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Case Report Korean J Otorhinolaryngol-Head Neck Surg 2015;58(10):699-703 / pISSN 2092-5859 / eISSN 2092-6529
http://dx.doi.org/10.3342/kjorl-hns.2015.58.10.699

A Case of Cavernous Hemangioma in the Submandibular Gland: A Review of Clinicoradiologic Features and Treatment Methods

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Received October 31, 2014
Revised January 20, 2015
Accepted January 21, 2015
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Cavernous hemangiomas in the submandibular gland (SMG) are rare; therefore, there are few clinicoradiologic pathognomonic findings upon which to base a diagnosis. Thus, a preoperative diagnosis of hemangioma is rare. We reviewed all English language literature on cavernous hemangioma involving SMG published after 1990. The characteristic clinical finding of cavernous hemangiomas involving the SMG is a fluctuant swelling that is unrelated to food intake. On computed tomography, cavernous hemangiomas appear as cystic or enhancing lesions either with or without calcification; however, this is not a pathognomonic finding for diagnosis. Most cavernous hemangiomas are best treated by complete surgical excision; preoperative angiography or embolization is not always required. Cavernous hemangioma should be added to the list of the masses presented a fluctuant swelling that is not associated with postprandial swelling. The treatment of choice is surgical excision without angiography and embolization.
Korean J Otorhinolaryngol-Head Neck Surg 2015;58(10):699-703

Key Words Angiography · Cavernous hemangioma · Embolization · Submandibular gland.

Introduction

Hemangiomas are the most common tumors found in the major salivary glands during infancy and early childhood.[1)] However, they rarely occur in the major salivary glands of adults, and submandibular involvement is particularly rare.[2)] Because of their anatomic location, the symptoms of submandibular hemangioma often resemble those of submandibular sialadenitis. In addition, a radiologist may often misdiagnose hemangiomas in the submandibular gland as mucoceles or sialadenitis with calculi.[3)]

Due to the rarity of hemangiomas involving the submandibular gland and their unusual presentation, inaccurate preoperative diagnosis and inappropriate treatment plans are common. Here, we present an unusual case of cavernous hemangioma involving the submandibular gland along with a review of the literature regarding clinicoradiologic characteristics and treatment methods.

Case

A 34-year-old woman presented to the otolaryngology clinic complaining of a painless swelling in the right submandibular area. She first became aware of the swelling 1 month earlier, and it had since grown gradually. The gland did not increase in size during meals. Clinical examination revealed a firm non-

