Clinical Case Report

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Sinonasal oncocytic Schneiderian papilloma accompanied by intravascular lymphoma

A case report on FDG-PET/CT imaging

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Abstract

Introduction: F-18 fluorodeoxyglucose positron emission tomography/computed tomography (FDG-PET/CT) is useful for the staging and assessment of treatment response in patients with lymphoma. Occasionally, benign lesions demonstrate avid FDG uptake and result in false positive findings.

Case: We report the case of an 82-year-old man presenting with cutaneous lesions, which were histopathologically diagnosed as intravascular lymphoma. FDG-PET/CT for staging demonstrated an FDG-avid mass extending from the right maxillary sinus to the nasal cavity, moderate uptake in the adrenal glands, mild uptake in the knee and the foot, and faint uptake in the skin and subcutaneous tissue of the legs. He subsequently underwent biopsy of the paranasal mass, which was diagnosed as oncocytic Schneiderian papilloma without lymphoma invasion. Glucose transporter (GLUT) 1 staining was highly positive in the papilloma cells, resulting in high FDG avidity. After completion of chemotherapy, the abnormal FDG uptakes in the skin, soft tissue, and adrenal glands disappeared on PET/CT. However, avid FDG uptake persisted in the sinonasal Schneiderian papilloma for 15 months before regression.

Conclusion: Benign tumors with oncocytic components may show avid FDG uptake. Therefore, correct diagnosis of oncocytic Schneiderian papilloma on FDG images is difficult when other accompanying malignant tumors, especially lymphoma, are present. If post-therapeutic PET/CT images show a discordant lesion, oncocytic tumors, albeit uncommon, should be considered in the differential diagnoses.

Abbreviations: FDG-PET/CT = F-18 fluorodeoxyglucose positron emission tomography/computed tomography, GLUT = glucose transporter, LDH = lactate dehydrogenase, MIP = maximum intensity projection, R-CHOP = rituximab-cyclophosphamide, doxorubicin, vincristine, and prednisolone, sIL-2R = soluble interleukin-2 receptor, SUV_{max} = maximum standardized uptake value.

Keywords: FDG-PET/CT, intravascular lymphoma, oncocytic Schneiderian papilloma

1. Introduction

F-18 fluorodeoxyglucose positron emission tomography/computed tomography (FDG-PET/CT) is useful for the staging and assessment of treatment response in patients with lymphoma. Occasionally, benign lesions demonstrate avid FDG uptake and result in false positive findings. We experienced a case of oncocytic Schneiderian papilloma, the rarest type of sinonasal papilloma,[1,2] which mimicked a lymphoma lesion with high FDG uptake.

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2. Case report

2.1. Ethics review and patient consent

This retrospective study dealt only with the patient's medical records and related images. Ethics committee approval was not thought to be necessary because the entire clinical course of the case was within standard medical care. Informed consent on diagnostic examinations and therapeutic procedures was given by the patient.

2.2. Case

An 82-year-old man presented with several months of erythema on the legs, which was diagnosed as erythema nodosum. The lesions resolved by steroid therapy, but progressed after withdrawal of the therapy. Thereafter, he complained of edema on the lower abdomen and lower extremities, accompanied by fever. Splenomegaly and elevated serum levels of lactate dehydrogenase (LDH) and soluble interleukin-2 receptor (sIL-2R) were noted. These symptoms spontaneously remitted, but relapsed after several months. No significant weight loss was noted.

