Submandibular Gland Duct Giant Sialolith: Case Report and Literature Review

Maryam Sahwan, MUDr; CABS* Fatema Alasfoor, MD**

Sialolithiasis of submandibular gland duct is a common salivary gland pathology. Giant Sialolithiasis, on the other hand is quite rare. This report illustrates a unique case of an uncomplicated submandibular gland duct sialolith of an abnormal size that was managed with an uneventful and simple office-based transoral sialolithotomy, emphasizing the favorable outcome of such minimally invasive treatment option. This paper summarizes relevant published studies mainly from PubMed and MEDLINE databases on the clinical entity of giant sialolithiasis and its management.

Keywords: Sialolithiasis, Giant Sialoliths, Submandibular Gland Duct Sialoliths, Wharton's duct

INTRODUCTION

Sialolithiasis is defined as formation of stones in the salivary glands or their ducts and the term sialolith indicates a calcified mass within the gland or its duct. It is considered as a common non-neoplastic major salivary gland pathology. Submandibular gland is the most commonly affected[1-3] due to its anatomical and physiological characteristics that favor the formation of stones. Hence, it may harbor stones of greater than 15 mm that are termed giant sialoliths or megaliths[3-7]. In the year 2009, a published review of the literature reported 54 cases since 1942[4]. Since then, multiple cases have been reported with varying presentations, treatment approaches and complications. The infrequency of the condition along with the diversity of its clinical encounters renders it an important and interesting topic for reporting and discussing.

CASE REPORT

A young Bahraini gentleman who is not known to have any medical illness presented to our tertiary care hospital otolaryngology emergency referrals' clinic with a gradually increasing painful right submandibular area swelling of around 6 months' time. He reported that it worsened with eating. His history was otherwise unremarkable, and he did not seek any medical attention prior to that visit. On local examination, there was no significant palpable lump at the submandibular region. Trans-oral examination showed minimal congestion and edema over the submandibular duct orifice. Bimanual palpation revealed a hard bump over the right Wharton's duct. Head and neck examination were insignificant. Baseline blood investigations were within normal limits. Panoramic dental x-ray has been arranged to be done prior to the next follow up visit.

The provisional management plan was conservative, and patient was sent home on a course of oral antibiotic and analgesia. He was advised to increase the intake of sialagogues as well as to try to maintain good hydration and oral hygiene. One week later, he attended the outpatient clinic for follow up and he was still symptomatic. The Requested x-ray was not done due to patient reluctance. On trans-oral examination, a stone was seen projecting through Wharton's duct orifice. Bimanual palpation revealed the same hard swelling that was previously palpable in the duct area and in continuity with the projecting tip. At that time, no clue was evident regarding extent or size. Trans-oral sialolithotomy under local anesthesia was attempted as an office-based procedure. It delivered a large irregular elongated stone through the duct orifice. The duct orifice was marsupialized with an absorbable suture. The stone took the shape of the duct measuring around 40 mm in its largest diameter (Figure 1). The procedure was tolerated well with no acute complications. The patient was sent home a couple of hours after the procedure on oral antibiotics and analgesia. At two weeks follow up assessment, he was asymptomatic. Examination was unremarkable. Duct orifice started to heal and salivary flow looked normal.

Figure 1 : This picture illustrates the giant submandibular sialolith taken out by simple marsupialisation technique resembling the shape of wharton's duct with the longest dimension of 40 mm.

DISCUSSION

Sialolithiasis is one of the commonest non-neoplastic major salivary gland pathologies. Its incidence is estimated at 1.2%[4,5,7-9] in the general population. It usually affects adults with a male predominance[1-3,5,7-9]. Children are rarely affected[3,7,8]. It affects the submandibular gland in 80-90 % of the cases and the parotid in 10-20%[1-3]. Giant sialolithiasis on the other hand is not as common. It is defined as formation of a salivary gland stone larger than 15 mm in its greatest dimension[4,5,7]. Some authors define it by weight more than 1 gram[5,6]. Such stones are

* Fellowship Paediatric ENT
Senior fellowship in Paediatric ENT at Royal Manchester Children hospital, UK
ENT and Head and Neck department, Salmaniya Medical Complex
Kingdom of Bahrain
E-mail: Maryam.sahwan@gmail.com
** Resident
ENT, Head and Neck Department

termed megalith, giant sialolith or a salivary stone of unusual size. Usually, salivary stones size varies from 1 mm to 10 mm[3]. It rarely exceeds 10 mm[2,8]. Hence the peculiarity of the condition. There are less than 100 cases reported in the literature since 1942[4]. The largest reported to date was 72 mm[10].

