

CASE REPORT

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# The key role of magnetic resonance sialography in the differential diagnosis between sialolith and phlebolith of the floor of the mouth

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## Abstract

**Background** The confusion between a phlebolith and a sialolith in the floor of the mouth can pose serious surgical risks, making it essential to define appropriate complementary imaging studies.

**Case presentation** We present the case of a patient with right submandibular sialolithiasis suspected by computed tomography. A previous attempt at extraction in another centre, under local anesthesia, was interrupted due to profuse bleeding, preventing the localization and removal of the calculi. Due to our pre-surgical study protocol using magnetic resonance sialography (MR-Si), a low-flow vascular malformation with internal phleboliths was revealed, without involvement of the salivary duct. As a result, the planned sialendoscopy was cancelled, and a follow-up approach was chosen given the asymptomatic nature of the condition.

**Conclusions** This case underscores the importance of standardizing radiological studies for salivary gland and floor-of-mouth pathology, highlighting MR-Si for its high sensitivity and specificity. Therefore, MR-Si enables more precise surgical planning and helps prevent unnecessary intraoperative complications.

**Keywords** Sialolith, Sialolithiasis, Phlebolith, Floor of the mouth, Magnetic resonance sialography, Low-flow vascular malformation, Sialendoscopy, Sialadenitis

## Background

Sialolithiasis is one of the most common causes of chronic obstructive sialadenitis (COS) [1], which may present as recurrent inflammation of the salivary gland or be an incidental finding in imaging studies. Its prevalence in postmortem studies is estimated at 0.115% [2].

The presence of phleboliths in the head and neck region is well-documented in the literature [3–6], predominantly in pediatric patients [7, 8]. Previous reports have described head and neck lesions that may contain internal phleboliths, mimicking sialolithiasis, such as submandibular hemangioma [3] or low-flow vascular

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malformations (LFVM). LFVMs typically present as soft masses with a tendency toward inflammation or thrombosis [9], with the potential to develop phleboliths. The inflammatory nature of LFVMs, sometimes exacerbated by meals, combined with imaging findings suggestive of sialolithiasis, often leads to a misdiagnosis of COS.

The diagnosis of phleboliths is primarily radiological, based on their intrinsic characteristics and their relationship with the vascular lesion. Generally, phleboliths appear as circular radiopacities, whereas sialoliths tend to have an elongated shape due to their location within the salivary duct [10]. Magnetic resonance (MR) imaging allows visualization of the dilated vessels characteristic of vascular malformations, enabling the assessment of LFVMs. Computed tomography (CT) excels in identifying calcified concretions, though it may not always clearly distinguish between vascular and salivary tissue. Doppler ultrasound is useful for detecting increased blood flow in vascular lesions, while sialography remains the most precise method for differentiating between these entities. Sialoliths appear as filling defects within the duct, whereas phleboliths are located outside the ductal system. However, conventional sialography is becoming obsolete, being replaced by MR sialography (MR-Si), which is a highly sensitive and specific imaging technique for assessing salivary duct pathology [1]. Therefore, for an accurate diagnosis, it is crucial to combine appropriate radiological evaluation with clinical findings.

Based on this case, which involved two phleboliths within a submandibular LFVM, we aim to highlight the risk of misdiagnosis and the importance of MR-Si in the differential diagnosis between phlebolith and sialolith.

### Case presentation

A 64-year-old female patient was referred for suspected right submandibular sialolithiasis, based on a prior non-contrast CT scan that revealed two calculi located in

the ductal and hilar regions, with no associated lesions (Fig. 1a and b). The patient had experienced two episodes of floor-of-mouth inflammation over the past 10 years, initiating the diagnostic process after the most recent episode two years ago. There was no previous history of vascular malformations. Additionally, she reported a previous attempt at stone removal at another centre, under local anesthesia, which was interrupted due to profuse bleeding, preventing continuation of the procedure.