When he was referred to our hospital, mottled erythema and edema were found on both legs. Laboratory evaluation revealed elevated serum levels of LDH (770 U/L; normal range, 124–222 U/L) and sIL-2R (564 U/mL; normal range, 145–519 U/mL). With a suspicion of lymphoma, he underwent FDG-PET/CT (Fig. 1), which showed a soft tissue mass with increased FDG uptake (maximum standardized uptake value [SUV_{max}], 13.7) extending

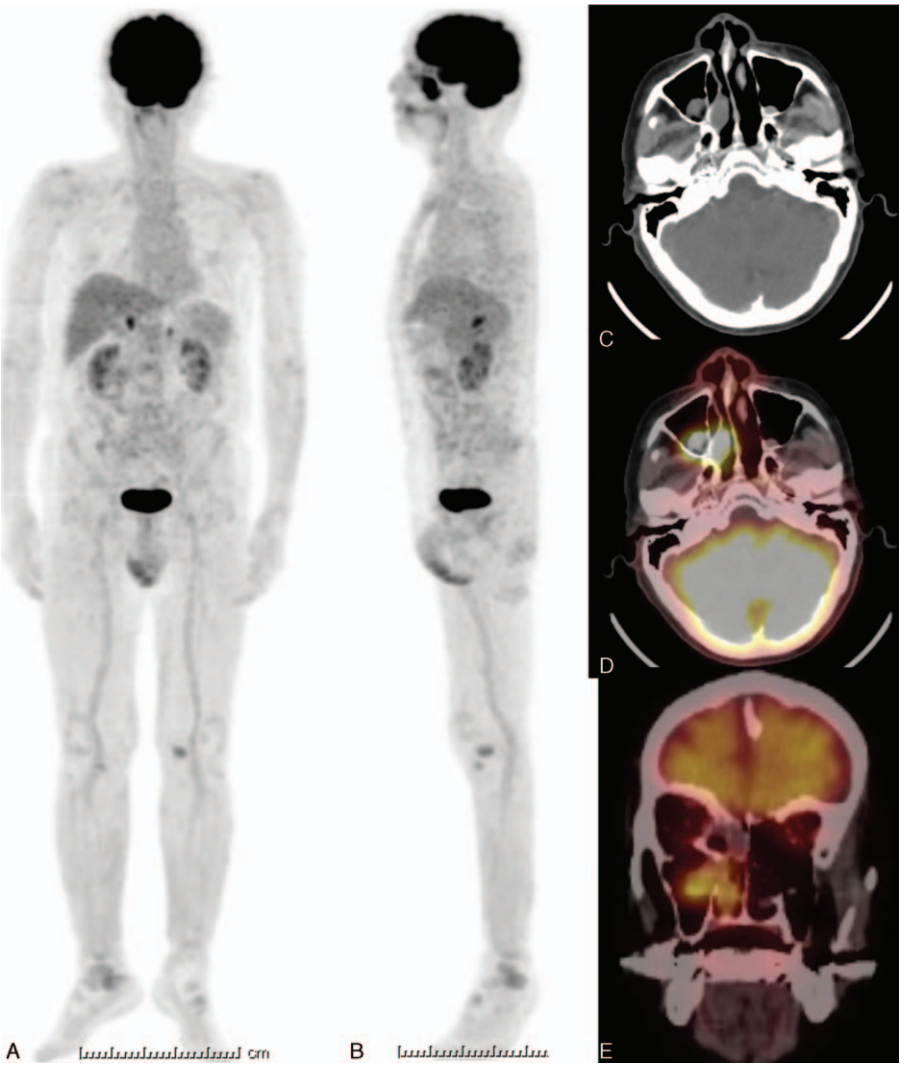

Figure 1. On PET/CT (A, anterior and B, lateral MIP) and CT (C, plain; D, transaxial fusion; and E, coronal fusion) images, a soft-tissue mass with significantly increased FDG uptake (SUV_{max}, 13.7) is seen extending from the right maxillary sinus to the lateral wall of nasal cavity. On whole body PET scan (A and B), increased FDG uptake is also noted in the adrenal glands (SUV_{max}, right, 5.6; left, 3.9), medial condyle of the left femur (SUV_{max}, 3.1), medial condyle of the right tibia (SUV_{max}, 2.2), and the tarsal bones (SUV_{max}, right, 3.0; left, 2.2). Subtle FDG uptake (SUV_{max} up to 1.5) is shown in the skin and subcutaneous tissue of the legs. FDG = F-18 fluorodeoxyglucose, MIP = maximum intensity projection, PET/CT = positron emission tomography/computed tomography, SUV_{max} = maximum standardized uptake value.

from the right maxillary sinus to the lateral wall of the nasal cavity. This lesion was highly suggestive of a malignant process, probably lymphoma. Lesions with abnormal FDG uptake were also noted in the bilateral adrenal glands, medial condyle of the left femur, medial condyle of the right tibia, and the tarsal bones, predominantly on the right. These lesions were suspected to be invasion of lymphoma. Faint uptake was observed in the skin and subcutaneous tissue of the legs.

