Int J Clin Exp Pathol 2016;9(7):7696-7699
www.ijcep.com /ISSN:1936-2625/IJCEP0023995

Case Report

Marginal zone lymphoma with prominent lymphoepithelial sialadenitis in heterotopic salivary gland tissue

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Received January 13, 2016; Accepted May 20, 2016; Epub July 1, 2016; Published July 15, 2016

Abstract: This is the first reported case of a marginal zone lymphoma with prominent lymphoepithelial sialadenitis of heterotopic salivary gland tissue. A 67-year-old female patient presented neck masses. Neck 3-dimensional computed tomography suggested Sjögren's syndrome in both parotid glands and also revealed several cervical lymph nodes enlargement. Histologic findings of superficial parotid glands were consistent with marginal zone lymphoma with prominent lymphoepithelial lesions. Several paraparotid and infraparotid lymph nodes showd morphologic features similar to parotid glands with prominent lymphoepithelial sialadenitis.

Keywords: Lymphoma, marginal zone, sialadenitis, heterotopia

Introduction

Heterotopic salivary gland tissue (HSGT) has been found in the head and neck including cervical lymph node [1, 2]. Benign and malignant tumors of salivary glands can arise from HSGT. The most common neoplasm reported is Warthin's tumor [2]. Rarely, salivary inclusions of the lymph node revealing lymphoepithelial sialadenitis (LESA) in the background of malignant lymphoma have been reported [3]. Herein, we report an unusual case of marginal zone lymphoma with prominent LESA of HSGT in the cervical lymph node.

Case report

A 67-year-old female was admitted because of a right neck mass present for one year. She received a total thyroidectomy due to malignancy 19 years ago. She had suffered from dry eye and dry mouth for several years. However, serologic studies were not performed. Neck 3-dimensional (3D) computed tomography (CT) showed diffuse heterogeneities in both parotid glands, suggesting an autoimmune disease, such as Sjögren's disease. Neck 3D CT also revealed a few intraparotid enhancing lesions with perilesional infiltration in both superficial lobes and enlargement of several cervical lymph nodes. Therefore, bilateral superficial parotidectomy and right modified radical neck dissection were performed.

Histologic sections of both superficial parotid glands showed a monotonous lymphoid infiltrate with prominent lymphoepithelial clusters comprised of ductal epithelium and lymphocytes. Lymphocytes were small to medium-sized cells with pale to clear cytoplasm. They expressed CD20, CD43 and bcl-2. This case was diagnosed as marginal zone lymphoma with LESA feature. Several paraparotid and infraparotid lymph nodes revealed histologic features similar to parotid glands with prominent LESA (**Figure 1**). Nodular configuration with the presence of peripheral encapsulation (**Figure 2A**) and subcapsular sinus (**Figure 2B**) but the absence of multilobular architecture was considered as a nodal-based disease. Monoclonal immunoglobulin heavy chain (IgH) gene rearrangement was identified by BIOMED-2 multiplex polymerase chain reaction (PCR) assays (**Figure 3**). Remaining cervical lymph nodes had lymphomatous involvement without salivary gland inclusions.

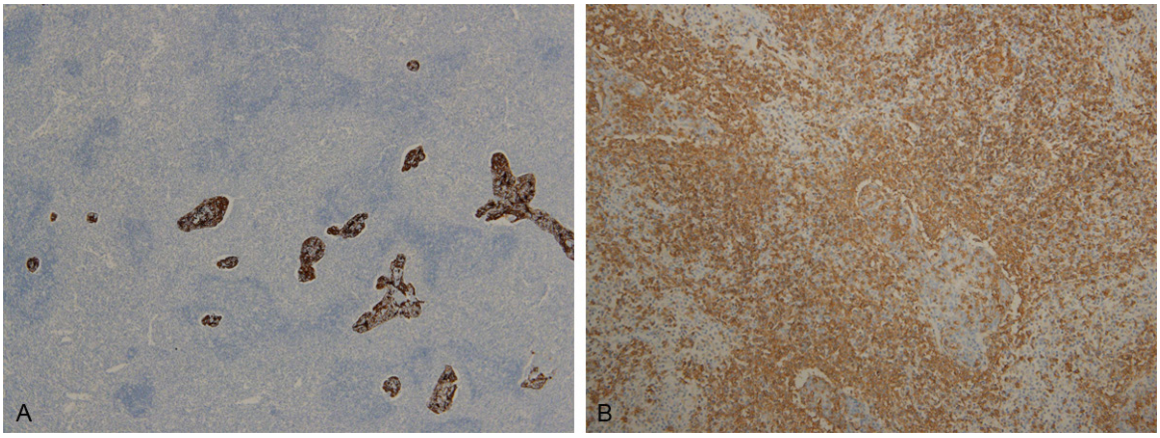

Figure 1. Immunohistochemical staining of cytokeratin revealing prominent lymphoepithelial lesions (A) and small to medium-sized lymphoma cells positive for CD20 (B).

