

## CASE REPORT

# A rare case of multiple minor salivary gland sialoliths in the masseteric region

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## Abstract

We report a case of a 59-year-old woman who presented with swelling of the right cheek. CT Scan showed three calculi located anteromedial to the masseter, and well isolated from other major salivary glands. Calculi were surgically removed with standard intraoral incision and exploration.

## KEYWORDS

calculi, Ghana, masseter, minor salivary gland, sialolithiasis

## 1 | INTRODUCTION

Sialolithiasis is the most common condition of the major salivary glands occurring mainly in the submandibular gland or its duct. In contrast with major salivary glands, sialolithiasis of the minor salivary glands is rare (less than 1%). We report a case of a 59-year-old woman who presented with swelling of the right cheek of a week's duration. CT scan showed three calculi located anteromedial to the masseter, and well isolated from other major salivary glands, while the parotid gland and its duct were free from lithiasis and inflammation. Two calculi were surgically removed with standard intraoral incision and exploration. Adequate clinical evaluation is important for acceptable diagnosis and treatment outcome.

The occurrence of calcified structures located in the parenchyma or ductal system of the salivary gland, referred to as sialolithiasis, is well described in the literature.<sup>1,2</sup> Sialolithiasis is the most common salivary gland disorder, characterized by the obstruction of the salivary secretion apparatus. With

a reported incidence of 1:10 000 to 1:30 000, the condition accounts for approximately 33% of salivary gland disorders.<sup>3</sup> It occurs mainly in the submandibular gland or its duct (80%-90%) and, to a lesser degree, in the parotid gland and the sublingual gland or their ducts (9%-20%).<sup>4,5</sup> In contrast with these, however, sialolithiasis of minor salivary glands is very rare (<1%).<sup>6,7</sup>

There have been some reports of minor salivary gland sialoliths in different locations, but none so far, to the best of our knowledge, describing multiple calculi formation in a minor salivary gland around the masseteric region.

## 2 | CASE REPORT

A 59-year-old Ghanaian woman presented to the Oral and Maxillofacial Surgery Unit of the Korle-Bu Teaching Hospital with recurrent swelling in the right cheek of 2 months duration, but with an acute exacerbation of 1 week. There was no

**Institution where study was conducted:** Oral and Maxillofacial Surgery Unit, Korle-Bu Teaching Hospital, Accra, Ghana

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difficulty in mouth opening, nor associated toothache. The patient's past medical history was unremarkable.

Examination revealed a swelling of about  $4 \times 3$  cm extending over the right posterolateral aspects of the mandible, with normal overlying skin. The swelling was soft in consistency, mildly tender, and nonfluctuant (Figure 1), with mild differential warmth.

Ultrasonography of right cheek region indicated an ovoid nodular lesion in masseter region measuring 1.14 cm with eccentric calcification suggestive of sialolithiasis. A follow-up CT scan further showed three well-circumscribed hyperdense lesions suggestive of calculi, the largest measuring  $5.6 \times 5.2$  mm, and demonstrated the reactive inflammation around the right masseteric region (Figures 2-4). The location of the calculi was well isolated from all major salivary glands. Furthermore, the ipsilateral parotid gland and its duct were free from lithiasis and inflammation. An impression of multiple minor salivary gland sialolithiasis was formed. Routine hematological investigations showed no obvious abnormality.

The patient underwent excision of the sialoliths via intraoral approach under local anesthesia. Two calculi were surgically removed through a standard intraoral incision; however, the smallest calculus was not found during the procedure (Figure 5). The patient was put on oral Cefuroxime and analgesics following the procedure, with oral hygiene instructions. She recovered uneventfully, confirmed at 1-week,

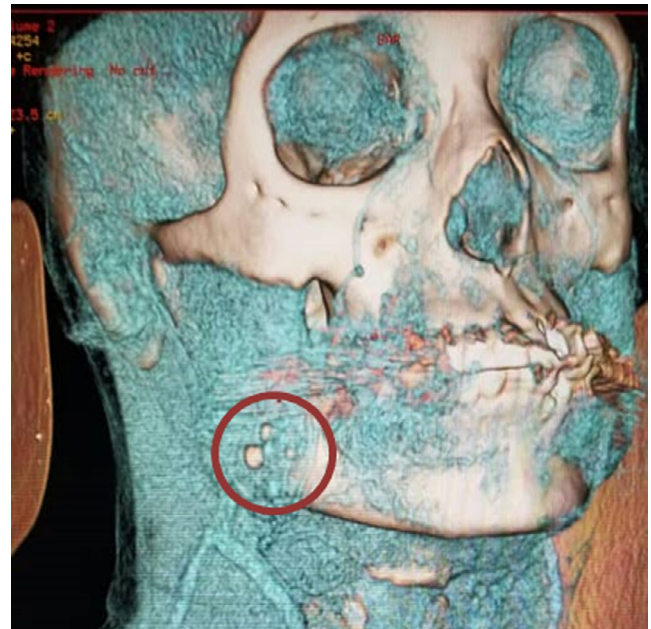
6-week, and 3-month reviews (Figures 6 and 7). Biochemical analysis of the stone revealed it to consist of carbonate apatite (Dahlite) 62%, amorphous calcium phosphate carbonate 28%, and calcium oxalate dihydrate (Weddellite) 20%.



