

Gynecomastia, as an Extremely Rare Presentation of Chest Wall Lymphoma: A Case Report and Review of the Articles

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Abstract- Male breast enlargement as gynecomastia may be as a sign of underlying systemic diseases. Male breast malignancy is also considered in differential diagnosis. We present a young man with primary chest wall lymphoma as gynecomastia, without pre-existing problem or other disease. Here, we present his clinical manifestation, management, and his early outcome. Chest wall lymphoma as an initial presentation of isolated chest wall mass in males is a rare clinical entity and its presentation as gynecomastia is even extremely unusual.

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Introduction

Breast enlargement may originate from the breast, chest wall or even representing a secondary metastasis. Due to overlapping morphological characteristics of malignant and benign male breast masses, any palpable breast enlargement should be differentiated, interpreted and approached cautiously (1).

Primary tumors of the chest wall are not so prevalent, and the incidence of malignancy is reported up to 50% in some studies (2,3). Chest wall masses are usually invasive or metastatic tumors derived from the other organs like breast and lung tumors. Primary chest wall malignancies include only 5% of chest wall neoplasms and among them primary chest wall lymphoma is a rare entity and can mimic breast cancer. It accounts for 2.4% of chest wall tumors. Chest wall lymphoma is closely associated with preceding chronic pleural diseases such as pyothorax and pneumothorax with longstanding tuberculosis (2,3).

Although Breast cancer in men accounts for 1% of all breast malignancies, it is 100 times less common than in women. Surprisingly, its prevalence was rising from 1 in 100,000 in the 1970s to 1.5 in 100,000 in 2012 (1,4,5).

Breast lymphoma is also rare and may mimic

carcinomas clinically. Its prevalence is 1.6% of all identified cases with non-Hodgkin lymphoma and 0.5% of cases with breast cancer. B-cell lymphoma is found in 94% of the cases (6,7).

Some genetic factors such as genetic mutations, Klinefelter's syndrome in addition to factors such as alcohol, obesity and liver disease are considered as the major risk factors in male breast cancers (5).

Male breast enlargement as gynecomastia-both the localized type and the diffuse type- may be due to drug effects or even a sign of underlying systemic diseases, so male breast malignancy should also be considered in the differential diagnosis (8,9).

Because of the paucity of lymphoid tissue, primary breast lymphoma is a rare entity in the male and can mimic other benign or malignant masses. It is likely to be misdiagnosed, and its treatment still remains controversial (10,11).

There are few reports of primary chest wall lymphoma in which diffused large B-cell Lymphoma (DLBCL) is the most common subtype (12).

DLBCL is a subgroup of lymphoid B cell malignant proliferation that involves about 30% of all non-Hodgkin lymphomas (12).

Case Report

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The patient was a 30-year-old male that was admitted to our department due to gradually left breast enlargement during the previous 5 months ago. He was referred for surgery as gynecomastia three months ago, but the patient did not return for treatment. After 5 months, he came with a pale appearance and a large tense left breast lump measured about 10×10 Cm (Figure 1).



Figure 1. Patient's image reveals the apparent left breast and chest wall enlargement

A spiral chest CT scan was performed for further evaluation and revealed a solid round mass in the left anterior chest wall (Figure 2,3).

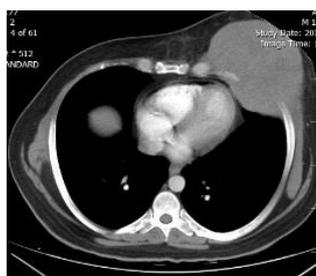


Figure 2. Chest CT scan revealed a left chest wall mass (mediastinal view)

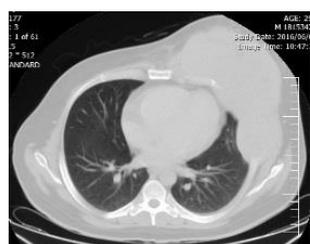


Figure 3. Chest CT scan revealed a left chest wall mass (lung view)

Subsequent chest wall mass core needle biopsy with immunohistochemistry (IHC) showed neoplastic cells positive for CD 20, LCA. His Ki 67 index was more than 70%. Tumor cells were negative for MYO-D1, CD99 and S-100. According to the above IHC profile, diffuse high grade B-cell lymphoma was confirmed (Figure 4).

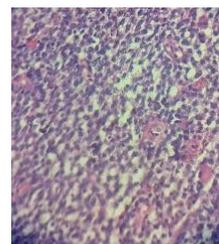


Figure 4. Small round cell tumor (X10)

His bone marrow aspiration revealed a mild cellular and erythroid hyperplasia and zero iron stores. No evidence of lymphomatous involvement is identified.

Chemotherapy was performed and the mass resolved successfully. He was treated properly with six cycles of R-CHOP regimen (Rituximab plus cyclophosphamide, Doxorubicin, vincristine, and Prednisolone) chemotherapy followed by local radiotherapy.

In the first course of chemotherapy, he had a severe leukopenia and infection that was resolved by G-CSF treatment. During the treatment, he experienced severe episodes of gastroenteritis. During the 12 months follow up, he was symptom free without any evidence of malignancy. His brain CT and whole body scans were negative for metastasis (Figure 5).

