ISSN 2307-8960 (online)

World Journal of *Clinical Cases*

World J Clin Cases 2021 September 16; 9(26): 7614-7962

Published by Baishideng Publishing Group Inc

World Journal of Clinical Cases

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Thrice Monthly Volume 9 Number 26 September 16, 2021

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NAME OF JOURNAL
World Journal of Clinical Cases

ISSN
ISSN 2307-8960 (online)

LAUNCH DATE
April 16, 2013

FREQUENCY
Thrice Monthly

EDITORS-IN-CHIEF
Dennis A Bloomfield, Sandro Vento, Bao-Gan Peng

EDITORIAL BOARD MEMBERS
https://www.wjgnet.com/2307-8960/editorialboard.htm

PUBLICATION DATE
September 16, 2021

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World J Clin Cases 2021 September 16; 9(26): 7870-7875

DOI: 10.12998/wjcc.v9.i26.7870

ISSN 2307-8960 (online)

CASE REPORT

Uniportal video-assisted thoracoscopic surgery for complex mediastinal mature teratoma: A case report

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Author contributions: Hu XL and Zhu WY were the patient's doctor-in-charge, who performed the operation; Zhu WY provided the main idea of the manuscript; Hu XL and Zhang D reviewed the literature and drafted the manuscript; Zhu WY was responsible for the revision of the manuscript for important intellectual content; all authors issued final approval for the version to be submitted.

Informed consent statement: Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

Conflict-of-interest statement: The authors declare that they have no conflict of interest.

CARE Checklist (2016) statement: The authors have read the CARE Checklist (2016), and the manuscript was prepared and revised according to the CARE Checklist (2016).

Open-Access: This article is an open-access article that was selected by an in-house editor and fully peer-reviewed by external

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Abstract

BACKGROUND

Mediastinal mature teratoma is the most common histological type of primary extragonadal germ cell tumor. In this report, we describe a rare case of giant mature teratoma located primarily in the anterior mediastinum and causing partial atelectasis of the upper and middle lobes of the right lung, as well as extrinsic compression of the right atrium.

CASE SUMMARY

A 31-year-old male with a giant mediastinal mature teratoma presented with progressive exertional dyspnea and chest pain for 1 mo. Computed tomography of the chest indicated the diagnosis of anterior mediastinal teratoma. The patient underwent right uniportal anterior approach video-assisted thoracoscopic surgery (VATS). *En bloc* resection of the giant teratoma, wedge resection of the upper and middle lobes of the right lung, resection of the thymus and partial excision of the pericardium were successfully performed. The pathological diagnosis revealed a mature cystic teratoma with foreign-body reaction that was closely related to the right lung, atrium dextrum, superior vena cava and ascending aorta. An atrophic thymic tissue was also discovered at the external teratoma surface. The patient was discharged on postoperative day 7.

CONCLUSION

This is the first report of the use of uniportal VATS for complete resection of a teratoma in combination with wedge resection of the right upper and middle lung lobes and partial resection of the pericardium.

Manuscript source: Unsolicited manuscript

Specialty type: Medicine, research and experimental

Country/Territory of origin: China

Peer-review report's scientific quality classification

Grade A (Excellent): A
Grade B (Very good): B, B
Grade C (Good): 0
Grade D (Fair): 0
Grade E (Poor): 0

Received: March 29, 2021
Peer-review started: March 29, 2021
First decision: June 24, 2021
Revised: July 6, 2021
Accepted: August 13, 2021
Article in press: August 13, 2021
Published online: September 16, 2021

P-Reviewer: Kermenli T, Kumar A, Yalçinkaya İ
S-Editor: Gao CC
L-Editor: A
P-Editor: Zhang YL

Key Words: Uniportal video-assisted thoracoscopic surgery; Mediastinal mature teratoma; Complex adhesions and infiltration; Video-assisted thoracoscopic surgery; Case report

Core Tip: Mediastinal mature teratoma accounts for the majority of primary germ cell tumors in the mediastinum, which can be resected by traditional surgery. We present a rare case of mediastinal mature teratoma with complex adhesions and infiltration resected using uniportal video-assisted thoracoscopic surgery (VATS) technology. The patient had no postoperative complications and was completely asymptomatic at the first-year postoperative follow-up. As VATS currently plays an increasingly significant role in the therapy of teratomas, our report indicated that uniportal VATS for resection of mediastinal teratomas with complex adhesions and infiltration can be successfully conducted.

