CASE REPORT

Spontaneous cutaneous extrusion of a parotid gland sialolith

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Accepted 16 April 2016

SUMMARY

Parotid gland sialolithiasis is an uncommon condition that can cause pain and recurrent infection in affected patients. Migration of a stone through a fistula is a rare but possible complication of untreated sialolithiasis. We present a case of parotid gland sialolithiasis in a 63-year-old woman with recurrent episodes of parotitis and facial pain, which resolved through spontaneous extrusion of the stone (11 mm) through a cutaneous fistula while awaiting surgery. Management is typically conservative or surgical, depending on the location and size of the stone, and the clinical presentation.

BACKGROUND

Salivary stones, while benign, can be troublesome for patients who develop obstructive symptoms on eating and are predisposed to recurrent bacterial infection. While the majority of salivary gland calculi are found in the submandibular gland, between 5% and 20% of stones are located in the parotid gland.¹ Patients with parotid sialolithiasis usually report of unilateral painful swelling over the parotid region, caused by an obstructing stone in the salivary duct of the gland, and present to either emergency medical or dental centres for assessment.² The management of the condition depends on the size and location of the stone, and the severity of the patient's symptoms.³ Smaller stones may be responsive to conservative treatments such as hot compresses, massage, increased oral fluid intake and encouraging salivation with sialogogues.³⁻⁴ Surgical treatment is, however, indicated to prevent recurrent parotitis. This patient had been correctly identified to require surgical management after a prolonged trial of conservative measures, however, significant delay resulted in spontaneous extrusion of a large stone through the skin. The purpose of this case report is to highlight a potential pitfall of conservative management for patients with parotid stones.

CASE PRESENTATION

A healthy 63-year-old woman with recurrent parotitis initially presented to our otolaryngology, head and neck clinic following discovery of a parotid sialolithiasis on ultrasonography, in 2010. The scan revealed an 11 mm calculus lying in the intraglandular duct near the anterior margin of the gland, with heterogeneous tissue suggestive of vascular parotitis. The patient was managed conservatively at the time, with massage, hot compresses and antibiotic therapy.

She presented to the emergency department the following year, with right-sided preauricular facial swelling on a background of 3 months of recurrent obstructive symptoms described as right facial pain exacerbated by consumption of food. She was afebrile on assessment and examination of the face demonstrated a large painful right facial swelling with pus expressed from the parotid duct papilla. This was suspicious for an abscess, and she was further investigated with imaging in the emergency department. An ultrasound was repeated, demonstrating the 11 mm calculus situated within the proximal aspect of the right parotid duct causing dilation of the mid to distal portions. The scan also showed significant inflammation of the parotid gland parenchyma around the calculus, with no drainable abscess demonstrated (figure 1). The patient went on to have a CT scan, which diagnosed obstructive parotitis on the right side due to ductal stenosis, the latter secondary to the extruded calculus lying externally at the mid-portion of the main parotid duct (figure 2). The patient was given a further course of oral antibiotics and subsequently referred for surgical excision of the stone via parotidectomy.

Several months later, she re-presented to clinic prior to surgical intervention reporting spontaneous discharge of the stone through a cutaneous fistula. On examination, there was evidence of wound healing on the skin overlying the parotid (figure 3) and resolution of the painful palpable mass in the right parotid region.

OUTCOME AND FOLLOW-UP

The patient was discharged from the outpatient clinic as her symptoms had resolved, demonstrating a rare case of a favourable outcome for this patient with a functional parotid gland and minimal scarring.

Figure 1 Ultrasound demonstrating 11 mm calculus within the right parotid duct, causing mid to distal ductal dilation with associated oedema.

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To cite: Brown K, Cheah T, Ha JF. *BMJ Case Rep* Published online: [please include Day Month Year] doi:10.1136/bcr-2016-214887

Figure 2 CT scan showing extruded calculus situated externally to the mid-portion of the main parotid duct.

