Case Report

Fine-Needle Aspiration Cytology of Follicular Dendritic Cell Sarcoma of Cervical Lymph Node: A Challenging Diagnosis

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Abstract

Follicular dendritic cell sarcoma (FDCS) is a rare malignant tumor that could arise in both nodal and extranodal sites, with only nine previously reported cases demonstrating cytologic features. In this report, we describe a case of FDCS in a 60-year-old female who presented with neck mass. Fine-needle aspiration cytology and subsequent core biopsy were suggestive of metastatic carcinoma. The cytologic features were epithelioid-to-spindle cell morphology, vesicular nuclei, prominent nucleoli, intranuclear inclusions, and occasional binucleated and multinucleated forms. However, absence of cytokeratin expression was against the diagnosis of metastatic carcinoma. The definitive diagnosis was reached by the demonstration of CD21 and CD23 expression. The pathologist should be aware of this rare malignant tumor, especially its cytologic features in aspirated material. The differential diagnosis in the above case was metastatic carcinoma, melanoma, and malignant granular cell tumor. The demonstration of expression of one or more dendritic cell marker is the clue for the diagnosis, which could be applied on cytological preparations with sufficient material.

Keywords: CD21, CD23, follicular dendritic cell sarcoma, fine-needle aspiration cytology, neck swelling

Introduction

Follicular dendritic cell sarcoma (FDCS) is a rare malignant tumor that could arise in both nodal and extranodal sites. It is a nonlymphoid primary nodal malignant tumor that has an indolent course.[1] Some studies demonstrated the liability for recurrence and metastasis of FDCS and therefore, it has been viewed as an intermediate-grade malignancy.[2] There are limited number of studies that investigated the cytologic features of FDCS in fine-needle aspiration cytology[3-10] [Table 1]. In this report, we describe a case of FDCS in a female aged 60 years who presented with neck mass, demonstrating the common pitfalls and differential diagnosis.

Case Report

A 60-year-old female patient presented with right lateral neck swelling before 5 years. Computed tomography and magnetic resonance imaging suggested a neoplasm with neurogenic features and recommended biopsy. Fine-needle aspiration cytology revealed cohesive clusters of malignant epithelioid cells that showed abundant eosinophilic cytoplasm and pleomorphic vesicular nuclei with prominent nucleoli and intranuclear inclusions [Figure 1]. Occasional binucleation and multinucleation forms were seen [Figure 1]. Positive for malignant epithelial cells, suggestive of metastatic carcinoma was the diagnosis of cytology specimen. The patient was further submitted to ultrasound-guided trucut biopsy that revealed rim of lymphocytes admixed with sheets of malignant ovoid-to-spindle-shaped cells that showed syncytial pattern with indistinct cell borders together with whorly pattern of arrangement, similar to that seen in meningioma [Figure 2]. The diagnosis was made as a metastatic carcinoma for clinical and radiological correlation. However, clinically and after extensive investigation, no primary origin was detected. The core was submitted to cytokeratin (CK) and S100 staining. The tumor cells were negative for CK and positive for S100 [Figure 2d]. At this time, the patient was submitted to complete excision of the mass and excision of two deep cervical lymph

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Table 1: Summary of published articles concerning fine-needle aspiration cytology of follicular dendritic cell sarcoma

Articles	Age/year	Clinical picture	Site of presentation	Recurrence
Gaffney *et al*., 2000[3]	33/female	Abdominal mass with metastasis to lung, liver, and lymph node	Extranodal	Negative
Vicandi *et al*., 2000[4]	76/male	Lateral cervical mass	Nodal	Positive
Loo *et al*., 2001[5]	80/male	Intra-abdominal mass	Extranodal	Negative
Ren *et al*., 2005[6]	65/female	Liver mass with a history of splenectomy for FDCS	Extranodal	Negative
Fan *et al*., 2007[7]	Female Male	Tonsillar Nasopharynx	Extranodal	Positive
Granados *et al*., 2008[8]	57/female	Left hepatic lobe mass positive for EBV	Extranodal	Negative
Tokyol *et al*., 2008[9]	41/female	Neck swelling	Nodal	Positive
Kure *et al*., 2010[10]	47/male	Right neck mass, hypertension, and HIV	Nodal	Negative
Wang *et al*., 2010[1]	26/female	Neck mass	Nodal	Negative
The present case	60/female	Neck mass	Nodal	Negative