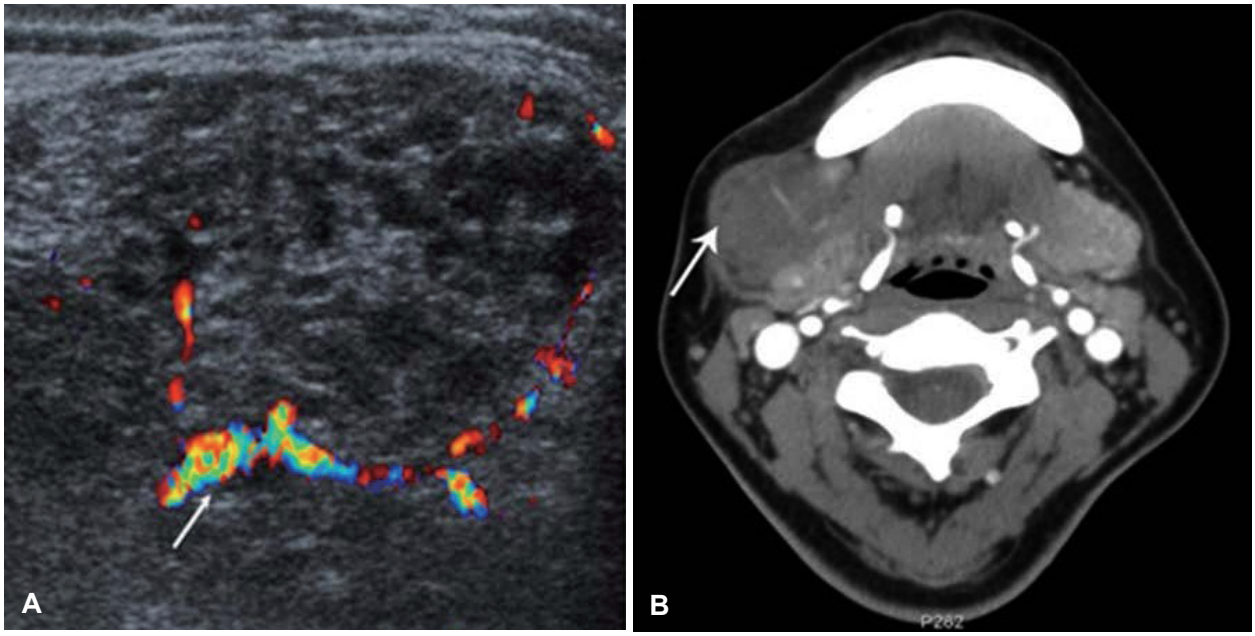

Fig. 1. Doppler ultrasonography showing a well-circumscribed hypoechoic heterogeneous mass (arrow) with color flow in the right submandibular gland (A). Axial contrast-enhanced CT scan showing a minimally enhanced, enlarged submandibular gland (arrow) (B).

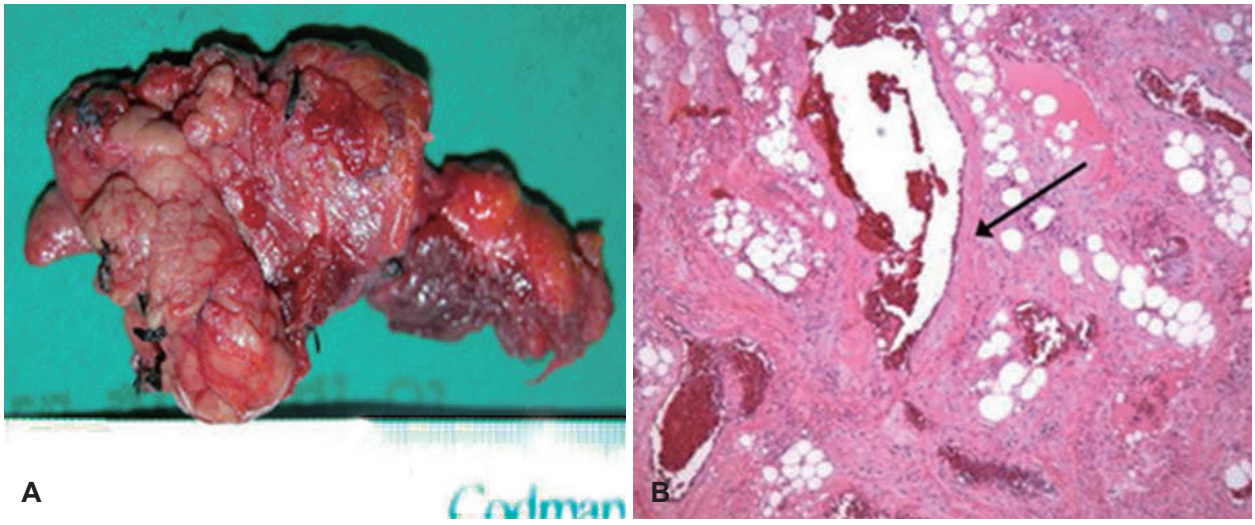

Fig. 2. An excised specimen shows high vascularity at the surface (A). A microscopic view of a cavernous hemangioma reveals a dilated, thin-walled vascular space filled with blood and lined by endothelial cells (arrow) (hematoxylin and eosin stain, ×200) (B).

tender swelling measuring about 4×4 cm in the region of the right submandibular gland. There was no bluish coloration of the overlying skin or any dilated blood vessels. There was no recent history of oropharyngeal (or other head and neck) infection. Plain radiographs showed no evidence of calcifications.

