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Case report

Extranodal pulmonary marginal zone B-cell lymphoma☆

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ABSTRACT

Aim: Extranodal mucosa-associated lymphoid tissue (MALT) arises a number of epithelial tissues, including the stomach, salivary gland, lung, small bowel, and elsewhere. Here we present a male patient with an uncommon site of extranodal MALT such as a pelvic mass diagnosed after a long period of evaluation, which initially presented with an incidental pulmonary nodule.

Method: We report a 60 years old man presenting with pulmonary nodules and consolidation. He refused invasive procedures and 3 years later was administered to our clinic with disseminated pulmonary nodules on chest X-ray. Subsequently a thin needle aspiration biopsy was performed and candida geotrichum was suspected in the specimen of the lung biopsy by light microscopic examination. After this time the patient was referred to our clinic, bronchoscopy, mediastinoscopy and abdominal computerized tomography (CT) scans were performed.

Results: Lymphoid hyperplasia was seen in the mediastinal lymph nodes biopsy specimens and the pelvic mass (52 × 18 mm) on the superior iliac muscles not related to any organs. Thin needle biopsy revealed MALT lymphoma and pathological examination of pulmonary nodule was similar to pelvic mass (MALT lymphoma). After the diagnosis, the thin needle biopsy of lung was repeated. The specimen appeared to be similar to the pelvic mass (MALT lymphoma) in the pathologic examination. The patient survived 5 years after initial diagnosis.

Conclusion: MALT has an affinity for the different tissues however has not been located in the pelvis. Our case represent an unusual presentation in a 60 years old man with lung and a pelvic mass.

1. Introduction

Extranodal marginal zone B-cell lymphoma arises in a number of epithelial tissues, such as the stomach, salivary gland, lung and small bowel. It was originally often referred to as a “pseudo-lymphoma” because of its tendency to remain localized to the tissue of origin for long periods of time, but it has been reported that it is a clonal B cell neoplasm that frequently recur locally and has potential for systemic spread and transformation to a high-grade B cell lymphoma.

The clinical presentation of extranodal marginal zone lymphoma (MZL) differs depending upon the tissue involved. Patients can present with symptoms of peptic ulcer disease, abdominal pain, Sicca/Sjögren's syndrome, or with a mass effect at the site of involvement [1,2]. MALTOMA is not commonly located in pelvic cavity, only 3 cases of MALTOMA of the bladder were reported previously [3]. Here we present a male patient with an uncommon site of extranodal MALTOMA diagnosed after a long period of evaluation, which initially presented with an incidental pulmonary nodule and existence of idiopathic thrombocytopenic purpura and

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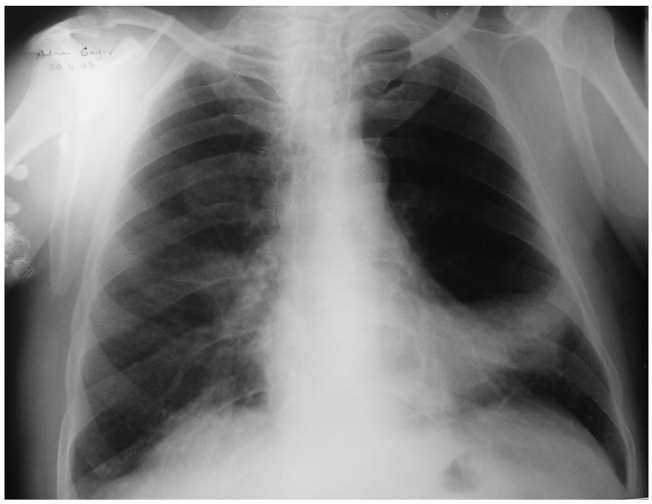

Fig. 1. Patient Chest X-Ray on admission.

pulmonary Candida Geothricum infection, which are the clues for malignancy.

