Marginal Zone B-cell Lymphoma Arising from Buccal Mucosa Resembling Inflammatory Myofibroblastic Tumor of the Soft Tissue

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Received: 19 March 2008 / Accepted: 13 May 2008 / Published online: 10 June 2008 © The Author(s) 2008

Abstract We report here a case of marginal zone B-cell lymphoma (MZBL) arising from buccal mucosa resembling inflammatory myofibroblastic tumor (IMT) of the soft tissue. On a low-power field, the lesion is characterized by fibrous granulation tissue and numerous lymphoid follicles with or without atrophic small germinal center. A portion of the lymphoid follicles were surrounded by partial and/or complete thin pale cuff of centrocyte-like (CCL) cells on high power field. The thin rim of the lymphoid follicles contained CCL-cells, plasma cells, cells showing plasma cell differentiation and mature eosinophils. In contrast, the granulomatous areas contained were characterized by haphazardly arranged spindle cells, mature plasma cells, mature eosinophils, small-to-medium lymphocytes and histiocytes. Histologically, IMT was suspected. However, flow cytometry and immunohistochemical study demonstrated a monotypic nature of centrocyte-like cells, plasma cells and their precursor and confirmed the diagnosis of MZBL arising from the buccal mucosa. The differential diagnostic problems between IMT of the soft tissue and classical Hodgkin lymphoma and T-cell lymphoma have been discussed previously. However, the present case indicated that MZBL should be added to the differential diagnosis of IMT of the soft tissue.

Keywords Marginal zone B-cell lymphoma · Buccal mucosa · Inflammatory myofibroblastic tumor · Immunohistochemistry · Flow cytometry

Introduction

Although the pathologic diagnosis of lymphoma traditionally depends on the major criteria of cellular cytologic atypia and monomorphism, marginal zone B-cell lymphomas (MZBL) occasionally lack significant cytological atypia [1]. Moreover, polymorphous lymphoid infiltrate of mature plasma cells, eosinophils, macrophages and centrocyte like (CCL)-cells is sometimes found in the MZBL [1]. Occasionally, plasma cell differentiation has been noted in various degrees in MZML, and the plasma cells may occasionally obscure the CCL-cells [1].

We report here a unique case of MZBL arising from the buccal mucosa showing prominent plasma cell differentiation and associated with and fibroblastic proliferation and tissue eosinophilia whose histological findings resembled inflammatory myofibroblastic tumor (IMT) of the soft tissue.

Case Report

A 78-year-old Japanese woman presented with a two-month history of left buccal mucosa mass measuring 2 cm in
diameter. Physical examination was noncontributory. Laboratory data on admission including peripheral blood count, liver function tests and serum immunoglobulin level were all within normal limits. No paraproteinemia was noted preoperatively. Clinically, lobular capillary hemangioma was suspected due to the gross appearance of after resection. Initially, IMT of the soft tissue was suspected due to the histological findings. However, by the information from flow cytometry of operatively resected specimen, a distinct clonal population of B-cells were identified which comprised 76% of the total sample and expressed CD19, CD20, CD38 and were lambda light chain restricted. The clonal population of B-cells were negative for CD5, CD10 and CD56. The patient did not receive any medication after tumorectomy because the tumor was completely resected and there was no other evidence of disease. The patient is currently alive and disease-free at the last follow up five months postoperatively.

On low-power field, the lesion was characterized by fibrous granulation tissue and numerous lymphoid follicles with or without atrophic small germinal centers (Fig. 1a). Some of the lymphoid follicles were surrounded by partial and/or complete thin pale rim of cells with clear cytoplasm (Fig. 1a). On high power field, the fibrous granulation tissue contained numerous haphazardly arranged spindle cells, mature plasma cells, mature eosinophils, small-to-medium lymphocytes and histiocytes (Fig. 1b). The spindle cells usually had plump nuclei with small but conspicuous nucleoli (Fig. 1b). However, there were no mitotic figures. A few binucleated plasma cells or cells containing numerous intracytoplasmic immunoglobulins (Russell bodies) were observed. However, there were no plasma cells containing intranuclear inclusion bodies (Dutcher bodies). Vascular proliferation was not prominent. The thin rim of lymphoid follicles composed of small to medium lymphocytes, mature plasma cells, cells showing plasma cell differentiation and mature eosinophils. Some of the medium-sized lymphocytes had round or slightly indented nuclei with small conspicuous nucleoli and a moderate amount of clear cytoplasm (centrocyte-like cells) (Fig. 1c) [1].

