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Schwannoma Derived from Lingual Nerve Occurring in Floor of Mouth

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Abstract

A schwannoma is a benign tumor composed of schwann cells which forms on the periphery of nerves. We report a case of a schwannoma derived from a lingual nerve occurring in the floor of the mouth. The patient was a 27-year-old woman who presented with the complaint of a swelling in the floor of the mouth. It is difficult to distinguish a swelling from a sublingual gland tumor, cyst, or malignant tumor by MRI alone. Therefore, a biopsy and cytological examination were performed one week prior to surgery to determine whether the growth represented a malignancy. The results revealed a class II growth which was suspected to be a schwannoma. Intraoperatively, it became clear that the tumor and lingual nerve were inseparable, making excision of the nerve unavoidable. On the other hand, there was a clear border between the tumor and the sublingual gland, so it was possible to preserve the sublingual gland. In the postoperative pathological diagnosis, a definitive diagnosis was difficult based solely on H-E staining. Therefore, immunohistochemical staining was performed, resulting in a diagnosis of schwannoma. Currently, the patient is still being followed up. The results of this case indicate that preoperative aspiration biopsy cytology is useful in deciding the operative method to be employed.

Key words: Schwannoma — Lingual nerve — Floor of the mouth

Introduction

A schwannoma is a benign tumor composed of schwann cells, which are covered by neurilemma in myelinated nerve fibers. These tumors usually occur under the skin of the limbs or head and neck, and are only little found in the oral area. Here, we report a case of a schwannoma derived from the lingual nerve occurring in the floor of the mouth. Clinically, it appeared to be a benign tumor, but MRI findings suggested a malignancy. Here, we discuss the radiographic findings of this case and review the literature.
Case

Patient: 27-year-old woman.
Chief complaint: Swelling on floor of mouth.
First visit: February, 2010.

Current medical history: She noticed discomfort in the left-hand side of the floor of the mouth in 2005. On attending her local dental clinic for tooth treatment in January 2010, her dentist pointed out the lesion and advised her to visit our hospital for further evaluation and treatment.

Previous medical history: Grave’s disease at 25 years of age.

Present illness:

Facial appearance: Symmetrical. Mild dysarthria due to a mass on the floor of the mouth was noted.

Oral cavity: There was a palpable mass on the floor of the mouth on the left-hand side. It was $18 \times 15$ mm with hard elasticity; no fluctuation or tenderness was observed (Fig. 1). The surface of the mucosa showed normal coloring, with no redness or ulceration.

MRI findings: The tumor was located on top of the mylohyoid muscle and had displaced the existing structure to the right. The tumor produced a low signal on T1-weighted images, while the inside of the growth produced a high and heterogeneous signal on T2-weighted images. The margin was well defined. The area of low signal indicated necrosis inside the tumor. These findings indicated a malignant salivary gland tumor (Fig. 2).

Clinical diagnosis: Tumor on left-hand side of floor of the mouth.

Treatment and course: Fine needle aspiration (FNA) cytology was performed at one week before surgery as a malignant tumor

Fig. 1 Mass occurred on left-side floor of mouth
Size: $18 \times 15$ mm; hard elasticity; painless growth; uneven and irregular in shape; mucosa showed healthy coloring.

Fig. 2 MRI findings revealed clearly defined mass, producing high signal on T1-weighted and low signal on T2-weighted images; images in horizontal plane revealed heterogeneous contents
a: Horizontal section (T2-weighted image), b: Coronal section (T2-weighted image).
Schwannoma in Floor of the Mouth

was suspected. Cells with oblong or spindle-shaped nuclei were arranged in a palisade. No epidermal cells, which are characteristic of salivary tumors, or myoepithelial cells were observed (Fig. 3). The results of FNA cytology indicated a diagnosis of schwannoma (class II).