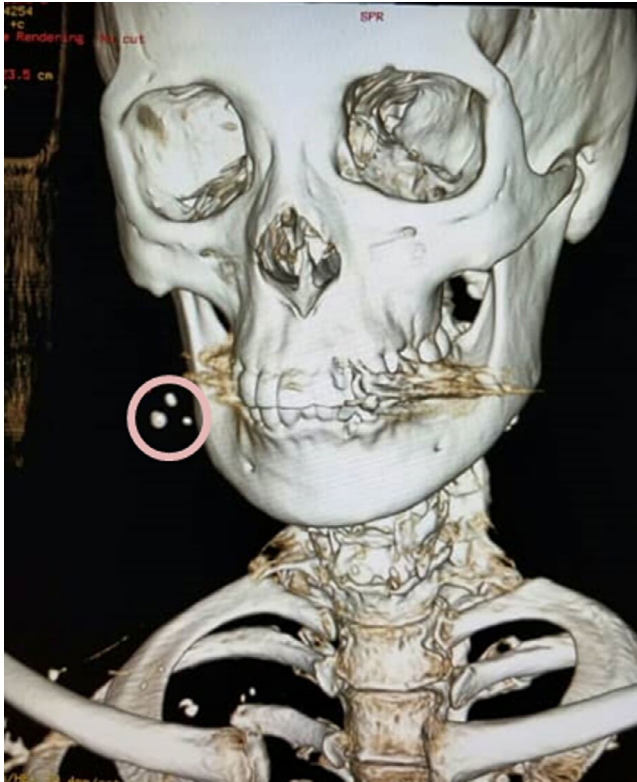
**FIGURE 2** Axial CT showing calculi



**FIGURE 1** Swelling on the right cheek



**FIGURE 3** Contrast CT showing calculi (red circle) in the lower masseter region



**FIGURE 4** 3D CT showing definite three calculi (white circle)



**FIGURE 5** Two calculi were surgically removed through a standard intraoral incision

### 3 | DISCUSSION

Despite being rare, there are several reports of sialolithiasis of the minor salivary glands. A study by Wang et al reviewing 17 cases of minor salivary calculi reported the most frequent sites to be the buccal mucosa (41.2%), upper lip



**FIGURE 6** The incision site had been healed without complication



**FIGURE 7** The swelling had been subsided

(29.4%), and lower lip (17.6%), respectively. In their series, they also had calculi occurring in the labial vestibule and retromolar spaces.<sup>7</sup> Kimura et al also reported a similar finding in the buccal mucosa,<sup>6</sup> while Souza et al reported three cases of sialolithiasis of the upper and lower lips.<sup>8</sup>

Generally, as for major salivary glands, lithogenesis is thought to be influenced first by factors such as dehydration,

reduced salivary flow rate, abnormalities in calcium metabolism, infections, altered solubility of crystalloids, neck radiotherapy, Sjogren's disease, and medications such as antihistamines and antipsychotics.<sup>9,10</sup> The pathogenesis of sialoliths is unclear, much so for minor salivary glands.

While Wang et al's patients ranged from 35 to 82 years, Anneroth and Hansen suggested calculi in the minor salivary glands to occur more in the 5th, 6th, or 7th decade.<sup>11</sup> Our patient, likewise, was in her sixth decade of life. Wang et al however reported it to be more common in males.

Though the occurrence of multiple stones in this current report makes it more peculiar, sialoliths of minor salivary glands are described to clinically appear as single, small, round, submucosal asymptomatic nodule, which is hard or firm and moves freely in the surrounding tissue.<sup>11</sup> The two calculi found in our patient had similar characteristics.

It is evident that depending on the location of a minor salivary gland, its presenting symptoms mimic other conditions. In a report of minor salivary gland sialolithiasis, clinicians had initially suspected fibroma, mucocele, and squamous cell carcinoma.<sup>7</sup> In this current report, for instance, there was inflamed soft tissues around the masseteric region which could have obscured the diagnosis in the absence of special imaging. Thorough examination and investigations are therefore required for appropriate clinical diagnosis and treatment planning.

Traditional imaging modalities, namely, conventional radiographs and sialograms, are more useful in detecting calculi in submandibular and parotid glands and the draining ducts.<sup>12</sup> Whereas plain radiographs can only detect radio-opaque stones which form about 70-90% of all calculi, sialograms depict both radiolucent and radio-opaque calculi as round filling defects within dilated ducts.<sup>13</sup> Minor salivary glands however do not have large prominent ducts as with the submandibular and parotid glands, and hence are better demonstrated with modalities allowing for good soft tissue differentiation such as ultrasonography, computed tomography (CT) scanning, and magnetic resonance imaging (MRI). These 3 modalities are also able to demonstrate most, if not all types of calculi, and the soft tissues within which they are located. CT and MRI scanning can also readily demonstrate the presence of associated inflammation. It is worth noting that whereas CT is inferior to MRI in demonstrating soft tissues such as minor salivary glands, CT is superior to MRI in the demonstration of calculi and calcifications.<sup>14</sup>

## 4 | CONCLUSION

Sialolithiasis of the minor salivary gland is an uncommon clinical finding which when present could be masked by its clinical presentation. Adequate clinical evaluation is important for acceptable diagnosis and treatment outcome.

## CONFLICT OF INTEREST

The authors declare that there are no conflicts of interest.

## AUTHOR CONTRIBUTIONS

MOB and EKB: contributed to conceptualization; SC and PKB: contributed to writing—original draft preparation; GEP, MOB, and EKB: contributed to writing—review and editing.

## ETHICAL APPROVAL

Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

## DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available from the corresponding author upon reasonable request.

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