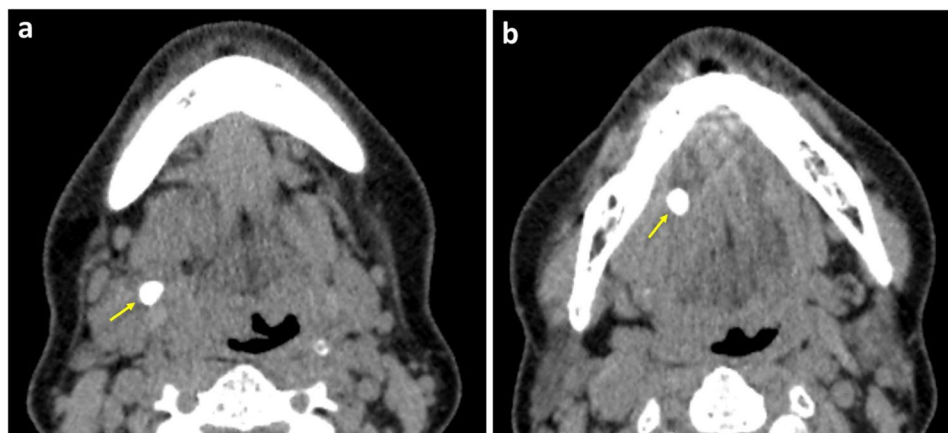
On physical examination, the floor of the mouth was soft, and both calculi were easily palpable. The glandular parenchyma was not swollen, and the ductal papilla was patent and permeable. Following the protocol of our salivary gland unit, an MR-Si was requested prior to performing a scheduled sialendoscopy. The MR-Si showed that the major salivary glands maintained their normal morphology and signal, with no evidence of dilation of the excretory ducts or any relationship or impact of the calcified concretions on the salivary system (Fig. 2a). Additionally, a T2-hyperintense lesion with small (<5 mm) hypointense foci was identified, suggestive of phleboliths within a LFVM (Fig. 2b).

Given these findings, the scheduled sialendoscopy was cancelled, and a conservative approach was adopted. The patient remains asymptomatic to date.

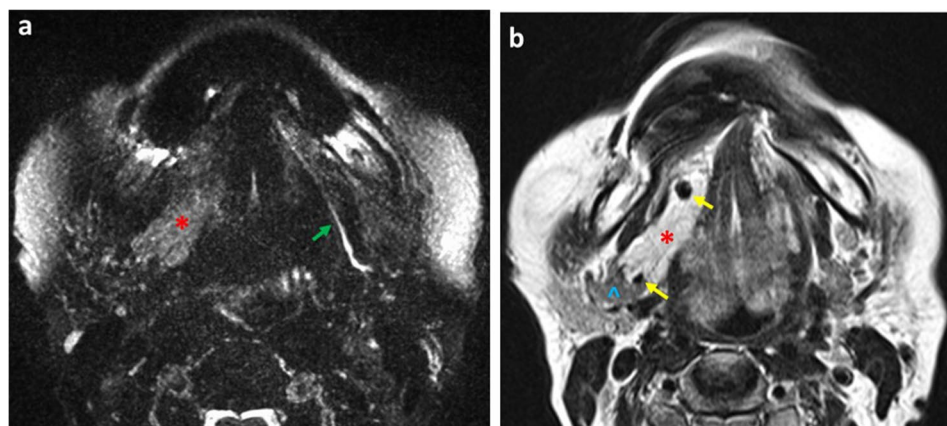
### Discussion

The confusion between phlebolith and sialolith is possible, frequent, and may lead to erroneous and potentially harmful decisions for patients. Therefore, diagnostic protocols must be established to enable their differential diagnosis.

