(<50%) and 3% of patients are asymptomatic.The duration of symptoms varies from days to years and depends on its onset and the size and site of the sialolith.[7]The rate of enlargement of calculi is approximately only 1–1.5 mm per year.[12]Acute sialadenitis of the submandibular gland is the most common form of inflammation to the major salivary glands. Obstruction of the salivary duct due to sialolithiasis is the most frequent etiology of sialadenitis.Hence patients with sialolithiasis can have acute pain due to sialadenitis.[11]This was consistent with our case who presented with acute sialadenitis secondary to a childhood onset Wharton's duct calculus which gradually enlarged over years(due to the slow rate of enlargement of calculi).However,as pointed out by Shetty et.al,it is difficult to explain how such a large calculus can remain for years within its duct without causing any significant symptoms.[9]

Sialography is useful in patients showing signs of sialadenitis related to radiolucent stones or deep submandibular/parotid stones. Sialography is contraindicated in acute infection or patients who are having contrast allergy,and was thus not performed in our acute case.[11]Ultrasonography was performed,which detected sialadenitis. Sialolithiasis was detected on CT following a course of antibiotic. Szalma et al. showed that the sensitivity of ultrasonography for salivary stones was 75.0% and for inflammation was 77.4%.[13] According to Thomas et al. in sialolithiasis,the sensitivity and specificity were 65% and 80% for ultrasonography and 98% and 88% for CT,respectively.[14]

Different management options may be selected according to the size and location of the sialolith.Large submandibular calculi measuring several centimetres,symptomatic or silent,have to be removed promptly for prevention of possible complications that may occur in the future.[9]

Removal of the calculi from the duct transorally is the preferred treatment for anteriorly localised sialoliths in the absence of infection,as was planned in our case following resolution of acute inflammation.Submandibular gland excision is recommended for the posteriorly localised or intraglandular sialoliths.[15] Traditionally, recurring episodes necessitate treatment by open surgery.Once the diagnosis of an intraglandular salivary stone with destruction of the gland is established,removal of the entire gland via an extraoral approach is recommended.[16]Shock wave lithotripsy and interventional sialoendoscopy have also been used in the management of small sialoliths in recent years.

Perforation through the floor of the mouth by a submandibular sialolith,as seen in our case,is a rare phenomenon.In our case,the site of extrusion was allowed to heal secondarily without complications as is done usually following a sialodochotomy where there is no need to suture the incision,allowing the duct to heal spontaneously.[9]

CONCLUSION

There are various methods available for the management of sialolithiasis depending on the glandular involvement,the location and the size of the stone.Management modalities whether conservative or surgical, aim to restore normal salivary flow following the elimination of the calculus.This case demonstrates the rare self extrusion of a long standing giant sialolith and its successful resolution .

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