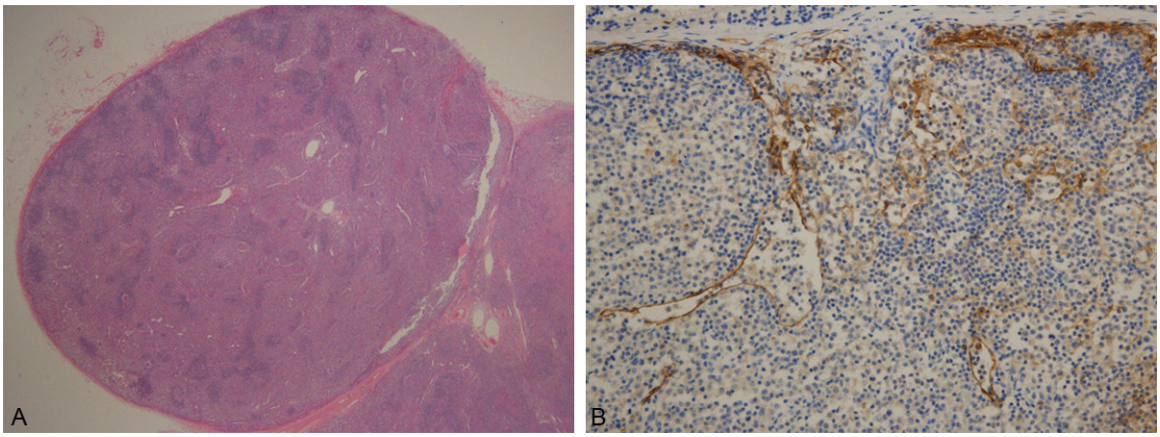

Figure 2. Nodular configuration with discrete fibrous capsule considered as lymph node-based lesion (A) and subcapsular sinus ascertained by immunohistochemical staining for D2-40 (B).

Discussion

HSGT, also known as ectopic or choristomatous salivary glands, consists of salivary gland tissue outside of the major and minor salivary glands [1]. This rare congenital lesion typically can present as a draining sinus and/or asymptomatic nodule of the neck along the low anterior sternocleidomastoid muscle [1]. It also has been reported in numerous locations of the head and neck including the pituitary, middle and external ear, mandible, gingiva, hypopharynx, thyroglossal duct, capsule of the thyroid and parathyroid glands, cervical lymph node [2, 4], mediastinum [1] and rectum [1]. HSGT is susceptible to the same disorders of the major and minor salivary glands, including infectious, inflammatory, and neoplastic diseases [2]. Both benign and malignant neoplasms can occur in HSGT [2]. Most common neoplasm is Warthin's tumor (WT) [2], followed by pleomorphic adenoma [4].

WT is a benign salivary gland neoplasm composed of cystic or glandular spaces, which are lined by oncocytic epithelium, and lymphoid stroma [5]. The most popular hypothesis for the pathogenesis of WT is neoplastic proliferation of heterotopic salivary ducts that are present in preexisting intrapatotid or parapatotid lymphoid tissue. WT may be a lesion of the lymph nodes rather than the salivary glands [6]. Uncommonly, lymphoid stroma in WT can be infiltrated by malignant lymphoma [5] that may represent secondary proliferative and metaplastic changes of salivary inclusions in lymph nodes affected by the lymphoma. A review of the English literature identified 28 cases of WT

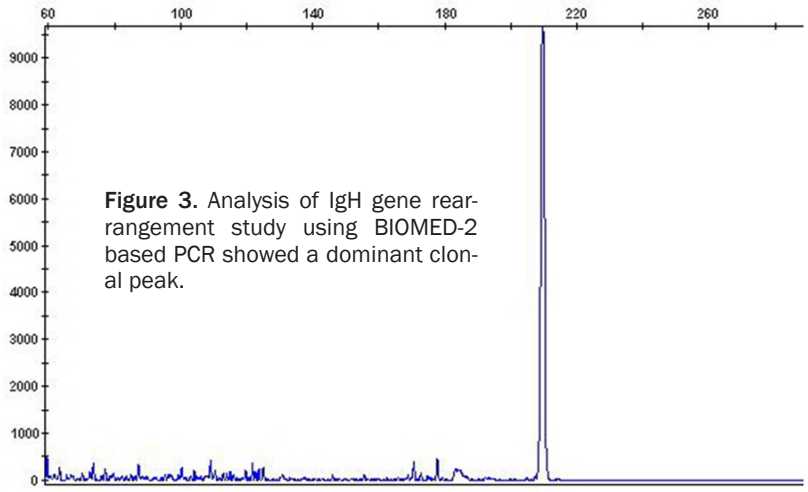

Figure 3. Analysis of IgH gene rearrangement study using BIOMED-2 based PCR showed a dominant clonal peak.

related to malignant lymphomas [7-10]. Among them, four cases arose in cervical lymph nodes. They might be WT arising from HSGT in lymph nodes affected by malignant lymphomas. In these cases, it is possible that the presence of WT in lymph nodes might have provided a source of long-term antigenic stimulation, thus predisposing patients to the development of malignant lymphoma [11]. In 28 cases of WT related to malignant lymphomas, only one case of marginal zone lymphoma (MZL) was identified. It was present in the parotid gland [12].

