Case report

A refractory pleural effusion caused by a pleural capillary hemangioma

Kazumasa Akagi a,*, Toyoshi Matsutake b, Kiyoyasu Fukushima a, Hiroshi Mukae c, Hirokazu Kurohama d, Hirofumi Matsumoto e

a Department of Respiratory Medicine, Japanese Red Cross Nagasaki Genbaku Ishihaya Hospital, Ishihaya City, Nagasaki, 859-0497, Japan
b Department of Respiratory Medicine, Koseikai Hospital, Nagasaki City, Nagasaki, 852-8053, Japan
c Department of Respiratory Medicine, Nagasaki University Graduate School of Biomedical Sciences, Nagasaki City, Nagasaki, 852-8501, Japan
d Department of Diagnostic Pathology, Isahaya General Hospital, Isahaya City, Nagasaki, 854-8501, Japan
e Department of Thoracic Surgery, Isahaya General Hospital, Isahaya City, Nagasaki, 854-8501, Japan

ARTICLE INFO

Keywords:
Capillary hemangioma
Pleural effusion
Refractory
Nonspecific pleuritis

ABSTRACT

A 69-year-old man presented with a left pleural effusion. Even after repeated drainage, the pleural effusion had been increasing for more than two years. Thoracoscopy unexpectedly showed a pleural mass on the parietal pleura, and it was completely removed. The diagnosis was pleural capillary hemangioma, and the effusion has not recurred after the resection. Pleural hemangioma is one of the crucial differential diagnoses of refractory pleural effusion.

1. Case report

A 69-year-old man presented with a left pleural effusion. Laboratory data showed slightly increased white blood cells (9170/μL) and C-reactive protein (0.99 mg/dL). The carcinoembryonic antigen level was high (10.2 ng/mL), but it decreased spontaneously. The pleural effusion was exudative and lymphocyte-dominant (99%). On evaluation of the effusion, the cytology was class II, and the carcinoembryonic antigen level was not increased (<5.8 pg/mL). Positron emission tomography-computed tomography did not show fluorodeoxyglucose uptake. The pleural effusion was exudative and lymphocyte-dominant (99%).

Generally, hemangioma is a frequently used term, but the definition of a hemangioma is controversial. For convenience, the diagnosis has been made according to the size of increased vessels, such as cavernous hemangioma. Recently, in the International Society for the Study of Vascular Anomalies classification [2], abnormalities of endothelial cells or mitoses and the lesion’s development are considered, and the present case was classified as a capillary malformation.

2. Discussion

A pleural biopsy is performed to evaluate a pleural effusion, but about 30% of the cases are of unknown etiology and diagnosed as nonspecific pleuritis [1].

A pleural biopsy is performed to evaluate a pleural effusion, but about 30% of the cases are of unknown etiology and diagnosed as nonspecific pleuritis [1].

There were increased small capillaries (CD34-positive), with no abnormalities of endothelial cells or mitoses (Fig. 2).

There were increased small capillaries (CD34-positive), with no abnormalities of endothelial cells or mitoses (Fig. 2). Providing supported that the mass was a malformation rather than a tumor, and it was diagnosed as a pleural capillary hemangioma. After the surgery, the pleural effusion resolved and did not recur.
treatment of hemangiomas, dissection, laser radiation, and sclerotherapy are considered. For pleural hemangiomas, dissection could be reasonable and radical, and no recurrences of the pleural effusions were seen after dissection in the reported cases [4].

In the clinical course of nonspecific pleuritis, malignancies could ultimately be identified, and one year of follow-up has been considered sufficient [5]. If a pleural effusion remains after one year of follow-up, thoracoscopy is recommended and could lead to an unexpected diagnosis as this capillary hemangioma case.

3. Conclusion

Though extremely rare, pleural hemangioma is one of the crucial differential diagnoses of refractory pleural effusion.

Formatting of funding sources

This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

Author statement

Kazumasa Akagi: Conceptualization, Investigation, Writing – original draft preparation, Toyoshi Matsutake: Writing-Reviewing and Editing, Kiyoyasu Fukushima: Writing-Reviewing and Editing, Hiroshi Mukae: Supervision, Hirokazu Kurohama: Resources, Writing-Reviewing and Editing, Hirofumi Matsumoto: Resources, Writing-Reviewing and Editing

Declaration of competing interest

None.
References

[1] H.E. Davis, J.E. Nicholson, N.M. Rahman, E.M. Wilkinson, R.J.O. Davies, Y.C.G. Lee, Outcome of patients with nonspecific pleuritis/fibrosis on thoracoscopic pleural biopsies, Eur. J. Cardio. Thorac. Surg. 38 (4) (2010) 472–477, https://doi.org/10.1016/j.ejcts.2010.01.057.

[2] International society for the Study of vascular Anomalies classification 2018 [internet] [cited 2020 July 19] Available from: https://www.issva.org/classification.

[3] H. Mimura, S. Akita, A. Fujino, M. Jinnin, M. Ozaki, K. Osuga, et al., Japanese clinical practice guidelines for vascular Anomalies 2017, Pediatr. Int. 62 (3) (2020) 257–304, https://doi.org/10.1111/ped.14077.

[4] G. Sindhwani, R. Khanduri, S. Nadia, V. Jethani, Pleural haemangioma: a rare cause of recurrent pleural effusion, Respir. Med. Case Rep. 17 (2016) 24–26, https://doi.org/10.1016/j.rmcr.2015.12.004.

[5] Z.S. DePew, A. Verma, D. Wigle, J.J. Mullon, F.C. Nichols, F. Maldonado, Nonspecific pleuritis: optimal duration of follow up, Ann. Thorac. Surg. 97 (6) (2014) 1867–1871, https://doi.org/10.1016/j.athoracsur.2014.01.057.