

Non-Hodgkin's Lymphoma in the Parotid Gland Similar to Benign Lymphoepithelial Lesion: A Case Indonesian Male

Muhammad Arif Sudianto Utama¹, Widodo Ario Kentjono²

¹Resident, ²Assistant Professor, Department of Otorhinolaryngology - Head and Neck Surgery, Faculty of Medicine, Universitas Airlangga - Dr. Soetomo General Academic Hospital, Surabaya 60286, Indonesia

Abstract

Background: Primary parotid non-Hodgkin's lymphoma (NHL) is a very rare case. **Case presentation:** A 66-year-old Indonesian male with a complaint of a lump in front of the right ear about 1 year with a history of postoperative primary parotid NHL 3 years ago. Patients underwent Fine Needle Aspiration Biopsy (FNAB) examination, showing a distribution of lymphoid cells with a diverse population (benign lymphoepithelial lesion / BLL) different from the results of FNAB 3 years ago. The patient underwent a right superficial parotidectomy, and the results of the histopathological examination revealed mature lymphocyte cells mixed with histiocytes, forming foci of the follicle in part with an active germinal center. **Conclusion:** NHL reports are necessary to minimize misdiagnosis between primary parotid NHL and BLL.

Keywords: non-Hodgkin's lymphoma, benign lymphoepithelial lesion, parotidectomy

Introduction

Non-Hodgkin lymphoma (NHL) is found in 86% of all lymphoma cases, with a rough estimation of 90% covering B cell lymphoma. About 2-5% of primary NHL occurs in the head and neck region, one of which is in the parotid gland. Parotid gland NHL cases are very rare, limited to small and closed case series reports. Primary parotid gland NHL accounts for only 0.5-0.87% of all NHL cases, with increasing prevalence in the last few decades [1,2].

The diagnosis of primary parotid gland NHL in patients is difficult because of the extremely rare number of cases. In NHL case, lymphatic nodules are generally

biopsied, whereas computed tomography (CT scan) / magnetic resonance imaging (MRI) only informs about the origin of the malignancy and extraparotid expansion. To date, there have been no pathognomonic findings that indicate lymphoma on CT or MRI scans [3-5]. The purpose of reporting this case is to inform that there are similarities between the diagnosis of primary parotid gland NHL and benign lymphoepithelial lesion (BLL).

Case Presentation

A 66-years-old Indonesian male with complaints of a lump in front of the right ear about 1 year. The lump is enlarged slowly. The patient had surgery for the right parotid gland tumor 3 years ago. The results of the Fine Needle Aspiration Biopsy (FNAB) at that time showed the distribution of lymphoid cells of various maturity with some forming germinal centers. Visible groups of salivary gland epithelial cells also showed BLL.

The results of histopathological examination of postoperative tissue showed pieces of tissue consisting of dense proliferating lymphoid cells, small to medium sized, monotonous, smooth chromatin, diffused narrow cytoplasm, infiltrative between salivary glandular tissues. There were still follicles with germinal centers among them. Mitosis was difficult to find, and there

Corresponding author:

Muhammad Arif Sudianto Utama

Department of Otorhinolaryngology – Head and Neck Surgery, Faculty of Medicine, Universitas Airlangga – Dr. Soetomo General Academic Hospital, Jl. Mayjen Prof. Dr. Moestopo No. 6-8, Airlangga, Gubeng, Surabaya, East Java 60286, Indonesia.

Phone: +6231-5501649 / +628113530889

Mail: arf.utamaa@gmail.com

Orcid id: 0000-0002-1378-089X

was a normal salivary gland remained at the edge. A low-grade lymphoma was found in accordance with the description of lymphoepithelial sialadenitis (Mikulicz disease). Advice from anatomic pathology to ensure immunohistochemical examination in this preparation. Immunohistochemical results showed positive CD20 in lymphocyte cells surrounding the epimyoeipithelial island, positive CD3 in T cells between follicles, and Ki67 was limited to germinal center. In accordance with the description of benign lymphoepithelial lesion (Mikulicz disease).

