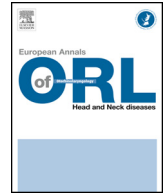




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What is your diagnosis?

A solitary asymptomatic submandibular mass

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1. Case description

A 32-year-old woman was referred to our department with a painless swelling in the right submandibular region. The elastic hard mass was mobile in the bimanual palpation. The mucosa of the oral floor was normal. The salivary flow was normal, and there was no paresthesia of the right lingual or hypoglossal nerve. Contrast-enhanced computed tomography (CT) showed a well-circumscribed, heterogeneous low-density tumor ($21 \times 17 \times 15$ mm) in the right submandibular gland (Fig. 1A,B). There was no cystic or necrotic degeneration. Magnetic resonance imaging (MRI) showed a well-circumscribed submandibular tumor with homogenous low-signal intensity in T1-weighted image, heterogeneous high-signal intensity in fat-suppressed T2-weighted image, heterogeneous enhancement in gadolinium-enhanced T1-weighted image (Fig. 1C-E). Apparent diffusion coefficient (ADC) value in diffusion-weighted image was $1.3 \times 10^{-3} \text{ mm}^2/\text{s}$ (Fig. 1F). Fine needle aspiration cytology (FNAC) showed benign tumor.

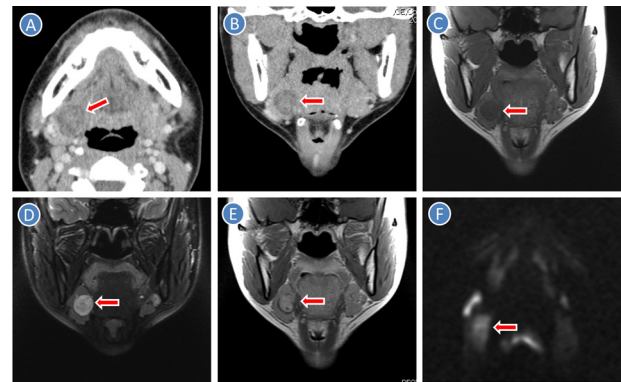


Fig. 1. A Contrast-enhanced CT (Axial image), B: Contrast-enhanced CT (Coronal image), C: MRI (T1-weighted image), D: MRI (Fat-suppressed T2-weighted image), E: MRI (Contrast-enhanced T1-weighted image), F: MRI (Diffusion-weighted image). Arrow indicates tumor.

What is your diagnosis?

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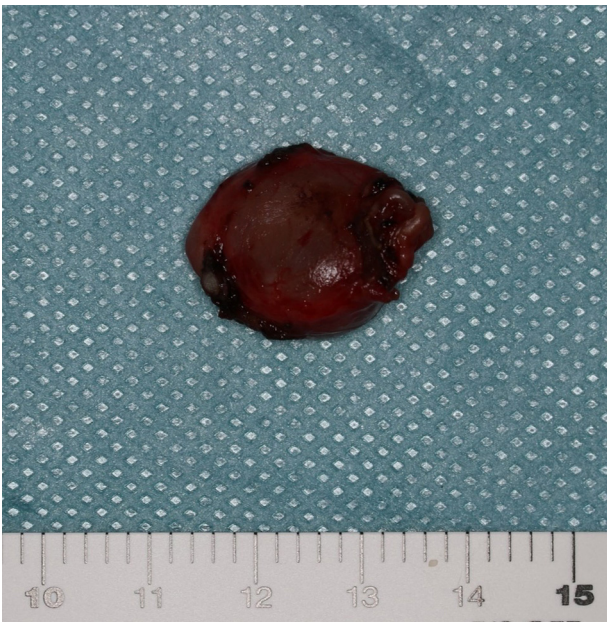


Fig. 2. The removed specimen.

2. Answer

The lesion was diagnosed radiologically as a benign submandibular gland tumor, and the intraoral submandibulectomy was performed under general anesthesia. However, there was no definitive continuity between the tumor and the submandibular gland, and the tumor originated from a thin branch of the hypoglossal nerve. Complete removal of the tumor was performed after cutting of the branch (Fig. 2). The pathological diagnosis of the specimen was schwannoma with highly cellular areas (Antoni A) and hypocellular areas (Antoni B). Postoperatively, there was a slight hypoglossal nerve palsy, which completely recovered 2 months after surgery.

3. Discussion

Schwannomas are benign, slow-growing, solitary, and encapsulated tumors that are composed of Schwann cells, and commonly arise from the sensory division of the major and cranial nerve [1,2]. Schwannomas in the submandibular space are extremely rare [1,3–5], and are located lateral to the genioglossus and hypoglossus muscles and compress the submandibular gland [4]. Therefore, some submandibular schwannomas may be diagnosed preoperatively as submandibular gland tumors [3]. The present case had schwannoma arising from the thin branch of the hypoglossal nerve masquerading as a submandibular gland tumor.

The differential diagnosis of submandibular lesion includes lymph node metastasis, lymphadenitis, malignant lymphoma, and

submandibular gland tumor such as pleomorphic adenoma or metastatic tumor [1,6]. Common clinical presentation is painless swelling. FNAC is commonly not helpful for the diagnosis of schwannoma [3], but may rule out a malignant tumor. The radiological diagnosis of schwannoma is performed by several imaging modalities. The CT features are well-circumscribed tumors with low- or soft-tissue attenuation and enhancement reflecting its histological features. MRI is most sensitive and specific in the diagnosis of schwannoma in the head and neck region [2]. MRI shows low-signal intensity in T1-weighted image and high-signal intensity in T2-weighted image [2,3]. Target sign (biphasic pattern of peripheral hyperintensity and homogeneous central hypointensity on T2-weighted images) and reverse target sign (biphasic pattern central high intensity and peripheral low intensity on gadolinium-enhanced T1-weighted images) are specific to schwannomas [2]. On diffusion-weighted image, schwannomas show high ADC values [2,7], and high ADC values, which indicate ancient change, may prove useful when distinguishing schwannoma from other lesions [7].

In salivary gland tumors, benign tumors have higher ADC values than malignant tumor, and pleomorphic adenomas have the highest ADC values [8]. Warthin tumors has lower or similar ADC values compared with malignant tumors [8]. Consequently, schwannoma, which involved in the submandibular gland could not distinguished from pleomorphic adenoma by ADC value in the present case.

In conclusion, radiologists should consider schwannoma in the radiological differential diagnosis of submandibular tumors, although submandibular schwannomas are very rare.

Disclosure of interest

The authors declare that they have no competing interest.

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