



Contents lists available at ScienceDirect

International Journal of Surgery Case Reports

journal homepage: www.casereports.com**Thymoma in middle mediastinum that induced tracheal compression—Case report and literature review**Teiko Sakurai ^{a,*}, Tetsuki Sakamoto ^a, Tomoko Wakasa ^b, Yoshio Ohta ^b, Hiroyuki Shiono ^a^a Department of Thoracic Surgery, Kindai University Nara Hospital, Otoda-Cho 1248-1, Ikoma, Nara 630-0293, Japan^b Department of Diagnostic Pathology and Laboratory Medicine, Kindai University Nara Hospital, Otoda-Cho 1248-1, Ikoma, Nara 630-0293, Japan**ARTICLE INFO****Article history:**

Received 5 September 2016

Received in revised form

10 December 2016

Accepted 10 December 2016

Available online 13 December 2016

Keywords:

Thymoma

Middle mediastinum tumor

Tracheal compression

Case report

ABSTRACT

BACKGROUND: A thymoma, an epithelial neoplasm of the thymus, mainly occurs in the anterior mediastinum, while few are seen in the middle mediastinum.

CASE PRESENTATION: An 83-year-old male was referred for an incidental mass in the middle mediastinum. He had severe dementia and denied symptoms. Our follow-up computed tomography (CT) examinations had revealed the progress of tracheal compression along with tumor enlargement for 2 years. At 85 years old, we performed a thymomectomy via a median sternotomy to avoid complete trachea obstruction. The pathological diagnosis was WHO type A thymoma, Masaoka stage II. One year after surgery, the patient was free of disease.

DISCUSSION: Thymomas occurring in the middle mediastinum are rare. In our review of 13 such cases, none were Masaoka stage III or IV, while the majority (9/13, 69.2%) were WHO type A or AB.

CONCLUSION: We encountered a thymoma in the middle mediastinum that showed enlargement over a 2-year period, inducing severe tracheal compression. Thymomas can occur widely in pharyngeal pouch-derived locations and should be considered in differential diagnosis of a middle mediastinum tumor.

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1. Introduction

A thymoma is a low-grade malignant epithelial neoplasm of the thymus commonly located in the anterior mediastinum [1], though limited numbers have been found in other locations, such as the neck, another mediastinum site, a lung, and the pleural cavity [2]. A normally located thymoma rarely induces tracheal compression. We present a rare case of thymoma in the middle mediastinum that induced tracheal compression, along with a review of previous reports.

2. Case presentation

An 83-year-old male was referred for an incidental tumor in the middle mediastinum detected by computed tomography (CT) prior to emergency surgery for a gastric carcinoma. The tumor was anterior to the trachea and dorsal to the innominate vein, and found to compress adjacent vessels and especially the trachea, with no invasion of surrounding tissues (Fig. 1). The serum anti-acetylcholine

receptor antibody was negative. The patient had severe dementia and denied symptoms, thus we did not immediately perform surgical treatment and provided follow-up examinations. At 85 years old, the solid mass had enlarged to 6.7 cm in length, with a 5-mm minimum tracheal inner diameter. Since the tumor showed more rapid growth than initially expected and the physical condition of the patient was generally good, except for mild short-term memory disturbance, his family members gave consent to our recommendation for surgical treatment. We performed a thymomectomy via a median sternotomy, and found that the tumor was not continuous with either the thyroid or surrounding thymic tissue.

Macroscopically, the resected specimen was a firm tumor (6.5 × 5.0 × 5.0 cm) (Fig. 2). Microscopically, the tumor considered of uniform epithelial cells with only very few small lymphocyte. Surrounding the small vessels, the perivascular lymphoid spaces were shown (Fig. 3). The diagnosis was thymoma type A, according to the World Health Organization (WHO) classification fourth edition, and Masaoka staging system Stage II. No postoperative adjuvant therapy was given and the patient was alive without recurrence 1 year after surgery.

3. Discussion

A thymoma, an epithelial neoplasm of the thymus, is the most frequent type of anterior mediastinum neoplasm [1], with only

Abbreviations: CT, computed tomography; WHO, World Health Organization; MG, myasthenia gravis; VATS, video-assisted thoracoscopic surgery.

