

Hemangioma with Phleboliths in the Sublingual Gland: As a Cause of Submental Opacity

HAKAN, ÖZKAN ÜNAL,¹ SERDAR UGRAS,² KOKSAL YUCA and MUZAFFER KIRIŞ

Department of Otorhinolaryngology, ¹Department of Radiology, and ²Department of Pathology, Medical Faculty, Yüzüncü Yıl University, Van, Turkey

ÇANKAYA, H., ÜNAL, Ö., UGRAS, S., YUCA, K. and KIRIŞ, M. *Hemangioma with Phleboliths in the Sublingual Gland: As a Cause of Submental Opacity.* Tohoku J. Exp. Med., 2003, **199** (3), 187-191 — Hemangiomas are the most common lesions of the major salivary glands during infancy and early childhood. Changes in blood flow dynamic within hemangioma results in thrombus and phleboliths. There have been a number of reports of hemangiomas with phleboliths in parotid and submandibular glands. We present the first case of a hemangioma with multiple phleboliths in the sublingual gland as a cause of submental opacity, and discussed the diagnosis of radiopaque masses in the sublingual and submental regions———sublingual gland; phlebolith; sublingual mass; hemangioma

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Hemangiomas are the most common lesions of the major salivary glands during infancy and early childhood. The parotid gland was the anatomic site for 85% of hemangiomas encountered in major salivary glands (McMenamin et al. 1997). Changes in blood flow dynamic within hemangioma result in thrombus and phleboliths (Li 1990). Phlebolith formation reported as a characteristic feature of hemangioma was first described in the splenic vein (Dempsey and Murley 1970). Although hemangioma is a relatively common lesion in the head and neck region it is rarely associated with phlebolith in the region. There have been a number of reports of hemangioma with

phlebolith in buccal mucosa, the parotid and submandibular glands, the preauricular area, and mentalis muscle (Dempsey and Murley 1970; Ingalls et al. 1985; Sano et al. 1988; Li 1990; McMenamin et al. 1997). But there is no report of hemangioma with phlebolith in the sublingual gland in English literature.

We present the first case of a hemangioma with multiple phleboliths in the sublingual gland as a cause of submental opacity and discussed the diagnosis of radiopaque masses in the sublingual and submental regions.

CASE REPORT

An 18-year-old male patient was admitted

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Address for reprints: Dr. Hakan Çankaya, Yüzüncü Yıl Üniversitesi, Tıp Fakültesi, Kulak Burun Bogaz Anabilim Dalı, 65100 Van/Türkiye.

e-mail: hcankaya2@hotmail.com

to the Department of Otorhinolaryngology for examination of swelling of his right submental region. The swelling had been present for 8 years and had gradually increased in size.

Clinical examination showed a painless and hard swelling of the right submental region. Saliva discharging from the right submandibular duct was normal. An ultrasonography of the submandibular area revealed many opac-

ities in the swollen sublingual gland and a diagnosis of sublingual sialolithiasis was reached (Fig. 1). Computed tomography (CT) showed several round calcified foci (Fig. 2) and, magnetic resonance imaging (MRI) revealed a 25×15 mm lesion in the right submental region (Fig. 3). The lesion had high intensity on T2-weighted images and relatively lower intensity on T1-weighted images. Calcifications were

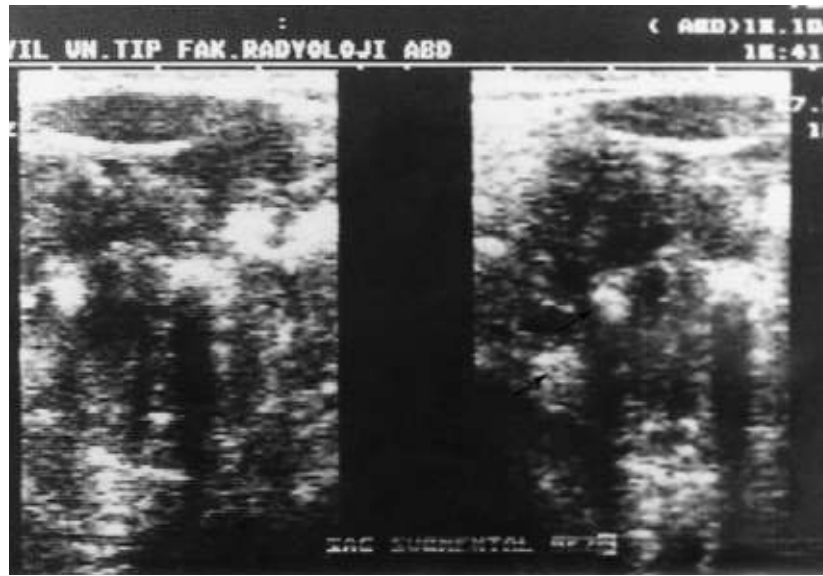


Fig. 1. Multiple calcified foci on ultrasonography. (arrow pointed calcified foci).

