

Spontaneous Extrusion of Parotid Sialolithiasis through a Cutaneous Fistula: Case Report

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Abstract

Parotid sialolithiasis is a rare pathologic disorder when compared to those of the submandibular salivary glands. Furthermore, infection arising parotid sialolithiasis is very rare to be a causative factor for the development of cutaneous parotid fistulae. Only 3 cases had been reported earlier in which these salivary stones expelled spontaneously through the related skin fistulae with total remission of all signs and symptoms of the affected gland. A case of two parotid stones inducing inflammation, infection, and discharging cutaneous fistula in a 77 years old Saudi male that had been initially misdiagnosed spontaneously extruded through the cutaneous fistula with total remission of all symptoms will be presented.

Keywords: Parotid Sialolithiasis; Parotid Salivary Stones; Cutaneous Parotid Fistula; Spontaneous Extrusion

Introduction

The parotid gland represents the largest of three pairs of major salivary glands; namely the parotid, the submandibular and sublingual glands. Different pathologic lesions may affect these major salivary glands varying from inflammation to neoplasm with sialolithiasis being the commonest pathologic changes. The parotid gland sialolithiasis is an uncommon condition and account only for 6% of all major salivary glands sialolithiasis [1]. The parotid sialolith (stones or calculi) are characterized by having less inorganic (49%) and more organic component (51%) as compared to the submandibular gland stones which composed of 82% inorganic and 18% organic material only [2]. This makes the diagnosis and identification of parotid salivary stones by plain x-ray a more difficult task.

The size and location of these salivary stones are the main factors that determine the severity of their induced symptoms and their treatment of choice. Pain, recurrent infections, and facial swelling, in addition, to reduce salivary flow from the affected gland are the main presenting symptoms for parotid gland sialolithiasis. Very rarely the presence of a cutaneous fistula will be associated with the presence of these salivary stones and more rarely the stones will be extruded from these fistulae without any surgical intervention [3-5].

A case of a 77 years old Saudi male patient with two parotid stones that spontaneously extruded from the related cutaneous fistula will be presented.

Case Report

A 77 years old Saudi male patient was referred eventually to the oral and maxillofacial surgery clinic after repeated failures of treatment of a cutaneous fistula with pus discharge in the left cheek area by his treating general surgeon, dermatologist and general dentist. The treatment provided by the patient's general surgeon and dermatologist includes antibiotic therapy and local debridement and surgical dressing on the assumption that the infection is from an infected sebaceous gland cyst. Whereas, the treating general dentist assumed

that the facial sinus is due to infection arising from the remaining roots of the left maxillary teeth and performed extraction of the affected teeth similarly with no improvement of the presenting symptoms (Figure 1).



Figure 1: Clinical view at the time of presentation showing a cutaneous fistula on the left cheek (arrow) with indurated margins.

The clinical findings at the time of presentation include; a cutaneous fistula with pus discharging over the left cheek area without any facial asymmetry, intra-oral satisfactory clear salivary flow from the left parotid duct's orifice, and no noticeable regional lymphadenopathy on the left side was noted. When the patient's ortho-pantomo-graphic (OPG) x-ray view was carefully reviewed two small lightly radio-opaque bodies were noted over the maxillary molars region (Figure 2). Therefore, the presence of parotid stones was suggested and the patient was then referred for CT scan examination.

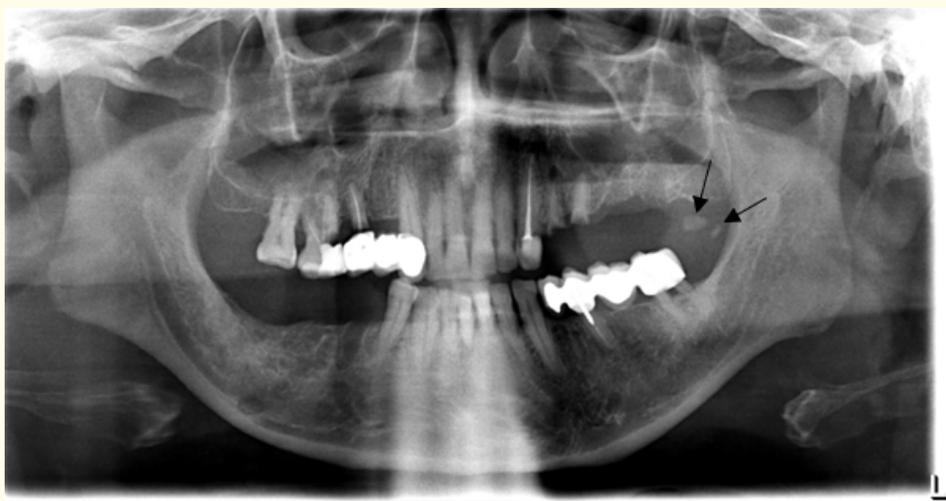


Figure 2: Initial Ortho-pantomo-graphic (OPG) x-ray view showing two lightly calcified bodies (arrows) on the left side parotid area unnoticed by treating physicians and dentist.

