CASE REPORT – OPEN ACCESS

International Journal of Surgery Case Reports 41 (2017) 292–295

Contents lists available at ScienceDirect

International Journal of Surgery Case Reports

journal homepage: www.casereports.com

A giant mediastinal liposarcoma weighing 3500 g resected with clam shell approach, a case report with review of literature

Yasoo Sugiura*, Toshinori Hashizume, Hiroyuki Fujimoto, Etsuo Nemoto

Department of General Thoracic Surgery, National Hospital Organization, Kanagawa National Hospital, 666-1 Ochiai Hadano, Kanagawa 257-8585, Japan

ARTICLE INFO

Article history:
Received 7 September 2017
Received in revised form 30 October 2017
Accepted 30 October 2017
Available online 7 November 2017

Keywords:
Liposarcoma
Mediastinum
Calm shell approach
Adjuvant therapy
Case report

ABSTRACT

INTRODUCTION: Liposarcoma is rare in the mediastinum and is less than 1% of all mediastinal tumors. In the present report, we demonstrated our case and summarized the principal treatment of the mediastinal liposarcoma with literature review.

PRESENTATION OF CASE: A 50-year-old man presented at our hospital with complain of dyspnea. Chest radiography showed remarkable cardiomegaly. Computed tomography revealed an anterior mediastinal tumor from the level of the cephalic vein to the diaphragm of bilateral thoracic cavity with fat component. Using clam shell approach, complete en bloc resection of the tumor was performed. The weight of the tumor was 3500 g. The pathological findings were 0that size of adipocyte and lipoblast were different, and the nuclei of atypical stromal cell were misshapen. Immune-histologic examination was negative for MDM2 and cyclin-dependent kinase 4. The diagnosis was liposarcoma, well-differentiated type. He could discharge 10 days after surgery. Without adjuvant therapy, disease free survival for three years has passed.

DISCUSSION: From 1990–2016 in Japan, 60 cases of the mediastinal liposarcoma were reported. In analysis of the 61 cases including the present case, adjuvant therapy was performed in 14 cases, subsequently, and recurrence was recognized in 5 cases. Adjuvant therapy did not significantly suppress the recurrence.

CONCLUSION: Mediastinal liposarcoma weighing 3500 g could be resected using calm shell approach, and no recurrence interval for 3 years has been achieved without adjuvant therapy. Complete resection is the only means to achieve the favorable outcome in mediastinal liposarcoma.

1. Introduction

Liposarcoma is rare in the mediastinum [1,2]. Complete resection is the only means to achieve the favorable outcome in the cases of sarcoma including liposarcoma [3]. In the cases of mediastinal liposarcoma, resection with fully margin is difficult because the vital organs surround the mediastinum. In the present case, we accomplished complete resection for the giant liposarcoma weighing 3500 g, using clam shell approach. We report the case with literature review of 60 cases from 1990 to 2016 in Japan, because there is hardly study with analysis about the significance of an adjuvant therapy for the mediastinal liposarcoma. This work has been reported in line with the SCARE criteria [4].

2. Presentation of case

A 50-year-old man presented at our hospital with the complain of dyspnea. Chest radiography showed remarkable cardiomegaly (Fig. 1A). He did not have any past medical history, family history and any relevant genetic information. His smoking history was 2 packs per day for 30 years and his occupation was a truck driver. Computed tomography and magnetic resonance imaging revealed a giant anterior mediastinal tumor with fat component and there was a nodule with calcification partially (Fig. 1B, C and D). The tumor extended from the space of the mediastinum at the level of the cephalic vein to the diaphragm of bilateral thoracic cavity. We considered that the difference diagnosis of the tumor was mediastinum liposarcoma or teratoma. Surgical resection was planned to relive the dyspnea.

We chose clam shell approach at bilateral the fifth intercostal level to assure the safe operation field in the both the anterior mediastinum and the bilateral thoracic cavity. The first author who has been a surgeon for 10 years was the operator. The tumor adhered loosely to the chest wall, the cardiac sac and the diaphragm. The tumor could be mobilized with energy devices from the surrounded anatomical structures. Two thin veins flowing into the cephalic vein from the tumors were ligated. A complete en bloc resection of the tumor including an entire capsule was performed. The weight of the tumor was 3500 g (Fig. 2A). In the findings of hematoxylin and eosin stain, the size of adipocyte and lipoblast were different, and the nuclei of atypical stromal cell were misshapen (Fig. 2B). The nodule lesion was surrounded a scar-formed fibrosis and fell into necro-