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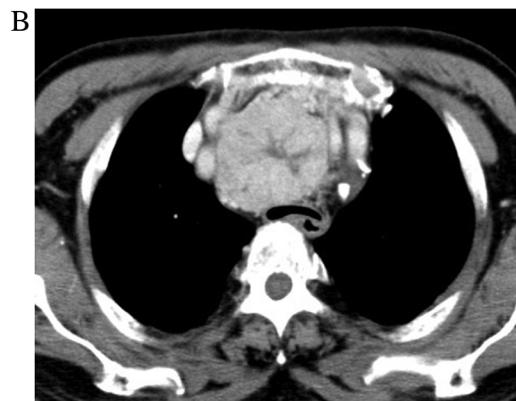


Fig. 1. Enhanced CT revealed a solitary mass severely compressing the trachea and sized $5.5 \times 5.0 \times 4.0$ cm (A). Two years after the initial diagnosis, the tumor had enlarged lengthwise to $6.7 \times 5.1 \times 5.0$ cm (B, C).

a limited number arising outside of the anterior mediastinum reported.

Our literature review found 13 cases of thymomas in the middle mediastinum, including the present (Table 1) [2–13]. Such location can induce localized symptoms caused by airway or esophagus compression [1]. In those reports, no characteristic symptom was described, though trachea compression was reported in 2 cases, the same as in the present [7]. This is the first known report of a thymoma inducing severe tracheal compression. Regarding systemic symptoms related to thymoma, only 2 cases (15.4%) presented MG,

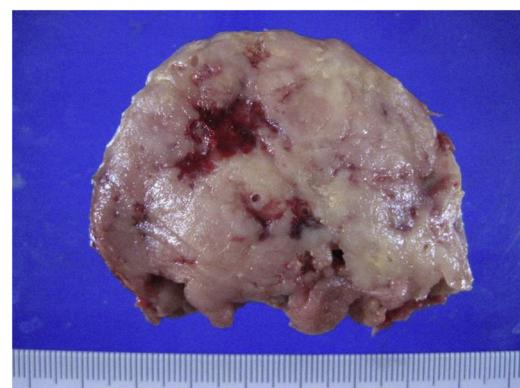


Fig. 2. The cut surface of the tumor showed a pale tan color and irregular lobulation.

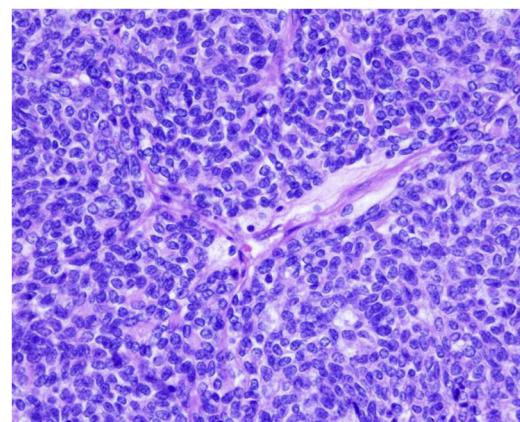


Fig. 3. Histological findings showed epithelial cells with oval nuclei and were compatible with type A thymoma (HE staining).

with the WHO pathological classification in both type B1. It has been reported that type B occurs more frequently in thymoma patients with MG than type A and AB [14].

Previous studies have described use of the WHO pathological classification system for predicting outcome of thymoma patients [14,15]. As for histologic distribution, 7% were type A, 22% type AB, and 71% type B [15]. In our review, the major histologic type was type A or AB (9/13, 69.2%), and more frequent than type B. Furthermore, all were classified as Masaoka stage I or II, and the tumors were surgically resected. Although why less invasive tumors are relatively frequent is unclear, there may be a correlation between WHO type and Masaoka stage in thymoma case reports.