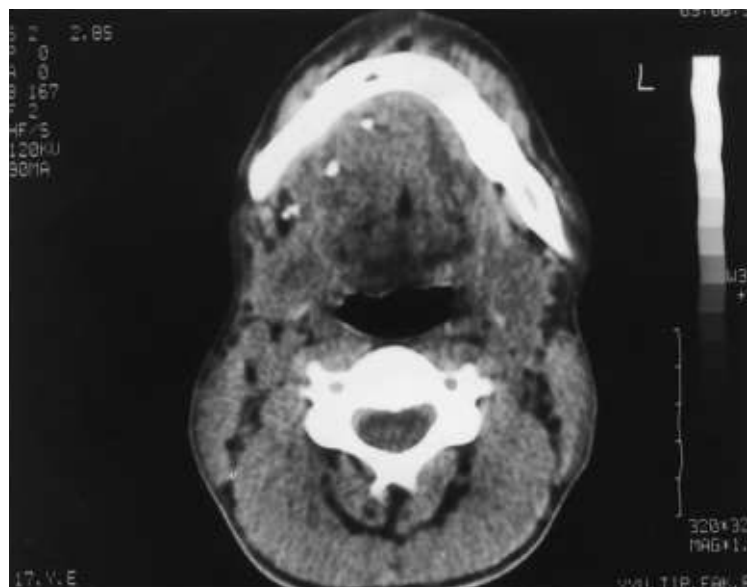


Fig. 2. Axial non-contrast CT shows the mass and calcification.

not demonstrated on MRI. Excisional surgery was performed under general anesthesia and several hard bodies within the mass were observed. Blood transfusion was not needed and no complications occurred.

Microscopically, hemangioma was observed in the mucous glands of the sublingual gland (Fig. 4). Additionally, phleboliths and thrombus were also observed in hemangioma (Fig. 5).

The lesion was diagnosed as hemangioma with phleboliths in the sublingual gland.

DISCUSSION

The most common causes of masses in the sublingual or submental regions are Ludwig's angina, congenital sublingual dermoid cyst, amiloidosis, ranula, branchial cleft cyst, thyroglossal duct cyst, and cystic hygroma (Ichimura

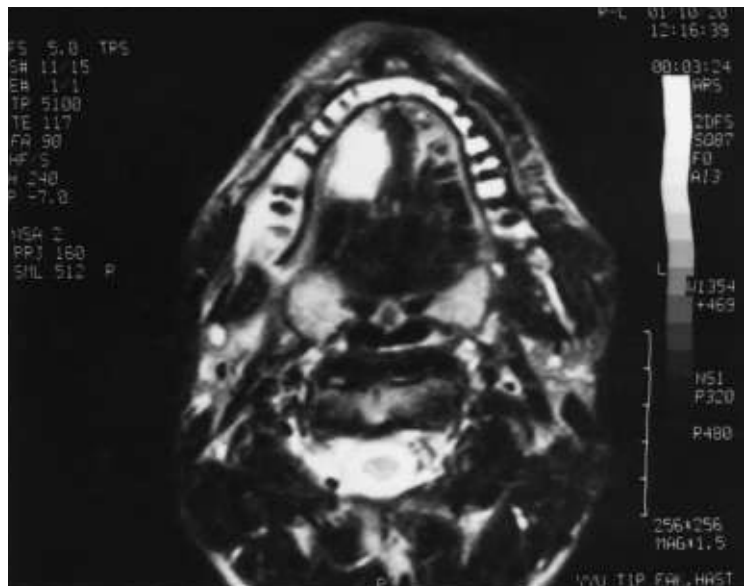


Fig. 3. T₂-weighted MR image shows high intensity mass in right submental area.

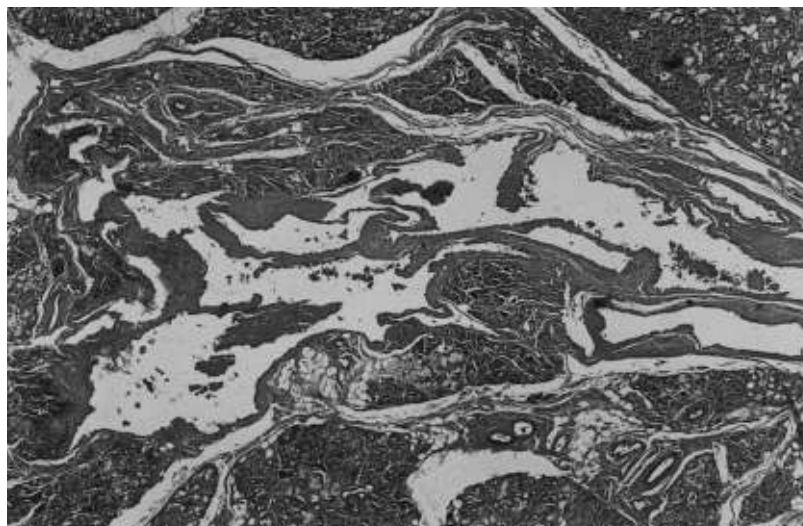


Fig. 4. Microscopically, specimen demonstrates hemangioma in mucous glands of the sublingual gland (H&E, $\times 10$).