Ultrasonography showed enlargement of the right submandibular gland, with a hypoechoic lesion measuring about 4×3.5 cm within the gland (Fig. 1). A computed tomography (CT) scan revealed an enlarged submandibular gland with minimal enhancement (Fig. 1). The patient underwent fine-needle aspiration, but the cytological results were not diagnostic as the specimen contained too many red blood cells. Our clinical, radiologic, and pathological impression was that of a benign epithelial salivary gland tumor. At surgery, an ill-defined vascular lesion was identified within and surrounding the submandibular gland. No unusual bleeding was encountered during surgery and the patient was discharged from hospital on the sixth post-operative day after an uneventful recovery.

Pathological examination of the specimen revealed that the right submandibular gland was attached to a blood-filled spongy vascular lesion (Fig. 2). Microscopic examination revealed large, dilated, blood-filled vessels lined by flattened en-

dothelium (Fig. 2). The vessels were arranged in a lobular configuration. No thrombosis or fibrosis was detected. A diagnosis of cavernous hemangioma was made. At the 12-month follow-up, the functional and cosmetic results were excellent and there was no recurrence.

Discussion

Hemangiomas involving the major salivary glands are uncommon in adults, and those that do occur are more common in the parotid gland (87.5%) than in the submandibular gland (12%).[1] According to a recent report, only four out of 230 patients who underwent submandibular gland excision had a submandibular gland hemangioma.[4]

Hemangiomas are classified as either capillary or cavernous. The cavernous type is characterized by dilated, thin-walled, vascular spaces that are filled with blood and lined by markedly flattened endothelial cells. The vessels may be arranged in a roughly lobular arrangement or in a diffuse haphazard pattern.

The walls are occasionally thickened as a result of adventitial fibrosis, and inflammatory cells may be scattered throughout the stroma.[5] Consistent with previous reports, our patient's pathologic report showed that the vessels revealed large, dilated, blood-filled vessels lined by flattened endothelium with a lobular configuration.

Some 90% of all hemangiomas arise during the first three decades of life, and the average age of a patient with a hemangioma is 10 years;[5,6] however, cavernous hemangiomas, as seen in this case, typically occur in older children and adults.[5] Nagao, et al.[7] examined a series of 20 patients with cavernous hemangioma of the parotid gland, and found that the mean age was 26 years (range: 4 months−50 years). The age of our patient was consistent with this finding. We reviewed all English language papers on cavernous hemangioma involving the submandibular gland published after 1990. We found that the average age of a patient with a cavernous hemangioma involving the submandibular gland was 39 years (range: 16−65 years) (Table 1). Hemangiomas in the submandibular gland occur later in life than those in the parotid gland, and are twice as common in females than in males because they are influenced by circulating hormones.[4] The sex of our patient was consistent with this findings. We also confirmed that hemangiomas tend to occur more frequently in the submandiblar glands of females than males (female to male ratio=3:1) (Table 1).

Clinical examination and radiologic investigations are important to confirm a diagnosis. However, hemangiomas of the

Table 1. Clinical and radiologic features of submandibular cavernous hemangiomas

Author	Sex/age (years)	Fluctuant swelling	Pain or tenderness	Postprandial swelling	Skin color change	CT	MRI	Sonography	FNA
This study	F/34	No	No	No	No	Enhancement			Not diagnostic
Cho, et al.[3]	F/41	Yes	No	No	No				Not diagnostic
	F/16	Yes	No	No	No				Not diagnostic
	F/49	Yes	No	No	No				Not diagnostic
	M/44	Yes	Yes	No	No				Not diagnostic
	M/57	Yes	No	No	No				Not diagnostic
El-Hakim and El-Khashab[2]	F/35	No	Yes	No	No			Hypoechoic	
McMenamin, et al.[5]	F/37	Yes	Yes	Yes	No	Enhancement and calcification			
Chuang, et al.[1]	M/65	Yes	Yes	No	No	Calcification			
Ozturk, et al.[10]	M/27	Yes	No	No	No	Cystic mass	T1-hypointense	Anechoic	Not diagnostic
Kumar, et al.[4]	M/20		Yes	No	No	Enhancement			
	F/35		Yes	No	No	Enhancement			
	F/47		Yes	No	No	Enhancement			
	F/37		Yes	No	No	Enhancement			

CT: computed tomography, FNA: fine needle aspiration, MRI: magnetic resonance imaging

submandibular gland often present diagnostic difficulties because there are no diagnostic clues. To the best of our knowledge, no study has investigated the clinical characteristics of hemangiomas in the submandibular gland. According to our literature review, the majority of patients with submandibular gland hemangiomas (80%) complain of fluctuant swelling in the submandibular triangle, although the swelling is not related to food intake. Approximately half of patients (57%) complain of pain in the submandibular triangle, but changes in skin color are not evident in all cases.

