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## Three-dimensional image simulation of primary diaphragmatic hemangioma: A case report

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### Abstract

#### BACKGROUND

Fewer than 200 cases of diaphragmatic tumors have been reported in the past century. Diaphragmatic hemangiomas are extremely rare. Only nine cases have been reported in English literature to date. We report a case of cavernous hemangioma arising from the diaphragm. Pre-operative three-dimensional (3D) simulation and minimal invasive thoracoscopic excision were performed successfully, and we describe the radiologic findings and the surgical procedure in the following article.

#### CASE SUMMARY

A 40-year-old man was referred for further examination of a mass over the right basal lung without specific symptoms. Contrast-enhanced computed tomography revealed a poorly-enhanced lesion in the right basal lung, abutting to the diaphragm, measuring 3.1 cm × 1.5 cm in size. The mediastinum showed a clear appearance without evidence of abnormal mass or lymphadenopathy. A preoperative 3D image was reconstructed, which revealed a diaphragmatic lesion. Video-assisted thoracic surgery was performed, and a red papillary tumor was found, originating from the right diaphragm. The tumor was resected, and the pathological diagnosis was cavernous hemangioma.

#### CONCLUSION

In this rare case of diaphragmatic hemangioma, 3D image simulation was helpful for the preoperative evaluation and surgical decision making.

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**Core tip:** Diaphragmatic hemangioma is rare, and its diagnosis is challenging. We present the case of a 40-year-old man with incidental abnormal findings in chest imaging studies. Contrast-enhanced computed tomography revealed a poorly-enhanced lesion in the right basal lung, abutting to the diaphragm. Three-dimensional (3D) image simulation revealed a supra-diaphragmatic tumor. Successful tumor resection followed by primary repair of diaphragm was performed *via* minimally invasive thoracoscopic surgery. The pathological findings confirmed a primary cavernous hemangioma of the diaphragm. In this rare case of diaphragmatic hemangioma, 3D image simulation was helpful for the preoperative evaluation and surgical decision making.

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## INTRODUCTION

Diaphragmatic tumors are very rare. Fewer than 200 cases have been reported in the past century<sup>[1]</sup>. Most diaphragmatic tumors are benign, including bronchogenic cyst, lipoma, and hemangioma. Diaphragmatic hemangiomas are extremely rare, and only nine cases have been reported in the English literature to date<sup>[2-10]</sup>. We report a rare case of cavernous hemangioma arising from the diaphragm. Pre-operative three-dimensional (3D) simulation and minimally invasive thoracoscopic excision were performed successfully, and we describe the radiologic findings and the surgical procedure in the following article.

## CASE PRESENTATION

### Chief complaints

A mass over the right basal lung field was found incidentally.

### History of present illness

A 40-year-old man presented to our hospital with a one-day history of hematuria that was diagnosed due to a ureteral stone after examination. Computed tomography (CT) of the abdomen revealed a mass over the right basal lung field found incidentally. He was referred to a chest surgeon. Contrast-enhanced CT of the chest was arranged for further evaluation.

### History of past illness

The patient denied a history of hypertension, diabetes mellitus, or coronary artery disease. He had no known drug or food allergies. He also denied a history of operation, trauma, or blood transfusion.

### Personal and family history

The patient had no significant personal or family history.

### Physical examination upon admission

The patient's vital signs upon arrival were as follows: Body temperature 36.5°C, heart rate 74 beats/min, respiratory rate 18 times/min, blood pressure 156/84 mmHg, and oxygen saturation 98% on room air. He was alerted and oriented. The physical examination disclosed clean breathing sounds bilaterally, no chest tenderness, and no chest wall deformity. There was no specific finding of other systems.

### Laboratory examinations

A complete blood count was obtained showing a white blood cell count of  $8.75 \times 10^6/L$ , hemoglobin level of 15.7 g/dL, and platelet count of  $222 \times 10^9/L$ . Electrolyte, blood biochemistry, and coagulation tests were all in normal range.

