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Unexpected Rare Association of Secondary Breast, Thyroid, and Ovarian Lymphomas in a Female Patient with Burkitt Lymphoma: PET-CT Findings

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Abstract

We report the case of a 15-year-old female, followed for Burkitt lymphoma with initial ovarian presentation. Positron emission tomography-computed tomography (PET-CT) with ^{18}F-fluorodeoxyglucose (^{18}F-FDG) performed to evaluate initial extension assessment showed an unexpected association with secondary breast, thyroid, and ovarian lymphomas. To our knowledge, this association is very rare, unusual, and these extranodal presentations have been reported in literature as a separate case report. Authors highlight performance of PET-CT with ^{18}F-FDG to determinate the secondary origin of this multiglandular involvement, to establish a lesional map of the whole body, and to contribute to the therapeutic decision.

Keywords
- secondary breast
- thyroid and ovarian lymphomas
- Burkitt lymphoma
- PET-CT with ^{18}F-FDG
- case report

Introduction

The association of secondary breast, thyroid, and ovarian lymphomas is very rare at the time of the initial diagnosis of Burkitt lymphoma. Primitive or secondary, these are practically the same varieties of lymphomas, and diffuse large B cell lymphoma seems to be the most common origin. Positron emission tomography-computed tomography (PET-CT) with ^{18}F-fluorodeoxyglucose (^{18}F-FDG) is recommended to evaluate initial extension assessment, response to treatment, and to followup this group of patients with poorly prognosis.

Case Presentation

A 15-year-old female teenager presented with heavy menstrual bleeding and abdominal pain. She had no previous gynecological issues and any pathological history. On examination, she had large palpable pelvic and abdominal masses with no hepatosplenomegaly. Pelvic ultrasound suspected two small solid ovarian masses in the two ovarian glands without ascites, measuring 30×30 and 30×10mm, respectively. Computed tomography confirms the ovarian masses and showed at this time a suspected large digestive enlargement without significant lymphadenopathy or distant metastasis. Lactate dehydrogenase was 3,250 IU/L with normal serum β-human chorionic gonadotropin, α-fetoprotein (6ng/mL with normal value $<$ 15ng/mL), carcinoembryonic antigen ($<$ 5ng/mL), and cancer antigen-125 (19U/mL with normal value $<$ 35U/mL) levels. Biopsy of digestive tract and the left solid ovarian mass revealed focal CD20 staining and nuclear positivity for terminal deoxynucleotidyl transferase and scattered CD3 positivity that suggested a B cell lymphoblastic lymphoma. Peripheral blood and bone marrow biopsy showed evidence of malignant cells. PET-CT with ^{18}F-FDG was performed for initial staging, showing an unexpected association of secondary breast, ovarian, and thyroid lymphomas (►**Fig. 1A**). The intense hypermetabolism concerned the thyroid, more markedly in the right

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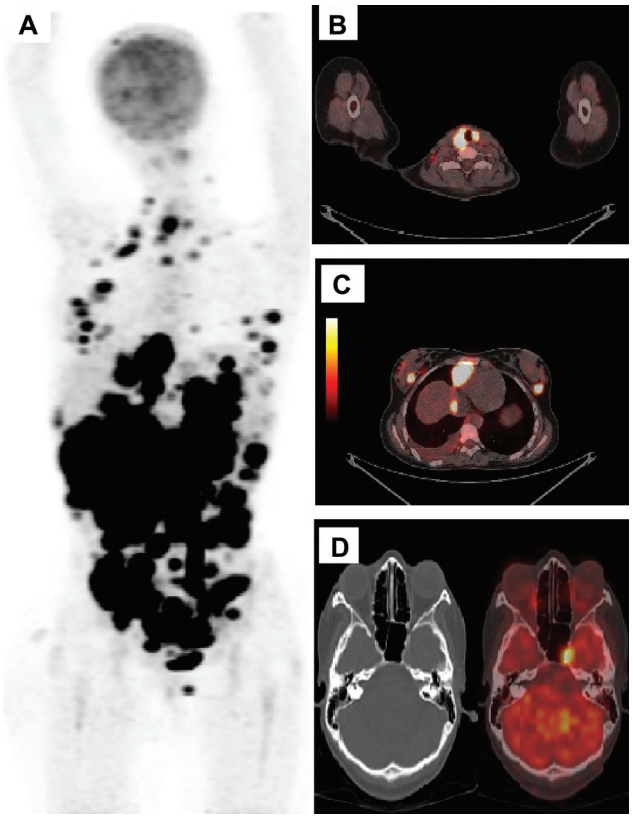

Fig. 1 (**A**) Maximum intensity projection of our patient, showing in addition to an above diaphragmatic nodes involvement, a rare association of breast, thyroid, ovarian, pleural, digestive, and osseous lymphoid diffusion as a concurrent extranodal disease at distant sites. Note that the ovarian involvement was taken in the pelvic hypermetabolism, difficult to characterize in our exploration. (**B** and **C**) Fusion images in axial sections showing intense hypermetabolism in breasts (maximum standard uptake value [SUVmax] = 17.1) and thyroid, more markedly in the right side (SUVmax = 17.7). (**D**) Computed tomography and fusion image in axial sections showing bone hypermetabolic foci the left base of the skull behind the sphenoid sinus (SUVmax = 5.0).

side (►**Fig. 1B**), breasts with multi-hypermetabolic foci (►**Fig. 1C**), digestive tract, and the right pleural effusion, like an extranodal diffusion. However, nodes involvement concerned just cervical, mediastinal, and right axillary regions.

