CASE REPORT

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Selective intra-arterial Y-90 microsphere therapy in hemangioendothelioma

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Epithelioid hemangioendothelioma is a malignant soft tissue tumor originating from the endothelial cell. Its clinical course varies from benign hemangioma to angiosarcoma. The therapy procedure remains unclear. Although resection of the tumor is the most frequently suggested treatment, due to its multinodular type, transplantation is frequently performed. Therapy regimens other than transplantation and resection are not effective and the median survival in hemangioendothelioma with unresectable lesions is approximately eight months. In this case, we report the therapeutic effect of selective intra-arterial radionuclide yttrium (Y)-90 microspheres and the value of F-18 fluorodeoxyglucose positron emission tomography/computed tomography to evaluate therapy response in hemangioendothelioma.

Key words: Hemangioendothelioma, intra-arterial radionuclide therapy with yttrium (Y)-90 microspheres, F-18 fluorodeoxyglucose positron emission tomography/computed tomography

Hemanjioendoteliomada intraarteriyel radyonüklid tedavi

Epiteloid hemajioendotelioma, endotelial hücrelerden köken alan yumuşak doku malign tümörüdür. Klinik bulguları hemanjiomdan anjiosarkoma kadar değişir.Tedavi rejimi henüz net değildir. Rezeksiyon en sık önerilen yöntem olmasına rağmen, multinodüler olgularda transplantasyon sıklıkla uygulanmaktadır. Transplantasyon ve rezeksiyon haricindeki tedavi şekilleri etkili değildir ve anrezektabl epiteloid hemajioendotelioma olgularında ortanca yaşam süresi 8 aydır. Bu olguda, epiteloid hemajioendotelioma olgularında intrarateriyel radyonuklid Y-90 mikroküre tedavisinin etkinliği ve tedavi cevabının değerlendirmesinde F18 florodeoksiglukoz pozitron emisyon tomografi/bilgisayarlı tomografi'nin yeri değerlendirilmiştir.

Anahtar kelimeler: Hemangioendothelioma, Y-90 mikroküre ile intraarterial radionuclid tedavi, F18 FDG PET/CT

INTRODUCTION

Hemangioendothelioma (HE) is a rare tumor originating from the vascular system. It is usually observed in soft tissue and bone and infrequently in the liver and spleen (1,2). It is an intermediate entity among well-differentiated hemangioma and angiosarcoma. The median survival of HE with unresectable lesions is approximately eight months.

The clinical manifestations are nonspecific: upper abdominal pain, anorexia, weight loss, and hepatosplenomegaly. Computed tomography (CT) is commonly used to evaluate HE. Although peripheral confluent masses with capsular retraction and evidence of multiple calcifications are observed in CT (3,4), findings of recurrent tumor have not been well described. The value of F-18 fluorodeoxyglucose positron emission tomography (FDG PET)/CT in detecting primary lesions and recurrence was recently reported (5-7).

The therapy regimen is still unclear. While resection is the most adequate treatment, transplantation is commonly performed, since HE often presents in a multinodular diffuse type (8-11). Results of chemotherapy and radiotherapy are not

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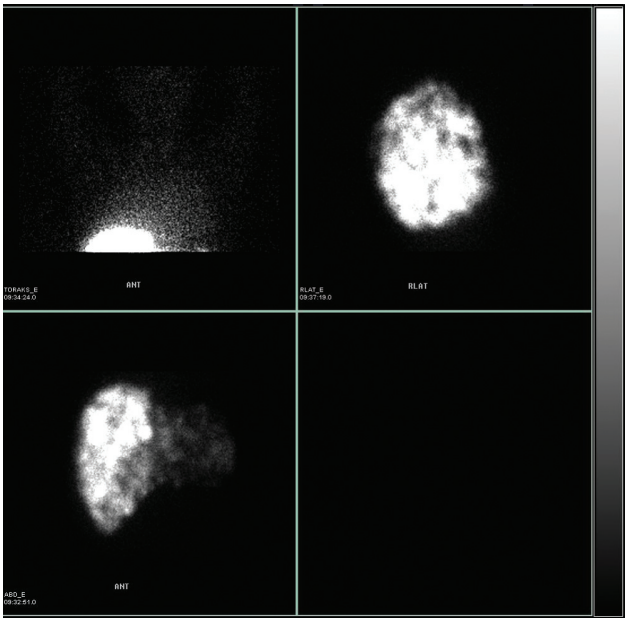

Figure 1. Shunt analysis from liver to lung was evaluated with Tc99m MAA given intra-arterially during the angiography. The shunt was calculated as <10%.

encouraging. Interferon-alpha 2 is still being experimented (10). Selective intra-arterial radionuclide therapy has not been reported for HE therapy previously.