### **Imaging examinations**

Contrast-enhanced CT of the chest revealed a poorly-enhanced lesion in the right basal lung, abutting the right diaphragm, measuring 3.1 cm  $\times$  1.5 cm in size (Figure 1). The mediastinum showed a clear appearance without evidence of abnormal mass or lymphadenopathy.

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## **FINAL DIAGNOSIS**

The resected tumor was sent for pathology examination after the operation. Grossly, the tumor was 3.5 cm  $\times$  1.5 cm  $\times$  1.3 cm in size and brown in color, with a soft consistency. Microscopically, the section showed a vascular lesion composed of dilated cavernous vascular spaces separated by irregular vascular walls (Figure 2). The pathological findings were compatible with those of cavernous hemangioma.

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## **TREATMENT**

It was difficult to differentiate its origin from that of lung parenchyma or diaphragm tumor. A preoperative 3D image was reconstructed, which revealed a diaphragmatic lesion (Figure 3). The patient underwent video-assisted surgery. A thoracoscope was inserted into the thoracic cavity *via* an incision in the 8<sup>th</sup> intercostal space. Intraoperative findings revealed a lobulated reddish tumor located at the lateral aspect of the right diaphragm with a wide-based pedicle (Figure 4). The patient underwent a thoracoscopic procedure with tumor resection, followed by primary repair of the diaphragm. An energy device (LigaSure™ vessel sealing technology, Medtronic) was used because of the presence of an abundant feeding vessel which bled easily on touch.

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## **OUTCOME AND FOLLOW-UP**

The patient's postoperative course was uneventful, and he was found to be tumor free during the 2-year follow-up.

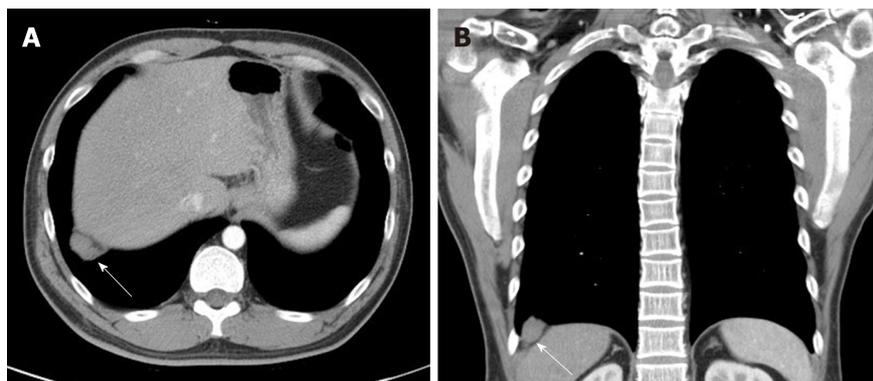
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## **DISCUSSION**

Primary diaphragmatic tumors are very rare. In the past century, fewer than 200 cases have been reported<sup>[1]</sup>. Most diaphragmatic tumors are benign, including cystic mass (bronchogenic cyst, for example), lipoma, hemangioma, angiofibroma, neurofibroma, schwannoma, leiomyoma, teratoma, and endometrioma. Malignant diaphragmatic tumors include rhabdomyosarcoma, fibrosarcoma, sarcoma, hemangiopericytoma, germ cell tumors, pheochromocytoma, and leiomyosarcoma. Diaphragmatic hemangiomas are extremely rare, and only nine cases have been reported in the English literature to date<sup>[2-10]</sup> (Table 1).

Most diaphragmatic tumors are asymptomatic and, therefore, often incidentally found. Associated symptoms are often related to the compression effect of huge tumors, including chest pain, shortness of breath, or abdominal pain. CT discloses more information about these tumors, such as size, location, and relationship with other structures. Using CT, we can also distinguish whether a diaphragmatic tumor is cystic or solid in nature. Magnetic resonance imaging (MRI) with contrast could help evaluate the internal components of the tumor without any concern of radiation dosage.

