

CASE REPORT

Radiation Medicine: Vol. 23 No. 6, 451–455 p.p., 2005

CT and MRI Findings of Carcinoma Showing Thymus-like Differentiation

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Carcinoma showing thymus-like differentiation (CASTLE) is a rare tumor that occurs in the soft tissue of the neck or in the thyroid gland. We report a case of CASTLE in a 54-year-old man presenting with hoarseness. Computed tomography (CT) and magnetic resonance imaging (MRI) showed a mass enclosing the right carotid artery with the shape of lobulation and invasion to the trachea. The mass was hypointense on T1-weighted imaging and, on T2-weighted imaging, showed a hyperintense periphery with isointense center relative to muscle. In dynamic MRI, the mass revealed gradual but slight heterogeneous enhancement from the periphery. Diagnosis was confirmed by histopathology.

Key words: thyroid, thymus, carcinoma showing thymus-like differentiation (CASTLE), magnetic resonance imaging (MRI), computed tomography (CT)

INTRODUCTION

CARCINOMA SHOWING THYMUS-LIKE DIFFERENTIATION (CASTLE) was first named by Chan and Rosai in 1991.¹ Since then, only a few cases have been reported. It is presumed to arise from ectopic thymic tissue entrapped within the thyroid gland or remnants of branchial pouches.²⁻⁴ Only one case of the computed tomography (CT) and magnetic resonance imaging (MRI) findings has been reported.⁴

CASE REPORT

A 54-year-old man presented with gradually worsening hoarseness, sputum, and dyspnea, but with no pain. He had a 5-year history of hoarseness. Physical examination revealed a thyroid mass on the right side.

Noncontrast CT demonstrated that a soft tissue density mass was located inside the right lobe of the thyroid

gland and the right side of the trachea. At the upper end of the mass, the boundary with the thyroid gland was well defined. The mass displaced and compressed the trachea, narrowed its air space lumen, and invaded the tracheal cartilage (Figs. 1A, B). On MRI study, the mass was hypointense on T1-weighted imaging and showed a hyperintense periphery relative to muscle, with an isointense center on T2-weighted imaging. The right common carotid artery was enclosed by the mass. It was in contact with the right internal jugular vein (Figs. 2A, B). In dynamic MRI, the mass revealed gradual but slight heterogeneous enhancement from the periphery (Figs. 3A, B). Coronal contrast-enhanced T1-weighted images showed a locally elevated tracheal wall; moreover, the endotracheal surface was smooth (Fig. 4). Our diagnosis had been thyroid cancer with tracheal invasion. At surgery, total thyroidectomy, resection of the segmental trachea, right common carotid artery, and internal jugular vein, and reconstruction of the common carotid artery were performed.

Grossly, a grayish-white tumor, measuring 3.5×3.5×3.2 cm, was located between the right lobe of the thyroid gland and the trachea, with invasion to both organs. The tracheal cartilage was displaced and compressed. Microscopic examination revealed the tumor consisted of irregularly shaped islands of polygonal epithelioid cells separated by abundant collagenous or hyalinized stroma. The tumor invaded to the adjacent soft tissue, tracheal submucosa, and thyroid gland in pushing fronts or infiltrative growth and

Received December 9, 2004; revision accepted March 23, 2005.

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We presented this paper at the oral session of the 2003 JRS meeting.

