Glomus Tumor of the Upper Lip

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Abstract
A case of glomus tumor of the upper lip in a 57-year-old Japanese male is described. The patient had a tender swelling of the upper lip, and this was completely excised. The lesion was found to be encapsulated by fibrous connective tissue and composed of blood capillaries surrounded by sheets of epithelioid cells. Immunohistochemical examination revealed that the neoplastic cells were positive for S-100 protein, vimentin, desmin and actin. Although the endothelial cells of blood capillaries in the tumor tissue were positive for factor VIII, no reaction product was found in the tumor cells. These results suggested that the tumor cells had characteristics of smooth muscle cells.

Introduction
Glomus tumor is a well defined histopathologic entity thought to be derived from the glomus body[1]. Histologically, the neoplasm is composed of blood capillaries surrounded by epithelioid cells which have the characteristics of smooth muscle cells[1]. The tumor typically occurs in the skin of the finger, especially beneath the fingernails and fingertips[1,3]. Cutaneous and subcutaneous glomus tumors in various locations have been reported[2,3]. A small number of glomus tumors have also been described in oral locations such as the hard palate[4,5], gingiva[6-8], cheek[9,10], tongue[11,12] and lip[13,14]. We report a case of glomus tumor occurring in the upper lip, in which the nature of the tumor cells was examined by immunohistochemistry.

Case report
A 57-year-old Japanese man presented with a complaint of midline swelling of the upper lip, which had been present for several years. Clinically, the lesion was a well demarcated, spherical nodule with tenderness and partial erosion on its surface (Fig. 1). It was removed surgically under local anesthesia. The tumor was well circumscribed and easily excised, and there has been no evidence of recurrence for 4 years after surgery.

Materials and Methods
The surgical specimen was fixed routinely in 10% neutral buffered formalin and embedded in paraffin wax. Sections were subjected to routine histologic examination. Immunohistochemical analysis was also performed to examine the localization of S-100 protein, smooth muscle actin, desmin and vimentin in the tumor. The avidin-biotin peroxidase complex (ABC) method was applied to deparaffinized sections.

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Results

The tumor was completely encapsulated by fibrous connective tissue (Fig. 2) and composed of blood capillaries surrounded by glomus cells, characterized by relatively large nuclei and homogeneous cytoplasm (Fig. 3). Immunohistochemical analysis revealed that the tumor cells were positive for S-100 protein (Fig. 4), smooth muscle actin, vimentin and desmin (Fig. 5). A positive reaction product for S-100 protein was observed in the cytoplasm as well as in the nucleus (Fig. 4). Endothelial cells of the blood capillaries were positive for factor VIII, but the tumor cells were negative (Fig. 6).

Discussion

Glomus tumor is a relatively rare neoplasm derived from the glomus body and typically occurs in the skin of the finger, especially beneath the fingernails and fingertips[1-3]. The tumor also occurs uncommonly in oral region, and a small number of cases in the hard palate[4,5], gingiva[6-8], cheek[9,10], tongue[11,12] and lip[13,14] have been reported (Table 1). Although glomus tumors usually occur as a single lesion, a few cases of multiple lesions have been described[2,15,16]. Charles[15] reported a case of multiple glomus tumors involving the face, palate, lip, cheek, eyelid and anterior orbit. Tsuneyoshi et al.[2] reported that the tumors were more common in female (41/63) than in male (22/63) patients. On the other hand, Heys et al.[3] have reported that they are more common in men (27/43) than in women (16/43). Of twelve cases of glomus tumor of the oral cavity, the frequency has been the same between male (6/12) and female (6/12) patients (Table 1). The majority of glomus tumors occur in patients in the third, fourth and fifth decades of life[1-3]. In the present case, the patient was a 57-year-old man.

Tenderness or pain is the most characteristic symptom of glomus tumors[1-3]. However, Ficarra et al.[13] have pointed out that lesions of the oral cavity are painless. In this case, the patient had noticed tenderness with the lesion.

The histopathological features of glomus tumor resemble those of the normal glomus body. The tumor is composed of endothelial-lined vascular spaces surrounded by round epitheloid cells. The tumors are separated into three varieties, depending on the predominant or characteristic histological pattern: (1) the vascular form; (2) the myxoid form; (3) the solid form[2]. In this case, the tumor was well encapsulated by fibrous connective tissue and showed solid form.

It has been reported that glomus tumor cells have the characteristics of smooth muscle cells, based on immunohistochemical[10,14,17-21] and ultrastructural[10-13,17,18,20,22-25] studies. Miettinen et al.[17] and Googe et al.[21] reported that glomus tumor cells were negative for desmin, as the same result in the glomus body. Brooks et al.[20] mentioned that the cells of one solid and microcystic glomus tumor were positive for desmin using the avidin-biotin peroxidase complex (ABC) method, in spite of a lack of reactivity by immunofluorescence. In this study we employed the ABC method and the tumor cells were positive for S-100 protein, smooth muscle actin, desmin and vimentin.

These results suggest that the cells of glomus tumor have the characteristics of smooth muscle cells, as described previously.

Acknowledgement

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References

Fig. 1 Swelling of the upper lip with tenderness

Fig. 2 Glomus tumor encapsulated by fibrous connective tissue

Fig. 3 Blood capillaries surrounded by glomus cells

Fig. 4 Immunoreactivity for S-100 protein in the nucleus and cytoplasm of the tumor cells

Fig. 5 Immunoreactivity for desmin in the cytoplasm of the glomus cells

Fig. 6 Immunoreactivity for factor VIII in the cytoplasm of endothelial cells
Table 1  Summary of previous cases of glomus tumor of the oral cavity

<table>
<thead>
<tr>
<th>Case report</th>
<th>Age</th>
<th>Sex</th>
<th>Location</th>
</tr>
</thead>
<tbody>
<tr>
<td>Von Langer 1949</td>
<td>52</td>
<td>M</td>
<td>Hard palate</td>
</tr>
<tr>
<td>King 1954</td>
<td>32</td>
<td>M</td>
<td>Gingiva</td>
</tr>
<tr>
<td>Kirschner and Strassburg 1962</td>
<td>56</td>
<td>M</td>
<td>Gingiva</td>
</tr>
<tr>
<td>Harris and Griffin 1965</td>
<td>35</td>
<td>F</td>
<td>Gingiva</td>
</tr>
<tr>
<td>Frenkel 1965</td>
<td>13</td>
<td>M</td>
<td>Cheek</td>
</tr>
<tr>
<td>Sidhu and Subherwal 1967</td>
<td>10</td>
<td>F</td>
<td>Hard palate</td>
</tr>
<tr>
<td>Charles 1976</td>
<td></td>
<td>F</td>
<td>Multiple</td>
</tr>
<tr>
<td>Sato et al. 1979</td>
<td>29</td>
<td>M</td>
<td>Tongue</td>
</tr>
<tr>
<td>Tajima et al. 1981</td>
<td>63</td>
<td>F</td>
<td>Tongue</td>
</tr>
<tr>
<td>Saku et al. 1985</td>
<td>45</td>
<td>M</td>
<td>Cheek</td>
</tr>
<tr>
<td>Ficarra et al. 1986</td>
<td>51</td>
<td>F</td>
<td>Upper lip</td>
</tr>
<tr>
<td>Moody et al. 1986</td>
<td>65</td>
<td>F</td>
<td>Upper lip</td>
</tr>
<tr>
<td>Present case</td>
<td>57</td>
<td>M</td>
<td>Upper lip</td>
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