

Recurrent Follicular Dendritic Cell Sarcoma of the Parotid Gland Imaged with 18F-FDG PET/CT

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Abstract Follicular dendritic cell sarcoma (FDCS) is an extremely rare tumor with only 67 cases of head and neck FDCS reported in the literature. A 65-year-old female had a 6-cm follicular dendritic cell sarcoma resected from the left parotid gland with close margins. It recurred 1 year later as a 5-cm mass that was intensely [18F] fluoro-2-deoxy-D-glucose (18F-FDG) avid on positron emission tomography/computed tomography (PET/CT) and was re-excised. A follow-up PET/CT did not show any metastatic disease. The use of 18F-FDG PET/CT in the management of FDCS warrants further research. We present the 18F-FDG PET/CT imaging findings of this rare tumor.

Keywords Follicular dendritic cell sarcoma · FDCS · Parotid gland · 18F-FDG · Fluorodeoxyglucose · PET/CT

Introduction

Soft tissue sarcomas are uncommon tumors that arise from the embryonic mesoderm and account for approximately 0.7% of all adult malignancies. They have a high mortality rate and account for 3–4% of annual cancer-related deaths [1]. Diagnostic imaging plays a major role in the evaluation of patients with sarcoma. Anatomic imaging, which is used in

the evaluation of sarcomas, includes computed tomography (CT), magnetic resonance imaging (MRI) and bone scintigraphy. During the initial workup, MRI provides a better description and definition of the extent of the tumor than CT or PET/CT [2]. 18F-FDG PET/CT imaging has been investigated in soft tissue sarcomas for biopsy guidance [3], response assessment [4], grading [5], follow-up [6] and prognostication [7]. Several recent studies have found a correlation between the maximum standardized uptake value (SUVmax) of sarcoma tumors and histological characteristics such as histologic grade, mitotic counts, and presence of tumor necrosis [8]. 18F-FDG PET/CT can also be used as a prognostic indicator for progression-free survival and overall survival [9].

FDCS is a very rare type of sarcoma of low to intermediate malignant potential, originating from follicular dendritic cells, which are non-lymphoid and non-phagocytic accessory cells of the lymphoid system [10, 11]. Most cases arise within lymph nodes, but extranodal sites including the mediastinum, gastrointestinal tract, liver and spleen have been described. Only 67 cases of head and neck FDCS have been reported in the literature, with none in the parotid gland [12–16]. The use of 18F-FDG PET/CT imaging in the evaluation of head and neck FDCS has not been previously described.

Case Report

A 65-year-old female had a 6-cm FDCS tumor resected from the left parotid gland. The tumor was arising from the gland, and the margins were negative but close (less than 0.5 mm from both the deep and superficial margins of the resection). A follow-up 18F-FDG PET/CT (dose: 604 MBq) done 18 months later showed a new left parotid gland mass measuring 5.0 × 4.2 cm, which was intensely 18F-FDG avid with a SUVmax of 9.1, consistent with sarcoma recurrence.

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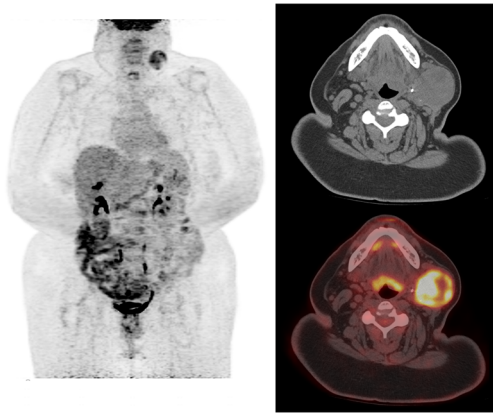


Fig. 1 PET/CT MIP, transaxial CT, and PET/CT fusion images showing an 18F-FDG-avid left parotid mass, consistent with recurrent FDCS

Maximum intensity projection (MIP) and transaxial images showed the 18F-FDG-avid left parotid sarcoma recurrence with no evidence of nodal involvement or metastatic disease (Fig. 1). Excision of the mass showed a well-circumscribed, lobulated 5-cm tumor with multiple hemorrhagic areas consistent with recurrent FDCS. The proliferative index by MIB-1 was 10%. Histological evaluation showed tumor cells with histiocytoid appearance,

