CASE REPORT OPEN ACCESS

Bilateral Parotid Duct Stones: A Case Report

Aijaz Ali, Atif Hafeez Siddiqui, Salman Ahmed and Unsa Iqbal

Department of Otorhinolaryngology, Dow University Hospital, Karachi, Pakistan

ABSTRACT

Sialolithiasis predominantly affects major salivary glands with an incidence rate of 5.9 cases per 100,000 people yearly in the United Kingdom. Sialolithiasis is common in individuals aged between 30-60 years with higher prevalence in males. Among these, 80-90% of cases occur in the submandibular, 6-15% in the parotid, and 2% in the sublingual ducts or glands. Only 3-5% of cases occur in the minor salivary glands. Most parotid stones occur unilaterally involving either the right or left duct or gland parenchyma. However, bilateral parotid duct stones are an exceedingly rare condition. This article reports a unique case of a healthy adult male diagnosed as a case of bilateral parotid duct stones.

Key Words: Parotid duct stones, Salivary duct obstruction, Sialolithiasis.

How to cite this article: Ali A, Siddiqui AH, Ahmed S, Iqbal U. Bilateral Parotid Duct Stones: A Case Report. JCPSP Case Rep 2024; 2:186-188.

INTRODUCTION

Parotid glands are the largest of the major salivary glands secreting serous saliva which facilitates mastication and swallowing and initiates the digestion of starches in the mouth. 1,2 Although the aetiology of sialolithiasis is unknown, it can occur in any salivary gland or its duct and possible factors reported in studies include dehydration, poor diet particularly low fluid intake or high levels of calcium in the diet, anti-sialogogue medications, infections, and trauma to duct or gland. Comorbidities that help accelerate the formation of stones such as Sjogren syndrome (SS), diabetes mellitus, etc. can potentially lead to sialolithiasis and such cases have been reported.³ Salivary stones are composed of a mineralised centre as a nidus which progressively deposits layers of organic and inorganic substances in layers increasing their size with time. Bilateral parotid duct stones are an extremely rare diagnosis in clinical practice. Here, we present a case of bilateral parotid duct stones in a patient having no salivary gland disease.

CASE REPORT

A 60-year male smoker, diabetic and hypertensive, presented with a history of pain and swelling on the left side of the buccal cavity for one week. The patient was in his usual state of health 2 months ago when he first time noticed intermittent pain in his left cheek, particularly during meals. The pain was associated with mild-to-moderate swelling over the cheek which disappeared on its own a few hours after mealtime.

Correspondence to: Dr. Aijaz Ali, Department of Otorhinolaryngology, Dow University Hospital, Karachi, Pakistan E-mail: aijsoom@gmail.com

Received: January 25, 2024; Revised: May 18, 2024;

Accepted: May 22, 2024

DOI: https://doi.org/10.29271/jcpspcr.2024.186

A few days before reporting to OPD, he developed severe persistent pain and swelling on the left cheek and in the left side of the oral cavity associated with low-grade fever. The patient denied any pain or difficulty in swallowing liquids or solids. The patient had no history of trauma, dryness of mouth, dental and gum infection or discharge from the swelling on the face or inside the oral cavity. There was no history of facial weakness. There were no similar complaints for the right side of the mouth or face. The patient had 40 pack-year history of smoking. No other addictions were reported. His appetite was normal. Bowel habits and sleep were also normal. Past medical, surgical, and family history were insignificant.

General physical examination was normal. Oral cavity examination revealed redness and moderate tenderness in the left buccal cavity. A pea-sized hard mass, slightly movable, palpable under buccal mucosa in the left-upper gingivobuccal sulcus opposite 2nd molar tooth was noted. On pressing the parotid gland, frank purulent discharge was noted from the duct. Right-sided oral cavity examination also revealed a peasized hard mass palpable under buccal mucosa in the right upper gingivobuccal sulcus opposite 2nd molar tooth, with mild redness of mucosa and mild tenderness. The neck examination was normal. Cervical lymph nodes were not palpable.

CT scan revealed bilateral hyperdense structures at parotid duct levels on both sides (Figure 1). Based on the findings of clinical examination and CT, bilateral parotid duct stones were diagnosed.

Surgical exploration under general anaesthesia was performed. A stone measuring about 10 mm in length and 3 mm in width was retrieved from the left side. On the right side, stone of about 10 mm in length and 8 mm in width was retrieved from the right parotid duct (Figure 2). The sites of the incision were left open to heal naturally without closure.



Figure 1: CT scan showing bilateral parotid duct stones.



Figure 2: Stones after extraction through an open surgical approach.

DISCUSSION

Sialoliths represent approximately one-third of salivary gland diseases. Stone formation is more common in the sub-mandibular duct or gland due to sharp bends in Wharton's duct, viscous mucinous saliva, more calcium in the saliva, and the dependent position of the gland. Stone formation is less common in the parotid duct or gland due to the copious production of saliva which is serous.

Some studies have shown that tobacco smoking increases the cytotoxic activity of saliva, decreases polymorphonuclear phagocytic ability, a reduction of salivary amylase, and a reduction in salivary-protecting proteins, such as peroxidase. Studies also show a link between infection and sialolithiasis. A study showed no link between oral intake of calcium and hard water with sialolithiasis. A recent polymerase chain reaction (PCR)-based study found bacterial DNA, mainly of oral commensals belonging to the *Streptococcus* genus, in all examined sialoliths.

The exact pathogenesis is unknown, and various hypotheses have been proposed. The existence of intracellular microcalculi, which when excreted in the canal, may become a nidus for further calcification. Substances like food particles, abnormalmucous, or bacteria, desquamated cells and debris, and foreign bodies may obstruct the salivary duct and become the nidus for further calcification.

