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Case report

Successful removal of a giant recurrent mediastinal liposarcoma involving both hemithoraces

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Abstract

Primary liposarcomas of the mediastinum are unusual tumors. We report herein a case of a 52-year-old woman, who was found to have a mediastinal tumor involving both hemithoraces and radiologically showing non-resectable-invasive features to the adjacent vital structures. She had a history of left thoracotomy for mediastinal schwannoma 14 years previously. The patient underwent an exploratory thoracotomy following a preoperative misdiagnosis of an ancient schwannoma. Complete removal of the tumor was accomplished through a right posterolateral thoracotomy with a subsequent histological diagnosis of a recurrent low-grade liposarcoma. A resectable liposarcoma should be considered in the differential diagnosis of a mediastinal tumor, although radiologically, the tumor presents with invasive features. © 2001 Elsevier Science B.V. All rights reserved.

Keywords: Liposarcoma; Mediastinum; Thoracic surgery

1. Introduction

Liposarcomas are the most common soft tissue sarcomas of adult life [1]; however, those located in the mediastinum are extremely rare [1–4]. Mediastinal liposarcomas constitute 2.7 and 0.126% of all liposarcomas and soft tissue sarcomas, respectively [1,4]. To date, mediastinal liposarcomas regarding various clinicopathological features have been reported, however, we are not aware of any previous report of a mediastinal liposarcoma, involving both hemithoraces, which recurred 14 years later and eventually excised transmediastinally through a right thoracotomy with a single-stage procedure.

We report herein a case of a giant recurrent mediastinal liposarcoma involving both hemithoraces and radiologically showing non-resectable-invasive features, which we explored and totally excised following a preoperative histological misdiagnosis of an ancient schwannoma.

2. Case report

A 52-year-old woman, who was otherwise well, presented with mild shortness of breath with 1 year duration and a recent onset of chest pain. She had a history of a left thoracotomy for a mediastinal schwannoma 14 years previously. Physical examination showed dullness on percussion and bilateral decreased breath sounds in the lower zones of the lungs. Laboratory data, respiratory function tests and arterial blood gas analyses were within the normal limits. Chest X-ray showed an enlargement of the middle and lower mediastinum. On computed tomography (CT), a mass in the right hemithorax, extending to the left side, showing invasive features to the left atrium, left ventricle, esophagus, aorta, liver and diaphragma, was defined. It was a heterogeneous and hypodense mass with central necrosis and calcification foci. Evidence of pleural effusion also was seen. On the other hand, magnetic resonance imaging (MRI) revealed no direct invasion to the above indicated structures, but possible invasion to the esophagus (Fig. 1). Detection for distant metastases including bone scan, cranial and abdominal CTs showed no abnormal findings. Esophagoscopy and bronchoscopy revealed an extrinsic compression effect of the tumor, but no evidence of intraluminal tumor. A transthoracic CT-guided core-needle biopsy specimen showed spindle cells with pleomorphic

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Fig. 1. Coronal MR image shows a large, lobulated, hyperintense supradiaphragmatic mass encasing the distal esophagus (arrow) and the extension of the mass into the subcarinal region.

nuclear features, but no evidence of mitosis, and it was interpreted as an ancient schwannoma, consistent with the recurrence of the previously excised schwannoma.

On right posterolateral thoracotomy, a giant, lobulated and well-demarcated mass, located in the posterior mediastinum, was explored with 200 ml of serous pleural fluid. The tumor extended to the tracheal carina superiorly and to the diaphragma inferiorly. It showed a prominent compression on middle–lower lobes of the lung. The capsule was incised and the right hemithorax component of the tumor was excised. There was no evidence of invasion to the contiguous structures, whereas dense adhesions to the esophagus were detected and dissected meticulously under the guidance of a nasogastric tube. The esophagus was tapered and retracted to facilitate the dissection. The right component had an attachment with the left component behind the esophagus. With the guidance of this attachment, an intracapsular transmediastinal approach provided the exploration of the left component and showed that it was in close association with the left atrium and ventricle anteriorly and the thoracic aorta posteriorly, but showed no invasion to these vital structures. Following blunt dissection, the left component was eventually enucleated to the right hemithorax with resultant removal of the mass. The postoperative course was uneventful. The patient remains well and disease-free 3 months after surgery.

The entire tumor measured $33 \times 30 \times 17$ cm in diameter and weighed 1520 g in total. The mass was soft and pale yellow in color on cut section. On histological examination, the tumor consisted of loosely arranged fibroblast-like undifferentiated spindle cells with focal lipoma-like areas. Spindle cells showed mild to moderate pleomorphism with rare mitosis, some being atypical. A maze of branching narrow vessels was distinct in some areas. Focal areas of osseous metaplasia also were observed. An immunohistochemical study for cytokeratin, SMA, CD68, FVIII, CD34, and desmin was negative in the tumor cells. Vimentin was positive and S-100 stained the lipoma-like areas. Spindle cells only showed vimentin expression. The tumor was diagnosed as a low-grade liposarcoma (Fig. 2). Retrospective histological examination of the formerly excised tumor revealed that the present tumor was the recurrence of this tumor.