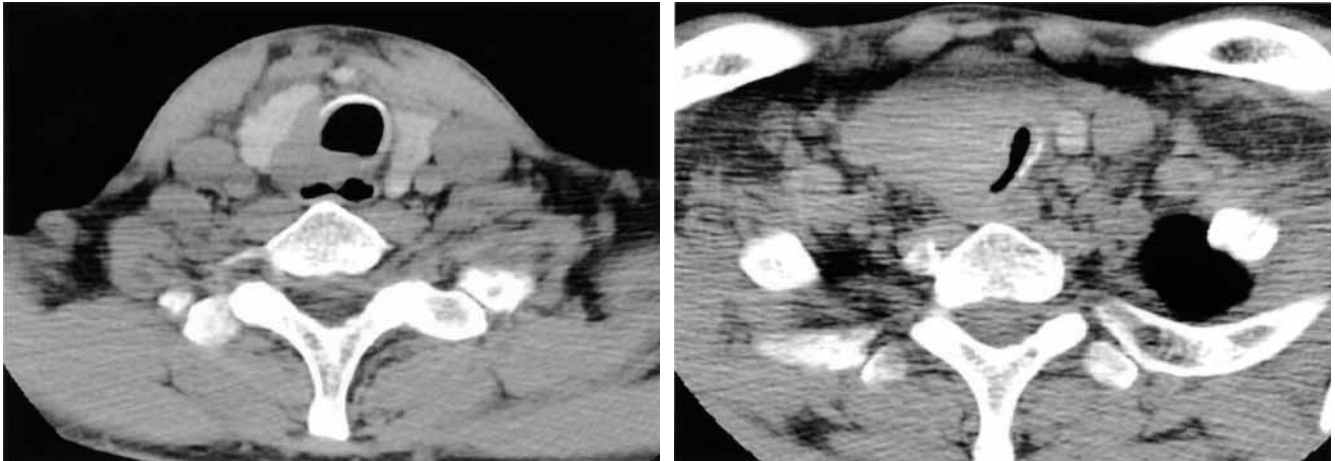


Fig. 1.
A: Noncontrast enhanced CT shows an isodense mass inside the thyroid gland.
B: The mass compresses the trachea and narrows the tracheal lumen.

A | B

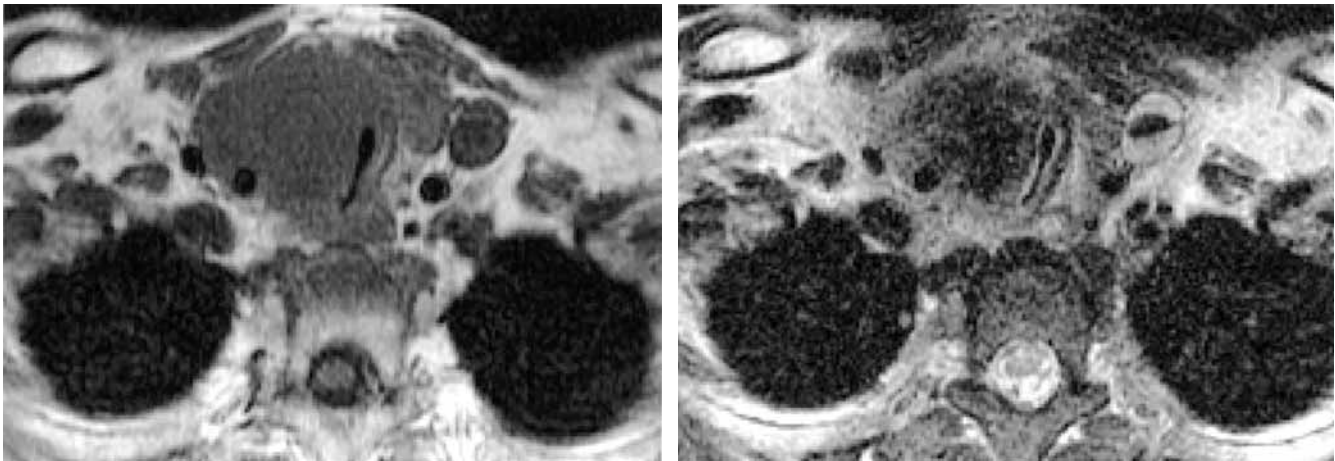


Fig. 2.
A: Axial T1-weighted MR image shows an isointense mass.
B: T2-weighted image shows an isointense center and hyperintense periphery. The mass surrounds the right common carotid artery.