It is common to diagnose hepatic hemangioma by abdominal dynamic three-phase CT. Kono *et al*<sup>[5]</sup> reported a case of diaphragmatic hemangioma using dynamic imaging findings. On contrast-enhanced dynamic MRI images of T1-weighted imaging, the lesion showed a gradual enhancement pattern, which was maintained in the late phase. It describes the possibility of differentiating diaphragmatic hemangioma from other diaphragm tumors. In our case, two-phase contrast-enhanced CT of the chest disclosed a poorly-enhanced lobulated soft-tissue lesion, measuring 3.1 cm  $\times$  1.5 cm in size, in the right basal lung. Dynamic three-phase CT might provide more information. However, other vascular-rich tumors, such as



**Figure 1** Contrast-enhanced chest computed tomography images illustrating a poorly-enhanced lesion (arrow) in the right basal lung, abutting the right diaphragm. A: Axial view; B: Coronal view.

hemangiopericytoma and angiofibroma, should be included in the differential diagnosis as well. Pathological examination is still required to confirm the diagnosis.

Benign diaphragmatic tumors could be observed. Image-guided needle biopsy is considered only for some patients because of limitations imposed by tumor size, diaphragm motion during inspiration and expiration, and risk of bleeding. Surgical treatment of a diaphragmatic tumor is indicated for patients with a symptomatic benign tumor or a resectable malignant tumor, or for patients requiring a definitive diagnosis. However, due to the small size of the tumor and risk of bleeding, we performed surgical intervention for our patient instead of biopsy.

Sometimes, it is difficult to distinguish between tumors originating from the lung, diaphragm, or intraabdominal organs using two-dimensional imaging. Their origin determines the surgical approach, which is either laparoscopy or thoracoscopy. Thapar *et al*<sup>[11]</sup> described a case of a diaphragmatic tumor that was thought to be an atypical hepatic mass preoperatively. 3D image simulation might be of help in such a situation.

The usefulness of 3D angiography using multiple detector CT in thoracic surgery was first reported by Watanabe *et al*<sup>[12]</sup> in 2003. Over the past decade, advanced software provides high-quality 3D models of the pulmonary vessels and the tracheobronchial tree. The clinical applications of 3D lung modeling in surgical simulation and navigation systems are safe and useful<sup>[13]</sup>. In this case, we built a 3D image simulation (Mimics Innovation Suite 21.0, Materialise) that clarified the relationship of the tumor and neighboring organs. The simulation revealed that it was a diaphragmatic tumor abutting the right basal lung, without any obvious extra blood supply. Thoracoscopic tumor resection was deemed suitable for this patient and it was performed smoothly.

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## CONCLUSION

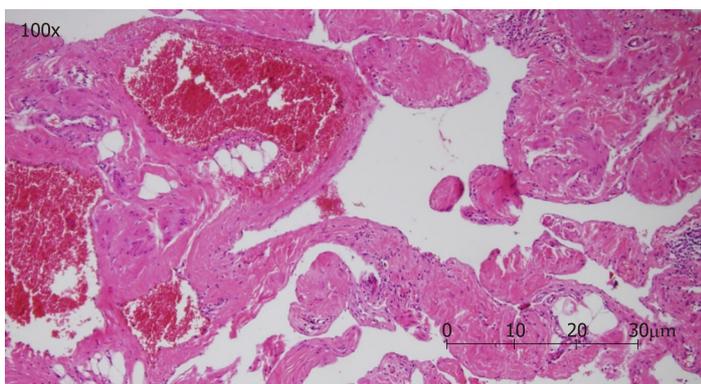
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3D image simulation clarified the relationship of the tumor and neighboring organs. In this rare case of diaphragmatic hemangioma, 3D image simulation was helpful for the preoperative evaluation of the tumor and surgical decision making.