Physical examination found no abnormalities in ear, nose, and throat, as well as no cervical lymph node enlargement. The parotid gland had a mobile mass with a size of about 3x2x2 cm, dense and springy, flat surface, no tenderness. Intact right facial nerve (House-Brackmann scale grade I). Physical examination of axillary and inguinal glands did not reveal a mass (Fig. 1). Laboratory test results were normal, serological hepatitis B virus and negative hepatitis C virus.

FNAB showed smears with diverse lymphoid cells distribution. Unclear epithelial cells; showed lymphoid hyperplasia. Tumor excision was recommended for histopathological and immunohistochemical confirmation if necessary, because it had not been able to get rid of low-grade lymphoid neoplasms. MRI examination with contrast obtained a solid mass of size 5.4x1.7x3.9cm; indistinct border of irregular edge in the right parotid region. Contrasting obtained heterogenous contrast enhancement. The lesion appeared attached to the right masseter muscle. The conclusion supported the picture of benign parotid mass of the right muscle (Fig. 2).

Based on available data, a BLL type of parotid tumor was established. A week after the diagnosis, a right superficial parotidectomy surgery was performed with Blair's modified incision. The skin incision was deepened into the subcutaneous tissue and platelet

musculature to identify and preserve the right facial nerve branches, then proceeded with tumor removal (Fig. 3). The size of the tumor obtained was about 40 cm³. The tumor tissue was sent to the anatomic pathology installation for histopathological examination (Fig. 4).

The results of post-operative histopathological examination showed extensive infiltration of lymphocytic cells surrounding the remaining glands and ducts that were pinched to form epimyoeipithelial nests. Lymphoid tissue consisted of mature lymphocyte cells mixed with histiocytes, forming foci of the follicle with some active germinal centres. This description supported Mikulicz disease or lymphoepithelial sialadenitis, concluded Lymphoepithelial sialadenitis/BLL (Mikulicz disease).

Furthermore, the patient was recommended to have immunohistochemical examination. The results showed negative CD3 in tumor cells, positive in mature lymphocyte cells, positive CD20 in diffuse and condense on tumor cell membranes, and Ki67 etched 505 in tumor nuclei, immunoarchitecture supported non-Hodgkin lymphoma B cell type which could come from MALT lymphoma.

The results of the data obtained concluded that the patient was diagnosed having a right parotid NHL, postoperatively obtained right facial fascia (House-Brackmann scale II-III) which was subsequently recovered with physiotherapy, especially the submandibular marginal branches and right buccal. Subsequent therapy included chemotherapy with a combination regimen according to NHL for 6 cycles. Control in the second month after surgery showed that the function of right facial fascia nerve had begun to recover, although it still looked a little paresis (House-Brackmann scale grade II), especially the right lip corner. Control in the fifth month after surgery did not show any residual mass on the right parotid, the right facial nerve function was visible again (House-Brackmann scale grade I).



Figure 1. Physical examination shows a mass in the right parotid gland

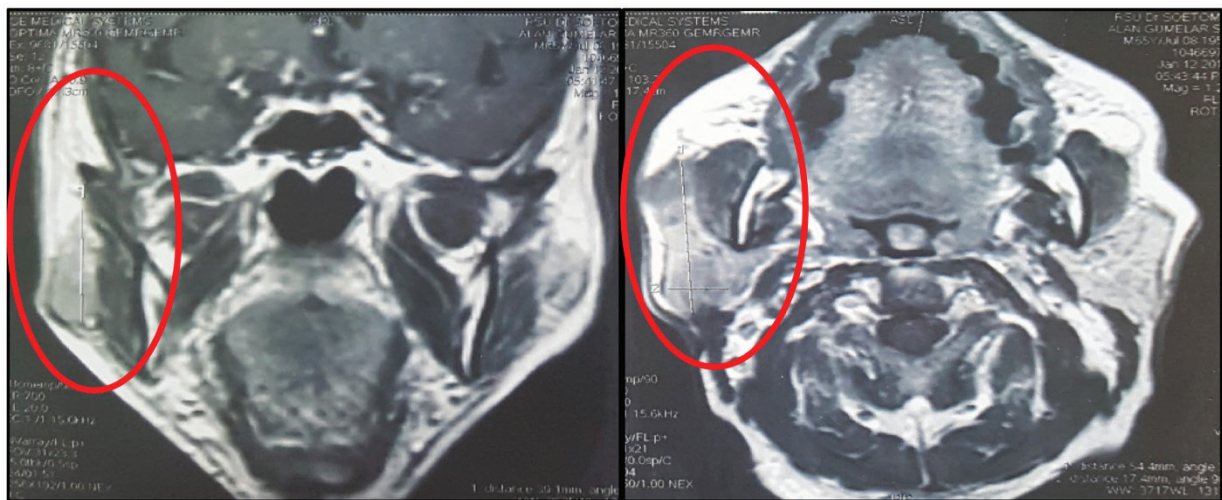


Figure 2. MRI with contrast shows indistinct solid borderline lesions, irregular margins in right parotid region