Surgical findings: Resection of the tumor was performed in April, 2010 under general anesthesia. An incision line was made above the left Wharton duct, which was directly above the tumor. Dissection of the mucosa and blunt dissection revealed a membrane-covered lesion. The tumor and lingual nerve were carefully decorticated, as the lingual nerve had penetrated the tumor (Fig. 4-a). The lesion and sublingual gland were clearly demarcated, allowing the sublingual gland and duct to be preserved (Fig. 4-b). After setting drains, the wound was sutured (Fig. 4-c). The excised tumor was $15 \times 30 \text{ mm}$ and covered in membrane; it had an irregular, bumpy surface, hard elasticity, and was solid (Fig. 5).

Histopathological findings: Differentiation between pleomorphic adenoma and schwannoma is important in arriving at a diagnosis. 

Fig. 3 Cells were oblong with spindle-shaped nuclei arranged in palisade
Results of FNA cytology indicated diagnosis of schwannoma (class II).

Fig. 4 a: Lingual nerve had penetrated tumor, b: Sublingual gland and duct were preserved, c: After setting of drains, wound was sutured
The tumor cells, which were spindle-shaped with an oval nucleus, had proliferated close together in a palisade arrangement, and there was no duct structure. Partially myxoid component sites were observed, together with an accumulation of mucosal fluid with floating foam cells (Figs. 6-a, b, c). The results of immunohistochemical staining were positive.
for S-100 (Fig. 6-d), GFAP, Vimentin, and NSE, but negative for p63. This was helpful in distinguishing the tumor from a neurilemmoma.

Postoperative course: There was desensitization of the left side of the tongue immediately after the operation, but no distortion of taste. The patient noted a strong bitter taste postoperatively, but this has abated at 26 months later. No recurrence of the tumor has been observed.

Discussion

A schwannoma is made up of neurilemma-derived schwann cells, and is characteristically a hard, elastic mass. This type of tumor can develop in various parts of the body, but only 4% are found in the oral cavity, of which 50% are on the tongue, with occurrence on floor of the mouth extremely rare. Although some reports have indicated no difference in prevalence between sexes, others have reported it to be higher in women. Occurrence is also greater between the ages of 10 and 20 years, showing a tendency to develop at a younger age. A number of factors have been reported to involved in their development, including trauma, internal secretion, and abnormal growth of the nervous system. It was impossible to determine which, if any, of these factors contributed to its development in the present case, however.

Twenty-four cases of this type of tumor have been treated at our institute between 1960 and 2010, and a male-to-female ratio of 7 to 17 indicates a tendency to occur in women. The patients’ age ranged from 12 to 65 years, although there were more young patients than old. By site, 5 tumors occurred on the palate, 4 in mandibular bone, 4 on the tongue, and 2 on the floor of the mouth.

Table 1 Clinical statistics of schwannoma at this college and in Japan

<table>
<thead>
<tr>
<th></th>
<th>Tokyo Dental College</th>
<th>Japan</th>
</tr>
</thead>
<tbody>
<tr>
<td>Case No.</td>
<td>24</td>
<td>138</td>
</tr>
<tr>
<td>Sex</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Male</td>
<td>7</td>
<td>69</td>
</tr>
<tr>
<td>Female</td>
<td>17</td>
<td>69</td>
</tr>
<tr>
<td>Average age</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Total</td>
<td>36.1</td>
<td>39.6</td>
</tr>
<tr>
<td>Male</td>
<td>27.3</td>
<td>38.1</td>
</tr>
<tr>
<td>Female</td>
<td>39.8</td>
<td>41.3</td>
</tr>
<tr>
<td>Site (%)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Oral floor</td>
<td>2 (8%)</td>
<td>10 (7%)</td>
</tr>
<tr>
<td>Tongue</td>
<td>4 (16%)</td>
<td>51 (37%)</td>
</tr>
<tr>
<td>Palate</td>
<td>5 (21%)</td>
<td>6 (4%)</td>
</tr>
<tr>
<td>Mandibular bone</td>
<td>4 (16%)</td>
<td>27 (20%)</td>
</tr>
<tr>
<td>Maxillary bone</td>
<td>0 (0%)</td>
<td>1 (0.7%)</td>
</tr>
<tr>
<td>Upper lip</td>
<td>3 (13%)</td>
<td>7 (5%)</td>
</tr>
<tr>
<td>Lower lip</td>
<td>2 (8%)</td>
<td>5 (4%)</td>
</tr>
<tr>
<td>Submandibular region</td>
<td>2 (8%)</td>
<td>11 (8%)</td>
</tr>
<tr>
<td>Buccal mucosa</td>
<td>1 (4%)</td>
<td>12 (9%)</td>
</tr>
<tr>
<td>Mandibular gingiva</td>
<td>0 (0%)</td>
<td>4 (3%)</td>
</tr>
<tr>
<td>Maxillary gingiva</td>
<td>1 (4%)</td>
<td>0 (0%)</td>
</tr>
<tr>
<td>Submental region</td>
<td>0 (0%)</td>
<td>2 (1%)</td>
</tr>
<tr>
<td>Pterygomandibular spatiun</td>
<td>0 (0%)</td>
<td>2 (1%)</td>
</tr>
</tbody>
</table>
Of 138 cases reported in Japan over the last decade (2000–2010), 69 occurred in men, showing no difference between sexes. Age ranged from 8 to 86 years. In terms of site, 51 occurred on the tongue, 27 in mandibular bone, 12 in buccal mucosa, 11 in the submandibular region, and 10 on floor of the mouth. Seven per cent occurred on the floor of the mouth (Table 1).