There were no eosinophilic microabscesses or Warthin-Finkeldy type polykaryocytes in the lesion.

Immunohistochemistry was performed on paraffin sections using a Ventana automated (BenchMarkTM) stainer. The spindle cells were vimentin+, desmin-, muscle specific actin-, cytokeratin-, P80-, CD3 and CD20 immunostain demonstrated the mixed nature of lymphocytes in the thin rim of the lymphoid follicles. A portion of the lymphoid follicles contained a small number of CD10+ germinal center cells. The majority of small lymphocytes of lymphoid follicles were CD20+, slgM+, slgD+, CD3-, CD5-, CD10-, CD23+, CD43+, bcl-2+. A cyclin D1immunostain was negative ruling out the mantle cell lymphoma. There were no CD5+, CD23+, CD43+, Cyclin D1+ B-cells in the thin rim of the lymphoid follicles. The majority of mature plasma cells in the fibrocollagenous

Fig. 1 (a) Low-power field of the lesion. The process was composed of fibrous granulation tissue and numerous lymphoid follicles with or without atrophic small germinal centers. Note a lymphoid follicle surrounded by a complete thin pale rim of cells with clear cytoplasm (+) (HE × 10). (b) High-power field of the fibrous granulation tissue. There were numerous haphazardly arranged spindle cells, mature plasma cells, eosinophils, small-to-medium lymphocytes and histiocytes. The spindle cells usually had plump nuclei with small but conspicuous nucleoli (HE × 100). (c) High-power field of the thin rim of the lymphoid follicles. These were composed of small lymphocytes, CCL-cells, plasma cells, cells with plasma cell differentiation and mature eosinophils. HE × 250
tissue had intracytoplasmic lambda light chain (Fig. 2a and b). Moreover, in the thin rim of the lymphoid follicle, plasma cells, cells showing plasma cell differentiation and a portion of CCL-cells also demonstrated monotypic intracytoplasmic lambda light chain (Figs. 2a–d).

There was germinal center deposition of IgE in a portion of lymphoid follicles.

There were no Epstein-Barr virus (EBV)-encoded small RNA (EBER)-positive cells on in situ hybridization using a Ventana automated (BenchMarkTM) stainer.

**Discussion**

The histological finding of the present lesion is quite unusual in MZBL. On a low-power field, the lesion composed of fibrous granulation tissue and lymphoid follicles. The fibrous granulation tissue was characterized by the presence of numerous haphazardly arranged spindle cells, mature plasma cells and eosinophils. The presence of the lymphoid follicles seen in our case is also present in IMT of the soft tissue [2]. Ballesteros et al. also reported two cases of IMT of the oral cavity containing numerous mature plasma cells [3]. The overall histological findings of the present lesion were somewhat similar to those of IMT of the soft tissue including oral cavity [2–6]. However, the spindle cells were vimentin+, desmin−, muscle specific actin−, cytokeratin−, p80− [5, 6].

In addition, in this lesion, a portion of the lymphoid follicles were surrounded by partial and/or complete thin pale cuff of the CCL-cells, cells with plasma cell differentiation and mature plasma cells. This histological finding is one of the histological characters of MZBL namely, a marginal zone distribution pattern [1]. Moreover, flow cytometry and immunohistochemical study demonstrated the monotypic nature of the plasma cells and their precursor and confirmed the diagnosis of MZBL arising from buccal mucosa.

Kimura’s disease appears another differential diagnosis because of the presence of lymphoid follicles and numerous eosinophils and plasma cells in the fibrous granulation tissue in this lesion [7–9]. Moreover, there was germinal center deposition of IgE which is one of the characteristic immunohistochemical findings of Kimura’s disease [8, 9]. However, there were no eosinophilic microabscesses or Warthin-Finkeldy type polykaryocytes in the lesion [7, 8]. Moreover, there was no peripheral blood eosinophilia.

Tissue eosinophilia is frequently observed in MZBL [10]. However, the association of MZBL and prominent tissue eosinophilia and fibroblastic proliferation appears to be a rare event [1]. Previously, the differential diagnostic problems between IMT of the soft tissue and classical Hodgkin lymphoma and T-cell lymphoma have been well discussed in the literature [2, 4]. However, the present case indicated that MZBL should be added to the differential diagnosis of IMT of the soft tissue. Both IMT arising from
buccal mucosa and primary buccal mucosa MZBL appear to be rare disorders [2, 3, 11]. However, it is important to discriminate MZBL and IMT on clinical practice.

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