Case Report

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A Malignant Cystic Midline Neck Mass

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Keywords

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Thyroglossal duct cysts (TDCs) are the most common congenital neck cysts.[1] They often contain ectopic thyroid tissue (71%), with up to 3% of TDC excisions demonstrating papillary thyroid carcinoma.[2] Mucoepidermoid carcinoma (MEC) typically arises from salivary glands and has rarely been associated with thyroid tissue or TDC.[3] This rare case of concurrent MEC and TDC highlights the differential diagnosis for a midline neck mass. Informed consent was obtained, as was ethics approval from the University of Manitoba Research Ethics Board.

Report of a Case

A healthy 51-year-old woman presented with an 8-month history of enlarging midline neck mass, with persistent pain, dysphagia, and dysphonia. Examination revealed a 3-cm compressible midline neck mass at the level of the thyroid cartilage, which was firmer to the right of midline. It elevated on tongue protrusion. Fiberoptic nasolaryngoscopy demonstrated submucosal fullness in the base of tongue and right vallecula, with posterior displacement of the right supraglottic larynx. Vocal cords were mobile.

Ultrasound identified a solid 2.7-cm right neck mass, separate from a thyroid with small colloid nodules. Computed axial tomography described an irregular 2.9-cm lesion that was more solid laterally and cystic in the midline, centered within the right strap muscles (**Figure 1**). It straddled the hyoid bone without obvious bone destruction, with suspicion for a midline cystic tract through the thyroid cartilage. There was a mass effect on the supraglottic airway, distortion of the larynx, and no cervical lymphadenopathy. Fine-needle aspiration cytology revealed highly atypical cells in a background of cystic content. A malignancy within a TDC was suspected. A Sistrunk procedure with a submucosal base of tongue resection was performed. Pathology revealed high-grade MEC with associated TDC (**Figure 2**). The tumor had a high mitotic index (23 per 10 high-power fields), with tumor necrosis and lymphovascular invasion but no perineural invasion. It invaded the hyoid bone and extended focally to inked margins. Adjuvant treatment was discussed at multidisciplinary rounds. Due to positive margins, intensity-modulated radiotherapy was delivered with 70 Gy to the tumor bed and 63 Gy to the vicinity of tumor in 35 fractions. Chemotherapy was considered but not pursued. Further surgical resection was reserved for salvage. She remains free of recurrence at 18 months.

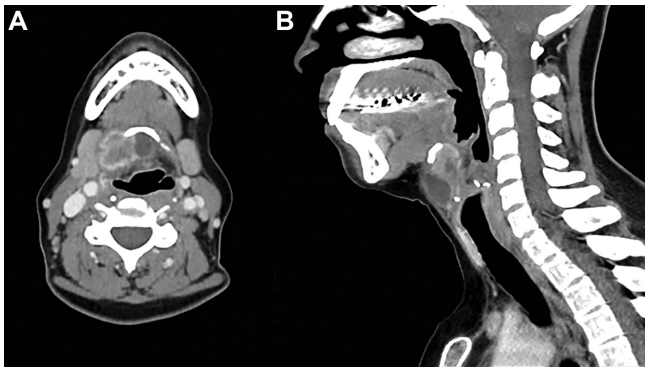

Figure 1. Head and neck computerized axial tomography scan images, illustrating the solid and cystic components of the mass: (A) axial view at the level of the hyoid bone and (B) sagittal view.