Skin biopsy on the leg demonstrated infiltration of atypical large lymphoid cells in the small vessels of the skin and subcutaneous fat tissue. Immunohistochemical staining for CD20 was positive. Based on these findings, intravascular large B-cell lymphoma was proven (Fig. 2). However, cytology of the cerebrospinal fluid was negative for malignancy. On the other hand, the histopathologic examination of the sinonasal mass revealed oncocytic Schneiderian papilloma or cylindrical cell papilloma (Fig. 3A). There was no evidence of lymphoma cell invasion. Immunohistochemistry staining for glucose transporter (GLUT) 1 was performed with anti-GLUT1 rabbit polyclonal antibody (IBL, Gunma, Japan) and N-Histofine Simple Stain MAX PO (Nichirei Biosciences Inc., Tokyo, Japan), and showed high positivity in the papilloma cells (Fig. 3B); these findings explained the high FDG avidity of the sinonasal mass.

He underwent chemotherapy with 8 cycles of rituximab plus 6 cycles of cyclophosphamide, doxorubicin, vincristine, and prednisolone (R-CHOP) for 5 months, including 4 cycles of intrathecal injection of methotrexate, cytarabine, and prednisolone. Grade 3 to 4 neutropenia was noted during each cycle and was treated with granulocyte colony stimulating factor. After completion of chemotherapy, the abnormal uptake in the adrenal glands and bones disappeared on PET/CT (Fig. 4). Edema on both legs improved. The lymphoma lesions responded well to the chemotherapy and were considered to be in complete remission. However, the sinonasal mass persisted to have avid FDG uptake (SUV_{max}, 23.8) and slight morphologic deformity. There was a slightly enlarged component in the maxillary sinus, which was

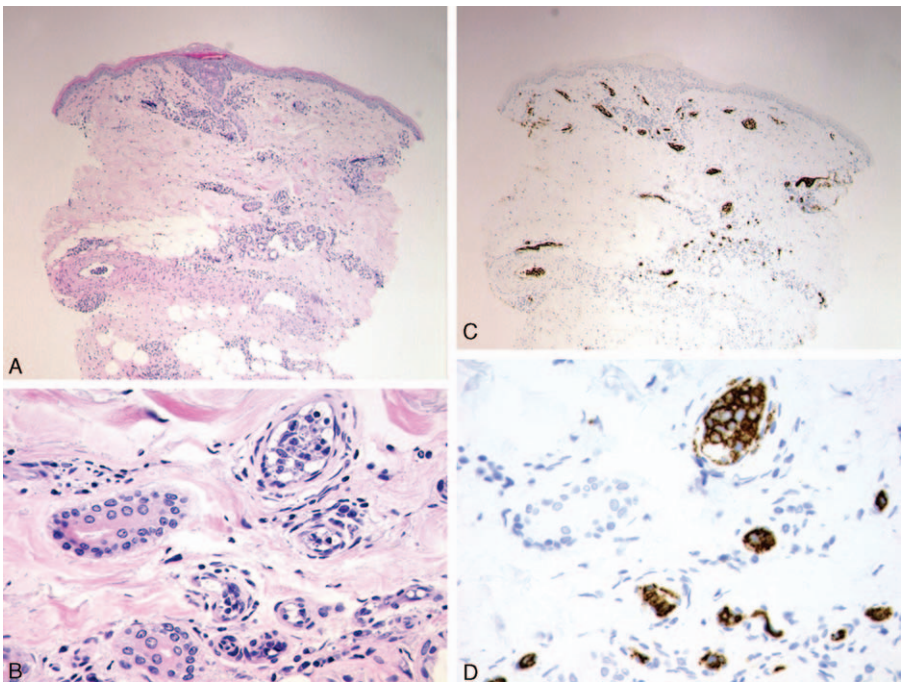

Figure 2. Skin biopsy of the leg demonstrates infiltration of large atypical lymphoid cells in the small vessels and subcutaneous fat tissue (hematoxylin and eosin. A, × 4; B, × 40). Immunohistochemical staining for CD20 was positive (C, × 4; D, × 40).

