

Interdigitating dendritic cell sarcoma of the supraglottic larynx

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ABSTRACT

Background: Interdigitating dendritic cell sarcoma (IDCS) is an extremely rare neoplasm with sparse published literature, limited to case reports. This neoplasm arises from dendritic cells mainly within lymph nodes, however extranodal localization has been noted in the head and neck.

Methods: Herein we present the first report of IDCS of the supraglottic larynx. A 35-year-old woman presented with odynophagia and dysphagia and was found to have an epiglottic primary lesion with bilateral regional lymphadenopathy. She was treated with transoral laser supraglottic laryngectomy, bilateral neck dissections, and postoperative radiation therapy.

Results: Patient has no evidence of disease with 4 years of follow-up.

Conclusion: Surgical resection with postoperative radiation therapy is a reasonable approach for management of loco-regionally advanced extranodal IDCS of the head and neck.

Introduction

Interdigitating dendritic cells (IDCs) are nonlymphoid, non-phagocytic antigen-presenting cells involved in regulation of cellular immune response and initiation of primary T-lymphocyte immune responses [1–4]. IDCs are commonly located in T-cell zones of lymphoid tissues including paracortex and deep cortex of lymph nodes, inter-follicular areas of mucosa-associated lymphoid tissue (MALT) and splenic periarteriolar lymphoid sheaths but can also be found in extranodal sites [1]. IDCs originate from hematopoietic precursors predominantly through conversion of Langerhans cells but can also develop through myeloid or lymphoid pathways [5]. Neoplasms of dendritic cell origin are rare with only about 100 cases of interdigitating dendritic cell sarcoma (IDCS) reported in the literature [6,7]. Although surgery is the primary treatment modality for localized disease, IDCS can recur locally and distally [7–9]. Limited information is available regarding use of postoperative radiation therapy [6–8]. We present the case of a patient treated for IDCS of the supraglottic larynx.

Case report

A 35-year-old female presented with 7-month history of dysphagia and odynophagia. Computed tomography (CT) scan of the neck showed enhancing multifocal lesions of the right epiglottis and a level 3 heterogeneous lymph node abutting and partially encircling the right common carotid artery. Following non-diagnostic biopsy of the epiglottis, patient underwent excisional biopsy of the enlarged level 3 lymph node and was found to have malignant spindle cell neoplasm consistent with IDCS. She was subsequently referred to our institution.

On physical exam patient had normal true vocal cord mobility bilaterally and no focal abnormalities. Flexible fiberoptic laryngoscopy showed a frond-like lesion emanating from the laryngeal surface of the epiglottis without involvement of the vocal folds or glottis.

Subsequent CT of the neck showed an enhancing soft tissue lesion extending from the right aspect of the epiglottis and aryepiglottic fold measuring 1 × 2.1 × 3.3cm, a right plaque-like infraglottic lesion and bilateral neck subcentimeter level 2, 3, 4 lymph nodes which were fluorodeoxyglucose (FDG) avid on corresponding positron emission tomography (Fig. 1). There was no evidence of distant metastatic disease.

Abbreviations: Dendritic cells, (DCs); Interdigitating dendritic cells, (IDCs); Interdigitating dendritic cell sarcoma, (IDCS); Tumor-infiltrating dendritic cells, (TIDCs); Mucosa-associated lymphoid tissue, (MALT); fluorodeoxyglucose, (FDG); Computed tomography, (CT); intensity-modulated radiation therapy, (IMRT); planning target volumes, (PTV)