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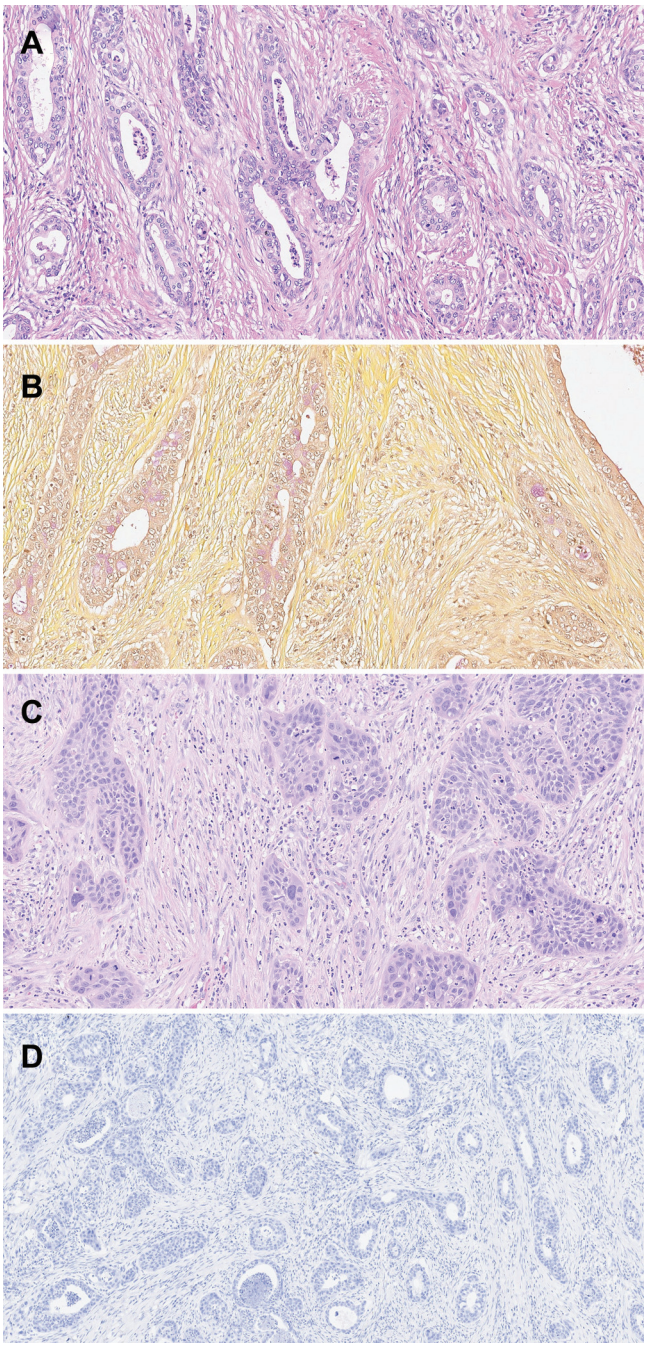

Figure 2. Histopathologic sections: (A) gland formation, (B) positive mucin stain, (C) pleomorphic nuclei and frequent mitotic figures in hematoxylin-eosin stain, and (D) negative thyroid transcription factor 1 immunohistochemical stain.

Discussion

Classical presentation of a TDC is a midline neck mass adjacent to the hyoid bone. The differential diagnosis includes ectopic thyroid, thyroid neoplasm, dermoid cyst, sebaceous cyst, lipoma, and submental lymphadenitis. Although ectopic thyroid tissue has been reported within the cyst, rarely with malignant transformation (3%), only 1 previous case of MEC occurring within a thyroglossal duct remnant has been reported.[3] In the present case, however, a TDC and a minor salivary gland tumor occurred concurrently in the central neck.

MEC is the second-most common malignancy of minor salivary glands, more frequent in females and in the fifth decade of life.[4] Recommended treatment for MECs involves surgery, with local resection for less aggressive tumors and wide resection including involved adjacent structures with possible selective neck dissection for high-grade tumors.[4] Radiotherapy is reserved for high-grade tumors and tumors with aggressive features such as perineural invasion, lymphovascular invasion, and metastatic nodes. The RTOG 1008 trial is currently investigating radiotherapy with cisplatin chemotherapy.[5]

Based on histologic features and imaging, this tumor probably arose from the minor salivary glands of the base of the tongue and less likely from minor mucoserous glands that can be seen in a TDC wall, since the tumor was negative for thyroid transcription factor 1 by immunohistochemistry. This tumor was staged as a T4aN0M0 stage IVa MEC of the minor salivary glands per the seventh edition of the American Joint Committee on Cancer's TNM staging system.

In this unusual case, MEC arose from minor salivary glands at the base of tongue, presenting as a midline neck mass in association with a concurrent TDC. We had suspected a TDC carcinoma based on the atypical cells identified by fine-needle aspiration cytology. If MEC had been suspected, panendoscopy and wider excision of the mass with frozen section would have been performed to achieve negative margins. Positive surgical margins necessitated adjuvant radiotherapy. Minor salivary gland tumors should be considered in the setting of a malignant midline neck mass, as this can affect disease management.

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Author Contributions

Heather Sigvaldason, contributed to the conception of the work and acquired data for the work, drafted the work, provided approval of the version to be published, and agrees to be accountable for the work; **Sheena Graham,** contributed to the conception of the work and acquired data for the work, drafted the work, provided approval of the version to be published, and agrees to be accountable for the work; **Raena Buksak,** contributed to the acquisition, analysis, and interpretation of data for the work, revised the paper critically, provided approval of the version to be published, and agrees to be accountable for the work; **K. Alok Pathak,** contributed to the conception of the work, the acquisition, analysis, and interpretation of data, critically revised the paper, provided approval of the version to be published, and agrees to be accountable for the work.

Disclosures

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