DISCUSSION

Lifetime prevalence of sialolithiasis in any salivary gland is around 1–2%, however, many cases are asymptomatic and are found incidentally.³ The exact aetiology of sialolithiasis is unclear, however, it has traditionally been attributed to reduced or obstructed salivary flow rate, dehydration, changes in salivary pH and the precipitation of salts.^{4 5} In addition, it has been hypothesised that retrograde migration of oral materials and bacteria act as a nidus for development of sialoliths.⁶ Stones are usually comprised of calcium and phosphorus, however, serum

Figure 3 Healing skin overlying the right parotid gland with closure of the cutaneous fistula.

levels of these ions are not associated with increased occurrence.⁷ Development of salivary calculi within the parotid gland is less common as the saliva produced is entirely serous and has lower pH, calcium and phosphate content in comparison to the submandibular gland, in which the majority of stones occur.⁸ Parotid stones are most commonly located in the hilum or parenchyma of the gland, with only 10–20% of stones found in the parotid duct.³ Presence of intraductal calculi leads to obstruction, which causes spasm on eating and predisposes patients to recurrent bacterial infection leading to chronic bacterial sialadenitis.⁴

In all patients with a salivary gland stone, a trial of conservative measures is recommended prior to proceeding with more invasive treatment options. These conservative measures include stimulation of salivary flow by massage or using sialogogues, anti-inflammatories and external heat, and judicious use of antibiotics.^{3 4 9} If conservative approaches are unsuccessful at relieving symptoms, surgical removal of the sialolith is then indicated. With parotid sialoliths, the various surgical options are dictated by the location of the stone and the anatomy of the parotid duct in relation to the gland.³ Traditionally, if the stone is positioned medial to the masseter muscle, then an intraoral incision of the duct may be attempted, whereas it may be possible to express the stone if it lies close to the surface of the duct. However, if the stone is lateral to the masseter muscle and therefore intraparenchymal, then parotidectomy is considered the procedure of choice if minimally-invasive surgical options are not available.^{3 9}

In recent years, the advent of sialendoscopy and application of extracorporeal shockwave lithotripsy (ESWL) in the management of sialolithiasis has modified treatment algorithms in countries in which these are readily available. Depending on their location and size, parotid stones can be removed with sialendoscopy, ESWL, or a combination of both.^{5 9 10} If ESWL is not available, larger or intraparenchymal parotid stones may be managed using a combined sialendoscopic and open approach.^{11–14} In these cases, a transcutaneous incision is made over the stone and duct, which has been transilluminated through the skin with the sialendoscope.^{11 13} The parotid gland is thereby preserved with a reduction in the risk of facial nerve damage.¹¹

Commonly recognised complications of chronic obstructive sialolithiasis include infection and ductal strictures. Drage *et al*¹⁵ reported migration of salivary calculi as a potential complication of conservative management and presented three patients with stones that migrated to the adjacent dermis and were removed under anaesthesia. The authors postulated that migration may be a consequence of abscess formation, as was their finding in their limited series, however, this was not demonstrated in our patient. Furthermore, migratory salivary stones with cutaneous extrusion are a rare complication and have not been reported frequently in the literature.¹⁶ Only three papers were identified reporting spontaneous extrusion of parotid sialoliths through the skin, one of which occurred following iatrogenic perforation of the parotid duct during sialendoscopy.^{15–17} A common feature of these cases was a history of recurrent infective episodes, in keeping with our patient's history.

This case describes an 11 mm migratory stone that was well defined on CT scan and ultrasound sonography; it is one of the most well-imaged migratory stones described in the literature.

Learning points

- ▶ Sialolithiasis of the parotid gland is a relatively uncommon condition that can present to both dental and acute medical practices.
- ▶ Traditionally, the management of parotid sialolithiasis is either surgical or conservative depending on the position and size of the stone within the parotid gland.
- ▶ While conservative management is well tolerated, a prolonged clinical course of obstructive sialolithiasis results in recurring infection and ductal strictures regardless of stone location, and should prompt referral for surgery. Formation of a cutaneous fistula is a rare outcome.

Contributors KB wrote the initial manuscript. JFH supplied the figures and supervised the writing of the manuscript. TC edited, prepared and submitted the article.

Competing interests None declared.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

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