Case Reports and Reviews

Case Report

A Case of Primary Malignant Lymphoma of The Tongue

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Abstract

We experienced a very rare case of primary malignant lymphoma of the tongue. The patient was an 83-year-old Japanese woman. Her chief complaint was swelling of the tongue for half a year. Fine needle biopsy from her tongue mass was performed at a previous hospital and showed no malignant cells. This hospital introduced her to our hospital for the purpose of scrutiny.At the first visit to our Department of Otorhinolaryngology, the tongue surface showed no erosion or ulcer, but hard tumors were palpable in the subepithelium of the central region of the tongue. Open biopsy of this tumor was performed and was diagnosed as T-cell lymphoma. PET-CT revealed no metastasis findings and bone marrow biopsy showed abnormal findings. This lymphoma was classified as Stage I (Ann Arbor classification). We consulted the Department of Hematology and she received chemotherapy by hematologists. Two years have passed since the start of chemotherapy. At present, this lymphoma shows no growth in the tongue or whole body.

Introduction

Malignant lymphoma is known to be divided into two types: node lymphoma that arises in the lymph nodes and extra-nodular lymphoma derived from organs and tissues other than lymph nodes.

Some researchers [1-4] reported that extra-nodular malignant lymphoma with initial symptoms in the oral cavity were uncommon. Epstein [1] reported that their occurrence was 3.5% of all oral malignancies. Clark [2] and Freeman [3] described that 3-5% of the extra-nodal lymphomas in the neck and neck arose in the oral and paraoral regions. The frequency of extra-nodular malignant lymphoma in the tongue was exceedingly rare. Primary extra-nodular malignant lymphoma with in the oral cavity would be difficult to be differential diagnosis. Because, it's clinical findings could be similar to inflammatory and other types of tumors [1,5]. We experienced a case of primary malignant lymphoma of the tongue. This case report presents the clinical findings, blood examinations, image examinations, histopathologic examination, treatment course and some considerations.

Case report

An 83-year-old Japanese woman presented the chief complaint of swelling of the tongue for half a year. A referring hospital tried fine needle biopsy for her tongue mass, but this histopathologic finding showed no malignant cells. This hospital introduced her to our hospital for the purpose of scrutiny. At first, in the examination by our Department of Otorhinolaryngology, there were palpable hard tumors in the sub epithelium of the central region of the tongue, but no erosion or ulcer on the tongue surface was observed (Figure1). Open biopsy of her tongue tumor was performed. In the histopathologic findings, H&E staining showed proliferating small lymphocytes in the subepithelial stroma but there was no solid cancer. CD3 and CD8 makers of T lymphocytes were positive immunohistochemically (Figure2). PET-CT showed no metastasis findings and bone marrow biopsy showed abnormal accumulation findings (Figure 3). Based on the above examinations, her lymphoma was diagnosed as T cell lymphoma and classified as Stage I (Ann Arbor classification). We consulted the Department of Hematology and she was administrated chemotherapy for 14 months by hematologists. In the present case, the chemotherapy was stopped because of severe pain on the tongue due to complications. Two years have passed since the start of chemotherapy and her lymphoma remains unchanged the range of the tongue lesions, and shows no growing to whole body.

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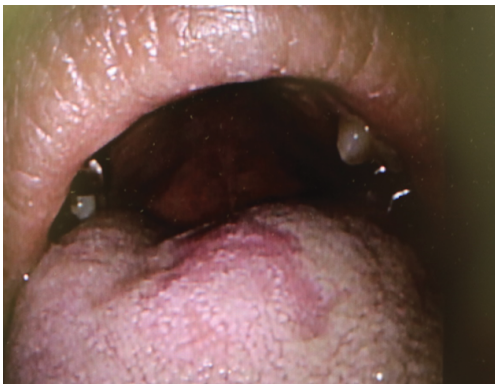

Figure 1. *At the first visit of our department.*
The hard tumors in subepithelium were palpable at the central region of the tongue (*), but the tongue surface showed no erosion or ulcer.

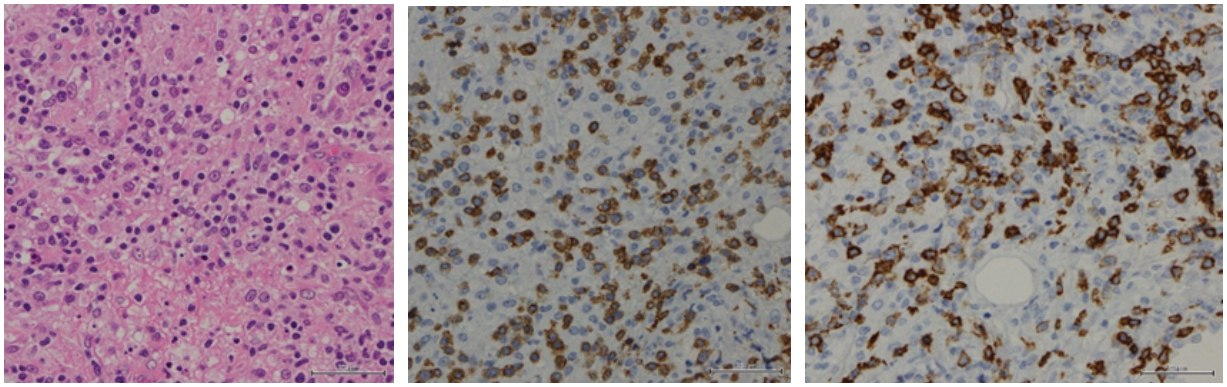

Figure 2. *The histopathologic findings of the open biopsy of the tongue H&E (X400Left) staining showed proliferating small lymphocytes in the subepithelial stroma but no solid cancer. And both CD3 (X400, Middle) and CD8 (X400, Right) markers of T lymphocytes showed a positive reaction immunohistochemically.*