The pre and post-I.V. CT scan study done showed relative heterogeneous density and enhancement of the left parotid gland. The oval shaped hypodense area is seen in the posterior part of the superficial parotid lobe measured 17 x 6 mm representing parotid lymph node. Dilated lumen and enhancing walls of the parotid duct was mostly due to two salivary stones appear at its distal end just lateral to the left parotid superficial lobe measuring 8 mm and 5 mm in maximum dimension. Skin and subcutaneous fat overlying the salivary stones show hypodense areas and stranding with relative enhancement suggesting inflammatory reaction. These findings are impressive of left parotid sialoadenitis and sialolithiasis (Figure 3).



Figure 3: Initial CT scan view showing two salivary stones of the left parotid gland (arrows).

Thereafter, the decision of surgical intervention was made to remove the salivary stones and excision of the cutaneous fistula together with superficial parotidectomy. The patient was reluctant to accept the proposed surgical treatment when it was discussed with him and demand more time to think about it. Therefore, an alternative more conservative treatment was planned for him to try during this time which includes; antibiotic therapy with Cefuroxime Axetil (Zinnat) 500 mg tablets twice daily for 5 days, frequent intake of citrus unsweetened drinks to enhance saliva production, and gentle massage over the affected gland aiming to enhance the salivary flow. The patient accepted the new conservative treatment willingly.

One week later, the patient attended for a follow-up visit and reported that two small seed-like bodies extruded from the cutaneous fistula about two days ago followed by cessation of the pus discharge from the facial fistula. Clinical examination showed that the cutaneous fistula starts to heal and only wound debridement and dressing were performed at that time. Unfortunately, the patient discarded the extruded masses, therefore, he was referred for a follow-up ortho-pantomo-graphic (OPG) x-ray view and CT scan to confirm presence or absence of the salivary stones. The results of both radiographic investigations confirmed the absence of the two salivary stone (Figures 4 and 5).



Figure 4: Ortho-pantomo-graphic (OPG) x-ray view one week after initial presentation confirming spontaneous extrusion of the two salivary stones of the left parotid gland.

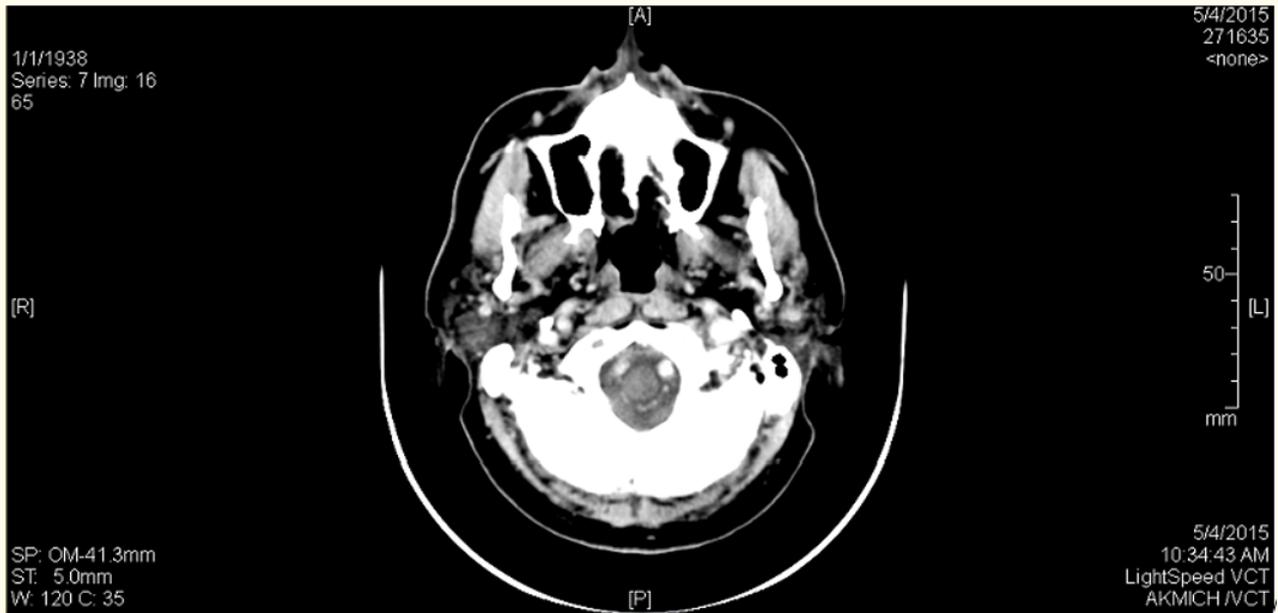


Figure 5: Follow-up CT scan view showing clear left side parotid area following extrusion of the two parotid stones.

