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A case of thymic non-papillary adenocarcinoma

Hiroshi Hashimoto, Yuichi Ozeki, Koji Kameda, Hideyuki Shimazaki, Shinichi Taguchi

ABSTRACT

Introduction: Primary thymic adenocarcinoma is a notably rare neoplasm. Here, we described a case of non-papillary thymic adenocarcinoma that was comparable to adenocarcinoma not otherwise specified (NOS) according to the recent WHO classification. Case Report: A 69-year-old woman presented to our hospital with an anterior mediastinal tumor. Thymothymectomy was performed. Histological examination revealed dense proliferation of irregular tubular and partially solid atypical cells with fibrous stroma. Based on pathological and immunohistochemical findings, and the absence of primary tumors in other sites, a diagnosis of primary thymic adenocarcinoma NOS was established. Recently, enteric-type adenocarcinoma was detected in a subset of adenocarcinoma NOS, but the current case was not of this type. The patient was alive without any sign of recurrence 30 months postoperatively. Conclusion: Our case report suggests that some adenocarcinomas NOS are not enteric and that this finding should be further evaluated.

Keywords: Adenocarcinoma, Anterior mediastinal tumor, Thymic carcinoma

Hiroshi Hashimoto[1], Yuichi Ozeki[1], Koji Kameda[1], Hideyuki Shimazaki[2], Shinichi Taguchi[1]

Affiliations: [1]Department of Thoracic Surgery, National Defense Medical College, Saitama 359-8513, Japan; [2]Department of Pathology and Laboratory Medicine, National Defense Medical College, Saitama, Japan.

Corresponding Author: Hiroshi Hashimoto, Department of Thoracic Surgery, National Defense Medical College, 3-2, Namiki, Tokorozawa, Saitama 359-8513, Japan; Email: hashimoh@ndmc.ac.jp

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INTRODUCTION

Primary thymic carcinomas are rare neoplasms that occur in the anterior mediastinum. The most common histologic subtype is squamous cell carcinoma, while adenocarcinoma is extremely rare [1]. Primary thymic adenocarcinoma was first reported in 1989 [2], and comprises papillary adenocarcinomas, mucinous adenocarcinomas, carcinomas with adenoid cystic carcinoma-like features, and not otherwise specified (NOS) subtypes according to the 2015 WHO classification [3]. Papillary adenocarcinoma and mucinous adenocarcinoma are the most common subtypes [4–7]. Here, we report a case of thymic non-papillary adenocarcinoma that was comparable to adenocarcinoma NOS based on the recent WHO classification.

CASE REPORT

A 69-year-old woman visited our hospital complaining of neck discomfort. Computed tomography (CT) showed no abnormal shadows in the neck, but a 2.5 cm tumor was detected in the anterior mediastinum (Figure 1). Chest radiography did not reveal an abnormal shadow (Figure 2). Magnetic resonance imaging showed a low-signal intensity on T1-weighted images, iso-signal intensity on T2-weighted images, and a heterogenous pattern on gadolinium-enhanced images (Figure 3). Abnormal hyper

uptake (maximum standardized uptake value [SUV_{max}] = 9.72) was found at the tumor on positron–emission tomography (Figure 4). The tumor was suspected to be an invasive thymoma or thymic carcinoma.

After median sternotomy, we found that the tumor did not invade the surrounding tissues. Thymothymectomy was performed. The mass was an apparently circumscribed, gray and solid tumor, 3.0×2.4×1.4 cm in size (Figure 5). Microscopically, the tumor demonstrated a proliferation of irregular tubular and partially solid atypical cells with fibrous stroma (Figure 6). Alcian blue-PAS stain showed a small amount of extracellular mucin. Immunohistochemically, the tumor cells were positive for CK7 and CD5, and negative for CK20, napsin-A, and TTF-1 (Figure 7). The features indicated a non-papillary adenocarcinoma.