FDCS: Follicular dendritic cell sarcoma, EBV: Epstein-Barr virus

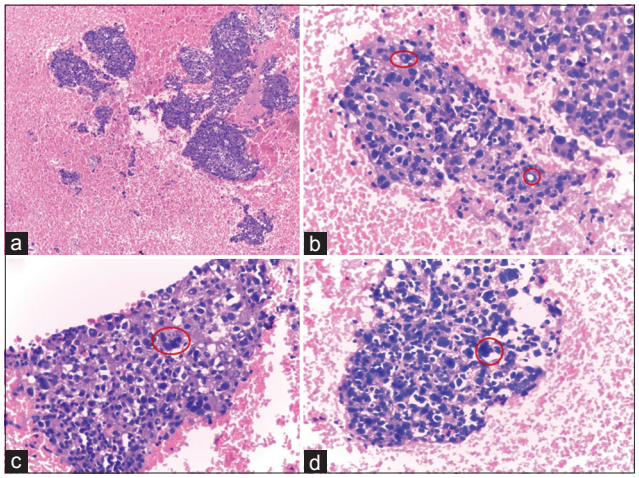

Figure 1: Cytology of aspirated cervical lymph node formed of cohesive sheets of epithelioid cells (a) that showed eosinophilic cytoplasm and occasional intranuclear inclusion (b, red circle) and binucleated or multinucleated forms (c and d, red circles) (H and E, ×100 for a and ×200 for b-d)

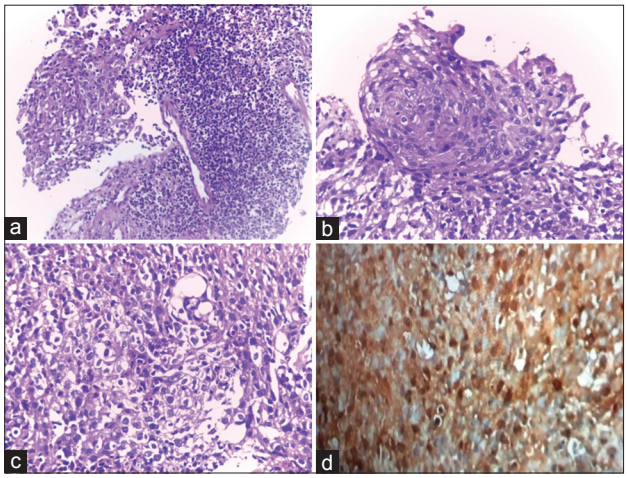

Figure 2: Core biopsy showing peripheral lymphocytic infiltrate adjacent to the malignant sheets of cells (a) that were arranged in syncytial sheets with whorly pattern (b) and formed of epithelioid cells with occasional spindling (c) (H and E ×100 for a and ×200 for b and c). Nuclear and cytoplasmic immunoreactivity for S100 (immunohistochemical, ×400)

nodes. Again, the histological picture of the excised mass was similar to that seen in core biopsy with confirmation of lymph node architecture replaced by this type of malignancy. The other excised deep cervical lymph nodes showed picture of Castleman's disease. The latter attracts the attention for the possibility of primary lymph node malignancy which arises on the top of Castleman's disease; therefore, CD21 [Figure 3a] and CD23 [Figure 3b] were investigated which showed positive expression with a final diagnosis of FDCS. A written consent was taken from the patient to publish her case.

Discussion

FDCS is commonly presented as neck mass similar to that reported in the present case. The cytologic features seen in the present case simulate that described in the previous studies[3-10] including syncytial and discohesive large epithelioid-to-spindled malignant cells with intranuclear inclusions and presence of binucleated and multinucleated forms. The cytology of FDCS represents a true challenge to reach a proper diagnosis, and it could be easily misdiagnosed as metastatic carcinoma as in the present study and in others.[1,5] We agreed with others that the diagnosis of FDCS is made after the demonstration of positivity for dendritic cell markers such as CD21, CD23, and CD35. We demonstrated positive expression for CD21 and CD23, which could be focal and not necessarily diffuse, agreeing with Yin *et al*., 2010.[11] We agreed with the latter study[11] in demonstrating S100 expression. The main differential diagnosis is from metastatic carcinoma, melanoma, and malignant granular cell tumor which are excluded by negativity for CK and HMB45, and although the present case showed S100 expression, it also showed positivity for dendritic cell markers. The latter positivity differentiates it from malignant granular cell tumor. FDCS could arise on the top of Castleman's disease, similar to that seen in the present case.[12] The etiology is unknown in most cases; however, some authors have found an association with Epstein–Barr virus by its detection by *in situ* hybridization.[8]

Declaration of patient consent

The authors certify that they have obtained all appropriate

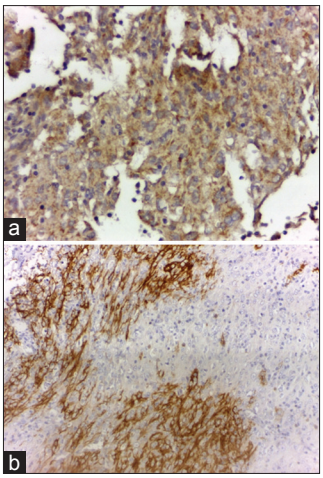

Figure 3: The malignant cells showing focal faint expression for CD21 (a) and strong expression for CD23 (b) (immunohistochemical, ×400 for a and ×200 for b)

patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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