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Follicular dendritic cell sarcoma in the submandibular region: A report of a rare case

KEYWORDS
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Follicular dendritic cell sarcoma (FDCS) is a rare malignant tumor exhibiting morphologic and phenotypic features of follicular dendritic cells (FDCs), which are mesenchymal-derived dendritic cells located in B-cell follicles.[1,2] The majority of FDCS cases in the head and neck are extranodal, but it can occur less frequently in cervical lymph nodes.[1] Here, we reported a rare case of nodal FDCS arising in the submandibular region.

A 27-year-old male presented with a 3-month history of swelling in the left neck. Computed tomography showed a 3.7-cm homogeneously enhancing mass in the left submandibular region (Fig. 1A). Magnetic resonance imaging revealed a T1-isointense and T2-hyperintense lesion (Fig. 1B and C). Surgical excision was performed. Gross examination showed a well-circumscribed mass with a fleshy cut surface. Histopathological examination revealed a sheet of oval or epithelioid neoplastic cells (Fig. 1D), effacing the normal nodal architecture. Tumor cells had eosinophilic cytoplasm with indistinct cell borders, resulting in a syncytial appearance (Fig. 1E). The nuclei of the tumor cells had vesicular chromatin and prominent nucleoli (Fig. 1E). A diffuse infiltrate of non-neoplastic lymphocytes was seen with focal perivascular cuffing (Fig. 1D and E). No necrosis was observed. Immunohistochemically, neoplastic cells were positive for vimentin and a panel of FDC markers, including CD21, CD23, and CD35 (Fig. 1F and G), but negative for pan-cytokeratin, S-100 protein, CD3, CD20, CD45, and CD68. The Ki-67 proliferation index was approximately 5%. *BRAF* mutation analysis was performed by Sanger sequencing as described previously,[3] and no *BRAF* V600E mutation was detected. The final diagnosis was FDCS. The patient received 45 Gy of postoperative radiotherapy. At the 26-month follow-up, no evidence of recurrence or metastasis was observed.

To date, there have been only a few reports of FDCS arising in the submandibular lymph nodes.[4] Since FDCS can histologically mimic other sarcomas, histiocytic tumors, and lymphomas, it is essential to confirm the expression of FDC markers, such as CD21, CD23, CD35, clusterin, and CXCL13, for its diagnosis.[1] In a similar context, the present case showed diffuse positivity for all FDC markers tested (CD21, CD23, and CD35), leading to a definitive diagnosis of FDCS. *BRAF* V600E mutations are found in a subset of FDCS cases and may serve as a diagnostic clue,[1] although not detected in the present case. Surgery is the mainstay of treatment for early-stage localized disease.[1,5] A previous

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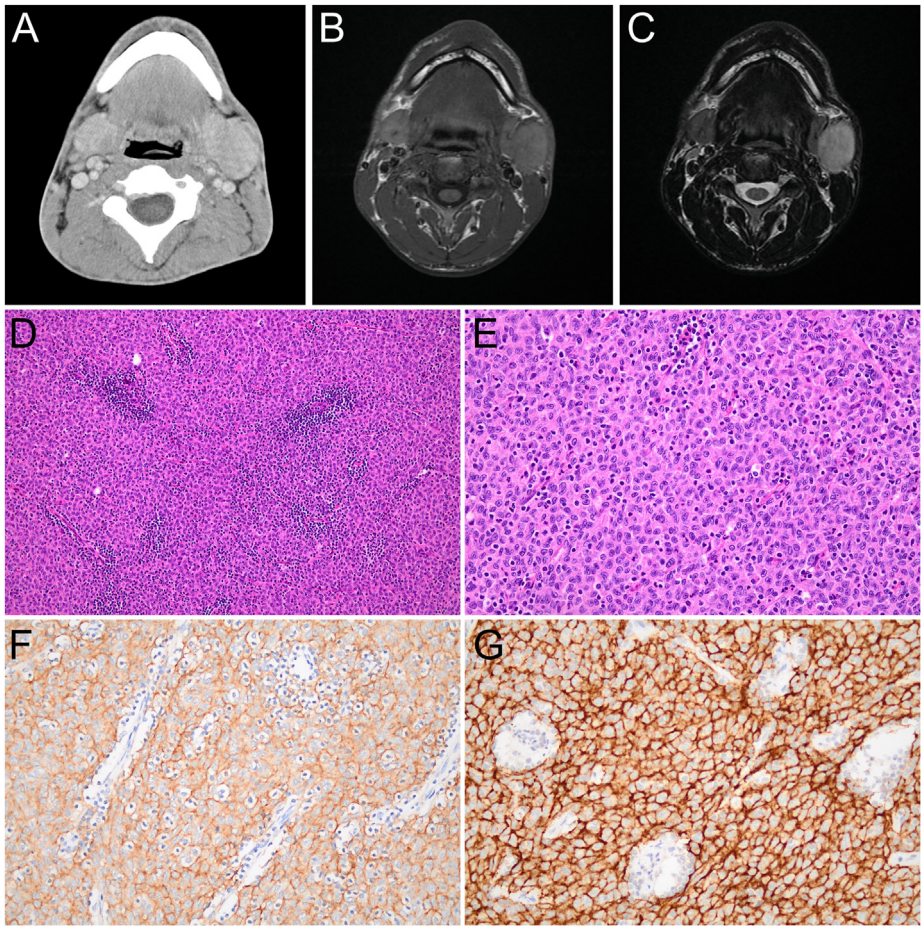

Figure 1 Radiological (A—C), histological (D and E), and immunohistochemical (F and G) findings of follicular dendritic cell sarcoma in the submandibular region. (A) A contrast-enhanced computed tomography image shows a homogeneously enhancing mass adjacent to the left submandibular gland. The mass is isointense in T1-weighted magnetic resonance imaging (B) and hyperintense in T2-weighted magnetic resonance imaging (C). (D) The mass histologically consists of a sheet of neoplastic cells with a lymphocytic infiltrate showing focal perivascular cuffing. (E) Tumor cells have eosinophilic cytoplasm, indistinct cell borders, and vesicular nuclei with small distinct nucleoli. Tumor cells are diffusely positive for CD23 (F) and CD35 (G).

study demonstrated that adjuvant radiotherapy may reduce the locoregional recurrence rate in patients with head and neck FDCS.[5] Although the present case can be classified as low-risk based on tumor size (<50 mm),[1] postoperative radiotherapy was performed, resulting in a good clinical outcome.

Declaration of competing interest

The authors have no conflicts of interest relevant to this article.

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None.

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