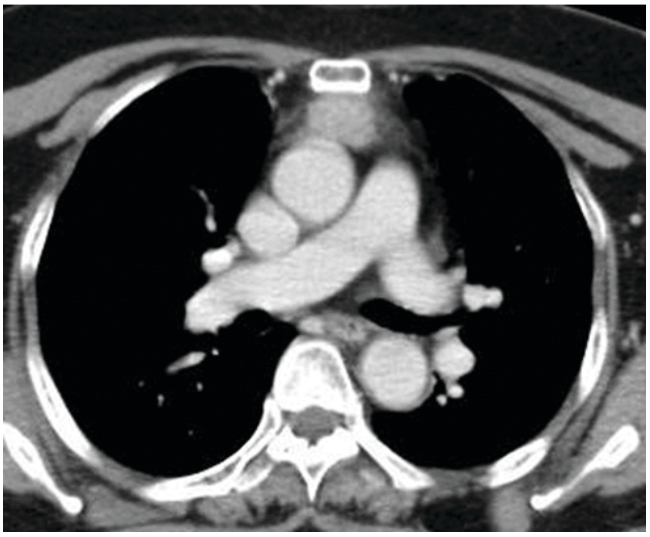

Figure 1: Chest computed tomography. A well-defined tumor (size 2.5 cm) was revealed in the anterior mediastinum.

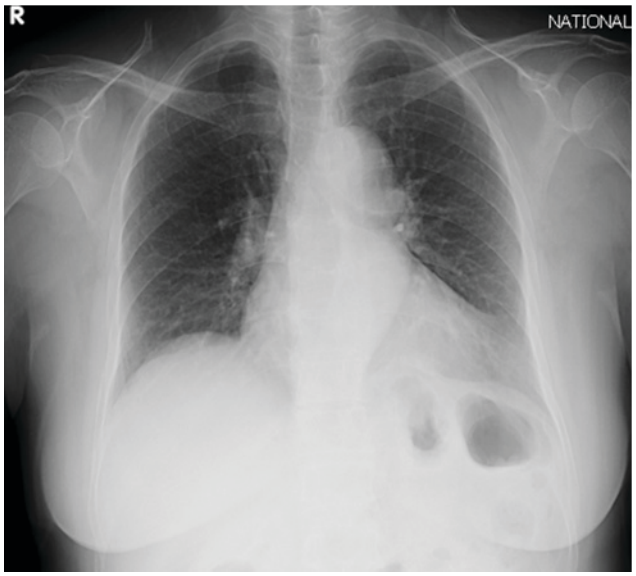

Figure 2: Chest radiography image. Chest radiography did not show an abnormal shadow.

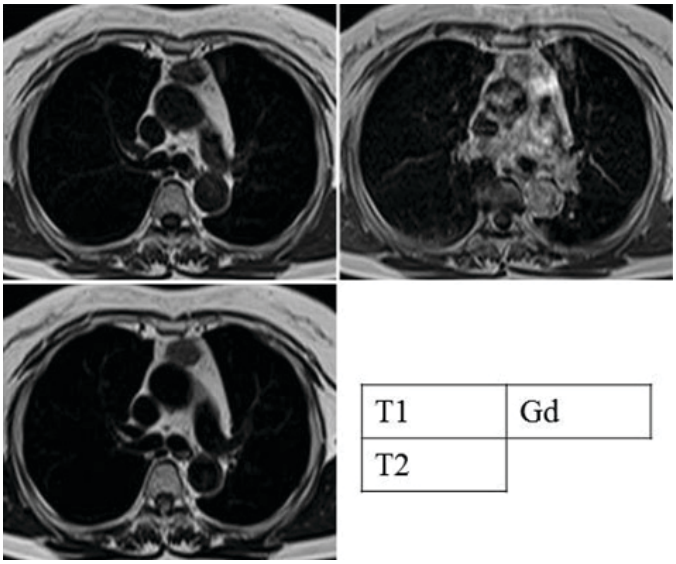

Figure 3: Magnetic resonance imaging (MRI) showed low-signal intensity on T1-weighted images, iso-signal intensity on T2-weighted images, and a heterogenous pattern on gadolinium-enhanced images.