Risk factors[2,8,11] for sialoliths formation are generally the same for ordinary and giant stones except for the possibility of chronicity that allows the larger size at diagnosis. Some factors are related to the salivary composition such as alkalinity, higher mucinous content and higher concentration of calcium and phosphate. Others are related to flow such as obstruction, stagnation or reduced salivary production secondary to dehydration. Besides, a diseased gland or duct due to infection, inflammation or trauma is more likely to form a stone. In addition to that, general medical condition of the patient such as tobacco smoking or exposure to certain medications may play a role[5,7]. Moreover, some states that aging increases likelihood of stone formation due to changes in the secretory activity, electrolyte composition and salivary gland cell membranes[3,11].

Submandibular gland is more likely to form stones hence more likely to harbor giant stones. It accounted for 46 out of 54 cases in a published review[4]. Its likelihood to form stones is secondary to its anatomical and physiological characteristics. Its duct has a wider diameter and longer course. It runs against gravity. Its salivary content is more alkaline and has a higher mucinous content and calcium concentration[2,7,8]. The megalith location could be ductal, hilar or intra-glandular. Generally submandibular gland sialolithiasis is commoner in Wharton's duct[2,5,8,9]. However, megaliths are believed to be commoner in the gland itself [2,8,9]. Nonetheless, published evidence is conflicting. In a review of 54 cases, there was an equal occurrence of ductal and glandular stones[4].

Though it is commonly mentioned that the exact pathophysiology of sialolithiasis is poorly understood[1,5,8,9], it is described to occur when mineral salts deposit around a nidus of bacteria, mucus or desquamated cells[5,7]. Amongst all, a mucus plug is believed to be the commonest nidus in a submandibular gland[9]. Stones are mainly formed of calcium and phosphate with traces of ammonia, potassium and magnesium[5,7,9]. For a stone to reach such unusual size it should cause an increased intra-ductal pressure without totally obstructing the salivary flow. This way, it leads to ductal dilatation and further maintenance of salivary flow. The continuous slow flow maintains continuous deposition and allows a marked increase in the stone size to reach unusual diameters. On top of that, at a stage that the stone is big enough to protrude in the oral cavity, it may further facilitate salivary flow and maintain deposition to create an even larger stone[3,5,7,11]. It is reported that a stone growth rate is estimated at 1-1.5 mm per year[6,9,11].

Patients presentation is generally like other salivary gland diseases. In a symptomatic patient, history is usually significant for obstructive salivary symptoms manifesting as recurrent acute attacks of submandibular pain exacerbated with meals[8]. Swelling in the submandibular area or floor of mouth may be present[1]. Some other patients are asymptomatic. These patients are diagnosed as incidental findings[5,7,9]. Another category of patients present with complications such as perforation of floor of mouth and sialo-oral fistula formation[2,5,11]. Natural history of the disease progresses as recurrent obstructive attacks with superimposed infective and inflammatory process. That may lead to neck space infections in forms of cellulitis or abscess formation, acute airway compromise, chronic sialadenitis, orocutaneous fistulous tract or floor of mouth ulceration or perforation with formation of a sialo-oral fistula which is a relatively rare occurrence[2,5,11]. If obstruction is long standing with no recurrent infections, it may lead to reduced salivary production and result in gland atrophy and ultimately fibrosis[9].

Examination is helpful to identify the underlying pathology. Bimanual palpation of the gland and duct may reveal an enlarged gland or a palpable stone. Intra-oral examination exposes the duct orifice inflammatory changes, any pouring pus, floor of mouth cellulitis or protruding stones.

