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Case Report

Submandibular triangle cavernous hemangioma: Case report and review of literature

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ABSTRACT

Hemangiomas in the submandibular triangle are relatively rare, and may be misdiagnosed as inflammatory lesions, benign tumors, IgG-related lesions, or salivary calculi. Diagnosis of these lesions is facilitated using appropriate imaging modalities. We report the case of a hemangioma occurring in the submandibular triangle region as well as a literature review of preoperative clinical diagnosis and image modalities. A 30-year-old woman was referred to our hospital from another dental clinic for treatment of a painful swelling of the left submandibular region. Contrast-enhanced magnetic resonance imaging (CE-MRI) showed a mass with clear boundaries in the left submandibular region. The CE-MRI dynamic study showed early enhancement and high washout. There was not abnormal accumulation of fluorodeoxyglucose (FDG) on positron emission tomography–CT (PET-CT), and it could not confirm a malignant lesion. Accordingly, the patient was suspected to have a submandibular gland benign tumor or arteriovenous malformation. The lesion is an arteriovenous malformation, but we judged it to be an extractable hemangioma. Intraoperatively, the lesion was found to be continuous with the submandibular gland and extracted en bloc. Surprisingly, histological diagnosis of the specimen was cavernous hemangioma. There was no recurrence at 4.5 years after surgery. The diagnosis of tumor-like disease occurring in the submandibular triangle region requires both collection of clinical findings and evaluation of the internal properties of the lesion. Preoperative diagnosis based on these combined findings enables construction of an appropriate treatment plan.

1. Introduction

The lesions occurring around the submandibular triangle region may present as malignant or benign tumors, inflammatory lesions as salivary calculi, and autoimmune diseases including IgG4-related lesions. These lesions are often accompanied by swelling and pain. Munir and Bradley reported 107 patients diagnosed with submandibular triangle neoplasms (49 benign and 58 malignant) [1]. Among the cases with benign lesions, 76% were pleomorphic adenoma cases, and 8% were cases of Warthin tumors and adenolymphoma. Among the cases with malignant lesions, 82.7% were primary malignancy cases and 17.2% were metastatic lesion cases (non-Hodgkin lymphoma, 38%; adenoid cystic carcinoma, 15.5%; mucoepidermoid carcinoma, 15.5%; Hodgkin lymphoma, 7%). Hemangioma was not diagnosed [1]; therefore, hemangioma in the submandibular triangle are very rare. These lesions are often misdiagnosed as other diseases (Table 2). The clinical and radiologic features of hemangioma occurring in submandibular triangle have not been systematically reported. Generally, hemangiomas with phlebolith resemble cystic lesions with calcified nodules on computed tomography (CT) but it is difficult to distinguish them from salivary calculi [2]. Magnetic resonance imaging (MRI) of hemangiomas showed enhancement on T1- and T2-weighted images. Dynamic MRI of cavernous hemangioma shows slow enhancement and low washout. Herein, we report a rare case of cavernous hemangiomas around a submandibular salivary gland that showed rapid increase in signal intensity on dynamic contrast-enhanced MRI (CE-MRI).

2. Case report

A 39-year-old woman was referred to the Maxillofacial Surgery Department at the Dental Hospital of Tokyo Medical and Dental University for evaluation of pain and swelling in the submandibular region. These symptoms had been present for 5 years, but increased swelling and pain resulted in referral from a local dental clinic. The patient's medical and family histories were unremarkable. On clinical examination, a mildly tender 25 × 15 mm elastic hard mass was

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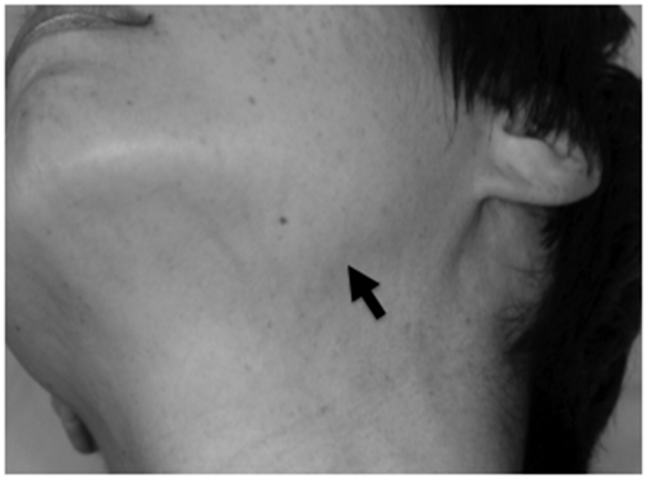

Fig. 1. Facial photograph taken during initial diagnosis.Mass formation was confirmed in left submandibular region.