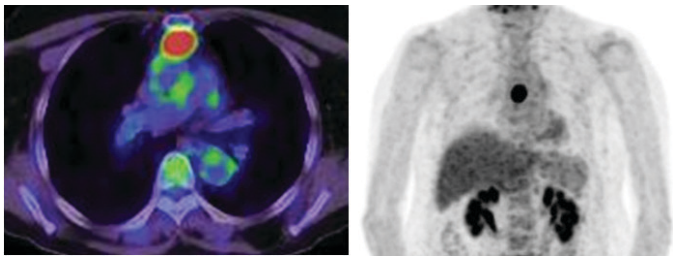

Figure 4: Positron–emission tomography. Abnormal hyper uptake (SUV_{max} = 9.72) was found at the tumor.

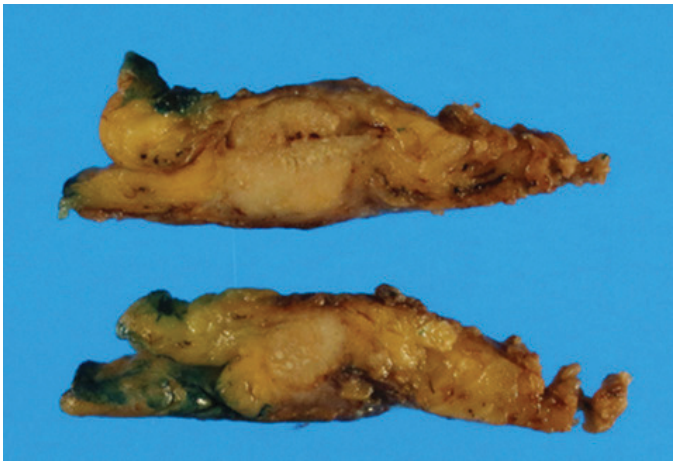

Figure 5: Macroscopic findings. Gross examination of the specimen revealed an apparently circumscribed, gray and solid tumor, sized 3.0×2.4×1.4 cm.

Physical and radiologic examinations did not reveal primary tumors elsewhere, including in the gastrointestinal tract. Based on these observations, we finally reached a diagnosis of thymic non-papillary adenocarcinoma that was comparable to adenocarcinoma NOS according to the recent WHO classification.

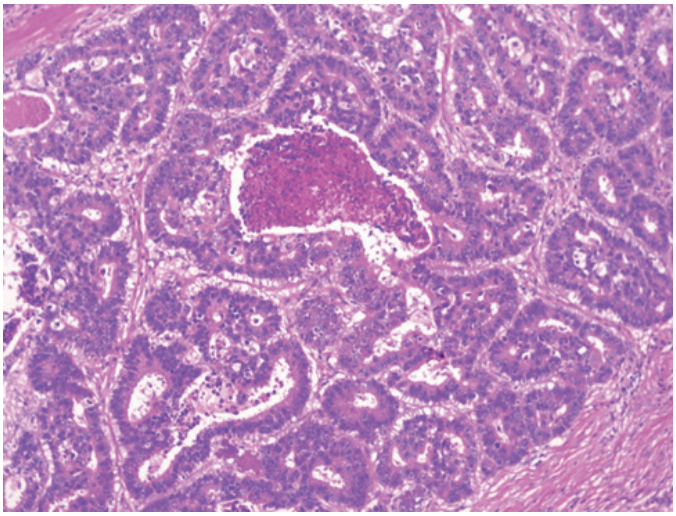

Figure 6: Microscopic findings. The tumor demonstrated a proliferation of irregular tubular and partially solid atypical cells with fibrous stroma.