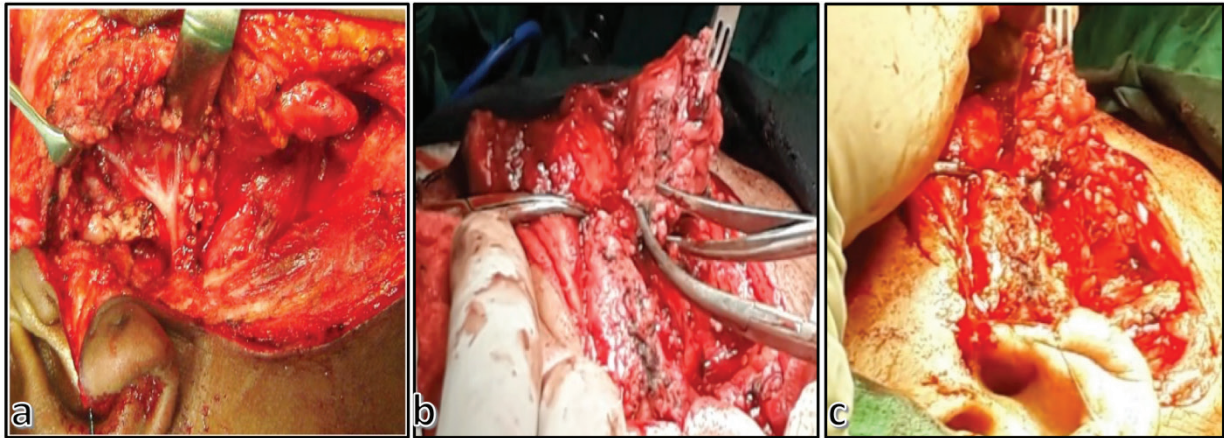


Figure 3. Surgery performed with superficial parotidectomy approach of the right parotid gland (a), preservation of the right N. Facial fascia (b), followed by tumor removal (c).

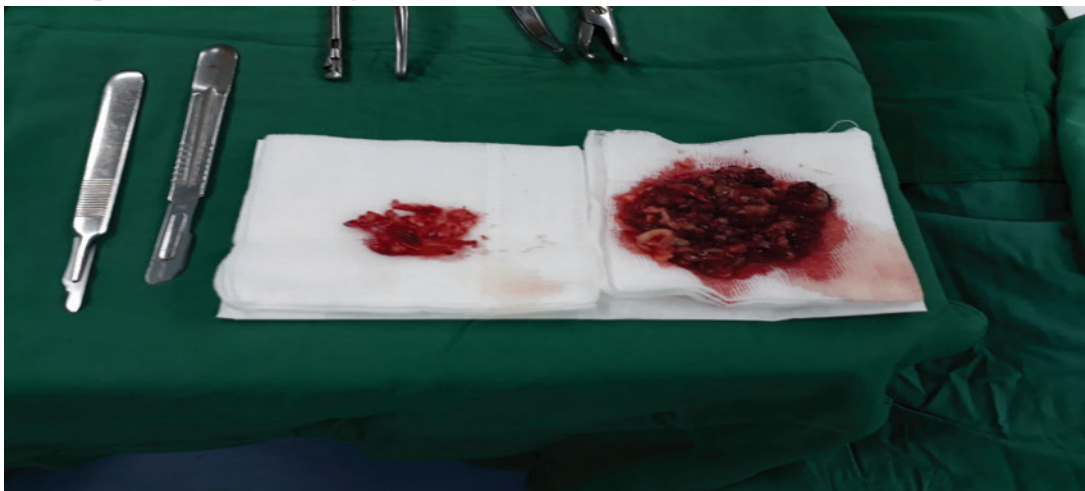


Figure 4. Mass of the right parotid gland after superficial parotidectomy.

