

¹⁸F-FDG and ⁶⁸Ga-FAPI-04 PET/CT Findings of a Rare Epithelialmyoepithelial Carcinoma Arising From Ex Pleomorphic Adenoma of Parotid

Parotidin Eski Pleomorfik Adenomundan Kaynaklanan Nadir Bir Epitelyal-miyoepitelyal Karsinomun ¹⁸F-FDG ve ⁶⁸Ga-FAPI-04 PET/CT Bulguları

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Abstract

Epithelial-myoepithelial carcinoma (EMC) is a rare low-grade salivary gland neoplasm. Distant metastasis is rare, and ¹⁸F-fluorodeoxyglucose positron emission tomography/computed tomography (¹⁸F-FDG PET/CT) has been used to determine the metastatic disease in EMC. ⁶⁸Ga-fibroblast activation protein inhibitors (FAPI) PET/CT is a promising imaging modality for diagnostic and theognostic purposes in various malignancies. Comparison studies with ¹⁸F-FDG have investigated the role of ⁶⁸Ga-FAPI PET/CT. Herein, we present ¹⁸F-FDG and ⁶⁸Ga-FAPI-04 PET/CT findings of a 51-year-old woman with metastatic EMC arising from ex-pleomorphic adenoma of the parotid.

Keywords: 68Ga-FAPI, 18F-FDG, PET/CT, epithelial myoepithelial carcinoma

Öz

Epitelyal-miyoepitelyal karsinom (EMK), nadir görülen düşük gradlı tükürük bezi neoplazmıdır. Uzak metastazlar nadir görülmekte olup EMK'de metastatik hastalığı tespit etmek için ¹⁸F-florodeoksiglukoz pozitron emisyon tomografisi/bilgisayarlı tomografi (¹⁸F-FDG PET/BT) kullanılmaktadır. ⁶⁸Ga-fibroblast aktivasyon proteini inhibitörleri (FAPI) PET/BT, çeşitli malignitelerde tanıda ve teranostik amaçla kullanılan umut verici bir görüntüleme yöntemidir. ¹⁸F-FDG PET/BT ile karşılaştırmalı yapılan çalışmalar ⁶⁸Ga-FAPI PET/BT'nin rolünü araştırmaktadır. Bu olguda parotisin eks pleomorfik adenomundan kaynaklanan metastatik EMK tanılı 51 yaşında kadın hastanın ¹⁸F-FDG ve ⁶⁸Ga-FAPI-04 PET/BT bulgularını sunuyoruz. **Anahtar kelimeler:** ⁶⁸Ga-FAPI, ¹⁸F-FDG, PET/BT, epitelyal miyoepitelyal karsinom

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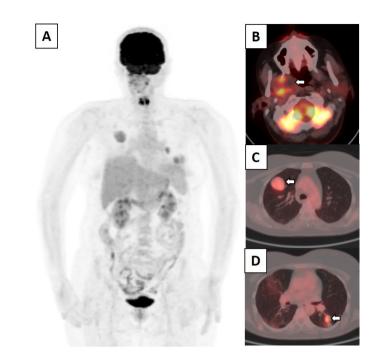


Figure 1. A 51-year-old woman with a history of parotidectomy due to pleomorphic adenoma five years ago was referred for ¹⁸F-fluorodeoxyglucose positron emission tomography/computed tomography (¹⁸F-FDG PET/CT) due to suspicious lung nodules. Maximum intensity projection (A: MIP) and transaxial fused ¹⁸F-FDG PET/CT images showing a hypermetabolic mass located in the right parapharyngeal region (B: arrow), in addition to multiple lung lesions with mild to moderate FDG uptake (C, D: arrows).

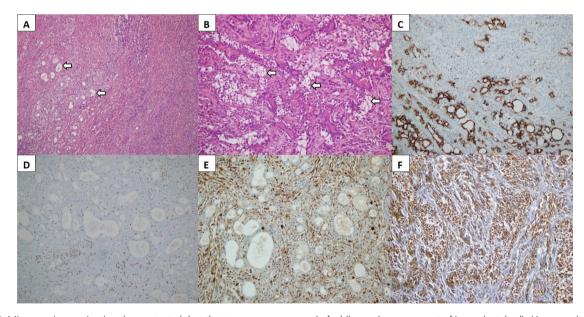


Figure 2. Microscopic examination demonstrated that the tumor was composed of a bilayered arrangement of inner ductal cells (A: arrows) and outer myoepithelial cells (B: arrows). The inner luminal cells were immunoreactive for cytokeratin 7 (C) and EMA, whereas the outer myoepithelial layer exhibited p63 (D), S100 expression (E), and calponin (F). Histomorphological and immunohistochemical findings confirmed the diagnosis of epithelial-myoepithelial carcinoma (EMC). In the literature, few cases reports have demonstrated distant metastasis in EMC, and therapy management remains unclear based on the limited efficacy results of recommended therapies (1,2). Because the tumors had mild to moderate FDG avidity and therapy options were limited, ⁶⁸Ga-fibroblast activation protein inhibitors-04 (⁶⁸Ga-FAPI-04) PET/CT was planned to improve diagnostic accuracy and assess eligibility for radionuclide therapy.