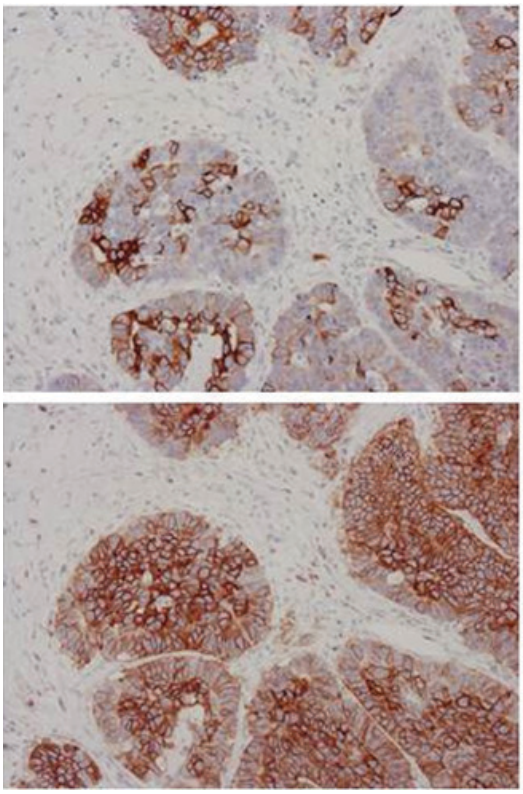

Figure 7: Immunohistochemical findings. The tumor cells were positive for CK7 and CD5, and negative for CK20, napsin-A, and TTF-1.

The patient was followed up with no additional postoperative treatment, and there was no evidence of recurrence 30 months after the procedure.

DISCUSSION

Primary thymic carcinoma is an uncommon malignant tumor that accounts for 14.6% of all thymic epithelial tumors [8]. Squamous cell carcinoma is the most common subtype and accounts for 61.8% of thymic carcinomas. Adenocarcinoma is a notably rare subtype, accounting only for 1.6–2.7% of thymic carcinomas [1, 9]. Within the subtypes according to the WHO classification, papillary adenocarcinoma usually occurs along with type A and AB thymomas and is now defined as a low-grade carcinoma [3]. High-grade adenocarcinomas with the usual focal papillary growth, such as the present case, are considered as adenocarcinoma NOS [3]. Recently, enteric-type adenocarcinoma, as shown by immunohistochemistry, was detected in a subset of mucinous adenocarcinomas and adenocarcinoma NOS [10–15]. According to Kwon et al., their nine cases (seven thymic mucinous adenocarcinomas and two thymic adenocarcinomas NOS) showed a histology similar to that of lower gastrointestinal tract adenocarcinoma [16]. Moreover, their nine cases also demonstrated an immunoreactive profile similar to that of enteric-type adenocarcinomas, which are reactive for CK20 and/or CDX2. Furthermore, they reviewed 58 previously reported thymic adenocarcinomas including their nine cases, and noted 22 mucinous adenocarcinomas and 13 adenocarcinomas NOS. Of 28 (28/35) cases for which CK20, CDX2, or MUC2 immunostaining had been performed, 25 were immunoreactive for at least one of these markers. They suggested that mucinous adenocarcinoma and adenocarcinoma NOS are morphologically and immunohistochemically similar, resembling enteric-type adenocarcinoma. However, in the present case, we also performed CK20 and CDX2 immunostaining, and confirmed the tumor cells were negative for these markers; thus, indicating that our case of adenocarcinoma NOS was not an enteric type. Therefore, it appears that some types of adenocarcinoma NOS are not enteric and further research seems necessary.

It is also important to exclude the possibility of metastasis from other sites. CD5 is a leukocyte marker expressed on thymocytes. It is considered to be useful in the diagnosis of thymic carcinoma. It is also important to exclude tumors derived from the lung and pleura by evaluating TTF-1 and calretinin. In addition to the pathological and immunohistochemical findings, clinical and radiological evaluations are also essential to exclude the presence of a primary tumor for an accurate diagnosis.

