Bilateral cavernous sinus thrombosis as first manifestation of primary Burkitt lymphoma of the thyroid gland

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Abstract

Cavernous sinus thrombosis (CST) is a rare condition that is usually associated with infections, pregnancy, vasculitis and some types of medication, such as the contraceptive pill and paraneoplastic. Primary Burkitt lymphoma (PBL) of the thyroid gland is very uncommon and the clinical description of such cases has been largely limited to case reports. In this paper, we present a case of CST as the first manifestation of PBL of the thyroid gland. To the best of our knowledge, our patient is the first case report of PBL of the thyroid gland that presents with bilateral CST.

Introduction

Cavernous sinus thrombosis (CST) is a rare condition that usually due to infections, pregnancy, vasculitis and some types of medication, such as contraceptive pill.[1] Burkitt lymphoma is a form of non-Hodgkin's lymphoma that starts in immune cells called B-cells and is highly aggressive and fatal without treatment.[2] Primary Burkitt lymphoma (PBL) of the thyroid gland is a very uncommon and the clinical description of such cases has been largely limited to case reports. The early clinical effects of PBL of the thyroid gland are dyspnea, dysphagia, pain and hoarseness of voice that might be due to compression effects of tumor and lymphadenopathy. The early manifestation of this tumor with CST is very rare.[3,4] This report describes a 34-year-old woman who had the CST as the initial manifestation of Primary Burkitt lymphoma of the thyroid gland.

Case Report

A 47-year-old Iranian women was referred to our hospital with diplopia. The early Symptoms had begun 1 months earlier with binocular diplopia, headache and nausea. A headache was bifrontal, pulsatile and exacerbated by supine position. Previous medical history was unremarkable. There is no family history of heart disease, liver cancer and stroke. On examination, he was conscious and had stable vital signs, there was no peripheral lymphadenopathy palpable. He had normal rate and regular rhythm, no murmur. Abdomen was flat, soft and non-tender with normal liver and spleen. Extremities had normal findings. Neurological examination shows complete ophthalmoplegia of left eye and sensory loss of the first and second division of the left trigeminal nerve. Muscle strength was 5/5 in all four limbs. All deep tendon reflexes were normal and plantar response was bilateral flexor. On 3 day of admission, she experienced a diplopia and a left ptosis in other eye. On MR (Figure 1), there was symmetrical enlargement of both cavernous sinuses that enhanced after contrast. After the anticoagulation therapy with enoxaparin and warfarin, in follow up MRI, complete improvement was seen. The laboratory studies, including complete blood count, creatine kinase, erythrocyte sedimentation rate, and C-reactive protein level were normal. The thyroid function tests, anti-thyroglobulin and thyroid-stimulating hormone receptor antibody also were normal. Cerebrospinal fluid (CSF) was normal in cell count, protein, glucose and cytology. The work up for occult malignancy including chest CT, abdominopelvic CT, and mammography was normal, the PET scan was not done but Due to the neck pain that developed in the course of hospitalizations, the neck sonography was done that showed enlargement of thyroid gland with lymphadenopathy. The surgical biopsy was done for the patient and PBL of thyroid finally was diagnosed. The bone marrow also biopsy was done that was negative. The chemotherapy was started for her in ward chemotherapy but she succumbed 1 month after the diagnosis due to lack of response to the treatment and upper respiratory tract involvement.

Discussion

Primary thyroid lymphoma (PTL) is a rare thyroid malignancy that accounts for about 1 to 5% of all thyroid cancers and 2 % of all malignant extra nodal lymphomas. It is found more in older patients and has predominance in women and is usually associated with Hashimoto thyroiditis.[5] The common subtypes are diffuse large B cell lymphoma, mucosa-associated lymphoid tissue (MALT) lymphoma and primary Burkitt lymphoma.[6] Burkitt lymphoma of the thyroid gland are very rare and has been largely limited to case reports. In study by Campagno *et al*. retrospectively reviewed 245 patients with lymphoma of the thyroid gland, Burkitt's lymphoma was observed in 3 (1.2%) of patients.[7] Clinically, PBL like anaplastic thyroid carcinoma has rapid grow with compression symptoms consisted dyspnea, dysphagia, pain, hoarseness of voice and lymphadenopathy. Paraneoplastic neurological syndromes (PNSs) rarely associate with lymphoma. They include cerebellar degeneration, dermato/polymyositis, sensory neuronopathy, Lambert- Eaton myasthenic syndrome, granulomatous angiitis, cerebrovascular disease, subacute necrotizing myelopathy and encephalomyelitis.[8,9] Cavernous sinus thrombosis (CST) as an initial presentation of lymphoma, especially thyroid Burkitt lymphoma is very rare. To the best of our knowledge, our patient is the first case report that CST present as first manifestation of PBL of thyroid. The clinical effects of CST are headache, diplopia, proptosis, impaired visual acuity and ophthalmoplegia due to involvement of cranial nerves, optic nerve and venous drainage that are located within or along the cavernous sinus.[10-12] CT and MR provide useful information to the diagnosis and exclude other pathology that can mimic this sign and symptoms.[13] In our patient on MRI, there was a*symmetrical cavernous sinus enhancement*. In differential diagnosis, in addition to CST, Carcinomatous meningitis also should be considered. But due to, in the course of disease that we was seen the complete improvement in follow up MRI, before the onset of chemotherapy and after anticoagu-

