Giant Leiomyoma Arising from the Mediastinal Pleura: A Case Report

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This report presents a rare case involving a patient with a giant leiomyoma originating from the mediastinal pleura. The patient underwent a medical examination, and chest radiography revealed a giant tumor. Computed tomography (CT) and magnetic resonance imaging (MRI) showed a well demarcated, heterogeneous mass which seemed to originate from the posterior mediastinum. Positron emission tomography (PET) showed the uptake of this tumor with a standardized uptake value of 4.9. We suspected that this tumor was a solitary fibrous tumor, and the patient underwent a surgical resection. Intraoperative exploration revealed a well-encapsulated tumor measuring 15 × 11 cm that appeared to originate from the mediastinal pleura. Immunohistochemical findings revealed a benign leiomyoma. We finally diagnosed the patient with a mediastinal leiomyoma. The present report describes CT, MRI, and PET findings of leiomyoma, and presents a review of relevant literature.

Keywords: leiomyoma, mediastinal pleura, computed tomography, magnetic resonance imaging, positron emission tomography

Introduction

A leiomyoma is a benign soft tissue tumor originating from the smooth muscles. Leiomyomas usually occur in the uterus, urinary tract, and gastrointestinal tract; however, pleural leiomyomas are rare. Additionally, the imaging findings of leiomyomas, including those obtained by computed tomography (CT), magnetic resonance imaging (MRI), and positron emission tomography (PET), are not well known.

The present report describes a rare case of a giant pleural leiomyoma and its imaging characteristics obtained by CT, MRI, and PET.

Case Report

A 42-year-old woman presented with a giant tumor in the left lower field on chest radiography (Fig. 1A). Both chest CT and MRI showed a well demarcated and heterogeneous mass measuring 12 × 9 cm. The mass originated from the posterior mediastinum or pleura and was positioned adjacent to the lower lobe of the lung and descending aorta (Fig. 1B). PET showed significant uptake of this tumor with a standardized uptake value (SUV) of 4.9. Laboratory findings, including tumor marker measurements, were negative. We suspected that this tumor was a solitary fibrous tumor (SFT), and the patient underwent a surgical resection. A posterolateral thoracotomy was performed through the sixth rib bed. The tumor, measuring 15 × 11 × 5 cm in diameter, seemed to originate from the mediastinal pleura and did...
Fig. 1  (A) Chest X-ray showed a huge mass in the left lower field. (B) Chest computed tomography showed a well demarcated and heterogeneous mass measuring $12 \times 9$ cm, originating from the posterior mediastinum or pleura and adjacent to the left lower lobe and descending aorta.

Fig. 2  (A) Macroscopic findings of resected tumor: the tumor was well encapsulated by fibrous tissue, and the borderline was sharp. (B) Intrathoracic findings: the stalk of the tumor seemed to originate from the mediastinal pleura. (C) Histological findings of the tumor (hematoxylin and eosin staining): mature smooth muscle cells proliferated in an interlacing, fascicular, or irregular pattern associated with focal liquefaction and fibrosis. (D) Immunohistochemical findings of the tumor: the tumor cells were positive for alpha-smooth muscle actin.
not invade adjacent organs. After ligating and cutting the stalk, tumor extirpation was completed (Fig. 2A). The stalk’s diameter was 1.8 cm. The tumor was well encapsulated by fibrous tissue, and the borderline was sharp (Fig. 2B). Mature smooth muscle cells proliferated in an interlacing, fascicular, or irregular pattern and were associated with focal liquefaction and fibrosis (hematoxylin and eosin stain, × 40) (Fig. 2C). Tumor cell mitotic figures were rarely seen. Immunohistochemically, the tumor cells were positive for alpha-smooth muscle actin (Fig. 2D), HHF-35, and desmin, but negative for S-100 protein and CD34. There was no evidence of malignancy. These features were compatible with a benign leiomyoma. And the tumor had no positional relationship with adjacent structures such as esophagus, bronchus, and vascular except the mediastinal pleura in the operation findings, then we considered that the origin of this leiomyoma was the mediastinal parietal pleura.

The patient had an uneventful postoperative recovery without any complications. At the time of this writing, the patient had been in good clinical condition with no recurrence for 5 years after surgery.

**Discussion and Conclusion**

Leiomyomas originate from smooth muscles and were first described in 1854. Leiomyomas usually occur in the uterus, small intestine, and esophagus.1) Intrathoracic leiomyomas are rare; moreover, leiomyomas originating from the mediastinal pleura are extremely rare. Table 1 summarizes the patient and tumor characteristics of 12 previously described cases and our case. Based on these reports, it appears that pleural leiomyoma occurs more frequently in young to middle-aged female patients (9 of 12 cases).

Of the 12 cases of pleural leiomyomas, 6 tumors originated from the vascular smooth muscle or microvascular wall, and 6 reports did not define the origins of the tumors. In the present case, we found no microscopic vascular channels among the smooth muscle cells, and the tumor had no positional relationship with adjacent structures except the mediastinal pleura. These tumors, including our case, may have arisen from mesodermal cells that have acquired the capacity to differentiate along smooth muscle lines.3,4) Then, we considered that the origin of this leiomyoma was the mediastinal parietal pleura as previously reported.5)

The CT, MRI, and PET findings can reveal the tumor size, location, level of vascularization, and anatomic

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**Table 1: Clinical features of published case reports of primary pleural leiomyoma**

