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Mucosa-Associated Lymphoid Tissue Lymphoma of the Cheek Mimicking Benign Entities: a Case Report

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Case Report

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The prevalence of cheek lymphoma, especially a mucosa-associated lymphoid tissue lymphoma (MALT), is very rare. Non-specific symptoms and image findings of cheek lymphoma may mimic benign entities and make it difficult to diagnose. In this case report, we present a case of MALT lymphoma of the cheek mimicking benign entities on computed tomography and magnetic resonance imaging.

Keywords: Lymphoma; Cheek; Head and neck neoplasms; Magnetic resonance imaging; Computed tomography

INTRODUCTION

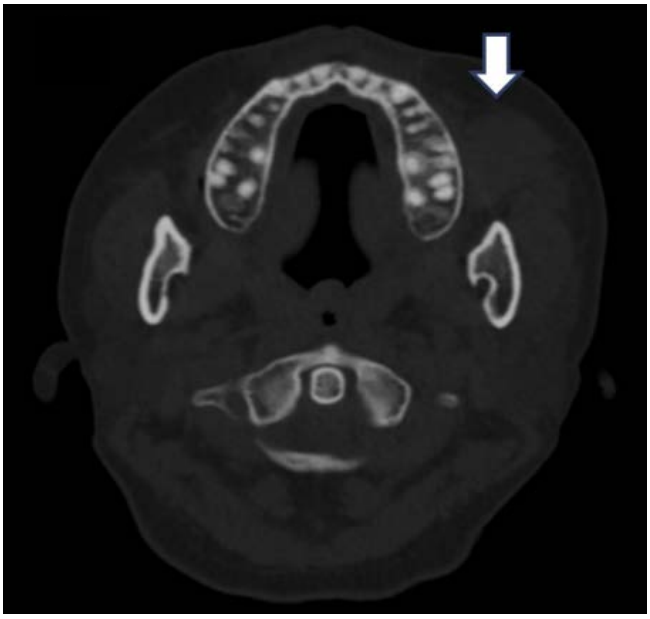
Mucosa-associated lymphoid tissue (MALT) lymphoma, a form of low-grade lymphomas of the mature B-cell, may involve numerous extranodal sites, such as the gastrointestinal tract, lung, oropharynx, or skin, and may occur in chronic inflammation or autoimmune disease patients (1-3). Although the prevalence of MALT lymphoma accounted for 19% of all non-Hodgkin's lymphoma in a study conducted by Kim et al. (4), MALT lymphomas involving the head and neck are rare. Considering typical location of head and neck lymphomas is in the Waldeyer's ring, isolated cheek involvement of non-Hodgkin's lymphoma such as a MALT lymphoma is very rare (2, 5-7).

Clinically, head-and-neck lymphoma may present as cervical lymphadenopathy and B-symptoms such as weight loss, fever, and night sweats (2, 8). However, diagnosis of lymphoma involving the head and neck region, including the subcutaneous tissue of the cheek, can be difficult because of its non-specific symptoms and non-specific radiological appearances. In this case report, we present a rare case of an immunocompetent, Epstein-Barr virus (EBV)-negative, mucosa-associated lymphoid tissue (MALT) lymphoma of the cheek, mimicking benign entities on image modalities.

CASE REPORT

This case report was approved by our Institutional Review Board and waived the requirement for informed consent.

A 49-year-old woman with no history of underlying illness or recent trauma presented with painless swelling and a palpable mass in the left cheek that had persisted for two months. The swelling worsened progressively, and anti-inflammatory medications, prescribed at different institution, did not alleviate it. The initial evaluation, performed at our outpatient center in the otorhinolaryngology department, included a blood work-up, computed tomography (CT), and magnetic resonance imaging (MRI). The initial serum laboratory and blood chemistry results were generally within normal limits, except for slightly low red blood-cell counts, hemoglobin concentration, and hematocrit percentage (3.62×10^{12}/L, 10.2 g/dL, and 31.7%, respectively). There was no bleeding