CASE REPORT

A 39-year-old male with unresectable multiple liver lesions and proven HE with liver biopsy was evaluated for selective intra-arterial radionuclide yttrium (Y)-90 microsphere therapy. Liver transplantation could not be performed due to multinodular lesions. Despite his undergoing chemotherapy and alpha-2 interferon, progression in his lesions was observed. His biochemical tests including liver and kidney functions were normal. Multiple liver lesions and hepatomegaly were observed in F-18 FDG-PET/CT scan before the therapy. The angiogram was performed two weeks prior to the therapy to check the vascular system, and the coils were used where necessary. The shunt from the liver to the lung evaluated with Tc-99m micro-aggregated albumin (MAA) was less than 10% before the therapy (Figure 1).

Microsphere therapy (1.6 GBq Y-90) was administered selectively from the hepatic artery to the right lobe and 1 GBq to the left lobe in three months. No significant side effects were observed. Initiation of the clinical improvement was seen in two weeks. The metabolic response was observed in the first PET/CT scan six weeks after therapy (Figs. 2-5). His performance status improved. The follow-up period of this patient was 18 months, and he died due to peritoneal involvement and liver insufficiency.

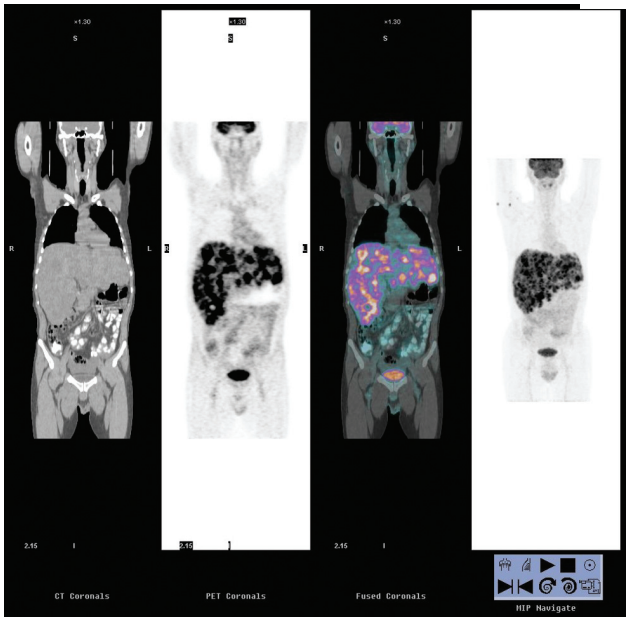

Figure 2. F-18 FGD PET/CT before the therapy. Hepatomegaly and multiple focal hypermetabolic uptake were observed.

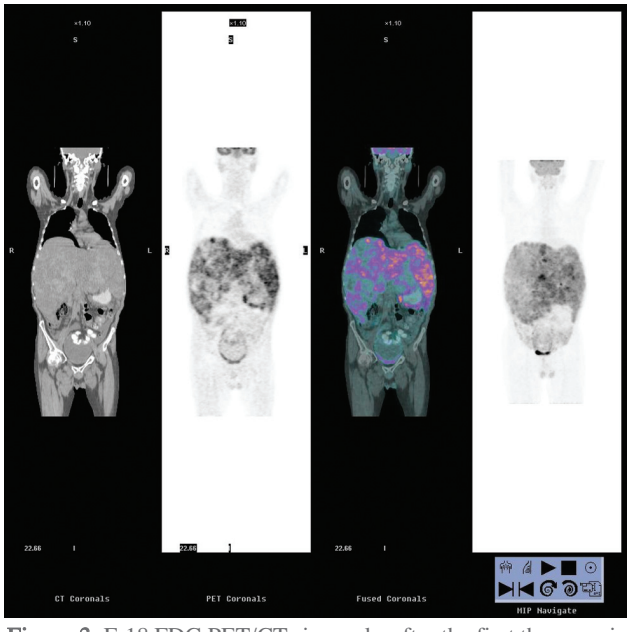

Figure 3. F-18 FDG PET/CT six weeks after the first therapy given to the right lobe (1.6 GBq) intra-arterially. The metabolic activity of the lesions in the right lobe was significantly decreased compared to that of lesions in the left lobe.