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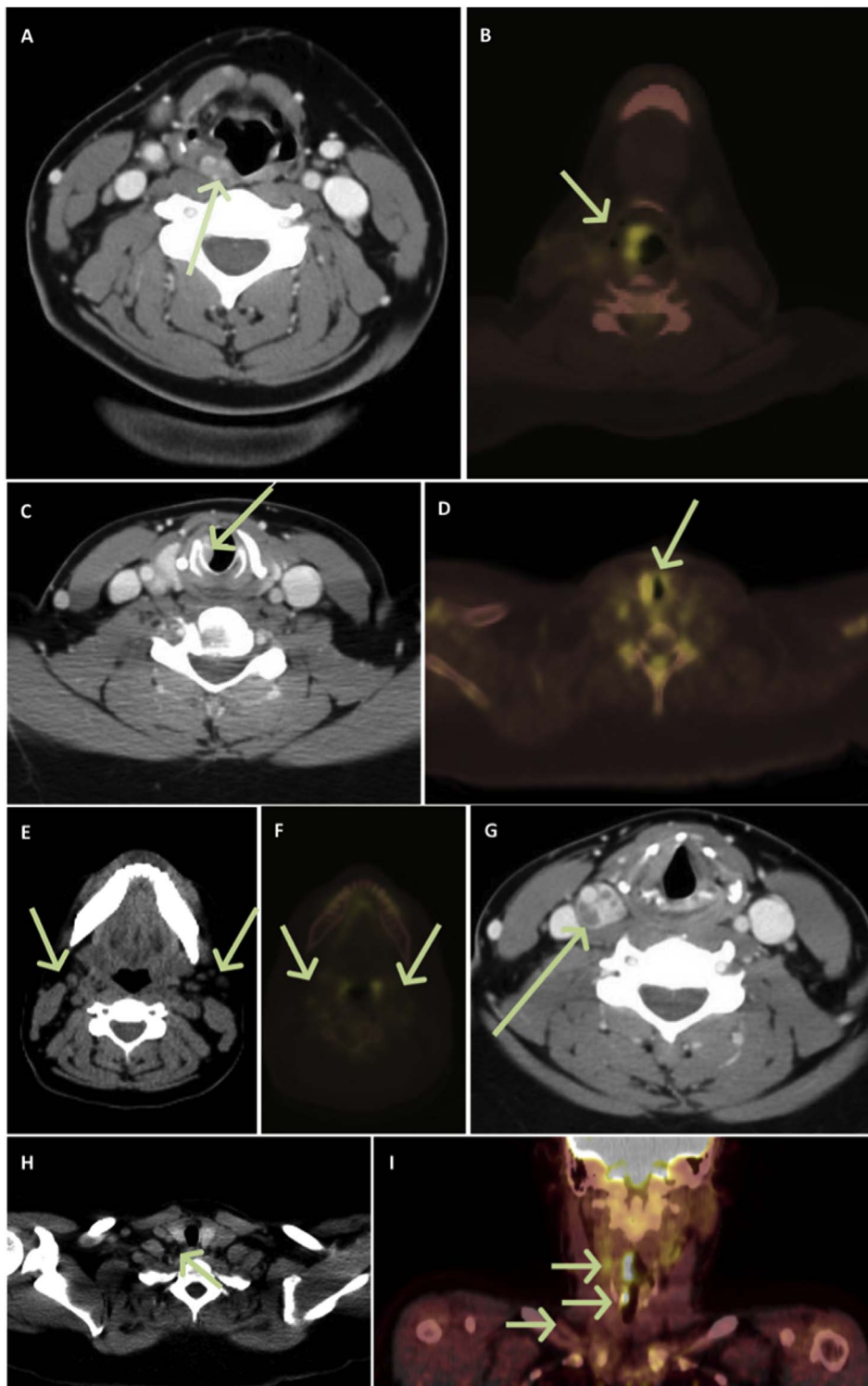


Fig. 1. Diagnostic imaging. A: Axial neck CT with contrast shows asymmetric, heterogeneous rim-enhancing soft tissue mass measuring 1.0 cm × 2.1 cm × 3.3 cm originating from the right aspect of the epiglottis and aryepiglottic folds and protruding into the supraglottic larynx. B: Epiglottic mass demonstrates intense FDG avidity on axial PET overlaid on CT. C: Axial neck CT with contrast shows a mass arising from the infraglottic region of the right laryngeal wall. D: Infraglottic mass demonstrates intense FDG avidity on axial PET overlaid on CT. E: Bilateral subcentimeter enlarged cervical lymph nodes. F: Bilateral node FDG avidity in PET overlaid on CT is concerning for metastatic disease. G: Axial neck CT with contrast shows a heterogeneous mass, likely a level 3 necrotic lymph node, abutting and partially encircling the right common carotid artery. H: Subcentimeter level IV nodes. I: Sagittal image from PET/CT demonstrating asymmetric FDG uptake in right larynx, right vocal cords and bilateral hypermetabolic cervical lymph nodes at levels II, III, IV.

The multidisciplinary treatment team recommended surgery and postoperative radiation therapy. Four months following initial presentation patient underwent transoral laser supraglottic laryngectomy with bilateral neck dissections. Pathology demonstrated bilateral metastasis to the following lymph nodes: right levels 2 (1 of 5 nodes positive, 7 mm metastasis), 4 (1 of 6 nodes positive, 5 mm metastasis) and left level 2 (1 of 6 nodes positive, 3 mm metastasis). No extracapsular extension of lymph node metastasis was noted. The primary lesion was removed in standard multi-block transoral laser microsurgery fashion with negative surgical margins.

Surgical specimen morphology was similar to that of the previously biopsied mass. Review of immunohistochemical staining of the enlarged level 3 lymph node sample revealed a neoplasm consisting of a proliferation of spindle to ovoid cells with eosinophilic cytoplasm and pleomorphic nuclei. Infiltrates composed of small lymphocytes and eosinophils were present. Background vascular proliferation with hemorrhage was noted. Reticulin stain demonstrated the lobular nature of the mass. Neoplastic cells were diffusely positive for CD31, CD99, vimentin, S-100, fascin and focally positive for EMA and pancytokeratin (AE1/3). Neoplastic cells were negative for MART-1, ACTIN, desmin,

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