

BILATERAL MULTIPLE PAROTID CALCULI

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ABSTRACT

Bilateral multiple parotid calculi are rare entity. Calculi are found in normal individuals as well as in association with some local and systemic diseases. A case of bilateral multiple intra-parenchymal parotid calculi in and otherwise healthy patient who developed recurrent parotid swelling and had unusual cutaneous exfoliation of parotid calculi from the parotid region, is reported.

Human research ethics committee of Directorate Royal Medical Service agreed to prepare this case report, and the consent of the patient was obtained.

Key words: Sialolithiasis, parotid, bilateral, intraparenchymal, calculi

INTRODUCTION

Sialolithiasis is a common disease. Its incidence is higher than it is generally realized. Postmortem studies indicate that the incidence of sialolithiasis among general population is 1.2%.¹ Sialolithiasis occurs mainly in the submandibular gland (80-90%), and to a lesser degree in the parotid gland (5-20%). The sublingual and minor salivary glands are rarely affected.²

Parotid sialolithiasis usually involves one gland at a time, and the stones are usually solitary and often involves the ductal system of the gland. Simultaneous sialolithiasis of both parotid glands has a low incidence,² and the presence of multiple stones in the parenchyma of the gland is a rare finding.³

An unusual case of bilateral multiple intra-parenchymal parotid calculi occurring in the absence of any systemic or local disease and demonstrating unusual cutaneous exfoliation of calculi in the parotid region is described in this report.

CASE REPORT

A 32-year-old woman presented to oral and maxillofacial surgery department of this hospital in 2009

with a 4-year history of recurrent pain and swelling in the right parotid gland with fluctuation in size during meals and episodes of pus discharge from right Stenson's duct. Previous investigations included ultrasonography (US) of both parotids, and fine needle aspiration cytology (FANC) and sialogram of right parotid gland, and a diagnosis of obstructive sialadinitis with sialolithiasis was made.

When the patient reported her chief complaint was the presence of a subcutaneous nodule in the right parotid region. The patient's medical history was uneventful. Physical examination of the patient revealed an infra-auricular, hard and tethered subcutaneous nodule, and the skin overlying the nodule was mildly darkened. There was absence of swellings in both parotid regions. Intra-oral examination revealed patency of orifices of Stenson's ducts with clear salivary flow on gentle manipulation of glands. A computerized tomographic (CT) scan without contrast was taken. It revealed the presence of bilateral multiple intra-parenchymal parotid calculi of varying sizes and the presence of two calculi, one on each side, situated superficially in the subcutaneous tissues. The right and nearly fistulated calculus was the reason for the patient's chief complaint.

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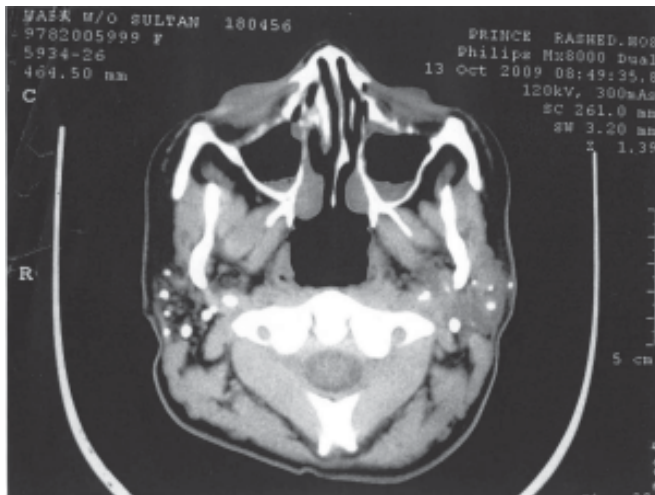


Fig 1: CT scan showing bilateral multiple parotid calculi of varying sizes. On the right side they are more.

The patient underwent an extensive investigation to exclude any systemic or local diseases or the coexistence of lithiasis in other body organs. The patient's uric acid, calcium and phosphorus levels were all within normal ranges. There was no subjective sensation of dry mouth or dry eyes. Unstimulated and stimulated whole saliva collection performed at 10:00 AM when the patient was fasting and hadn't brushed her teeth before the procedure. Stimulation was induced by asking the patient to chew on a paraffin block. The saliva was then collected by asking the patient to spit out actively into a calibrated container. The values for the whole saliva collection were 2.3 ml per 15 minutes for the unstimulated and 3.2 ml per 5 minutes for the stimulated. Labial minor salivary glands biopsies revealed absence of chronic inflammatory infiltrates. Autoimmune serology showed negative results for anti-Ro/SS-A, anti-La/SS-B and RF. ANA was 1/80. FNAC of both parotids was consistent with chronic sialadinitis. Kidney CT and gallbladder US revealed no stones.

In the face of long-standing and recurrent complaint, the decision was made to do right superficial parotidectomy. However, the patient didn't accept the possible complications and refused the surgery. The right subcutaneous stone was extirpated and the patient kept under follow-up examination. On the last recall in April 2011, both parotids were asymptomatic but the patient recalled cutaneous exfoliation of seven stones from both parotids.

DISCUSSION

Sialolithiasis may occur in normal people. Its exact pathogenesis remains unknown, and various hypotheses have been proposed.⁴ Lustmann et al² in a survey on 245 patients with sialolithiasis found no correlation between sialolithiasis and systemic diseases and only 6 of the 56 patients who were available for follow-up examination were reported to suffer from nephrolithiasis. The mineral composition of salivary calculi is different from that of biliary and urinary stones,⁵ and contrary to nephrocalcinosis and uroliths formation, the effect of hypercalcemia on the major salivary glands seems quite weak.³ Furthermore, Sherman et al⁵ found no correlation between water hardness and salivary calculi in England and concluded that local factors as yet unknown are likely to be important in salivary calculogenesis.