* Corresponding author.
E-mail address: dryasoo@outlook.com (Y. Sugiura).

https://doi.org/10.1016/j.ijscr.2017.10.055

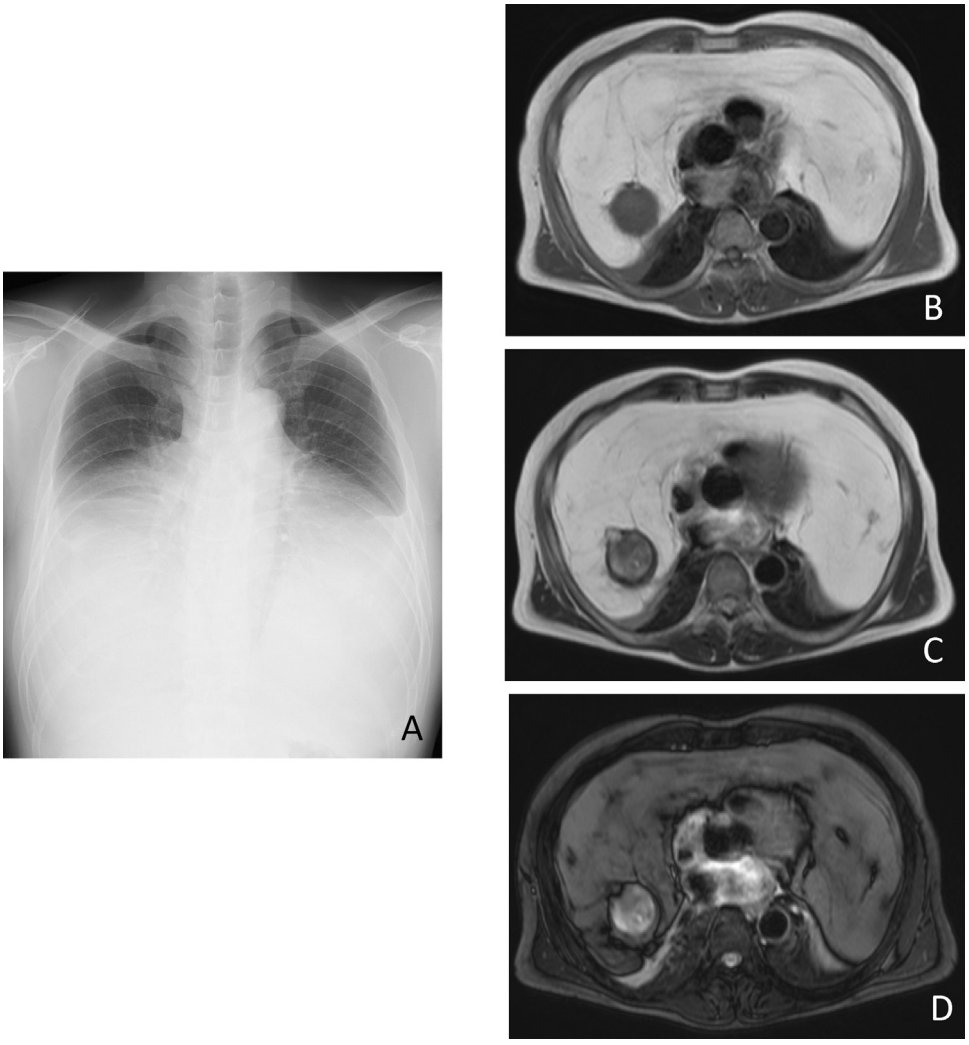

Fig. 1. Chest radiography (A) showed remarkable cardiomegaly. T1-weighted image (B), T2-weighted image (C) and fat suppression T2-weighted image of magnetic resonance revealed a giant anterior mediastinal tumor with fat component, suggesting liposarcoma.