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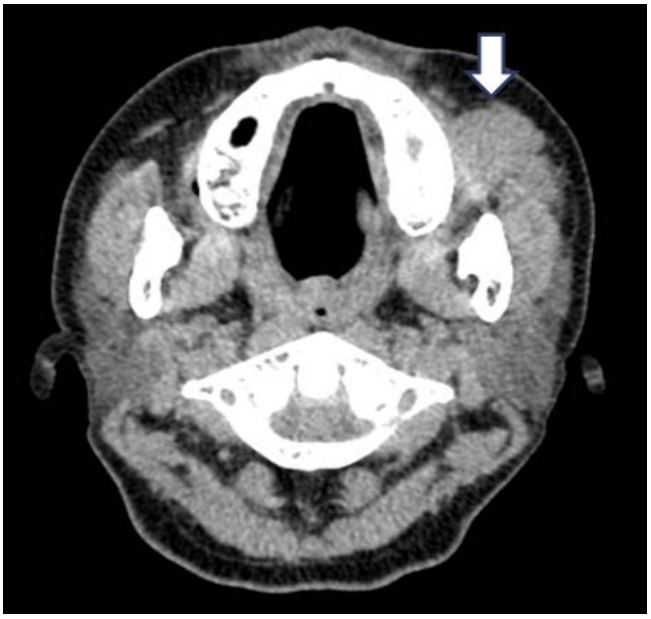

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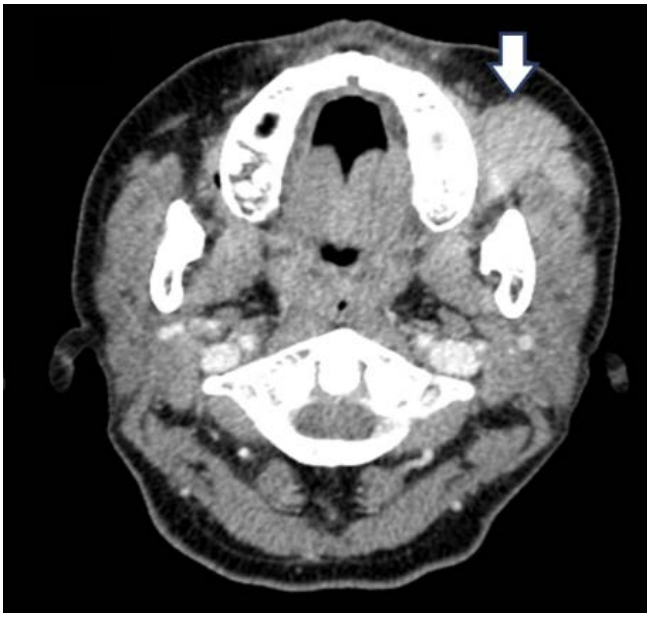

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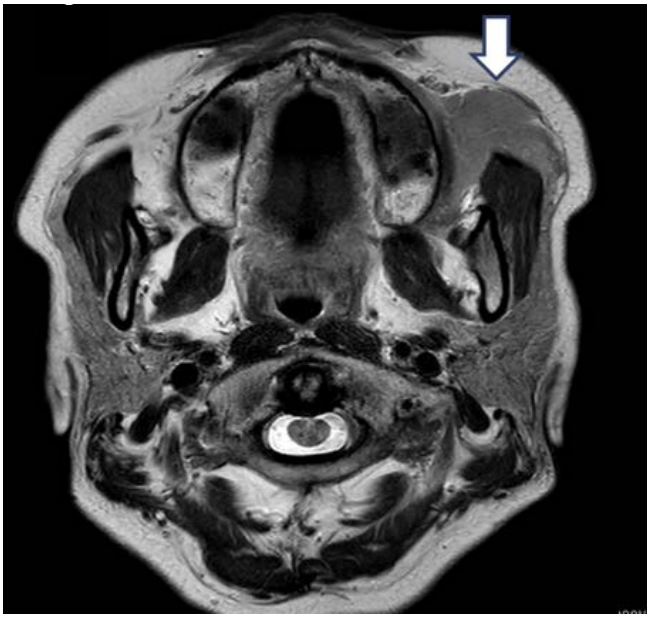
Fig. 1. Computed tomography (CT) findings of cheek mucosa-associated lymphoid tissue lymphoma. (a, b) Axial scan of non-contrast CT of the face demonstrated a smooth-margin mass (arrows), approximately 2.8 cm (transverse) × 3.5 cm (anteroposterior), located at the left cheek subcutaneous region with 50 Hounsfield units (HU) without evidence of osteolysis or sclerotic changes of adjacent facial bones. (c) Contrast-enhanced axial CT demonstrated the mass (arrow) with uniform enhancement of 110 HU.

tendency, and there were no abnormalities in the bleeding time, prothrombin time, or activated partial thromboplastin time. The initial fine-needle aspiration biopsy at the left cheek mass revealed lymphocytic cell infiltration.