Parotid calculi are usually single, unilateral and located within the ductal system of the gland. Simultaneous bilateral multiple intra-parenchymal calculi like the present case are rarely encountered and few case reports or case series are found in the medical literature describing such finding. Lindamn and Woolley⁶ described one case in a child with absence of any local or systemic disease. Wickramasinghe et al⁷ reported a case with MALT lymphoma of the salivary glands who also had multiple calculi in the parotid sialolithiasis in a patient who had acquired immunodeficiency syndrome and was affected by multiple myeloma. Shimizu et al⁸ reported three cases of Sjogren's syndrome in whom multiple sialolithiasis were observed in the parenchyma of the parotid glands. Konstantinidis et al⁹ reported a case of bilateral multiple calculi of the parotids in a patient with primary Sjogren's syndrome. However, as the incidence of bilateral multiple parotid calculi, is very low their occurrence in those more common systemic diseases may be an incidental finding. Furthermore, this entity may occur in the absence of any systemic or local disease as is shown in the present case and in other case reports.⁶

The present case had undergone an unusual course of the disease. The cutaneous exfoliation or erosion of several calculi at different occasions without fistula formation may be attributed to the long course of the disease and the elevated intra-glandular pressure at times of recurrent obstruction. The secretory function

of the parotid glands haven't suffered considerably as clear saliva was expressed from the glands, however, the salivary flow rate hadn't been measured individually for the parotid glands. Although it is commonly believed that a gland with sialolithiasis is no longer functional, Marchal et al¹⁰ in their study on submandibular glands removed because of sialolithiasis demonstrated that there was no correlation episodes and despite appropriate indication for submandibular gland removal, close to 50% of the removed glands were histopathologically normal or close to normal.

Sonography is the first-line imaging for the evaluation of many diseases affecting the salivary glands including sialolithiasis.^{11,12,13} Its accuracy in assessment of sialolithiasis is high.^{13,14} However, US is less accurate than CT in distinguishing multiple clusters of stones from single large stones.¹⁴ Due to its anatomic position, a little portion of the parotid gland may be hidden by the acoustic shadow of the mandible, and therefore not visualized in US.¹³ Furthermore, the deep part of the gland is sometimes difficult to visualize.¹⁴ In the present case, US did reveal the presence of a number of stones, however, there was underestimation of the true multiplicity and location of the stones, and non-enhanced CT was invaluable in detecting the true extent of the disease.

An important differential diagnosis of the present case would be phlebolithiasis. Phleboliths are calcified thrombi found within vascular channels. Although phleboliths can occur in the absence of vascular lesion, they are often associated vascular anomalies,¹⁵ and in particular anomalies of low-flow such as venous malformations and cavernous hemangiomas. In fact, the presence of phleboliths is considered by some authors to be suggestive of cavernous hemangioma.¹⁶ Vascular lesions of the adult parotid gland is considered to be rare^{17,18} and the cases reported in literature have been described as being vascular malformations or hemangiomas, predominantly cavernous.¹⁸ Phleboliths of head and neck vascular lesions are usually multiple, varied in size and randomly distributed,¹⁹ and appear as dense, rounded opacities in contrast to the faintly opaque, scattered shadows of salivary stones.¹⁸ However, parotid stones occurring in the parenchyma of the gland may assume similar characteristics of a phlebolith. Nonetheless, in the present case a vascular lesion was

excluded by the cumulative analysis of patient's history, physical examination and investigations.

Multiple intra-parenchymal parotid stones are considered not straightforward cases. The management of such cases is not clear-cut. In the past, such cases were treated by superficial parotidectomy. Minimally invasive techniques, especially endoscopy of the salivary glands, had reduced the number of cases in which superficial parotidectomy is indicated.²⁰ Complicated cases have been treated by many surgeons by a combination of techniques. Nahlieli et al²¹ in their prospective study assessed a combined external lithotripsy-sialoendoscopic method for advance salivary gland sialolithiasis and the success rate for complete removal of the stones after lithotripsy sessions was easier and less complicated. In another article, Nahlieli et al²⁰ discussed a new approach to impacted parotid stone included endoscopy and US technique to perform precise removal. They achieved complete removal of stones in 75% of cases, and multiple sialoliths were detected and removed from 4 glands. Kulkens et al²² demonstrated a connection between the location of the parotid calculi and the success of extracorporeal shock wave lithotripsy (ESWL), in their study 10 patients had intra-parenchymal calculi, and after ESWL treatment 5 of them were free of calculi.

In the present case, the decision was to perform superficial parotidectomy for the effected gland due to the unavailability of the minimally invasive technique. The patient refused such radical treatment and so was kept under follow-up examination. As the patient remained symptom-free during the 2-year follow-up, no intervention was indicated. If symptoms supervene in the future, hopefully ESWL would be an available option as a last resort before superficial parotidectomy.

CONCLUSION

A bilateral intra-parenchymal parotid calculus is a rare entity. Its association with any systemic or local disease is uncertain. CT is invaluable in detecting the disease. Such cases may be mistaken with other local diseases that show calcifications, such as phlebolithiasis. The management of such cases is not straightforward, and superficial parotidectomy may be inevitable.

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