Citation: Hu XL, Zhang D, Zhu WY. Uniportal video-assisted thoracoscopic surgery for complex mediastinal mature teratoma: A case report. *World J Clin Cases* 2021; 9(26): 7870-7875
URL: https://www.wjgnet.com/2307-8960/full/v9/i26/7870.htm
DOI: https://dx.doi.org/10.12998/wjcc.v9.i26.7870

INTRODUCTION

Germ cell tumors, frequently observed in the gonads, can also be found in the (anterior) mediastinum, which is the most predominant extragonadal site, and manifests as a mature teratoma accounting for the majority of primary germ cell tumors in the mediastinum[1]. Most mediastinal mature teratomas are insidious and therefore are generally discovered during abnormal medical examinations. Teratomas usually compress adjacent structures, predominantly altering the respiratory and circulatory systems[2]. Another typical feature of mediastinal mature teratoma is tumor bleeding or rupture into the bronchus, pleura, vessel, or pericardium[3]. Mediastinal mature teratoma can be definitively diagnosed by histopathological examination as the tumors are generally composed of ectodermal, mesodermal, and endodermal derivatives[4]. Video-assisted thoracoscopic surgery (VATS) technology has evolved from its multiport origins to even less invasive approaches due to its identified advantages compared with open surgery for a series of thoracic cavity tumors[5]. During the course of its evolution, Uniportal VATS (UniVATS) has emerged, which has proved to be superior to multiport VATS and has extended the surgical boundaries[6]. In the present case, we describe a giant mature teratoma in the anterior mediastinum of a 31-year-old male with complex rupture into the right lung and compression of the right atrium, as well as the superior vena cava and ascending aorta, which was completely resected using UniVATS.

CASE PRESENTATION

Chief complaints

A 31-year-old male patient was admitted to our hospital complaining of progressive chest pain, exertional dyspnea and cough, accompanied by recurrent hemoptysis, and a low-grade fever. The patient had been treated with antibiotics for 5 d, but this was unsuccessful in relieving his symptoms.

History of present illness

Chest pain and exertional dyspnea in this patient started 6 wk ago, and recurrent hemoptysis developed 10 d previously.

History of past illness

The patient had no previous medical history.

Personal and family history

The patient had no disease-related personal or family history.

Physical examination

Physical examination on admission showed that the patient was normothermic and normotensive, without tachypnea, cyanosis or tachycardia. In addition, no abnormalities of features, thyroid, lung fields, abdomen, skin, arms and legs, lymph nodes, or testicles were observed during the examination. Neurological examination and routine stool as well as urine analysis were generally within normal limits. A 12-lead resting electrocardiogram demonstrated sinus rhythm.

Laboratory examinations

Routine blood test showed a slightly higher C-reactive protein level of 12.98 mg/L, suggesting a mild inflammatory response. Tumor markers, such as CEA, AFP, CA-125, NSE, β-HCG, and CA 19-9, were within normal ranges, except for TK1 (5.60 pmol/L). In addition, no abnormalities were found following respiratory function tests. Transthoracic echocardiography in the parasternal view revealed a normal-sized heart with normal function and blood flow velocities.

Imaging examinations

An earlier computed tomography (CT) report from a local hospital showed a large oval mass measuring approximately 9.0 cm × 6.0 cm × 5.0 cm in the anterior mediastinum. To further define the location, nature, composition, characteristics, and operability of the tumor, contrast-enhanced CT of the chest and soft tissue three-dimensional reconstruction were performed, which confirmed the presence of a right anterior mediastinal mass with extrinsic compression of the right atrium and right lung (Figure 1).

Further diagnostic work-up

Moreover, contrast-enhanced CT further revealed an oval, polycystic, heterogeneous mass with fatty, liquid and calcified components in the right anterior mediastinum, measuring 8.3 cm × 4.4 cm × 4.8 cm in size. The mass was closely related to the surrounding tissues and organs, presenting as infiltration of the right upper and middle lobes with pneumonitis, and encased the origin or terminal of the superior vena cava as well as the ascending aorta. Heterogeneous enhancement of the mass was confirmed following administration of Ultravist 300.

FINAL DIAGNOSIS

The final diagnosis in the presented case was mediastinal mature teratoma.

