Clinical Case Report

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Plasmablastic lymphoma exclusively involving bones mimicking osteosarcoma in an immunocompetent patient

A case report

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Abstract

Background: It has been known that plasmablastic lymphoma (PBL) is a neoplasm of immunocompromised patients occurring in soft tissue of oral cavity or in the vicinity whereas bone is an unlikely site to harbor PBL. However, its occurrence is increasingly being reported in immunocompetent individuals in either osseous or extra-oral sites. To our best knowledge, F-18 FDG PET/CT findings of PBL involving bones in an immunocompetent patient have not been reported, yet .

Case summary: We report a case of PBL involving multiple bones in an immunocompetent patient. Features of different imaging modalities including F-18 Fluoro-deoxy glucose (FDG) positron emission tomography/computed tomography (PET/CT) were correlated well as findings of osteosarcoma in mandible with metastatic lesions. However, the histopathology and immunohistochemistry (IHC) of bone tissues from 2 separate biopsy sites revealed features of PBL.

Conclusion: awareness to F-18 FDG PET/CT findings of PBL involving bones in an immunocompetent patient may prevent misdiagnosis.

Abbreviations: CD = cluster of differentiation, FDG = fluorodeoxyglucose, HHV = human herpes virus, HIV = human immunodeficiency virus, IHC = immunohistochemistry, Ki 67 = antigen Ki 67, a marker of proliferation, LANA 1 = latency-associated nuclear antigen, MDP = methyl diphosphonate, MRI = magnetic resonance imaging, PBL = plasmablastic lymphoma, PET/CT = positron emission tomography/computed tomography, PNS = paranasal sinus, SUV = standard uptake value, T2W = T2 weighted, WHO = World Health Organization.

Keywords: case report, FDG, osteosarcoma, PET/CT, plasmablastic lymphoma, skull

1. Introduction

Plasmablastic lymphoma (PBL), according to the World Health Organization (WHO)'s classification 2008, is characterized by diffuse proliferation of large neoplastic cells that has a morphologic resemblance to B immunoblasts with immonophenotype of plasma cells.[1] Immunophenotyping is the favored technique for a definite diagnosis since the first case of PBL was reported.[2] There has been a number of reports of PBL that occurred predominantly in adult males who are immuno-compromised due to human immunodeficiency virus (HIV) infection or due to solid organ transplantation with a predilection for soft tissue of oral cavity or in the vicinity.[3,4] However, its occurrence is increasingly being reported in immunocompetent individuals and also in osseous and extra-oral sites.[3,5–8] It is observed that there is less frequent association of extraoral PBL with HIV.[9,10] There are several reported cases of PBL with extraoral and osseous manifestation in addition to soft tissue PBL in immuno-competent patient .[5,6,8] No case has yet been reported PBL arising exclusively from osseous tissue in an immuno-competent person. We report a case of PBL that was confirmed by immunohistochemistry (IHC) from the biopsy of lesions in mandible and parietal bone in an immuno-competent patient.

2. Case report

A 38-year-old male had a right-sided sub-mental swelling and a right sided mandibular mass. His management was started with the working diagnosis of primary neoplastic lesion in right mandibular ramus and condyle with metastatic tumor in right posterior temporo parietal skull bone.

X-ray skull (Fig. 1) revealed round-shaped osteolytic lesion in right parieto temporal bone that was likely to be meatastatic

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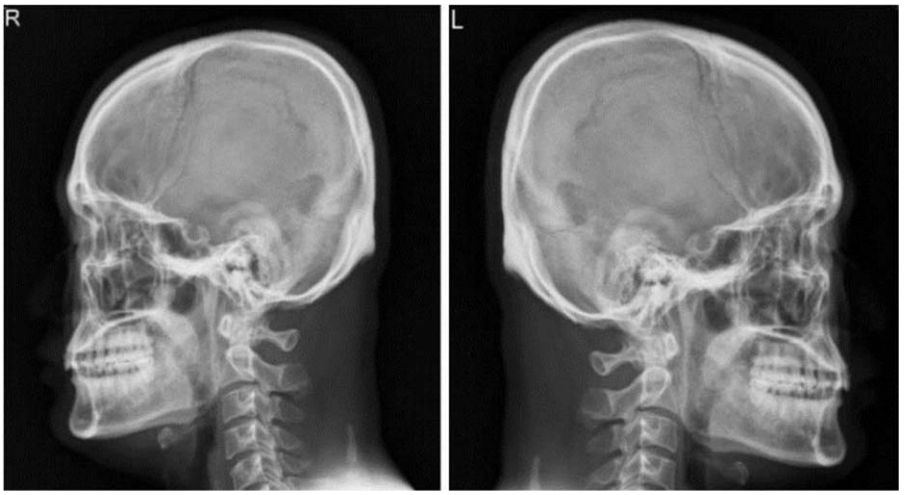