considered to be an inflammatory process, and a somewhat shrunken component in the nasal cavity, probably due to the previous biopsy. We confirmed regression of the sinonasal mass after 15 months.

3. Discussion

Sinonasal papillomas are categorized into 3 histologic types:[1] (1) inverted; (2) fungiform; and (3) cylindrical cell papilloma or oncocytic Schneiderian papilloma, as proposed by Barnes and Bedetti.[2] Oncocytic Schneiderian papilloma is the rarest of these types, accounting for 3% to 5% of the total, and typically involves the maxillary sinus or in conjunction with the lateral wall of the nasal cavity or ethmoid sinus. This tumor has a tendency for recurrence, if inadequately excised, and for malignant transformation in up to 17% of patients.[1,2] Oncocytic Schneiderian papilloma is known to show high FDG uptake despite its benign nature.[3–5] The etiology of increased FDG uptake is poorly understood, but it is hypothesized that oncocytic tumors possess a high number of mitochondria that cause increased metabolism and uptake of metabolites, such as FDG.[6] In this case, we demonstrated GLUT1 expression in the sinonasal tumor, which could explain the increased FDG uptake. Benign tumors with oncocytic components sometimes show avid FDG uptake and may present incidentally.[7,8] Therefore, it is difficult to rule out malignancy on FDG images when these are accompanied by other malignant tumors, especially systemic malignancy, such as lymphoma.

Cutaneous lesions are the dominant presenting feature in 39% of intravascular lymphomas.[9] Our case presented with cutaneous lesions that were histologically confirmed as intravascular lymphoma. Furthermore, a large majority (83%) of patients with intravascular lymphoma showed positive findings on biopsy of normal-appearing skin.[10] In patients with suspected intravascular lymphoma, random skin biopsy has been proposed, even in the absence of evident skin lesions, because diagnosis based on biopsy of tissues other than the skin is usually difficult.[11]

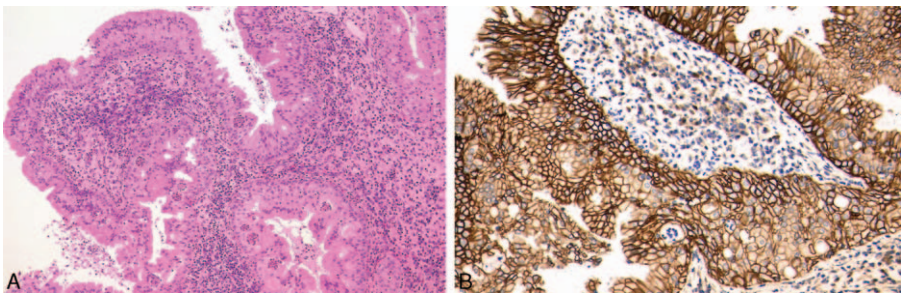

Figure 3. Histologically, the sinonasal mass shows papillary structures covered by columnar cells with eosinophilic cytoplasm, compatible with oncocytic Schneiderian papilloma or cylindrical cell papilloma (A, hematoxylin and eosin, × 10). There is no evidence of lymphoma cell invasion. Immunohistochemistry staining for GLUT1 is strongly positive on the membrane of tumor cells (B, × 20). GLUT = glucose transporter.