The patient was called for another follow-up appointment a week later and clinical examination confirmed complete uneventful healing of the cutaneous fistula with normal clear salivary flow from the left parotid gland. The patient was followed up for another 16 months and did not show any pieces of evidence of recurrence of his initial symptoms and/or any complaints in regards to his left parotid gland function and health which is confirmed by a CT scan performed at that follow up visit (Figure 6).

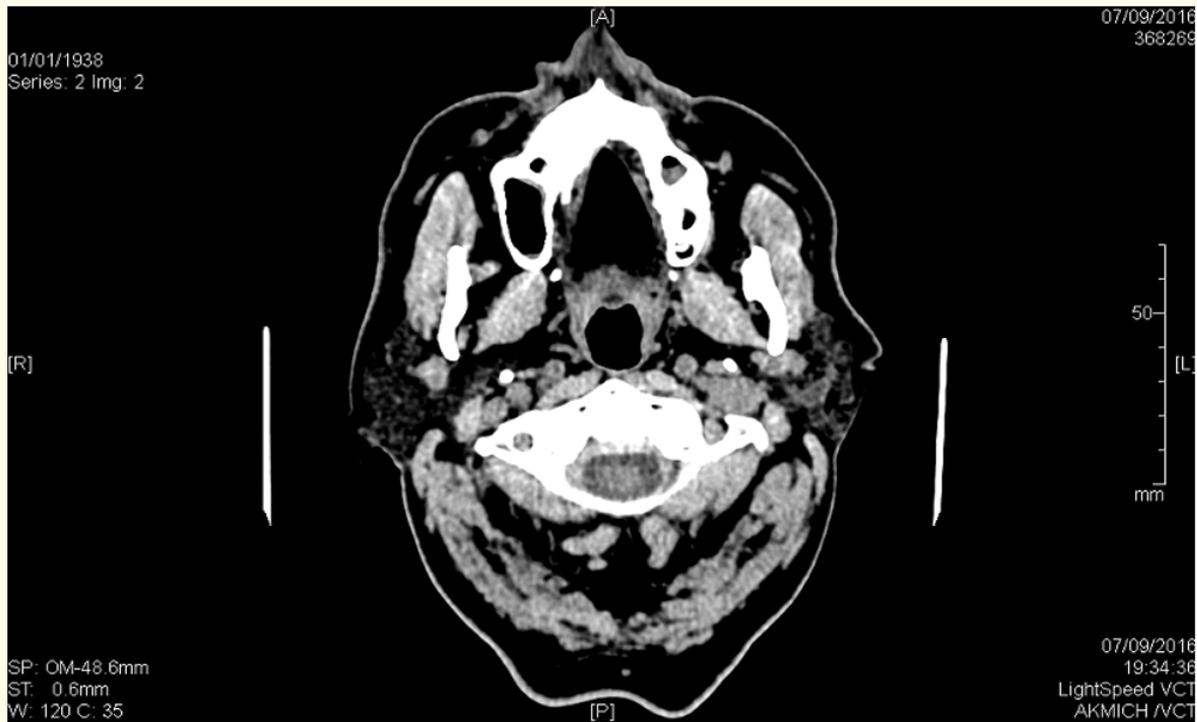


Figure 6: Follow-up CT scan view 16 months later following the extrusion of the salivary stones showing clear left parotid gland area.

Discussion

The presence of salivary stones in the parotid gland is quite rare as it represents only 6% as compared to its existence in the submandibular salivary glands where it counts for more than 80% of all stones of major salivary glands [1]. The differences in the composition of parotid stones since it is formed of more organic than inorganic component as compared to the submandibular ones creates extra difficulties in the ease of its diagnosis without CT scan and ultrasound examination. The development of cutaneous salivary fistulae as a result of present parotid stone is being extremely rare in medical literature, and spontaneous extrusion of these stones through the related fistulae was reported only three times as an exceptionally rare phenomenon. The first case was being reported by Goudot, *et al.* in 1986 [3] where they report a case in a 42-year-old male, followed after 27 years in 2013 by the case of a 59-year-old male patient reported by Abadi, *et al.* [4]. Later on in 2016 Brown, *et al.* [5] presented the third case of a 59-year-old Australian lady with recurrent episodes of parotitis and facial pain, which resolved through spontaneous extrusion of the stone through a cutaneous fistula while awaiting surgery.

The case presented represents the fourth case in the medical literature to describe such an extremely rare presentation of parotid sialolithiasis. In addition, it shares with previous publications the similarity of spontaneous healing of the cutaneous fistula and the disappear-

ance of any symptoms of inflammation and/or infection of the affected gland following the extrusion of the salivary stones. These findings may suggest a more conservative management of the parotid gland in cases of related cutaneous fistula formation as a result of salivary stones existence.

Conclusion

Parotid glands salivary stones although they are rare as compared to the submandibular salivary stones still, they have to be considered in the differential diagnoses of facial cellulitis and infection. Plain x-ray views in such cases may add little to diagnosis but still, they should be carefully reviewed. Additional investigations including CT scan, MRI, and ultra-sound examination should be requested to confirm the diagnosis.

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