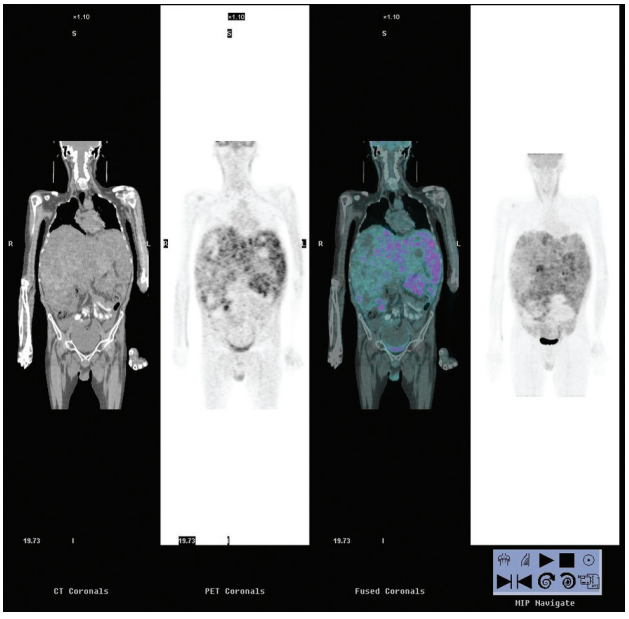

Figure 4. F-18 FDG PET/CT six weeks after the second therapy given to the left lobe (1.0 GBq) intra-arterially. The uptake of the right lobe was still significantly decreased. The uptake of the left lobe's lesions became less metabolic than before the therapy.

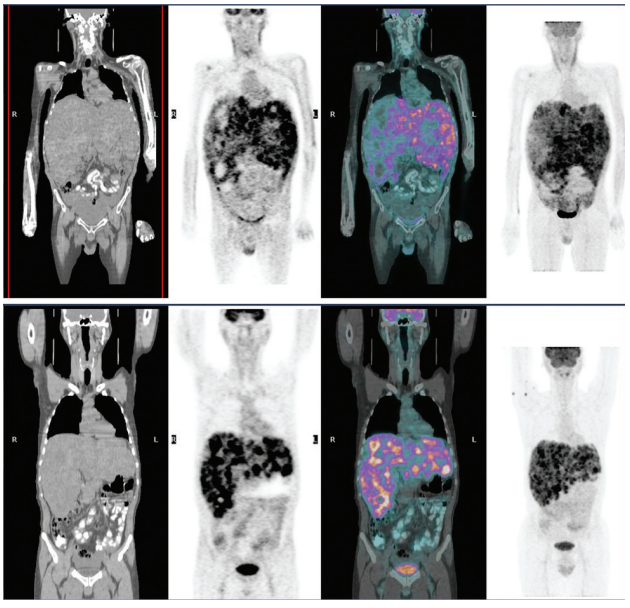

Figure 5. Comparative transaxial images of before (A) and after (B) selective intra-arterial radionuclide therapy.

In conclusion, selective intra-arterial radionuclide therapy is commonly used for unresectable liver metastases of colorectal cancer and primary liver tumors such as hepatocellular cancer (12,13). The reported results were hopeful; therefore, selective intra-arterial radionuclide therapy with Y-90 is recommended with first-line chemotherapies. Unresectable liver tumors showing metabolic activity in F-18 FDG PET/CT and enhancement of tumor with contrast medium on CT can be treated with this therapy. The most important parts of the pretreatment evaluation with this therapy are biochemical tests, especially bilirubin levels and liver functions, and shunt from liver to lung. The liver function tests should be less than 5 times the upper limit of normal levels and bilirubin should be <2 mg/dl. The shunt analysis is performed with Tc 99m MAA administered to the hepatic artery during the angiography. The images are obtained and quantitative evaluation is done. The shunt should be <20%. In this patient, the biochemical tests and shunt analysis were acceptable for therapy.

Selective intra-arterial radionuclide Y-90 microsphere therapy seemed to be an effective therapy method for unresectable HE. It should be kept in mind as a choice for successful therapy in HE patients.

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