Figure 1. X-ray skull: round-shaped osteolytic lesion in the right parieto temporal bone.

lesion. Ultrasound of mandibular mass showed a mixed echogenic mass with minimal Doppler flow. Magnetic resonance imaging (MRI) of Paranasal sinus (PNS) (Fig. 2) showed soft tissue mass involving right mandibular ramus, angle, posterior part of body, and surrounding masticator space with T2-weighted (T2W) iso to slightly low-signal intensity, homogeneous enhancement, and sunburst periosteal reaction. MRI also revealed another soft tissue mass involving right supero-posterior parietotemporal bone. In addition, there was also diffuse infiltrative change in the subcutaneous spaces of the right neck which were more likely to be due to inflammatory change rather than tumor infiltration. A few indeterminate lymph nodes in level II on the right side were also noted.

PET/CT (Fig. 3) revealed hypermetabolic lesion on the right side of mandible with increased FDG uptake along periosteal reaction with maximum Standard Uptake Value (SUV) of 4.0. The mandibular lesion showed a sunburst appearance on CT. The lesion in right parietal bone was hypermetabolic with a maximum SUV of 3.02. In addition, hypermetabolic lesions were also seen in right scapula and left 9th rib with maximum SUV being 2.89 and 3.07, respectively. The lesion in right parietal bone including other 2 in scapula and rib appeared osteolytic on CT images. Skeletal scintigraphy (Fig. 4) showed increased Tc-99m-methyl diphosphonate (MDP) uptake in the mandible, right parietal bone, right scapula, and left 9th rib.

Based on the imaging findings osteosarcoma of mandible with multiple metastases in parietal bone, scapula and rib were the most likely differential diagnosis. Biopsy specimen from mandible and parietal bone lesions were taken for histopathological examination. The histopathology and IHC features of both tumors were compatible with plasmablastic lymphoma; HIV serology in the patient was negative.

In situ hybridization of the tumor cells revealed most of the cells to be positive for Epstein Burr virus. Human herpes virus (HHV)-8 latency-associated nuclear antigen (LANA 1) was found negative. IHC of the tumor cell revealed their positivity for Cluster of differentiation (CD) 38, CD138 and CD79α. Ki67 antigen was also positive in 20% tumor cells. The tumor cells were found to be negative for CD45, CD56, and CD20. These features were compatible with the WHO criteria 2008 for PBL.

Patient was then treated with chemotherapy in the protocol of non-Hodgkin's lymphoma for the treatment of PBL.

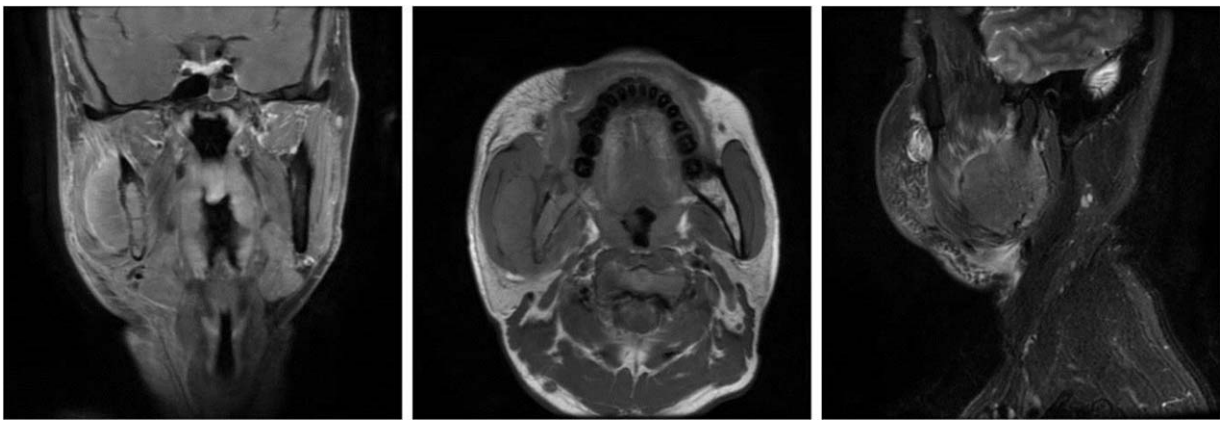

Figure 2. MRI of PNS: soft tissue mass involving right mandibular ramus, angle, posterior part of body and surrounding masticator space, soft tissue mass involving right supero-posterior parieto temporal bone, diffuse infiltrative change in the subcutaneous spaces of the right neck and a few indeterminate lymph nodes in level II on the right side. MRI = magnetic resonance imaging, PNS = paranasal sinus.