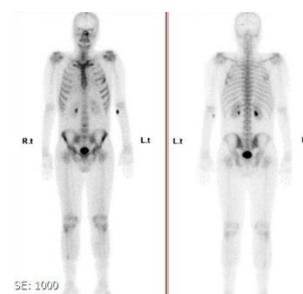


Figure 5. Whole body scan during the patient follow up

Among the few reported cases in the literature, our patient is one of the rare specific ones in clinical presentation of chest wall lymphoma.

Discussion

Breast cancer in male is a rare event; however male breast enlargement or a palpable lump is also prevalent. In some cases, it was diagnosed as a lump in breast and lymphadenopathy.

Chest wall tumors may represent the underlying diseases such as metastatic malignant tumors or invasion from a local tumor or even representing a Burkitt lymphoma originating from the chest wall (12,13). It may present as a chronic plural effusion or even other

pre-existing plural disease (2,14).

Most of the identifying chest wall tumors have been usually large with plural effusion or lung atelectasis, but some studies presented the chest wall tumors as slow growing with no other symptoms (3).

Qui *et al.*, reported a case of chest wall lymphoma in a woman, presenting as a chronic chest pain for about six months (15).

In another study, an EBV associated chest wall lymphoma was reported in the base of previous implanted mesh (15).

Chest wall lymphoma was also detected as a chronic post blunt traumatic rib fracture (15).

Lokesh *et al.*, described 2 cases of male breast lymphoma without B symptoms such as weight loss, fever or night sweats that were not the proper response to chemotherapy and failed treatment. One had a cerebrovascular accident with hemiparesis and died; the other developed to a progressive disease and lost to follow up (16).

in the pediatric population, Rich *et al.*, presented 3 cases of pediatric chest wall lymphoma as rare cases (17). Yaris *et al.*, reported a 5-year-old boy with a Burkett's lymphoma presenting as an isolated chest wall mass with soft tissue origination and in a rare another case report breast lymphoma is represented as gynecomastia (18,19).

Chest wall lymphoma may even present itself with a tender mass, hemothorax pain and plural effusion or rib erosion. There are some reports of pyothorax- associated malignant lymphoma adjacent to surgical mesh, in patients with a history of lung resection and chest wall reconstruction (20). A case of pyothorax-associated lymphoma of the chest wall was reported by Santini *et al.*, in a patient with chronic empyema. Their case had previous pulmonary tuberculosis (21).

The chest wall abscess was another rare presentation of lymphoma in a young man that confirmed later by cervical lymph node excisional biopsy (22).

Treatment

As local surgical resection is not often so effective, Chemotherapy and radiation therapy, or combination of both, are frequently done for treatment of primary breast lymphoma, but some authors agreed with this opinion that surgical resection followed by chemotherapy could improve the outcome in patients with locally chest involvement (2,15,16,23).

Pfreundschuh *et al.*, believed that Rituximab in addition to CHOP like chemotherapy protocol lead to a long term outcome improvement in patients with

DLBCL. As improving the overall survival in different follow ups, some other studies agreed with this idea (2,12,24,25).

In a few other cases, the patient died after an aggressive chemotherapy due to sepsis or other complications (13,16,25).

Chemotherapy is the first treatment of choice for lymphoma, even with or without chest wall involvement. Some studies suggested that the surgical debulking associated with chemotherapy will improve the survival in the early stages. In some others, chemotherapy and local irradiation are the best choice in poor local chest wall lymphoma (15,25). During the 15 months follow up in Santini's study, the patient had no problem in the post operation chemotherapy (21).

Ueda *et al.*, showed a slow growing chest wall lymphoma in a 57-year-old male. In his case as a solitary tumor, radiation therapy was the curative treatment and his two year follow up revealed no recurrence (3).

Mpallas *et al.*, suggested a multidisciplinary approach, including adjuvant chemotherapy accompanied with surgical resection to achieve a more favorable outcome (26).

Malignant lymphoma presenting as a solitary chest wall mass is not frequently seen. Only a few case reports have been found in the English literature. The treatment of primary chest wall lymphoma remains controversial (25).

Against the previously reported cases, our patient was young and had a slow growing isolated chest wall tumor (3,15,25).

According to the preceding studies, the outcome is different in various cases of chest wall lymphoma. Correct diagnosis and more effective treatment is made by clinical presentation incorporating with imaging findings (9).

A great index of suspicion, early biopsy and immunohistochemistry assessment and then proper treatment leads to better survival. To the best of our knowledge, primary DLBCL of the chest wall is rare and here is the extremely rare case of chest wall lymphoma presenting as gynecomastia. It is very important to have an accurate clinical examination, use of appropriate para-clinics such as ultrasonography, CT scan and MRI may involve some informative data for early detection and cost effective treatment of the breast tumors. This will achieve by sustained education of the general practitioners to be more familiar with the problem. In addition to gynecomastia "as a benign proliferation of male breast glandular tissue" other

benign breast tumors such as lipoma, schwannoma, granular cell tumor, fibromatosis, myofibroblastoma, pseudoangiomatous stromal hyperplasia and hemangioma are considered as differential diagnosis (1,27).

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