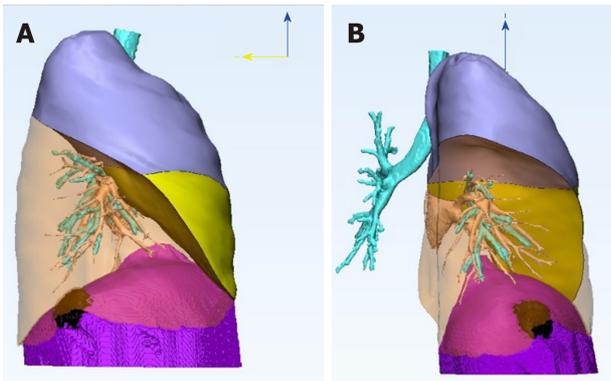
**Table 1** Nine cases of diaphragmatic hemangiomas reported in the English literature to date

Ref.	Gender	Age (yr)	Clinical magnification	Diagnostic modality	Tumor location	Treatment	Outcome
Kaniklides <i>et al</i> <sup>[2]</sup> , 1999	Male	4	Large numbers of subcutaneous vessels over left upper abdominal wall	CT, MRI, angiography	Supraphrenic, left	Surgical resection with laparotomy plus thoracotomy	Good
Ohsaki <i>et al</i> <sup>[3]</sup> , 2000	Female	31	Chest pain on the deep inspirations	CT, MRI, bone scintigraphy (99mTc-hydroxymethylene diphosphonate)	Supraphrenic, right	Surgical resection <i>via</i> thoracic approach	Good
Cacciaguerra <i>et al</i> <sup>[4]</sup> , 2001	Female	0	Neonatal respiratory failure and hydrops fetalis	Cardiac sonography, CT	Supraphrenic, right	Surgical resection with median sternotomy	Good
Kono <i>et al</i> <sup>[5]</sup> , 2006	Female	75	No specific discomfort	CT, MRI	Supraphrenic, right	Surgical resection <i>via</i> thoracoscopic approach	good
Ino <i>et al</i> <sup>[6]</sup> , 2010	Male	64	No specific discomfort	CT, PET scan	Subphrenic, left	Surgical resection <i>via</i> laparoscopic approach	Good
Tsang <i>et al</i> <sup>[9]</sup> , 2011	Male	0	Massive pleural effusion, pericardial effusion with cardiac tamponade	Chest X ray, cardioechography	Supraphrenic and subphrenic, left	Surgical resection with median sternotomy, extended to upper abdomen	Good
Ueno <i>et al</i> <sup>[7]</sup> , 2013	Male	51	No specific discomfort	CT, PET scan	Supraphrenic, right	Surgical resection <i>via</i> thoracic approach	Good
Yao <i>et al</i> <sup>[8]</sup> , 2013	Unknown	0	Dyspnea	CT, MR	Supraphrenic, right	Interventional vascular embolization	Good
Wu <i>et al</i> <sup>[10]</sup> , 2015	Female	0	Progressive respiratory distress and massive right hydrothorax	Sonography, CT, MRI	Supraphrenic, right	Interventional vascular embolization	Good

CT: Computed tomography; MRI: Magnetic resonance imaging; PET: Positron emission tomography.



**Figure 2** Pathologic image showing a vascular lesion composed of dilated cavernous vascular spaces separated by irregular vascular walls microscopically. The pathologic findings (hematoxylin and eosin staining; magnification, 100×) are compatible with those of cavernous hemangioma.



**Figure 3** Three-dimension reconstruction image illustrating the relationship of the tumor and neighboring organs. We mark the right upper lobe in lavender color, the right middle lobe in yellow, and the diaphragm in purple. The right lower lobe is transparent to see the black lesion clearly. The three-dimension reconstruction image reveals a diaphragmatic lesion, without lung parenchyma involvement. A: Right lateral view of the right lung; B: Posterior view of the right lung.



**Figure 4** Intraoperative image of a red papillary tumor arising from the right diaphragm. The resected tumor measured 3.5 cm × 1.5 cm × 1.3 cm in size, was brown in color, and had a soft consistency.

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