Surgical resection is the most important treatment for a thymoma [14,15], with a median sternotomy considered to be the standard approach, though video-assisted thoracoscopic surgery (VATS) is universally performed as a less invasive technique. Since a tumor in the middle mediastinum is surrounded by the superior vena cava, trachea, aorta, and left brachiocephalic vein, other surgical approaches have been selected, including a thoracotomy in 3 cases (Table 1). We chose a median sternotomy for resection of the relatively large tumor in order to prevent injury to the phrenic and recurrent nerves.

During embryological development, the thymus is mainly derived from the third and fourth pharyngeal pouch. Failure of the thymic gland to migrate into the anterior mediastinum results in isolated foci in unusual locations, described as ectopic thymic tissue [16,17]. A term 'ectopic' thymoma was used in all of previous reports (Table 1). However, using surgical-anatomic study findings, Jaretzki noted that thymic tissue is frequently distributed in pre-tracheal tissue, mediastinal fat from the thyroid level to the

Table 1

Previous reports of thymoma in the middle mediastinum.

Author (Ref. #)	Age	Sex	Symptom	MG	Approach	Thymectomy	Diameter (cm)	Type/stage ^b	Outcome
Kojima [2]	60	F	–	No	Rt. thoracotomy	No	5.5	AB/II	–
Kanzaki [3]	60	F	–	No	Median sternotomy	Total	6	AB/I	–
Minniti [4]	53	M	–	No	Not described	No	7	AB/I	–
Kuzdzal [5]	69	F	–	No	Anterolateral thoracotomy	No	10	Metaplastic/I	–
Nakamura [6]	69	F	Precordial discomfort	No	VATS	No	7.5	A/I	2y4m/DFS
Huang [7]	71	F	–	No	Median sternotomy	No	7	A/I	3y/DFS
Sakurai [8]	61	M	–	No	VATS	No	3.5	AB/I	6m/DFS
Chung [9]	59	F	–	No	VATS	No	4.5	A+B2/I	–
Shikada [10]	56	M	–	No	Anterolateral mini-thoracotomy	No	6.5	AB/II	9y8m/DFS
Takizawa [11]	69	F	Ptosis, dysphagia, muscle weakness	Yes	Median sternotomy	ET	2	B1/II	5y/DFS
Koezuka [12]	51	M	Ptosis, diplopia, muscle weakness	Yes	VATS	ET	2	B1/I	–
Tokuno [13]	42	F	–	No	Median sternotomy ^a	ET	4.9	AB/I	1y/DFS
Present case	85	M	–	No	Median sternotomy	No	6.5	A/II	1y/DFS

MG: myasthenia gravis, VATS: video-assisted thoracoscopic surgery, ET: extended thymectomy.

^a Conversion from VATS.^b According to WHO classification/Masaoka staging system.

diaphragm, and even subcarinal fat [17,18]. Thus, tumors can arise from so-called ‘ectopic’ thymic tissue in an unusual location. Such tissues were subsequently defined as ‘a variation of normal’ and not ‘ectopic’ thymic tissue in a monograph [19]. Therefore, we consider that a thymic tumor should not be ruled out when occurring in an unusual location, but rather be considered in differential diagnosis of a neoplasm in a pharyngeal pouch-derived site.

4. Conclusion

We report the first case of a thymoma in the middle mediastinal that induced severe tracheal compression. A thymoma can occur widely in locations derived from the pharyngeal pouch and should always be considered in differential diagnosis of an undefined neoplasm, even those in the middle mediastinum.

Conflicts of interest

The authors have no conflicts of interest to disclose.

Funding

There are no sources of funding for this research.

Ethical approval

For this type of report, the Ethics Committee of Kindai University gave their approval without a specific judgement, thus there is no reference number.

Patient consent

Written informed consent was obtained from the patient and family members for publication of this case report and related images.

Author contribution

T. Sakurai wrote the manuscript.

T. Sakamoto and H. Shiono provided treatment to the patient, and contributed to writing the manuscript.

T. Wakasa and Y. Ohta diagnosed the disease as pathologists, and provided advise for composing the manuscript.

Guarantor

The Guarantor of this paper is H. Shiono, who accepts full responsibility for this work, had access to the data, and controlled the decision to publish.

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