

Giant Wharton's duct sialolithiasis causing sialo-oral fistula: a rare case report with literature review

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ABSTRACT

Giant submandibular sialolithiasis is uncommon and sialoliths causing sialo-oral fistula are rare. We report a case of giant Wharton's duct sialolithiasis causing sialo-oral fistula in a 40 years male who presented with pain and swelling over right side of floor of mouth for more than two years and visualisation of yellow coloured stone like structure at floor of mouth in the last two days. With diagnosis of Submandibular gland sialolithiasis with sialo-oral fistula, sialolithotomy and marsupialization of Wharton's duct was done under local anaesthesia.

Keywords: Giant, masupilization, sialo-oral fistula, Sialolith, submandibular gland

Introduction

Sialolithiasis is one of the commonest diseases of salivary glands. Submandibular gland sialoliths account for 80-90% cases while parotid gland accounts for 5-10% cases, sublingual and minor salivary glands account for the rest.^[1] Sialolithiasis can occur at any age but is most commonly seen between fourth to sixth decades of life with males effected twice as females.^[2] Typical symptoms of sialolithiasis is described as 'mealtimes syndrome' which manifests as pain and swelling over the affected site during meals. Commonly sialoliths measure from 1 millimetre (mm) to 10 mm while giant sialoliths are characterised by size more than 15 mm and weight more than one gram. Sialoliths more than 35 mm in largest dimension are rare.^[3] Here we report a case of giant submandibular gland sialolithiasis measuring 42 mm

causing sialo-oral fistula. Such presentation of sialolithiasis is extremely rare.

Case Report

A 40-year-old male farmer reported with complaints of swelling over right side of floor of mouth and pain during meals for around 2 years and protrusion of stone-like structure from floor of mouth for the last 2 days. The swelling over floor of mouth had gradually increased in size and for the past 2 days patient started experiencing excessive salivation and a pale-yellow coloured swelling became visible over right side of floor of mouth. Patient's medical history was insignificant. Extraoral examination showed no significant findings. Intraoral examination showed a pale-yellowish stony hard structure protruding from right Wharton's duct orifice [Figure 1]. There was clear mucoid discharge seen from the duct. Rest of the oral cavity was unremarkable. The orthopantomogram was done and reported as single crescent-shaped radiopaque calculus superimposed over right canine, molars and right mandible extending from midline to distal molars over the edentulous ridge

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suggestive of right submandibular sialolithiasis [Figure 2]. The stone extraction with marsupialization of duct was done under local anaesthesia. The procedure was uneventful. Stone measured 42 mm in size and 15 gram in weight. [Figure 3] which is one of the largest reported in medical literature. After a year in follow up patient is asymptomatic.



Figure 1: Showing right Wharton's duct stone at floor of mouth



Figure 2: Orthopantomogram showing right submandibular calculus



Figure 3: Showing giant sialolith measuring 42 mm

Discussion

Presence of calculi in salivary gland or its ducts is known as sialolithiasis. Although it is second most common salivary gland disease after mumps affecting 12 per thousand adults, exact etio-pathogenesis of sialolithiasis is unknown.^[3] Sialoliths are most commonly found in submandibular gland with minor incidences in parotid and other salivary glands. Various factors have been attributed to submandibular sialolithiasis. Due to longer, tortuous and wider diameter of Wharton's duct, more alkaline and viscous content of secretion having mucin proteins, calcium and phosphate salts which flow against gravity predispose submandibular gland to frequent stone formation.^[4] Infections, salivary gland dysfunction, anomalous ducts, foreign body impaction and ductal epithelial metaplasia are other potential causes for sialolithiasis.^[5] Two phases of sialolith formation have been described namely central core formation and layered periphery formation. In first phase organic substance bound mineral salts precipitate to form the nidus or core. Thereafter in second phase, layered deposition of organic and inorganic material around the core forms the peripheral layer.^[2] Pain and swelling of involved gland are cardinal features of most of sialolithiasis due to blockage of lumen of gland ducts by stones especially during meals also called 'mealtime syndrome'. The pain and swelling may remain for hours followed by weeks to months of remission.^[3,5] However submandibular gland sialoliths may be asymptomatic. Recurrent infections of gland due bacterial translocation from duct to gland parenchyma may cause pain and swelling over gland.^[2] Sialoliths larger than 15 mm and heavier than 1 gram are called giant sialoliths. Sialoliths may attain considerable sizes to be accommodated in ducts. Giant sialoliths may causes various complications like ulceration of duct causing duct perforation and subsequent sialo-oral fistula formation as reported in this case. Giant Stones present in anterior ducts and in old age patients are more likely to cause duct perforation and fistula.^[6,7]

