Case Report

A Resected Case of Malignant Fibrous Histiocytoma of the Parotid Salivary Gland

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Abstract: We report here a resected case of malignant fibrous histiocytoma (MFH) of the parotid salivary gland. Only eleven previous cases of MFH involving the parotid salivary glands have been reported in the English-language literature. A 48-year-old Japanese man was admitted complaining of a slowly growing but not painful tumor that developed in the right cheek, and under the clinical diagnosis of pleomorphic adenoma of the parotid salivary gland, superficial parotidectomy was performed. Macroscopically, an encapsulated nodular tumor involving the parotid salivary gland was seen in the resected specimen, that measured 3 cm in the largest dimension. The cut surface was hard and rubbery, grayish-white in color and lacked necrotic foci. Microscopically, the tumor consisted of scattered atypical spindled and rounded mononuclear cells with hyperchromatic nuclei, which tended to be arranged in a storiform pattern, and occasionally multi-nucleated bizarre cells. The cells did not involve the parotid salivary gland. Immunohistochemically, the cells were negative for keratin, but stained positive for vimentin and focally for smooth muscle actin. This tumor was pathologically diagnosed as ordinary MFH of the parotid salivary gland arising from the para-glandular tissue. No recurrence was observed two months after surgery.

Key words: malignant fibrous histiocytoma, sarcoma, parotid salivary gland

Introduction

Sarcomas, including malignant fibrous histiocytoma (MFH), of the parotid salivary glands are very rare and must be distinguished from other spindle cell tumors, particularly those of epithelial and myoepithelial origins. Auclair et al. reported the following criteria to make a diagnosis of salivary gland sarcoma: 1) the neoplasm was clinically interpreted by the contributing institution as primarily involving a major salivary gland; 2) no other malignant tumors of the face, scalp or distant sites were noted in the clinical or follow-up records; 3) slides and/or paraffin blocks were available for study; 4) there was no morphologically identifiable neoplastic epithelial component present on routine hematoxylin and eosin-stained sections. Only eleven previous cases of MFH involving the parotid...
salivary glands have been reported in the English-language literature\(^2\text{-}^6\). We report here a resected case of MFH of the parotid salivary gland, which was clinically interpreted as pleomorphic adenoma of the parotid salivary gland.

**Clinical Summary**

A 48-year-old Japanese man was admitted to our hospital on the 22nd of November 2000 complaining of a slowly growing but not painful tumor that developed in the right cheek. Computed tomography revealed a solitary, well demarcated solid tumor without calcification, measuring 3 cm in dimension in the lower part of the right parotid salivary gland. No lymph node swelling or any other tumor was found in the pre-operative examination. The patient had been healthy until this admission, with no prior surgery. Under the clinical diagnosis of pleomorphic adenoma of the parotid salivary gland, superficial parotidectomy was performed and no recurrence was observed two months after surgery.

**Pathological Findings**

The resected parotid tissue measured \(3 \times 2.5 \times 3\) cm in size, with a nodular tumor involving the parotid salivary gland that measured 3 cm in the largest dimension. On sectioning, the tumor was solid, hard and rubbery, and grayish-white in color without necrotic foci (Fig. 1). Microscopically, the tumor consisted of scattered atypical spindled and rounded mononuclear cells with hyperchromatic nuclei, which tended to be arranged in a storiform pattern (Fig. 2A), admixed with lymphocytes and proliferating small vessels within a dense fibrous or myxomatous matrix. Multi-nucleated bizarre cells were also present in the tumor, and occasionally showed phagocytized erythrocytes (Fig. 2B). More than five mitotic cell figures were detected per ten high power fields. This tumor showed an expansive growth, and the cells did not involve the parotid salivary gland (Fig. 2C).

Immunohistochemically, the tumor cells, including multi-nucleated bizarre cells, showed diffusely positive staining with vimentin (DAKO Japan, Kyoto, Japan) and focally positive staining with a smooth muscle actin (DAKO), but were negative with cytokeratin (AE1/AE3; DAKO), epithelial membrane antigen (DAKO), factor-\(\text{VIII}\) related antigen (DAKO), CD34 (Novocastra, Newcastle-upon-Tyne, UK), S-100 protein (DAKO), HMB45 (DAKO), CD68 (DAKO) and desmin (DAKO). Furthermore, the cells were negative with Sudan-
Fig. 2. Microscopic findings of malignant fibrous histiocytoma of the parotid salivary gland
A. The tumor consisted of scattered atypical spindled and rounded mononuclear cells, which tended to be arranged in a storiform pattern (Hematoxylin and eosin, original magnification ×50)
B. Multi-nucleated bizarre cells were occasionally seen, and some cells (arrow) showed phagocytozed erythrocytes (Hematoxylin and eosin, original magnification ×100)
C. The tumor showed an expansive growth, and the cells did not involve the parotid salivary gland (Hematoxylin and eosin, original magnification ×2.5)

III staining.
From these findings, this tumor was pathologically diagnosed as typical MFH of the parotid salivary gland arising from para-glandular tissue.
Discussion

Sarcomas arising in and around the salivary glands (salivary gland sarcomas) are rare, accounting for only 0.3 to 1.5% of all salivary gland tumors\(^2\),\(^7\), and they are mostly MFH, malignant schwannoma and rhabdomyosarcoma\(^7\). Auclair et al.\(^2\) classified salivary gland sarcomas into three groups based on the relationship of the tumor with the surrounding tissue at the time of surgery; tumors arising from within the gland (Group I), tumors that could not be determined whether they arose in the gland or in the paraglandular tissues (Group II), and tumors of para-glandular tissues secondarily involving the gland (Group III). Sarcomas classified into Group I, which account for less than half, are regarded as primary salivary gland sarcomas.