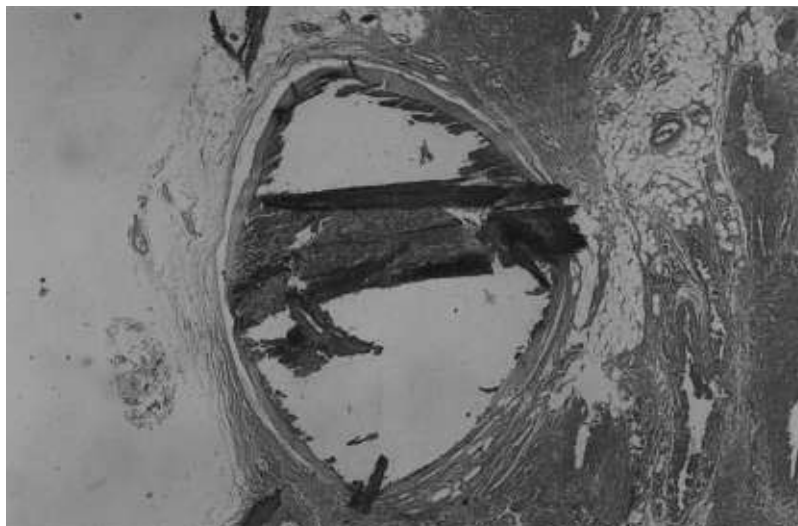


Fig. 5. Specimen demonstrates phlebolith in hemangioma (H & E, $\times 4$)

et al. 1996; Jonsson et al. 1998; Gossett et al. 1999; Obiechina et al. 1999). If an opacity is present in areas of major salivary glands, sialolithiasis should be first considered. Cases of sublingual gland calculi are very rare and it is possible not to encounter one even after examining a great number of cases of salivary gland calculi (Lustmann et al. 1990; Zenk et al. 1999).

Phleboliths imaged as opacities in radiological examinations are among the characteristic properties of cavernous hemangioma with phleboliths. There have been a number of reports of hemangioma with phlebolith of the parotid and submandibular glands. Additionally, Branch et al. (1981) reported a case that had sublingual hemangioma but not phlebolith. Our report is a first case of hemangioma with phleboliths in the sublingual gland.

Diagnosis of benign tumors of mesenchymal origin such as hemangioma that involve major salivary glands is difficult (McMenamin et al. 1997). If only based on clinical features and history, the preoperative assessment may suggest a benign epithelial tumor of the salivary gland or sialolithiasis. Radiological assessment is necessary.

In plain radiographies, calcified structures

such as phleboliths will be seen as opacities. However, plain radiographies are reported to be not always helpful in the differentiation of sialolithiasis and phleboliths such as in the case reported by Dempsey and Murley (1970).

Ultrasonography and CT are generally able to distinguish intrinsic from extrinsic masses of salivary glands. CT is also of little use in evaluating generalized paranchymal disease or ductal architecture (Rice 1998). In MRI, high intensity has been reported on T2-weighted images, and isointensity with muscle has been described with T1-weighted images. Enlarged vessels may be seen as signal voids within and around lesions. In the case reported by McMenamin et al. (1997), panoramic radiograph showed multiple calcific foci in the region of the right submandibular gland, and by CT the case was diagnosed as chronic sialadenitis associated with salivary calculi because it was intraglandularly localized. Nevertheless, the result of its pathology was hemangioma with phleboliths. In our case, hemangioma was localized in the sublingual gland. The lesion was seen clearly on T2-weighted images. But neither MRI nor CT was sufficient for diagnosis as in the case of McMenamin and therefore diagnosis has not truly been established.

Sialography can easily differentiate a phlebolith from sialolithiasis as it can show localization of the calcification within salivary ducts (McMenamin et al. 1997). Unfortunately, in either the case of McMenamin or in our case sialography was not performed.

Finally, hemangioma with phleboliths should be considered in differential diagnosis of calcified lesions of the submental area and, sialography is still an important investigation method in the evaluation of radiopaque lesions localized intraglandularly in the submental and sublingual areas.

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