Recently, eight cases of Hodgkin lymphomas (HLs) have been reported in intraparotid lymph nodes with minimal or no involvement of parenchyma. Six cases revealed salivary inclusions within lymphoma. Among them, one case revealed numerous lymphoepithelial lesions, reminiscent of LESA [3]. However, it did not show WT like oncocytic changes, similar to our case. Those findings suggested an inherent tendency, especially of HL, to induce secondary lymphoepithelial proliferation of HSGT through a tendency for epitheliotropism or through as yet unclear immunological mechanisms [3]. However, other histologic types of malignant lymphomas other than HLs have not yet been reported.

MZL of salivary gland usually arises in a setting of LESA and a long-term inflammatory stimulus, such as in Sjögren's syndrome, where chronically activated B-cells that formed LESA, could be an initial event in the lymphomagenesis [12]. Therefore, Sjögren's syndrome might be a cause of malignant lymphomas in HSGT containing lymphoid organs in our case, leading to marginal zone lymphomagenesis through LESA.

Disclosure of conflict of interest

None.

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References

[1] Haemel A, Gnepp DR, Carlsten J, Robinson-Bostom LJ. Heterotopic salivary gland tissue in the neck. Am Acad Dermatol 2008; 58: 251-256.

[2] Cannon DE, Szabo S, Flanary VA. Heterotopic salivary tissue. Am J Otolaryngol 2012; 33: 493-496.

[3] Agaimy A, Wild V, Märkl B, Wachter DL, Hartmann A, Rosenwald A, Ihrler S. Intraparotid classical and nodular lymphocyte-predominant Hodgkin lymphoma: pattern analysis with emphasis on associated lymphadenoma-like proliferations. Am J Surg Pathol 2015; 39: 1206-1212.

[4] Vegari S, Naderpour M, Hemmati A, Baybordi H. Pleomorphic adenoma of the cervical heterotopic salivary gland: a case report. Case Rep Otolaryngol 2012; 2012: 470652.

[5] Giaslakiotis K, Androulaki A, Panagoulias G, Kyrtsonis MC, Lazaris AC, Kanakis DN, Patsouris ES. T cell lymphoblastic lymphoma in parotidectomy for Warthin's tumor: case report and review of the literature. Int J Hematol 2009; 89: 359-364.

[6] Gorai S, Numata T, Kawada S, Nakano M, Tamaru J, Kobayashi T. Malignant lymphoma arising from heterotopic Warthin's tumor in the neck: case report and review of the literature. Tohoku J Exp Med 2007; 212: 199-205.

[7] Chu CY, Pan SC, Chang KC. EBV-positive diffuse large B-cell lymphoma of the elderly involving Warthin tumor. Pathol Int 2015; 65: 677-679.

[8] Liu YQ, Tang QL, Wang LL, Liu QY, Fan S, Li HG. Concomitant lymphocyte-rich classical Hodgkin's lymphoma and Warthin's tumor. Oral Surg Oral Med Oral Pathol Oral Radiol 2013; 116: e117-120.

[9] Di Napoli A, Mallel G, Bartolazzi A, Cavalieri E, Becelli R, Cippitelli C, Ruco L. Nodular lymphocyte-predominant Hodgkin lymphoma in a Warthin tumor of the parotid gland: A case report and literature review. Int J Surg Pathol 2015; 23: 419-423.

[10] Ozkök G, Taşlı F, Ozsan N, Oztürk R, Postacı H. Diffuse large B-cell lymphoma arising in Warthin's tumor: Case study and review of the literature. Korean J Pathol 2013; 47: 579-582.

[11] Park CK, Manning JT Jr, Battifora H, Medeiros LJ. Follicle center lymphoma and Warthin tumor involving the same anatomic site. Report of two cases and review of the literature. Am J Clin Pathol 2000; 113: 113-119.

[12] Marioni G, Marchese-Ragona R, Marino F, Poletti A, Ottaviano G, de Filippis C, Staffieri A. MALT-type lymphoma and Warthin's tumour presenting in the same parotid gland. Acta Otolaryngol 2004; 124: 318-323.