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Submandibular gland obstruction caused by phlebolith masquerading as sialolith

KEYWORDS
Obstruction;
Phlebolith;
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Generally, salivary gland obstructions occur by sialolith, stricture, or mucous plug.[1,2] Because the obstruction caused by venous malformation (VM)/cavernous hemangioma with phleboliths is rarer,[2,3] phleboliths may be misdiagnosed as sialoliths. We reported a rare submandibular gland (SMG) obstruction caused by phlebolith masquerading as sialolith.

A 38-year-old man with swelling and pain of the left SMG during eating was referred to our hospital. Non-contrast-enhanced computed tomography (CECT) showed a large sialolith (11.5 × 10.5 × 9.2 mm) and small sialolith (4.0 × 3.5 × 3.1 mm) in the left SMG, and there were multiple calcifications around the SMG (Fig. 1A–I). The lesion with multiple calcifications was found extending from the submandibular space to the parapharyngeal space. Clinical and radiological diagnosis was SMG sialoliths and submandibular VM with multiple phleboliths. Two weeks after the first visit, the patient underwent intraoral removal of SMG with 2 sialoliths under general anesthesia. A 3-cm mucosal incision was made, and Wharton's duct and lingual nerve were identified. The submandibular ganglion and Wharton's duct were ligated and cut, and the lingual nerve was preserved. Soft tissues surrounding the SMG were dissected bluntly, and the SMG was gripped with forceps and pulled up through the incision. After the facial artery was ligated and cut, the SMG was removed completely in an intraoral approach. A 30°, 4-mm diameter, endoscope with tissue retractor (Karl Storz, Tuttlingen, Germany) was used for ligation of the facial artery and detecting residual gland and hemostasis in the submandibular space after SMG removal. Because the large sialolith was not found in the removed SMG, the large stone remaining in the surgical field was diagnosed as the phlebolith in VM by the palpation and endoscopic view, and the phlebolith remained for the risk of hemorrhage.

VMs/cavernous hemangiomas sometimes occur in the submandibular region and may include phleboliths, but they in the SMG are extremely rare.[3,4] Phleboliths within VMs/cavernous hemangiomas in or adjacent to the SMG may be misdiagnosed as sialoliths.[1–5] Phleboliths are usually multiple and circular radiopacities with a laminated morphology and a radiopaque or radiolucent center, while sialoliths are commonly uniform radiopaque and there is more often a single sialolith rather than multiple sialoliths.[2] CECT, gadolinium-enhanced magnetic resonance imaging, and ultrasonography are useful for the diagnosis of VMs. But, non-CECT is commonly performed, when sialolithiasis is clinically suspected. In the present case, 2 sialoliths in SMG and multiple phleboliths in VM were diagnosed by non-CECT. As a result, a large calcification was not sialolith but phlebolith, and it was thought that the increase of the phleboliths had compressed the SMG, causing obstruction. Although the VM with multiple phleboliths could be found in non-CECT images, the anatomical relationship between the SMG and VM should have been confirmed by CECT. But, McMenamin et al.[3] and Chuang et al.[5] misdiagnosed phleboliths as sialoliths on CECT images and performed SMG removal, because painful swelling of the SMG during eating was suspicious of sialoliths. Phleboliths should be included in the differential diagnosis of SMG obstruction, because

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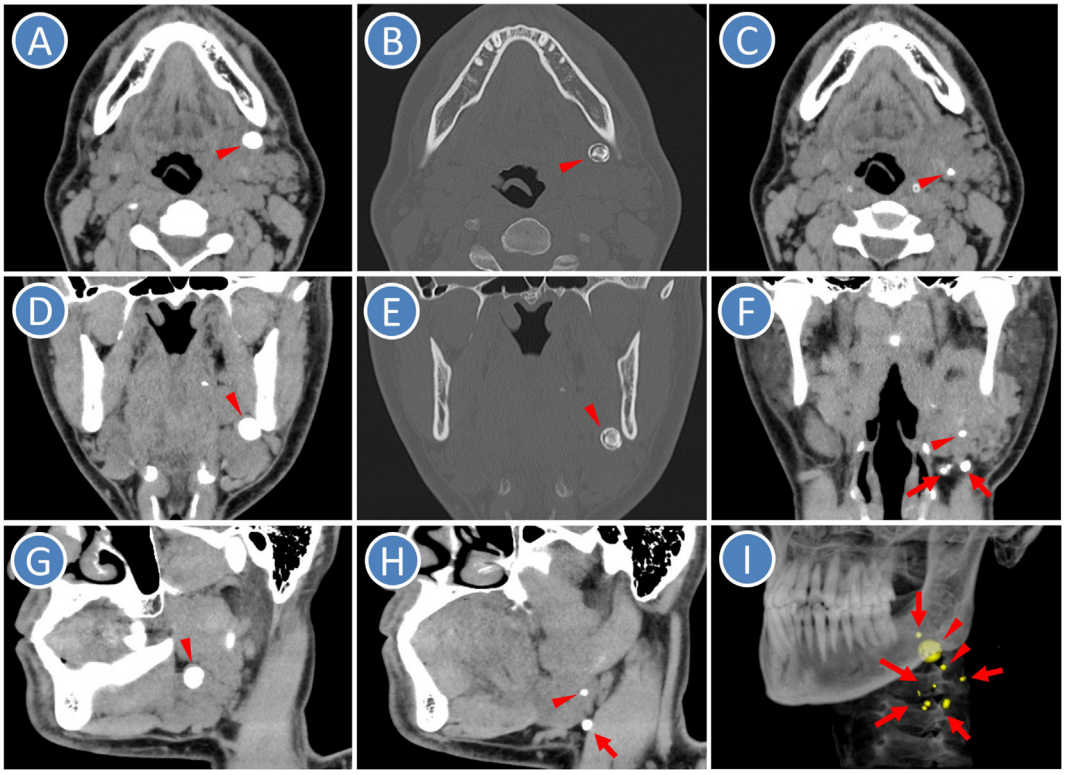

Figure 1 Computed tomography (CT) images.
(A–C) Axial images. (D–F) Coronal images. (G, H) Oblique sagittal images. (I) Three-dimensional image. CT showed a large sialolith (11.5 × 10.5 × 9.2 mm) and small sialolith (4.0 × 3.5 × 3.1 mm) in the left submandibular gland, and there were multiple calcifications around the submandibular gland. Arrowheads indicate sialoliths in the submandibular gland. Arrows indicate calcifications around the submandibular gland.

the differentiation may be challenging in the case of VMs/cavernous hemangiomas including phleboliths in or adjacent to the SMG.

Declaration of competing interest

The authors have no conflicts of interest relevant to this article.

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None.

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