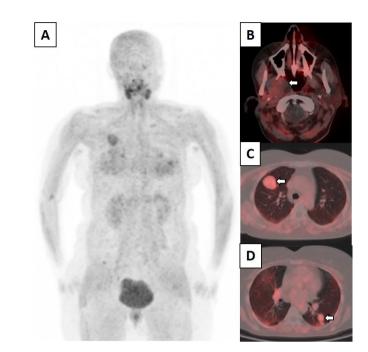


Figure 3. ⁶⁸Ga-FAPI-04 PET/CT (A: MIP) showed no significant uptake in the parapharyngeal mass (B: transaxial fusion, arrow), and only mild uptake was demonstrated in the lung lesions (C, D:, arrows). The patient was referred to chemotherapy regime (capecitabine + cisplatin). EMC is a very rare low-grade neoplasm of the salivary gland in which distant metastasis has been reported in only 4.5% of all cases (3). The histological diagnosis of EMC could be challenging because other benign salivary gland tumors present similar features (4). EMC mostly shows high FDG avidity based on the case reports, and it should be considered for differential diagnosis, especially in patients with a history of salivary gland surgery (5). Distant metastasis of EMC has been demonstrated in only a few case reports, and therapy options are uncertain due to the limited results in the literature (1,2). Recently, ⁶⁸Ga-FAPI PET/CT has opened up new opportunities, including the theognostic approach using cancer-associated fibroblasts (6). Dendl et al. (7) emphasized the value of ⁶⁸Ga-FAPI PET/CT in different rare malignancies, including epithelial carcinomas. Although intense FAPI expression has been demonstrated in salivary gland malignancies, especially for adenoid cystic carcinoma (8), FAPI expression has not been demonstrated in EMC. We herein present the findings of ¹⁸F-FDG and ⁶⁸Ga-FAPI-04 PET/CT in this rare metastatic EMC case. ¹⁸F-FDG PET/CT seems feasible for determining the spread of disease; however, ⁶⁸Ga-FAPI-04 PET/CT does not seem to have theranostic potential in EMC.

Ethics

Informed Consent: Patient consent was obtained.

Authorship Contributions

Surgical and Medical Practices: C.C., D.H.Ş., D.V.B., S.K., Concept: C.C., D.H.Ş., Design: C.C., D.H.Ş., D.V.B., Data Collection or Processing: C.C., D.H.Ş., D.V.B., S.K., Analysis or Interpretation: C.C., D.H.Ş., Literature Search: C.C., D.V.B., Writing: C.C., D.H.Ş., D.V.B., S.K.

Conflict of Interest: No conflicts of interest were declared by the authors.

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References

- Wakasaki T, Kubota M, Nakashima Y, Tomonobe E, Mihara T, Fukushima J. Invasive myoepithelial carcinoma ex pleomorphic adenoma of the major salivary gland: two case reports. BMC Cancer 2016;16:827.
- Pierard S, Gregoire V, Weynand B, Machiels JP. Epithelial-myoepithelial carcinoma of the submandibular gland with symptomatic lung metastases treated with chemotherapy. Eur Arch Otorhinolaryngol 2006;263:1158-1160.
- 3. Gore MR. Epithelial-myoepithelial carcinoma: a population-based survival analysis. BMC Ear Nose Throat Disord 2018;18:15.
- Kusafuka K, Yamashita M, Muramatsu A, Arai K, Suzuki M. Epithelialmyoepithelial carcinoma ex-pleomorphic adenoma of the parotid gland: report of a rare case with immunohistochemical and genetic analyses. Med Mol Morphol 2021;54:173-180.
- Takumi K, Fukukura Y, Kamiyama T, Nakajo M, Ohori J, Kurono Y, Higashi M. Epithelial-myoepithelial carcinoma of the parotid gland: correlation of dynamic magnetic resonance imaging, (18)F-fluorodeoxyglucose-

positron emission tomography, and pathological findings. Jpn J Radiol 2010;28:618-622.

- Kratochwil C, Flechsig P, Lindner T, Abderrahim L, Altmann A, Mier W, Adeberg S, Rathke H, Röhrich M, Winter H, Plinkert PK, Marme F, Lang M, Kauczor HU, Jäger D, Debus J, Haberkorn U, Giesel FL. 68Ga-FAPI PET/CT: tracer uptake in 28 different kinds of cancer. J Nucl Med 2019;60:801-805.
- Dendl K, Finck R, Giesel FL, Kratochwil C, Lindner T, Mier W, Cardinale J, Kesch C, Röhrich M, Rathke H, Gampp H, Ristau J, Adeberg S, Jäger D,

Debus J, Haberkorn U, Koerber SA. FAP imaging in rare cancer entitiesfirst clinical experience in a broad spectrum of malignancies. Eur J Nucl Med Mol Imaging 2022;49:721-731.

 Röhrich M, Syed M, Liew DP, Giesel FL, Liermann J, Choyke PL, Wefers AK, Ritz T, Szymbara M, Schillings L, Heger U, Rathke H, Kratochwil C, Huber PE, von Deimling A, Debus J, Kauczor HU, Haberkorn U, Adeberg S. 68Ga-FAPI-PET/CT improves diagnostic staging and radiotherapy planning of adenoid cystic carcinomas - Imaging analysis and histological validation. Radiother Oncol 2021;160:192-201.