Non-contrast CT of the patient's face demonstrated a smooth-margin mass in the subcutaneous tissue of the left cheek, with dimensions of approximately 2.8 cm (transverse) × 3.5 cm (anteroposterior) × 2.8 cm (craniocaudal) (Fig. 1). The mass was measured to be 50 Hounsfield units (HU). After the contrast-media infusion, the mass showed a uniform enhancement of 110 HU. There were no signs of osteolysis or sclerotic changes of the adjacent bony structures, and none of internal calcifications. The initial differential diagnosis based on the CT findings included benign entities, such as venous malformation, Schwannoma, and pleomorphic adenoma, arising from accessory parotid gland. A small, uniformly enhancing venous malformation was a possible diagnosis; however, there were no typical



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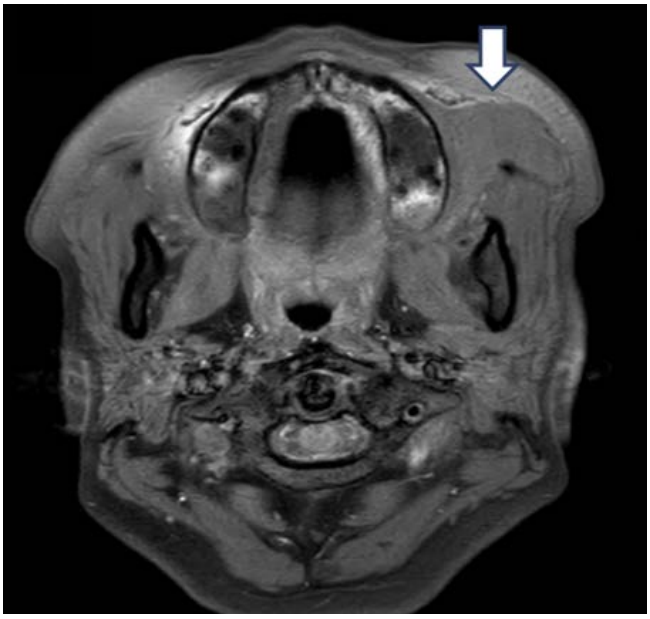

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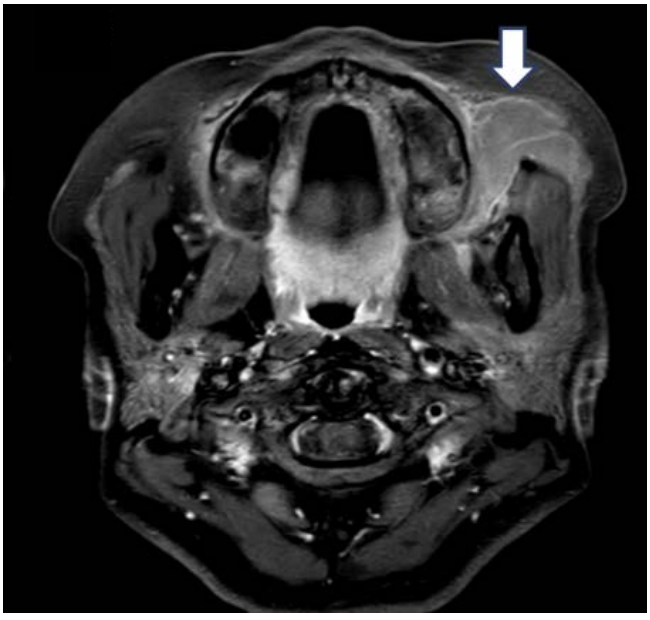

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Fig. 2. Magnetic resonance (MR) findings of cheek mucosa-associated lymphoid tissue lymphoma. (a) T2-weighted image and (b) T1-weighted image axial scans demonstrated a smooth-margin insinuating mass (arrows) at the left cheek subcutaneous region with slightly high T2 signal and iso-intense T1 signal intensity relative to surrounding muscle signal. (c) T1 contrast-enhanced axial scan demonstrated uniform enhancement of the mass (arrow) with minimal or no mass effect. There was no definite evidence of perilesional infiltration or skin thickening.