Therefore, hemangioma should not be excluded when the overlying skin does not appear bluish.

Several groups have attempted to define the radiologic characteristics associated with cavernous hemangioma involving the submandibular gland. Odabasi, et al.[8] reported that magnetic resonance imaging can show focal heterogeneities, which represent areas of thrombosis, fibrosis, or calcification. Others suggest that the presence of a color Doppler signal in a hypoechoic mass with a heterogeneous Echo texture on sonography suggests the presence of a hemangioma.[9] Cho, et al.[3] suggested that calcified nodules on CT images are pathognomonic for a diagnosis of cavernous hemangioma.

On the other hand, other reports suggest that radiologic imaging studies may lead to suspicion of the anomaly, but are not diagnostic because they do not show definitive evidence; however, they are necessary to exclude other pathologies.[10] Our patient's finding and literature review suggest that submandibular gland hemangioma appears as a cystic or enhancing lesion either with or without calcification (Table 1). Calcification is observed in 50% of cases. Calcification occurs subsequent to thrombus and phlebolith formation due to changes in blood flow dynamics within the hemangioma.[1] Differential diagnoses of cystic or enhancing lesions with or without calcification in the submandibular triangle must rule out sialadenitis with sialoliths and other benign or malignant tumors. Occasionally, the distinction between a plunging ranula in the submandibular space, with little or no visible sublingual component, and a second branchial cleft cyst may be problematic. Careful evaluation of the location of the cystic mass in relation to the submandibular gland may be helpful in making this distinction. Sialadenitis with sialoliths can lead to submandibular gland swelling related to food intake, whereas other benign or malignant tumors (such as pleomorphic adenoma) do not present with fluctuant swelling. As mentioned earlier, the swelling of cavernous hemangiomas is not associated with food intake and tends to be fluctuant.

Although fine-needle aspiration cytology was performed in 50% of patients including this case, none were diagnosed by fine-needle aspiration cytology before surgery (Table 1). Thus, fine-needle aspiration cytology is not helpful for the diagnosis of cavernous hemangioma.

Because cavernous hemangiomas tend to be larger and less well circumscribed than capillary and juvenile hemangiomas, and show no tendency to regress, most require treatment. Most cavernous hemangiomas are best treated by complete surgical excision. All 14 patients we reviewed were treated by surgical excision alone. There were no cases of serious intraoperative bleeding and/or recurrence (Table 2). Preoperative angiography and embolization were performed in only two cases. Angiography was not performed in the remaining cases because there

Table 2. Treatment of submandibular cavernous hemangiomas

Author	Preoperative angiography and embolization	Massive intraoperative bleeding	Treatment	Recurrence
This study	No	No	Surgical excision	No
Cho, et al.[3]	No	No	Surgical excision	No
	No	No	Surgical excision	No
	No	No	Surgical excision	No
	No	No	Surgical excision	No
	No	No	Surgical excision	No
El-Hakim and El-Khashab[2]	No	No	Surgical excision	No
McMenamin, et al.[5]	No	No	Surgical excision	No
Chuang, et al.[1]	No	No	Surgical excision	No
Ozturk, et al.[10]	No	No	Surgical excision	No
Kumar, et al.[4]	No	No	Surgical excision	No
	Yes	No	Surgical excision	No
	Yes	No	Surgical excision	No
	No	No	Surgical excision	No

was no suspicion of hemangioma before surgery. There were no incidences of serious intraoperative bleeding, even in the absence of preoperative embolization.

Therefore, we do not think that preoperative angiography and embolization are necessary. Kumar, et al.[4] also reported that preoperative vascular embolization in patients with cavernous hemangioma did not result in a significant reduction in total blood loss compared with that in patients who had not undergone embolization.

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