Some hypermetabolic bone foci were visualized at the level of the skull and at the left pterygoid region suggestive for early bone involvement (►**Fig. 1D**).

Follow-up cervical ultrasound revealed a multinodular goiter. She had no symptoms consistent with either hyperthyroidism or hypothyroidism. She denied family history of thyroid lymphoid malignancy, and she had no prior history of neck irradiation. Thyroperoxidase antibody was 20.25 IU/mL (normal range: 0–34 IU/mL). About breast exploration, mammography confirmed the breast involvement and cancer antigen (CA 15–3= 17U/mL with normal value < 28U/mL) level was at the normal level. Our patient has been treated with heavy chemotherapy including rituximab, which plays an essential role in the treatment of CD20-positive B cell lymphoma, and posttherapy ^{18}F-FDG PET/CT scan showed a total response (►**Fig. 2**).

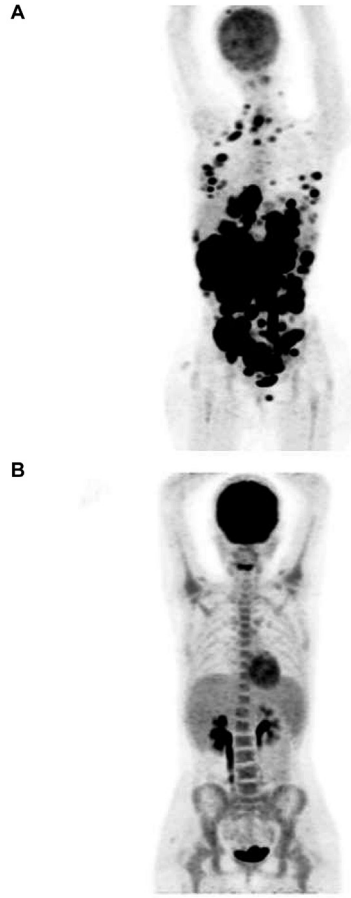

Fig. 2 (**A**) Maximum intensity projection (MIP) before treatment. (**B**) MIP performed after treatment showing a total response.

Discussion

Diffuse large B cell lymphoma is the most common subtype of non-Hodgkin lymphoma. Although the majority of diffuse large B cell lymphoma cases present with nodal involvement, approximately 25 to 33% present with extranodal involvement.[1]

Gastrointestinal sites are the most common locations for extranodal presentations.[1] Secondary breast, ovarian, and thyroid lymphomas represent 0.1, 0.5, and 3.4%,respectively, of these extranodal presentations, and have been reported in literature as a separate case report.[2–4]

Initially, for our patient, primary or secondary ovarian lymphoma was important in terms of prognosis. Lymphomas of the ovary should be regarded as local manifestations of systemic disease for therapeutic purposes.[5,6] At this time of diagnosis, if the lymphoma is clinically not confined to the ovary and the complete investigation shows evidence of extranodal lymphomatous lesions (imaging exploration, peripheral blood, and bone marrow with abnormal cells), the secondary origin can be considered.

PET-CT with ^{18}F-FDG findings was very consistent in our context, showing an unexpected and very rare association of breast, thyroid, ovarian, pleural, digestive, and osseous

lymphoid diffusion as a concurrent extranodal disease at distant sites.

For thyroid involvement, Hashimoto thyroiditis has been proposed as a potential predisposing condition for the subsequent development of thyroid lymphoma, perhaps via the infiltration of inflammatory lymphocytes and chronic antigen stimulation. Patients with chronic thyroiditis have a greater than 60-fold relative risk of developing thyroid lymphoma over baseline expected risk, which persists up to 10 years following the diagnosis of thyroiditis.[7] We do eliminate Hashimoto thyroiditis by dosing thyroperoxidase antibody that was at normal level.

However, the secondary breast lymphoma in our context was considered in the multiglandular involvement. CA (15–3) level was normal. No histological confirmation was obtained, to start chemotherapy as soon as possible.

Patients with multifocal extra nodal lymphomas have a very poorly prognosis.[1] The best treatment option seems to be a heavy chemotherapy[6] including rituximab that plays an essential role in the treatment of CD20-positive B cell lymphoma.[8]

Conclusion

Extranodal diffuse large B cell lymphoma can rarely and separately present with thyroid, breast, and ovarian involvement. The association of the three multiglandular involvement with Burkitt lymphoma is very unusual. PET-CT with ^{18}F-FDG is recommended to evaluate initial extension assessment. This hybrid imaging technique can help clinicians to determinate the secondary origin and to establish a lesional map of the whole body.

Conflict of Interest
None.

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