TREATMENT

The patient underwent VATS *via* a uniportal right-sided anterior approach with surgical resection *via* the 5th intercostal space on day 7 after admission. The extracorporeal circulation system was prepared in advance for possible intraoperative haemorrhage. Intraoperative thoracoscopic exploration revealed extensive adhesions around the thorax, including a large mass measuring approximately 9 cm × 6 cm × 5 cm, located in the right anterior mediastinum. The tumor was located above the superior border of the left brachiocephalic vein and was partially solid and cystic in nature. Moreover, the tumor was adjacent to the pericardium, the terminal superior vena cava and the left brachiocephalic vein, and infiltrated the right upper and middle lung lobes and the right phrenic nerve, and it was extremely difficult to separate the mass. We therefore performed en bloc resection of the giant teratoma, a wedge resection of the upper and middle lobes of the right lung, and partial pericardial resection in this patient. The tumor was resected with utmost care to preserve vital structures, such as the phrenic nerve, recurrent laryngeal nerve, aorta, superior vena cava, left brachiocephalic vein, pulmonary vessels, and subclavian vessels. In addition,

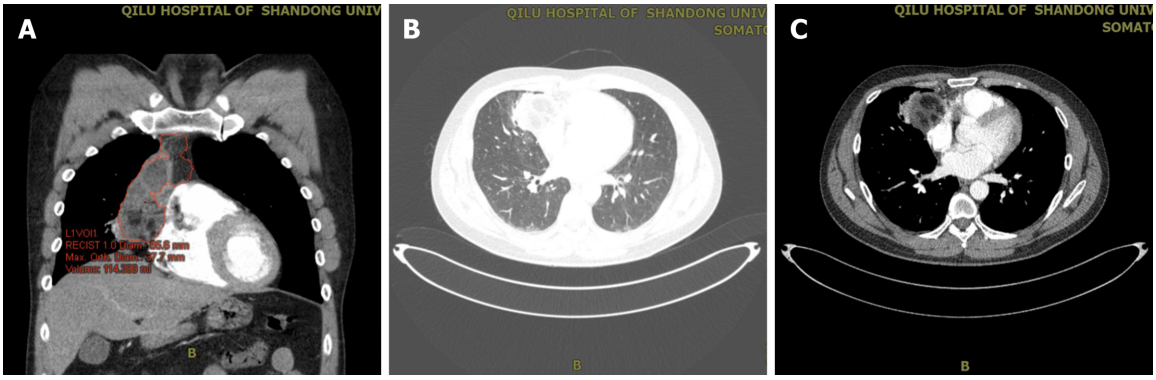

Figure 1 Enhanced chest computed tomography scan before surgery. A: In the coronal mediastinal window, the presence of a right hilar enlargement.an emerging cavity with high-density consolidation, high-density opacities occupying the right upper and middle lobe, diffuse high-density opacities with shadows and stripes, irregular soft tissue of mixed density in the anterior right upper mediastinum, and consolidation with cavitation and calcification adjacent to the mediastinum are shown; B: In the mediastinal window; C: In the lung window.

the enlarged mediastinal lymph nodes and the right portion of the thymus were also resected to ensure that no tumor tissue remained. Basal thoracostomy tubes were placed.

The gross pathological diagnosis revealed a predominantly mature cystic teratoma with a foreign-body reaction that measured 10 cm × 6 cm × 4.5 cm (Figure 2). The tumor infiltrated the lung, and the tissue was filled with sebaceous materials. An immunohistochemical diagnosis was conducted and showed positive markers of epithelial as well as T cells. However, neither mesenchymal nor neural elements were observed (data not shown). Moreover, there was no histological or morphological evidence of malignancy in the tumor (Figure 3). In addition, atrophic thymus and inflammatory lymph nodes were observed in the tissue surrounding the tumor.

OUTCOME AND FOLLOW-UP

The patient had no post-operative complications, and was discharged on the 7th post-operative day. Moreover, he was completely asymptomatic at the first-year post-operative follow-up.

DISCUSSION

Mature teratomas comprise the vast majority of all histological types of germ cell tumors, which predominantly originate from the gonads. Extragonadal germ cell tumors are found in the mediastinum, peritoneum, sacral pineal gland, thyroid, and gastrointestinal tract, of which the anterior mediastinum is the most frequent extragonadal site and these tumors are named mediastinal teratomas[7]. Mediastinal teratoma should be differentiated from a series of thoracic tumors, including mediastinal lymphomas, thymomas, esophageal cysts, bronchogenic cysts, pericardial tumors, lymphangiomas, neurogenic tumors, and mesenchymal tumors[8].

