CASE REPORT

Transcervical approach to the removal of a deep-seated giant submandibular calculus and the submandibular gland

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SUMMARY

Sialolithiasis is among the most common disease affecting the major salivary glands whereby the submandibular gland or its duct is affected in the majority of cases. We report a case of the surgical removal of a giant sialolith along with the submandibular gland using the transcervical approach and its clinical outcome.

BACKGROUND

The submandibular gland is the most common major salivary gland affected by calculi. It is said to affect 12 per 1000 of the population¹ and is two times more likely to affect middle-aged men than women.² This is owing to the fact that the submandibular gland contains a long and tortuous upward submandibular duct and the mucoid secretions produced by the gland itself. The calculus, formed by the deposition of calcium salts, may organise around a collection of debris located within the submandibular duct (Wharton's duct) or the gland itself.³ Occasionally, a giant salivary gland calculus (GSGC) (defined as a calculi measuring >15 mm)⁴ may present as a slow-growing lump in the neck with recurrent sialadenitis and abscess formation. A giant submandibular calculus such as that seen in this case may be firmly adherent to the submandibular gland and may require the removal of the calculi along with the underlying gland.

CASE PRESENTATION

A man aged 34 years with a history of recurrent episodes of left submandibular swelling for the past 5 years had presented to the otorhinolaryngology department with the chief symptom of increasing pain, fever and discharge coming from his oral cavity. On further history taking, it was noted that he had been visiting multiple doctors in the past few years and was treated for recurrent sialadenitis with antibiotics. On completion of each course of antibiotics, his fever and oral discharge had subsided and the submandibular swelling had reduced in size but had never resolved.

On examination, there was a vague mass at the left submandibular region measuring ~4×4 cm (figure 1) which was mobile but hard in consistency. Oral examination revealed no calculi within the submandibular duct and the floor of mouth was not raised. There was pus discharge arising from the submandibular duct when the patient had

presented to us during the suppurative phase of the disease but resolved after the completion of a course of antibiotics. Flexible nasopharyngolaryngoscopic examination was unremarkable.

INVESTIGATIONS

An orthopantomogram (OPG) revealed a calcified structure within the left submandibular region (figure 2). This was further supported by a CT scan of the neck which revealed an enlarged submandibular gland with evidence of a calculus within measuring ~2.5×3.5 cm (figure 3) associated with a dilated submandibular duct. The postoperative histological examination of the submandibular gland revealed chronic inflammatory changes to the submandibular gland (chronic sialadenitis) and was negative for malignancy.

DIFFERENTIAL DIAGNOSIS

The most likely differential diagnosis was that of a giant submandibular gland calculus within the submandibular gland. This is supported by the clinical presentation of recurrent left submandibular swelling, discharge from the oral cavity and the radiological evidence of a calcified structure at the submandibular gland. Other differentials include a calcified lymph node, retained foreign body or Kuttner's tumour (Chronic sclerosis sialadenitis).

Figure 1 Preoperative picture showing a vague swelling arising from the left submandibular region.

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Figure 2 Plain OPG (orthopantomogram) showing a calcified structure within the left submandibular region.

Figure 3 CT scan of the neck showing a large calcified structure (red arrow) within the left submandibular region.

TREATMENT

The patient was then planned for a submandibulectomy with the removal of the giant calculus after the infection had resolved. A trans-cervical approach was used for the removal of the gland due to the close relation of the calculus to the underlying gland. A horizontal incision was made below the lower border of the left mandible after infiltrating 10 mL of local anaesthesia (bupivacaine+epinephrine 1:100 000). The gland and the calculus were carefully dissected ([figure 4](#)) while preserving the underlying structures such as the hypoglossal, lingual and marginal mandibular nerves. The submandibular duct was then ligated and the calculus was then inspected. The length and width of the calculus measured 3.5×2.5 cm ([figures 5 and 6](#)), respectively, and weighed ~10 g which was among the largest submandibular calculus removed by us in our clinical practice. On intraoral examination, there was small defect at the floor of mouth after the removal of the calculus which suggested an underlying attachment to the floor of mouth. The intraoral defect was then closed using absorbable (Polyglactin 3/0) sutures. A nasogastric tube was then inserted and the patient was then fed exclusively through the nasogastric tube for 1 week until the wounds had healed.

OUTCOME AND FOLLOW-UP

The patient had made a full recovery on being discharged a week after the surgery. At follow-up 1 month after the surgery, there was no evidence of recurrence or swelling at the operated site and he had been asymptomatic ever since.

DISCUSSION

Sialolithiasis is a common but poorly understood disease that affects the major salivary glands. Its pathogenesis is still highly debated, but it is generally agreed that the formation of a

Figure 4 The left submandibular gland and calculus being slowly dissected from the underlying tissue.

calculus requires three conditions: salivary stagnation, a nidus and precipitation of salivary salts.⁴ Subsequent infection, inflammation and abscess formation may further precipitate the formation of a calculus⁵ or enlarge the calculus further.