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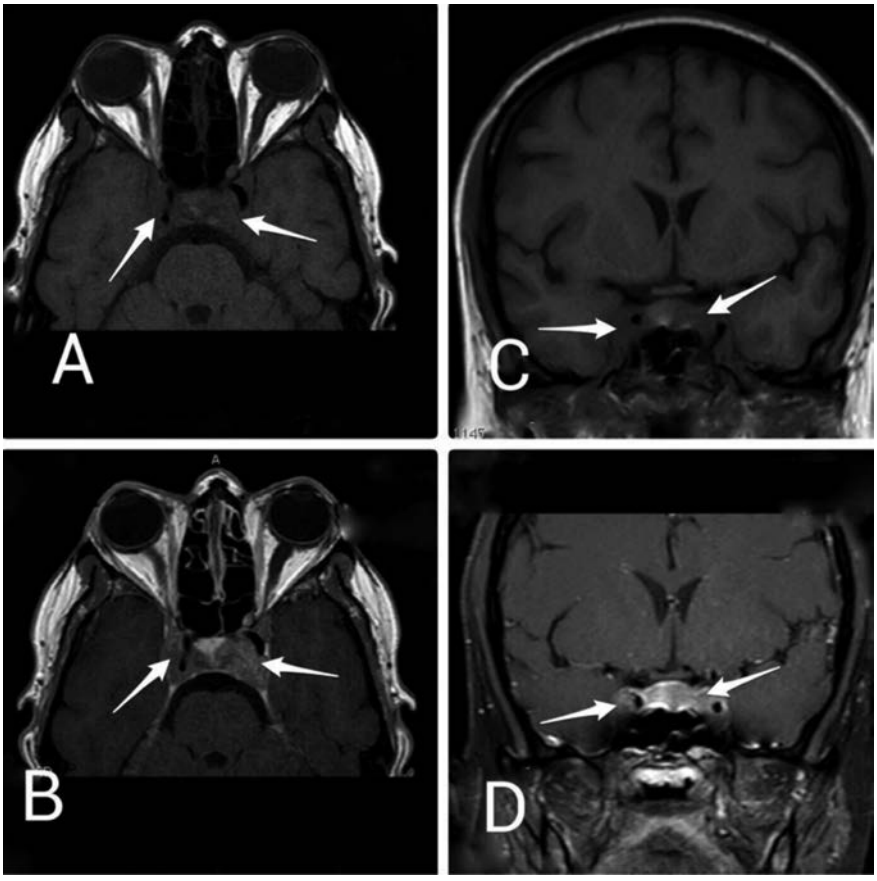

Figure 1. Axial and coronal T1 weighted images (A and C) show bilateral asymmetrical enlargement of cavernous sinuses with inhomogeneous contrast enhancement (B and D).

lant therapy and normal CSF cytology, the diagnosis of CST is more likely in the presence of this clinical features. For diagnosis of thyroid Burkitt lymphoma, core needle biopsy has higher sensitivity and positive predictive value than FNA for the diagnosis of thyroid gland lymphomas because the presence of concurrent Hashimoto thyroiditis, decrease the accuracy of FNA.[14,15] In our patient at first FNA was done for the patient that the Hashimoto thyroiditis was diagnosed but after surgical biopsy, Burkitt lymphoma diagnosed. The first step in treating is to eliminate the underlying cause to reduce the risk of recurrence, but due to prevent further thrombosis, anticoagulation with heparin should be considered.[16,17] In our case heparin and chemotherapy were started but she succumbed one month after the diagnosis of the disease.

Conclusions

In conclusion, paraneoplastic bilateral CST as the first presentation of lymphoma is very unusual. But in any case without evidence of common causes, hypercoagulability state and malignancy should be considered.

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