The majority of mediastinal teratomas are benign, which means the tumors grow quite slowly and are almost asymptomatic in the early stages. Few cases of rapidly growing mediastinal mature teratomas have been published. Thus, most of these tumors were diagnosed incidentally during medical examination[9]. However, the specific mechanism of action of mediastinal mature teratoma development is unclear. It is documented that tumors which proliferated and grew to a certain extent could induce extrinsic compression of adjacent vital structures, which results in representative clinical symptoms including chest congestion, bronchial cough, blood-streaked phlegm, hemoptysis, as well as dyspnea[10]. Moreover, the tumors can also rupture into adjacent structures, such as the pericardium, bronchus, and pleural space, where sebaceous materials or hair can be observed by electric bronchoscope or fine needle aspiration biopsy. As an extreme example, a case report illustrated a mature mediastinal teratoma with complex rupture into the skin with a cysto-cutaneous fistula, which produced sebum and hair[7]. Some teratomas contain malignant components, indicating that radiotherapy and chemotherapy may be acceptable

Figure 2 The images of surgical specimen histopathology. Surgical specimen' largest diameter was 10 cm. A: The macroscopical image of the mass; B: Cut of the mass shows sebaceous material; C: Image of the cartilage tissue of the mass.

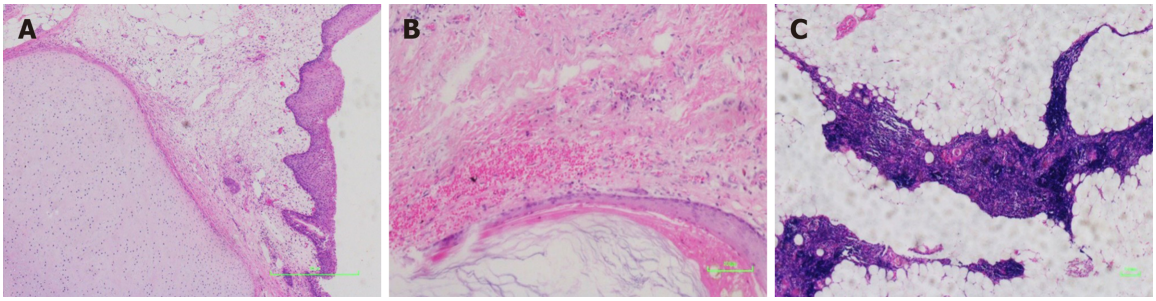

Figure 3 Hematoxylin and eosin staining of teratoma. Histopathologic analysis revealed mature tissues, cartilage tissue and sebaceous material with the mass. A: Lower magnification (40 ×, scale bar = 250 μm) shows cartilage tissue; B: Loupe image of the tumor shows mature tissues (100 ×, scale bar = 100 μm); C: Higher magnification (200 ×, scale bar = 33 μm) shows sebaceous material.

management options[11].

In clinical practice, the most effective diagnostic modality for mediastinal teratomas is CT, which provides unambiguous signs of tumor rupture as well as infiltration of adjacent structures, crucial for early diagnosis and surgical decisions[12].

Surgical resection is the mainstay of teratoma treatment, particularly for mature teratomas. The choice of surgical approach for mediastinal teratomas depends to a large extent on the size, location, invasiveness, histological classification, metastases, the relationship of the tumor to the relevant vital structures, as well as the experience of the surgeon. Traditionally, a lateral, anterolateral or median sternotomy is the primary approach for resecting teratomas[11]. Moreover, robotic surgery and VATS-assisted surgery are increasingly used in the surgical excision of mediastinal tumors [13]. In recent years, the application of VATS has played an important role in thymectomy, which also provides the rational for teratoma resection[6,14]. However, there are few reports on the application of VATS to resect mediastinal teratomas. There are always extensive and widespread adhesions between mediastinal teratomas and adjacent vital organs and tissues. In addition, some teratomas are also inflamed, infiltrated, and ruptured. Therefore, great care must be taken during resection of the tumor. Serious cardiopulmonary complications, such as hemorrhage, cardiac arrest, and diaphragmatic paralysis, may occur when adjacent vital structures are damaged [15]. In addition, some anesthesia-related problems may occur during mediastinal tumor excision due to adjacent structures such as the superior vena cava. For example, Kumar *et al*[16] reported a case of intraoperative catastrophe during benign mediastinal tumor mass excision, which demonstrated that adequate preoperative assessment, as well as standby extracorporeal circulatory support is critical for successful management. In the present case, we successfully applied the UniVATS technique to resect a large infiltrating mediastinal teratoma with a significant degree of structural and functional preservation of the adjacent tissues and organs.

CONCLUSION

Mediastinal mature teratomas are uncommon, asymptomatic benign tumors that are

often diagnosed on CT scans obtained for unrelated reasons. Complete surgical excision is the best treatment for mediastinal teratomas. Currently, VATS plays an increasingly significant role in the treatment of teratomas. Our report revealed that UniVATS for the resection of mediastinal teratomas with complex adhesions and infiltration can be successfully conducted.

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