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Case Report

Castleman Disease in the Buccal Space Mimicking Lymphoma: A Case Report

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ABSTRACT

Castleman disease is a benign lymphoproliferative disorder of unknown etiology, more commonly appearing in the mediastinum. Craniofacial involvement is rare. Surgical excision is the only intervention needed for the unicentric type, while systemic treatment would be necessary for the multicentric type. Here, we report a rare case of unicentric castleman disease of the buccal space of a 23-year-old woman. Additionally, we review the radiological and histopathological features of this lesion.

INTRODUCTION

Castleman disease (CD) is an infrequent lymphoproliferative disorder with an unknown etiology. It is a lymphadenopathy with specific histologic characteristics and is generally benign. The clinical manifestations can be categorized into two groups: unicentric (localized) and multicentric (systemic) (1). Unicentric CD, the more frequent type, is defined as a localized lymphadenopathy, while multicentric CD has systemic manifestations such as flulike symptoms and multiple lymphadenopathies probably associated with high cytokine levels. It is also believed that there is a strong relationship between multicentric CD and HIV or HHV-89 (2).

We herein describe a rare case of unicentric CD with presentation of a mass located in the buccal space that was completely excised.

CASE REPORT

A 23-year-old woman presented with swelling on the left buccal region first noticed 2 years ago. Medical history was unremarkable and the initial laboratory findings were normal. The patient was HIV negative.

In the clinical examination, the lesion was slightly firm, non-tender and mobile. There was no sensory alteration or loss of motor function in the facial region.

Ultrasound examination of the buccal region revealed a 2 x 1 cm, well defined and hypoechoic mass located at the left buccal space on the buccinator muscle.

Midface non-enhanced computed tomography (CT) demonstrated a homogenous, well defined, oval-shaped and isodense entity in the left buccal space on the buccinator muscle, anterior to the masseter. No effect on the maxilla was detected (Figure 1). The differential diagnoses included reactive buccinator lymph node, fibroma and lymphoma.

After general anaesthesia, access to the lesion was provided by a vestibular incision. The lesion was tightly adherent to the buccinator muscle. The buccal branch of the facial nerve was exposed and retracted. The lesion was carefully dissected and excised from the adjacent buccinator muscle and subcutaneous fatty tissue without complications. Grossly, it was a brownish lobulated soft tissue, with yellow-tan color on cut surfaces. The pathologic examination revealed numerous follicles scattered in a mass of lymphoid tissue with no sinuses. Higher magnification showed marked

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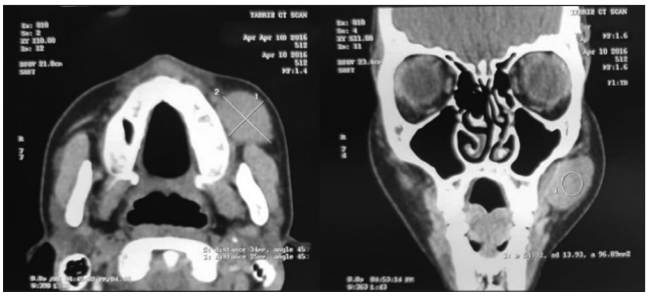

Figure 1. Midface non-enhanced computed tomography (CT) demonstrating a homogenous, well-defined, oval-shaped and isodense entity in the left buccal space on the buccinator muscle, anterior to the masseter

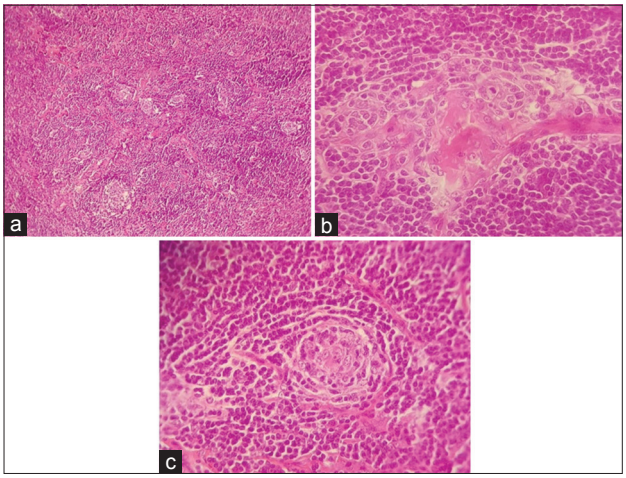

Figure 2. Microscopic view (H&E staining) (a) Numerous follicles scattered in a mass of lymphoid tissue with the absence of sinuses, (40x) (b) Higher magnification shows vascularization and hyalinization of the abnormal germinal centers. (100x) (c) Tight concentric layering of lymphocytes at the periphery of new follicles also known as "the onion skin appearance"

vascular proliferation and hyalinization of the abnormal germinal centers. There was also a tight concentric layering of lymohocytes at the periphery of new follicles (Figure 2). The definite diagnosis was hyaline vascular-type castleman disease.

No further treatment was given. The first 6-month follow-up, revealed no recurrence.

DISCUSSION

Castleman disease is an uncommon lymphoproliferative disorder. The incidence is estimated at 21-25 cases per million person-years (3); however, its epidemiology remains unclear due to the low incidence. Generally, CD is of unknown etiology but increased production of interleukin-6 by the lymph nodes is suggested as a possible cause in cases with systemic manifestations (4).

There are 2 clinical manifestations of CD: unicentric disease (localized enlargement of a single lymph node) and multicentric disease (including systemic involvement). Unicentric CD is commonly asymptomatic. The most common site (up to 70%) is the mediastinum followed by the cervical region (15%-20%), bearing in mind that unicentric CD may occur at any site with a lymphatic tissue (5).

It must be taken into consideration that CD could develop in patients with Kaposi's sarcoma, lymphoma, and POEMS syndrome (6).

CD appears in two histologic forms: (1) hyaline vascular and (2) plasma cellular. The hyaline vascular type is distinguished by lymphatic follicles spread in hypervascular and hyalinized stroma with the lymphocytes aligned in a concentric pattern around the follicles. It usually has a benign nature and develops as a single entity without systemic symptoms. The plasma cellular type, on the other hand, is scarce and also more invasive noted by mature plasma cell groups scattered in the inter-follicular zone (7).

Despite the common sites of involvement, CD is probable at any site with lymph nodes as the buccal space in the current case. Hyperplastic lymph nodes showing enhancement with contrast on CT supports CD. Accurate diagnosis, however, necessitates histopathological assessment. Surgery with no further treatment is considered the treatment of choice for unicentric CD, with follow-ups to detect any recurrence.

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