Diagnosis of sialolithiasis is usually clinical, based on thorough history and clinical examination. Radiological and special investigations may be required for confirmation of diagnosis. Location and size of stones is important for planning treatment of sialolithiasis.^[6,8] Standard mandibular occlusal radiograph is the standard imaging modality to view radiopaque sialoliths. Ultrasound, sialography, computed tomography and magnetic resonance sialography are other investigations which are good for locating radiolucent as well as radiopaque stones.^[4] Ultrasound provides accurate non-invasive imaging which can detect stones greater than 1.5 mm with 99% accuracy. Ultrasonography is also best method of demonstrating salivary flow after stones removal. Computed tomography although expensive is most accurate non-invasive imaging technique in locating stones. Sialography allows visualisation of whole glandular ductal system and calculi of all sizes along with functional assessment of gland.^[8]

Treatment of sialolithiasis should aim at preservation of gland function with minimal risk and discomfort to the patient.

Treatment is tailored based on size, location and number of stones.^[2] Conservative non-invasive management of small stones include administration of sialagogues and bimanual palpation and milking of stones through duct orifice after each meal. If stone is large extracorporeal shock wave lithotripsy, laser sialolithectomy or surgical removal of stone or gland should be planned.^[1,4] Giant sialoliths can be removed by sialendoscopy.^[8] Surgical management of sialolithiasis depends on location of stone in relation to gland and its duct. Submandibular gland duct called Wharton's duct is anatomically divided into three parts, anterior third, middle third and posterior third.^[3] If stone is located in anterior or middle third of duct then dilatation of duct or sialolithotomy is treatment of choice. If stone is located in posterior third of duct or in gland then sialolithotomy or sialadenectomy is treatment of choice.^[9] According to Cherian *et al.*^[1] sialadenectomy should be done only when small stones are present in vertical portion of duct or in gland parenchyma itself which produce symptoms and are not surgically accessible intraorally. Potential complications of sialolithotomy and sialadenectomy are duct stenosis and lingual nerve damage respectively.^[10] Medical treatment is indicated in case of gland infection to be followed by sialolithotomy. Post sialolithotomy patients should be advised to adopt diet rich in proteins, liquids and acidic foods. It helps in prevention of new stone formation in salivary glands.^[5] Post-operative medications for adults aim at infection control and analgesia. It usually includes amoxicillin 2 gram/day in divided doses for six days, antispasmodics for six days, analgesics for three days and chlorhexidine mouth wash three times a day for seven days.^[3]

Conclusion

Giant sialolithiasis is uncommon and stone causing sialo-oral fistula is rare. Pain and swelling during meals should raise suspicion of sialolithiasis. Various modalities of management of sialolithiasis like milking of stone, sialagogues, sialolithotomy, sialadenectomy are available and should be tailored according to patient and stone characteristics. Treatment should aim at complete stone clearance and preservation of gland function. Clinical knowledge of sialolithiasis, its complications and

treatment would help early diagnosis and management especially in rural areas by rural surgeons and primary care physicians.

Patient Consent

Informed written consent taken from patient for publication of images and clinical data.

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Conflicts of interest

There are no conflicts of interest.

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