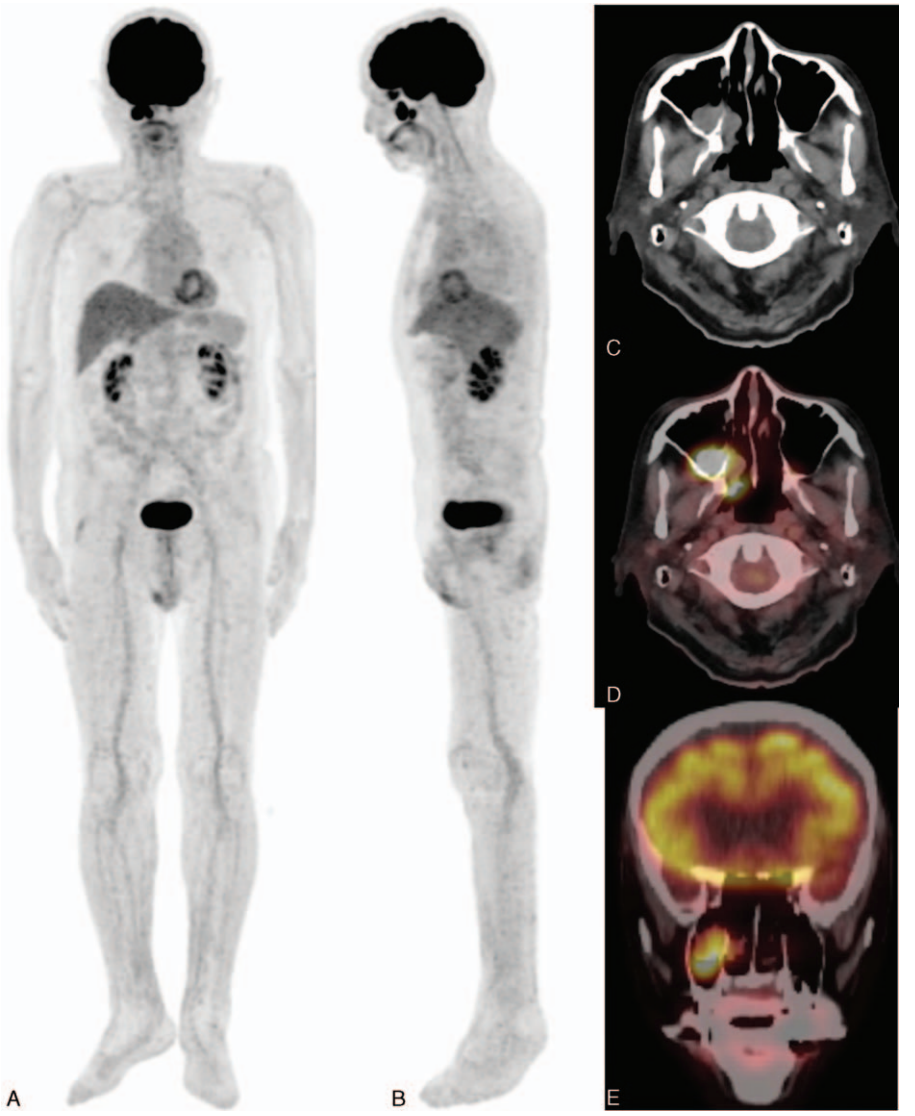

Figure 4. After completion of chemotherapy, the abnormal uptakes in the skin, soft tissue, and adrenal glands disappeared on PET/CT (A and B). However, the sinonasal mass had persistent avid FDG uptake (SUV_{max}, 23.8) and slight morphologic deformity (C, D, and E). FDG = F-18 fluorodeoxyglucose, PET/CT = positron emission tomography/computed tomography, SUV_{max} = maximum standardized uptake value.

4. Conclusion

In oncocytic Schneiderian papilloma, GLUT1 was highly expressed and resulted in high FDG avidity on PET/CT images. When accompanied by malignancy, such as lymphoma, oncocytic tumors are difficult to diagnose correctly on pre-therapeutic PET/CT images. If post-therapeutic PET/CT images show a discordant lesion, oncocytic tumors, albeit uncommon, should be considered in the differential diagnoses.

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