To make a pathologic diagnosis of salivary gland sarcoma, carcinosarcomas should be excluded\(^8\). Furthermore, pleomorphic adenomas of predominantly spindle cell pattern are the tumors for which mesenchymal tumors are most likely mistaken\(^9\). Immunohistochemistry is of special value in excluding epithelial tumors with a spindle cell pattern, and cytokeratin immunoreactivity in a spindle cell tumor of the salivary gland is considered sufficient to classify the neoplasm as carcinoma, except for synovial sarcoma or epithelioid sarcoma\(^3\).

The present case satisfied Auclair's criteria for salivary gland sarcomas\(^2\); the neoplasm was clinically interpreted as primarily involving a parotid salivary gland, no other malignant tumors were detected, paraffin blocks were available for study, and no morphologically identifiable neoplastic epithelial component was present in the hematoxylin and eosin-stained sections. Furthermore, completely negative immunoreactivity with cytokeratin or epithelial membrane antigen in the spindled and rounded tumor cells suggested that this tumor originated from mesenchymal cells. On differential diagnosis, negative reactivity with fact or-VIII related antigen, CD34, S-100 protein, desmin and HMB45 was compatible with excluding sarcomas derived from vessels, peripheral nerves, adipose tissues, cross-striated muscles and malignant melanoma. From these immunohistochemical results and the histological storiform pattern of the tumor cells with the presence of multi-nucleated bizarre cells, this case was diagnosed as ordinary MFH of the parotid salivary gland, and classified into Auclair's Group III. Focal positive immunoreactivity with a smooth muscle actin, although rare in MFH\(^10\) and suggesting the differentiation toward myofibroblast, as well as negative immunoreactivity with CD68 are also compatible with MFH\(^11\).

To the best of our knowledge, only eleven previous cases of MFH involving the parotid salivary glands have been reported in the English-language literature\(^2\)\(^-\)\(^6\). The main features of these, together with the case presented here, are summarized in Table 1. Six occurred in males and six in females, the ages of the patients ranging from 28 to 91 years: the maximum incidence was in the 8th decade. Four died of disease within 4 years (range, 2 to 4 years) of the operation, and five were still alive (range, 0.2 to 5.3 years). The prognosis for patients with salivary gland sarcomas has been generally poor\(^2\)\(^-\)\(^4\),\(^7\) and about 50% of patients either died of their diseases or were alive with tumor\(^6\). A poor prognosis is also suggested by the summarized cases in Table 1. On the other hand, some articles referred to cases with a favorable prognosis\(^3\) reporting that patients with Group I sarcomas have a far better prognosis than the patients with Group III sarcomas, and Luna et al.\(^3\) described that all the living patients had sarcomas with low-grade malignancy measuring less
Table 1. Reported cases of MFH involving the parotid salivary gland

<table>
<thead>
<tr>
<th>Author</th>
<th>Age, y/Sex</th>
<th>Size, cm</th>
<th>Surgery</th>
<th>Follow-up, y</th>
</tr>
</thead>
<tbody>
<tr>
<td>Auclair et al. (2)</td>
<td>91/F</td>
<td>Uncertain</td>
<td>SP</td>
<td>Dead from other cause/2</td>
</tr>
<tr>
<td>Auclair et al. (2)</td>
<td>67/M</td>
<td>Uncertain</td>
<td>SP</td>
<td>Uncertain</td>
</tr>
<tr>
<td>Auclair et al. (2)</td>
<td>74/F</td>
<td>Uncertain</td>
<td>SX</td>
<td>Dead from other cause/5.8</td>
</tr>
<tr>
<td>Auclair et al. (2)</td>
<td>78/F</td>
<td>Uncertain</td>
<td>SX</td>
<td>Alive and well/5.3</td>
</tr>
<tr>
<td>Luna et al. (3)</td>
<td>75/M</td>
<td>6.0</td>
<td>RP</td>
<td>Dead from other cause/2.6</td>
</tr>
<tr>
<td>Luna et al. (3)</td>
<td>52/M</td>
<td>5.4</td>
<td>RP</td>
<td>Dead from other cause/3</td>
</tr>
<tr>
<td>Luna et al. (3)</td>
<td>29/M</td>
<td>2.2</td>
<td>TP</td>
<td>Alive and well/5</td>
</tr>
<tr>
<td>Benjamin et al. (4)</td>
<td>28/F</td>
<td>2.0</td>
<td>SP</td>
<td>Alive and well/0.5</td>
</tr>
<tr>
<td>van Wingerden et al. (5)</td>
<td>41/M</td>
<td>5.0</td>
<td>RP</td>
<td>Alive and well/0.5</td>
</tr>
<tr>
<td>Danielson et al. (6)</td>
<td>58/F</td>
<td>Uncertain</td>
<td>RP</td>
<td>Dead from other cause/4</td>
</tr>
<tr>
<td>Danielson et al. (6)</td>
<td>77/F</td>
<td>Uncertain</td>
<td>SP</td>
<td>Dead from other cause/2</td>
</tr>
<tr>
<td>Present case</td>
<td>48/M</td>
<td>3.0</td>
<td>SP</td>
<td>Alive and well/0.2</td>
</tr>
</tbody>
</table>

SP : Superficial parotidectomy ; SX : Surgical excision ; RP : Radical parotidectomy ; TP : Total parotidectomy

than 3 cm in dimension. The literature suggests that the present case, showing ordinary MFH measuring 3 cm in dimension arising from para-glandular tissue, is likely to have a poor prognosis.

In conclusion, the possibility of sarcomas, represented by MFH, should be considered in the exploration of parotid salivary gland tumors, although pleomorphic adenoma is the most common and it would be difficult to distinguish from other solitary demarcated tumors in the routine clinical examination.

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References

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