observed in the left submandibular area. There was no pulsation in the lesion (Fig. 1). There was no blockage of the submandibular duct and outflow of saliva from the sublingual papilla was unobstructed. Although an IgG4-related lesion was suspected, blood tests showed unremarkable values of IgG4. Her white blood cell count was 2.13×10^9/L and C-reactive protein (CRP) level was 8.8 mg/L (Table 1), which ruled out inflammatory lesions. CE-MRI revealed a $28 \times 19 \times 15$ mm lesion with clear boundaries in the lower left submandibular region. The lesion showed heterogeneity of signal intensity including high signal on T2-weighted imaging (Fig. 2). Furthermore, inhomogeneous enhancement of gadolinium within the tumor was confirmed using CE-MRI. Time-signal intensity curves showed a peak time of 43 s and washout rate of 35.8%, by CE-MRI dynamic studies. This showed early enhancement and high washout. There was not abnormal accumulation of fluorodeoxyglucose (FDG) on positron emission tomography–CT (PET-CT), thus ruling out a malignant tumor, lymphoid tumor, or neurogenic tumor. Accordingly, a submandibular gland benign tumor or arteriovenous malformation was suspected. Although arteriovenous malformation was suspected in the differential diagnosis we did not perform FNA to avoid the high risk of bleeding. Signal void and blood flow into the tumor were not observed on MRI. We performed tumor excision via a submandibular incision under general anesthesia. A number of vessels into the submandibular gland were observed (Fig. 3). The tumor had not invaded the salivary gland but was close to the capsule of the submandibular gland (Fig. 4A, B). The tumor was continuous with the surface of submandibular gland, and the tumor body was elastic hard and dark red. The tumor was removed from the submandibular gland with difficulty; then, the tumor was excised with the salivary gland en bloc. Pathological findings revealed vascular endothelial hyperplasia of various sizes and shapes. Inflammatory cell infiltration was occasionally observed in the submandibular gland and tumor. Acinar and ductal tissue showed relatively little change. There were many expanded conduits, and the lumen was accompanied by an accumulation of mucus (Fig. 4C). These findings led to a histopathological diagnosis of cavernous hemangioma, which contrasted with the results seen on dynamic MRI. Postoperatively, nerve paralysis of the mandibular inferior margin of the facial nerve was observed. Currently, paralysis is slowly improving with medication. No recurrence was observed at the follow-up examination 4.5 years after surgery.

Table 1
The blood examination data during initial diagnosis.

IgG1	691	mg/dl	53.90%
IgG2	489	mg/dl	38.15%
IgG3	13.3	mg/dl	1.04%
IgG4	88.6	mg/dl	6.91%
WBC	2130	/μL	
CRP	0.88	mg/dl	
AMY	146	U/L	

3. Discussion

According to the International Society for the Study of Vascular Anomalies classification, hemangiomas are classified as congenital vascular tumors and vascular malformations [3]. Vascular malformations are subdivided into venous malformations, arteriovenous malformations, capillary malformations, and lymphatic malformations. When the lesion exists in deep tissue, it is often difficult to diagnose preoperatively. For preoperative diagnosis, internal characteristics can be assessed using technetium-labeled red blood cell scintigraphy, CT, CT angiography (CTA), dynamic MRI, and/or ultrasonography. Dynamic MRI shows that late enhancement, absence of flow void, and the presence of dilated venous space are indicative of venous malformation; early enhancement, the absence of flow voids, and the presence of dilated venous spaces are indicative of capillary-venous malformations; and early enhancement and the presence of flow voids are indicative of arterial or arteriovenous malformations [5]. Van Rijswijk et al. [5] reported that they could differentiate venous and non-venous malformations with 100% sensitivity using MRI, but with the addition of dynamic MRI, specificity increased to 95%, with acceptable sensitivity remaining at 83%. CTA reveals what artery or vein supplies the hemangioma, and whether there is nidus. If necessary, preoperative vascular embolization is performed. Kumar et al. [4] reported that the hemangioma had been supplied by the facial artery and lingual artery by CTA, so they performed preoperative vascular embolization. However, this did not result in a significant decrease in total blood loss. So it is considered important to diagnose in combination. In case of findings of non venous malformation by dynamic MRI, we should think that it is important to conduct CTA. Cavernous hemangiomas are classified as venous malformations. Using dynamic MRI to evaluate the flow rate [6], cavernous hemangiomas often show slow enhancement and low washout [5]. In contrast, the present case showed atypical findings of early enhancement and high washout of signal intensity on dynamic MRI at the preoperative image examination.

Most lesions occurring around the submandibular glands are inflammatory lesions, malignant or benign tumors, salivary calculi, or autoimmune diseases including IgG4-related lesions. These lesions are often accompanied by swelling and pain. Isacsson et al. reported that salivary calculi caused inflammation in the submandibular gland in 83% of patients who presented with pain in the region [7].

Kumar et al. reported that 82.6% of patients undergoing submandibular resection had chronic sialadenitis, 10.9% had polymorphous adenomas, 4.8% had adenocarcinomas, and 1.7% had hemangiomas [4]. In addition, Munir and Bradley reported the proportions of tumorous lesions occurring in the submaxillary triangle that had been surgically removed. In their report, there was no mention of hemangioma [1]. Azadarmaki et al. reported that it is difficult to detect this disease in the early stages because of a lack of obvious symptoms; in their case, a submandibular gland hemangioma progressed to the nasopharyngeal space before detection [8]. Hemangioma of the submandibular gland and submandibular triangle is rare. Also, clinical findings are limited, and diagnosis is often difficult. It is important to maintain clinical suspicion for hemangioma in the submandibular triangle, and question a diagnosis of sialadenitis including sialolith. Inflammatory findings were excluded in this case based on the results of blood tests. No radiopaque areas were observed on panoramic radiographs and CT images in this case. Therefore, a Kuttner tumor was suspected. The first choice of therapy for salivary gland tumors is surgical resection, and surgical approach differs according to a benign or malignant presentation [9]. When the radiographic and clinical findings cannot distinguish between benign and malignant tumors, FNA or