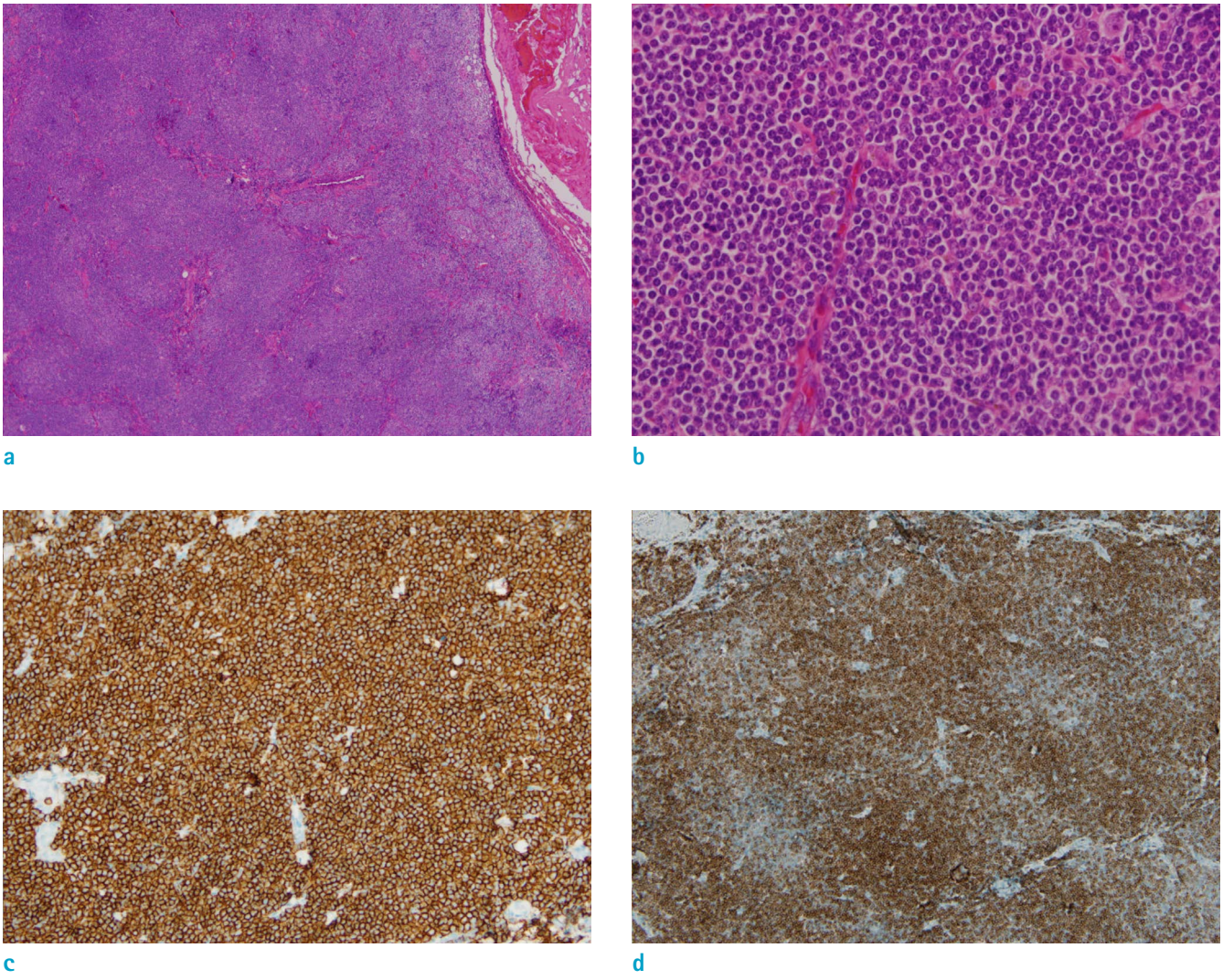

Fig. 3. Histopathologic findings of excision biopsy at the left cheek mass. (a) The lymphoma cells surrounded germinal centers, expanding the marginal zones and interfollicular areas. (b) The lymphoma cells showed relatively monotonous medium-sized nuclei and abundant pale cytoplasm. (c) The tumor cells demonstrated diffuse expression of CD20. (d) Immunohistochemistry for Bcl-2 highlighted the lymphoma cells (Bcl-2 positive) and the small remnants of the germinal center (Bcl-2 negative).

findings of venous malformation, such as phleboliths or calcifications. We considered Schwannoma and pleomorphic adenoma originating from the accessory parotid gland, since the enhancing smooth-margin mass with minimal mass effect was located at the anterolateral aspect of the masseter muscle. Although cheeks are rare extranodal sites of lymphoma, lymphoma can be also considered from its features, such as insinuating nature and uniform enhancement. These features were clarified upon further evaluation using 3-Tesla MRI (Ingenia, Siemens, Germany).