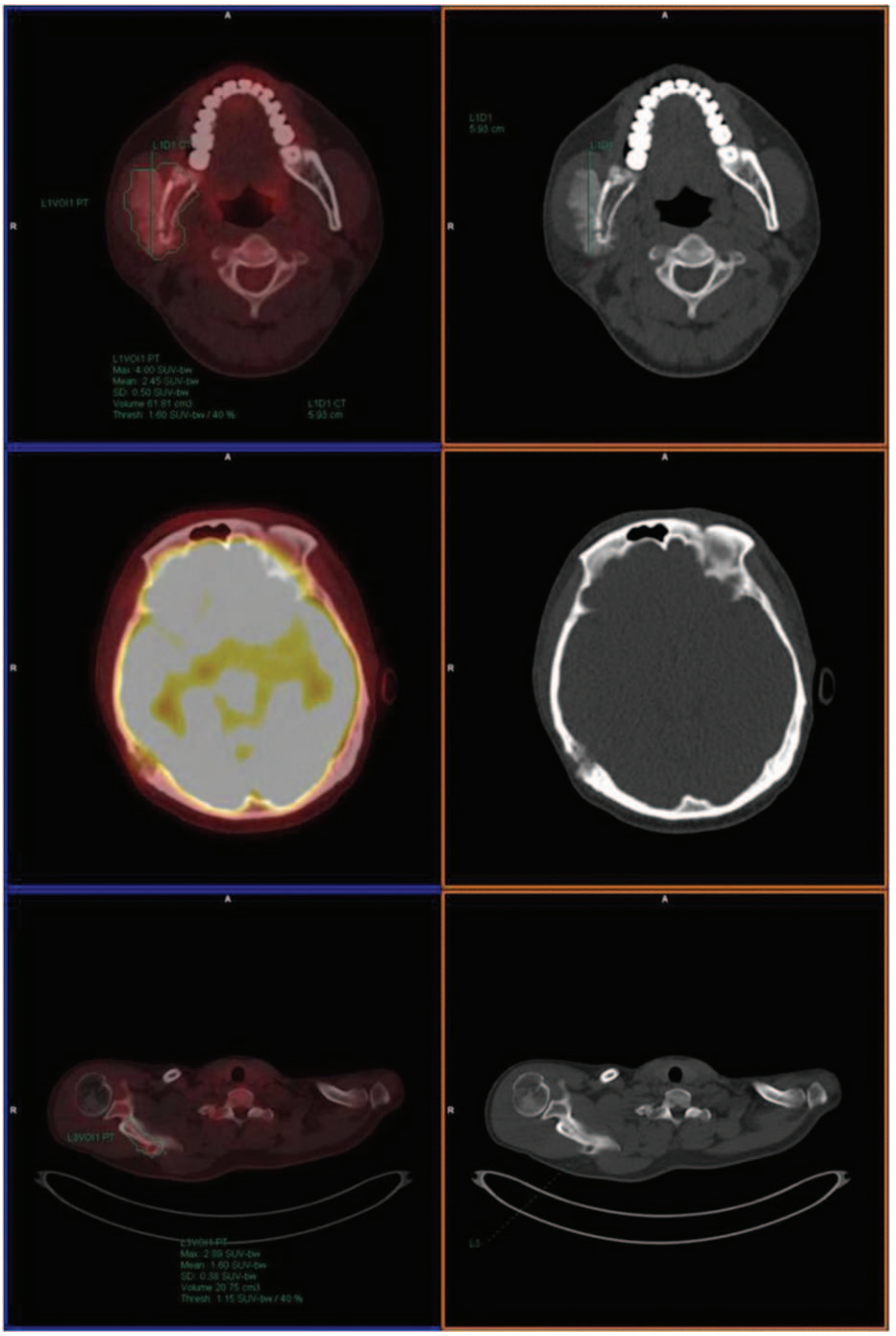

Figure 3. F-18 FDG PET/CT: hypermetabolic lesion on the right side of mandible, with sunburst appearance and increased FDG uptake along the periosteal reaction. Hypermetabolic lesions in the right parietal bone and right scapula. FDG = fluorodeoxyglucose, PET/CT = positron emission tomography/computed tomography.

3. Materials and methods

Approval from institutional review board of Seoul National University Hospital was obtained.