3. Discussion

Liposarcomas take their origin from primitive mesenchymal cells. They are almost always encountered in deeper

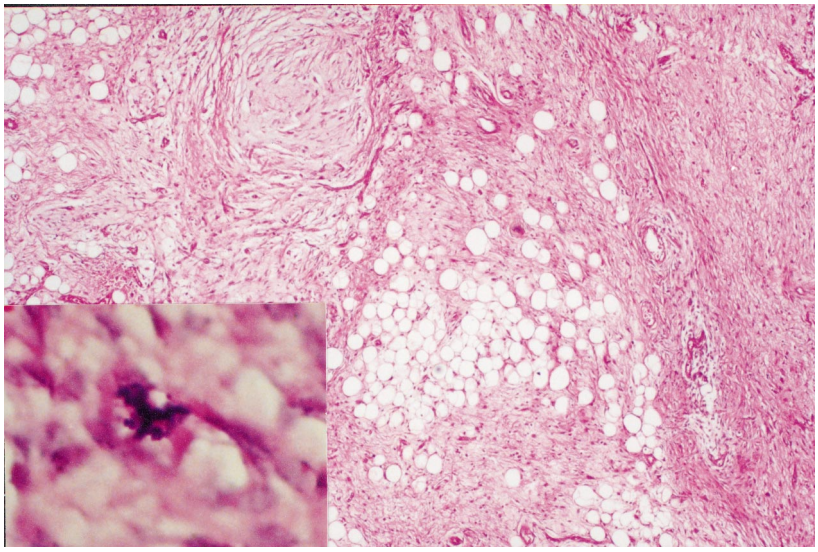


Fig. 2. Lipoma-like areas and spindle cells (hematoxylin eosin, ×40). Tripolar mitosis in spindle areas (inset; hematoxylin eosin, ×200).

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structures as insidiously growing tumors. Liposarcomas occurring in the mediastinum extend into the pleural spaces and they may achieve a large size before detection [1]. The presenting signs and symptoms are related to size and direct invasion of contagious structures. Dyspnea, tachypnea and wheezing are the most common symptoms, followed by chest pain. Asymptomatic cases discovered by radiological imaging also have been reported [2,3]. Despite the enormous size of the mass, our patient had a good performance status, and showed only a mild shortness of breath.

Mediastinal liposarcomas, most commonly arising from the thymus-related fatty tissue in the anterior mediastinum also might occur in the posterior mediastinum [2,5]. To our knowledge, our patient is unique, in that the liposarcoma was located in the posterior mediastinum and involved both hemithoraces.

On CT, the appearance of mediastinal liposarcomas, as of liposarcomas located in any part of body, varies from a predominantly fat-containing mass to a solid mass [5]. Low attenuation values between −50 and −150 Hounsfield Unit (HU) are consistent with a tissue composed of fat. Greater values are related to the necrosis, heterogeneity and soft tissue component in liposarcomas [6]. On MR imaging, T1-weighted images show the fatty tissue with a high signal intensity, whereas the signal intensity diminishes in T2-weighted images [7]. A differential diagnosis should be made between lipoma, thymolipoma, and teratoma [5]. As a superior imaging technique to CT in delineating the mediastinal invasion, MRI revealed a possible invasion of the tumor only to the esophagus, whereas CT showed the tumor as an unresectable mass in our patient.

Liposarcomas have very low sensitivity, both to radiotherapy and chemotherapy. High-doses of radiation may result in mediastinal fibrosis, thus precluding radiotherapy in this location [2]. Data on the efficacy of chemotherapy is limited and its role has yet to be determined [8]. Hence, complete surgical removal is the optimal treatment for a mediastinal liposarcoma. Partial excision or debulking of the tumor may relieve the compression effect of more advanced and infiltrating tumors [2–4]. In our case, surgical removal, at least, in part may be attributed to the misdiagnosis of an ancient schwannoma, which led us to explore the tumor through a thoracotomy. Exploration revealed a well-demarcated tumor and enabled its complete removal, which would otherwise gradually enlarge and invade to the vital structures.

Recurrence is common in deep-seated liposarcomas and it becomes apparent within the first 6 months in most cases, but it may be delayed for 5 or 10 years following the initial excision. Recurrence is related to the incomplete excision and tumor tissue left behind at the time of surgery [1]. In the presented case, recurrence occurred 14 years after the initial surgery, which draws attention to the significance of long-term follow-up in these patients.

In conclusion, should any suspicion arise about the resectability of a radiologically invasive tumor located in the mediastinum with a possible histological diagnosis of a low-grade liposarcoma, surgical exploration may result in complete removal of the tumor.

References

[1] Enzinger FM, Weiss SW, editors. Liposarcoma. Soft tissue tumors, 3rd ed. St. Louis, MO: Mosby, 1995. pp. 431–466.
[2] Grewal RG, Prager K, Austin JHM, Rotterdam H. Long-term survival in non-encapsulated primary liposarcoma of the mediastinum. Thorax 1993;48:1276–1277.
[3] Attal H, Jensen J, Reyes CV. Myxoid liposarcoma of the anterior mediastinum. Diagnosis by fine needle aspiration biopsy. Acta Cytol 1995;39:511–513.
[4] Burt M, Ihde JK, Hajdu SI, Smith JW, Bains MS, Downey R, Martini N, Rusch VW, Ginsberg RJ. Primary sarcomas of the mediastinum: results of therapy. J Thorac Cardiovasc Surg 1998;115:671–680.
[5] Eisenstat R, Bruce D, Williams LE, Katz DS. Primary liposarcoma of the mediastinum with coexistent mediastinal lipomatosis. AJR Am J Roentgenol 2000;174:572–573.
[6] Epler GR, Mcloud TC, Munn CS, Colby TV. Pleural lipoma. Diagnosis by computed tomography. Chest 1986;90:265–268.
[7] Munk PL, Lee MJ, Janzen DL, Connell DG, Logan PM, Poon PY, Bainbridge TC. Lipoma and liposarcoma: evaluation using CT and MR imaging. AJR Am J Roentgenol 1997;169:589–594.
[8] Mclean TR, Almassi GH, Hackbarth DA, Janjan NA, Potish RA. Mediastinal involvement by myxoid liposarcoma. Ann Thorac Surg 1989;47:920–921.

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