Non-contrast T2-weighted images and T1-weighted axial scans revealed a smooth-margin, insinuating mass with slightly high T2 signal and iso-intense T1 signal relative to the surrounding muscle signal (Fig. 2). Similar to the CT findings, contrast-enhanced T1-weighted images showed uniform enhancement of the mass with minimal or no mass effect exerted on adjacent muscles or on the left maxillary sinus. There was no definite evidence of perilesional infiltrations or skin thickening. Unlike venous malformation and Schwannoma, the mass showed a relatively uniform signal intensity (lower T2 signal intensity than that of a typical venous malformation T2 signal) and uniform

enhancement on T1WI with contrast media. Pleomorphic adenoma arising from the accessory salivary gland was a possibility based on the CT findings. However, MRI revealed that the mass was insinuating rather than exerting mass effect. Considering the MR findings of insinuating mass with uniform enhancement, our initial diagnoses based on CT findings were less likely, and we included lymphoma as a possible diagnosis. Other than the left cheek region, there were no lymphoma involvements in lacrimal glands, central compartment, or lateral compartment of the scanned neck.

Open, excisional biopsy under general anesthesia was performed in the otorhinolaryngology department. A horizontal excision at the left buccal mucosa just underneath the orifice of Stensen's duct was performed and dissection was performed through the left buccal fat pad and buccinators muscle. A soft, yellowish mass was observed in the superior buccal space and excision biopsy was performed. The histopathological findings showed that the lymphoma cells were expanding the marginal zones and interfollicular areas. The tumor cells showed monotonous medium-sized nuclei and abundant pale cytoplasm (Fig. 3). Immunohistochemical staining demonstrated diffuse CD20 expression of the tumor cells, suggestive of B-cell lymphoma, and Bcl-2 staining highlighted the tumor cells proliferating in the marginal zones and infiltrating into the residual germinal centers. In *in situ* hybridization, EBV was negative. The pathologic diagnosis was extranodal marginal-zone lymphoma of MALT. After the histopathological confirmation, the patient was referred to the hematology-oncology department for treatment planning and further evaluation.

DISCUSSION

Some studies report that prevalence of isolated MALT lymphoma of the cheek is very rare (2-4). Although cheek lymphoma may present with B-symptoms, their symptoms may be non-specific and similar to those of benign entities (2, 3, 5, 7, 8). For these reasons, the diagnosis of MALT lymphoma arising in the cheek subcutaneous tissue is challenging. Furthermore, when visualized using image modalities such as CT and MRI, MALT lymphoma of the cheek may mimic benign entities such as venous malformation, Schwannoma, and pleomorphic adenoma.

In our case, the patient presented only with swelling at the left cheek. Laboratory work-up results were within normal limits, except for a mild decrease in red blood-cell counts, and the initial fine-needle aspiration demonstrated lymphocytic infiltration. CT showed a soft-tissue mass with little or no mass effect in the left cheek subcutaneous tissue. The initial differential diagnosis included benign entities, such as venous malformation, Schwannoma, and pleomorphic adenoma. The follow-up MR images further clarified the insinuating, molding nature of the mass without phleboliths and its T2 signal intensity lower than that of venous malformation T2 signal, which lowered the possibility of the initial diagnoses based on the CT scan. The mass showed uniform signals among different MRI sequences and uniform enhancement in T1-weighted image with contrast media; however, the Schwannoma of head and neck may exhibit heterogeneous enhancement, such as a nodular rim enhancing pattern (9). There have been reports of comparable imaging findings of lymphoma in the head and neck region. Malaguarnera et al. (3) reported ultrasound and CT findings of cheek lymphoma, describing it as a well-demarcated, hypervascular mass. Kim et al. (10) reported MRI findings of B-cell lymphoma in the buccal space, describing it as a well-demarcated, uniform mass with a molding rather than infiltrative nature, although they noted that T-cell lymphoma may exhibit an infiltrative nature.

MR findings of MALT lymphoma of the cheek, such as insinuating nature and uniform T1- and T2-weighted image signals, may be helpful with a differential diagnosis; however, there are no definite pathognomonic MRI features of MALT lymphoma of the cheek, especially when it is small. In our case, MRI findings leaned towards lymphoma rather than toward venous malformation, Schwannoma, or pleomorphic adenoma arising from the accessory parotid gland. The final diagnosis was confirmed via excisional biopsy and histopathology.

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