The literature has demonstrated that specific radiological features, such as the presence of hypointense areas within the lesion on T2-weighted MR sequences, are highly suggestive of a venous malformation [11]. In contrast, CT imaging can be misleading, failing to detect



**Fig. 1** Non-contrast axial CT images reveal a sialolith (arrow) in the hilum of the submandibular gland (a) and another suspected lithiasis on the right side of the floor of the mouth, following the presumed course of Wharton's duct (b). No retrograde dilatation of Wharton's duct is observed



**Fig. 2** MR sialography image (a) and T2-weighted MR image (b) show an increased signal intensity lesion on the right side of the floor of the mouth (\*), suggestive of a low-flow vascular malformation, containing two phleboliths (yellow arrows). The right submandibular gland is posteriorly displaced by the lesion (^). Note the normal appearance of Wharton's duct on the left side (green arrow) and the absence of visualization of the duct on the right side

the presence of an LFVM and mistaking a phlebolith for a sialolith. This is particularly significant given that CT remains the standard diagnostic tool for salivary gland pathology in most centres.

Additionally, sialendoscopy may serve as the gold standard for differential diagnosis by confirming, *in situ*, the absence of lithiasis in the ductal salivary system. However, with the advancement of MR, combining MR sequences with sialography protocol, the need for a minimally invasive procedure such as sialendoscopy, may be reduced. MR-Si is now considered the optimal imaging modality before performing sialendoscopy, given its high sensitivity and specificity for detecting lithiasis, as well as its accuracy in identifying strictures [1], which are the primary obstructive factors causing COS. MR-Si allows visualization of the ductal system, including its tertiary branches, and assessment of parenchymal tissue [12], without requiring radiation or intravenous contrast. Consequently, it enables simultaneous evaluation of both the glandular parenchyma and the ductal system. Due to these advantages, MR-Si is a highly reliable diagnostic tool and should be implemented in dedicated salivary gland pathology units.

Despite these advantages, MR-Si has practical limitations (higher cost, limited availability) and contraindications such as claustrophobia or metallic implants. In such situations, alternative non-invasive or minimally invasive tools, such as color Doppler ultrasound or cone-beam CT, can provide valuable diagnostic information [13, 14].

The treatment of LFVMs depends on their size, location, and clinical manifestations. In cases of small and asymptomatic malformations, observation may be sufficient, whereas in cases of significant symptoms or progressive growth, sclerotherapy has shown favourable outcomes [9].

This case underscores the importance of a thorough patient history, a comprehensive physical examination,

and the appropriate selection of complementary imaging tests. These steps enable the differentiation between sialolithiasis and other conditions affecting the salivary glands, the submandibular space, or the floor of the mouth. Although ductal obstruction was initially suspected, MR-Si facilitated the diagnosis of a vascular malformation, thereby avoiding an unnecessary invasive procedure—which had already been attempted at another centre under local anesthesia without success, posing significant haemorrhagic and life-threatening risks.

## Conclusion

LFVMs can occur in the floor of the mouth and may contain phleboliths, mimicking sialoliths. The confusion between these entities can lead to erroneous and potentially harmful clinical decisions for patients.

MR-Si is confirmed as the most appropriate imaging modality to distinguish between these two conditions and prevent unnecessary surgical procedures. Therefore, the implementation of MR-Si in salivary gland pathology units is essential.

## Abbreviations

|       |                                  |
|-------|----------------------------------|
| COS   | Chronic Obstructive Sialadenitis |
| CT    | Computed Tomography              |
| LFVM  | Low-Flow Vascular Malformation   |
| MR    | Magnetic Resonance               |
| MR-Si | MRSialography                    |

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## Authors' contributions

ASB, GDT, FGG, ESA and JMVA have made substantial contributions to the conception and design of the work. ASB, FGG, ESA have made substantial

contributions to acquisition, analysis, and interpretation of data. ASB, GDT, ESA have drafted the work. All authors have revised and approved the submitted version and have agreed both to be personally accountable for the author's own contributions and to ensure that questions related to the accuracy or integrity of any part of the work, even ones in which the author was not personally involved, are appropriately investigated, resolved, and the resolution documented in the literature.

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#### Data availability

No datasets were generated or analysed during the current study.

#### Declarations

#### Ethics approval and consent to participate

Not applicable.

#### Consent for publication

The patient provided explicit written informed consent for the publication of her clinical details and radiological images in this study.

#### Competing interests

The authors declare no competing interests.

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