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Submandibular schwannoma arising from the hypoglossal nerve radiologically masquerading as submandibular gland tumor

KEYWORDS

Core needle biopsy;
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Schwannomas in the submandibular space are very rare and originate from the hypoglossal nerve, lingual nerve, and parasympathetic nerve within the submandibular gland (SMG).^{1,2} Although submandibular schwannomas are located lateral to the genioglossus and hypoglossus muscles and compress the SMG, submandibular schwannomas are usually not included in the differential diagnosis of submandibular lesions.¹ Therefore, submandibular schwannomas are difficult to diagnose preoperatively and may sometimes mimic SMG tumors.^{1,3,4} We reported a submandibular schwannoma arising from the hypoglossal nerve radiologically masquerading as an SMG tumor.

A 52-year-old woman with right submandibular swelling was referred to our hospital for treatment. Contrast-enhanced computed tomography (CT) showed a well-circumscribed, heterogeneous low-density tumor ($21 \times 20 \times 19$ mm) in the right SMG (Fig. 1A and B). Magnetic resonance imaging (MRI) revealed a well-circumscribed SMG tumor ($23 \times 22 \times 18$ mm) with the homogenous low-signal intensity in the T1-weighted image and the heterogenous high-signal intensity in the short tau inversion recovery image (Fig. 1C and D). Ultrasonography (US) showed a well-circumscribed, heterogenous tumor with blood flow (Fig. 1E and F). Because US-guided core needle biopsy (CNB) revealed a schwannoma, the lesion was diagnosed as the submandibular schwannoma arising from the lingual or hypoglossal nerve. The patient underwent intraoral removal of the tumor under general anesthesia. Because the tumor was

arising from the hypoglossal nerve, the tumor was completely removed, including part of the nerve. Because the specimen presented the highly cellular areas (Antoni A) and the hypocellular areas (Antoni B) and immunohistochemical positivity to S-100 protein (Fig. 1G and H), the final pathological diagnosis was a schwannoma. Although there was no recurrence 10 months after surgery, the slight hypoglossal nerve palsy remained.

When schwannomas arising in the submandibular space are radiologically misdiagnosed as SMG tumors, unnecessary SMG removal may be performed.¹ Therefore, preoperative diagnosis of schwannomas is vital. The radiological diagnosis of schwannomas is performed by several imaging modalities. The CT features are the well-circumscribed tumors with the low or soft tissue attenuation and enhancement reflecting its histological features.^{1,3} MRI shows the low-signal intensity in the T1-weighted images and the high-signal intensity in the T2-weighted images.¹

For preoperative diagnosis of head and neck lesions, the fine needle aspiration cytology (FNAC) is commonly performed under the guidance of the US. However, the diagnostic sensitivity of the FNAC for schwannomas is relatively low (0–40%).^{1,3,5} In contrast, CNB can harvest histological specimens, but CNB is not commonly used to diagnose schwannomas.⁵ A comparative study of the US-guided FNAC and CNB in diagnostic accuracy of extracranial head and neck schwannomas showed that the incidence of unsatisfactory specimens in the US-guided FNAC and CNB were