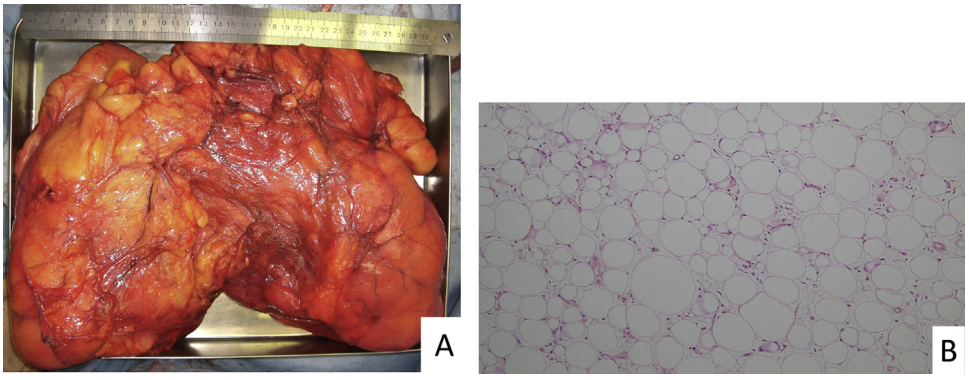

Fig. 2. The weight of the resected liposarcoma was 3500 g (A). In the findings of hematoxylin and eosin stain, the size of adipocyte and lipoblast were different, and the nuclei of atypical stromal cells were misshapen (B).

sis in the center. Immune-histologic examination was negative for MDM2 and cyclin-dependent kinase 4. The final pathological diagnosis was liposarcoma, well-differentiated type. He could discharge on 10 days after surgery without dyspnea. Without adjuvant therapy, disease free survival for three years has passed in checking the recurrence by CT every 6 months.

3. Discussion

Liposarcoma arises from precursors of adipocytes and mediastinal liposarcoma is less than 1% of all mediastinal tumors [2]. Liposarcoma is classified into 5 histologic subtypes: myxoid, well-differentiated, dedifferentiated, pleomorphic and mixed liposarcoma [1]. In general, the principle of the treatment for

liposarcoma is complete resection [5–7]. There are hardly reliable randomized control study nor series report to evaluate the significance of an adjuvant therapy for the mediastinal liposarcoma [8,9]. Our search with using Ichushi-Web by NPO Japan Medical Abstracts Society found 60 cases of mediastinal liposarcoma including Japanese and English literatures from 1990 to 2016. The clinical characteristics and outcome of 61 cases, including our case, are listed in Table 1. There were 47 men and 14 women with ages ranging from 12 to 82 years old (median ± standard error (SE): 62 ± 1.79 years old). All patients underwent surgical resection, including incomplete resection in 4 cases. The pathological subtype was that well differentiated liposarcoma was 30 cases (49%), dedifferentiated liposarcoma was 14 cases (23%), myxoid liposarcoma was 9 cases (15%), pleomorphic liposarcoma was 5 cases (8%), and mixed liposarcoma was 3 cases (5%). No recurrence cases were 30 (60%), recurrence cases were 20 (40%) and 11 cases were not mentioned. The median of no recurrence interval ± SE was 18 ± 11.74 months (6–480 months), of which the total observation interval ± SE was 23 ± 13.45 months (6–489 months). The cases without adjuvant therapy were 37 cases (72.5%). In cases with adjuvant therapy, 8 cases (15.7%), 5 cases (9.8%) and 1 case (2.0%) underwent radiation therapy, chemotherapy and chemo-radiotherapy, respectively.

From the perspective of the adjuvant therapy, there was not significant difference in the proportion between the adjuvant therapy and recurrence by chai square test (Table 2). Regarding each histologic subtype, in all 4 cases of dedifferentiated liposarcoma with adjuvant therapy, recurrence was not recognized (Table 3). Therefore, complete resection is the only means to achieve the favorable outcome in mediastinal liposarcoma.

The limitation of this analysis is that the data based on the case reports, which subsume publication bias. Further studies are needed to improve the outcome of cases of mediastinal liposarcoma, and an effective treatment will evolve.

Table 1
Clinical characteristics and outcome of patients in 61 Japanese cases from 1990 to 2016.

Gender		
male	47	
female	14	
Age (median ± SE, years old)	62 ± 1.79	Range: 12–82
No recurrence interval (median ± SE, months)	18 ± 11.74	Range: 6–480
Total observation interval (median ± SE, months)	23 ± 13.45	Range: 6–489
Histological subtype		
well differentiated	30	49.2%
dedifferetiated	14	23.0%
myxoid	9	14.8%
pleomorphic	5	8.2%
mixed	3	4.9%
Adjuvant therapy		
not performed	36	72.0%
radiation	8	16.0%
chemotherapy	5	10.0%
chemo-radiotherapy	1	2.0%
not mentioned	11	
Outcome		
recurrence	19	38.0%
no recurrence	31	62.0%
not mentioned	11	

SE; standard error.

Table 2
The distribution of the cases of mediastinal liposarcoma with or without adjuvant therapy and recurrence.

		recurrence		
		yes	no	
Adjuvant therapy	yes	15	21	36
	no	4	10	14
		19	31	50

$p = 0.595$

For comparisons of proportions, chai square test was used. The result was considered significant at $P < 0.05$. The statistical analysis was performed using Stat Mate IV software version 4.01 (ATM, Tokyo, Japan).