Discussions

In this case, a CT scan was not performed but an MRI examination was immediately performed. A solid border lesion was found with an irregular border in the right parotid region. A heterogeneous contrast enhancement appeared; the lesion was attached to the right masseter muscle. The radiological features of parotid gland NHL were rarely well described. According to Corr et al., who presented a cohort study of 10 HIV-infected children with parotid gland NHL, a CT scan of the lesion contained a muted hypoechoic solid nodule, which was similar to hyperplastic lymphoid tissue or lymphoma. These lesions are cystic (from compression of the parotid duct terminal with adjacent hypoplastic or neoplastic lymphoid tissue) and separate with calcification, both intracystic and parenchymal can coexist. These radiological features have been described

in the BLL found in patients with AIDS or Sjögren's syndrome [4,6-8].

An FNAB examination had been conducted twice, with first examination showing a Mikulicz disease. The second test was carried out three years ago because a lump appeared again in the same place, and the conclusion was a lymphoid hyperplasia. The results of this FNAB indicated the origin of a malignancy that leads to NHL, as this finding is consistent with the literature. Preoperative FNAB from a suspected tumor and radiological examination in certain cases is part of the preoperative examination of parotid gland lesions. Parotid gland NHL is difficult to diagnose through FNAB cytology. FNAB examination for head and neck region NHL is the subject with the highest error rate of all FNABs in the head and neck, with a false-negative rate of 32%. Although FNAB can definitively diagnose

lymphoma in some patients, tissue biopsy has a much higher accuracy [4,6,7,9].

A parotid surgery was conducted with a superficial parotidectomy approach with the aim of the diagnostic process and the therapeutic process. Patients with localized parotid gland tumors, in general, is highly recommended for surgical or surgical management to diagnose parotid gland NHL because histological evaluation is very important to treat NHL. Parotid surgery is recommended for therapy as well as to confirm histological diagnosis and further follow-up planning. The prognosis of patients with parotid gland NHL so far will be good if diagnosed immediately. Limited data shows that 80% of patients have a five-year survival rate [3,4,6,7].

This case, after being diagnosed as NHL type MALT parotid gland, immediately performed chemotherapy with NHL-compliant regimens. The patient was followed during chemotherapy and no residual mass was found after control for 6 months, showing an excellent therapeutic response. Diagnosis of NHL must involve an in-depth evaluation of the involvement and spread of the same disease must be carried out in other locations before starting local therapy. Radiotherapy and chemotherapy must be considered after surgery to eradicate spread or residue. Irradiation in cases of lesions localized in the early stages and chemotherapy in the advanced disease stage. Isobe et al., treated 37 patients with stage 1E extragastric MALT LNH with only radiotherapy, and around 97.3% of patients received an increase in the number of free survival rates over a three-year period of 91.9%. So far chemotherapy in patients with NHL is still a very sensitive therapy using the CHOP protocol (cyclophosphamide, doxorubicin, vincristine, and prednisone) [2,3,6,10].

This case has no lymph nodes or involvement of other organs, but only limited to the history and physical examination of the axillary and inguinal glands. This indicates that this NHL case is not a systemic type and is limited to the right parotid gland or primary to the parotid gland. Salivary gland NHL tends to be more aggressive and can occur in regional or distant lymph nodes in other organs. According to Wenzel et al., therapy with local surgery of patients with head and neck NHL has a relatively high risk of experiencing initial spread or

recurrence [2,5,10].

Conclusions

A very rare and suspicious parotid NHL case has been reported as a transformation from BLL or Mikulicz disease. Primary and secondary NHL in the parotid gland are clinically indistinguishable from benign tumors or other malignant lesions. The imaging modality and cytology of FNAB does not always help, so the majority of patients require parotidectomy for diagnosis and therapy.

Ethics Statement: The present case report adhered to the Declaration of Helsinki. Informed consent for publication was obtained from the patient.

Conflict of Interest: The authors report no conflict of interest in this publish.

Author's Contribution: All authors contributed toward data analysis, drafting and revising the paper, gave final approval of the version to be published and agree to be accountable for all aspects of the work.

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