A | B

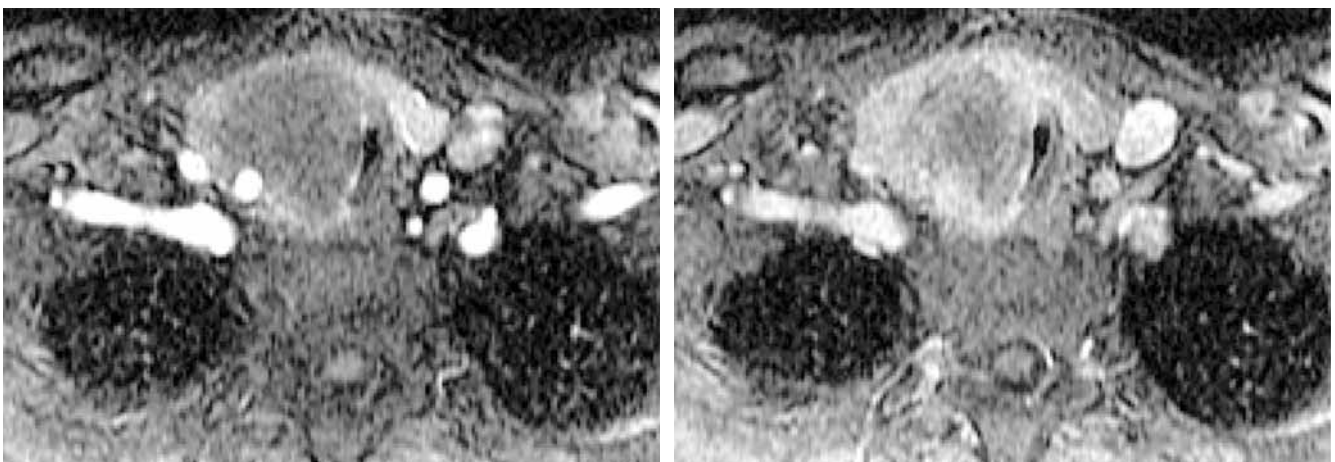


Fig. 3. On dynamic MRI with fat suppression, the mass shows mild contrast enhancement from the periphery with time.
A: Early phase.
B: Late phase.

A | B

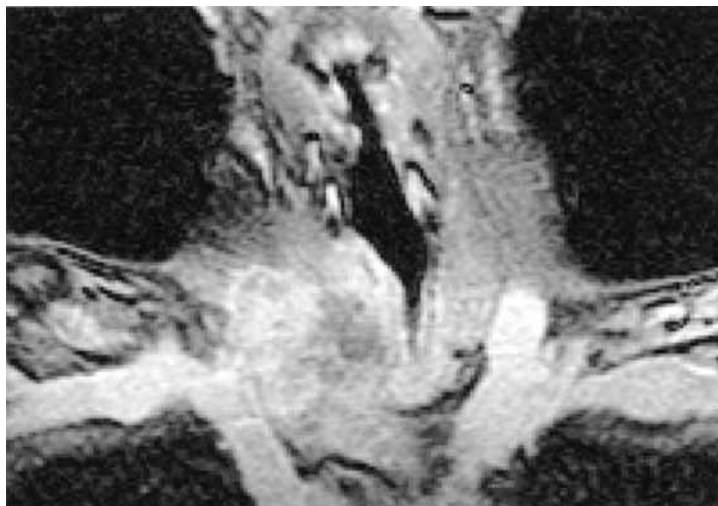


Fig. 4. Coronal contrast-enhanced MRI shows that the tracheal lumen is narrowed due to a locally elevated tracheal wall. The endotracheal surface is smooth.

did not invade to the carotid artery. The tumor cells had a high N/C ratio with vesicular nuclei and prominent nucleoli. Mitoses were few, and necrosis was very limited. Most of the tumor cells had a squamoid appearance with little keratinization and focal glandular differentiation (Figs. 5A-C). Focal cytoplasmic mucin was seen with mucicarmine stain. Immunohistochemically, the tumor cells were positive for cytokeratin, EMA, CEA, and CD5 (Fig. 5D), but negative for thyroglobulin, calcitonin, chromogranin A, and synaptophysin. Based on these morphological findings, the location of the tumor, and the results of immunohistochemistry, the diagnosis of CASTLE was made. After the operation, the symptom disappeared and no recurrence has been noted in a two-year period.