<table>
<thead>
<tr>
<th>Case</th>
<th>Author</th>
<th>Age/Sex</th>
<th>Tumor origin</th>
<th>Diameter (cm)</th>
<th>Operation</th>
<th>Prognosis</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>Tanaka T</td>
<td>40/F</td>
<td>MW</td>
<td>3.5 × 3.0</td>
<td>CR</td>
<td>17 months: alive without recurrence</td>
</tr>
<tr>
<td>2</td>
<td>Moran CA</td>
<td>21/F</td>
<td>VSM</td>
<td>Multiple fragments</td>
<td>Incomplete resection</td>
<td>4 months: alive</td>
</tr>
<tr>
<td>3</td>
<td>Moran CA</td>
<td>23/F</td>
<td>VSM</td>
<td>10.0 × 9.0</td>
<td>Incomplete resection</td>
<td>6 months: alive</td>
</tr>
<tr>
<td>4</td>
<td>Proca DM</td>
<td>32/M</td>
<td>No details</td>
<td>4.3 × 7.0</td>
<td>CR</td>
<td>12 months: alive without recurrence</td>
</tr>
<tr>
<td>5</td>
<td>Mochizuki H</td>
<td>33/M</td>
<td>No details</td>
<td>3.0 × 3.2</td>
<td>CR</td>
<td>Unknown</td>
</tr>
<tr>
<td>6</td>
<td>Nose N</td>
<td>55/F</td>
<td>MW</td>
<td>1.5 × 1.5</td>
<td>CR</td>
<td>26 months: alive without recurrence</td>
</tr>
<tr>
<td>7</td>
<td>Turhan K</td>
<td>50/F</td>
<td>VSM</td>
<td>4.0 × 4.0</td>
<td>CR</td>
<td>53 months: alive without recurrence</td>
</tr>
<tr>
<td>8</td>
<td>Rodriguez PM</td>
<td>48/F</td>
<td>No details</td>
<td>18.0 × 14.0</td>
<td>CR</td>
<td>18 months: alive without recurrence</td>
</tr>
<tr>
<td>9</td>
<td>Qiu X</td>
<td>45/M</td>
<td>No details</td>
<td>9.0 × 5.5</td>
<td>CR</td>
<td>14 months: alive without recurrence</td>
</tr>
<tr>
<td>10</td>
<td>Ziyade S</td>
<td>33/F</td>
<td>VSM</td>
<td>5.3 × 8.0</td>
<td>CR</td>
<td>Unknown</td>
</tr>
<tr>
<td>11</td>
<td>Arikura J</td>
<td>52/F</td>
<td>No details</td>
<td>6.5 × 5.5</td>
<td>CR</td>
<td>60 months: alive without recurrence</td>
</tr>
<tr>
<td>12</td>
<td>This case</td>
<td>42/F</td>
<td>Not clear</td>
<td>15.0 × 12.0</td>
<td>CR</td>
<td>Not clear</td>
</tr>
</tbody>
</table>

CR: complete resection, MW: micro-vascular wall, VSM: vascular smooth muscle
relationships. On CT, leiomyomas are solitary, well-circumscribed tumors with heterogeneous density. We also performed MRI, which is the first such reported examination of a pleural leiomyoma. Iso-signal intensity was observed on T1-weighted images. Heterogeneous high-signal intensity was observed on T2-weighted images. Heterogeneous enhancement was observed on contrast-enhanced T1-weighted images. On the other hand, PET findings showed heterogeneous increased uptake of fludeoxyglucose (FDG) in the entire tumor (SUVmax = 4.9). Arikura also reported a case of pleural leiomyoma examined by FDG-PET, which was the only case performed FDG-PET except for our case. The findings also showed heterogeneous increased uptake of FDG in the entire tumor (SUVmax = 6.1), similar to our case.

These CT, MRI, and PET findings are very similar to the features of SFTs, and our first diagnosis was SFT. Differential diagnosis between SFT and pleural leiomyoma was difficult. In addition, PET cannot reliably differentiate leiomyosarcoma from leiomyoma. However, few reports of pleural leiomyomas describe MRI or PET examination. Such studies may be useful for diagnosis if the numbers of cases increase.

On the other hand, MRI is useful for the evaluation of the tumor’s invasion with the lung, mediastinum, and chest wall, especially for the large tumor such as our case. With MRI, we could judge the large tumor was resectable.

To make an accurate preoperative diagnosis of intra-thoracic soft tissue tumor, CT-guided transthoracic fine-needle aspiration (FNA) is commonly useful. Qiu Zhu D performed FNA and made the preoperative diagnosis of pleural leiomyoma, which was the only case that made the accurate preoperative diagnosis. Leiomyomas appeared benign; however, it has a low but definite malignant potential. After a transthoracic fine-needle biopsy, leiomyoma has the possibility of metastasizing or disseminating through the needle tract. It may not be necessary for the intrathoracic soft tissue tumor to perform the FNA if the tumors can be resected completely. If the tumor is unresectable, FNA should be performed to make the pathologic diagnosis for further medical treatment.

According to our modest review of the literature, when located in atypical regions, leiomyomas are generally benign and usually completely resectable. Complete resection generally results in a cure. No recurrence was observed in the 10 cases involving complete resection. In our case, for 5 years after the operation (the longest postoperative follow-up term), no recurrence was observed. Considering the good prognosis after complete resection, the low malignant potential, and the possibility of serious symptoms after the tumor has grown very large, surgical excision of leiomyomas is justified and recommended.

In conclusion, pleural leiomyomas are very rare tumors of the intrathoracic space. Although we have described the CT, MRI, and PET characteristics of these tumors, an accurate preoperative diagnosis is difficult. Complete resection of pleural leiomyomas is recommended for an accurate diagnosis and good prognosis.

Consent

Written informed consent was obtained from the patient for publication of this case report and all accompanying images.

Disclosure Statement

The authors declare that they have no competing interests.

References