

Letter to Editor

Primary diffuse large B-cell lymphoma of the submandibular gland

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Received November 11, 2011; accepted January 7, 2012; Epub February 12, 2012; Published February 28, 2012

To the editor

Many kinds of malignant tumors occur in the major and minor salivary glands. Non-Hodgkin malignant lymphoma in the salivary glands is very rare, and account for about 1-5% of all salivary gland neoplasms [1]. The author herein reports a case of diffuse large B-cell lymphoma (DLBCL) occurring a submandibular gland. WHO classification of lymphoma was employed [2].

A 71-year-old Japanese woman consulted to our hospital because of left submandibular mass. She did not suffer from Sjogren syndrome and other connective tissue diseases. Physical examination and imaging modalities including CT and MRI revealed a left submandibular gland tumor measuring 6 × 6 × 5 cm (**Figure 1**). Clinical diagnosis was malignant pleomorphic adenoma. A large biopsy was performed. It revealed severe diffuse proliferation of atypical round cells (**Figure 2A**). No follicular structures were recognized. The atypical cells invaded the surrounding smooth muscle cells. Higher power viewer showed the atypical round cells are relatively large lymphoid cells with hyperchromatic nuclei and nucleoli (**Figure 2B**). Mitotic figures and apoptotic cells were scattered. Features of Burkitt lymphoma were not seen. An immunohistochemical study was performed with the use of Dako Envision method, as previously described [2, 3]. The atypical cells were positive for CD45 (**Figure 3A**), CD20 (**Figure 3B**), p53 protein (100%) (**Figure 3C**). The Ki-67 labeling was 100% (**Figure 3D**). In contrast, they were negative for various types of cytokeratins, epithelial membrane antigen, CD3, CD30, CD45RO, and TdT. Light chain restriction was

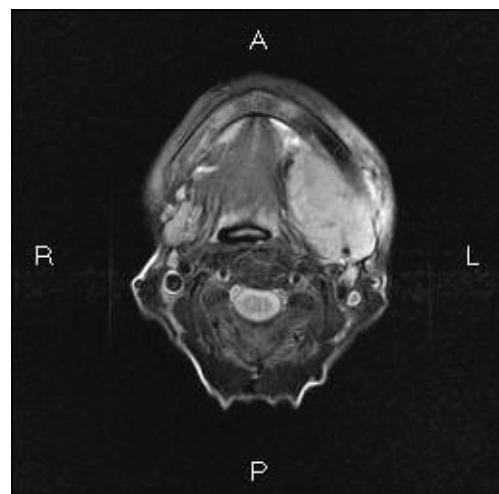


Figure 1. MRI features. A large tumor is seen in the left submandibular gland.

present; the tumor cells were positive for λ -chain (**Figure 3E**), but almost negative for κ -chain. The pathological diagnosis was DLBCL of the left submandibular gland. After the pathological diagnosis, systemic imaging study was performed, which revealed no other tumors. Lymph node swelling was not seen by these techniques. The patient underwent R-CHOP chemotherapy and radiation. The tumor markedly reduced in size 4 months after the diagnosis.

The present tumor showed diffuse proliferation of relatively large lymphoid cells. The histological, cytological, and immunohistochemical features are indicative of high grade B-cell neoplasm (DLBCL) [2]. Diffuse expression of p53

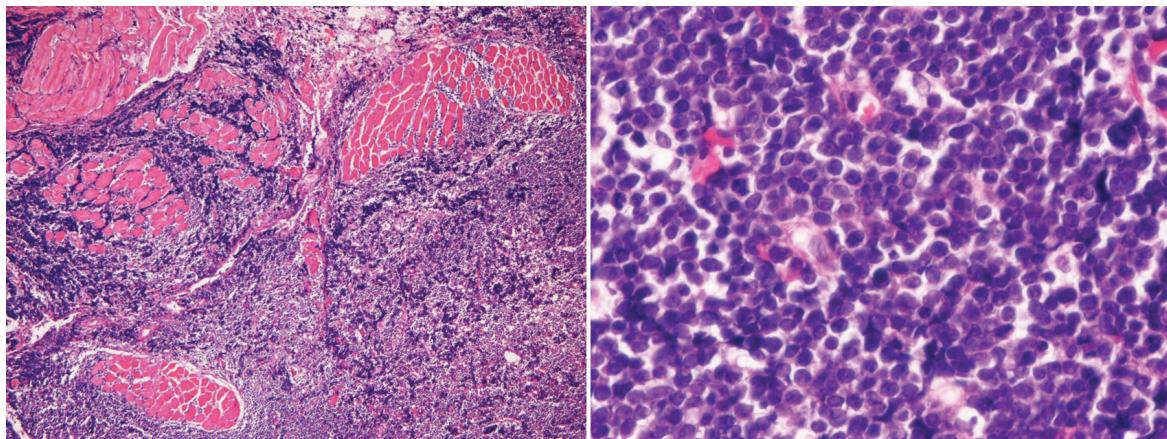


Figure 2. Histological features. A. Diffuse atypical lymphoid cell proliferation is seen. The lymphoid cells are invasive into the striated muscles. HE, $\times 20$. B. The atypical cells have hyperchromatic nuclei and occasional nucleoli. Some mitotic figures and apoptotic cells are seen. HE, $\times 400$.