DISCUSSION

A number of rare tumors that occur in the soft tissues of the neck or in the thyroid show complete to partial histologic resemblance to the fetal, mature, or involuted thymus and thymomas.⁵⁻⁷ In 1991, Chan and Rosai¹ divided these tumors into four groups on the basis of their morphologic features: ectopic hamartomatous thymoma, ectopic cervical thymoma, spindle epithelial tumor with thymus-like differentiation (SETTLE), and CASTLE. The CASTLE type of tumor is histologically similar to thymic carcinoma of the lymphoepithelioma or squamous cell variety. It is lobulated and expansive, and has fibrous septa. Generally, CASTLE occurs in adults. According to current reports,^{1,2,4} it is equally frequent in men and women. Lesions are on the left side in approximately two thirds of cases. It recurs in regional

nodes after long intervals in about one half of cases. Rarely, its clinical course is aggressive.¹

To our knowledge, there is only one report of imaging on MRI. It revealed an isointense mass on the T1-weighted image, was hyperintense on the T2-weighted image, and showed slight enhancement after the injection of contrast material.⁴

The thymus gland arises from the third and fourth pharyngeal pouches. A third pharyngeal pouch tract would pass from the pyriform sinus through the thyrohyoid membrane, between the carotid artery and vagus nerve, posterior and inferior to the glossopharyngeal nerve, then downward lateral to the thyroid gland. A fourth pharyngeal pouch tract would extend from the apex of the pyriform sinus inferiorly to the superior laryngeal nerve, externally to the recurrent laryngeal nerve, and pass inferiorly along the trachea to eventually swing forward between the arch of the aorta and the subclavian artery. A spectrum of abnormalities of the thymus is attributed to the embryonic cervical descent of the thymic primordium. The abnormalities may present as a solid mass or cystic changes in the associated epithelium derived from pharyngeal pouch endoderm.⁸⁻¹¹ CASTLE is postulated to arise either from the ectopic thymic tissue or remnants of branchial pouches, which retain the potential to differentiate along the thymic line.¹

CD5 is useful in the differentiation of thymic tumors. Some authors^{2,12-14} have reported that CASTLE and thymic carcinoma are CD5-positive, whereas the majority of thymomas or other malignancies are CD5-negative.