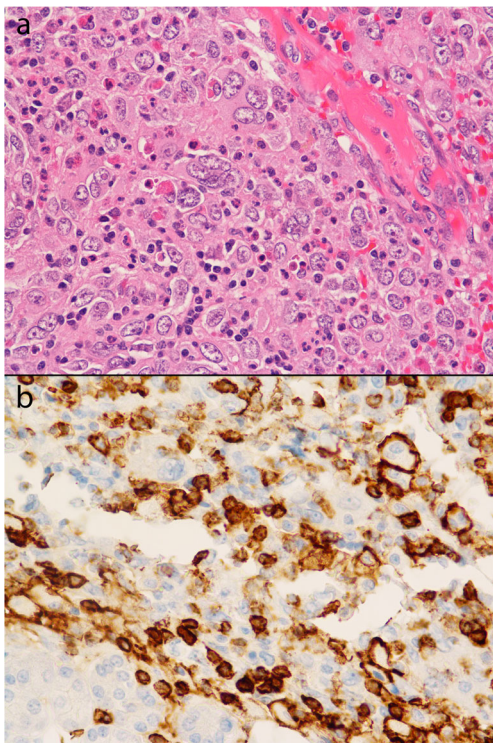


Fig. 2 **a** Histopathological evaluation showed tumor cells with histiocytoid appearance, pleomorphic nuclei, and small-to-intermediate-sized nucleoli. H&E stain, magnification 400 \times . **b** Immunohistochemical staining showing CD15-positive malignant cells, cytoplasmic staining. The immunomorphologic features were consistent with FDCS

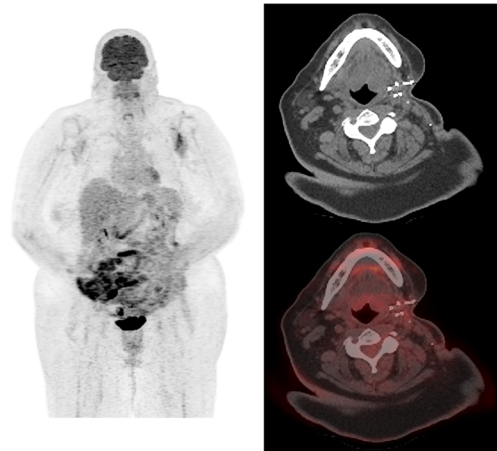


Fig. 3 Follow-up PET/CT performed 9 months later; MIP, transaxial CT and PET/CT fusion images do not show any evidence of recurrent disease in the left parotid surgical bed

pleomorphic nuclei, and small-to-intermediate-sized nucleoli (on H&E stain under 400 \times magnification) (Fig. 2a). Immunohistochemical staining showed CD15-positive malignant cells (Fig. 2b). The immunomorphologic features were consistent with FDCS. A follow-up PET/CT performed 9 months later did not show any evidence of recurrent disease (Fig. 3).

Discussion

FDCS is a very rare neoplasm with 129 cases reported in the literature, including 67 cases in the head and neck [12–16]. It affects males and females equally and at all ages with a mean age at presentation of 46. Approximately two thirds arise in a lymph node and one third arise in extranodal sites, including the oral cavity, GI tract, liver and spleen. Most extranodal sites occur in the head and neck; however, FDCS arising in the parotid gland has not been previously reported in the literature [17–19]. FDCS was traditionally understood to behave more like a low-grade sarcoma than lymphoma [20], with a local-regional recurrence rate of 40% and distant metastases of 25% (lymph nodes, lung, liver); however, recent studies suggest that FDCS tumors have a more aggressive clinical course than previously recognized [11, 16, 21, 22]. Radical surgery is curative in up to two thirds of cases [13], and radiotherapy and chemotherapy do not seem to be effective in improving patient's disease-free survival after radical tumor excision [11, 17]. The use of 18F-FDG PET/CT in the staging of FDCS has only been described in two cases of mediastinal FDCS [23, 24], and its use in the staging or surveillance of head and neck FDCS has not been previously described in the literature and warrants further research.

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Compliance with ethical standards

Conflict of Interest William Makis, EW Hudson and Brian Chiu declare that they have no conflict of interest.

Ethical statement The study was approved by an institutional review board or equivalent and has been performed in accordance with the ethical standards laid down in the 1964 Declaration of Helsinki and its later amendments. All subjects in the study gave written informed consent or the institutional review board waived the need to obtain informed consent.

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