

Coexistence of Cancer of Unknown Primary Site in Patients Diagnosed with Follicular lymphoma in situ

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Background: Since follicular lymphoma (FL) is an indolent lymphoma, the recommended treatment for follicular lymphoma in situ (FLIS) is watchful waiting. In our case, we initially diagnosed abdominal lymphadenopathy as FLIS, but during follow-up, we observed aggressive lymphadenopathy, leading to an additional diagnosis of cancer of unknown primary site (CUPS).

Case report: A 59-year-old woman was presented with mesenteric lymph node (LN) enlargement detected on abdominal ultrasonography (USG) due to lower extremity edema. She had no previous medical history or B symptoms. There were no specific findings on laboratory test. Deep vein thrombosis detected on doppler USG of lower extremities. An abdomen-pelvic CT showed enlarged lymph nodes in multiple regions, suggesting metastatic lymphadenopathy or lymphoma. PET-CT showed no additional malignant lesions. The diagnosis of FLIS was made by an excisional biopsy of the mesenteric LN based on positive CD20, CD10, BCL-2 staining, and negative Ki67 staining. Watchful waiting was chosen as treatment. 4 months later, CT showed multiple pulmonary nodules and worsening lymphadenopathy. Considering the atypical nature of this progression compared to the typical behavior of FLIS, an additional excisional biopsy of the left cervical LN at level 4 was performed. The pathological report indicated metastatic squamous cell carcinoma with positive staining for p16 and p40. Further investigations, including PAP smear, sigmoidoscopy, and PET-CT failed to identify a primary cancer site, except for a few atypical squamous cells in the PAP smear. Chemotherapy (bevacizumab, paclitaxel, cisplatin) was initiated for suspected metastatic cervical cancer, considering predominant lower abdominal LN metastasis and positive P16 staining. After 2 cycle of chemotherapy, CT showed regression of multiple pulmonary nodules and improving lymphadenopathy.

Conclusions: FL is the predominant type of indolent non-Hodgkin's lymphoma, and active surveillance is the recommended treatment for FLIS. Because aggressive progression is uncommon, physicians should consider another differential diagnosis when encountering aggressive lymphadenopathy.

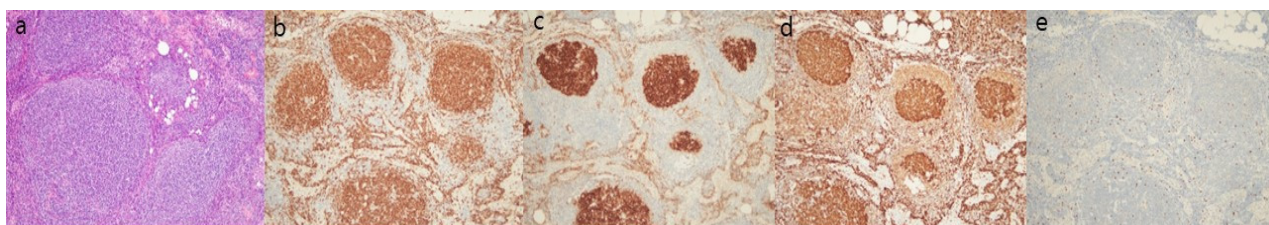


Figure 1. Histological diagnosis of FLIS of mesenteric lymph node (a) was H&E staining, and (b), (c), (d), (e) were immunohistochemistry staining of mesenteric lymph node. Germinal centers were positive for (b) CD20, (c) CD10, (d) Bcl-2 staining, and (e) Ki67 labeling index is very low, suggesting follicular lymphoma. (original magnifications X 100)

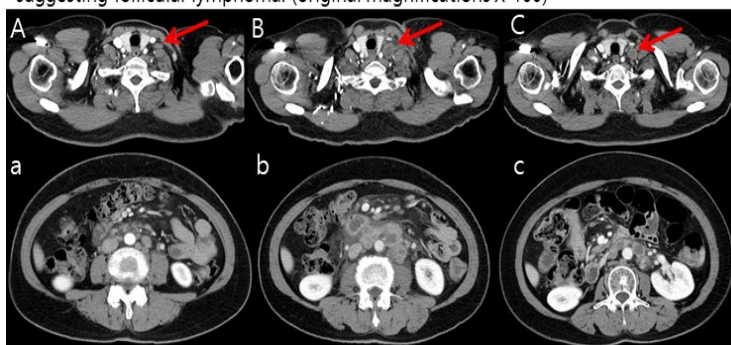


Figure 2. Computed tomography(CT). Initial chest CT (A) and abdominal-pelvic CT (a) showed enlarged lymph nodes in multiple regions including left supraclavicular lymph node (arrows). Images (B, b) after four month of "Watchful waiting" showed worsening lymphadenopathy. Images (C, c) after 2 cycle of chemotherapy showed improving lymphadenopathy.

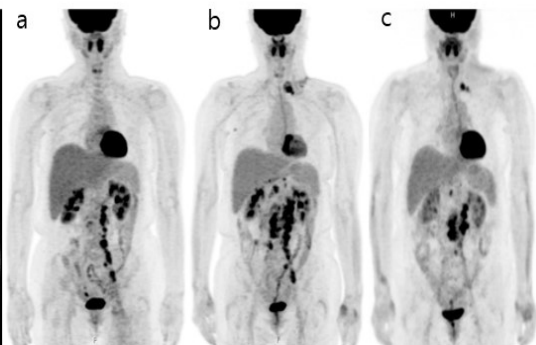


Figure 3. PET-CT. Initial PET-CT (a) showed enlarged lymph nodes in multiple regions. Images (b) after four month of "Watchful waiting" showed worsening lymphadenopathy and new lymphadenopathy in other regions. Images (c) after 13 cycle of chemotherapy showed improving lymphadenopathy.