Clinically, these tumors are characterized by painless growth and a covering membrane. The size is reported to be around 20–30 mm\(^1\)–\(^4\),\(^9\),\(^12\),\(^13\). The present case was, therefore, of average size, at 15–30 mm. Because it developed on the left-hand side of the floor of the mouth, the differential diagnoses were cystic diseases such as epidermoid, dermoid, or thyroglossal duct cyst, or neoplastic diseases such as hemangioma, leiomyoma, rhabdomyoma, or salivary gland tumor.

Contrast on the CT images was equal or at a low level in muscle, while it was moderately uneven on the inside of the tumor. Low-to-moderate signal intensity on T1W1 and high signal intensity on T2W1 images are reported to be characteristic MRI findings\(^1\),\(^5\),\(^9\),\(^12\). In the present case, a low signal was observed on T1W1 and a high, uneven signal on T2W1 images. The area of low signal indicated necrosis inside the tumor and adhesion to the lingual nerve was recognized. The mass was suspected to be a small salivary gland tumor or malignant tumor such as a sarcoma or metastatic tumor, as dense staining was observed from the early to late stage in contrast-enhanced CT.

As previously noted, schwannomas have been reported to be relatively small, at 20–30 mm. When such tumors grow larger, however, the following may be observed: a hyaline deposit inside the tumor; a cystic tumor; proliferation of blood vessels; bleeding; calcification; and necrosis. In such cases, the lesion is called an ancient schwannoma, and these are rarely reported in the oral cavity\(^7\),\(^11\).

It is possible that secondary change had occurred within the lesion, as several years had lapsed before treatment was commenced, and indeed the MRI findings appear to indicate this. Many studies have reported the usefulness of MRI in diagnosing such tumors\(^2\),\(^6\). However, it is sometimes difficult to distinguish them from malignant tumors, as in this case.

The following histopathological findings have been reported: 61% are Antoni A-type, which are characterized by proliferation of spindle-like cells and nuclei arranged in a palisade formation; 5% are Antoni B-type, in which the cells are patterned sparsely, with mucoid degeneration, bleeding, and production of cyst; and 34% are a combination of type A and type B\(^3\),\(^8\),\(^9\).

The standard treatment for such tumors is resection, as in the present case. Reports of relapse are rare, and it is said to have a good prognosis. A median, vertical incision is usually selected for small tumors occurring on the floor of the mouth. In the present case, as described earlier, synechia was observed between the lingual nerve and tumor, suggesting that it had developed from the lingual nerve. Preoperative MRI findings suggested a malignancy, and a diagnosis based on these images alone was difficult. However, needle aspiration cytology indicated a schwannoma, demonstrating the usefulness of this procedure in diagnosing this type of tumor.

**Conclusion**

We have reported a lingual nerve-derived schwannoma developing in the floor of the mouth.

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