The treatment and prognosis of thymic adenocarcinoma have not been established because of its rarity. In general, low-grade histology (well-differentiated squamous cell carcinoma, mucoepidermoid carcinoma, and basaloid carcinoma), low Masaoka stage, and complete resection are factors associated with better survival in thymic carcinoma [1, 9]. Jung et al. reviewed 39 reports of primary thymic adenocarcinomas, consisting of 16 mucinous adenocarcinomas, 13 papillary adenocarcinomas, seven tubular adenocarcinomas, and three papillotubular adenocarcinomas [11]. Their report suggested that modified Masaoka stage I or II disease had better disease-free survival than stage III or IV disease, and that the mucinous/tubular subtype showed a trend

toward poorer overall survival than the papillary or papillotubular subtype.

CONCLUSION

In summary, we reported a rare case of primary thymic non-papillary adenocarcinoma that was comparable to adenocarcinoma NOS according to the recent WHO classification. Our report suggests that some adenocarcinomas NOS are not enteric, and this finding should be evaluated.

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Author Contributions

Hiroshi Hashimoto – Conception of the work, Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Yuichi Ozeki – Conception of the work, Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Koji Kameda – Conception of the work, Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Hideyuki Shimazaki – Conception of the work, Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Shinichi Taguchi – Conception of the work, Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all

aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Guarantor of Submission
The corresponding author is the guarantor of submission.

Source of Support
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Written informed consent was obtained from the patient for publication of this article.

Data Availability
All relevant data are within the paper and its Supporting Information files.

Conflict of Interest
Authors declare no conflict of interest.

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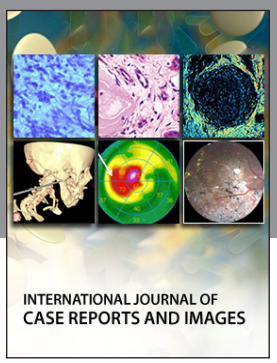

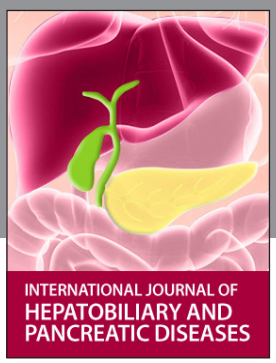

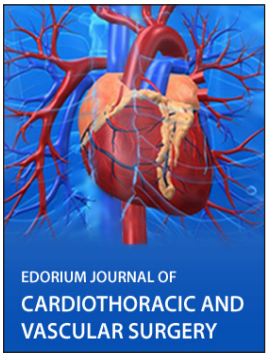

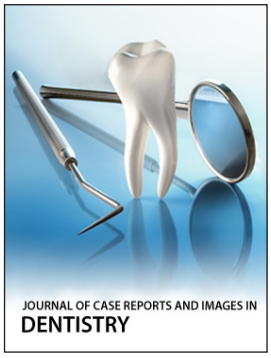

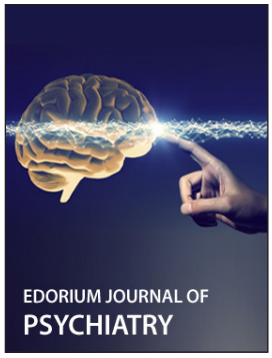

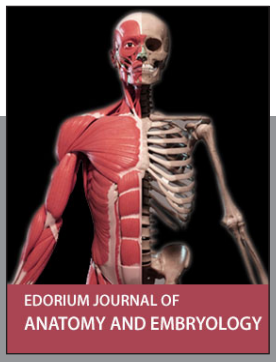

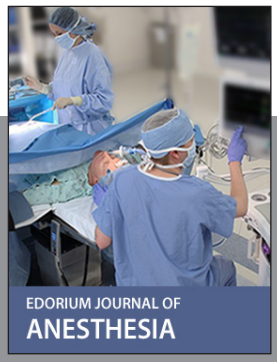

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