Table 3
The outcome of each histologic subtype of mediastinal liposarcoma with or without adjuvant therapy.

			well-differentiated		pleomorphic		myxoid		dedifferentiated		mixed	
			recurrence									
			no	yes	no	yes	no	yes	no	yes	no	yes
adjuvant therapy	no		14	6	0	4	2	1	4	4	1	0
	yes	radiation	3	1	1	0	1	1	0	0	0	0
		chemotheray	0	1	0	0	0	1	3	0	0	0
		chemoradiation	0	0	0	0	0	1	0	0	0	0
		heavy-ion laser	0	0	0	0	0	0	1	0	0	0

4. Conclusion

Mediastinal liposarcoma weighing 3500 g could be resected using calm shell approach, and no recurrence interval for 3 years has been achieved without adjuvant therapy. The literature review of 61 Japanese cases of mediastinal liposarcoma demonstrated that the adjuvant therapy did not have value to suppress the recurrence of the mediastinal liposarcoma. Therefore, complete resection is the only means to achieve the favorable outcome in mediastinal liposarcoma.

Consent

Written informed consent for the *peri*-operative care and the operation in this case was obtained on admission in accordance with institutional guidance based on the Helsinki Declaration. At the time of discharge, we reconfirmed that the patient gave permission to publish a case report about his clinical course with any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

Author contribution

Yasoo Sugiura – Study concept or design, data collection, literature search, writing paper, final decision to publish.

Toshinori Hashizume – Data collection, final decision to publish.

Hiroyuki Fujimoto–Study concept or design, final decision to publish.

Etsuo Nemoto–Study concept or design, final decision to publish.

Funding

There were no sponsors involved in the study.

Ethical approval

As this was a case report, informed consent has been taken from the patient.

Registration of research studies

Not applicable, because this study did not deal with clinical trial and any experiment. The present study described the clinical course about the surgery for the giant mediatinal liposarcoma and the literature review.

Guarantor

Etsuo Nemoto is responsible for all aspects of management as president of Kanagawa National Hospital and chief of the department of pulmonary and thoracic surgery.

Conflicts of interest statement

The author and co-authors have no potential conflict of interests to disclose.

References

[1] C.D. Fletcher, The evolving classification of soft tissue tumours – an update based on the new 2013 WHO classification, Histopathology 64 (1) (2014) 2–11.
[2] R.G. Grewal, K. Prager, J.H. Austin, H. Rotterdam, Long term survival in non-encapsulated primary liposarcoma of the mediastinum, Thorax 48 (12) (1993) 1276–1277.
[3] A.M. Crago, S. Singer, Clinical and molecular approaches to well differentiated and dedifferentiated liposarcoma, Curr. Opin. Oncol. 23 (4) (2011) 373–378.
[4] R.A. Agha, A.J. Fowler, A. Saeta, I. Barai, S. Rajmohan, D.P. Orgill, et al., The SCARE Statement: Consensus-based surgical case report guidelines, Int. J. Surg. 34 (2016) 180–186.
[5] M. Burt, J.K. Ihde, S.I. Hajdu, J.W. Smith, M.S. Bains, R. Downey, et al., Primary sarcomas of the mediastinum: results of therapy, J. Thorac. Cardiovasc. Surg. 115 (3) (1998) 671–680.
[6] C.N. Catton, B. O'Sullivan, C. Kotwall, B. Cummings, Y. Hao, V. Fornasier, Outcome and prognosis in retroperitoneal soft tissue sarcoma, Int. J. Radiat. Oncol. Biol. Phys. 29 (5) (1994) 1005–1010.
[7] S.W. Kendall, E.A. Williams, J.B. Hunt, M.C. Petch, F.C. Wells, B.B. Milstein, Recurrent primary liposarcoma of the pericardium: management by repeated resections, Ann. Thorac. Surg. 56 (3) (1993) 560–562.
[8] D.S. Klimstra, C.A. Moran, G. Perino, M.N. Koss, J. Rosai, Liposarcoma of the anterior mediastinum and thymus. A clinicopathologic study of 28 cases, Am. J. Surg. Pathol. 19 (7) (1995) 782–791.
[9] H.P. Hahn, C.D. Fletcher, Primary mediastinal liposarcoma: clinicopathologic analysis of 24 cases, Am. J. Surg. Pathol. 31 (12) (2007) 1868–1874.