4. Discussion

In this current case, an initial differential diagnosis of osteosarcoma was made based on hybrid imaging. The reason for this was high FDG uptake on PET combined with classic feature of osteosarcoma, sunburst appearance,[11] in mandible. Patient's age and site of lesion were also compatible with the diagnosis. The immuno-competent state of the person prevented the clinical minds to think about PBL. Also compatible with the existing knowledge of tumor metabolism, osteosarcoma like most other malignancies demonstrates increased FDG uptake on PET images.[12] Osteosarcoma and other malignant bone lesions tend to have SUV of >2.[13] Some recent case series reports SUV of osteosarcoma ranging from 3.15 to 16.5.[13,14] Remarkable FDG uptake is reported in osteolytic bone metastases[15,16] which is compatible with the fact that lesions in parietal bone, scapula, and rib appeared osteolytic on CT images while demonstrating increase FDG uptake on PET. However, there are reports of bone

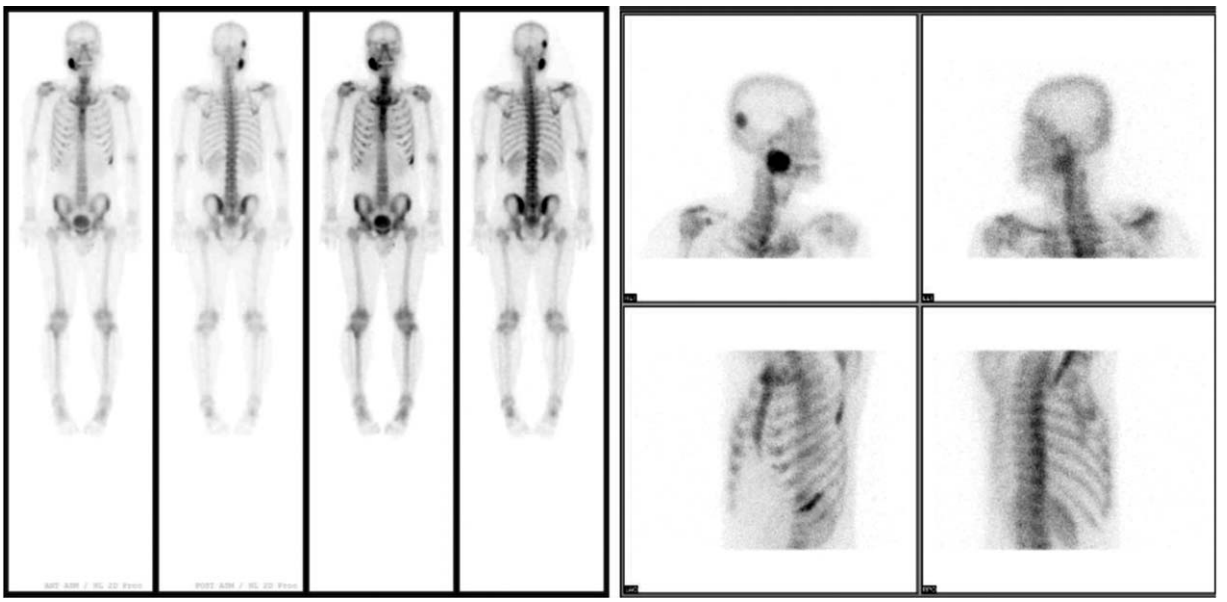

Figure 4. Tc-99m-MDP bone scan: increased tracer uptake in the mandible, right parietal bone, right scapula, and left 9th rib. MDP = methyl diphosphonate.

metastases from osteosarcoma that displayed low uptake of FDG on PET scan[17] or was false negative.[18]

In the current case, all 4 bone lesions were hypermetabolic with maximum SUV between 3 and 4. There are few reported cases of soft tissue PBL with hypermetabolic activity on PET images.[6,19,20] Cazaentre et al[21] reports a case of soft tissue PBL showing no abnormal FDG uptake but associated hypermetabolism in retroperitoneal lymph node with SUV of 7.5.

CT images of the current case showed that among the bone lesions the 1 in mandible was osteoblastic whereas the other 3 were osteolytic. In the reported cases of PBL either the lesions themselves have osteolytic appearance on CT scan[21–23] or there was CT evidence of osteolysis adjacent to soft tissue lesions.[6,21,22,24–26]

Though there is a report of osteolytic appearance of PBL on bone scan,[22] in this current case, concordant with the FDG uptake on PET images, bone scan demonstrated increased MDP uptake in all 4 bone lesions.

5. Conclusion

The present case reports F-18 FDG PET/CT findings of a case of PBL arising from bone in an immunocompetent patient. Although the definite diagnosis of PBL is reliant on IHC, awareness to possible diverse finding on PET/CT images can prevent misdiagnosis.

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