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Case Report

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BILATERAL PAROTID GLANDS SWELLING AND FACIAL PALSY AS RELAPSING SYMPTOM IN ADULT PATIENT WITH ACUTE LYMPHOBLASTIC LEUKEMIA: A CASE REPORT

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ABSTRACT

Parotid glands are uncommonly involved in leukemia. Moreover, parotid gland relapse is an extremely rare event. We report a young adult male with acute lymphoblastic leukemia who presented with bilateral parotid glands involvement and facial palsy as first presenting sign of relapse. Unfortunately in spite of aggressive treatment the outcome was dismal.

KEYWORDS: Parotid gland, Acute lymphoblastic leukemia, Facial palsy.

INTRODUCTION

About one half of adults with acute lymphoblastic leukemia are not cured of the disease. After front-line therapy, one third of patients with standard-risk and two thirds of high-risk patients will relapse.^[1] Extramedullary relapse is a recurrence of leukemia in sites other than the bone marrow.^[2] Relapse in lymphoid tissue and sanctuary sites, such as the central nervous system and testes, is common in acute lymphoblastic leukemia. It is rarely isolated, and almost always exists with systemic marrow disease.^[3,4] Relapse of acute lymphoblastic leukemia occurring in the parotid is extremely rare.^[5] Infiltration of blast into parotid gland lead to swelling with or without lacrimal gland involvement. This unique clinical presentation may be misdiagnosed as mumps and leading to delay both diagnosis and treatment.^[6,7] Hematology/Oncology Department, Imam Al- Hussein Medical City, Karbala, Iraq.

CASE REPORT

An 18-year-old male presented with a fatigue for two months. On examination, he was pale, no lymph nodes were palpable and no organomegaly. Blood film suggested acute lymphoblastic leukemia. Bone marrow aspiration revealed a heavy infiltration by lymphoblast. PAS stain 1% positive and SBB stain was negative. Picture was consistent with acute lymphoblastic leukemia (Figure 1). Peripheral blood fluorescence in situ hybridization was undetected for BCR-ABL. Flow cytometric immunophenotypic analysis was consistent with B-cell acute lymphoblastic leukemia (CALLA +). Cerebrospinal fluid analysis showed no malignant cells. Patient received UKALL XII regime, after phase I induction, patient achieved remission (Figure 2).

After phase II induction, patient presented with drooping of his mouth to the right side, loss of his ability to close the left eye and bilateral non tender parotid swelling for more than ten days. The parotid swelling was not painful but associated with difficulty in opening the mouth and mastication. Neurological examination suggested lower motor neuron lesion left side facial palsy (Figure 3 & 4). FNA from parotid suggested clusters and individual lymphoblast infiltration (Figure 5). Bone marrow aspiration revealed heavy infiltrated by blasts (Figure 6). Central nervous system disease (CNS) was ruled out by cerebrospinal fluid examination (cytospin and cell count) which was negative for malignant cells. Patient received hyper-CVAD regime, his parotid swellings dramatically decreased in size but patient didn't achieved bone marrow remission. Unfortunately the patient died after second cycle because of disseminated fungal sinusitis.

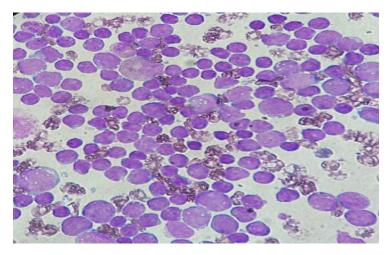


Figure 1: First bone marrow showing infiltration by lymphoblast.

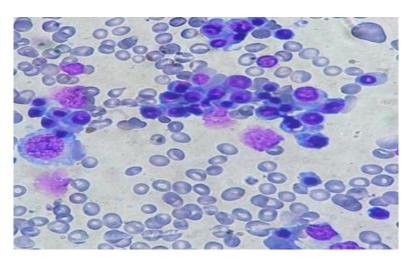


Figure 2: Bone marrow in remission after chemotherapy.





Figure 3: Left side facial palsy.



Figure 4: Bilateral parotid swelling.

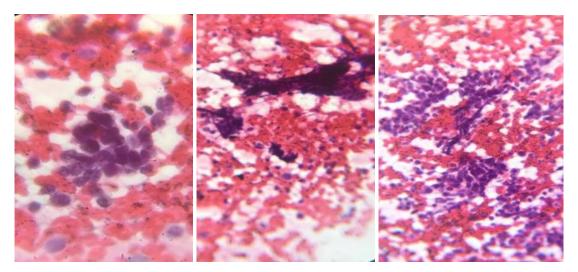


Figure 5: Cellular and hemorrhagic smears showed clusters and individual lymphoblast infiltration.

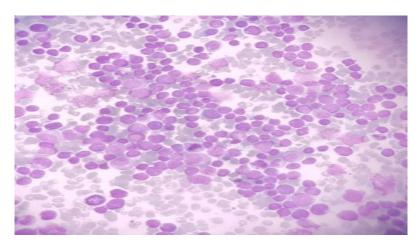


Figure 6: Bone marrow infiltration by lymphoblast (patient in relapse).

DISCUSSION

Data on extramedullary relapses of ALL is limited in the adult population. The majority of the literature either in pediatric patients or in the postallogeneic hematopoietic stem cell transplantation setting.^[8]

Parotid gland could to be a sanctuary site for relapse of acute leukemia either in isolation, or in association with central nervous system.^[5,9,10] Parotid gland involvements in ALL has been reported as post-chemotherapy second malignancy, first presentation and as relapse either after primary treatment or post-transplant.^[5,11-12]

Presentation as bilateral or unilateral swelling, mostly misdiagnosed as infection especially in pediatrics patients leading to delay proper mangement.^[11,13] Facial nerve involvement which co-exists with enlargement of the parotid gland in AML has been described in a previous reports while parotid gland involvement in ALL along with facial palsy has not yet been reported. Theoretically, lymphoblast from parotid mass could infiltrate facial nerve and cause palsy^[13,14] To the best of our knowledge this is the first adult ALL patients presented with parotid gland involvement and facial nerve palsy had been reported.

The outcome in such patients was poor possibly related to the bulk and biology of their disease.^[7] Although parotid swelling may subsided rapidly after initiation of therapy, the prognosis was dismal.^[13] Our patient treated with hyper CVAD protocol which is proved to improve outcome in relapsed acute lymphoblastic leukemia.^[15] Our patient was refractory to the treatment and we failed to achieve second remission. Unfortunately after two cycles patient died from disseminated fungal sinusitis.

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