In our case, the preoperative diagnosis was considered

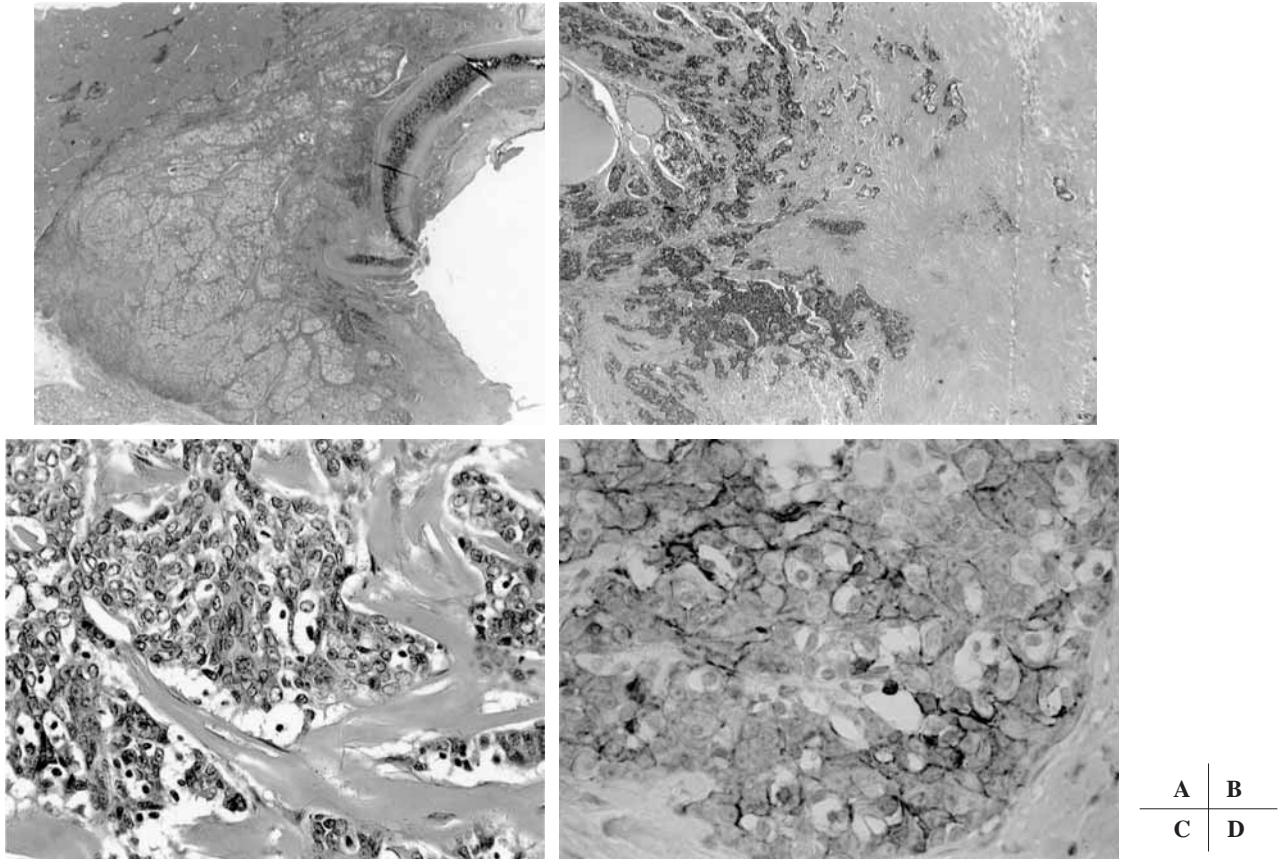


Fig. 5.
A: Photomicrograph of the resected specimen shows the tumor located between the thyroid gland and trachea. The tumor invades the thyroid gland slightly (hematoxylin-eosin stain, original magnification $\times 4$).
B: The tumor is rich in stromal tissue (hematoxylin-eosin stain, original magnification $\times 10$).
C: The neoplastic cells have a high N/C ratio and a squamoid appearance (hematoxylin-eosin stain, original magnification $\times 40$).
D: Immunohistochemical staining is positive for CD5 (original magnification $\times 40$).

to be thyroid cancer with tracheal invasion. However, in retrospect, the facts that the tracheal wall was locally elevated, the cartilage was markedly compressed, the thyroid gland was displaced to the right, and there was a well-defined border between it and the tumor, were not consistent with thyroid origin. The differential diagnosis includes malignant lymphadenopathy and neurogenic tumor. If the tumor had consisted of malignant nodes or been a neurogenic tumor, it would not have been lobulate in shape, had an isointense center on T2-weighted imaging relative to muscle, or have invaded the tracheal cartilage with enclosure of the common carotid artery. An isointense center on T2-weighted imaging and gradual enhancement from the periphery well reflected the pathologic feature, which was abundant stromal tissue. As the imaging features differed from those of the previous case report, further examination and future case reports are required. We assume that the mass arose from a remnant of the fourth pharyngeal pouch tract because it was located inside the right lobe of the thyroid

gland and the right side of the trachea.
 In conclusion, we reported a case of CASTLE. We suggest that in the case of cervical masses in the thyroid gland or juxtathyroid gland that appear to enclose the common carotid artery with the shape of lobulation and invasion to the surrounding tissue, cervical thymic abnormalities should be considered in the differential diagnosis.

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