2. Case report

In 2003 during a routine check-up of an asymptomatic man aged 60 years, a pulmonary opacity was seen on the P-A chest X-ray (Fig. 1). Subsequently the multislice thorax CT revealed a pulmonary nodules and consolidation. The patient refused the bronchoscopy and discontinued his follow-up in the outpatient clinic for four years. In 2006, he was diagnosed as having idiopathic thrombocytopenic purpura (ITP) in another center and steroid treatment was administered for one year, after which his thrombocytes returned to normal levels in hematology department. In late 2007 he was advised to undergo surgery for cholelithiasis. During the preoperative evaluation disseminated pulmonary opacities were seen on the chest X-ray and multiple pulmonary nodules and enlarged lymph nodes (maximum 21 mm) on the multislice thorax CT (Fig. 2); however, he had no complaint of respiratory system. Thin needle lung tissue aspiration biopsy was performed and microorganisms that mimicked Candida were revealed under light microscopic examination. After this evaluation he was re-administered to our department in January 2008. At that

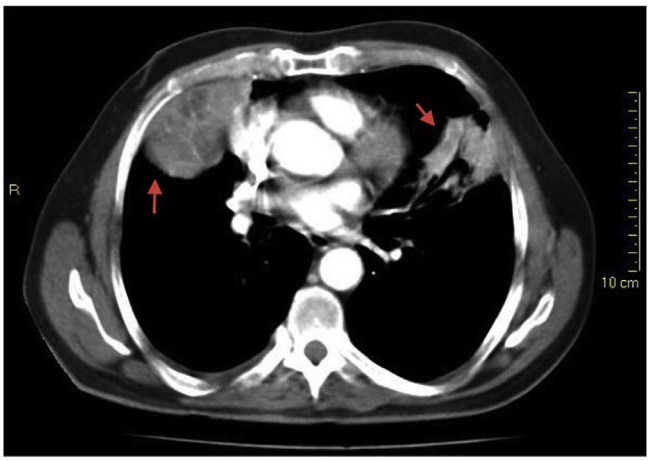

Fig. 2. Patient Thorax CT before diagnosis.

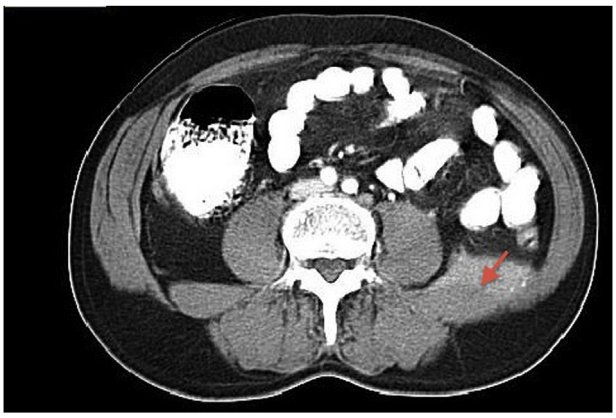

Fig. 3. Patient Pelvic CT with mass.

time our hospital did not have positron emission tomography (PET-CT) neither endobronchial ultrasound (EBUS) so conventional bronchoscopy and mediastinoscopy were performed; the bronchoalveolar lavage and brush specimen revealed no pathology; however, lymphoid hyperplasia was seen in the lymph node biopsy specimens of the mediastinum. No treatment was given for the candida infection because the clinical presentation was not related to a fungal infection.

An ITP relapse was diagnosed 1 month after the bronchoscopy and mediastinoscopy. The pathological examination of bone marrow aspiration and biopsy excluded hematologic malignancies and diagnosed again ITP. Steroid treatment was initiated and the thrombocyte level increased. To exclude lymphoproliferative diseases as a cause of immune thrombocytopenia, computed tomography of the abdomen was performed. In the abdominal tomography revealed a left pelvic mass (52 × 18 mm) on the superior iliac muscles (Fig. 3). Subsequently, the patient underwent a fine needle biopsy from the detected abdominal mass. Marginal zone B-cell lymphoma was diagnosed from the biopsy specimen. After the diagnosis, the fine needle biopsy of the lung was repeated. Examination of the pathological specimen was similar to the pelvic mass (marginal B cell lymphoma).

After the diagnosis 6 cures of CVP (cyclophosphamide, vincristine, prednisolone) On the follow up of the patient pelvic mass and pulmonary nodules regression was shown with the abdominal and

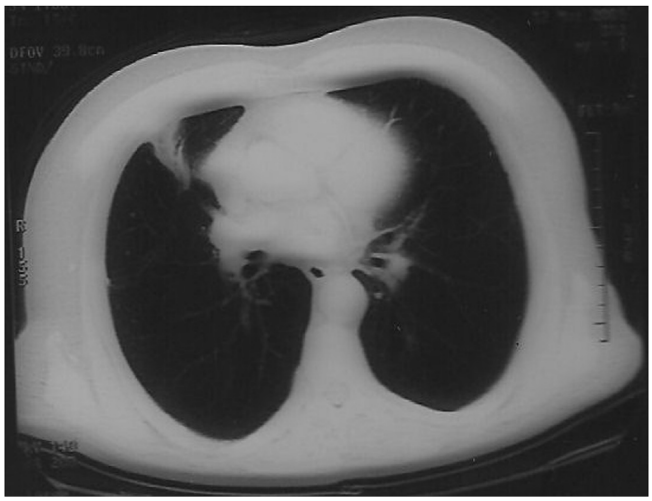

Fig. 4. Patient thorax CT after Chemotherapy [1].