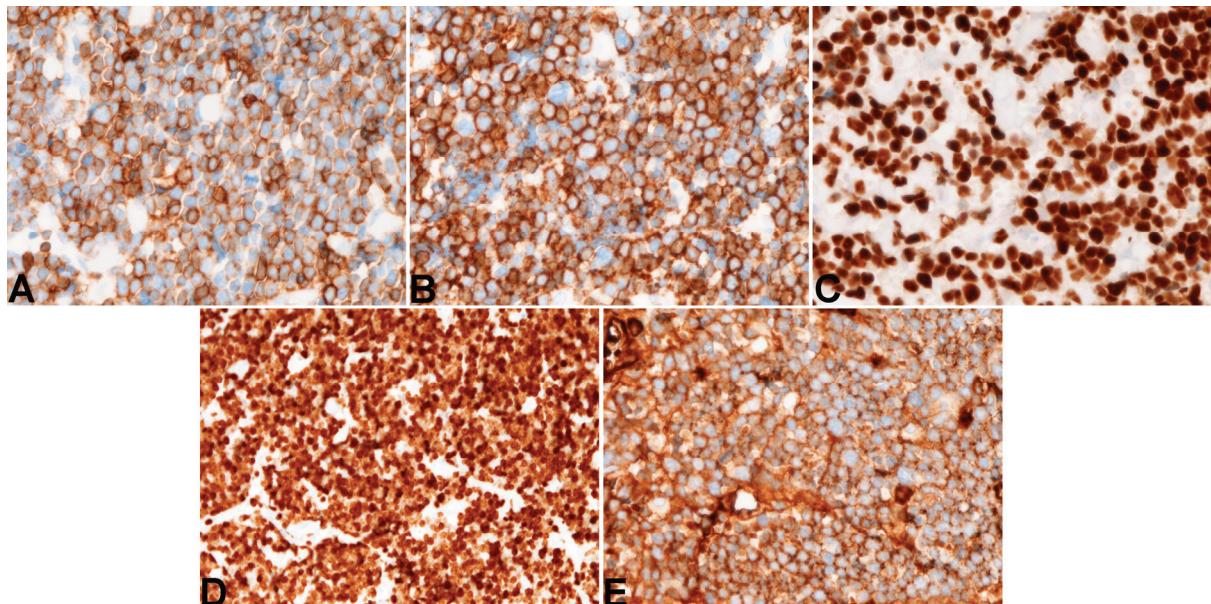


Figure 3. Immunohistochemical features. The tumor cells are positive for CD45 (A) and CD20 (B). They are diffusely positive for p53 protein (95%) (C). The Ki-67 labeling is 100% (D). The tumor cells are positive for λ -chain (E), but not κ -chain. $\times 400$.

protein (100%) and high Ki-67 labeling (100%) support this. The present case is not low grade B-cell lymphomas, such as follicular lymphoma, mantle cell lymphoma, and MALT lymphoma [2]. The present case is not Burkitt lymphoma. The present case is not immunoblastic lymphoma histologically, and also because of negative TdT [2].

The present tumor developed in the subman-

dibular gland. Most cases of salivary gland lymphoma develop in the parotid gland because of its large size and because of the presence of intraparotid lymph nodes [1]. In the largest series [5], of 40 salivary gland lymphomas, only 3 cases were submandibular gland lymphomas. Schmid et al [6] described that of 25 salivary gland lymphomas, 21 were parotid gland lymphomas and 4 were submandibular gland lymphomas. Thus, submandibular gland lymphoma

is very rare. The present case was DLBCL. Dun et al [7] described that of 23 salivary gland lymphomas, 19 were MALT lymphomas, 3 were DLBCL, and 1 was follicular lymphoma. Kojima et al [8] described that of 30 salivary gland lymphomas, 15 were MALT lymphomas, 7 were follicular lymphomas, 6 were DLBCL with MALT lymphoma, and 2 were DLBCL. Therefore, DLBCL of the salivary gland is very rare. The prognosis of salivary gland lymphomas is different among cases, depending on the stage and histological type. Dun et al [7] described that the overall survival and relapse-free survival rates at 5 years were 95% and 51%, respectively; thus salivary gland lymphoma is an indolent disease.

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