Investigation of choice is plain film x-ray with preferably two views; not to miss a stone due to its posterior location or to visualize possible multiple stones[7]. Occlusal or panoramic orthopantomogram can be utilized[3,7]. Sometimes stones are missed on x-ray either due to their posterior location or their radiolucency. Thus, some indicates that ultrasound is the method of choice[5]. Computed Tomography (CT), Magnetic Resonance Imaging (MRI) and radionuclide imaging are indicated options if stone was radiolucent[7]. This may happen in around 20% of all submandibular gland stone cases[10]. However, it is worth noticing that most if not all giant sialoliths are radio-opaque secondary to their chronic formation that allows for calcification to take place[3,10]. Sialography is another modality of investigations that is mmentioned[7],but some authors do not recommend it due to its invasive nature[8].

Initial treatment is conservative[1] with use of sialagogues and gland massage. Concurrent sialadenitis need to be treated with analgesia and antibiotics. Classically the definite treatment aims to restore normal salivary flow with the least invasive technique[3,5,9]. There are various options available where the choice amongst them depends on several factors such as stone site, size, quality, number and shape[9]. Amongst well documented options are: transoral sialolithotomy, submandibular gland resection and interventional sialoendoscopy[9].

It is traditionally believed that the treatment of choice for intra-ductal stone is transoral sialolithotomy under local or general anesthesia[3,11]. The stone location is identified with palpation or probing[9]. It is then stabilized with proximal suture to avoid slippage[7]. The duct is identified prior to incision to avoid direct blind cut down that is associated with high risk of ductal stenosis[5,9,10]. An incision is made either parallel to the duct[7] or longitudinally over the stone[1,5,8-10] with caution to avoid lingual nerve injury[10]. The stone is then delivered. The wound is either left to heal via secondary intention[1], marsupialized with absorbable sutures [10] or repaired in layers[2] with a catheter in situ as a stent[1]. Some advocates resection of the overlying edematous mucosa prior to repair and keeping a vacuum drain[2].

Surgical excision of the submandibular gland via classic external approach if stone is hilar or intraglandular was and is till advised by some[3,4]. The aim is to remove the sialolith and the gland altogether to prevent recurrence of obstructive symptoms, avoid the possibility of proximal spillage of stone if accidentally fragmented and remove non-functional salivary gland after chronic inflammation[8]. Classical indications described include: significantly large stone within the gland, posteriorly located ductal stone, involvement of vertical part of the duct, atrophied non-functional gland and failure of conservative treatment[2,4-8,10,11]. Complications listed includes injury to marginal mandibular, lingual and hypoglossal nerves[1,2,4] where the commonest is the marginal mandibular nerve[1].

In the era of interventional endoscopy, some favor endoscopic retrieval when stone is ductal[8,9]. Using an interventional sialo-endoscope the accurate location of the stone is identified. Then it can be retrieved carefully. It carries the risk of proximal dissemination of the fragmented stone leading to recurrence[8]. Multiple emerging treatment modalities has been described with varying outcomes. Some mentioned transoral CO2 laser sialolithectomy[7,9]. A promising combined approach sialoendoscopically assisted open sialolithectomy has been first described in 2007 and was not utilized in giant sialoliths till later [2,4]. The stone is precisely localized with the use of an endoscope then delivered via limited transoral incision and dissection. It offers the option for proximal duct examination and wash. It was shown to have higher

rates of gland preservation and less risk of nerve injury independent of stone location[4]. Shock wave lithotripsy has been investigated as an option too. There are two means of use: Extracorporeal Shock Wave Lithotripsy (ESWL) versus Endoscopic Intracorporeal/ Intraductal Shock Wave Lithotripsy (EISWL)[2,5,9,11]. It is used to break the stone into smaller fragments that can be flushed away with normal salivary flow. It has limitations if a stone is relatively large and in the setting of acute infection[5].

Regardless of the modality of treatment used, patients are required to have interval follow up to review healing, asses for recurrence and monitor return of normal salivary flow[8]. However, it was not precisely mentioned if an optimal follow up timeline is documented for various modalities.

CONCLUSION

Submandibular gland sialolithiasis is a common encountered pathology in the otolaryngology general practice. However, presentation can vary from one patient to another and so is the approach of the management. This case presents an uncomplicated giant submandibular gland duct sialolith that was managed with an uneventful simple office-based extraction.

SUMMARY

• Giant sialolith is easily managed in clinic-setting.

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