Investigations for sialolithiasis include standard x-ray films. The occlusal film shows ductal stones. Upto 20% of sialoliths are radiolucent and hence not detected on plain x-ray films. CT is the imaging modality of choice for the evaluation of salivary stones. This is because many calcified sialoliths are not detected by conventional radiography until they are 60–70% calcified. Ultrasonography (US) is a non-invasive method, but operator-dependent.

A study comparing US, sialography, and endoscopy demonstrated a sensitivity of 81%, a specificity of 94%, and an accuracy of 86% for US. In a study comparing magnetic resonance (MR) sialography and US, Jager *et al.* found a specificity and sensitivity of 80% for US.

Sialography is the gold standard investigation, it shows a clear image of stones and ductal morphologic structure. Sialography can be diagnostic for conditions such as Sjogren disease, or therapeutic due to the injection of dye in the duct, which sometimes extrudes the stone.

Sialolithiasis is managed conservatively, endoscopically, and by an open surgical approach. For small salivary stones (3 mm in the parotid duct and <4 mm in the submandibular duct), antibiotics and gentle massage, increased fluid intake, sialogogues, and moist heat may be enough. Some of them may require endoscopic removal with a retrieval basket. For large salivary stones, combined approach of sialendoscopy and ductotomy are required. In some cases, vein graft widening plasty or stenting may be required to prevent ductal stenosis.

In the literature review, five similar case reports have been found. The first ever reported case of bilateral multiple parotid calculi was in 1997. A 38-year male, AIDS patient, also affected by multiple myeloma, was reported by Ottaviani *et al.* The other four case reports involved patients suffering from SS. Dobai *et al.* reported a 41-year male, a case of SS, having multiple calculi in parotid glands bilaterally. Konstantinidis *et al.* reported a 49-year female patient, a case of primary SS, presenting with multiple stones in the parotid parenchyma bilaterally. Noreikaite *et al.* reported a 41-year female with a 2-year history of bilateral facial pain and swelling. This was a case of SS and hypothyroidism. The family history included severe rheumatoid arthritis in her mother.

In a study by Overton *et al.*, 55 consecutive patients underwent surgical removal of parotid stones from 57 glands, two patients having bilateral procedures with bilateral involvement of approximately 3.64% among cases of parotid sialolithiasis.¹⁰

Parotid duct stones presenting as bilateral pathology are extremely rare. Patients having comorbidities such as SS can present with bilateral parotid stones. Bilateral parotid sialolithiasis may have an association with smoking and can present without having any history of salivary gland morbidity.

PATIENT'S CONSENT:

Verbal and written consent was taken from the patient.

COMPETING INTEREST:

The authors declared no conflict of interest.

AUTHORS' CONTRIBUTION:

AA: Manuscript writing.

AHS: Concept, design, and analysis.

SA: Interpretation of data and analysis.

UI: Analysis and literature review.

All authors approved the final version of the manuscript to be published.

REFERENCES

- Chason HM, Downs BW. Anatomy, head and neck, parotid gland. [Updated 2022 Oct 24]. In: StatPearls [Internet]. Treasure Island (FL): StatPearls Publishing; 2024. Available from: http://www.ncbi.nlm.nih.gov/books/NBK534225/.
- Ghannam MG, Singh P. Anatomy, head and neck, salivary glands. [Updated 2023 May 29]. In: StatPearls [Internet]. Treasure Island (FL): StatPearls Publishing; 2024. Available from: http://www.ncbi.nlm.nih.gov/books/NBK538325/.
- Goli RR, Manesh R, Landry-Wegener B. bilateral sialolithiasis in a patient with sjogren syndrome. J Gen Intern Med 2020; 35(10):3091-2. doi: 10.1007/s11606-020-06054-z.
- Marchal F, Dulguerov P. Sialolithiasis management: The state of the art. Arch Otolaryngol Head Neck Surg 2003; 129(9):951-6. doi:10.1001/archotol.129.9.951

- Jager L, Menauer F, Holzknecht N, Scholz V, Grevers G, Reiser M. Sialolithiasis: MR sialography of the submandibular duct-an alternative to conventional sialography and US? Radiology 2000; 216(3):665-71. doi: 10.1148/ radiology.216.3.r00se12665.
- Ottaviani F, Galli A, Lucia MB, Ventura G. Bilateral parotid sialolithiasis in a patient with acquired immunodeficiency syndrome and immunoglobulin G multiple myeloma. *Oral* Surg Oral Med Oral Pathol Oral Radiol Endod 1997; 83(5):552-4. doi: 10.1016/s1079-2104(97)90119-0.
- Dobai A, Pataky L, Barabas J. Multiple microlithiasis in bilateral parotid glands as the initial clinical manifestation of primary Sjogren's syndrome. *Oral Radiol* 2018; 34(3): 267-72. doi: 10.1007/s11282-017-0294-8.
- 8. Konstantinidis I, Paschaloudi S, Triaridis S, Fyrmpas G, Sechlidis S, Constantinidis J. Bilateral multiple sialolithiasis of the parotid gland in a patient with Sjogren's syndrome. *Acta Otorhinolaryngol Ital* 2007; **27(1)**:41-4.
- Noreikaite G, Toscano ML, Shermetaro CB. Bilateral parotid gland punctate calcifications in Sjogren's syndrome: A case report. Radiol Case Rep 2021; 17(1):265-7. doi: 10.1016/ j.radcr.2021.10.049.
- 10. Overton A, Combes J, McGurk M. Outcome after endoscopically assisted surgical retrieval of symptomatic parotid stones. *Int J Oral Maxillofac Surg* 2012; **41(2)**: 248-51. doi: 10.1016/j.ijom.2011.10.010.

.