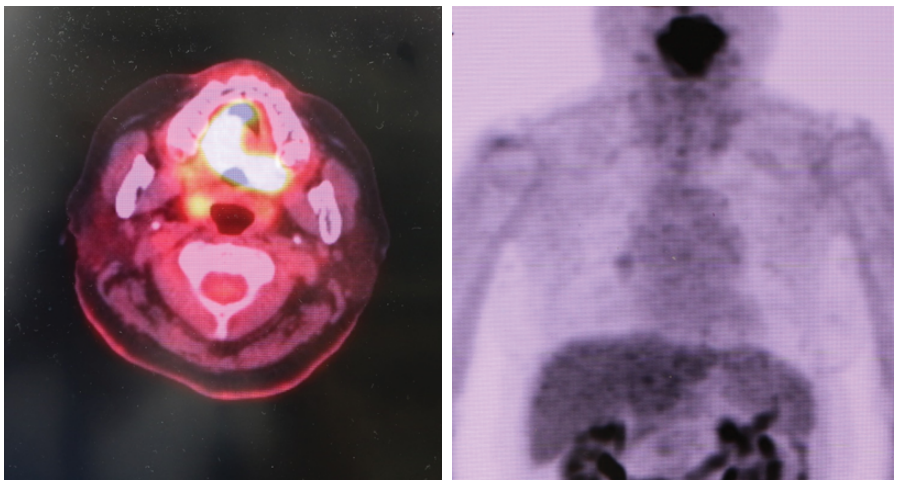

Figure 3. *PET-CT showed abnormal accumulation at the tongue, but no whole body abnormality accumulation could be observed.*

Discussion

Singh [6] described that malignant lymphoma could be divided into Hodgkin‘s disease and non-Hodgkin`s lymphoma. Non-Hodgkin`s lymphoma comprises approximately 5% of head and neck malignancies and shows a wide range of appearances comparable with Hodgkin's disease. Hodgkin's disease and non-Hodgkin`s lymphoma are seen in the head and neck region, but extranodal disease, with or without lymph node involvement, was more common among non-Hodgkin`s lymphoma patients. Clark [2] noted that extranodal lymphomas in the neck occurred in the oral and paraoral region, mainly from Waldeyer's ring. But tongue lesions are noted in the literature to be extremely rare. Moreover, extranodal T lymphoma of the tongue as our case is even rarer than B cell lymphomas. In Japan, Sugiyama [5] reviewed 125 extranodal malignant lymphomas with initial symptoms in the oral cavity. The primary site with the highest incidence in the head and neck was the gingiva (47.2% (59/125) and in the tongue it was 4.8% (6/125). The incidence of non-Hodgkin's lymphoma was 94.7%. In histologic type, B cell lymphoma and T cell lymphoma, the incidence was 84.8% (106/125) and 14.4% (18/125), respectively. Concerning the staging classification, Eisenbud [4] reviewed 31 cases with oral presentations in non- Hodgkin's lymphoma and found that 58.0% (18/31) were in stage I or II. Kawamata [7], in a review of 140 Japanese cases, described that most of the cases were stage I (approximately 40%). The treatments for extra-nodular malignant lymphoma in the oral cavity have generally been chemotherapy or radiotherapy [4,5,7]. Our case with stage I underwent chemotherapy for 14 months before being stopped because of severe pain, as mentioned above. Two years have passed since the start of chemotherapy and her lymphoma has remained unchanged in the tongue lesions and has shown no growth in the whole body.

Some reports have [5,7,8] mentioned that primary extra-nodular malignant lymphoma of the oral cavity could be difficult to diagnose in the differential diagnosis because of the many various symptoms. Anifin [8] described that the most reported symptom of oral cavity lymphoma was local, painless swelling with or without ulceration, though they could present in various ways and mimic other diseases. In Kawamata's [7] review of 140 Japanese cases, he noted the difficulty in diagnosing malignant lymphoma of the oral regions, with 16.4% (23/140) of the cases requiring multiple biopsies to make the diagnosis. In particular, the cases with ulcer or necrosis could not be definitely diagnosed. Therefore, it was necessary to aggressively or repeatedly biopsy lesions that were difficult to diagnose. Practical Guidelines of Hematological Malignancies [9] described that needle biopsy for histopathologic diagnosis of malignant lymphoma was often insufficient. In our case, needle biopsy at the referral hospital did not lead to a diagnosis. Open biopsy in our department was successful by adding immunohistochemistry. Eisenbud [4] described that some cases might be difficult to distinguish a benign lymphoproliferative response from a true malignant lymphoma. In addition, it was difficult at the times to discriminate between a poorly differentiated carcinoma and malignant lymphoma by light microscopy. Immunologic techniques could prove to be helpful in such cases. Clarke [2] also recommended immunohistochemistry, which would be useful not only for the diagnosis, but also for classifying such lesions.

Conclusion

We experienced a very rare case of primary malignant lymphoma of the tongue. The cases with ulcer or necrosis could not be definitely diagnosed. Some cases have been reported to be difficult to distinguish a benign lymphoproliferative response from a true malignant lymphoma. In addition, it was difficult at the times to discriminate between a poorly differentiated carcinoma and malignant lymphoma by light microscopy. Immunologic techniques could prove to be helpful in such cases.

Author Contributions

Takeshi Kusunoki: diagnosis and therapy, composition of this manuscript. Ryo Wada: pathological diagnosis.

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Ethics and Informed Consent Statement

We have protected the patient`s anonymity and obtained consent for publication of the clinical findings, image examination and pathological examination.

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Conflicts of Interest

The authors declare no conflict of interest.

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