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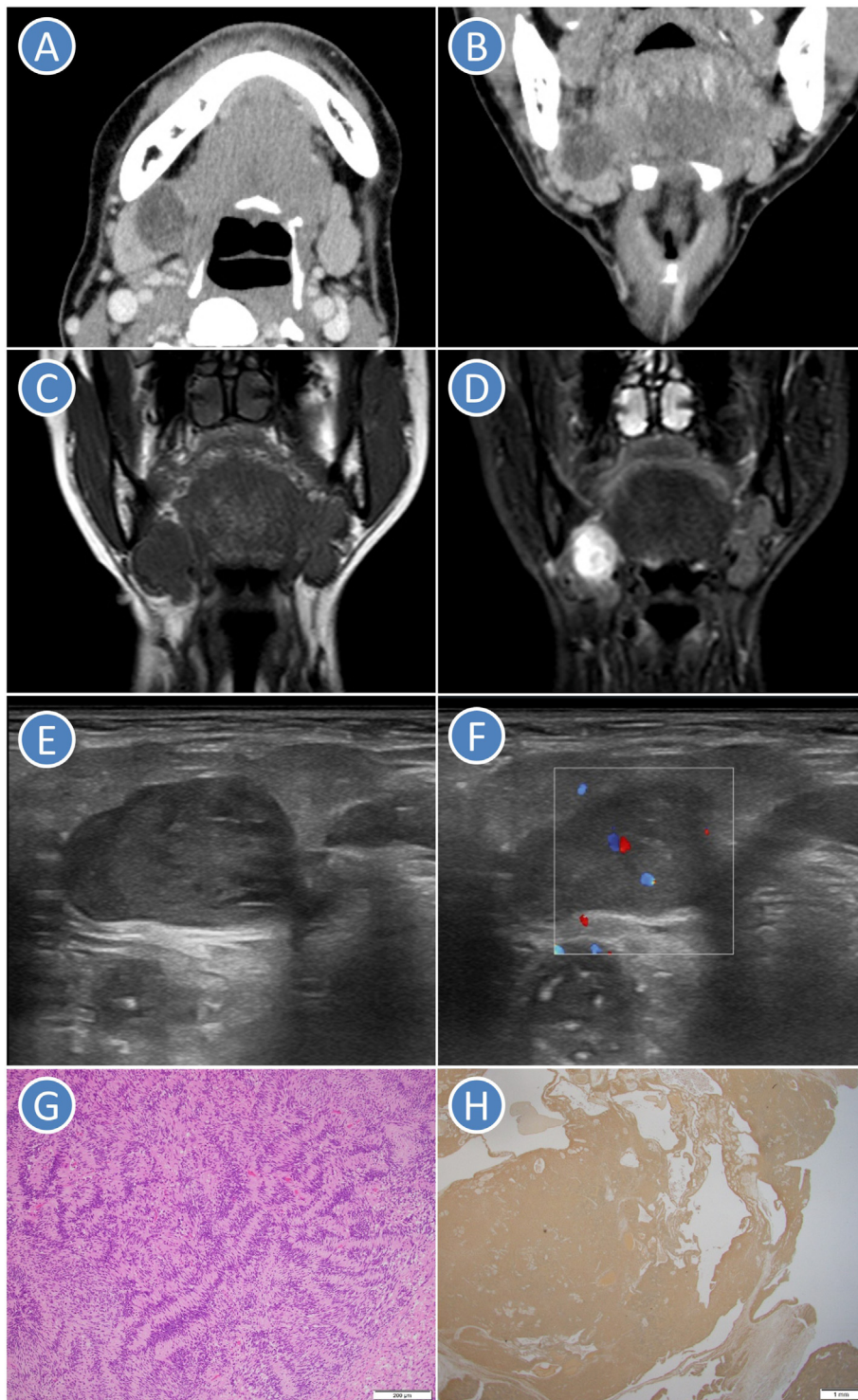


Figure 1 Radiological images and histopathological photomicrographs. (A and B) Contrast-enhanced CT (Axial and coronal image) showing a well-circumscribed, heterogeneous low-density tumor in the right submandibular gland (SMG), (C) MRI (T1-weighted coronal image) revealing a well-circumscribed SMG tumor with the homogenous low-signal intensity, (D) MRI (Short tau inversion recovery coronal image) demonstrating a well-circumscribed SMG tumor with the heterogenous high-signal intensity, (E and F) Ultrasonography showing a well-circumscribed, heterogenous tumor with blood flow. (G) Hematoxylin and eosin staining showing the highly cellular areas (Antoni A, main components) and the hypocellular areas (Antoni B), (H) Immunohistochemical staining revealing positivity for S-100 protein.

46.2 % and 0 %, respectively.⁵ Specific diagnoses of schwannomas, including “suspicious schwannoma” and “consistent with schwannoma”, were obtained from 19.2 % of the US-guided FNAC samples and 96.6 % of the US-guided CNB samples.⁵ In the present case, the preoperative diagnosis of schwannoma rather than SMG tumor could be obtained, because the US-guided CNB with the high diagnostic accuracy of schwannomas was performed.

In conclusion, oral surgeons should consider schwannomas in the radiological differential diagnosis of submandibular tumors and the CNB should be performed for the preoperative diagnosis, although submandibular schwannomas are very rare.

Declaration of competing interest

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

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