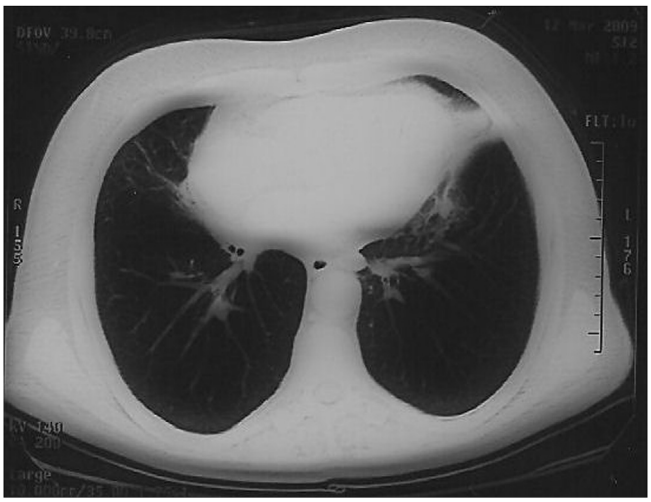

Fig. 5. Patient Thorax CT after Chemotherapy [2].

thorax CT, without any pulmonary and abdominal symptoms (Figs. 4 and 5). He died 5 years after the diagnosis of MALTOMA because of.

3. Discussion

Clinical presentation of extranodal MALTOMA differs according to the tissue involved and it may be asymptomatic [4,5]. Similarly there was no presenting symptom at the initial diagnosis of the pulmonary nodule n our patient, he also presented with an unusual localization of the extranodal MALTOMA without any symptom. To our knowledge this is the first case of extranodal MALTOMA involving the pelvic cavity but not confirmed to a specific organ.

The final diagnosis was unable to be made until 4 years after the incidental pulmonary nodule diagnosis due to the patient diagnosis refuse of invasive diagnostic procedures. His preoperative evaluation revealed nodular images on chest X-ray. The initial biopsy of the pulmonary nodule mimicked Candida Geothricum infection and this is usually found together with hematologic malignancies and immunosuppressed patients [6,7]. Steroid responsive ITP diagnosis was another clue. Therefore, we suspected a hematologic malignancy and performed the necessary evaluations (thorax CT, abdominal CT and bone marrow biopsy). Finally, we were able to diagnose extranodal MALTOMA within the pelvis. MALT lymphomas comprise 5% of non-Hodgkin lymphomas. There is no age prediction and a slight female predominance has been reported. Gastric involvement is the most frequent location of extranodal MALT lymphoma. Ocular adnexa involvement may compose up to 90% of the cases. Pulmonary involvement of the disease is reported to be 10% [8]. Fifty percent of patients with pulmonary disease are asymptomatic and 80% of patients who receive treatment are cured completely or partially. Peripheral blood disease and bone marrow involvement are seen together and solitary peripheral blood disease in MALT has not been reported. However, our patient had experienced several ITP attacks and there was no proven bone marrow association. To our knowledge the only case of pelvic MALTOMA was reported by Takara Y et al. [9]. However, their case was organ confined (bladder) while our case had lung involvement.

The treatment of extranodal MALTOMA depends principally upon the stage of disease at diagnosis.

In our case the disease had atypical presentation and multiorgan involvement. Patients with advanced stage disease are usually not cured with conventional treatment protocols and treatment therapies varies widely [10]. Chemotherapy has been administered to these patients as we did in our case. Remission was sustained in our patient with the CVP protocol.

To conclude, this rare case was initially referred to the hospital for routine check-up and incidentally diagnosed with a consolidation on the routine chest X-ray. Further investigation revealed an ITP and growth of Candida Geothricum in pulmonary specimen by thin needle aspiration sample. Given the paramalign characteristics of these findings, the patient was evaluated for further investigations, which finally was resulted in MALTOMA diagnosis with an usual localization and atypical presentation.

Conflict of interest

None.

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