More than 80% of sialolithiasis tend to occur at the submandibular gland or duct and the remaining in the parotid, sublingual and minor salivary glands.⁶ The higher predisposition of a calculus forming in the submandibular gland is thought to be due to higher alkaline and mucinous secretions from the submandibular gland and also due to the longer and larger calibre of the submandibular duct.⁷ Occasionally, GSGC may form after years of precipitation and its formation may be either intraductal or within the gland itself such as the one seen in the presented case. The shape of the calculus obtained conforms to the site where the calculus forms. Hence, if the calculus precipitates within the submandibular gland, it tends to form an ovoid or round mass, whereas if the calculus precipitates within the submandibular duct, it tends to form an irregular or elongated-shaped mass.⁸ The calculus obtained in our case appears to be a rounded mass with faceted ends which suggests that the calculus had precipitated within the submandibular gland and may have partially extended into the submandibular duct.

Diagnosis of a giant submandibular calculus is fairly straightforward with detailed history and examination. However, cases are still being misdiagnosed as it may be confused with other conditions such as lymphadenitis as a submandibular calculus rarely enlarges to such an extent. Investigations such as a plain OPG radiograph can be used to confirm the diagnosis and a CT scan can be used for detailed localisation of the calculus and its relation with other underlying structures.⁷ Ultrasound scanning is a cheaper, faster and well-established method for the diagnosis of a submandibular calculus as it is able to detect submandibular calculi of above 2 mm with reliable accuracy. It visualises the calculus and the underlying submandibular gland.⁹ Other investigations include sialograms⁷ and sialoendoscopy which can be useful in other modalities of calculus removal

Figure 5 The length and height of the removed calculus measuring 3.5×2.5 cm, respectively.

Figure 6 The calculus and submandibular gland removed en bloc.

such as the endoscopic extracorporeal short-wave lithotripsy and stone removal,¹⁰ especially when the stone is at about 5 mm in size and is freely lying within the lumen of the submandibular duct.

Although it is generally agreed that the calculus should be removed with the least invasive method available, the calculus may be firmly adherent to the gland which makes minimal invasive techniques like the transoral approach difficult. In such cases, the removal of the calculus and the submandibular gland is generally carried out using a transcervical approach. There are disadvantages in using the transcervical approach such as an unsightly scar, more postoperative pain, longer hospital stay and higher risks of nerve injury in comparison with the transoral

approach¹¹ and this should be discussed with the patient prior to surgery. Thankfully, we did not detect any long-term sequelae as a result of the surgery aside from a horizontal scar above the operated site. It should also be remembered that when approaching a giant submandibular calculus, there is a high likelihood that it may have adhered to the floor of mouth such as the one seen in this case. Careful manipulation of the calculus while removal is crucial as the floor of mouth runs a risk of being torn while the calculus is being removed.

Learning points

- ▶ Although rare, a giant submandibular calculus should be considered as part of the differential diagnosis when dealing with a chronic submandibular swelling with recurrent episodes of submandibular pain, fever and intraoral discharge.
- ▶ The trans-cervical approach of the removal of a submandibular calculus should be performed when transoral approach is impractical. In this case, the calculus was seen to have adhered to the submandibular gland and was fairly distal to the floor of mouth; hence the transcervical approach was employed.
- ▶ Careful manipulation of a giant submandibular calculus is needed when attempting to remove the calculus as the floor of mouth may be torn when trying to remove the calculus.

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Contributors LCG drafted the article, revised it and managed the case accordingly. BKC and MMS managed the case and performed the surgery. WSJN was involved in patient care and revision of the article.

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REFERENCES

- 1 Rauch S, Gorlin RJ. Diseases of the salivary glands. In: Gorlin RJ, Goldman HM, eds. *Oral pathology*. St Louis: CV Mosby, 1970:962–1070.
- 2 Cawson RA, Odell EW. *Essentials of oral pathology and oral medicine*. 6th edn. Edinburgh: Churchill Livingstone, 1998:239–40.
- 3 Brad WN, Douglas DD, Angela CC, et al. *Oral and maxillofacial pathology*. 4th edn. Elsevier, 2016:427–9.
- 4 Bodner L. Giant salivary gland calculi: diagnostic imaging and surgical management. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2002;94:320–3.
- 5 Raksin SZ, Gould SM, William AC. Submandibular duct sialolith of unusual size and shape. *J Oral Surg* 1975;33:142–5.
- 6 Siddiqui SJ. Sialolithiasis: an unusually large submandibular salivary stone. *Br Dent J* 2002;193:89–91.
- 7 Fowell C, MacBean A. Giant salivary calculi of the submandibular gland. *J Surg Case Rep* 2012;2012:6.
- 8 Oteri G, Procopio RM, Cicciu M. Giant salivary gland calculi (GSGC): report of two cases. *Open Dent J* 2011;5:90–5.
- 9 Jäger L, Menauer F, Holzknecht N, et al. Sialolithiasis: MR sialography of the submandibular duct—an alternative to conventional sialography and US? *Radiology* 2000;216:665–71.
- 10 Wallace E, Tazuin M, Hagan I, et al. Management of giant sialoliths: review of the literature and preliminary experience with interventional sialendoscopy. *Laryngoscope* 2010;120:1974–8.
- 11 Eun YG, Chung DH, Kwon KH. Advantages of intraoral removal over submandibular gland resection for proximal submandibular stones: a prospective randomized study. *Laryngoscope* 2010;120:2189–92.

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